On Your Own Feet

**Adolescents with chronic conditions
and their preferences and competencies for care**

Op Eigen Benen

**Jongeren met chronische aandoeningen
en hun preferenties en competenties voor de zorg**

AnneLoes van Staa

On Your Own Feet

Adolescents with chronic conditions
and their preferences and competencies for care

Op Eigen Benen

Jongeren met chronische aandoeningen
en hun preferenties en competenties voor de zorg

Proefschrift

ter verkrijging van de graad van doctor aan de Erasmus Universiteit Rotterdam
op gezag van de rector magnificus prof.dr. H.G. Schmidt
en volgens besluit van het College voor Promoties.

De openbare verdediging zal plaatsvinden op
woensdag 6 juni 2012 om 13.30 uur

door
AnneLoes van Staa
geboren te Johannesburg (Z-A)

ERASMUS UNIVERSITEIT ROTTERDAM

Promotiecommissie

Promotores: Prof.dr. M.J. Trappenburg
Prof.dr. H.A. Moll

Overige leden: Prof.dr. R. Bal
Prof.dr. J.M.W. Hazes
Prof.dr. A.J. van der Heijden

Contents

Introduction

Introduction to the research topic; aims & overview of the research projects

1 A small epidemiologic transition

Thirty years ago, in the summer of 1982, I was introduced to pediatric nursing during my internship in the Sophia Children's Hospital, Academic Hospital Rotterdam (now Erasmus MC). One of my unforgettable patients was Dirk, a 24-year-old man with cystic fibrosis (CF), who spent his time in hospital in a small room with glass windows (called "the box") where he could watch the toddlers play. At that time, he was our oldest CF-patient still alive. Even though Dirk was an adult, he wasn't treated like one, and he certainly did not look like one with his small stature, his "chicken breast", his swollen belly, and clubbed fingers. By the end of the summer, two of our CF-patients, Dirk and little Claudia (6 years old), were dead proving that CF was indeed a fatal disease.

Thirty years later, the scene for many of such fatal conditions in pediatric care has changed dramatically. The ability to successfully manage childhood-onset chronic diseases is one of the greatest advances in pediatric medicine.[1] Drastic improvements in life expectancy, as well as better functional outcomes, fewer disabilities, and less hospitalization have been reported for many congenital conditions, including cystic fibrosis, sickle cell disease, congenital heart disease, and metabolic disorders. Similar advances in survival were reached in acquired conditions such as childhood cancer, diabetes mellitus type 1, juvenile rheumatoid arthritis, and kidney failure (Box 1). When managed appropriately, many patients with previously lethal conditions can now expect to live near-normal lifespans.[1] This trend is sometimes called the *small epidemiologic transition* - a continuation of the large epidemiologic transition describing the change from acute to chronic diseases originating in the middle of the last century.[2] Due to an expansion in knowledge and possibilities to observe and intervene, infant and child mortality declined, but many congenital and acquired conditions became chronic instead.

Box 1   *Advances in the treatment of Cystic Fibrosis, Cancer, Congenital Heart Disorders, Diabetes Type 1, and Cerebral Palsy – and some drawbacks too*

> **Cystic fibrosis** (CF; also known as mucoviscidosis) is a recessive multi-system genetic disease characterized by abnormal transport of chloride and sodium across epithelium, leading to thick, viscous secretions in the lungs, pancreas, liver, and intestine. Estimated survival to 30 years increased from 6% in the 1950-to-1954 cohort, to 36% in the 1970-to-1973 cohort in the Netherlands. Exact survival to 15 years increased from 72% from the 1974-to-1979 cohort, to 91% in the 1985-to-1989 cohort. Contributors to the better survival include improved nutritional management and dietary recommendations, new airway clearance techniques, new antibiotics, as well as centralized care in specialized CF centers.[3] Better survival, however, has come at considerable societal cost due to intensified treatment with more lung transplants, and substantial (co)-morbidity.
>
> In **childhood cancers**, survival rates today are nearly 80%.[4] In the Netherlands, some 500 children each year are diagnosed with a form of neoplasm. One of the commonest childhood cancers is **acute lymphoblastic leukemia** (ALL), the most frequent form of leukemia in children. Leukemia is a type of cancer of the blood or bone marrow characterized by an abnormal increase of white blood

cells. Due to aggressive treatments, involving chemotherapy and radiotherapy, the survival rates for ALL in children near 85%, and are much better than for adults. However, nearly 40% of all survivors in a Dutch study suffered from medium to severe long-term effects.[5]

Congenital Heart Disease (CHD), the most common congenital disorder in newborns, used to be one of the major causes of infant death, but advances in cardiovascular medicine and surgery have enabled most patients to reach adulthood.[6, 7] The one-year survival rate in children with CHD born between 1985-1994 in one health region in the UK was 82%, and 96% of those who survived their first year were predicted to be alive at the age of 16 years.[8] Nevertheless, almost all patients operated upon in childhood for a congenital cardiac malformation require life-long, specialized medical care and are at increased risk for decreased life expectancy compared to healthy peers. However, studies indicate that the number of CHD survivors should be much higher than the number of patients actually seen in specialized centres.[9] This loss to follow-up starts to become substantial in the adolescent and teenage years.[10]

Diabetes mellitus type 1 (DM1) is a progressive autoimmune disease that destroys the cells that produce insulin. It was previously called insulin-dependent diabetes mellitus or juvenile-onset diabetes as it usually first develops in childhood or adolescence. Type 1 diabetes accounts for 10% of all diabetes cases. In the Netherlands, there are some 6000 children with diabetes type 1 and the incidence increases. In the 1950s, about one in five people diagnosed with type 1 diabetes died within 20 years, and one in three within 25 years after the diagnosis was made. Eye, nerve, kidney and cardiovascular complications were very prevalent. The long-term survival of those with type 1 diabetes has dramatically improved in the last 30 years. For people born between 1975 and 1980, about 3.5% died within 20 years after diagnosis, and 7% within 25 years. Intensive glucose control delays or helps prevent complications. However, adolescents and young adults generally poorly adhere to control of blood glucose levels, which increases their risk for acute complications (such as ketoacidosis) and for permanent damage in small vessels (NDF Zorgstandaard Addendum Diabetes type 1; Part 2, 2009; www.diabetesfederatie.nl).

Today, more than 90% of children with chronic conditions survive to adulthood. This implies that, unlike in the past, they will transition both to adulthood and to adult health care.[15] The small epidemiologic transition in pediatrics was first recognized around 1984, when a national invitational conference was held in Minnesota (USA): "*Youth with Disability: The Transition Years*". Five years later, Surgeon General C. Everett Koop convened a second conference: "*Growing Up and Getting Medical Care: Youth with Special Health Care Needs*".[16] Since then, we have witnessed a worldwide explosion of editorials and expert opinion articles about the impact of this transition for adult and pediatric health care services, and many professional organizations and government bodies published consensus statements and policy documents on this topic.[17-19] In the Netherlands, however, pediatricians were slow on the uptake. The first contribution about the 'transition problem' in a Dutch medical journal was written by an Australian pediatrician, Phelan (*Tijdschift voor Kindergeneeskunde*, 1993).[20] Almost ten years later (in 2002), the Dutch pediatricians Donckerwolcke and Van Zeben discussed the issue in the *Nederlands Tijdschrift voor Geneeskunde*.[21] Since then, only a handful of (expert opinion) articles in Dutch medical journals have followed.[7, 22-25] No general professional guidelines on transition or consensus statements have been published. A 2010 review of all available pediatric treatment guidelines (*n* = 25) in the Netherlands revealed that only six paid (some) attention to issues related to transition of care.[26]

The issue has also remained outside recent policy debates although it is beyond doubt that the rise in chronic conditions early in life already has a substantial impact on the demand for

health care and on public expenditure. Government policy papers and reports on the organization of care for the chronically ill in the Netherlands invariably focus on the adult or the ageing population. The same is true for the overview study *Zorg voor chronisch zieken* (Care for the chronically ill) published in 2011 by the Netherlands Institute for Health Services Research (Nivel).[27]

Now survival is no longer the major challenge, the focus should shift to optimal living with the chronic condition. We should realize, however, that young people with chronic conditions have specific needs, especially during their transition to adulthood.[28] Listening to their experiences and views may help to understand their concerns and daily struggles and open the way to a better future, with meaningful participation in society and the best possible quality of life.

We still know very little about the lived experiences of young people who grow up with chronic conditions. Adolescents' own perspectives on their health condition and health care have been underresearched. In the Netherlands, this information is almost entirely absent, which lack provided the rationale for the research program 'On Your Own Feet' described in this thesis.

In this Introduction chapter, I will sketch the general epidemiologic context of chronic conditions in childhood (prevalence, definitions) and discuss the consequences of growing up with chronic disease for society in general, for health care and for the involved individuals. I will specifically look at what is known about the epidemiology, social participation and health-related quality of life in adolescents and young adults with chronic conditions in the Netherlands. Finally I will describe the context, study population and overall aims of the 'On Your Own Feet' research projects.

2 The prevalence of chronic health conditions in childhood

Numbers of children and youth with chronic health conditions have increased considerably in the past decades, which was well documented in the United States.[29] In 1960, only 1.8% of all US children and adolescents were noted by their parents to have limited activity due to a health condition of more than 3 months duration; in 2004 this rate had risen to more than 7%.[29] However, comparing prevalences of chronic disease over time and across countries is complicated by diversity in concepts, definitions and operationalizations of chronic health conditions and disability in childhood.[30]

The dominant method for diagnosing and classifying a chronic condition in children has relied on the presence of an individual health condition of lengthy duration. Back in 1989, Stein and Jessop[31] tested the usefulness of this approach by examining correlates of illness in two large databases. They concluded that diagnosis is not a helpful categorization when examining the psychological and social variables related to chronic illness because there is more variability within diagnostic groupings than between them.[31] In view of the limitations

of a condition-specific or 'categorical' approach, a 'generic' approach may be recommended, focusing on elements shared by many conditions, children, and families.[32]

Such a definition encompasses the consequences of conditions and reflects the child's functional status or ongoing use of medical services. A typical example is the USA federal Maternal and Child Health Bureau definition of 'Children with Special Health Care Needs' (CSHCN): *"those who have or are at increased risk for a chronic physical, developmental, behavioral, or emotional condition and who also require health and related services of a type or amount beyond that required by children generally"*.[33, 34] According to the 2005-2006 National Survey of Children with Special Health Care Needs, which excluded the "at risk" component, approximately 10.2 million children aged 0–17 years in the United States (13.9%) had special health care needs. An even higher prevalence rate emerged from the 2007 National Survey of Children's Health: 19.2% of all American children aged 0-17 years had special health care needs (www.childhealthdata.org). The CSHCN Screener identifies children that currently experience health consequences due to a medical, behavioral, or other type of health condition that has lasted or is expected to last 12 months or longer.[35] Of those children, 91% suffered from one or more of the sixteen conditions listed in Table 1. One quarter even suffered from three or more of these conditions. In the above-mentioned 2007 National Survey of Children's Health, 42.9% of parents of children suffering from one or more conditions (Table 1), rated the condition(s) as moderate or severe, 31.3% as mild; the other children (25.8%) suffered from other conditions (www.childhealthdata.org).

In the Netherlands, Mokkink *et al* (2007) attempted to estimate the number of children and adolescents with a chronic disease and to evaluate the consequences of living with this.[36] In a consensus procedure they established that a disease or condition is considered to be a chronic condition in childhood if (1) the onset occurs between ages 0 and 18 years; (2) the diagnosis is based on medical scientific knowledge and established using reproducible and valid methods or instruments according to professional standards; (3) it is not (yet) curable; and (4) it has been present for longer than three months or will very probably last longer than three months, or it has occurred three times or more during the past year and will probably reoccur.[36] Using this definition and the International Statistical Classification of Diseases and Related Health Problems-10th revision (ICD-10), the authors established that 284 diagnoses were considered to be of chronic nature. Adding upon research findings and available prevalence rates, it was estimated that at least 14% of children in the Netherlands are growing up with a chronic condition; this implies at least 500.000 children and adolescents.

However, this estimation is definitely an underestimation because reliable information was available for only few diseases.[36] According to 2008 CBS (Statistics Netherlands) data, collected in the *Permanent Onderzoek Leefsituatie* 2001-2007 (www.cbs.nl), one in five Dutch children aged 4-12 years suffers from a chronic somatic disorder. Like in the US,[37] the most commonly reported chronic conditions in the Netherlands included asthma (10.0%), eczema (6.2%), frequent or severe headaches, and chronic bowel disorders. Less prevalent chronic conditions (< 1%) are congenital heart diseases, diabetes mellitus type 1, epilepsy, rheumatoid arthritis, neuromuscular disorders, cystic fibrosis, hemophilia, HIV, plus a large group of rare conditions. Cancer is not very common in childhood: about 0.1% of all children have ever suffered from neoplasms.

Despite differences in definition (categorical or generic) and in operationalization, the percentage of chronic conditions in childhood in the US compares well with that in the Netherlands. Stein & Silver compared different definitions of chronic conditions in a US national data set and found substantial overlap between several different conceptual definitions in the numbers and characteristics of the children identified.[38] Irrespective of these definition issues, the increased prevalence of chronic conditions in childhood bears considerable consequences: not only on the level of society and the entire health care system, but also for the individuals affected by them: the young people living with chronic disease and their families.

Table 1   *Sixteen most common chronic conditions in US-children with Special Health Care Needs[a]*

Chronic condition	% of CSHCN[b]
Allergies	53
Asthma	38
ADD/ADHD	30
Depression, anxiety, or emotional problems	21
Migraine / frequent headaches	15
Mental Retardation	11
Autism or Autism Spectrum Disorder	5
Joint problems	4
Seizure disorder	4
Heart problems	4
Blood problems	2
Diabetes Mellitus	2
Cerebral Palsy	2
Down Syndrome	1
Muscular Dystrophy	0.3
Cystic Fibrosis	0.3

[a] *2005-2006 National Survey of Children with Special Health Care Needs;*
   www.childhealthdata.org.
[b] *CSHCN stands for Children with Special Health Care Needs, ie, children that currently experience health consequences due to a medical, behavioral, or other type of health condition that has lasted or is expected to last 12 months or longer.*

3 Health care consequences of chronic health conditions in childhood

Increasing rates of childhood chronic conditions imply, subsequently, higher rates of these conditions during adulthood, which will have major impact on public and private health expenditures.[29] Not surprisingly, medical consumption in children with special health care needs is considerably higher than in healthy children: they consume much specialist care and are often admitted to a hospital emergency room (www.childhealthdata.org). Also, their parents reported more unmet needs for care and rated their health status more often as fair or poor than of children without special health care needs.

Young people with chronic conditions require lifelong treatment, follow-up, and adequate support, while they also face sequelae, long-term complications or impairments, co-morbidity, and in some instances even early death. They will transfer to the adult health care system at one time or another – in order to receive age- and developmental appropriate health care. However, the adult health care system seems poorly equipped to provide integrated (team) care for young people who present with complex, multifaceted health problems[39] and to ensure a long-term care perspective across the lifespan.[40, 41] Health care utilization of young adults is reported to decline in the transitional phase despite ongoing health problems. Dutch adults with cerebral palsy (aged 25-36 years) frequently reported pain (59%) and joint deformities (19-57%), but consumed less health care since they had turned 18. Only 32% visited their rehabilitation physician during the past year and 15% had seen a neurologist in this period.[26] An explanation for this decline may be that adult specialists may not be knowledgeable about or experienced in treating (the consequences of) specific childhood conditions.[39]

Studies on congenital heart disease[9, 10] and other congenital disorders, childhood cancer[5] and type 1 diabetes have made clear that structural follow-up needs to be improved, notably by preventing dropout. This means that a transition program must start at the beginning of the adolescent years and appropriate adult health care teams should be implemented.[8, 10, 42] For a study in young adults and older adolescents with moderate or complex CHD found that they expect to live almost as long as their healthy peers. They also had similar optimistic, but rather unrealistic expectations about their risks for complications.[43] There is an international challenge to incorporate a lifespan perspective in pediatric, transition, and adult health care services for persons with a childhood-onset disability.[41]

4 Social functioning for those growing up with childhood chronic health conditions

The label 'chronically ill' does by itself not provide much information about the medical, psychological and social consequences for the young people suffering from those conditions. Chronic conditions have widely varying impact on child or adolescent activity levels.[37] Within each diagnostic category large variability exists, suggesting that diagnosis is not a very helpful

categorization in the examination of these consequences.[31] For example, functional impairments differ greatly between children with spastic cerebral palsy. Those suffering from spastic quadriplegia (*ie,* all four limbs affected) are the least likely to be able to walk, in contrast to those with other forms. There is also a wide variation in the presence of secondary conditions, such as seizures, epilepsy, apraxia, communication disorders, eating problems, sensory impairments, intellectual and learning disabilities, urinary or fecal incontinence, and/or behavioral disorders.[31] This variability within diagnostic categories is a well known feature of chronic conditions in general, limiting the possibility to predict the functional outcome or the 'objective' impact of the condition. Nevertheless, many children with chronic conditions suffer one or more functional difficulties, as is demonstrated in Table 2, presenting data from the 2005-2006 US National Survey of Children with Special Health Care Needs.

Health status and functional limitations, however, do not tell us everything about the impact on social functioning. The majority of these children function quite well, but a substantial subgroup develops clinically significant psychological or social problems. Wellbeing is determined not only by health status but also by the emotional response to these problems. A chronic illness may interfere with their development and make children and adolescents more vulnerable to psychological and social problems.[44] A Dutch study into psychosocial adjustment of 84 adolescents aged 13-16 years with a chronic illness found that they showed more submissive

Table 2 *Functional difficulties in US-children with Special Health Care Needs[a]*

Functional difficulties	% of CSHCN[b]
Respiratory problems	43
Learning, understanding, or paying attention (ages 2–17 yrs old)	41
Feeling anxious or depressed	29
Behavior problems	28
Speaking, communicating or being understood	23
Making & keeping friends (ages 3–17 yrs old)	20
Chronic pain	18
Gross motor	14
Self care (ages 3-17 yrs old)	12
Fine motor	11
Swallowing, digesting food, or metabolism	10
Difficulty seeing even with glasses	4
Blood circulation	2
Uses a hearing aid	1

[a] 2005-2006 *National Survey of Children with Special Health Care Needs; www.childhealthdata.org.*
[b] CSHCN *stands for Children with Special Health Care Needs.*

behavior and tended to be more socially withdrawn, in particular when they use avoidant or passive coping strategies.[45] Overall, children with chronic conditions have a slightly elevated risk of psychosocial distress, although only a minority experience clinical depression.[46] A recent meta-analysis found a small negative effect across 57 studies indicating that chronically ill children had poorer social competence, (defined as the quality of social interactions with other children) than children without chronic illness.[47] Effects varied according to illness type: children with neurological disorders and obesity had the poorest social competence of all groups while children with asthma and diabetes did not suffer deficits in social competence and other illness groups had small effect sizes.[47]

The emotional responses from parents or peers also have an important effect. Compared to nonchronically ill adolescents, those with chronic conditions are more likely to be victims of bullying. They run a higher risk to be depressed, to have more physical symptoms, to have a poorer relationship with their parents, and to perceive a poorer school climate.[48] However, it is often difficult to determine if problems in psychosocial functioning are caused by the underlying illness, by treatment, or by the resultant effects of either illness or treatment on physical growth or cognitive development.[49] Assessment of wellbeing and mental health should therefore be an integral component of the comprehensive care of chronically ill children and adolescents,[50-51] as well as prevention and intervention programs to increase their social skills and social support.[45, 47]

4.1   Definition of quality of life

The concept of wellbeing is closely related to the concept of the quality of life. The World Health Organization (WHO) defines quality of life as *"individuals' perceptions of their position in life, in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards and concerns"* (www.who.int). The concept of Health Related Quality of Life (HRQoL) refers to the impact of health and illness on an individual's quality of life. HRQoL is a multidimensional construct incorporating at least three broad domains: physical, psychological (emotional, mental) and social / behavioral components of wellbeing and functioning as perceived by patients and/or other individuals. In the case of young people with chronic illness, adult-focused definitions should be extended to take developmental changes and young people's construction of health and illness into consideration.[52] Definitions therefore should include goals and aspirations as well as the constraints imposed through ill-health and treatment.[52] HRQoL is usually measured through generic or disease-specific questionnaires assessing dimensions such as physical and psychological wellbeing, family life, school performance and peer relations.[53]

4.2   Definition of social participation

Adolescents' transition to adulthood is critical to autonomous social participation in adult life.[41] A chronic illness may complicate this transition. Puberty, autonomy, personal identity, sexuality, education, and vocational choices all may be influenced by impaired physical or mental abilities, pain, medical setbacks, forced dependence, and perceived prognosis.[16]

The WHO's International Classification of Functioning (ICF, 2001) offers a model of human functioning which shifts the focus onto the social environment and the contextual conditions which impact on functioning. In the ICF, the term *functioning* refers to all body functions, activities and participation, while *disability* is similarly an umbrella term for impairments, activity limitations and participation restrictions. The ICF defines *activity* as the execution of a task or action by an individual; *participation* is defined as involvement in a life situation. The ICF discerns four major participation domains:

1   Domestic life (involvement in the running of your home);
2   Interpersonal life (informal social relationships, family relationships, intimate relationships, formal relationships);
3   Major life activities (education, training, employment, voluntary work);
4   Community, civic & social life (recreation & leisure, sports, arts & culture, religious practice, socializing) (www.who.int/classifications/icf/en/).

The ICF definition of participation has been criticized for ignoring the subjective experience of meaning, the client's autonomy, and the environmental influences on participation. In response, Hemmingsson and Jonsson proposed an alternative definition of participation: *"the subjective experience of meaning in connection to participation in a life situation and the subjective experience of autonomy and self-determination"*.[54]

Limitation in activities and restrictions in social participation have been reported for children with diverse chronic conditions. The 2005-2006 National Survey of Children with Special Health Care Needs asked parents whether medical, behavioral or other health conditions interfered with their child's ability to attend school regularly, participate in organized sports or clubs, or make friends. 68.7% of parents reported no limits on participation, 17.2% one limitation in, and 14.2% two or more limitations (www.childhealthdata.org). The next paragraph will describe what is known about these issues in the Netherlands.

5   Social participation and wellbeing of Dutch adolescents with chronic conditions

Already twenty years ago, the Dutch psychologist Sinnema pointed to the challenge to support young people with chronic conditions in their quest for economic and social participation.[55] Although on the rise since 2000, studies exploring the consequences of growing up with a chronic disease for children, adolescents, young adults, their parents, and siblings in the Netherlands are still scarce.[56] The available evidence indicates that having a chronic disease often negatively affects one's social participation and health-related quality of life. The overview below only presents studies comparing chronically ill with 'healthy' peers. Both across and within diagnostic categories we find large differences. However, as study samples tended to be small, and different instruments were used, the findings presented here should be interpreted with some caution.

5.1   Social participation

Lower educational outcomes and impaired cognitive functioning were reported for young people with spina bifida,[57, 58] and also for young adults with end-stage renal disease.[59] Compared with age-matched Dutch citizens, young adults with childhood end-stage renal disease were more often involuntarily unemployed, had a lower occupational level, more often still lived with their parents, and more often had no partner.[60]

Quite many adolescents and young adults with cerebral palsy without severe learning disabilities are restricted in daily activities (mobility, self-care, nutrition) and social participation (taking responsibility, community living, leisure activities, and employment). These problems are mainly attributable to restricted gross motor functioning, low level of education and younger age.[61] For young people with disabilities such as spina bifida or cerebral palsy, participating in social activities and intimate relationships, and experiencing a fulfilling sexual life is often very challenging.[62, 63] At least 60% of a sample of adults with spastic bilateral cerebral palsy had difficulties with mobility, recreation and housing, and 44% had difficulty with personal care and employment.[64] A review of rehabilitation-based samples of (young) adults with childhood-onset conditions summarized functioning, quality of life, and lifespan care issues.[41] It shows that many young adults with a childhood-onset disability experience health-related problems such as functional deterioration, pain or fatigue, and lead an inactive life. A considerable number are restricted in participation in work, housing, and intimate relationships – as reported for men with Duchenne Muscular Dystrophy in Box 2.

Box 2   *Becoming a "futureless" adult with Duchenne Muscular Dystrophy*

When there was not yet treatment available, the life expectancy of boys with Duchenne Muscular Dystrophy (DMD) was 15-20 years. Owing to the introduction of chronic mechanical ventilation a new generation of young men with DMD may live up to 30-40 years. Despite this and other medical advances, many young men with DMD struggle to lead meaningful lives once they have finished full-time education. A Danish study described the 'ordinary adult man with DMD' as follows: "He states his quality of life as excellent; he is worried neither about his disease nor about the future. His assessment of income, hours of personal assistance, housing, years spent in school and ability to participate in desired activities are positive. Despite heavy immobilization, he is still capable of functioning in a variety of activities that are associated with normal life. He lacks qualifying education and he is in painful need of a love life. The frequency of pains is surprisingly high; nearly 40% has pains daily".[65] A qualitative study labelled these men as "futureless": social and symbolic isolation was also a temporal isolation whereby the men had lived every day anticipating that it could be their last for as much as a decade.[66] In a recent study among 40 young men with DMD and their families in the UK almost all families interviewed described their overall experience of service provision as problematic; 80% of parents reported clinical levels of anxiety and depression, most young men with DMD had very limited social opportunities or friends beyond their immediate family, and a third of young men were at home during the day and not doing any kind of education, training or work. Young men and their parents said that they tended to 'live for the day', largely because the future was so uncertain.[67]

In the Netherlands, adult men with DMD mostly live with their parents and their social participation is limited. Their desire for meaningful activities is shared by men who live in institutions: a qualitative study involving six mechanical ventilation-dependent adult men with neuromuscular

disorders including DMD living in 'Het Dorp' (an assisted living institution), established that they wished to be taken seriously in all aspects of life, including decisions concerning risk and end-of-life issues. They also wanted meaningful activities and "be treated as an employee" rather than being just a client.[68] Social participation, and adequate service provision are major challenges for this neglected generation of young men and their families.

Having a chronic digestive disease, in particular chronic liver disease and inflammatory bowel disease, negatively affected adolescents' social position: problems occurred in education, leisure activities, labor participation, financial situation, partnership and sexuality.[69] Adults with hemophilia were less involved in full-time paid work and suffered more from occupational disability than men from the general population, despite advances in treatment.[70] In contrast, in a large sample of Dutch young adults with type 1 diabetes, no negative effects on employment careers were established. Their chances of entry into the labor market were not decreased and there was no higher unemployment in the study population than in the general population.[71]

Recent Dutch studies found that young adults aged 18-30 years with chronic conditions achieved significantly fewer milestones, or reached these at older age, than did their healthy peers, in all course-of-life-domains.[72] Type of condition made a difference, however. Young adults with esophageal atresia did not differ from their peers in this respect, and course of life was delayed most in survivors of childhood cancer and patients with end-stage renal disease.[72] A recent study assessing the achievement of psychosocial milestones in course of life of young adult disability beneficiaries with somatic limitations (22-31 years, $n = 415$) confirmed substantial negative outcomes compared to healthy peers from the general Dutch population. They achieved fewer milestones or at a later age than peers.[73]

Bullying, too, affects Dutch children with chronic conditions. The Netherlands Handicapped Child Foundation (Stinafo) indicated that children with disabilities who attend regular school are five times more likely to be bullied than their nondisabled peers (www.stinafo.nl). In young people with galactosemia, an inherited metabolic disorder, special school attendance was higher than that of the general population, and their educational attainment was lower.[74] Also, their social and psychosexual development was delayed; they were significantly less frequently married or living together and significantly less frequently employed than the general population.[75]

5.2   Health-Related Quality of Life (HRQoL)

To provide insight into how growing up with a chronic condition may affect one's HRQoL, I present findings from recent studies in the Netherlands. In a large study that collected reference data on the Pediatric Quality of Life Inventory (PedsQL) in almost 500 Dutch children, HRQoL of chronically ill children was significantly lower in the youngest (5-7 years) and in the adolescent (13-18 years) age groups.[76]

Adolescents with inflammatory bowel disease (IBD) had lower HRQoL on four domains (body complaints, motor functioning, autonomy and negative emotions) than had healthy peers.[77] These adolescents were worse off than younger children with IBD. Another study confirmed this and showed that adolescents with IBD, especially boys, had more internalizing

problem behavior compared with healthy peers. Self-esteem was an important predictor of their HRQoL.[78] Adolescents with galatosemia had significantly lower scores on the domains of cognitive and social function.[74] HRQoL of patients with severe hemophilia between ages 31 and 64 years was lower than that of the general population; however, the difference in HRQoL with the general population was less pronounced for patients between ages 16 and 30 years.[70] HRQoL of children with another bleeding disorder, Von Willebrand disease, was lower than that in reference populations, particularly in school age children. Apart from physical scales, emotional and social scales were affected.[79]

In contrast, other studies do not confirm that HRQoL is always impaired in internal diseases. Young women with Turner Syndrome (TS) who reached normal height and had age-appropriate pubertal development after long-term hormone therapy reported normal HRQoL. They even reported higher HRQoL scores on some of the scales, including social functioning and role-emotional.[80] Also, a study among children and adolescents with juvenile rheumatoid arthritis found that they coped quite well with the psychological and social consequences of their long term condition.[81] A sample of adolescents with type 1 diabetes reported optimal wellbeing, although glycemic control often was poor, which was related to lower social and family functioning.[82]

Survivors of cancer treatment are reported to adjust well to the cancer experience and HRQoL improved with time. Shortly after the end of successful cancer treatment, physical well-being was affected for all ages, but 12- to 18-year-olds had significantly better HRQOL than the norm on social scales. Compared to chronically ill norms, 8- to 18-year-olds demonstrated no differences, except for adolescents who experienced significantly more physical limitations.[83] In another study, survivors of retinoblastoma treatment even reported a very good HRQoL compared with the Dutch reference group.[84]

In rehabilitation samples, HRQoL is often negatively affected.[41] Young adults with meningomyelocele (*spina bifida manifesta*) perceived lower physical HRQoL than a Dutch reference population. 63% had difficulties in daily activities and 59% in social roles. Higher levels of physical activity and fitness were related to fewer difficulties in participation and higher physical HRQoL.[85] Compared to healthy controls, children and adults with Muscular Dystrophy had lower HRQoL scores.[86] Adults with bilateral cerebral palsy perceived low HRQoL for physical, but not for mental functions.[64]

Conflicting findings are reported for HRQoL in those who were surgically treated for serious congenital malformations. A study of adults treated for congenital anorectal malformations and congenital hernia diafragmatica showed little difference with the general population. Even though there is considerable morbidity and mortality in childhood, the ultimate prognosis of survivors of those birth defects is favorable.[87] In another study, the HRQoL of children and adolescents with congenital anorectal malformations or Hirschsprung's disease was also comparable to that of healthy peers, even though both patient groups encountered overall physical health problems. Only patients with congenital anorectal malformations reported pain and limitations in role functioning due to physical problems.[88] The HRQoL of most adult patients with a congenital heart disease (CHD) was good and comparable with that of the standard Dutch population,[7] but HRQoL of a sample of recently treated CHD children and adolescents was lower than that of reference groups. CHD patients (aged 8-15) on average scored signifi-

cantly lower on motor functioning, cognitive functioning, and positive emotional functioning than did reference peers.[89]

We conclude that HRQoL is often affected in adolescents with chronic conditions in the Netherlands, but not always, nor in all domains. In general, we hardly know whether factors such as medical determinants, personality or family characteristics could predict functioning and HRQoL later in life. The importance of such factors was demonstrated in children with sickle cell disease.[90] Their HRQoL was lower than that of the Dutch norm population on five domains (physical wellbeing, moods and emotions, autonomy, parent relation, and financial resources), but was comparable to that of their healthy siblings. Apart from the disease-specific effects on the physical and autonomy domain, the lower HRQoL was mainly related to (low) socio-economic status.[90]

Delayed development and low social participation also contribute to impaired HRQoL. For example, those young adults with end-stage renal disease who achieved fewer milestones than peers with respect to autonomy, social, and psycho-sexual development, also experienced more emotional problems and less vitality, and had a lesser overall mental quality of life.[91] The interplay between psychological factors, participation and HRQoL was demonstrated in a study of adults with bilateral spastic cerebral palsy: higher general self-efficacy or a greater willingness to expend effort in achieving behavior was related to better participation and a higher physical and mental health-related quality of life.[64]

Finally, having a child with a chronic condition may also have serious consequences for family functioning. Dutch parents of chronically ill children in 10 diagnosis groups had a significantly lower HRQoL compared to parents of healthy children.[92] Subgroup analysis showed lower health-related quality of life on sleep, social functioning, daily activities, vitality, positive emotions, and depressive emotions in disease-specific groups. On average, 45% of the parents were at risk for HRQoL impairment.[92]

All in all, the (often hidden) psychosocial consequences of the pediatric success story deserve attention. Not only for the young people themselves, but also for their families. But is measuring quality of life enough to gain understanding of these consequences?

6   The limitations of HRQoL to capture the experience of living with chronic illness

Measuring HRQoL is aimed at capturing the patient's subjective evaluation of health and illness. However, we should be challenged by the lack of consensus on the conceptualization of child health, illness, wellbeing and quality of life, and the fact that we do not know well how these outcomes are expressed among children at different developmental levels.[93] For example, the current WHO definition of health, formulated in 1948, describes health as *"a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity"* (www.who.int). This definition of 'health as a state' is no longer fit for purpose given the rise of chronic disease. Therefore, a conference of international health experts held in the

Netherlands in 2009 preferred that this definition should be replaced by a dynamic conceptual framework of health: *"health as the ability to adapt and to self manage"*.[94]

Quantitative HRQoL measurements may be criticized for their inability to grasp this dynamic process of experiencing health in the context of having a chronic condition. Qualitative studies may reveal another view on how chronic conditions affect daily life or influence the life course, and on the meanings attached. To give an example, although fecal incontinence did not affect their HRQoL,[88] adults with congenital anorectal malformations retrospectively, during in-depth interviews, often labeled puberty as *"a dark period"* because the unpleasant consequences of fecal incontinence landed them in social isolation. They responded with grotesque behavior, such as playing the clown or by withdrawing from peer activities.[95]

In the seventies, sociologists Glaser and Strauss[96] turned attention to how people manage to live as normal a life as possible in the face of disease. Managing chronic illness needs to be understood in the context of people's lives, but also in the context of the disease course (*the illness trajectory*).[97] Corbin and Strauss described chronic illness as continuous, life-long work, not only by the persons suffering from the chronic illness, but also by those around them.[97] Other sociologists have centered on the importance of restructuring meaning during illness, by describing the onset of the disease as an acute crisis or a biographical disruption.[98]

The progression into a chronic illness or disability is a turning point in an individual's life, or in other words – a transition. In this context, transition is often cited as: *"a passage from one life phase, condition, or status to another that disrupts normal life and demands for adaptation".*[99] The concept of transition refers to both the process and the outcome of complex person-environment interactions.[100] The views on change people have, their expectations of events, level of knowledge and skill, availability of new knowledge about a change event, the resources available in the environment, capacity to plan for change, and emotional and physical wellbeing; all these conditions have an impact on transitions. A successful transition is one where feelings of distress are replaced with a sense of wellbeing and mastery of a change event.[99] Health care providers are challenged to provide support to people in transition.[99, 100]

While a 'normal' HRQoL may indicate a successful transition, this outcome does not properly reflect the amount of daily work needed to cope with the stressors and adaptive tasks that the chronic condition continuously imposes on individuals and families. To fully appreciate what it means to grow up with chronic illness, we would have to grasp the 'lived experiences' of the young people themselves. To this aim it would be important to understand the processes by which they adjust to illness and, in many cases, find positive meaning in their experiences. An important concept in this respect is *resilience*, the ability to bounce back from set backs.[101] Many young people with chronic illness appear resilient in the face of the adjustment challenges presented by their illness.

Still, little is known about how young people with chronic conditions actually perceive (the impact of the condition on) their lives and development. Moreover, there is a lack of studies relating to the way young people with chronic conditions regard their daily lives and factors that are important for their wellbeing.[102] In 2008, Taylor identified no more than 22 (often small) qualitative studies covering many different conditions.[103] Seven common themes were found:

developing and maintaining friendships; being normal/getting on with life; the importance of family; attitude to treatment; experiences of school; relationship with health care professionals; and the future. As there was considerable commonality in themes across conditions, the authors conclude that the burden of chronic illness during adolescence is not necessarily disease-specific.[103] There are indications that adolescents with long-term illness generally experience wellbeing like everybody else. While illustrating the difficulties of managing a chronic medical condition during adolescence, an Australian study concluded that a generally positive message emerges about these young people.[104] A Swedish study found three factors needed to feel good: 'a feeling of acceptance of illness/disability as a natural part of life', 'a feeling of support' and 'a feeling of personal growth'.[102] This demonstrates that young people use personal and social resources in managing acute and chronic stressors, but also that life crises and transitions may even enhance their development and maturation.[99, 105] Indeed, the challenge of adjusting to a chronic illness can provide an excellent opportunity to master crucial skills, such as emotion regulation and problem-solving, engendering strong self-esteem and confidence.[106]

Different health problems encompass many comparable adaptive tasks, such as managing physical symptoms, accepting one's illness, giving up ordinary activities, adapting to an altered social identity, and finding new ways to maintain social relationships.[107, 108] These adaptive tasks can be divided into two groups: three related to the health condition and its treatment (managing symptoms; managing treatment; forming relationships with health care providers); the other four more general and applying to all types of life-crises (managing emotions; maintaining a positive self-image; relating to family members and friends; and preparing for an uncertain future).[108] These seven adaptive tasks are very similar to the seven themes described above in chronically ill adolescents. Typically, these tasks and themes are encountered with all chronic conditions, but their relative importance varies, depending on the person, the specific health problem, and the unique set of circumstances.

We embarked on the research described in this thesis since there are no Dutch studies exploring the lived experiences and the responses to the adaptive tasks of adolescents with a variety of chronic conditions, and their preferences and their developmental competencies for health care are largely unknown.

7   Context and study population

This thesis presents the results of four research projects in a program named 'On Your Own Feet', initiated by Rotterdam University, university of applied sciences, and carried out together with Erasmus University Medical Center – Sophia Children's Hospital and Erasmus University Rotterdam. The program was initiated to increase our knowledge about young people's experiences with chronic conditions in order to provide adequate, age-appropriate services for them and support them in their transition to adulthood and adult care.

The program (presented in Figure 1) took a generic, noncategorical approach, implying that all projects were aimed at youth with somatic chronic conditions irrespective of their medical diagnosis. This approach seemed most appropriate as we did not focus on medical

and treatment-related issues, but on the experiences and viewpoints of the young people themselves. A generic approach may help to appreciate the similarities between specific diseases and groups of disorders.[109] In one project ('Experiences with transition'; Figure 1, #3), seven specific diagnostic groups were targeted because this allowed us to compare transitional care practices in different pediatric subspecialties. All projects but one focused on grasping young people's voices to reveal their views on living with a chronic condition. The only exception, the 'On Your Own Feet Ahead' study (Figure 1, #4), wished to map the current practices of health care professionals involved in adolescent health care and identify their priorities for improvement.

The 'On Your Own Feet' studies were conducted in Erasmus University Medical Center, Sophia Children's Hospital. This is the largest university pediatric hospital in the Netherlands and is made up of the departments of Pediatrics, Pediatric Surgery, and Child and Youth Psychiatry and Psychology. The department of Pediatrics and Pediatric Surgery were involved in the studies. The department of Pediatrics offers tertiary specialized pediatric care (in 8 subspecialties), (neonatal) intensive care, general pediatrics, radiology, and laboratory services. The hospital has no specialized adolescent in-patient facilities and in 2004, when the research started, offered only a handful of outpatient youth / transition clinics. Only the Hemophilia and IBD subspecialty departments had recently set up transition clinics. Not all subspecialty departments employ nurse specialists or work with multidisciplinary teams, but all have (specialized) pediatricians. Yearly, the Sophia Children's Hospital treats thousands of children, from birth up to the age of 18. In the Netherlands, this is the general age for transfer to adult care, although older children may still attend pediatric outpatient clinics.

Some 4000 adolescents (aged 12-19) formed the study population for the qualitative and quantitative 'On Your Own Feet' studies (Figure 1, #1 and 2). The criterion for being a chronic patient was having had checkups in the hospital on a regular basis for the past three years. In the 'Experiences with transition' study (Figure 1, #3), we invited young adults who had transferred from seven departments in the Sophia Children's Hospital to adult care in the past two years, and interviewed their parents and health care professionals as well. In the 'On Your Own Feet Ahead' study, we invited a national sample of teams of health care providers that work with young people with somatic chronic conditions.

Figure 1 *Overview of studies and participants in the 'On Your Own Feet' program*

1) Qualitative research 'On Your Own Feet'(2004-2006)	**A**	Semi-structured *interviews* with youth with chronic conditions (*n* = 31; 12-19 years)	**Chapters 2, 5**
	B	*Q-methodology* with same interviewees (*n* = 31; 12-19 years)	**Chapters 3, 4**
	C	*Peer-research* with 9 co-researchers (15-17 yrs) and interviewees (*n* = 25; 12-19 years)	**Chapters 1, 2**
	D	*Observations* of medical and nursing consultations in 8 outpatient clinics (*n* = 39; 12-22 years)	**Chapter 5**
	E	*Focus group discussions* (*n*=3) with 27 health care providers	**Chapter 5**
2) Quantitative research 'On Your Own Feet' (2006-2007)		*Web-based questionnaire* (*n* = 1087 adolescents (12-19 years); *n* = 830 parents; 593 pairs of adolescents and parents)	**Chapters 2, 4, 5, 6, 7, 8**
3) Qualitative Research 'Experiences with transition' (2004-2007)		*Semistructured interviews* with young adults with 7 different chronic conditions (*n* = 24; 15-22 years); their parents (*n* = 24); and 17 health care providers (pediatric and adult care)	**Chapter 9**
4) Quality Improvement Collaborative 'On Your Own Feet Ahead' (2008-2012)		*Survey* (*n* = 115) teams of health care providers treating adolescents with somatic chronic conditions	**Chapter 10**

8   Overall aims and research questions

Four major aims were formulated for the research projects. Three are related to the overall aim of the 'On Your Own Feet' research program: *to map preferences and competencies for health care of adolescents with chronic conditions in their transition to adulthood and adult care*. The fourth aim is concerned with contributing to the knowledge base on methodological issues.

Aim I

To investigate the preferences for (participation in) health care and self-management of adolescents with chronic conditions, and determine the associations between profiles for self-management and health care, socio-demographic, and disease-related variables.

Related research questions

Ia.   What are the preferences of chronically ill adolescents for health care professionals, outpatient and inpatient service delivery, and what improvements are recommended for adolescent health care provision in Erasmus MC – Sophia? (Chapter 2)
Ib.   Which profiles for self-management and preferences for care (Q-Care Profiles) can be elicited in adolescents with chronic conditions? (Chapter 3)
Ic. What is the distribution of the Q-Care Profiles in adolescents with chronic conditions and what are the associations between fit to the Q-Care Profiles and relevant variables? (Chapter 4)

Preferences for care reflect what patients consider the most important elements of particular health care services and are also referred to as patients' desires and expectations of receiving health care.[110] We use the most common definition of self-management: *"the ability to manage one or more chronic conditions (symptoms, treatment, physical and psychosocial consequences, and life style changes) and integrate them in day-to-day life with the aim of achieving optimal quality of life".*[111]

> *Aim II*
>
> To study the competencies for self-management, transfer readiness, and the self-efficacy of adolescents with chronic conditions and determine the extent to which these competencies can be observed during hospital consultations.

Related research questions

IIa. *What are the perceived and observed competencies for hospital consultations of adolescents with chronic conditions?* (Chapter 5)
IIb. *What constitutes adolescents' readiness to transfer to adult care?* (Chapter 6)
IIc. *What are the essential, generic tasks for self-management and how can self-efficacy for these be measured in adolescents with chronic conditions?* (Chapter 7)
IId. *What is the extent and direction of agreement between Health-Related Quality of Life adolescent self-reports and proxy reports of their parents and which factors are associated with child-parent agreement?* (Chapter 8)

Successful self-management encompasses three major tasks for the person with a chronic illness: managing the medical aspects of the illness, changing social roles, and dealing with the psychological consequences.[97] These tasks demand skills such as problem solving, decision-making, resource utilization, forming partnerships with healthcare providers, and taking action.[112] Self-efficacy refers to *"the beliefs in one's capabilities to organize and execute the courses of action required to produce given attainments"*.[113] This concept lies at the center of Albert Bandura's social cognitive theory; most prominent health behavior theories include this concept. One's sense of self-efficacy can play a major role in how one approaches goals, tasks, and challenges, and in that sense self-efficacy is an important predictor for self-management. Self-efficacy can be characterized mainly as being competence-based, prospective, and action-related.[113] The concept of competency is used to describe the knowledge, attitude, and skills required to successfully perform tasks or to approach new challenges.

Aim III

To explore the experiences of young adults, parents, and health care providers with the transition from pediatric to adult health care services and to collect their recommendations and priorities for improving transitional care.

Related research questions

IIIa. What are the experiences with the transfer to adult care of young adults with chronic conditions, their parents, and health care providers and what are their recommendations for improvement of transitional care? (Chapter 9)

IIIb. What are the current practices regarding transitional care and which priorities for change are identified by teams of health care providers? (Chapter 10)

The most widely cited definition of 'transition of care' in the adolescent health literature is that of Blum *et al*: *"the purposeful, planned movement of adolescents and young adults with chronic physical and medical conditions from child-centered to adult-oriented health care systems"*.[16] Transition of care is further characterized as *"a multi-faceted, active process that attends to the medical, psychosocial and educational / vocational needs of adolescents as they move from child- to adult centered care"*.[16] It implies an increase in independent behaviors and autonomy in adolescents. The goal of transitional care is: *"to maximize lifelong functioning and potential through the provision of high quality, developmentally appropriate health care services that continue uninterrupted as the individual moves from adolescence to adulthood"* – with the development of successful self-management in young people with chronic conditions as the ultimate aim.[114] Transfer (the actual move from pediatric to adult services) is merely an event within the total process of transition.

Related research questions

IVa. What are the feasibility, the advantages, and the limitations of collaborating with chronically ill adolescents in participatory research? (Chapter 1)

IVb. What does Q-methodology add to our understanding of adolescents' preferences related to self-management and health care? (Chapter 3)

IVc. What is the added value of a Mixed Methods approach integrating qualitative and quantitative data in studying adolescents' preferences and competencies for care? (Chapter 2, 5)

IVd. How can a new generic self-efficacy instrument for adolescents with chronic conditions be developed and validated? (Chapter 7)

Mixed methods research is defined as a single study in which qualitative data collection and/ or analysis is combined with quantitative data collection and/or analysis either in a concurrent or sequential design.[115] Q-methodology can be seen as a methodology combining qualitative and quantitative approaches in itself.[116] Q-methodology provides a scientific foundation for the systematic study of subjectivity, such as people's opinions, attitudes and preferences.[117, 118] Individuals represent their viewpoints by ranking a set of statements, after which by-person factor analysis reduces these many viewpoints to a few shared perspectives. Participatory research is a collaborative undertaking aimed at more involvement of the persons being studied in all aspects of the research process. It is carried out *with* and *by* the research subjects rather than *on* them.[119, 120]

Self-efficacy is an important variable associated with various outcomes such as self-management, HRQoL, etc. The measurement of self-efficacy requires precisely developed and validated instruments but for chronically ill adolescents only disease-specific instruments are available. [121]

9   Outline of the thesis

Part I Preferences for care

The thesis opens with four chapters related to the study of chronically ill adolescents' preferences for health care and attitudes toward self-management. **Chapter 1** presents a critical exploration of the value and the pitfalls of the participatory approach in research with adolescents. It concerns a peer-research project where nine adolescent co-researchers helped investigate their fellow patients' evaluation of the quality of care in the Erasmus MC – Sophia. In **Chapter 2**, the results of this peer-research study are combined with qualitative interviews on adolescents' preferences for providers and health care; and are compared with quantitative data gathered in a questionnaire survey. We discuss adolescents' suggestions for the improvement of adolescent care in the Erasmus MC – Sophia.

The next two chapters deal with the development, distribution and validation of the Q-Care Profiles. These profiles for self-management and health care preferences, representing distinct self-management styles, were uncovered with Q-methodology during the interview study (**Chapter 3**). Distribution of the profiles and associations with socio-demographic and disease-related variables are further explored in the questionnaire survey (**Chapter 4**).

Part II Competencies for care

Adolescents' competencies for health care are investigated in the second part of the thesis comprising four chapters. In **Chapter 5,** we turn attention toward clinical encounters in the outpatient department. Adolescents' preferences and competencies for health communication during hospital consultations are presented based on the integration of data from three different substudies, hereby also exploring the added value of mixed methods research itself. The issue whether adolescents consider themselves to be ready for the transfer to adult care is raised in **Chapter 6**, exploring the predictors of transfer readiness collected in the questionnaire.

Chapter 7 describes the development and the validation of a new generic instrument measuring self-efficacy in coping with chronic condition, in knowledge about the condition, and in managing independent hospital visits. In **Chapter 8**, we compare adolescent self-reports with proxy (parent) reports of HRQoL, in order to resolve whether we need parent proxy reports to explore chronically ill adolescents' quality of life.

Part III Transitional care: the need for action

The third and final part of this thesis deals with transitional care. **Chapter 9** reports on a qualitative study comparing the experiences of young adults with seven different chronic conditions, their parents and their (ex) health care providers with the recent transition of care. Their recommendations to improve preparation for transition and the delivery of adolescent health care inspired us to move from research to action. We therefore set up a Quality Improvement Collaborative called *'On Your Own Feet Ahead!'*. **Chapter 10** presents data from a national

survey among 115 teams of health care providers in hospitals. The study identifies current transitional care practices in Dutch adolescent chronic care and presents professionals' priorities for change.

Finally, the **Conclusion and Discussion** section reviews the results of the studies in this thesis, comparing them with the current state-of-the-art knowledge and discussing future research and clinical implications. The thesis concludes with a summary of the findings.

References

1. Okumura MJ. Growing up and getting old(er) with childhood-onset chronic diseases: paving the way to better chronic illness care worldwide. *J Adolesc Health.* 2009;45(6):541-542.

2. van der Veen WJ. De kleine epidemiologische transitie: verdere daling van de zuigelingensterfte door medische interventie rond zwangerschap en bevalling, maar geen daling van handicaps op de kinderleeftijd. [The small epidemiologic transition: further decrease in infant mortality due to medical intervention during pregnancy and childbirth, yet no decrease in childhood disabilities]. *Ned Tijdschr Geneeskd.* 2003;147(9):378-381.

3. Slieker MG, Uiterwaal CS, Sinaasappel M, Heijerman HG, van der Laag J, van der Ent CK. Birth prevalence and survival in cystic fibrosis: a national cohort study in the Netherlands. *Chest.* 2005;128(4):2309-2315.

4. Freyer DR, Kibrick-Lazear R. In sickness and in health: transition of cancer-related care for older adolescents and young adults. *Cancer.* 2006;107(7 Suppl):1702-1709.

5. Blaauwbroek R, Groenier KH, Kamps WA, Meyboom-de Jong B, Postma A. Late effects in adult survivors of childhood cancer: the need for life-long follow-up. *Ann Oncol.* 2007;18(11):1898-1902.

6. van der Bom T, Zomer AC, Zwinderman AH, Meijboom FJ, Bouma BJ, Mulder BJ. The changing epidemiology of congenital heart disease. *Nat Rev Cardiol.* 2011;8(1):50-60.

7. Meijboom FJ, Roos-Hesselink JW, Bogers AJJC. Aangeboren hartafwijkingen op volwassen leeftijd. [Congenital heart disorders in adulthood]. *Tijdschr Kindergeneeskd.* 2006;74 (Dec):236-241.

8. Wren C, O'Sullivan JJ. Survival with congenital heart disease and need for follow up in adult life. *Heart.* 2001;85(4):438-443.

9. Meijboom F, Mulder B. Problems in the organization of care for patients with adult congenital heart disease. *Arch Cardiovasc Dis.* 2010;103(6-7):411-415.

10. Moons P, Meijboom FJ. Healthcare provision for adults with congenital heart disease in Europe: a review. *Curr Opin Pediatr.* 2010;22(5):573-578.

11. Strauss DJ, Shavelle RM, Anderson TW. Life expectancy of children with cerebral palsy. *Pediatr Neurol.* 1998;18(2):143-149.

12. Strauss D, Brooks J, Rosenbloom L, Shavelle R. Life expectancy in cerebral palsy: an update. *Dev Med Child Neurol.* 2008;50(7):487-493.

13. Hilberink SR, Roebroeck ME, Nieuwstraten W, Jalink L, Verheijden JM, Stam HJ. Health issues in young adults with cerebral palsy: towards a life-span perspective. *J Rehabil Med.* 2007;39(8):605-611.

14. Nieuwenhuijsen C, Donkervoort M, Nieuwstraten W, Stam HJ, Roebroeck ME. Experienced problems of young adults with cerebral palsy: targets for rehabilitation care. *Arch Phys Med Rehabil.* 2009;90(11):1891-1897.

15. Blum RW. Transition to adult health care: setting the stage. *J Adolesc Health.* 1995;17(1):3-5.

16. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, *et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 1993;14(7):570-576.

17. American Academy of Pediatrics. Transition of care provided for adolescents with special health care needs. American Academy of Pediatrics Committee on Children with Disabilities and Committee on Adolescence. *Pediatrics.* 1996;98(6 Pt 1):1203-1206.

18. American Academy of Pediatrics. The role of the pediatrician in transitioning children and adolescents with developmental disabilities and chronic illnesses from school to work or college. American Academy of Pediatrics Committee on Children With Disabilities. *Pediatrics.* 2000;106(4):854-856.

19. American Academy of Pediatrics, American Academy of Family Physicians, American College of Physicians, Transitions Clinical Reporting Group. Supporting the health care transition from adolescence to adulthood in the medical home. *Pediatrics.* 2011;128(1):182-200.

20. Phelan PD. The chronically ill child as an adult. *Tijdschr Kindergeneeskd.* 1993;61 Suppl:22-26.

21. Donckerwolcke RAMG, van Zeben-van der Aa DMCB. Overdracht van de zorg voor adolescenten met chronische ziekten: van kindergeneeskunde naar specialismen voor volwassenen [Transfer of care for adolescents with chronic illnesses: from pediatrics to specialists for adults]. *Ned Tijdschr Geneeskd.* 2002;146(14):675-678.

22. Maassen H. Een soepele overgang. Van kinderarts naar 'gewone' dokter. [A smooth transition. From pediatrician to 'ordinary' doctor]. *Medisch Contact.* 2005;60(17):700-703.

23. Escher JC, van der Woude CJ. Transitie van de adolescent met een chronische inflammatoire darmziekte. [Transition of the adolescent with a chronic inflammatory bowel disease]. *Tijdschr Kindergeneeskd.* 2005;73(Feb):36-41.

24. van der Ent CK, Drubbel I, de Vrankrijker AMM, Arets HGM, Heijerman HGM. Cystic fibrosis; een vergrijzend ziektebeeld? [Cystic fibrosis: an ageing disorder?]. *Tijdschr Kindergeneeskd.* 2006;74(Dec):242-248.

25. de Muinck Keizer-Schrama SMPF. Het belang van zorgvuldige transitie van zorg voor patienten met het Syndroom van Turner. [The importance of a careful transition of care for patients with Turner's Syndrome]. *Ned Tijdschr Geneeskd.* 2007;151:1612-1615.

26. van den Berg M, van Staa AL, Havers J. *De kloof tussen wat we weten en wat we doen in transitiezorg voor jongeren en adolescenten. Een Gap-analyse van de richtlijnen en standaarden.* [The gap between what we know and what we do in transitional care for adolescents. A gap analysis of guidelines and standards.] Rotterdam: XX Science / Hogeschool Rotterdam; 2010. www.opeigenbenen.nu

27. Ursum J, Rijken M, Heijmans M, Cardol M, Schellevis F. *Zorg voor chronisch zieken. Organisatie van zorg, zelfmanagement, zelfredzaamheid en participatie. [Care for the chronically ill. Organization, self-management, and participation]* Utrecht: Nivel; 2011.

28. Chamberlain MA, Kent RM. The needs of young people with disabilities in transition from paediatric to adult services. *Eura Medicophys.* 2005;41(2):111-123.

29. Perrin JM, Bloom SR, Gortmaker SL. The increase of childhood chronic conditions in the United States. *JAMA.* 2007;297(24):2755-2759.

30. van der Lee JH, Mokkink LB, Grootenhuis MA, Heymans HS, Offringa M. Definitions and measurement of chronic health conditions in childhood: a systematic review. *JAMA.* 2007;297(24):2741-2751.

31. Stein RE, Jessop DJ. What diagnosis does not tell: the case for a noncategorical approach to chronic illness in childhood. *Soc Sci Med.* 1989;29(6):769-778.

32. Perrin EC, Newacheck P, Pless IB, Drotar D, Gortmaker SL, Leventhal J, *et al*. Issues involved in the definition and classification of chronic health conditions. *Pediatrics.* 1993;91(4):787-793.

33. McPherson M, Arango P, Fox H, Lauver C, McManus M, Newacheck PW, *et al*. A new definition of children with special health care needs. *Pediatrics.* 1998;102(1 Pt 1):137-140.

34. Newacheck PW, Strickland B, Shonkoff JP, Perrin JM, McPherson M, McManus M, *et al*. An epidemiologic profile of children with special health care needs. *Pediatrics.* 1998;102(1 Pt 1):117-123.

35. Newacheck PW, Kim SE. A national profile of health care utilization and expenditures for children with special health care needs. *Arch Pediatr Adolesc Med.* 2005;159(1):10-17.

36. Mokkink LB, van der Lee JH, Grootenhuis MA, Offringa M, van Praag BMS, Heymans HSA. Omvang en gevolgen van chronische aandoeningen bij kinderen. [Extent and consequences of chronic conditions in children]. *Tijdschr Kindergeneeskd.* 2007;75(4):138-142.

37. Newacheck PW, McManus MA, Fox HB. Prevalence and impact of chronic illness among adolescents. *Am J Dis Child.* 1991;145(12):1367-1373.

38. Stein RE, Silver EJ. Comparing different definitions of chronic conditions in a national data set. *Ambul Pediatr.* 2002;2(1):63-70.

39. Binks JA, Barden WS, Burke TA, Young NL. What do we really know about the transition to adult-centered health care? A focus on cerebral palsy and spina bifida. *Arch Phys Med Rehabil.* 2007;88(8):1064-1073.

40. Stein RE. Challenges in long-term health care for children. *Ambul Pediatr.* 2001;1(5):280-288.

41. Roebroeck ME, Jahnsen R, Carona C, Kent RM, Chamberlain MA. Adult outcomes and lifespan issues for people with childhood-onset physical disability. *Dev Med Child Neurol.* 2009;51(8):670-678.

42. Reid GJ, Irvine MJ, McCrindle BW, Sananes R, Ritvo PG, Siu SC, *et al*. Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics.* 2004;113(3 Pt 1):e197-205.

43. Reid GJ, Webb GD, Barzel M, McCrindle BW, Irvine MJ, Siu SC. Estimates of life expectancy by adolescents and young adults with congenital heart disease. *J Am Coll Cardiol.* 2006;48(2):349-355.

44. Gortmaker SL, Walker DK, Weitzman M, Sobol AM. Chronic conditions, socioeconomic risks, and behavioral problems in children and adolescents. *Pediatrics.* 1990;85(3):267-276.

45. Meijer SA, Sinnema G, Bijstra JO, Mellenbergh GJ, Wolters WH. Coping styles and locus of control as predictors for psychological adjustment of adolescents with a chronic illness. *Soc Sci Med.* 2002;54(9):1453-1461.

46. Barlow JH, Ellard DR. The psychosocial well-being of children with chronic disease, their parents and siblings: an overview of the research evidence base. *Child Care Health Dev.* 2006;32(1):19-31.

47. Martinez W, Smith Carter J, Legato LJ. Social competence in children with chronic illness: a meta-analytic review. *J Pediatr Psychol.* 2011;36(8):878–890.

48. Pittet I, Berchtold A, Akre C, Michaud PA, Suris JC. Are adolescents with chronic conditions particularly at risk of bullying? *Arch Dis Child.* 2010;95(9):711-716.

49. Turkel S, Pao M. Late consequences of chronic pediatric illness. *Psychiatr Clin North Am.* Dec 2007;30(4):819-835.

50. de Wit M, Delemarre-van de Waal HA, Bokma JA, Haasnoot K, Houdijk MC, Gemke RJ, *et al.* Monitoring and discussing health-related quality of life in adolescents with type 1 diabetes improve psychosocial well-being: a randomized controlled trial. *Diabetes Care.* 2008;31(8):1521-1526.

51. Haverman L, Engelen V, van Rossum MA, Heymans HS, Grootenhuis MA. Monitoring health-related quality of life in paediatric practice: development of an innovative web-based application. *BMC Pediatr.* 2011;11:3.

52. Taylor RM, Gibson F, Franck LS. A concept analysis of health-related quality of life in young people with chronic illness. *J Clin Nurs.* 2008;17(14):1823-1833.

53. Ravens-Sieberer U, Gosch A, Rajmil L, Erhart M, Bruil J, Duer W, *et al*. KIDSCREEN-52 quality-of-life measure for children and adolescents. *Expert Rev Pharmacoecon Outcomes Res.* 2005;5(3):353-364.

54. Hemmingsson H, Jonsson H. An occupational perspective on the concept of participation in the International Classification of Functioning, Disability and Health--some critical remarks. *Am J Occup Ther.* 2005;59(5):569-576.

55. Sinnema G. Youths with chronic illness and disability on their way to social and economic participation: a health-care perspective. *J Adolesc Health.* 1992;13(5):369-371.

56. Mokkink LB, van der Lee JH, Offringra M, van Praag BMS, Heymans HS, Grootenhuis MA. Sociaal-maatschappelijke gevolgen en kwaliteit van leven in Nederland bij kinderen en jongvolwassenen opgegroeid met een chronische aandoening. [Social consequences and quality of life in Dutch children and young adults with a childhood-onset chronic condition]. *Tijdschr Kindergeneeskd.* 2007;75(6):229-235.

57. Barf HA, Verhoef M, Post MW, Jennekens-Schinkel A, Gooskens RH, Mullaart RA, *et al*. Educational career and predictors of type of education in young adults with spina bifida. *Int J Rehabil Res.* 2004;27(1):45-52.

58. Barf HA, Verhoef M, Jennekens-Schinkel A, Post MW, Gooskens RH, Prevo AJ. Cognitive status of young adults with spina bifida. *Dev Med Child Neurol.* 2003;45(12):813-820.

59. Groothoff JW, Grootenhuis M, Dommerholt A, Gruppen MP, Offringa M, Heymans HS. Impaired cognition and schooling in adults with end stage renal disease since childhood. *Arch Dis Child.* 2002;87(5):380-385.

60. Groothoff JW, Grootenhuis MA, Offringa M, Stronks K, Hutten GJ, Heymans HS. Social consequences in adult life of end-stage renal disease in childhood. *J Pediatr.* 2005;146(4):512-517.

61. Donkervoort M, Roebroeck M, Wiegerink D, van der Heijden-Maessen H, Stam H. Determinants of functioning of adolescents and young adults with cerebral palsy. *Disabil Rehabil.* 2007;29(6):453-463.

62. Verhoef M, Barf HA, Vroege JA, Post MW, van Asbeck FW, Gooskens RH, *et al*. Sex education, relationships, and sexuality in young adults with spina bifida. *Arch Phys Med Rehabil.* 2005;86(5):979-987.

63. Wiegerink DJ, Roebroeck ME, Donkervoort M, Cohen-Kettenis PT, Stam HJ. Social, intimate and sexual relationships of adolescents with cerebral palsy compared with able-bodied age-mates. *J Rehabil Med.* 2008;40(2):112-118.

64. van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, Wensink-Boonstra AE, Stam HJ, Roebroeck ME. Participation and health-related quality of life in adults with spastic bilateral cerebral palsy and the role of self-efficacy. *J Rehabil Med.* 2010;42(6):528-535.

65. Rahbek J, Werge B, Madsen A, Marquardt J, Steffensen BF, Jeppesen J. Adult life with Duchenne muscular dystrophy: observations among an emerging and unforeseen patient population. *Pediatr Rehabil.* 2005;8(1):17-28.

66. Gibson BE, Zitzelsberger H, McKeever P. 'Futureless persons': shifting life expectancies and the vicissitudes of progressive illness. *Sociol Health Illn.* 2009;31(4):554-568.

67. Abbott D, Carpenter J. *Becoming an adult: transition for young men with Duchenne Muscular Dystrophy (DMD).* Bristol University: School for Policy Studies;2010. www.muscular-dystrophy.org

68. van Huijzen SS, van Staa AL. Social participation and chronic ventilation: experiences of adults with neuromuscular disorders. *Scand J Occup Ther.* Accepted.

69. Calsbeek H, Rijken M, Bekkers MJ, Kerssens JJ, Dekker J, van Berge Henegouwen GP. Social position of adolescents with chronic digestive disorders. *Eur J Gastroenterol Hepatol.* 2002;14(5):543-549.

70. Plug I, Peters M, Mauser-Bunschoten EP, de Goede-Bolder A, Heijnen L, Smit C, *et al*. Social participation of patients with hemophilia in the Netherlands. *Blood.* 2008;111(4):1811-1815.

71. Bergers J, Nijhuis F, Janssen M, van der Horst F. Employment careers of young type I diabetic patients in The Netherlands. *J Occup Environ Med.* 1999;41(11):1005-1010.

72. Stam H, Hartman EE, Deurloo JA, Groothoff J, Grootenhuis MA. Young adult patients with a history of pediatric disease: impact on course of life and transition into adulthood. *J Adolesc Health.* 2006;39(1):4-13.

73. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis MA. Growing into disability benefits? Psychosocial course of life of young adults with a chronic somatic disease or disability. *Acta Paediatr.* 2012;101(1):e19-26.

74. Bosch AM, Grootenhuis MA, Bakker HD, Heijmans HS, Wijburg FA, Last BF. Living with classical galactosemia: health-related quality of life consequences. *Pediatrics.* 2004;113(5):e423-428.

75. Bosch AM, Maurice-Stam H, Wijburg FA, Grootenhuis MA. Remarkable differences: the course of life of young adults with galactosaemia and PKU. *J Inherit Metab Dis.* 2009;32(6):706-712.

76. Engelen V, Haentjens MM, Detmar SB, Koopman HM, Grootenhuis MA. Health related quality of life of Dutch children: psychometric properties of the PedsQL in the Netherlands. *BMC Pediatr.* 2009;9:68.

77. Loonen HJ, Grootenhuis MA, Last BF, Koopman HM, Derkx HH. Quality of life in paediatric inflammatory bowel disease measured by a generic and a disease-specific questionnaire. *Acta Paediatr.* 2002;91(3):348-354.

78. de Boer M, Grootenhuis M, Derkx B, Last B. Health-related quality of life and psychosocial functioning of adolescents with inflammatory bowel disease. *Inflamm Bowel Dis.* 2005;11(4):400-406.

79. de Wee EM, Fijnvandraat K, de Goede-Bolder A, Mauser-Bunschoten EP, Eikenboom JC, Brons PP, *et al*. Impact of von Willebrand disease on health-related quality of life in a pediatric population. *J Thromb Haemost.* 2011;9(3):502-509.

80. Bannink EM, Raat H, Mulder PG, de Muinck Keizer-Schrama SM. Quality of life after growth hormone therapy and induced puberty in women with Turner syndrome. *J Pediatr.* 2006;148(1):95-101.

81. Huygen AC, Kuis W, Sinnema G. Psychological, behavioural, and social adjustment in children and adolescents with juvenile chronic arthritis. *Ann Rheum Dis.* 2000;59(4):276-282.

82. de Wit M, Delemarre-van de Waal HA, Bokma JA, Haasnoot K, Houdijk MC, Gemke RJ, *et al*. Self-report and parent-report of physical and psychosocial well-being in Dutch adolescents with type 1 diabetes in relation to glycemic control. *Health Qual Life Outcomes.* 2007;5:10.

83. Engelen V, Koopman HM, Detmar SB, Raat H, van de Wetering MD, Brons P, *et al*. Health-related quality of life after completion of successful treatment for childhood cancer. *Pediatr Blood Cancer.* 2011;56(4):646-653.

84. van Dijk J, Huisman J, Moll AC, Schouten-van Meeteren AY, Bezemer PD, Ringens PJ, *et al*. Health-related quality of life of child and adolescent retinoblastoma survivors in the Netherlands. *Health Qual Life Outcomes.* 2007;5:65.

85. Buffart L, van den Berg-Emons R, Roebroeck M, Stam H. Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele. *Dev Med Child Neurol.* 2009;51(11):886-894.

86. Grootenhuis MA, de Boone J, van der Kooi AJ. Living with muscular dystrophy: health related quality of life consequences for children and adults. *Health Qual Life Outcomes.* 2007;5:31.

87. Poley MJ, Stolk EA, Tibboel D, Molenaar JC, Busschbach JJ. Short term and long term health related quality of life after congenital anorectal malformations and congenital diaphragmatic hernia. *Arch Dis Child.* 2004;89(9):836-841.

88. Hartman E, Oort F, Aronson D, Hanneman MJ, van der Zee DC, Rieu PN, *et al*. Critical factors affecting quality of life of children and adolescents with anorectal malformations or Hirschsprung's disease. *Am J Gastroenterol.* 2004;99(5):907-913.

89. Spijkerboer AW, Utens EM, De Koning WB, Bogers AJ, Helbing WA, Verhulst FC. Health-related Quality of Life in children and adolescents after invasive treatment for congenital heart disease. *Qual Life Res.* 2006;15(4):663-673.

90. Hijmans CT, Fijnvandraat K, Oosterlaan J, Heijboer H, Peters M, Grootenhuis MA. Double disadvantage: a case control study on health-related quality of life in children with sickle cell disease. *Health Qual Life Outcomes.* 2010;8:121.

91. Grootenhuis MA, Stam H, Last BF, Groothoff JW. The impact of delayed development on the quality of life of adults with end-stage renal disease since childhood. *Pediatr Nephrol.* 2006;21(4):538-544.

92. Hatzmann J, Heymans HS, Ferrer-i-Carbonell A, van Praag BM, Grootenhuis MA. Hidden consequences of success in pediatrics: parental health-related quality of life - results from the Care Project. *Pediatrics.* 2008;122(5):e1030-1038.

93. Bevans KB, Riley AW, Moon J, Forrest CB. Conceptual and methodological advances in child-reported outcomes measurement. *Expert Rev Pharmacoecon Outcomes Res.* 2010;10(4):385-396.

94. Huber M, Knottnerus JA, Green L, van der Horst H, Jadad AR, Kromhout D, *et al.* How should we define health? *Brit Med J.* 2011;343,d4163.

95. van Staa AL, Van Huijzen SS. Transitie in zorg bij anusatresiepatiënten. Aan de overgang van kinder- naar volwassenenzorg valt nog veel te verbeteren. [The transition from pediatric to adult care for patients with anorectal malformation: much needs to be improved]. *ErVaring (Vereniging Anusatresie).* 2008(Dec):7-8.

96. Strauss AL, Glaser B. *Chronic illness and the quality of life*. St. Louis: Mosby; 1975.

97. Corbin JM, Strauss AL. *Unending work and care: managing chronic illness at home*. San Francisco: Jossey-Bass; 1988.

98. Bury M. Chronic illness as biographical disruption. *Sociol Health Illn.* 1982;4(2):167-182.

99. Schumacher KL, Meleis AI. Transitions: a central concept in nursing. *Image J Nurs Sch.* 1994;26(2):119-127.

100. de Lange J, van Staa AL. *Transities in ziekte en zorg. Op zoek naar een nieuw evenwicht*. Openbare Les. [Transitions of illness and care. Searching for a new balance. Inaugural Speech]. Rotterdam: Hogeschool Rotterdam; 2003. ISBN 90-801776-2-8.

101. Olsson CA, Bond L, Burns JM, Vella-Brodrick DA, Sawyer SM. Adolescent resilience: a concept analysis. *J Adolesc.* 2003;26(1):1-11.

102. Berntsson L, Berg M, Brydolf M, Hellstrom AL. Adolescents' experiences of well-being when living with a long-term illness or disability. *Scand J Caring Sci.* 2007;21(4):419-425.

103. Taylor RM, Gibson F, Franck LS. The experience of living with a chronic illness during adolescence: a critical review of the literature. *J Clin Nurs.* 2008;17(23):3083-3091.

104. Olsson CA, Bond L, Johnson MW, Forer DL, Boyce MF, Sawyer SM. Adolescent chronic illness: a qualitative study of psychosocial adjustment. *Ann Acad Med Singapore.* 2003;32(1):43-50.

105. Moos RH. Life stressors, social resources, and coping skills in youth: applications to adolescents with chronic disorders. *J Adolesc Health.* 2002;30(4 Suppl):22-29.

106. LeBlanc LA, Goldsmith T, Patel DR. Behavioral aspects of chronic illness in children and adolescents. *Pediatr Clin North Am.* 2003;50(4):859-878.

107. Heijmans M, Rijken M, Foets M, de Ridder D, Schreurs K, Bensing J. The stress of being chronically ill: from disease-specific to task-specific aspects. *J Behav Med.* 2004;27(3):255-271.

108. Moos RH, Holahan CJ. Adaptive tasks and methods of coping with illness and disability. In: Martz E, Livneh H, eds. *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. New York: Springer; 2007.

109. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.

110. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics.* 2004;114(5):1272-1280.

111. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J. Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns.* 2002;48(2):177-187.

112. Lorig KR, Holman H. Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med.* 2003;26(1):1-7.

113. Bandura A. *Self-efficacy: the exercise of control*. New York: W.H. Freeman and Company; 1997.

114. American Academy of Pediatrics, American Academy of Family Physicians & American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics.* 2002;110(6 Pt. 2):1304-1306.

115. Creswell JW. *Research design: qualitative, quantitative, and mixed methods approaches*. 2nd Edition ed. London: Sage Publications; 2003.

116. Jedeloo S, van Staa AL. Q-methodologie, een werkelijke mix van kwalitatief en kwantitatief onderzoek? [Q-methodology: a true mix of qualitative and quantitative research?]. *KWALON.* 2009;14(2):5-15.

117. Cross RM. Exploring attitudes: the case for Q methodology. *Health Educ Res.* 2005;20(2):206-213.

118. Watts S, Stenner P. Doing Q methodology: theory, method and interpretation. *Qual Res Psychol.* 2005;2(2):67-91.

119. Hart RA. *Children's participation: from tokenism to citizenship.* Innocenti Essays No 4. Florence: UNICEF; 1992. http://www.unicef-irc.org/publications/100

120. Litt IF. Research with, not on, adolescents: community-based participatory research. *J Adolesc Health.* 2003;33(5):315-316.

121. Frei A, Svarin A, Steurer-Stey C, Puhan MA. Self-efficacy instruments for patients with chronic diseases suffer from methodological limitations--a systematic review. *Health Qual Life Outcomes.* 2009;7(86):1-10.

Part 1

Preferences for care

1

Exciting but exhausting: experiences with participatory research with chronically ill adolescents

AnneLoes van Staa, Susan Jedeloo, Jos M Latour, Margo J Trappenburg

Abstract

BACKGROUND: Adolescents with chronic conditions are major users of pediatric hospitals, but seldom participate in the evaluation of services or in research. Little is known about the usefulness of the participatory approach in adolescent health research.

OBJECTIVE: To evaluate the feasibility, benefits and limitations of a participatory research project involving chronically ill adolescents as coresearchers.

DESIGN, SETTING AND PARTICIPANTS: Nine adolescents, aged 15–17 years, acted as coresearchers in a hospital-based participatory research project. They codeveloped an interview protocol and during a disco party held for this purpose interviewed each other and 25 fellow patients (12-19 years). They provided advice on the draft report and participated in the dissemination of the results, but were not involved in the design of the project or analysis of results.

RESULTS: Involving adolescents in participatory health research was feasible and appreciated by researchers and youth alike, but had its drawbacks too. The peer-research attracted few participants, the interviews lacked depth and did not yield substantial new insights. Maintaining a high level of participation of the chronically ill coresearchers also proved difficult.

CONCLUSIONS: Adolescents with chronic conditions like to have a say in the design and evaluation of hospital services. But their participation as coresearchers demands ample resources from all parties involved without automatically improving research quality. Participatory research does not seem the most effective and efficient way to make services more responsive. We therefore recommend further exploration of other creative and sustainable ways for involving youth in health care service development and innovation.

1   Introduction

Over the last decade, the active involvement of patients in decision-making processes has become a policy priority aimed at making health care services more patient-centered. The Netherlands Ministry of Health, for example, stimulates transition from a supply-driven to a demand-driven health care system, expecting users to be active consumers selecting those services that best suit their needs.[1] Following the consumerist rhetoric, patient or user participation refers to consultation and involvement of patients in all health care decisions on the individual and collective level; in the development and evaluation of services; and also in health research.[2] Although the notion of patients as partners in the research process is of fairly recent date, the participatory approach is quite popular with health researchers.[3-5]

Participatory Research (PR) is a collaborative undertaking aimed at more involvement of the community being studied in all aspects of the research process. It is carried out *with* and *by* the research subjects rather than *on* them.[6, 7] Core elements are colearning and reciprocal transfer of expertise, shared decision-making power and mutual ownership of process and products of the research enterprise.[3, 6, 8, 9]

From a review by Viswanathan *et al,*[8] it appears that well-conducted PR enhances competencies and capacities of the community; improves research quality; leads to more effective and efficient interventions; and results in better health outcomes. In contrast, dissenting voices question the effectiveness and impact of consumer involvement in research[10] and there are indications that the benefits of PR are not equitably spread, nor gained without considerable cost.[11]

The call for securing patients' voice and choice in health care does not only apply to adults, but also to minors.[12-14] Children and adolescents are major users of pediatric health care services and have special needs.[15] This holds true in particular for those with chronic illnesses or disabilities, who will need to take over responsibility from their parents to manage their own health.[16, 17]

The right of minors to express their views and be involved in decision making in all matters affecting them is grounded in the United Nations Convention on the Rights of the Child (UNCRC 1989; Article 12). However, participation of adolescents with chronic conditions in medical decision making, in services evaluation or in health research is by no means self-evident, nor easy to accomplish. For example, the Dutch Medical Treatment Act (WGBO; 1995) states that adolescents aged 12-15 years have the right to codecide with parents in treatment decisions, and that those over 16 may decide for themselves. Still, to date their active participation is still fairly limited in primary care[18, 19] or hospital care[20] in the Netherlands. Similar observations come from other countries.[21-25]

Giving young people a collective voice in the development and evaluation of health care services, implies their views should be listened to[14, 26-28] and acted upon.[29] Given the opportunity, young people are quite willing and able to make decisions about services they want,[30-33] but disabled children tend to be less involved in participation activities than nondisabled children, according to Franklin and Sloper.[34]

An important barrier to children's partnership in health care is related to concerns about offering choices to them.[33, 35, 36] Nevertheless, the traditional view of children's incompetence

to give consent for treatment and for research participation has been contested. Children are now seen as social actors, competent to be commentators on their own lives and to be involved in decision making.[37] Chronically ill children often prove to be capable of making informed, 'wise' decisions in their own best interests.[17, 29, 38] Still, young people are often not recognized as service users,[30] seeing that the parents' perspective tends to be the basis for measuring quality of pediatric hospital care.[39] However, the parents' perspective may not accurately represent their children's views.[40, 41]

Inclusion of young people themselves in research is therefore being considered essential. The Guidelines for Adolescent Health Research[35] propose a task for adolescent research subjects in working out research priorities and purposes. According to Litt,[7] the next logical step would be inviting adolescents' input into a study's design and execution. Participatory research has been used for children suffering from cancer,[42, 43] diabetes,[44, 45] and for disabled children.[28, 34, 46] Furthermore, it was applied in exploring children's preferences for hospital care or design,[30, 47-50] and in promoting health and providing services to marginalized or underserved groups, such as homeless or human immunodeficiency virus positive (HIV+) youth.[51-55]

In view of the potential benefits of participation, we used several forms of consultation and involvement in the 'On Your Own Feet' Project, launched to explore chronically ill adolescents' self-care competencies and their preferences. In the study described here, adolescents participated as *coresearchers*, ie, they interviewed fellow patients about their preferences and experiences with hospital care.

The aim of this study is to assess the feasibility, advantages and limitations of working with chronically ill adolescents in participatory research.

2 Methods and sample

The 'On Your Own Feet' Project took shape between 2004 and 2007 in the Erasmus MC - Sophia Children's Hospital, Rotterdam, the Netherlands. This hospital treats over 4000 adolescents with chronic conditions, but has no specialized adolescent in-patient facilities and only a handful of outpatient youth clinics. The hospital board supported the idea of consulting patients in a participatory project in order to better adjust services to their needs. However, a hospital is not an ideal setting for community-based research. Thus we decided to organize a disco party to be held outside the hospital at an attractive location, in a real discotheque, which usually is not easily accessible to adolescents with chronic conditions. During this event the coresearchers were to interview their fellow patients. We describe the design and execution of the study in four subsequent stages.

2.1 Recruitment of coresearchers

Adolescents were sought who were both motivated to act as coresearchers and willing to give their opinion about the care they received. A representative sample of adolescents was not intended. Ten nurse specialists working in different subspecialty departments in the hospital were asked to identify adolescents > 15 years of age whom they thought would be interested

in participating. These nurses had cooperated in a previous 'On Your Own Feet' study. We expected that young people would be more willing to participate if they were invited by familiar care providers.

Together with the Communications Department we developed a leaflet explaining the aim, method and expected requirements of potential coinvestigators. The study was also announced on two websites, the project-related site http://ww.opeigenbenen.nu and a hospital-based site for patients: http://www.sophiakids.nl.

Eight of 10 nurse specialists eventually approached adolescents. Sixty information leaflets were printed, but we do not know how many were handed out. Ten adolescents consented, but as one girl with cystic fibrosis needed to be hospitalized, nine (two boys and seven girls; aged 15-17 years) actually participated (Table 1). Their chronic conditions included dermatologic disorders (n = 2), blood disorders (n = 2), neuromuscular diseases (n = 2), renal failure (n = 2), and diabetes mellitus (n = 1).

Table 1   *Characteristics of study participants*

	interviewees n = 25 (%)	coresearchers n = 9 (%)	*P*-value[a]
Gender			
boys	9 (36.0)	2 (22.2)	0.68
girls	16 (64.0)	7 (77.8)	
Age (years)			
12-15	20 (80.0)	3 (33.3)	0.03
16-19	5 (20.0)	6 (66.7)	
Age at diagnosis chronic condition (years)			
at birth & < 6 years	10 (40.0)	4 (44.4)	1.00
> 6 years	15 (60.0)	5 (55.6)	
Visits outpatient department past three years			
less than 12 visits	21 (84.0)	1 (11.0)	<0.01
over 12 visits	4 (16.0)	8 (89.0)	

[a] *Fisher's Exact Test; 2-sided.*

2.2   Training program and designing of interview protocol

As the coresearchers had no research experience at all, introducing them to interviewing techniques was necessary. In order to make a training session appealing to participants, the research team (AvS, SJ, JML) organized a site-visit to the national newspaper *nrc.next* for a training session on location. The aim of the training was to jointly develop an interview protocol and prepare the coresearchers for their task as interviewers.

The chief editor received our coresearchers as VIPs and first gave a short introduction on running a newspaper. They then visited the newspapers' offices before the actual training session, which lasted three hours. Following a brief introduction on the aim of interviewing, the coresearchers worked in pairs to interview each other while another coresearcher and a member of the research team observed them. They discussed their experiences with hospital staff by posing the sample question: *"What is a good doctor?"* Afterwards, the observers evaluated the technique of interviewing. Next came a single role play acted out in front of the group demonstrating how to pose probing questions to a noncooperative respondent. The researchers provided the adolescents with tips and tricks. Finally, the themes and topics to be included in the interview protocol were discussed. The research team presented a short draft protocol, divided into several general themes. Some open-ended questions were inspired by our previous interview study about adolescents' health care preferences[20] and by a child-friendly questionnaire designed in another Dutch pediatric hospital.[56] Working in small groups, the coresearchers re-phrased the questions where needed and added new topics.

In the weeks after the training session, the research team categorized and merged the provisional questions. After several discussion rounds with the coresearchers through e-mail, the interview protocol was finalized (Box 1).

Box 1 *Interview protocol designed in collaboration with coresearchers*

I   Your general impression of the hospital
1 If I say, Sophia Children's Hospital, what do you think?
2 What is best in the Sophia Children's Hospital?
3 What is worst in the Sophia Children's Hospital?

II   Doctors, nurses and other staff
4 According to you, what is a good doctor?
5 How are your experiences with doctors at Sophia Children's Hospital? Please give examples of both positive and negative experiences.
6 How are your experiences with other health care workers, such as nurses, dieticians and social workers at Sophia Children's Hospital? Please give examples of both positive and negative experiences.
7 What is your number one advice for hospital staff?
8 How do you feel about the fact that you'll have to leave Sophia Children's Hospital when you turn eighteen?
9 Do you think you're ready to transfer to adult care?

III When visiting the outpatient department
10 What is most important to you when you're at the outpatient department?
11 Do you feel that the doctor focuses mostly on you?
12 What happens if you have a different opinion than the doctor?
13 Would you prefer to talk with the doctor alone sometimes?
14 If you could change the organization of the outpatient department, what would you do?

IV   When admitted to hospital
15 What do you miss most when you have to stay in hospital?
16 How are your experiences with the Acute Care department at Sophia Children's Hospital?
17 Suppose, you get a bag full of money for the hospital, what would you buy?

V Activities in the hospital
18 How do you feel about the activities organized in the hospital?
19 What is your opinion on the hospital clowns?
20 Do you think it is important to meet fellow patients? How should the Sophia Children's Hospital make this possible?
21 Do you think young patients should get a greater say in the hospital? How should this be organized?
22 Is there anything else you have on your mind about the Sophia Children's Hospital?

2.3   Setting of research: disco party

The research team organized a disco party on a Saturday afternoon in a Rotterdam club, in May 2006. Recruitment of potential guests started four weeks before. Parents were not allowed, but the patients could bring a friend. Most adolescents would not know other participants and we anticipated that a friend's presence could help them feel more comfortable. Colorful posters announcing the event were put up throughout the Sophia Children's Hospital and an information flyer was distributed via the counters of the six outpatient departments to all visiting patients over 12 years of age. Eight nurse specialists and the Communications Department sent this flyer to their adolescent patients or contacts with a personal invitation. Approximately 1000 leaflets were distributed in this short period.

The disco party was staged with the help of many volunteers including a well-known Dutch radio DJ, club staff, students, and graffiti and break-dance artists. Several workshops (such as break dance, rap, graffiti and nail art) were offered in addition to the disco dancing itself.

The coresearchers had been briefed before. They were instructed to use the interview protocol as a guide - not as a structured questionnaire. The interviews were held in a designated area: the discotheque's café. The coresearchers worked in pairs to support each other and were seated at round tables. Each pair was assisted by a student who directly typed the respondents' answers into a laptop.

All in all, 25 young patients – predominantly younger girls (12-15 years) - and 26 healthy friends attended. Thirty-four patients participated in the peer-interviews, ie, 25 attendants and 9 coresearchers.

2.4   Data-analysis and dissemination of results

The coresearchers were also invited to help with the data-analysis. Regrettably, establishing a date for a 'real-life' meeting to discuss the preliminary results with the coresearchers proved impossible. Instead, the research team analyzed the anonymous transcripts and invited coresearchers to comment on draft versions of the report through e-mail. To establish the additional value of the participatory approach, we compared the peer-interviews to qualitative interviews.[20]

The coresearchers participated in several media activities: David was interviewed by a national newspaper, others were interviewed on national radio or on the hospital-based Sophia

Television. Dorine and Britt contributed to a popular article about the project in *ZieSo*; a maga-zine for patients and parents distributed in the hospital's waiting areas.

The results of the overall project 'On Your Own Feet' including those of the disco party were disseminated in a national conference that started with a theatre play created by adolescents (April 2007, Rotterdam). For the recruitment of the young actors we used the same strategy as described above. The original coresearchers were also invited, but refrained from participat-ing. A group of nine young people worked with a professional drama teacher to create the play, using role-play and improvisation. The play was the highlight of the conference, attended by 500 delegates – including health professionals, young patients and their parents. After the show the young actors handed the first copy of the book summarizing the project's findings to representatives of the hospital board. Their self-created rap urged the hospital board to listen to young people and adjust care to their needs.

2.5   Ethical standards and procedures

The study was approved by the Erasmus MC Medical Ethics Review Board. All participants and their parents received written information about the project and provided their written consent. The coresearchers received €75 remuneration for their input, estimated at 15 h each. Participants of the disco party could win attractive prizes in a raffle. The young actors partici-pating in the drama project received €75 as well.

3   Results

In order to assess the benefits and limitations of this participatory approach, we describe the effects on (a) the coresearchers, respondents and the research team; (b) research quality; and (c) improvement of services in Sophia Children's Hospital. Table 2 presents an overview.

3.1   Effects on respondents, coresearchers, and research team

At first glance, the project seemed successful. For most *visitors,* a discotheque was a place they had never seen the inside of and all seemed to enjoy the party. The research team received many compliments from parents and young people alike. Adolescents felt the hospital should organize more age-appropriate activities during which they can meet fellow-patients. Also, all attending patients were willing to share their experiences and ideas about the hospital. They liked having the opportunity to speak out their appreciation for the hospital, while at the same time grabbing the opportunity to ventilate critical comments and make recommendations. and some recommended to consult youth more frequently. A national radio reporter asked one of them how she enjoyed being interviewed by peers and she answered it was "*a great idea*".

The *volunteers* who helped organizing the event were very willing to cooperate and were impressed about the adolescents' resilience as most of them were unfamiliar with youth with

Table 2 *Strengths and limitations of participatory research*

	Strengths	Limitations
Young patients	• Liking to give opinion about their own care • Opportunity to meet fellow patients in non medical context • Having a nice time and new experience	• Relatively few attendees. Disco party and contact with fellow patients probably not appealing to all ages and all patients
Coresearchers	• Enthusiasm to participate in data collection • Empowering experience • Increased self-esteem • Added status; earning some money	• Demanding in terms of energy and time investment • Difficult to maintain enthusiasm for participation • Representativeness
Hospital staff	• Enjoying the opportunity to contribute to a positive experience • Being inspired to giving young patients a greater say in their own care	• Difficult to guarantee that suggestions are being acted upon • Doubts about generalizability of results
Research team	• Stronger commitment to youth' needs and preferences • Enhanced public exposure; facilitating dissemination of results • Exciting to collaborate with adolescents	• High time and resources expenditure • Quality of peer-interviews disappointing • Recruitment and quality of data not better than with traditional (qualitative) research strategies

disabilities. *Parents* were also positive about the project; some were prepared to drive for hours to bring their child.

Hospital staff, notably the nurse specialists and the Communications Department, were very supportive. They relished the chance to invite their patients for an activity in which they could meet fellow patients. The staff members were also genuinely interested in young people's opinions. Through their personal approach, recruitment of peer-researchers had been rather easy.

As a consequence of this sampling strategy, most *coresearchers* were well-known to the hospital staff and they had extensive hospital experience. They were significantly older than the other disco-attendees and had visited the outpatient department of the hospital more often in the past 3 years (Table 1). The coresearchers felt grateful towards hospital staff and yearned to give something in return. Their motivations for wanting to be coresearcher included: opportunity to learn interviewing skills; enjoying the prospect of visiting a newspaper office; eager to meet fellow-patients and to give feedback to hospital staff. The opportunity to earn a little money was also attractive.

From most coresearchers we learnt they found the experience worthwhile, albeit strenuous. Our disease-burdened coresearchers had lower energy levels than their healthy peers. The training program and interview sessions at the disco party exhausted them. Britt commented in her report on the training session: *"it was a rather strenuous afternoon, but I learnt a lot".* She also commented that it would probably take her three days to recover from the disco party, but that it was worth it. The research team had organized support for the core-

searchers during the interviews and arranged for them to take turns, but we had not realized how burdensome this task was for some of them.

Participation of the coresearchers after data collection was limited. Attempts were made to involve them in the data analysis, but despite several recalls, no more than four of the nine coresearchers commented on the draft report, stating "*all is OK*" and claiming the results were recognizable to them. So most coresearchers seemed to have lost interest, but one girl agreed to be an advisor to the board of the research project together with her mother. Several attended the national conference.

Being a coresearcher was empowering in some respects. Not only did they learn new skills and felt useful as a researcher, they also enjoyed receiving a VIP-treatment. Being interviewed in the media added to their self-confidence. One mother commented that being a coresearcher helped her daughter to turn her illness into something positive. Another coresearcher decided to become a journalist. The remuneration also increased their feelings of self-esteem: for most this was the first money they had ever earned.

As *research team* we enjoyed working with these youth, and it certainly increased our sensitivity towards their perspective. It was exciting to work intensely with them during the research endeavor. Being connected to these adolescents and to share experiences with them strengthened our resolve to focus research efforts on improving quality of adolescent health care. Consequently, we invested more time and energy than anticipated in disseminating the research results and translating them into practice. All in all, the participatory process was exciting for all parties involved. It was fun, but also hard work and exhausting. While we had anticipated that the project would take some 500 working hours, we spent more than double. The money investment was some € 5000.

Despite the large investments of resources in recruitment and preparation of the disco party, a disappointing number of no more than 25 adolescents attended. As it was the end of the spring holidays, we had expected more visitors. The low number may be due to the somewhat rushed, indirect recruitment strategy, and choice for and timing of the event, but it also goes to show that a participatory approach does not necessarily facilitate recruitment.

3.2   Effects on research quality

The training program was limited to one afternoon. Busy school schedules and exams limited availability of the coresearchers and several lived outside Rotterdam or were seriously disabled, making them dependent upon adults for transportation. The coresearchers played an active part in the training session and provided several interesting interview topics, for example, their experiences in the Emergency Department and with hospital clowns. Yet, only general aspects of interviewing could be presented and practiced. Consequently, they were not very well prepared for interviewing their fellow-patients. Most of the interviews lasted no longer than 10 min and additional questions to clarify and explore answers were not often posed. The interview protocol was used as a structured questionnaire rather than a guide for open conversation as recommended. We had expected that these adolescents would speak more freely with each other than with trained researchers, but there are no indications that this was the case.

The typists' interpretation of what was said during the interviews cannot be verified. The café area where the interviews were held was a crowded and somewhat noisy spot. The interviews could therefore not be audiorecorded and were transcribed on the spot. Based on our observations, we conclude that the comments were recorded fairly literally but briefly, and that important details and nuances may have been missed.

Compared to our previous face-to-face interviews with adolescents with chronic conditions, the peer-interviews yielded few new insights, preferences for hospital care being fairly similar in both. Most useful proved the coresearchers' comments and suggestions on the sample research questions, and these were used, together with some quotes of the peer-interviews, in a follow-up questionnaire.[57]

One of the most interesting outcomes of the peer-interviews is related to the desired level of participation in service evaluation. To the question *'Do you think young patients should get a greater say in the hospital?'* many young people answered they did not think this was necessary, because *"you already have enough to say in Sophia's"* or *"all is fine as it is"*. Adolescents who were most explicit about the need of having a greater say, proved to be the coresearchers themselves. One felt that *"doctors should listen better to what children have to say".* Another said: "*we ought to do more such studies, so that young people themselves can tell what they prefer".* Some coresearchers recommended the installation of a youth forum in the hospital, although not all of them thought this was necessary. Compared to other participants, coresearchers seemed more articulate about the need for youth participation.

3.3   Effects on improvement of services

The project was designed in collaboration with hospital staff so as to ensure their involvement as well as a focus on relevant issues for daily practice. This would also ensure their commitment towards improving services based on adolescents' preferences and needs. During recruitment and organization of the disco party the level of participation of nurse specialists and staff from the Communications Department was high. Through them we not only gained access to young patients, but we also learnt from their practical experience. Nurses had valuable comments on the information leaflets and the way to involve youth. The idea to create a disco party came from the head of the Communications Department, who had organized several other youth activities before and managed to involve many volunteers and VIPs. Without the active involvement of hospital staff, this project would never have been realized. However, this does not guarantee that adolescents' recommendations will be acted upon.

There are not many direct results to boast about. A striking one, however, is the aroused interest in the overall project from the media and from health care providers both in the hospital and nationwide. Especially the drama play performed by adolescent actors was a great success. Partly as a consequence of the overall 'On Your Own Feet' Project, awareness increased among professionals in the hospital of the special health care needs and preferences of adolescents with chronic conditions. Several professionals developed new initiatives such as transition clinics, and involving young people in the design of peer activities and educational materials. These are the first steps in making health care services in Sophia Children's

Hospital more age-appropriate – an important recommendation from adolescents that were interviewed.

4 Discussion

Children and adolescents are able to express views on living with chronic illness or disability[58]; they can specify their health care needs and have constructive ideas about improving quality and service development.[26, 28, 29, 59] Most studies indicate that they want to be involved, respected and listened to,[30, 34, 38] but young people may also be critical about participation if not done properly, or if not meaningful to them.[30] Stafford *et al* consulted 200 young people about their experiences with consultation. It is judged meaningful when it concerns issues directly affecting them and when it is likely to yield results.[31]

Youth participation should not be used for window dressing or tokenism. In the overall 'On Your Own Feet' project this was avoided by having adolescents 'on board' during all stages. So on the one hand it proved feasible to involve young people with chronic conditions in the evaluation of health services. On the other hand, we feel that the participatory approach did not live up to our expectations.

Abma *et al*[9] reports on several projects involving patients as equal research partners. Our intention had indeed been to share responsibility with the adolescents in the design, execution and presentation of the study. This goal has not been attained. True, the adolescents acted as respondents, advisors and interviewers, but they were not equal research partners. Not only the number of young patients reached, but also their level of involvement and the outcomes were rather meager compared in view of the effort invested by the research team and coresearchers alike. Since mutual learning, openness, and respect are guiding principles for collaborations in PR,[9] we feel it is justified to discuss not only the benefits of the participatory approach, but also the limitations and drawbacks we were confronted with.

4.1 Strengths of participatory research

Increased commitment of the research team and hospital staff towards the adolescents' perspective may very well be the most important advantage of PR.[53] Also, young participants were positive of the opportunity given to them to be listened to - a feature of PR generally perceived as a strength.[11, 48, 53] Meeting fellow-patients outside a medical context can be seen as another advantage, as most of the visitors to the disco party appreciated this. Yet, not all young people with chronic conditions prefer to meet fellow-patients.[20]

For the coresearchers, the peer-research seemed empowering and rewarding, even though their participation had been limited in time and impact. Others have reported similar benefits for patient research partners.[9, 11, 53]

The research team enjoyed to collaborate with young people and it inspired us to disseminate the findings to a much wider public than academics alone and to experiment with alternative ways to involve youth such as the use of drama.[47] If user consultation and public relations are desired outcomes, participation of the researched subjects may be a good strategy.

4.2   Limitations of participatory research

An important claim for PR is that the *research itself* benefits from collaboration with the re-searched – through better questions, recruitment, data collection, analysis and dissemina-tion.[3, 8, 11, 53] On the whole, we were more content with the PR process than with the outcomes. The project implied a lot of work for the research team, hospital staff, volunteers and core-searchers alike. If research quality or recruitment really should improve, this would be worth it. In this case, however, the research benefitted less than we had hoped for, both in terms of quantity of participants and of research quality.

An explanation for the limited attendance to the disco party may be that young patients do not wish to focus on their disease or on hospital care, but rather on normalizing their condition and using their energy for participating in the 'healthy' world. Moreover, they were rather pleased with care-as-usual in the children's hospital compared to high-risk and out-of-the-mainstream youth in other projects.[51, 55] Lightfoot and Sloper's evaluation of six hospital-based initiatives involving adolescents in service provision in the UK also demonstrated that few young people actually participated.[30]

A possible issue for concern is *representativeness.*[10] The coresearchers differed from their fellow patients in being more outspoken about preferences, more critical of care received and more self-confident in talking to staff and peers. To enable informed decision making these viewpoints provided a good starting point, but they needed to be completed by survey data from a representative sample of users.

Despite our efforts, coresearchers' involvement was fairly limited compared to other re-ports.[9, 11] They fully participated in data collection only, not in analysis. Keeping their initial enthusiasm at the same level through all phases of the project proved rather difficult. Also, not all forms of participation were successful. For our adolescent coresearcher it proved virtually impossible to make a meaningful contribution as advisor to the research board. In contrast, the young persons' involvement in the drama project was a great way to stimulate self-expres-sion and to disseminate results.[47]

Disadvantages of using consumer involvement for research quality are not frequently re-ported.[8, 10] The data collected also did not substantially add to our previous knowledge gained from interviews. There are no indications that enhanced rapport between coresearchers and their peers reduced the bias resulting from lack of confidence between interviewer and interviewee, as is sometimes claimed.[3] The conversations lacked depth, demonstrating that interviewing is no easy job.

So, would we 'do it again'? Yes, but differently. We would search for more effective and efficient forms of youth participation than peer-research.

4.3   The more participation, the better?

Most PR initiatives report satisfaction with both the process and the results.[3, 8, 60] PR is claimed to add value to all stages of the research process for both academic and nonacademic part-ners.[3, 5, 9] The downsides of PR are not often discussed or are labeled as challenges only.[8, 53]

On closer examination, however, others have also reported PR to be more resource intensive, time-consuming, complicated and less efficient than traditional research.[8, 10, 11, 53]

Advocates of participation usually stress that obstacles need to be overcome by more, proper, genuine, sustainable participation.[32, 38] From isolated consultations like ours, we should move to 'embedded' participation in organizational cultures, according to Sinclair.[32] Indeed, few challenge the basic romantic notion that moving toward maximum patient participation in all aspects of research is optimal. And few wonder whether young people themselves want to be included in organizational structures or in decision-making. A recent study of Knopf *et al* demonstrated that the largest proportion of chronically ill adolescents under study tended to favor a passive role in treatment (preferred by 46%) over shared decision making (37%).[61]

Offering youth the choice to their level of participation would be more ideal than assuming that 'more is always better'.[11] 'Embedded' participation also involves the risk of creating participation elites. The level of participation should be negotiated, equitable rather than equal participation being the ultimate goal.[3] Participation seems a *hurray-word* like democracy or partnership.[62] But even advocates of participation note evidence of limited impact, of lack of measurable outcomes and of disillusionment in young people engaged in consultation and decision making.[34, 38]

According to Coad and Shaw, it is unclear whether having a choice for children leads to more responsive services.[33] Partnerships with young people in research require that their suggestions are not only listened to, but also acted upon. Researchers, however, are not the ones who change policies or practices. User consultation with those in charge of health care services could perhaps be more effective in achieving responsive services. In Experience-Based CoDesign, for example, user experiences are directly made accessible to the providers of services.[63] Staff and patients discuss the designing together. All in all, adolescents with chronic conditions may benefit more from being integral part of service improvement and innovation than from being research partners.

5   Conclusion

Adolescents with chronic conditions like to have a voice in the design and evaluation of health care services, but the desirable extent of patient partnership in research and consultation is still undefined. A participatory research approach may be helpful in increasing their social competency and disseminating research results. Adolescents are able to participate to a certain extent and researchers find it exciting to work with them. Yet, it participatory research is no easy job. It demands high commitment, extensive resources, and hard work while not always succeeding in providing meaningful results. Finding a balance between the benefits and costs of participation is necessary to ensure sustainability of efforts, commitment and credibility of results.

Acknowledgements

References

1. VWS. *Met zorg kiezen. [Choosing with care].* VWS Publication TK 2000-2001, 27 807 No. 2. The Hague: Netherlands Ministry of Health, Welfare and Sport; 2001. www.rijksoverheid.nl/ministeries/vws

2. van de Bovenkamp H, Grit K, Bal R. *Inventarisatie patiëntenparticipatie in onderzoek, kwaliteit en beleid.* [Inventarisation of patient participation in research, quality and policy]. Rotterdam: Erasmus MC University Medical Center; 2008. www.oldbmg.eur.nl

3. Cargo M, Mercer SL. The value and challenges of participatory research: strengthening its practice. *Annu Rev Public Health.* 2008;29:325-350.

4. Caron-Flinterman JF, Broerse JE, Teerling J, Bunders JF. Patients' priorities concerning health research: the case of asthma and COPD research in the Netherlands. *Health Expect.* 2005;8(3):253-263.

5. Abma TA. Patients as partners in a health research agenda setting: the feasibility of a participatory methodology. *Eval Health Prof.* 2006;29(4):424-439.

6. Hart RA. *Children's participation: from tokenism to citizenship.* Innocenti Essays No 4. Florence: UNICEF; 1992. www.unicef-irc.org/publications/100

7. Litt IF. Research with, not on, adolescents: community-based participatory research. *J Adolesc Health.* 2003;33(5):315-316.

8. Viswanathan M, Ammerman A, Eng E, Garlehner G, Lohr KN, Griffith D, *et al. Community-based participatory research: assessing the evidence.* Summary, Evidence Report/Technology Assessment: No. 99. AHRQ Publication No. 04-E022-1. Rockville (MD): Agency for Health care Research and Quality; 2004. www.ahrq.gov/clinic/epcsums/cbprsum.htm

9. Abma TA, Nierse CJ, Widdershoven GA. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qual Health Res.* 2009;19(3):401-415.

10. Boote J, Telford R, Cooper C. Consumer involvement in health research: a review and research agenda. *Health Policy.* 2002;61(2):213-236.

11. Flicker S. Who benefits from community-based participatory research? A case study of the Positive Youth Project. *Health Educ Behav.* 2008;35(1):70-86.

12. Hart C, Chesson R. Children as consumers. *Br Med J.* 1998; 316(7144):1600-1633.

13. Dixon-Woods M, Young B, Heney D. Partnerships with children. *Br Med J.* 1999;319(7212):778-780.

14. Aynsley-Green A, Barker M, Macfarlane A, Morgan J, Sibert J, Turner T, *et al.* Who is speaking for children and adolescents and for their health at the policy level? *Br Med J.* 2000;321(7255):229-232.

15. Viner RM, Barker M. Young people's health: the need for action. *Br Med J.* 2005;330(7496):901-903.

16. Britto MT, Slap GB, DeVellis RF, Hornung RW, Atherton HD, Knopf JM, *et al.* Specialists understanding of the health care preferences of chronically ill adolescents. *J Adolesc Health*, 2007;40(4):334-341.

17. Alderson P, Sutcliffe K, Curtis K. Children as partners with adults in their medical care. *Arch Dis Child.* 2006;91(4):300-303.

18. Tates K, Meeuwesen L. 'Let mum have her say': turntaking in doctor-parent-child communication. *Patient Educ Couns.* 2000;40(2):151-162.

19. Tates K, Elbers E, Meeuwesen L, Bensing J. Doctor-parent-child relationships: a 'pas de trois'. *Patient Educ Couns.* 2002;48(1):5-14.

20. van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg. [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?].* Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-7905-902-7.

21. Coyne I. Consultation with children in hospital: children, parents' and nurses' perspectives. *J Clin Nurs.* 2006;15(1):61-71.

22. Young B, Dixon-Woods M, Windridge KC, Heney D. Managing communication with young people who have a potentially life threatening chronic illness: qualitative study of patients and parents. *Br Med J.* 2003;326(7384):305.

23. Runeson I, Martenson E, Enskar K. Children's knowledge and degree of participation in decision making when undergoing a clinical diagnostic procedure. *Pediatr Nurs.* 2007;33(6):505-511.
24. Sartain SA, Clarke CL, Heyman R. Hearing the voices of children with chronic illness. *J Adv Nurs.* 2000;32(4):913-921.
25. Hallstrom I, Elander G. Decision-making during hospitalization: parents' and children's involvement. *J Clin Nurs.* 2004;13(3):367-375.
26. Oppong-Odiseng AC, Heycock EG. Adolescent health services--through their eyes. *Arch Dis Child.* 1997;77(2):115-119.
27. Sloper P, Lightfoot J. Involving disabled and chronically ill children and young people in health service development. *Child Care Health Dev.* 2003;29(1):15-20.
28. Cavet J, Sloper P. The participation of children and young people in decisions about UK service development. *Child Care Health Dev.* 2004;30(6):613-621.
29. Curtis K, Liabo K, Roberts H, Barker M. Consulted but not heard: a qualitative study of young people's views of their local health service. *Health Expect.* 2004;7(2):149-156.
30. Lightfoot J, Sloper P. Having a say in health: involving young people with a chronic illness or physical disability in local health services development. *Children & Society.* 2003;17(4):277-290.
31. Stafford A, Laybourn A, Hill M, Walker M. 'Having a say': children and young people talk about consultation. *Children & Society.* 2003;17(5):361-373.
32. Sinclair R. Participation in practice: making it meaningful, effective and sustainable. *Children & Society.* 2004;18(2):106-118.
33. Coad JE, Shaw KL. Is children's choice in health care rhetoric or reality? A scoping review. *J Adv Nurs.* 2008;64(4):318-327.
34. Franklin A, Sloper P. Supporting the participation of disabled children and young people in decision-making. *Children & Society.* 2009;23(1):3-15.
35. Santelli JS, Smith Rogers A, Rosenfeld WD, DuRant RH, Dubler N, Morreale M, *et al*. Guidelines for adolescent health research. A position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 2003;33(5):396-409.
36. Alderson P. Competent children? Minors' consent to health care treatment and research. *Soc Sci Med.* 2007;65(11):2272-2283.
37. Prout A. Researching children as social actors: an introduction to the Children 5-16 Programme. *Children & Society.* 2002;16:67-76.
38. Hill M, Davis J, Prout A, Tiall K. Moving the participation agenda forward. *Children & Society.* 2004;18(2):77-96.
39. Ygge BM, Arnetz JE. Quality of paediatric care: application and validation of an instrument for measuring parent satisfaction with hospital care. *Int J Qual Health Care.* 2001;13(1):33-43.
40. Chesney M, Lindeke L, Johnson L, Jukkala A, Lynch S. Comparison of child and parent satisfaction ratings of ambulatory pediatric subspecialty care. *J Pediatr Health Care.* 2005;19(4):221-229.
41. Battrick C, Glasper EA. The views of children and their families on being in hospital. *Br J Nurs.* 2004;13(6):328-336.
42. Fallon S, Smith J, Morgan S, Stoner M, Austin C. 'Pizza, patients and points of view': involving young people in the design of a post registration module entitled the adolescent with cancer. *Nurse Educ Pract.* 2008; 8(2):140-147.
43. Aldiss S, Horstman M, O'Leary C, Richardson A, Gibson F. What is important to young children who have cancer while in hospital? *Children & Society*. 2009;23(2):85-98.
44. Dedding C, Reis R, Ramaker C, Wolf B. Hoe denken kinderen over hun diabetes. [How do children view their diabetes]. *Ned Tijdschr Diabetol.* 2004;3:86-89.
45. Waller A, Franklin V, Pagliari C, Greene S. Participatory design of a text message scheduling system to support young people with diabetes. *Health Informatics J.* 2006;12(4):304-318.
46. Watson D, Abbott D, Townsley R. Listen to me, too! Lessons from involving children with complex healthcare needs in research about multi-agency services. *Child Care Health Dev.* 2007;33(1):90-95.
47. Jackson AM. 'Follow the fish': involving young people in primary care in Midlothian. *Health Expect.* 2003;6(4):342-351.
48. Moules T. 'They wouldn't know how it feels...': characteristics of quality care from young people's perspectives: a participatory research project. *J Child Health Care*. 2009;13(4):322-332.
49. Coad J, Coad N. Children and young people's preference of thematic design and colour for their hospital environment. *J Child Health Care.* 2008;12(1):33-48.

50. Coad J. Using art-based techniques in engaging children and young people in health care consultations and/or research. *J Res Nurs.* 2007;12(5):487-497.

51. Flicker S, Guta A, Larkin J, Flynn S, Fridkin A, Travers R, *et al*. Survey design from the ground up: collaboratively creating the Toronto Teen Survey. *Health Promot Pract.* 2010;11(1):112-122.

52. Powers JL, Tiffany JS. Engaging youth in participatory research and evaluation. *J Public Health Manag Pract.* 2006;12(Suppl.):S79-87.

53. Harper GW, Carver LJ. "Out-of-the-mainstream" youth as partners in collaborative research: exploring the benefits and challenges. *Health Educ Behav.* 1999;26(2):250-265.

54. Wilson N, Minkler M, Dasho S, Wallerstein N, Martin AC. Getting to social action: the Youth Empowerment Strategies (YES!) Project. *Health Promot Pract.* 2008;9(4):395-403.

55. de Winter M, Baerveldt C, Kooistra J. Enabling children: participation as a new perspective on child-health promotion. *Child Care Health Dev.* 1999;25(1):15-23.

56. Snel MC. Dokter, dit vind ik! Verbeterpunten voor een megacool ziekenhuis [Doctor, this is my opinion! Ideas for a megacool hospital]. *Kind en Ziekenhuis.* 2006;29(1):18-22.

57. van Staa AL, van der Stege HA, Jedeloo S. *Op Eigen Benen Verder. Jongeren met chronische aandoeningen op weg naar zelfstandigheid in de zorg. [On Your Own Feet Ahead. Young people with chronic conditions on their way to independence in health care].* Rotterdam: Hogeschool Rotterdam; 2008. ISBN 978-90-7905-902-7.

58. Birks Y, Sloper P, Lewin R, Parsons J. Exploring health-related experiences of children and young people with congenital heart disease. *Health Expect.* 2007;10(1):16-29.

59. Beresford BA, Sloper P. Chronically ill adolescents experiences of communicating with doctors: a qualitative study. *J Adol Health.* 2003;33(3):172-179.

60. Flicker S, Savan B, Kolenda B, Mildenberger M. A snapshot of community-based research in Canada: Who? What? Why? How? *Health Educ Res.* 2008;23(1):106-114.

61. Knopf JM, Hornung RW, Slap GB, DeVellis RF, Britto MT. Views of treatment decision making from adolescents with chronic illnesses and their parents: a pilot study. *Health Expect.* 2008;11(4):343-354.

62. Trappenburg M. *Genoeg is genoeg. Over gezondheidszorg en democratie. [Enough is enough. About health care and democracy].* Amsterdam: Amsterdam University Press; 2008. ISBN 978-90-5356-815-6.

63. Bate P, Robert G. Experience-based design: from redesigning the system around the patient to codesigning services with the patient. *Qual Saf Health Care.* 2006;15(5):307-310.

2

What we want: chronically ill adolescents' preferences and priorities for improving health care

AnneLoes van Staa, Susan Jedeloo, Heleen van der Stege,
On Your Own Feet Research Group

Abstract

PURPOSE: As important users of health care, adolescents with chronic conditions deserve to be consulted about their experiences and expectations. This study aimed to explore chronically ill adolescents' preferences regarding providers' qualities, outpatient and inpatient care. Furthermore, suggestions for improvement of service delivery were collected.

PATIENTS AND METHODS: This research was a sequential mixed methods study in adolescents aged 12-19 years with various chronic conditions treated in a university children's hospital. Methods comprised 31 face-to-face interviews at home, a hospital-based peer-research project in which 9 adolescents interviewed 34 fellow patients, and a web-based questionnaire (n = 990). Emerging qualitative themes were transformed into questionnaire items.

RESULTS: Having 'a feeling of trust' and 'voice and choice' in the hospital were central to these adolescents. Regarding providers' qualities, 'being an expert' and 'being trustworthy and honest' were ranked highest; followed by 'being caring and understanding'; 'listening and showing respect'; and 'being focused on me'. Regarding outpatient consultations, preferences were ranked as follows: 'answering all questions'; 'attending to my and my parents' needs'; 'clear communication', while 'limited waiting times' and 'attractive outpatient surroundings' scored lowest. Regarding hospitalization, adolescents most preferred to 'avoid pain and discomfort'; 'keep in touch with home'; 'be entertained' while 'being hospitalized with peers' and 'being heard' were least important. Regarding priorities for improvement, 52% of the respondents felt that more attention should be paid to older children, followed by enabling more contact with family and friends (45%), shorter waiting times (43%), and more activities to meet fellow patients (35%).

CONCLUSION: Adolescents prefer technically competent providers, who are honest, trustworthy and attend to their needs. As they gradually grow out of the pediatric environment, they desire staff attitudes to become less childish and more age-appropriate, and welcome being treated as an equal partner in care. Health care professionals should inquire into preferences and adjust their communication style accordingly.

1   Introduction

Children with chronic conditions are frequent users of health care services, yet they are rarely included in the evaluation of such services.[1, 2] Satisfaction with pediatric care is usually only measured in parents,[3-7] whereas children act as silent users of these facilties.[8] However, having parents evaluate children's care may not accurately represent children's views.[9-11] For example, Farrant and Watson found adolescents to be more critical about the received care than their parents, even though they identified the same qualities for good health care providers.[9]

Direct input from children and adolescents on their health care experiences, preferences and priorities is helpful for service evaluation and a prerequisite for improving patient-centeredness of pediatric health care.[12] This is especially relevant for young patients with chronic conditions, who may offer a valuable source of data with which to improve the overall effectiveness of the health care delivery system for adolescents.[12] Making services more responsive to their needs may have a positive effect on their adherence with treatment and appointment.[13]

Reasons for not involving children in the evaluation of services may be related to the idea that they are not interested in this or incompetent.[14] Both assumptions have been contested in studies that have included children as young as 4 or 6 years.[15-17] Children like to be seen as partners in medical care and in the planning, development and evaluation of services.[18, 19] The older they get, the more capable they are to provide rich and detailed descriptions of their preferences.[15]

The first studies of children's preferences for care focused on the factors affecting healthy adolescents' decision to seek preventive care.[20-22] Provider characteristics proved more important than site or system characteristics,[20] and issues of hygiene[21] and confidentiality[22] were of crucial importance. Chronically ill adolescents, on the other hand, may have different needs and experiences, because they have more frequent and often critical health care interactions. Also, these children and their families build long-lasting relationships with (the same) providers, which may affect their evaluation of care.[7]

Several studies (mostly in the USA, UK, or Canada) have explored experiences and preferences of adolescents with chronic conditions in three major domains: communication with health professionals,[9, 12, 23-26] outpatient facilities,[10, 11, 27] and inpatient services.[2, 15, 28-30] Some studies focused on themes related to privacy,[31] or adolescents' decision-making preferences.[32] The various studies applied either qualitative[2, 15, 23-26, 31, 33] or quantitative[9, 10, 12, 30] methodologies. In the Netherlands, an estimated 14% of all children under 18 have a chronic condition.[34] So far, only one study has explored their perspectives on hospital care.[35]

The aim of this paper was to explore the preferences of Dutch chronically ill adolescents (12-19 years of age) for health care professionals and outpatient and inpatient service delivery. We also aimed to collect their suggestions for improvement of adolescent health care.

2 Material and methods

2.1 Study design

In a project entitled 'On Your Own Feet' (2004-2008) we mapped preferences for hospital care and competencies required for self-management in adolescents with chronic conditions. These adolescents were treated in one Dutch university children's hospital. The research team consisted of social scientists, nurse researchers and a doctor. Here, we report on three substudies exploring preferences for health care. Data about competencies and readiness for transfer were published elsewhere.[36,37]

We chose a mixed methods design to account for the complexity of a multi-party context. Mixed methods research is defined as a single study in which qualitative data collection and/or analysis is combined with quantitative data collection and/or analysis.[38] Reasons for applying a mixed methods design were: first, *comprehensiveness*, ie, using different methods to address different aspects of the overall research question will provide a more complete insight. Second, *better validity and generalizability*, by combining qualitative and quantitative methods, inherent weaknesses of each methodology could be compensated for.[38,39] Third, because we wished to give adolescents a voice, *patient-centeredness* was another justification for including qualitative and participatory research.[39] Therefore, in one of the substudies we experimented with a participatory approach, inviting young people with chronic conditions to participate as coresearchers.[40]

Although mixed methods research has become popular in health research, integration of different strands of research is a big methodological challenge as there are no clear procedures for this.[41] To enhance the transparency and quality, we followed the guidelines of O'Cathain *et al* for good reporting of mixed methods studies.[42] We employed a sequential strategy of inquiry,[36] which implies that the studies were conducted in three consecutive steps presented in Figure 1. The qualitative studies preceded and guided the development of the quantitative survey. Design, execution and analysis of each step is presented below.

2.2 Participants and setting

All studies were performed in the Erasmus MC University Medical Center - Sophia Children's Hospital, Rotterdam, the largest tertiary referral center in the Netherlands. This hospital treats over 3500 adolescents with chronic conditions but has no specialized adolescent inpatient facilities and offered, at the time of the study, only a handful of outpatient youth clinics. We enrolled 12-19-year-old patients with somatic chronic conditions who had had at least one consultation in the past six months and had been under continuous treatment for the past three years. Adolescents with intellectual disabilities, and those already transferred to adult care were excluded.

Figure 1 *Overview of mixed methods research on preferences for care from the 'On Your Own Feet' project*

```
┌─────────────────────────────┐      ┌───┬──────────────────────────────────┐
│ Qualitative research        │─────→│ I │ Semistructured interviews with   │
│                             │      │   │ adolescents with chronic         │
│                             │      │   │ conditions                       │
│                             │      │   │ (n = 31; 12-19 yrs) [2004-2005]  │
└─────────────────────────────┘      └───┴──────────────────────────────────┘
           │                                          │
           │                         ┌────┬───────────▼──────────────────────┐
           │                         │ II │ Peer-research with adolescent    │
           │                         │    │ coresearchers                    │
           │                         │    │ (n = 34; 12-19 yrs) [2006]       │
           │                         └────┴──────────────────────────────────┘
           │                                          │
┌──────────▼──────────────────┐      ┌─────┬──────────▼─────────────────────┐
│ Quantitative research       │      │ III │ Web-based questionnaire in     │
│                             │      │     │ adolescents with chronic       │
│                             │      │     │ conditions                     │
│                             │      │     │ (n = 990; 12-19 yrs) [2006-2007]│
└─────────────────────────────┘      └─────┴────────────────────────────────┘
```
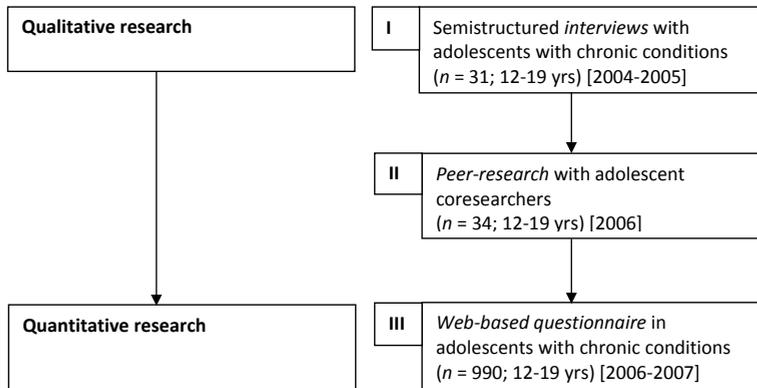
2.3   Qualitative research

2.3.1   Interviews

Two age groups were created, ie, younger adolescents aged 12-15 years and older adolescents aged 16-19 years. Equal numbers of random cases in both groups were drawn from the hospital database to facilitate purposive sampling.[38] Next, we aimed at sampling equal numbers of participants, even distributions of gender, hospital experience, and nature of the condition (ie, congenital or recently acquired, physically disabling or not) within both age groups. Our intent was to interview at least 15 adolescents in each age group.

Pairs of purpose-trained nursing and paramedical students or a researcher (SJ) conducted the interviews in the participants' homes. The semistructured interviews collected background information about their condition, their understanding of the condition; its impact on their day-to-day lives; and assessed their preferences for service delivery. This included asking about their experiences with hospital staff, their attributes of a good doctor, what they liked and disliked about hospital consultations, and their experiences with hospital admissions. Interviews lasted between 45-90 minutes, were audiotape recorded and transcribed verbatim. The data were analyzed in combination with those from the peer-research.

2.3.2   Peer-research

For the participatory research project, adolescents were sought who were both motivated to act as coresearchers and willing to give their opinion about the care they received. As a hospital is not

an ideal setting for community-based research, we organized a disco party in a real discotheque during which the coresearchers were to interview their fellow patients. A representative sample of adolescents was not intended. More information on the design and execution of the peer-research study is provided elsewhere.[40]

The nine coresearchers (all over 15 years of age) were recruited by nurse specialists in the hospital. They were briefly trained in interview techniques and we discussed the themes and topics to be included in the interview protocol with them. The research team presented some open-ended questions inspired by the previous interview study and by a child-friendly questionnaire designed and tested in another Dutch pediatric hospital.[35] The coresearchers rephrased the questions where needed and added new topics. The interview protocol was finalized in several discussion rounds with the coresearchers through e-mail. The questions related to adolescents' care experiences and preferences (Chapter 1; Box 1).[40] Data on socio-demographic characteristics were also collected.

Although we intended to involve the coresearchers in the data-analysis, setting up a "real-life" meeting proved impossible. Instead, the research team analyzed the anonymous transcripts and invited coresearchers to comment on draft versions of the report through e-mail.

2.3.3   Combined qualitative analysis

Because the topics discussed during the interviews and the peer-research project were very similar, all qualitative data were analyzed together, using the qualitative software package ATLAS.ti 5.5. Thematic analysis was chosen for its flexibility and theoretical freedom, and was applied in several phases.[43] As a first step, AvS and SJ read the interviews repeatedly to familiarize themselves with the data. They independently formulated initial codes (subthemes). Together, the researchers examined codes and reached consensus on the initial codes. Subsequently, these were modified, expanded or merged as new issues emerged. The third step was collating subthemes to identify potential themes; emerging themes were checked iteratively in other interviews. Possible relationships between patients' preferences and relevant demographic characteristics were identified. The research team examined the coding process, and the emerging themes were discussed continually until consensus was reached.

2.4   Quantitative research

2.4.1   Questionnaire

All adolescents who met the our aforementioned inclusion criteria stated on 1st July 2006 (n = 3648) were invited by letter to complete an online questionnaire accessible with a unique code on a secured Internet site. Response postcards were included in the invitation letter to encourage adolescents to state reasons for refusal. All received a written reminder after three weeks.

The questionnaire measured socio-demographic characteristics, disease-related and health care-related variables, competencies, and preferences for care. Data on gender, age, ethnicity, and hospital visits (outpatient departments; admissions) were retrieved from the electronic hospital database; all other data were self-reported. The questionnaire had been pilot-tested in face-to-

face interviews with five chronically ill adolescents and four parents. Information on the measures has been published elsewhere.[37]

Because there were no short and reliable measures available to assess preferences for health care providers, outpatient consultations, and inpatient care, we transformed the final themes of the qualitative studies into questionnaire items. Adolescents were asked to indicate what they considered to be the most important quality for a good doctor or nurse, and what is most important to them when coming to an outpatient appointment, and during hospitalization. Respondents were invited to rank-order five statements related to each topic from 1-5 (number 1 being the most important quality or issue and number 5 the least important).

Furthermore, respondents were asked to mark three out of nine items as priorities for improvement in the Sophia Children's Hospital. This list was adapted from a 7-item *"improvement indicator"* that had been constructed after consultation with 225 children between 7-16 years in another Dutch children's hospital.[35] Because half of their population was below 12 years of age, we added two items that had proved relevant in the peer-research project: *"I wish they would pay more attention to the needs of older children"* and "*I wish I could do more things with fellow patients".*

2.4.2   Quantitative analysis

Questionnaire data were analyzed using SPSS 17.0 (SPSS Inc, Chicago, IL). Descriptive statistics were applied. Ratings for preferences for providers, outpatient visits and hospital admissions were recoded (the most important preference receiving 5 points and the lowest 1 point) and then summated. Analysis of variance was used to test differences between the means on preference scores and priority listing between boys and girls, between older and younger adolescents, between higher and lower educated adolescents, those who visited the hospital more than four times a year versus those who came less frequently; and those who had, or had not been hospitalized in the past three years.

2.5   Validation and integration

Validation for the findings was primarily realized by method triangulation and peer-review. For example, preliminary analyses of interviews and peer-research were discussed within the research group, with the coresearchers, and with health care providers (data reported elsewhere).[36] *Integration* of the findings of the different study parts occurred at two stages. First, to establish the additional value of the participatory approach, results from the peer-interviews were compared to those from the home interviews.[40] All qualitative findings were thematically summarized in a popular book.[44] The qualitative findings also provided direct input for questionnaire development by transforming qualitative themes into questionnaire items. Second, for the present paper interpretations from the qualitative studies were systematically compared with the quantitative findings.

2.6   Ethics

All study procedures were approved by the Institutional Review Board of the Erasmus MC University Medical Center. The researchers had no access to participants' medical records. Participants were assured of confidentiality and data were processed anonymously. Eligible adolescents and their parents received written information about the study and gave informed consent. The interviewees were rewarded with a €20 gift voucher. The coresearchers involved in the peer-research received €75 remuneration for their input, while participants attended the disco party for free. Adolescents who completed the questionnaire were entered in a lottery for 2 iPods and a cell phone.

3   Results

3.1   Response

Characteristics and medical diagnoses of the participants in the substudies are presented in Table 1.

3.1.1   Interviews

Thirty-one of the 66 invited adolescents (47%) consented to an interview. Data on responders and nonresponders have been published elsewhere, revealing no significant differences in the selected variables for purposive sampling.[45] Five adolescents presented with surgical conditions, two were physically disabled, and the others had a variety of chronic illnesses.

3.1.2   Peer-research

Twenty-five adolescents, predominantly younger girls (12-15 years), attended the disco party. The nine coresearchers also interviewed each other, adding up to a total of 34 peer-interviews. Compared with the visitors, the coresearchers were older ($P < .05$) and they visited the hospital more frequently ($P < .01$).

3.1.3   Questionnaire

The study population for the questionnaire consisted of 3648 adolescents. Of the 1087 questionnaires received (response rate 29.8%), 97 were excluded because they were incomplete, leaving a total of 990 valid questionnaires. An analysis of response and nonresponse has been published elsewhere.[37] Nonresponders tended to be males with non Dutch surnames; in addition, they were older and had fewer consultations than responders ($P < .05$). Table 1 presents participants' characteristics. All major chronic conditions were represented, the majority suffered from life-long chronic conditions.

Table 1 *Adolescents' socio-demographic and disease-related characteristics in preference studies in the research project 'On Your Own Feet'*

	Interviews n = 31[a]		Peer-research n = 34[b]		Questionnaire n = 990[c]	
	n	*%*	*n*	*%*	*n*	*%*
Gender						
girls	15	48.4	23	67.6	560	56.6
boys	16	51.6	11	32.4	430	43.4
Age						
12-15 yrs	17	54.8	23	67.6	608	61.4
16-19 yrs	14	45.2	11	32.4	382	38.6
mean (SD)	*15.3 (2.1)*		*14.4 (1.7)*		*15.0 (1.9)*	
Ethnicity						
Dutch surname	26	83.9			851	86.0
non Dutch surname	5	16.1			139	14.0
missing data			34			
Educational level						
lower / middle	17	68.0			525	55.7
higher	8	32.0			417	44.3
missing data	6		34		48	
No. of outpatient visits in past three years						
≤ 12	19	61.3	22	64.7	501	50.6
≥ 13	12	38.7	12	35.3	489	49.4
mean (SD)					*17.2 (16.3)*	
Hospital admissions in past three years						
yes	12	38.7	30	88.2	238	24.0
no	19	61.3	4	11.8	752	76.0
Age at diagnosis						
at birth and during first 5 years	22	71.0	14	41.2	684	69.2
after 6 years of age	9	29.0	20	58.8	304	30.8
missing data					2	
Prescribed medications, diet or exercise						
yes	24	77.4			623	62.9
no	7	22.6			367	37.1
missing data			34			
Presence of physical limitations						
yes	11	35.5	5	16.7	285	28.8
no	20	64.5	25	83.3	705	71.2
General health score (range 1-5) *mean (SD)*	*3.0 (0.7)*				*3.6 (1.0)*	
missing data	2		34			

Notes:

[a] *Diagnoses represented in the interviews: scoliosis/kyphosis, facial schisis, benign intracranial hypertension, congenital bladder disorder, congenital heart disorders, rheumatoid arthritis, hemophilia, diabetes mellitus (DM), epilepsy, inflammatory bowel disease (IBD), cystic fibrosis (CF), various metabolic disorders, HIV, nephrotic syndrome, immune and hormone deficiencies, lung insufficiency, progressive kidney failure, congenital skin diseases, asthma, and neuromuscular diseases.*

[b] *Diagnoses represented in the peer-research study: congenital heart disorders (3), rheumatoid arthritis (3), hemophilia (3), diabetes mellitus (4), inflammatory bowel disease (4), end-stage renal insufficiency (8), skin diseases (2), neuromuscular diseases (3), cancer (1), various congenital conditions (2), unknown (2).*

[c] *In the questionnaire, the five largest diagnostic categories were (ICD-9 classification): congenital anomalies and conditions originating in the perinatal period (26.0%); neoplasm (13.0%); endocrine, nutritional, metabolic diseases, and immunity disorders (12.0%); diseases of the nervous system and sense organs (11.6%); and diseases of the musculoskeletal system and connective tissue (9.9%).*

3.2 Adolescents' preferences

Below, we first present the qualitative themes (summarized in Box 1) in each domain of care-related preferences, followed by the results from the questionnaire items developed from them.

Box 1   *Qualitative themes and subthemes related to preferences for health care providers, outpatient consultations and hospital care, and overall quality*

Preferences for health care providers
- Being trustworthy and honest
- Being caring and understanding
- Listening and showing respect
  - *Being taken seriously*
- Focusing on me
  - *Being treated as an adult*
- Being competent

Preferences for outpatient consultations
- Answering all my questions
- Attention to my and my parents' needs
- Clear and concise communication
- Limited waiting times
- Attractive outpatient surroundings

Preferences during hospital admissions
- Avoiding pain and discomfort
- Keeping in touch with home
- Being entertained
- Hospitalization with peers
- Being heard

Central themes related to quality
- Having a feeling of trust
- Having voice and choice

3.2.1   Preferences for interactions with providers

• Qualitative results

Adolescents regarded health care professionals as the most valuable asset of the hospital. Five themes emerged, ie, being trustworthy and honest, being caring and understanding, listening and showing respect, focusing on me, and being competent. One of the coresearchers summarized these attributes as follows: *"A good doctor is someone who is child-friendly. Someone who consults with you and with your parents; who doesn't treat you like a toddler. He doesn't need to tell you that you're ill, that's obvious. Furthermore, he or she should be able to listen well and to solve your problems"* (16-year-old girl).

The adolescents wanted a doctor who is trustworthy, ie, one whom they trust and who trusts them. This is why they preferred continuity in providers: *"there is some sort of trust that you feel with this doctor and not with someone else"* (16-year-old boy). It is also a matter of convenience: *"Having to explain everything every time is rather irritating"* (13-year-old girl). However, it is predominantly related to confidentiality: *"My own doctor knows all about me and that is confidential information"* (16-year-old girl). A 17-year-old boy saw his familiar providers as *"… more than just interested, you have the feeling you can tell them everything. They are not just passers-by, but confidantes".* It is vital that health care providers keep this information confidential. It also takes time to build a trusted relationship, and it requires *"getting to know each other".* The concept of trust seems closely intertwined with that of provider honesty. Therefore, we combined 'trust and honesty' in one theme. Many adolescents claimed to prefer honesty even when it implies being given unpleasant information. A 19-year-old boy thought: *"They should tell you what's going on and tell you straight in the face what the consequences of your behavior are, because that's in your best interest."* Mainly older adolescents did not fear confrontations: *"It startles me when they confront me. That helps me to correct my behavior; I guess I need that once in a while"* (18-year-old girl). Although not all adolescents wanted to be confronted directly with the consequences of sloppy adherence, they all felt that doctors should tell honestly *"what's up and what they're going to do"* (14-year-old girl). Withholding information or being overprotective is considered to be *"childish".*

A caring and understanding attitude was also much appreciated. Doctors and nurses should be *"kind, patient, and understanding"* (14-year-old boy). They should not treat adolescents *"like a number"* (15-year-old boy). Questions about their social life were appreciated: *"I like it when doctors and nurses take interest in the things I do in my free time"* (17-year-old girl). Some liked

jokes or small talk from doctors. This could even be useful, suggested a 16-year-old boy: *"Children should be made to feel at ease, so they don't withhold information".* But others wanted a doctor to be "*serious, taking his time"* (12-year-old girl). A 15-year-old girl recommended that health care staff should *"try to think from the patient's perspective: how would you like to be treated? Ask them if they can cope!"*

Adolescents wanted doctors to listen to them and consider their opinion. *"If I don't like some-thing, they should respect that"* (17-year-old girl). Not considering their opinion was experienced as a breach of confidence: "*Just one time the doctor did not take my opinion into account and then I got very upset"* (12-year-old girl)*.* In contrast, being given a choice in treatment options is appreciated: *"My doctor usually says that she only gives advice but that I have to decide for myself. That's fine."* (12-year-old girl).

Adolescents preferred health care professionals to focus on them rather than on their parents. Some complained that their parents were asked for information, rather than them themselves. A 16-years-old girl wished "… *they would talk more to the children and ask their permission if they want to change treatment. For example, they could ask children whether they would like to talk to the doctor alone. That would give you the opportunity to share things you would otherwise never tell."* Doctors *"should really talk to me and codecide with me"* thought a 16-year-old girl. Being focused on the adolescent patient was often related to a preference for being treated like an adult: *"They should treat teenagers in a less childish way, according to their age"* (15-year-old girl)*.* A 12-year-old girl complained: *"The doctor always says: "so, you have grown a lot" – that is so childish! They never talk to me in an adult way, like my parents talk to me. I don't like that. I wish they wouldn't treat me like a small child".* Only a minority of the adolescents, especially those who labeled themselves as *"… still being a child"* (12-year-old boy), did not prefer an adult approach, for example because *"… it would be too difficult for me"* (15-year-old girl). But a 14-year-old girl said that, even though she couldn't handle everything the way grownups do, she still wanted to be treated in an adult way.

Competence and professional expertise were valued highly. This was defined in terms of knowl-edge, attitude and practical skills*: "A good doctor is someone who knows exactly what he's doing and also takes into account that children find jabs very scary"* (12-year-old girl). It is *"someone who helps you and always tries to make you better and doesn't give up"* (14-year-old girl). Professionals should have the skill to explain matters in a way children can understand: *"A doctor should be able to explain everything very well, what it means and what you can expect"* (16-year-old girl).

•**Quantitative results**
The five themes were transformed into items. Table 2 presents the mean scores, standard devia-tions and priority listing (number of times this item was selected as being most important) of the questions on preferences for provider qualities. The most important attribution for a good doctor or nurse was being an expert and knowing what (s)he's doing, while issues related to honesty and trustworthiness came in second place. Significant differences were found between the genders and age groups. Boys attached more importance to providers' expertise (F[1, 986] = 5.48; P = .02) and trustworthiness and honesty (F[1, 986] = 7.24; $P < .01$) than did girls; girls rated the im-portance of careful listening (F [1, 986] = 5.53; P = .02) and being kind (F [1, 986] = 9.06; $P < .01$)

higher than did boys. Younger adolescents found providers' kindness and ability to reassure them more important than did older adolescents (F([1, 986] = 13.36; $P < .001$). The latter found it more important that health care providers are focused on them and consider their opinion (F[1, 986] = 8.54; $P < .01$).

Table 2   *Top 5: most important qualities of health care providers and most important issues related to hospital consultations and inpatient care (n = 988)*

Make your own top 5	mean (SD)[a]	% number 1[b]
I) What is the most important quality of a good doctor or nurse? (S)he should…		
be an expert and know what (s)he's doing	3.5 (1.5)	41.3
be trustworthy and honest	3.2 (1.3)	18.9
be kind and able to reassure me	3.0 (1.4)	18.1
be able to listen carefully and be patient	2.7 (1.2)	9.7
be focused on me and consider my opinion	2.5 (1.4)	11.9
II) What is most important to you when you have an appointment in the outpatient department?		
that all my questions are answered	3.7 (1.3)	35.4
that I don't have to wait too long	3.5 (1.3)	29.4
that I and my parents get all the time and attention we need	3.4 (1.2)	21.3
that the consultation does not take too long	2.3 (1.2)	5.9
that there is sufficient distraction in the waiting room	2.0 (1.3)	8.1
III) What is most important to you when you're hospitalized?		
that I don't have too much pain and other discomfort	3.7 (1.3)	38.2
that I can keep in touch with my friends and family	3.4 (1.4)	31.6
that health care providers listen to me	3.0 (1.2)	10.9
that there is sufficient distraction	2.6 (1.3)	9.8
that I am hospitalized together with peers / other adolescents	2.3 (1.4)	9.5

Notes:

[a] *Participants rank-ordered five statements related to each topic: number 1 was the most important quality or issue and received 5 points; the least important quality or issue received 1 point. Range for the means is 1-5 (5 being most important).*

[b] *Percentage of adolescents who rated this statement as most important (rated as number 1).*

3.2.2   Preferences for outpatient consultations

•**Qualitative results**

Five themes relating to effective and efficient consultations emerged from the thematic analysis, ie, answering all my questions, attending to my and my parents' needs, clear and concise communication, short waiting times, and attractive outpatient surroundings.

Adolescents wanted consultations to be meaningful and helpful, ensuring that all their questions are being answered: *"The doctor must listen to what is being said. He should answer all your questions without using difficult words"* (15-year-old boy). Doctors should *"talk steadily, not too fast"* (12-year-old girl). Some adolescents found consultations *"... boring, always the same".* Perhaps this is because they feel left out of the conversation: *"With all these difficult words, I don't understand what's going on and that is boring"* (12-year-old boy).

Both their own and their parents' needs should be attended to: "*They should explain things to me and to my parents as well. So that they too understand"* (17-year-old girl). Their parents' presence is important for many adolescents, especially the younger ones: "*It is convenient that my parents are there because I do not always understand everything"* (14-year-old boy). Still, many feel it would be a good idea to see the doctor alone sometimes: *"Certain things I can't discuss with my parents"* (16-year-old boy). In any case: *"Doctors should not only address my parents, but me in the first place"* (15-year-old girl).

Adolescents preferred clear and concise communication: *"They should come to the point straight away and not tell you nice stories"* (17-year-old girl). A 16-year-old boy was irritated by his doctor beating around the bush: *"If you ask my doctor a question, he spins a tale around it, but doesn't give an answer!"* Another 15-year-old boy was most concerned with the consultation *"going quick and smooth, without long silences".* Clear communication also implied avoiding jargon or difficult words.

Short waiting times for outpatient consultations were also thought to be important. Many complained: *"I wished I wouldn't have to wait so long!"* (16-year-old boy). Inefficient planning also bothered them: *"Consultations should be planned in one day, after each other"* (17-year-old girl).

Attractive outpatient surroundings also mattered: *"It should be cozy and pleasant"* (18-year-old girl). The waiting room should offer distraction and be an age-appropriate environment that is not too noisy: *"It should be quiet in the waiting area, so you won't get stressed out and can concentrate yourself"* (16-year-old girl). Unfortunately, this was not always the case as adolescents complained about *"... screaming young kids scooting about on tricycles".*

•**Quantitative results**

Table 2 presents the priority listing of preferences for outpatient visits. All questions being attended to and appointments starting on time were ranked as most important. Least importance was given to the waiting room environment. Only educational level accounted for differences in priorities: the higher educated found it more important that their questions were being answered (F[1, 939] = 9.47; P < .01), whereas the lower educated attached more importance to distraction in the waiting room area (F[1, 939] = 5.13; P = .02).

3.2.3   Preferences for hospital admissions

•**Qualitative results**

Not all interviewed adolescents had been hospitalized. Five themes associated with inpatient care emerged, ie, avoiding pain and discomfort, keeping in touch with home, being entertained hospitalization with peers, and being heard.

Avoiding pain and discomfort is important because hospital admission is associated with pain and unpleasant invasive procedures. Other associations included being locked up, being bored and being alone. Adolescents missed their family, friends, and animal pets.

Therefore, keeping in touch with family and friends during hospitalization was important to them and availability of a laptop with an Internet connection was highly appreciated for that reason.

Being entertained and being offered distraction (game computers and recreational activities) was also mentioned regularly: *"You should be entertained so you won't think of your illness all the time"* (18-year-old girl). These activities were not always age-appropriate. *"I would like more activities for older children – they are a little boring now, more for small kids"* (12-year-old girl). Also: "*the hospital clowns are nice for younger kids, but they should ask you if you want to be entertained by them. When you get older, you really do not have a need for that sort of entertainment anymore"* (17-year-old girl). While younger adolescents thought the clowns were *"… very funny: they cheered everything up"* (12-year-old girl), a 16-year-old girl thought they were "*stupid and boring. I am too old for that kind of humor. But it's good that they are there for the kids".*

Those with extensive hospital experience complained about being with younger children on the ward and favored being hospitalized with peers. Meeting fellow patients through activities offered by the hospital was important to about half of the attendants of the disco party, whereas the adolescents interviewed at home seemed less interested in meeting fellow patients. A 16-year-old-girl at the disco party said: *"That's very important. You can support each other in difficult times, exchange advice and hear stories about how others experience things"*. Suggested ways to meet fellow patients were chat rooms, group sessions, and activities outside the hospital. Not all adolescents were interested in such activities: *"I don't feel like there's something wrong with me, so I prefer to hang out with people who have nothing wrong with them"* (15-year-old-girl) and a 15-year-old boy thought he would *"go crazy"* if he would have to talk about hemophilia *"all the time"*.

Being heard and being empowered to participate in decisions formed the last theme. Some adolescents wanted more involvement in treatment decisions: *"I wish they would ask me more often to say what I really want. If only you should get the opportunity"* (14-year-old boy). On the other hand, not all participants felt the need to have a greater say in hospital matters because *"they already listen to you and I do not have any bad experiences"*. However, some did, especially the coresearchers: *"It's a children's hospital, so it should be child-friendly. Who can better judge whether it's a good or bad hospital than children themselves?"* (15-year-old girl). She suggested to form a youth council that could advice the hospital board how to further improve services.

•**Quantitative results**

It was most important to adolescents that they suffer the least possible pain and discomfort when being hospitalized (Table 2). Keeping contact with family and friends was also highly valued; being hospitalized with peers was least important. However, girls found the latter more important than did boys (F[1, 986] = 4.91; P < .03), whereas boys rated distraction as more important than did girls (F[1, 986] = 36.88; P < .001). Keeping in touch with family and friends was more important for younger adolescents (12-15 years) than it was for the older ones (F[1, 986] = 4.99; P < = .03). The latter rated the importance of staff listening higher than the younger group (F[1, 986] = 18.07; P < .001).

Those admitted to the hospital in the past three years attached more importance to experiencing the least possible pain and discomfort (F[1, 986]) = 6.41; P = .01) and staff listening to them than did those who had no inpatient experience (F[1, 986] = 4.16; P < .05). The latter rated maintaining contact with family and friends as more important than did the ones with inpatient experience (F[1, 986] = 7.75; P < .01).

3.2.4   Quality of care and priorities for change

•**Qualitative results**

The adolescents were very positive about the quality of care provided in the children's hospital. They pointed at the warm atmosphere, the caring attitude of the staff, and the child-friendly facilities: *"It is really a place for children. They help you and it's very beautiful there"* (13-year-old girl). *"They are really focused on children. They do their best to make your visit as pleasant as possible. I love the colors in the central hall. The hospital gives me a feeling of trust"* (15-year-old girl). *"The nurses are really nice. They have good computers there"* (12-year-old-girl). *"There are qualified doctors who listen to me"* (17-year-old-girl).

Having a feeling of trust was a central theme in the interviews and peer-research. The fact is that many have come here often from a very young age: "*The doctors are very nice, the building is nice and I've been coming here all my life, so it is all familiar"* (17-year-old girl). To the question what should be improved, quite a few adolescents responded *"nothing; everything is OK"*. However, others identified several areas for improvement, ie, lack of involvement in treatment decisions, staff attitude toward teenagers, and the lack of adolescent-focused services and facilities. Therefore, the other central theme related to the quality of adolescent care was having voice and choice.

•**Quantitative results**

Figure 2 displays adolescents' *priorities for improvement* based on the *'improvement indicator'*. The item selected most often (by 52.2% of all adolescents) was paying more attention to the needs of older children. Having more contact with family and friends through the Internet (44.6%), shorter waiting times (43.3%), and more activities with fellow patients (34.5%) came next. Only 14.6% of all adolescents wished that doctors and nurses would listen to them more often.

Here, we also tested for differences between those who did and did not select an item as priority for improvement. Girls more often than boys mentioned that more attention should be paid to

Figure 2   *Priorities for improvement in the children's hospital: percentage of adolescents that selected this item in the survey (n = 990)*

I wish...
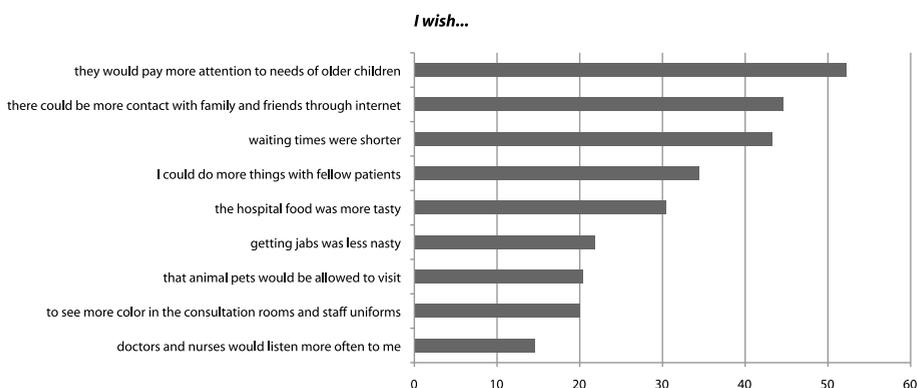
older children (F[1, 988] = 10.97; *P* = .001) and that there should be more color in the hospital (F[1, 988] = 10.37; *P* = .001). Boys more often than girls mention that the hospital food should be tastier (F[1, 988] = 4.54; *P* = .03) and that waiting times should be shorter (F[1, 988] = 5.78; *P* = .02). More of the younger ones wished that pets would be allowed to visit in the hospital (F[1, 988] = 8.17; *P* < .01); that more contact with family and friends through the Internet would be possible (F[1, 988] = 6.11; *P* = .01); and that getting jabs would be less unpleasant (F[1, 988] = 14.58; *P* < .001). The older ones were more concerned about the hospital paying greater attention to older children (F[1, 988] = 20.45; *P* < .001). For those with a lower education, having more activities with fellow patients (F[1, 940] = 9.98; *P* < .01) and having more contact with family and friends while in hospital (F[1, 940] = 3.95; *P* < .05) were higher priorities than for those with higher education. In reverse, the more highly educated adolescents were more concerned about shorter waiting times in the hospital (F[1, 940] = 7.25; *P* < .01). Finally, those who have been hospitalized in the past three years were more keen on having tastier hospital food (F[1, 988] = 8.88; *P* < .01), while shorter waiting times were more important to those without inpatient experience (F[1, 988] = 6.82; *P* < .01).

4   Discussion

This mixed methods study explored chronically ill adolescents' preferences for health care providers' attributes and for service delivery in outpatient clinics and during hospitalization in a children's hospital. Their suggestions for improvement of care delivery were also collected. These adolescents were generally very satisfied with the care provided and felt at home in the children's hospital, but they recommended paying more attention to the needs of older children and improving the age-appropriateness of providers' attitudes and services. Technical competence and good communication skills and attitudes were regarded as important qualities of health care providers. The adolescents were concerned about having their questions and needs attended to in outpa-

tient consultations as well as being seen on time. Having as little pain or discomfort as possible, as well as maintaining contact with family and friends, were considered most important during hospitalization. Environmental aspects were rated as less important.

4.1   Preferences

The themes we uncovered and those from other qualitative studies exploring adolescents' preferences for (communication with) providers share many similarities (Table 3). Honesty, respect, a caring and friendly attitude, being focused on adolescents, and technical competence are all important.[9, 23, 26, 33, 46] Trustworthiness seems a core attribute for professionals and is related to good communication skills and respect for teenagers' opinion and privacy.[20, 25, 31] Other studies have confirmed that adolescents favor direct communication with them (and not with their parents) and dislike being patronized or being approached in a condition-centered manner.[26, 47] In our study, the importance of receiving explanations and information did not emerge as a separate theme. We placed this under the theme 'being competent'. Receiving appropriate information during consultations was seen as very important. In contrast with some other studies, our respondents did not mention preferring a provider of the same gender.[9, 26, 46] Some mentioned a preference for continuity in providers in relation to trust, but this did not emerge as a separate theme.[26, 46]

The qualitative findings were reinforced by the survey outcomes, both in our study and among chronically ill adolescents in the US, who rated the honesty of their physician, attention to pain, and items related to respect as very important, while technical aspects of care were also highly appreciated.[12] Our study adds that professional expertise is most important (41% indicated this as their number 1 concern), and honesty and kindness of the provider came in second and third place, respectively. An important theme in the qualitative studies, ie, providers 'being focused on me', proved to be less important than the other qualities in our survey.

For outpatient facilities, our findings correspond with those of other studies that also indicated the importance of good explanations and having a choice,[33] as well as of more efficient services and reduced waiting times.[27, 47] A study by Wray and Maynard also demonstrated that both streamlined care processes in the outpatient department and care interactions are important to young people with congenital heart conditions who move to adult services.[47] Making the outpatient area more teen-oriented and less child-centered[27] was also a wish of our respondents, but they did not give it much priority.

With respect to clinic environment and inpatient care, our qualitative studies confirm the findings of others, ie, the interior design should be less child-centered but more teen-oriented, and feel more like home.[15, 27, 29] Adolescent inpatients would also appreciate more fun and distraction, more comfort, and attention to privacy.[15] Although few adolescents in our study complained of unsympathetic hospital staff,[30] friendliness is important to them. However, in our survey, they indicated that minimizing pain and discomfort was the most important aspect to them when hospitalized. Although our participants stressed the importance of being consulted and involved in their hospital care, as in the study by Coyne,[2] they did not give this a high priority in our survey. Only 15% indicated they wished doctors and nurses would listen to them more often. Some adolescents, especially those involved in the peer-research, would appreciate being hospitalized with

Table 3   *Comparison of themes in qualitative studies into adolescents' preferences for their interactions with health care professionals*

OUR STUDY	Review by Freake et al [46]	Woodgate [23]	Beresford and Sloper [26]	Farrant and Watson [9]	Klostermann et al [25]	Moules33
Being trustworthy and honest	"What I tell them is confidential" "I can trust them"		Behavior toward adolescent	Confidentiality	Confidential information / Honesty	Respect
	"They explain things and give me information and advice"	Give me some encouragement				Explanations
Listen and respect (being taken seriously)	"They listen to me" "They are nonjudgmental"	Don't force me		Good listener		Respect / choice
Being caring and understanding	"They are kind, caring, sympathetic, understanding"	Try to understand	Communication skills		Holistic trust	Friendly staff
	"I feel comfortable, and it is easy to talk"	Have a sense of humor				
Being competent	"They are competent, experienced and qualified"	Know what you're doing		Medical knowledge	Competence	Technical expertise
Focusing on me / (being treated as an adult)	"They do not patronize me or treat me like a child"	Don't treat me differently	Status issues			
	"I am treated as an individual, not just part of their job"	Treat me as a person	Condition-centered vs. person-centered		Fidelity	
	"For medical issues many girls prefer to see a female doctor"		Gender preferences	Same gender		
	"I get to see the same person each time"		Familiarity (continuity of person)			

peers,[30] or be offered activities where they would meet fellow patients (35% of survey participants indicated this was an area for improvement). An interesting, but unexplained association, was found between lower level of education and a stronger preference for meeting fellow patients. Our participants, however, did not voice a strong need for a dedicated adolescent unit.[48]

4.2   Differences between adolescents

Not all young people have the same preferences.[45] Our study confirmed some differences related to gender, age, inpatient experience, and educational level. Boys attached more importance to professional expertise and honesty of providers than did girls, who wanted more attention to older children and rated listening as a more important provider quality. We cannot explain these differences, and they have not been reported before. A large survey of adolescent preferences found only one significant association for gender: girls viewed the 'power/control' factor as more important than did boys.[12] In that survey, higher age was strongly associated with a preference for communication directly with the teen versus the parent,[12] a finding confirmed in our survey, in which older adolescents had a stronger preference for staff being focused on them and listening to them than did the younger ones. Younger adolescents in our study were more concerned about staff kindness, pets' visits and the discomfort of painful procedures, like the participants in another Dutch hospital.[35] For adolescents with inpatient hospital experience, the quality of hospital food and staff listening to them was more important than for those who had not been admitted before, indicating that both a higher age and more hospital experience raises adolescents' desire to be involved in decision making.

4.3   Mixed methods

Combining qualitative and quantitative research methods in one study proved successful. In two domains, the quantitative findings confirmed the interpretations from the interviews and the findings could also be explained from them, strengthening the validity and generalizability of the results. Still, the qualitative data suggested that communication issues were more important than issues related to professional expertise. This was not confirmed in the survey. Being listened to was even given lowest priority for improvement, with all other issues being considered more important. These findings seem contradictory, because adolescents in the interviews clearly indicated they liked to be consulted and wished to be involved in their own care. Perhaps this may be explained by adolescents' assumption that *"paying more attention to the needs of older children"* (listed as top priority) also encompasses their preference to be seen as a partner in care. For example, in the interviews the older adolescents highly valued *"being treated as an adult"*, which indicates a preference for direct communication, as well as for more voice and choice.[12]

There were few differences between the preferences reported in the face-to-face interviews at the adolescents' homes and in the peer-research interviews during the disco party. The most notable difference was that in the peer-research, adolescents were more convinced of the importance of meeting fellow patients and of being enabled to participate in hospital matters. Because the peer-interviews lacked depth and did not yield substantial new insights, we realized that the

participatory approach has its drawbacks. Adolescents with chronic conditions enjoy having a voice in the design and evaluation of health care services. However, the desirable extent of patient partnership (from patient perspective and in research) remains undefined.[40]

4.4   Limitations of the study

Although the mixed methods approach may be seen as a strength, our study also had some weaknesses. The research was carried out in one university hospital in the Netherlands and results may thus not apply to other settings and countries. Nevertheless, there are many similarities between countries in the reported care preferences of adolescents with chronic conditions.

We researched wide ranges of health conditions and preferences, because all chronic patients have many tasks and challenges in common.[49] As a logical consequence, differences in experiences and preferences related to the chronic conditions themselves cannot be accounted for. Also, the nonresponse rate was fairly high for both the interview study and the questionnaire, while only a small number of adolescents attended the disco party, implying that the results may not be representative for the total population. As girls and those with more extensive hospital experience were over-represented in the survey and among the peer-research participants, this may have affected the outcomes.

5   Conclusion

Young people with chronic conditions are able and willing to express their views on the perceived quality of health care services provided to them. They have a strong preference for providers who are technically competent, honest and straightforward. The older they are, the more concerned they are about providers focusing on them rather than their parents and treating them like an adult. Adolescents grow out of pediatric care and they wish that the pediatric environment and staff attitudes would be less child-centered and more age-appropriate. Different needs according to gender, age, and educational level should also be acknowledged. Health care professionals should be aware of preferences, inquire into them and adjust their communication style accordingly. This may strengthen adolescents' competencies on their road to adulthood and help build positive, trusting relations between professionals and their adolescent patients, which is a prerequisite of shared responsibility for treatment.

Acknowledgments

References

1. Hart C, Chesson R. Children as consumers. *Br Med J.* 1998;316(7144):1600-1603.
2. Coyne I. Consultation with children in hospital: children, parents' and nurses' perspectives. *J Clin Nurs.* 2006;15(1):61-71.
3. Homer CJ, Marino B, Cleary PD, Alpert HR, Smith B, Crowley Ganser CM, *et al.* Quality of care at a children's hospital: the parent's perspective. *Arch Pediatr Adolesc Med.* 1999;153(11):1123-1129.
4. Ireys HT, Perry JJ. Development and evaluation of a satisfaction scale for parents of children with special health care needs. *Pediatrics.* 1999;104(5 Pt 2):1182-1191.
5. Ygge BM, Arnetz JE. Quality of paediatric care: application and validation of an instrument for measuring parent satisfaction with hospital care. *Int J Qual Health Care.* 2001;13(1):33-43.
6. Co JP, Ferris TG, Marino BL, Homer CJ, Perrin JM. Are hospital characteristics associated with parental views of pediatric inpatient care quality? *Pediatrics.* 2003;111(2):308-314.
7. Mack JW, Co JP, Goldmann DA, Weeks JC, Cleary PD. Quality of health care for children: role of health and chronic illness in inpatient care experiences. *Arch Pediatr Adolesc Med.* 2007;161(9):828-834.
8. Carter B. Children--silent consumers of health care. *J Child Health Care.* 1998;2(2):57.
9. Farrant B, Watson PD. Health care delivery: perspectives of young people with chronic illness and their parents. *J Paediatr Child Health.* 2004;40(4):175-179.
10. Chesney M, Lindeke L, Johnson L, Jukkala A, Lynch S. Comparison of child and parent satisfaction ratings of ambulatory pediatric subspecialty care. *J Pediatr Health Care.* 2005;19(4):221-229.
11. Mah JK, Tough S, Fung T, Douglas-England K, Verhoef M. Adolescent quality of life and satisfaction with care. *J Adolesc Health.* 2006;38(5):e601-607.
12. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics.* 2004;114(5):1272-1280.
13. Litt IF. Satisfaction with health care: the adolescent's perspective. *J Adolesc Health.* 1998;23(2):59-60.
14. Alderson P. Competent children? Minors' consent to health care treatment and research. *Soc Sci Med.* 2007;65(11):2272-2283.
15. Lindeke L, Nakai M, Johnson L. Capturing children's voices for quality improvement. *MCN Am J Matern Child Nurs.* 2006;31(5):290-295.
16. Pelander T, Leino-Kilpi H. Quality in pediatric nursing care: children's expectations. *Issues Compr Pediatr Nurs.* 2004;27(3):139-151.
17. Aldiss S, Horstman M, O'Leary C, Richardson A, Gibson F. What is important to young children who have cancer while in hospital? *Children & Society.* 2009;23(2):85-98.
18. Alderson P, Sutcliffe K, Curtis K. Children as partners with adults in their medical care. *Arch Dis Child.* 2006;91(4):300-303.
19. Coad JE, Shaw KL. Is children's choice in health care rhetoric or reality? A scoping review. *J Adv Nurs.* 2008;64(4):318-327.
20. Ginsburg KR, Menapace AS, Slap GB. Factors affecting the decision to seek health care: the voice of adolescents. *Pediatrics.* 1997;100(6):922-930.
21. Ginsburg KR, Slap GB, Cnaan A, Forke CM, Balsley CM, Rouselle DM. Adolescents' perceptions of factors affecting their decisions to seek health care. *JAMA.* 1995;273(24):1913-1918.
22. Cheng TL, Savageau JA, Sattler AL, DeWitt TG. Confidentiality in health care. A survey of knowledge, perceptions, and attitudes among high school students. *JAMA.* 1993;269(11):1404-1407.
23. Woodgate RL. Health professionals caring for chronically ill adolescents: adolescents' perspectives. *J Soc Pediatr Nurs.* 1998;3(2):57-68.
24. Dovey-Pearce G, Hurrell R, May C, Walker C, Doherty Y. Young adults' (16-25 years) suggestions for providing developmentally appropriate diabetes services: a qualitative study. *Health Soc Care Community.* 2005;13(5):409-419.
25. Klostermann BK, Slap GB, Nebrig DM, Tivorsak TL, Britto MT. Earning trust and losing it: adolescents' views on trusting physicians. *J Fam Pract.* 2005;54(8):679-687.
26. Beresford BA, Sloper P. Chronically ill adolescents' experiences of communicating with doctors: a qualitative study. *J Adolesc Health.* 2003;33(3):172-179.
27. Tivorsak TL, Britto MT, Klostermann BK, Nebrig DM, Slap GB. Are pediatric practice settings adolescent friendly? An exploration of attitudes and preferences. *Clin Pediatr (Phila).* 2004;43(1):55-61.

28. Battrick C, Glasper EA. The views of children and their families on being in hospital. *Br J Nurs.* 2004;13(6):328-336.

29. Coad J, Coad N. Children and young people's preference of thematic design and colour for their hospital environment. *J Child Health Care.* 2008;12(1):33-48.

30. Ullán AM, Belver MH, Serrano I, Delgado J, Badía M. Perspectives of Youths and Adults to Improve the Care of Hospitalized Adolescents in Spain. *J Pediatr Health Care.* Epub 2010 Sept 20. DOI: 10.1016/j.pedhc.2010.08.005

31. Britto MT, Tivorsak TL, Slap GB. Adolescents' needs for health care privacy. *Pediatrics.* 2010;126(6):e1469-1476.

32. Knopf JM, Hornung RW, Slap GB, DeVellis RF, Britto MT. Views of treatment decision making from adolescents with chronic illnesses and their parents: a pilot study. *Health Expect.* 2008;11(4):343-354.

33. Moules T. 'They wouldn't know how it feels...': characteristics of quality care from young people's perspectives: a participatory research project. *J Child Health Care.* 2009;13(4):322-332.

34. Mokkink LB, van der Lee JH, Grootenhuis MA, Offringa M, van Praag BMS, Heymans HSA. Omvang en gevolgen van chronische aandoeningen bij kinderen. [Extent and consequences of chronic conditions in children]. *Tijdschr Kindergeneeskd.* 2007;75(4):138-142.

35. Snel MC. Dokter, dit vind ik! Verbeterpunten voor een megacool ziekenhuis [Doctor, this is my opinion! Ideas for a megacool hospital]. *Kind en Ziekenhuis.* 2006;29(1):18-22.

36. van Staa AL, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: The added value of mixed methods research. *Patient Educ Couns.* 2011;82(3):455–464.

37. van Staa AL, van der Stege HA, Jedeloo S, Moll HA, Hilberink S. Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors. *J Adolesc Health.* 2011;48(3):295–302

38. Creswell JW. *Research design: qualitative, quantitative, and mixed methods approaches*. 2nd Edition ed. London: Sage Publications; 2003.

39. O'Cathain A, Murphy E, Nicholl J. Why, and how, mixed methods research is undertaken in health services research in England: a mixed methods study. *BMC Health Serv Res.* 2007;7:85.

40. van Staa AL, Jedeloo S, Latour JM, Trappenburg MJ. Exciting but exhausting: experiences with participatory research with chronically ill adolescents. *Health Expect.* 2010;13(1):95–107.

41. Bryman A. Barriers to integrating quantitative and qualitative research. *J Mix Methods Res* 2007;1(1):8-22.

42. O'Cathain A, Murphy E, Nicholl J. The quality of mixed methods studies in health services research. *J Health Serv Res Policy.* 2008;13(2):92-98.

43. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res in Psychol.* 2006;3(2):77-101.

44. van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-8017-769-7.

45. Jedeloo S, van Staa AL, Latour JM, van Exel NJ. Preferences for health care and self-management among Dutch adolescents with chronic conditions: A Q-methodological investigation. *Int J Nurs Stud.* 2010;47(5):593-603.

46. Freake H, Barley V, Kent G. Adolescents' views of helping professionals: a review of the literature. *J Adolesc.* 2007;30(4):639-653.

47. Wray J, Maynard L. Specialist cardiac services: what do young people want? *Cardiol Young.* 2008;18(6):569-574.

48. Smith S. Adolescent units—an evidence-based approach to quality nursing in adolescent care. *European J Oncol Nurs.* 2004;8(1):20-29.

49. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.

3
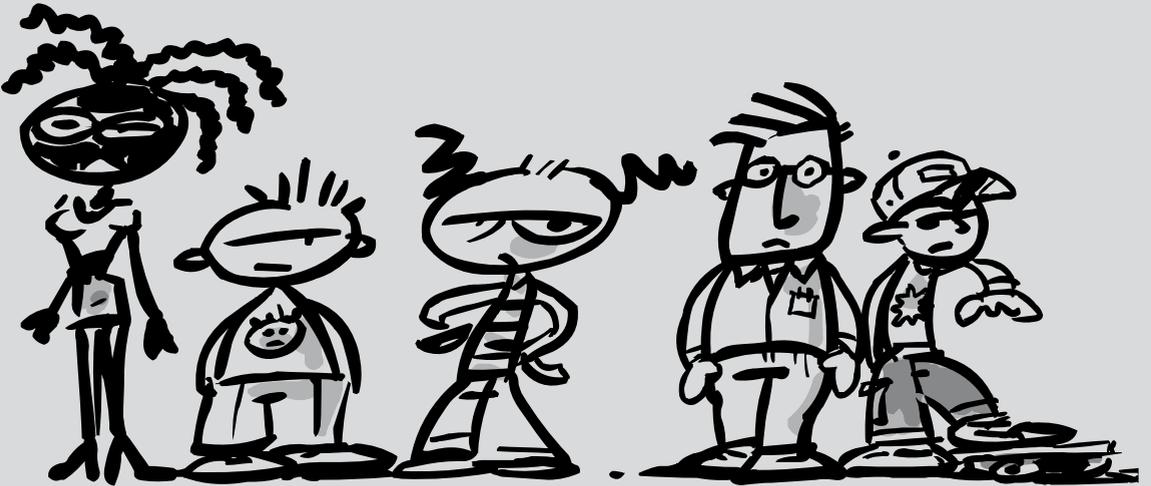
Preferences for health care and self-management among Dutch adolescents with chronic conditions: a Q-methodological investigation

Susan Jedeloo, AnneLoes van Staa, Jos M. Latour, N. Job A. van Exel

Abstract

BACKGROUND: Adolescents with chronic conditions have to learn to self-manage their health in preparation for transitioning to adult care. Nurses often struggle with how to approach youth with chronic conditions successfully. Little is known about the preferences and attitudes of these young people themselves.

OBJECTIVE: To uncover preferences for self-management and hospital care of adolescents with various chronic conditions.

DESIGN AND METHODS: A Q-methodological study was conducted. Semistructured interviews were held with adolescents who rank-ordered 37 opinion statements on preferences for care delivery and self-management. They were asked to motivate their ranking. By-person factor analysis was conducted to uncover patterns in the rankings of statements. The factors were described as preference profiles.

PARTICIPANTS AND SETTING: A purposive sample of 66 adolescents (12-19 years) treated in a university children's hospital in the Netherlands was invited to participate. Thirty-one adolescents, 16 boys and 15 girls with various chronic conditions eventually participated (response 47%). Eight participants (26%) had a recently acquired chronic condition, while the rest (74%) had been diagnosed at birth or in the first five years of life.

RESULTS: Four distinct preference profiles for health care delivery and self-management were identified: 'Conscious & Compliant'; 'Backseat Patient'; 'Self-confident & Autonomous'; and 'Worried & Insecure'. Profiles differ in the level of independence, involvement with self-management, adherence to therapeutic regimen, and appreciation of the parents' and health care providers' role. The desire to participate in treatment-related decisions is important to all preference profiles. The profiles are recognizable to adolescents and nurses alike. As Q-methodology allows no inferences with respect to the relative distribution of these profiles in a given population, only tentative hypotheses were formulated about associations between profiles and patient characteristics.

CONCLUSION: This study increases our understanding of different subjectivities of adolescents living with a chronic condition related to their treatment and health. There is no 'one size fits all' approach to adolescent health care, but rather a limited number of distinct preference profiles. This study demonstrates the value of a nondisease-specific approach in that adolescents with various chronic conditions were found to have much in common. The profiles seem a promising tool for nurses to actively seek adolescents' opinion and participation in health care and will be further explored.

What is already known about the topic?
- Adolescents with chronic disorders have to learn to manage their own condition, yet they are not often asked for their preferences.
- Actual involvement of adolescents in consultations and decision making is limited.
- Previous research revealed majority views and attitudes, while adolescents are known to have different preferences.

What this paper adds
- This Q-methodological study reveals four preference profiles among adolescents with different chronic conditions about health care and self-management.
- Profiles differ in attitudes toward independence, self-management, treatment adherence, and in appreciation of the parents' role. All profiles share a common desire to participate in decision making.
- The short profile descriptions are recognizable to nurses and seem a practical tool to seek adolescents' opinion and may increase young patients' participation in clinical encounters.

1   Background

Adolescents are rarely consulted as health care consumers, even though they are important users of health services whose preferences and concepts of care differ from those of adults.[1-3] Thus, little is known about priorities, desires and expectations of care from the adolescent perspective.[4-7]

As adolescence is a critical period for the establishment of health behaviours[8] and of a new working partnership with physicians and nurses, transition of adolescents with chronic conditions from pediatric to adult care is a major challenge for health care professionals in both settings.[9, 10] The first thing we should do is studying their perspectives and care-related attitudes.[11-13] In this regard, Britto and colleagues (2004) asked adolescents with various chronic illnesses to rate statements on quality of care and physician-patient communication styles in order of importance.[4] This produced a list of preferences from a majority perspective, with aspects of interpersonal care (especially honesty, attention to pain, and respect) ranking highest. However, the authors did not embed these rather specific preferences in adolescents' general views of their situation, their ideas about healthy lifestyles, coping with a chronic condition, and attitudes on compliance with treatment regimens. In addition, a majority view does not represent the variability of health care-related priorities and preferences among adolescents. Not all adolescents are alike and less dominant preferences do not emerge from such an approach, and may remain unrecognized and unattended. A recent study, for instance, revealed variation in preferences for decision making styles among adolescents with chronic illnesses.[14] Two other studies showed that younger and older adolescents with diabetes have differing attitudes with respect to preference for parents' involvement and responsibility in the management of their disease.[15, 16]

Everyday health care typically applies a disorder-specific approach. Different chronic health conditions nevertheless encompass many comparable tasks, such as managing symptoms and treatment, forming relationships with care providers, maintaining a positive self-image, relating to family and friends and preparing for an uncertain future.[17] The importance of such adaptive

tasks was established in a large study among older patients with different chronic diseases in the Netherlands,[18] but not yet in youth with chronic conditions.

Although the importance of consulting with chronically ill children and adolescents has been recognized, their views are rarely sought or acknowledged in health care settings and there is a need for strategies to facilitate and increase young patients' participation.[19] Most nurses will agree that there is no 'one size fits all' approach when it comes to stimulating youth to take responsibility for self-management and improving health care delivery for youth with chronic conditions. On the other hand, patients are not all different and it may thus be practical to identify sizeable and meaningful subgroups. Is this also the case for chronically ill adolescents? And do we go by age, socio-demographics, developmental milestones, or transition readiness scales? Or do we start with the viewpoint of young people themselves?

We decided on the latter. In this paper, therefore, results are presented of an exploratory study of attitudes of adolescents with chronic conditions toward health care delivery and self-management.

2   Method

Investigating a variety of accounts requires a methodology that is designed to identify the similarities and differences in attitudes from 'within'. Q-methodology[20, 21] was chosen because it allows identifying preference profiles of adolescents with a variety of chronic conditions sharing common viewpoints and to describe similarities and differences between these profiles.

2.1   Q-methodology

Q-methodology combines elements of qualitative and quantitative research methods and provides a scientific foundation for the systematic study of subjectivity, such as people's opinions, attitudes, and preferences.[22-25] While Q-methodology has had a place in science for almost seventy-five years,[21] it is fairly new in health research.[26-32] In nursing research, Q-methodology is increasingly popular.[33-35] Among adolescents, Q-methodology has been applied before to investigate their attitudes toward living with end stage renal disease,[36] therapy adherence in renal transplant receivers,[37] and toward healthy lifestyle attitudes.[38]

The aim of a Q-methodological study is to reveal principal views on a certain topic. Typically, respondents rank-order a sample of statements about the topic from their individual point of view, and thus reveal their subjective viewpoints.[23] The individual rankings, called Q-sorts, are then correlated in order to reveal similarities in viewpoint. Stephenson (1935) presented Q-methodology as an inversion of conventional factor analysis, in the sense that it correlates persons rather than statements.[21] If individuals each should have their own specific likes and dislikes, their Q-sorts would not correlate. If, however, significant clusters of correlations exist, these can be factorized, described as common viewpoints, and individuals can be mapped to them.

Q-methodology typically focuses on the range of viewpoints shared by specific groups of people.[25, 33] Therefore this method can be used to describe a *population of viewpoints* rather than

a population of people. For this purpose, a small sample of purposively selected respondents will do.[23] A Q-methodological study will thus *not* inform about proportions of people adhering to the viewpoints identified, or how these are associated with personal characteristics. Still, to map attitudes and subjective opinion, Q-methodology is a more robust technique than alternative methods.[22]

This Q-methodological study was conducted in four consecutive steps. Figure 1 presents an overview. Below we describe each of these steps.

Figure 1 *Steps in this Q-methodological study*

Step 1 Development of statement set (Q-set)
- Collection of statements through interviews with target population
- Thematic analysis of long-list of 104 opinion statements
- Selection of a representative set of 37 statements

Step 2 Selection of participants (P-set)
- Patients of Sophia Children's Hospital aged 12-19 years
- Purposive sampling based on age, sex, condition and hospital experience
- 31 patients participated

Step 3 Q-sorting
- Interviews at participants' home
- Participants rank-ordered the 37 opinion statements using a score sheet
- Participants explained their Q-sort

Step 4 Q-Analysis
- By-person factor analysis was conducted to reveal a limited number of corresponding ways the statements were sorted
- A composite sort of the statements was computed and consensus and differences in ordering of statements were examined
- Statistical results and explanations of participants defining the factor were used to interpret and describe the factors as preference profiles

2.1.1 Step 1. Statements (obtaining Q-set)

As the first step, we collected opinion statements regarding preferences for hospital care delivery. We did so through interviews with adolescents who recently transferred to adult care, by watching TV-documentaries, and by scrutinizing documentation and websites of patient organizations. All this resulted in a broad sample of 104 statements. Thematic analysis next identified seven major themes (Table 1). Each author independently assigned the statements to the seven themes and made a selection. These selections were discussed until consensus was reached on a final Q-set consisting of 37 representative statements, a number that was considered manageable for the population under study. Each of the themes was represented by at least three statements. The statements were randomly assigned a number and printed on cards (Table 2).

Table 1   *Major themes represented in final Q-Set*

	Themes	Statements
I	Clinical environment / organization of care	1,8,10,13,19,23,31,32
II	Physician/patient communication	2,4,12,16,22
III	Information provision	3,5,11,14,15
IV	Self-management / independence	6,7,9,17,18,20,21,24,30
V	Therapeutic regimen	26,29,33,37
VI	Disease perception	25,34,36
VII	Contact with fellow patients	27,28,35

Table 2   *List of statements (Q-set) with composite factor scores*

		Factor			
		A	B	C	D
1	Outpatient appointments should be scheduled according to my working hours / school hours	0	-1	-1	-1
2	It would be nice if you could also talk to the doctor or nurse in private, without your parents being present	1	1	1	1
3	If I want to know something about my disease, I'll look it up myself (on the Internet or in books)	1	-3*	-1†	0
4	Health professionals should not ask me personal questions in front of my parents	-1	-1	-1	0
5	I don't need any detailed information from health professionals about my disease or treatment	-1	1	-2	0
6	It's important for me to have my parents present during consultations	0*	3†	-1*	1†
7	I don't need any support from health professionals in becoming independent	1†	0	0	0
8	I want to have a say in when I transfer to adult care	1	1	1	-1*
9	Health professionals do not need to ask me about school, friends or how I spend my spare time	-1	1*	0	0
10	Apart from regular appointments with my doctor, I would also like to see a personal nurse or social worker	-1†	-2	0†	-2
11	I would like to know the consequences of a treatment for my daily life	3†	-1	2†	1
12	I would like health professionals to treat me like an adult	2*	0	1	0
13	There should be an outpatient clinic, particularly for young people (up to 25 years) with combined pediatric and adult care	0	0	0	-2
14	Health professionals should talk to me about sex, relationships and hereditary matters	0	-1	0	1*
15	Taking an 'exam' about your disease and treatment is a good idea	-3	-3	0*	-3
16	During consultations, I find it convenient if my parents do the talking for me	0†	2*	-2*	1†

		Factor			
		A	**B**	**C**	**D**
17	Health professionals should help me to set my own goals to become (more) independent	-1	0	0	-1
18	Health professionals should discuss my wishes and future plans with me	0	0	-1	-1
19	Care at Sophia's is okay the way it is. Nothing has to be changed	2	1	0†	2
20	Fortunately, my parents are there to remind me of my treatment and appointments	1	3*	0†	1
21	Health professionals should guide my parents and teach them how to 'let me go'	-1	-1	-2†	0
22	Health professionals should not be overprotective	0	1†	-1	0
23	I would like to ask my questions to health professionals by e-mail in between appointments as well	0	-2	0	-1
24	I want to have my own say in important matters about my health or treatment	2	2	2	2
25	I'd rather pretend as if there's nothing wrong with me	2*	0*	-3†	3*
26	Living easy now is more important than being completely treatment-compliant	0	0	3	2
27	I would like to have a buddy / pal to support me	-1	1	1	-2
28	It should be possible to ask difficult questions anonymously (by e-mail or a discussion panel)	1	0	2*	0
29	It's okay if health professionals give me a good talking about the consequences of being careless about my treatment	1	-2*	1	1
30	The hospital should also support you in finding a job, applying for services or living on your own	-2	0*	-2	-3
31	I'm not looking forward to go to another hospital when I'm about 18	-2	2*	-1	-1
32	I want to arrange my own hospital appointments	0	-1	0	0
33	I'll change my own treatment if this suits me better	-2	-1	2*	-1
34	I am just like anyone else, I just have a disorder on top	3	1*	3	3
35	I like meeting fellow patients through the hospital	-2*	0	1	0
36	I am worried about my health / my disease	-3	-2	-3	2*
37	I think it's annoying to get unasked-for advice on how to live	0	0	1	-2*

Notes:

A = 'Conscious & Compliant', B = 'Backseat Patient', C = 'Self-confident & Autonomous' and D = 'Worried & Insecure'.

"-3" indicates that the adolescents in that profile on (weighted) average disagree most with that statement; "+3" indicates adolescents in that profile on (weighted) average agree most with that statement (rank-ordered at extreme left/right in Fig. 1, respectively).

Distinguishing statements for a factor are indicated by an asterisk (*) P < .01; or cross (†) P < .05.

2.1.2 Step 2. Participants

A structured sample of respondents who are theoretically relevant to the problem under consideration was recruited to participate in the study.[20] Regarding characteristics of patients, gender, age, nature of the chronic condition (congenital or acquired in past five years), and hospital experience (number of outpatient and inpatients visits in the past two years) were considered most relevant to preferences for hospital care delivery.

The Erasmus MC Medical Informatics Department identified all patients between 12 and 19 years of age (n = 2202) who had visited Sophia Children's Hospital at least once in the past 6 months and had been under continuous treatment for at least the past 2 years. Two thirds had been under treatment for over 10 years. We did not select specific disorders or conditions because the study aimed to obtain a general understanding of adolescents' attitudes toward health care and self-management, and to research common adaptive tasks faced by all adolescents coping with a chronic somatic disorder. Adolescents with psychiatric diagnoses, a history of life-threatening illnesses, such as cancer, and known learning disabilities were excluded.

We distinguished into two groups: younger adolescents (12-15-year-olds, n = 1191) and older adolescents (16-19-year-olds, n = 1011). To facilitate the purposive sampling process, equal numbers of random cases were drawn from both groups. We aimed at equal numbers of participants from both age groups, and even distributions of gender, nature of the condition (congenital or acquired in past five years) and hospital experience within groups. Eligible adolescents and their parents received an invitation letter, a study information leaflet and a reply form from the research team. Upon written consent of both adolescent and parents, the primary researcher (SJ) arranged an interview.

2.1.3 Step 3. Q-sorting

The Q-set was administered during the interview at the participants' homes. Participants were first asked semistructured questions about their experiences with health care, knowledge and impact of their chronic condition, and about other issues related to care and daily living. Next, they performed the Q-sort by rank-ordering the statements using a score sheet (Figure 2). They were first asked to read through all statements and to sort them into three piles: cards containing statements, with which they agreed, disagreed and had no opinion about. Next, they read through the 'agreed' pile and placed the two they agreed with most on the two boxes at the right of the score sheet. Then they selected the next four cards they agreed with most and placed them on the score sheet, and so on, until the 'agreed' pile was exhausted. This procedure was repeated for the cards they disagreed with, now working from the left of the score sheet. The "neutral" statements were ranked in the middle. Finally, the interviewers asked the respondents to motivate the ranking of the four statements they (dis)agreed with most. Other choices were also discussed. All interviews were audio recorded and transcribed verbatim.

Figure 2 *Score sheet*

Least agree						Most agree
1	2	3	4	5	6	7
☐	☐	☐	☐	☐	☐	☐
☐	☐	☐	☐	☐	☐	☐
		☐	☐	☐	☐	
		☐	☐	☐	☐	
		☐	☐	☐		
		☐	☐	☐		
		☐	☐	☐		
			☐			
			☐			
			☐			
			☐			

2.1.4   Step 4. Q-Analysis

First, the Q-sorts were subjected to by-person factor analysis (centroid factor extraction with varimax rotation) using PQMethod version 2.11 (available on http://www.lrz-muenchen.de/~schmolck/qmethod/). The objective of the analysis was to reveal a limited number of corresponding ways the statements were sorted. For each factor a composite sort was computed, representing how a hypothetical adolescent with a 100% loading on that factor would have ordered the 37 statements.

   Then, the factors were interpreted and described as preference profiles. For this we used the characterizing statements (those with a factor score of +3, +2, -2 and -3 in the composite sort), the distinguishing statements (those with a statistically significantly different factor score as compared to all other factors; $P < .05$), and the verbal motivations by adolescents loading on that factor. This is extensive interpretative work, combining qualitative and quantitative analytic techniques.[39]

2.5   Ethical standards and procedures

The study protocol was approved by the Medical Ethics Review Board of the Erasmus Medical Center. All adolescents and their parents received information about the project and both child and parent gave their written consent. Participants received a € 20 gift voucher.

3   Results

3.1   Response

Of the purposive sample of 66 adolescents who were invited, thirty-one eventually participated. Twenty-four refused (36%) and 11 (17%) could not be reached by phone for a reminder. Thus, 31 (47%) consented to an interview. Most of those who refused said they had no time; some said they were too ill or too occupied with family problems while others were not interested to participate in the study. Parents were very supportive to the study and some even consented though their child refused. A nonresponse analysis based on the determinants used for the sample selection revealed no significant differences between participants and nonparticipants.

 Twenty-seven interviews were conducted by pairs of purpose-trained nursing and paramedical students and four by the primary researcher (SJ). Seventeen adolescents were aged 12-15 years; 14 aged 16-19 years (Table 3). Mean age for the total group is 15.3 (*SD* 2.1), 16 (52%) were male. Most had visited the outpatient department over three times a year and only a minority had been hospitalized over the past two years. Twenty-three (74%) suffered from a lifelong chronic condition (eg, congenital or diagnosed before six years of age); 8 (26%) were diagnosed in the past five years. Seven (23%) showed comorbidity. Five (20%) presented with surgical conditions: scoliosis, kyphosis, benign intracranial hypertension, congenital bladder disorder, ventricular septum defect and facial schisis. Internal conditions included rheumatoid arthritis, hemophilia, diabetes mellitus, epilepsy, inflammatory bowel disease, cystic fibrosis, various metabolic disorders, HIV, nephrotic syndrome, immune and hormone deficiencies, lung insufficiency, progressive kidney failure, congenital skin diseases, asthma, and neuromuscular diseases such as Duchenne muscular dystrophy, hereditary motor and sensory neuropathy, and spasticity.

Table 3   *Characteristics of 31 study participants*

	n (%)
Male	16 (52)
Age (years) range 12-19 *mean (SD)*	 15.3 (2.1)
Congenital chronic condition or acquired in early life	23 (74)
Over 7 outpatient visits in past 2 years	19 (61)
No. hospital admissions in past 2 years	22 (71)

3.2   Q-Analysis

By-person factor analysis of the 31 Q-sorts resulted in four distinct preference profiles (factors with Eigen value > 1 were retained). Adjacent factor solutions were also explored with respect to their content, but the four factor solution was also the most clear and comprehensible one. Based on a Q-set of 37 statements and $P < .01$, the factor loading of a Q-sort must be equal to or higher than .42 to be a defining variable for that factor.[20, 24] The four factors were defined by between three and eleven Q-sorts (21 in total). They explained from 7 to 19% of the variance, 42% in total. Correlation between factors B / C; and C / D is low ($r = .03$ and $r = .01$ respectively), while factor B correlates moderately with D ($r = .32$). Factor A has rather strong correlations with factor C ($r = .41$) and D ($r = .54$); and correlates to a lesser extent with B ($r = .27$), indicating that not only discrete viewpoints were identified, but also consensus.

Hereafter, we describe the four preference profiles, referring to the statement numbers shown in Table 2 [figures in parentheses]. The motivations by adolescents loading on a factor are given "*between quotation marks*". Then we discuss similarities and differences between the profiles.

3.3   Preference profile A: 'Conscious & Compliant'

Most characteristic of this profile is the high level of involvement with disease management. These adolescents want to know the consequences of a treatment [11] *"because simply I think it is very important to me, when something would have negative consequences I would rather not have it"*. Taking an exam about their disease and treatment [15] is seen, however, as *"nonsense"* and *"overdone"*. They *"already know most there is to know"*.

These adolescents prefer to pretend nothing is wrong with them [25]. They are not so much opposed to disclosure; it is rather felt not necessary: *"When it's not necessary for someone to know, I won't tell"*. They see themselves as being like anyone else, just with a disorder on top [34]: *"I try to be as normal as possible and this disease permits me to be"*. This may be related to the fact that these adolescents are not too worried about their health or disease [36]. Everything is going fine now, so *"I don't worry, it is not that bad"*, *"it's not life threatening or something"*.

These adolescents, more than those fitting in the other preference profiles, want to be treated as adults by health professionals [12]: *"I don't feel like a child anymore". "A doctor should come to the point immediately and not beat around the bush with nice stories".* They are equally firmly pronounced about not needing professional support in becoming more independent [7]: *"I think I am quite independent already and I don't really need help with it".* Nor do they feel the need to meet fellow patients through the hospital [35]: *"If I would feel that need, then I'd go and surf on the Internet".*

These adolescents least of all appreciate a role for their parents and strongly prefer being at the centre of the medical encounter [6;16]: *"It is convenient when my parents do the talking, but on the other hand it is nice when they are not present because they interrupt me all the time, and that's quite annoying"; "I am the patient; he (the doctor) should talk to me and not to my parents".* They do not see leaving pediatric care as problematic [31]. Adhering to treatment, they are not inclined to change treatment on their own initiative [33]. They wish to avoid problems and ap-

preciate doctors' knowledge and medical advice: *"Essentially, they do everything they can to make you healthy, so I think you have to put in some effort yourself"; "If you just do what the doctors tell you to, then you don't have to worry about your health".*

The eleven adolescents comprising this profile were seven boys and four girls with a mean age of 16 years (range 13-19 years). Six of them had been diagnosed in the past five years; most did not have extensive hospital experience.

3.4   Preference profile B: 'Backseat Patient'

The adolescents in this profile are less mature and lean more on their parents. They find it important to bring their parents to the hospital [6] and feel it is convenient the parents do the talking [16] because "*I don't like to talk, this is a little difficult", "because they take care of everything for me… it seems difficult to me".* They feel *"not capable of remembering everything very well".* Therefore they appreciate parents reminding them of their treatment regimens [20]. When careless about their treatment [29], they do not like *"other people confronting me with the consequences. I know the consequences, but I don't care".* They count on their parents to do so: *"I prefer that they tell me, better twice too often than one too few".*

Health professionals should not be overprotective [22]: *"My parents do this for me".* They do not see the need for professional support in issues related to school, friends or leisure activities [9], in finding a job [30], or in becoming more independent [17]: *"That's what I have my mother for!"* The opinions of professionals are not questioned: *"If the doctor says so, I believe him".*

Searching for information about the disease or treatment [3] is not something these adolescents would do: *"I surely wouldn't look it up myself", "I just don't want to know too much about it".* Accordingly, they are not in favor of taking an exam about their disease [15]: *"One surely wouldn't take an exam about one's disease! If you don't know, you can ask your doctor".*

These adolescents are also not very involved with their health. They consider themselves to be just like others, only with a disease on top [34]*: "Mentally I'm okay, but physically things are a bit less".* Still, they do not pretend nothing is wrong [25]: *"Because, actually there is something and if you try and pretend there is not, they will find out anyway. But I'm not upfront about it. If people ask, I tell."* They are also not particularly engaged with transition to adult care, but they are not looking forward to leaving pediatric care [31]: *"I really appreciate Sophia Children's Hospital, it is familiar".* Still, thinking about the future and discussing future plans with health professionals *"does not interest me at all"* [18]*.*

All four adolescents comprising this profile were girls, with a mean age of 15 years (range 12-17). They had serious congenital disorders with a considerable impact on daily life. They regularly visited the outpatient department and had been hospitalized as well.

3.5   Preference profile C: 'Self-confident & Autonomous'

These adolescents strongly feel the need to be upfront about their chronic condition [25]; they don't hide behind their disease, they live with it: *"If you suffer from something, you just have to come forward; otherwise you'll pay the price".* The chronic disorder does not make them differ-

ent from healthy peers [34]: *"It is a fact that I take medicines and sometimes have to stay in the hospital, but for the rest I am just the same and like to be treated as such"*. They strongly feel that enjoying life now is more important than being fully adherent to treatment [26]: *"Everyone sometimes skips their treatment. That should be possible! I don't think there is anyone who's always compliant with the rules. One should not give up things because of an illness..."*

In the same vein, they do not seem to worry much about their disease or health [36] and do not need help from health professionals in managing their care and independence [3; 17; 21] or from parents [6; 16; 20]: *"I can stand up for myself!"* They feel confident about their knowledge of their condition, but like to know the consequences of their treatment and of nonadherence [5; 11]. Compared to other profiles, they are the only ones who will change treatment if felt necessary [33], sometimes after consulting their doctor or parents, but not as a general rule: *"Well, if I should have to call the doctor for every little detail, he would go mad"*. Self-confidence and a strong desire to being autonomous in decision making characterize these adolescents.

The four adolescents comprising this profile were two boys and two girls from the older age group (mean age 17, range 16-19 years). All were diagnosed early in life and two of them were frequent hospital visitors.

3.6   Preference profile D: 'Worried & Insecure'

These adolescents are most of all worried about their disease [36]: *"there are so many consequences when things go wrong"*, *"one just doesn't know how things become later on, what you're still be able to do"*, *"for example, I might get a heart disease or die earlier"*. They prefer to pretend nothing is wrong with them [25] and to be like anyone else [34], mainly because it prevents them from worrying: *"That way I feel better... pretending I belong with the others"*. They think that living an easy life now is to be preferred over full therapy compliance [26].

Being insecure, they do not feel bothered by unsolicited health advice [37]. They feel they need more information about difficult and sensitive issues such as sexuality and procreation [14]. They are not opposed to welcoming professional support in learning how to manage their disease [7] as *"that makes becoming independent easier, if you don't have to do everything on your own"*. But they do not want support from health professionals in other areas, like job finding, applying for services or living independently [17; 30]; thinking about the future confronts them with the possible unpleasant consequences of their disease [18]. Unlike others, these adolescents do not want to have a say in when to transfer to adult care [8]. Adolescents in this profile appreciate their parents' support during consultations [6], ie, reminding them of therapy adherence and appointments [20].

Three girls comprised this profile (mean age 16 years; range 13-16). Two had recently acquired conditions with considerable impact on daily life. All had visited the outpatient department over six times in the past two years.

3.7   Differences and similarities between profiles

The four preference profiles differ particularly on five of the seven themes contained in the Q-set of statements (Table 1): (adherence to) therapeutic regimens; self-efficacy with relation to independent health behaviors; information provision; type of physician-patient communication and disease perception (being worried / feeling different). Differences are less pronounced with respect to adolescents' preferences for the organization of care and contact with fellow patients.

Adolescents in profiles A and C feel more independent and display a higher degree of self-efficacy than those in the other two. However, these profiles are characterized by different attitudes toward treatment adherence. While 'Conscious & Compliant' adolescents prefer to adhere to treatment to avoid future health problems, 'Self-confident & Autonomous' adolescents consciously decide to be nonadherent, preferring an easy life to continuous awareness of disease. The profiles B and D share a common feeling of dependency and lack of self-confidence, but for different reasons. 'Backseat Patient' adolescents lean on their parents because they feel uninvolved and incompetent. Leaving responsibility to their parents and having parents do the talking is easier for them. 'Worried & Insecure' adolescents lack confidence because they worry about their future health. They need their parents to support them in coping with insecurities.

With respect to information provision, adolescents in profiles A and C want to know about the consequences of their condition for daily life [11]. On the other hand, 'Worried and Insecure' adolescents would rather not know – so as to avoid anxiety. 'Backseat Patients' do not see the need of being informed, their parents ought to know.

Adolescents are positive about current care in the Sophia Children's Hospital. All but 'Worried & Insecure' adolescents want to have a say in when to transfer to adult care [8]. Only the 'Backseat Patients' agree that they are not looking forward to go to another hospital when they turn 18 [31]; the others do not seem to mind. Contact with fellow patients is desirable for 'Self-confident & Autonomous' adolescents only [28], preferably through the hospital [31]. This is in contrast to adolescents constituting the 'Conscious & Compliant' profile.

Apart from differences, adolescents' preferences also show similarities. Consensus among profiles was found on five statements (ie, no statistically significant difference in ranking of statements between any pair of profiles; *P* < .05). All adolescents want to have a say in important treatment-related issues [24]. Adolescents do not expect professionals to play a major role in supporting their independence [7] or discussing future plans [18]. They are not opposed to doctors asking personal questions in front of their parents [4], probably because they are used to having their parents present during consultations. Nevertheless, when sensitive issues such as sexuality and heredity are raised, parental presence *"may be sometimes inconvenient"*. All types of adolescents would like to have the opportunity to talk to doctors and nurses alone [2], even 'Backseat Patients' for who it is strongly important to have parents present during consultations [6].

4   Discussion

This study demonstrates that not all adolescents with chronic conditions share common ideas about their illness and treatment: some are care-free, others worry about their health. With respect to self-management and adherence to treatment regimens it appears that one group favors 'a good life' and autonomy over following a strict regimen; whereas another group is concerned about future consequences of sloppy adherence. A Q-methodological study among young adult renal transplant recipients found two profiles associated with nonadherent behaviour[37]; while in a third one, young adults ran a higher risk of depression. Unlike in the present study, preferences on the parents' role did not play a decisive role in differentiating between the profiles, possibly because the transplant recipients were older.

Preferences for information provision also vary, indicating that the desire to be fully informed is not a common trait, in contrast to what was suggested in a study of young cancer patients.[40] Other studies have stressed the importance of concrete, practical advice,[41] and of using understandable, jargon-free language.[42] The fact that most adolescents in our group desire to know the consequences of a treatment for their daily life [11] confirms this. Still, young people report they cannot always rely on professional advice, because it does not conform to their life situation.[43] Wanting to be informed does not imply, however, that adolescents will actively seek information themselves. 'Self-confident & Autonomous' and 'Backseat' adolescents indicate that they have no intention of doing so [3].

Several studies indicated that adolescents with chronic conditions give highest priority to a physician's honesty and expertise.[4, 5, 44] Honesty was not included in the present study, but the fact that most adolescents (except the 'Backseat Patient') do not mind it if health professionals give them a good talking about the consequences of suboptimal adherence to treatment [29] seems to corroborate this finding. Issues of confidentiality, familiarity and privacy have also been reported to be of importance,[42, 44, 45] but are not so prominent in our Q-set. Most adolescents do not worry about physicians asking private questions in front of their parents [4]. For that matter, adolescents with chronic illnesses are more comfortable involving parents in their care than are healthy adolescents.[44] Parental encouragement increases teenagers' certainty in performing self-management tasks.[43]

Yet, adolescents in profiles 'Conscious & Compliant' and 'Self-confident & Autonomous' prefer to be treated as adults [12] and to have communication directed to them rather than to their parents, as was reported before.[4, 46] Still, adolescents have different preferences with regard to communication, being treated as an adult,[13, 45] level of involvement in decision making,[13, 15, 40] and parental presence during consultations.[4, 46] Our study confirms this: 'Backseat Patients' do not mind their parents doing the talking for them, whereas 'Self-confident & Autonomous' adolescents insist on doing it themselves [16].

Knopf *et al* (2008) found that half of the adolescents studied favored a passive decision making style followed by one third preferring shared decision making.[14] Our study confirms these differences in preferred level of involvement, yet all adolescents appreciate being able to have their own say in important matters about their health or treatment [24]. This is related to adolescents' feeling that they are the very experts in self-management and decision making.[43]

Several studies indicated that adolescents expect emotional support and encouragement from health professionals.[12, 40, 45] Trust is a core concept in the relation between adolescents and health professionals.[4, 44] Our data suggest that young people appreciate health care staff 'being there' for them while at the same time they are reluctant to involve them in becoming independent young adults.

Finally, most adolescents empathically try to see themselves as normal individuals [34], an important aspect of growing up with chronic illness.[47] Only the 'Backseat Patient' is less pronounced regarding disease perception. Except for the 'Worried and Insecure' adolescents, worries about health do not figure prominently, like in the general adolescent population.[38]

4.1   Strengths and limitations of the study

A key element for interpretation of the results is combining the statistical results with the motivations by participants, marking the hybrid character of Q-methodology. Although this involves extensive interpretative work, it is firmly based on results from common statistical procedures.[39] Compared to conventional factor analysis, a strong point of Q-methodology is its use of the language of the population under study. Q-analysis does not only reveal scores on individual items but also links these to motivations provided in individual interviews.

Careful design of the Q-set is pivotal, as the opinion statements should be representative for the study topic.[33] We feel our Q-set to be broadly representative, but do not claim that all subjective constructions relevant to this field have been exhaustively identified.

Participants were sampled on age, gender, nature of the condition and recent hospital experience. Other potentially relevant characteristics, such as severity of the condition and impact on daily life could not be assessed beforehand.

The nonresponse in this study was fairly high (53%), but because there were no significant differences between participants and nonparticipants on selected variables, we do not expect that the nonparticipating individuals would have comprised a separate profile. We do not think that the single-centre recruitment strategy has influenced the findings of this study. The Erasmus MC – Sophia is the largest university children's hospital in the Netherlands, servicing a wide area and involving all pediatric subspecialties.

Q-methodology clusters respondents according to their ranking of the statements presented, whereas conventional factor analysis clusters statements according to respondents' ratings. The focus on similarities and differences elicits the diversity of viewpoints and helps avoid the tendency to concentrate on commonalities between participants.[33] But the results of a Q-methodological study can only be generalized to the study topic, not to the wider population of respondents. Therefore, based on this study, it is not possible to make inferences about the relative distribution of the profiles or their associations with personal characteristics of participants. This form of representativeness plays no role in Q-methodology.[25] Some tentative hypotheses about associations between profiles and patient characteristics can however be made. For example, younger teenagers and girls are more likely to be 'Backseat Patient' or 'Worried & Insecure', older teenagers more likely 'Self-confident & Autonomous'. Adolescents with a congenital disorder and a considerable burden of disease are more likely to be 'Backseat Patient' or 'Self-confident & Autonomous', while

those with a more recently acquired disorder will tend to be 'Conscious & Compliant' or 'Worried & Insecure'. Finally, the 'Worried & Insecure' might be prone to depression and experience a lower quality of life.

Because the study sample included adolescents with a wide range of chronic conditions, nothing can be said about possible relations between profiles and specific chronic conditions. A survey study which presents the four profiles to a representative sample of adolescents could reveal this type of information and is intended.

As demonstrated earlier,[37, 38] applying Q-methodology among adolescents is quite successful in triggering adolescents to speak freely and extensively about their own views and preferences. In the course of the study we learned that health care professionals valued this specific characteristic of the method, making Q-methodology potentially useful for clinical practice.

4.2   Clinical implications

This study demonstrates that there is no 'one size fits all' approach to adolescent health care, but that irrespective of individual differences between patients, a limited number of distinct preference profiles can be identified. There are also aspects that all adolescents share an interest in; they value the opportunity to have both voice and choice in decisions regarding their care. Knowing this, nurses and other health care professionals would do well to strive for a personalized approach and a more adolescent-centered health care system. Therefore, a priority for further research is developing more differentiated strategies, related to each of the four profiles, to stimulate adolescents' self-management competencies.

Most nurses and other health professionals involved in adolescent health care would welcome a simple screening instrument that helps identify potentially risky situations in clinical practice, such as over-dependence, lack of self-confidence and nonadherence, sooner and better. Motivated by this interest, we used abbreviated factor descriptions (Appendix A) as a first test of such an instrument. To avoid undesired response effects, names of the profiles were not presented to parents or adolescents. Although the profile descriptions refer to general attitudes and preferences for self-management and hospital care and do not address disease-specific tasks or skills, they still seem potentially useful in a range of clinical settings. Nurse specialists working with youth with different chronic conditions recognized the typologies and saw the descriptions as helpful in stimulating discussion with patients. Also, adolescents could easily recognize and identify themselves with the profiles.

Furthermore, adolescents' attitudes and preferences may develop over time. These abbreviated profile descriptions may therefore also be useful as part of a clinical assessment tool to measure changes in autonomy and preferences in adolescents with chronic conditions. These are issues of particular importance for future studies.

5   Conclusion

Q-methodology enabled us to identify four general clusters of adolescents with chronic conditions in terms of their priorities and preferences for health care provision, self-management and adherence. Irrespective of individual differences between adolescents, a limited number of distinct preference profiles could be identified. This study demonstrates the value of a nondisease-specific approach, as the preference profiles are recognizable to adolescents with various chronic conditions. There are also aspects that all adolescents share an interest in; they value the opportunity to have both voice and choice in decisions regarding their care. Knowing this, nurses and other health care professionals would do well to strive for a personalized approach and a more adolescent-centered health care system.

Use of these profiles in clinical practice could stimulate a much needed conversation between adolescent patients and their providers and needs to be further explored. Further research should also reveal the relative distribution of these four profiles in the broad population of adolescents with chronic conditions and the associations with personal characteristics such as gender, age and educational level and disease-related characteristics, such as type of chronic condition, severity of illness, and disease duration.

Appendix A   *Abbreviated descriptions of preference profiles*

How well does this profile fit you?

A = Conscious & Compliant

If I just do what the doctors say, I don't have to worry about my disease or health. After all, my disease permits living a normal life. I think I am pretty independent now and would like to be treated like an adult. That is why I want the doctors to talk to me and not to my parents. Health professionals should point out my own responsibility to me, for I don't want to regret it or be confronted with my disease later, for not following the rules now. I know enough about my disease, but I would like to know the consequences of a treatment for my daily life. Also, I don't need any support from the hospital in coping with my disease, I will take care of that myself, or my parents will help me.

B = Backseat Patient

I am not too bothered about my disease; my parents will take care of this. They are helping me with my treatment and see to it that everything is in control. I think it will be pretty difficult to take care of this myself. I don't need to know everything in detail, if my parents do, it's okay. When I'm at the doctor's, it's convenient to have my parents with me; they can do the talking for me. I find this difficult and actually, I'm not too interested. But the doctors shouldn't treat me like a child, nor tell me what I'm doing wrong. That's my parents' job. I'm not ready yet to leave the children's hospital and go to adult care.

C = Self-confident & Autonomous

Living easy now is more important to me than being completely compliant. It's me who decides how to live; I won't have my disease limit me in daily life. I'll change my own treatment if this suits me better. I am capable of doing this myself, for I know enough about it. I do want to be treated like an adult and do my own talking. I don't need my parents for that. No one needs to hold my hand, but I do find it convenient to get information. Though this does not mean I will always use it.

D = Worried & Insecure

I'm pretty worried about my disease. Suppose things will turn for the worse later. Sometimes, through my illness, I don't feel at ease with myself, but I try not to think about it too much. I'd rather pretend there's nothing wrong with me. I do not always take advice too seriously, that way I don't have to think about my disease. But I do need my parents and health professionals to help me discuss difficult subjects that are important for my future. I feel more secure about myself if they tell me how to live best.

Note: Profile names are not provided when the descriptions are presented to adolescents or parents

References

1. Cavet J, Sloper P. The participation of children and young people in decisions about UK service development. *Child Care Health Dev*. 2004;30(6):613-621.
2. Dixon-Woods M, Young B, Heney D. Partnerships with children. *Br Med J*. 1999;319(7212):778-780.
3. Sloper P, Lightfoot J. Involving disabled and chronically ill children and young people in health service development. *Child Care Health Dev*. 2003;29(1):15-20.
4. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics*. 2004;114(5):1272-1280.
5. Farrant B, Watson PD. Health care delivery: perspectives of young people with chronic illness and their parents. *J Paediatr Child Health*. 2004;40(4):175-179.
6. Litt IF. Satisfaction with health care: the adolescent's perspective. *J Adolesc Health*. 1998;23(2):59-60.
7. Oppong-Odiseng AC, Heycock EG. Adolescent health services--through their eyes. *Arch Dis Child*. 1997;77(2):115-119.
8. Viner RM, Barker M. Young people's health: the need for action. *Br Med J*. 2005;330(7496):901-903.
9. McDonagh JE, Viner RM. Lost in transition? Between pediatric and adult services. *Br Med J*. 2006;332(7539):435-436.
10. Viner RM. Transition of care from pediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child*. 2008;93(2):160-163.
11. Britto MT, Slap GB, DeVellis RF, Hornung RW, Atherton HD, Knopf JM, *et al.* Specialists understanding of the health care preferences of chronically ill adolescents. *J Adolesc Health*. 2007;40(4):334-341.
12. Woodgate RL. Health professionals caring for chronically ill adolescents: adolescents' perspectives. *J Soc Pediatr Nurs*. 1998;3(2):57-68.
13. Wray J, Maynard L. Specialist cardiac services: what do young people want? *Cardiol Young*. 2008;18(6):569-574.
14. Knopf JM, Hornung RW, Slap GB, DeVellis RF, Britto MT. Views of treatment decision making from adolescents with chronic illnesses and their parents: a pilot study. *Health Expect*. 2008;11(4):343-354.
15. Dovey-Pearce G, Hurrell R, May C, Walker C, Doherty Y. Young adults' (16-25 years) suggestions for providing developmentally appropriate diabetes services: a qualitative study. *Health Soc Care Community*. 2005;13(5):409-419.
16. Hanna KM, Juarez B, Lenss SS, Guthrie D. Parent-adolescent communication and support for diabetes management as reported by adolescents with type 1 diabetes. *Issues Compr Pediatr Nurs*. 2003;26(3):145-158.
17. Moos RH, Holahan CJ. Adaptive tasks and methods of coping with illness and disability. In: Martz E, Livneh H, editors. *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. New York: Springer; 2007.
18. Heijmans M, Rijken M, Foets M, de Ridder D, Schreurs K, Bensingt J. The stress of being chronically ill: from disease-specific to task-specific aspects. *J Behav Med*. 2004;27(3):255-271.
19. Coyne I. Children's participation in consultations and decision making at health service level: a review of the literature. *Int J Nurs Stud*. 2008;45(11):1682-1689.
20. Brown SR. *Political subjectivity: applications of Q methodology in political science*. New Haven, CT: Yale University Press; 1980.
21. Stephenson W. Correlating persons instead of tests. *Character and Personality*. 1935;4:17-24.
22. Cross RM. Exploring attitudes: the case for Q methodology. *Health Educ Res*. 2005;20(2):206-213.
23. Smith NW. *Current systems in psychology: history, theory, research, and applications*. Belmont, CA: Wadsworth/Thomson Learning; 2001.
24. van Exel NJA, de Graaf G. Q methodology: a sneak preview [Internet]; 2005. Available from: www.qmethodology.net.
25. Watts S, Stenner P. Doing Q methodology: theory, method and interpretation. *Qual Res Psychol*. 2005;2(2):67-91.
26. Aldrich S, Eccleston C. Making sense of everyday pain. *Soc Sci Med*. 2000;50(11):1631-1641.
27. Baker RM. Economic rationality and health and lifestyle choices for people with diabetes. *Soc Sci Med*. 2006;63(9):2341-2353.
28. Bryant LD, Green JM, Hewison J. Understandings of Down's syndrome: a Q methodological investigation. *Soc Sci Med*. 2006;63(5):1188-1200.

29.   Morecroft C, Cantrill J, Tully MP. Individual patient's preferences for hypertension management: a Q-methodological approach. *Patient Educ Couns*. 2006;61(3):354-362.

30.   Risdon A, Eccleston C, Crombez G, McCracken L. How can we learn to live with pain? A Q-methodological analysis of the diverse understandings of acceptance of chronic pain. *Soc Sci Med*. 2003;56(2):375-386.

31.   Stenner PH, Cooper D, Skevington SM. Putting the Q into quality of life; the identification of subjective constructions of health-related quality of life using Q methodology. *Soc Sci Med*. 2003;57(11):2161-2172.

32.   van Exel J, de Graaf G, Brouwer W. Care for a break? An investigation of informal caregivers' attitudes toward respite care using Q-methodology. *Health Policy*. 2007;83(2-3):332-342.

33.   Akhtar-Danesh N, Baumann A, Cordingley L. Q-Methodology in Nursing Research: A Promising Method for the Study of Subjectivity. *West J Nurs Res*. 2008;30(6):759-773.

34.   Barker JH. Q-methodology: An alternative approach to research in nurse education. *Nurse Educ Today*. 2008;28(8):917-925.

35.   Kim DS, Kim HS, Schwartz-Barcott D, Zucker D. The nature of hope in hospitalized chronically ill patients. *Int J Nurs Stud*. 2006;43(5):547-556.

36.   Snethen JA, Broome ME, Bartels J, Warady BA. Adolescents' perception of living with end stage renal disease. *Pediatr Nurs*. 2001;27(2):159-161, 164-157.

37.   Tielen M, van Staa AL, Jedeloo S, van Exel NJ, Weimar W. Q-methodology to identify young adult renal transplant recipients at risk for nonadherence. *Transplantation*. 2008;85(5):700-706.

38.   van Exel NJ, de Graaf G, Brouwer WB. "Everyone dies, so you might as well have fun!" Attitudes of Dutch youths about their health lifestyle. *Soc Sci Med*. 2006;63(10):2628-2639.

39.   Jedeloo S, van Staa AL. Q-methodologie, een werkelijke mix van kwalitatief en kwantitatief onderzoek? [Q-methodology: a true mix of qualitative and quantitative research?]. *KWALON*. 2009;14(2):5-15.

40.   Zwaanswijk M, Tates K, van Dulmen S, Hoogerbrugge PM, Kamps WA, Bensing JM. Young patients', parents', and survivors' communication preferences in pediatric oncology: Results of online focus groups. *BMC Pediatr*. 2007;7(35).

41.   Woodgate RL. Adolescents' perspectives of chronic illness: "it's hard". *J Pediatr Nurs*. 1998;13(4):210-223.

42.   Beresford BA, Sloper P. Information needs of disabled young people. *Research Works.* York (UK): Social Policy Research Unit, University of York; 2000 May. http://php.york.ac.uk/inst/spru/pubs/707

43.   Karlsson A, Arman M, Wikblad K. Teenagers with type 1 diabetes--a phenomenological study of the transition towards autonomy in self-management. *Int J Nurs Stud*. 2008;45(4):562-570.

44.   Klostermann BK, Slap GB, Nebrig DM, Tivorsak TL, Britto MT. Earning trust and losing it: adolescents' views on trusting physicians. *J Fam Pract*. 2005;54(8):679-687.

45.   Freake H, Barley V, Kent G. Adolescents' views of helping professionals: a review of the literature. *J Adolesc*. 2007;30(4):639-653.

46.   Beresford BA, Sloper P. Chronically ill adolescents' experiences of communicating with doctors: a qualitative study. *J Adolesc Health*. 2003;33(3):172-179.

47.   Taylor RM, Gibson F, Franck LS. The experience of living with a chronic illness during adolescence: a critical review of the literature. *J Clin Nurs*. 2008;17(23):3083-3091.
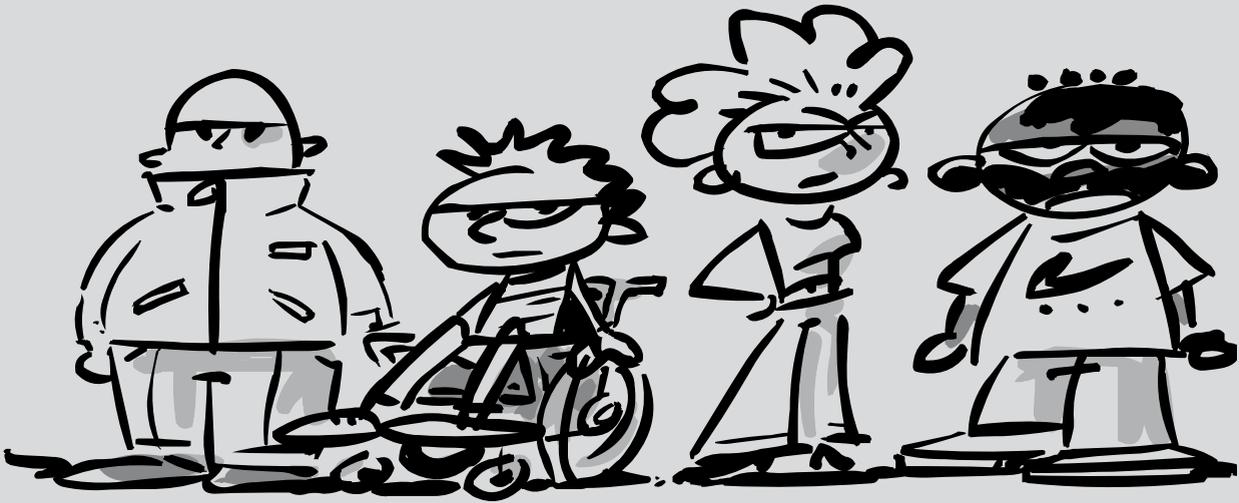
Four distinct self-management styles in chronically ill adolescents: distribution of and associations with Q-Care Profiles

AnneLoes van Staa, Susan Jedeloo, Heleen A. van der Stege, N. Job A. van Exel

Abstract

OBJECTIVE: Self-management is a major challenge for adolescents with chronic conditions. This study aimed to determine the distribution of four self-management styles called Q-Care Profiles, to establish construct validity, and to explore the associations between adolescents' fit to each Q-Care Profile and socio-demographic and disease-related variables.

PATIENTS AND METHODS: Cross-sectional study in a Dutch university children's hospital. All adolescents (12-19 yrs) with somatic chronic conditions in long-term treatment were asked to assess their fit to the different Q-Care Profiles in a web-based questionnaire. Differences were tested with logistic regression analyses.

RESULTS: 1087 adolescents responded (30%); 990 assessed their fit to the Q-Care Profiles. The 'Conscious & Compliant' profile fitted 56%, appealing more to older and higher educated youth. These experienced better health and felt confident to manage their condition. The 'Backseat Patient' profile fitted least (16%) and was associated with being younger, physically disabled, and lower educated. They experienced lower quality of life and felt less self-efficacious. The 'Self-confident & Autonomous' profile fitted 26%, but none of the variables considered was significantly associated with fit to this profile. The 'Worried & Insecure' profile fitted 25%, mostly younger, lower educated girls of non Dutch ethnicity. They experienced poor health, lower quality of life, and felt less self-efficacious.

CONCLUSIONS: The surveyed adolescents showed considerable variation in opinion on their self-management roles. Use of Q-Care Profiles in clinical care is promising, as they catch several important concepts in one short description. Further research should elaborate on the predictive value of the Q-Care Profiles to assess nonadherence or lack of independence.

What's known on this subject: Chronically ill adolescents have different preferences and attitudes toward health care and self-management. Previous Q-methodological research revealed four distinct self-management styles, or Q-Care Profiles: 'Conscious & Compliant', 'Backseat Patient', 'Self-confident & Autonomous', and 'Worried & Insecure'.

What this study adds: The four Q-Care Profiles distinguish between different self-management roles and may help identify risky behaviors. 'Backseat Patients' demonstrate lack of independence, 'Worried & Insecure' persons risk depression, while 'Self-confident & Autonomous' adolescents may be nonadherent to prescribed treatment.

1 Introduction

Adolescence is a critical period for the establishment of health behaviors in youth with chronic conditions.[1-3] As adolescents gradually grow out of the pediatric environment, a new working partnership with physicians and nurses must be formed. They prefer an age-appropriate approach, and welcome being treated as a partner in care.[4] Supporting adolescents through transitions is a major challenge for health care providers and fosters the development of self-management skills enabling to gain more control in, and over life.[5]

The concept of self-management goes beyond (non)adherence to treatment regimens. Self-management in chronic illness encompasses three major tasks: medical management, role management, and emotional management in order to achieve an optimal quality of life.[6,7] Chronically ill adolescents do not only have medical and treatment-related concerns, but also general health and psychosocial needs related to their developmental process of identity construction.[8,9] Most studies into self-management focus on specific diseases, disregarding the many similarities in the lived experiences of youth with different chronic conditions.[8]

Providing adequate self-management support requires studying and taking adolescents' own self-management-related attitudes into account.[9-11] Most pediatricians will agree that there is no 'one size fits all' approach when it comes to stimulating youth with chronic conditions to take responsibility for self-management. It may thus be practical to identify sizeable and meaningful subgroups.[12] Simple classification by 'compliant' versus 'noncompliant' would ignore the complex and multifaceted character of self-care behaviors, implying that more integrative taxonomies of self-management styles are needed. So how to identify these subgroups? Should we go by age, clinical outcomes, developmental milestones, or self-management assessment scales?

Instead, we decided to start with the viewpoints of adolescents themselves. Q-methodology provides a scientific foundation for the systematic study of subjectivity, such as people's opinions, attitudes and preferences.[13-16] Here, individuals represent their viewpoints by ranking a set of statements, after which by-person factor analysis reduces these many viewpoints to a few shared perspectives. A population of viewpoints is described rather than a population of people.[17] Q-methodology has proven to be successful in triggering adolescents to speak freely and extensively about their own views.[18,19] In a previous Q-methodological study we elicited four distinct profiles – or: Q-Care Profiles (QCPs) – regarding chronically ill adolescents' attitudes and preferences toward health care delivery and self-management. We labeled them as: (A) 'Conscious & Compliant'; (B) 'Backseat Patient'; (C) 'Self-confident & Autonomous'; and (D) 'Worried & Insecure'.[20]

However, a Q-methodological study neither informs us about the proportions of people adhering to the identified viewpoints, nor about associations with personal characteristics.[14,16,17] In the present study, the narrative descriptions were presented to a large population of chronically adolescents to survey fit to each QCP and to establish construct validity of the QCPs through exploring associations with outcome variables such as quality of life and self-efficacy. Finally, multivariable associations between preference for a particular QCP and socio-demographic and disease-related variables were explored.

2 Patient and methods

2.1 Participants

Eligible participants were 12-19–year-olds in active long-term treatment (over three years prior to July 1st 2006) for a somatic chronic condition in the departments of Pediatrics or Pediatric Surgery in the Erasmus MC - Sophia Children's Hospital, and who had made at least three outpatient visits and/or been hospitalized in these three years. Two exclusion criteria were applied: already transferred to adult care; documented diagnosis of intellectual impairment.

Approval for the study was obtained from the Erasmus MC Institutional Review Board. Eligible adolescents received written information and a unique access code, and were invited to complete a web-based questionnaire. All received a reminder after three weeks. Response postcards were included to encourage adolescents to state they did not qualify for the study, if this should be the case, or to explain why they did not wish to participate. There was no financial remuneration, although participants were entered in a lottery for two iPods and a cell phone.

2.2 Measures

Outcome variable: The questionnaire included short descriptions of the four QCPs developed in previous research (Appendix A; Chapter 3).[20] Adolescents were asked to assess their degree of fit to each QCP-description on a 5-point Likert scale by the question *"How well does this description fit you?"* (range: 1 = not at all; 2 = not; 3 = a little; 4 = well; 5 = very well). So, adolescents endorsed all QCPs. To avoid undesired response effects, the names of the profiles were left out.

As a first test of face validity, adolescents and health care providers during a study into hospital consultations had been asked whether they recognized the QCP-descriptions.[21] The initial analysis yielded tentative hypotheses about possible relations between each QCP and socio-demographic, disease-related and psychological variables (including quality of life, self-efficacy, and readiness for transfer),[20] so these were included in the questionnaire. The questionnaire was built on findings from a literature review, extensive data-analysis of our previously conducted interviews with 31 adolescents,[22] and pilot tests of the draft questionnaire in face-to-face interviews with 5 adolescents and 4 parents.

Socio-demographic characteristics: Participants' age and gender were retrieved from the hospital database. As ethnicity is not recorded, two researchers [AvS;HvdS] classified the family names into Dutch versus non Dutch using the Dutch Databank of Surnames. Educational level and type of education (regular versus special education for the physically disabled) were obtained through the questionnaire.

Disease-related characteristics: Medical diagnosis**,** numbers of outpatient visits, hospital admissions, and different outpatient departments visited were retrieved from the hospital database. Adolescents themselves were asked to state if their chronic condition had been diagnosed before or after the age of 6, and if they used any prescribed medication, diet or exercises. Presence of physical limitations in mobility was assessed with the 10-item AVO-99 scale[23] that was dichotomized. If any physical limitations were present, this was recoded as 1. General health status and

absenteeism from school or work due to illness in the past year were assessed through 1-item questions on a 5-point Likert scale.

Psychological measures included Health-related Quality of Life (HRQoL), self-efficacy and attitudes toward transition. HRQoL was assessed by the self-report short versions of the cross-culturally validated KIDSCREEN[24] and DISABKIDS[25] questionnaires using 5-point response scales. KIDSCREEN-10 provides a one-dimensional global HRQoL index with good internal consistency (Cronbach's α = .81).[24] The 10-item DISABKIDS chronic generic measure (DCGM-10) measures impact of the chronic disease on quality of life (α = .84).[25]

Validated instruments measuring self-efficacy and attitude toward transition in adolescents with a variety of chronic conditions were not available, so new scales were constructed. Self-efficacy was measured on three domains using 4-point response scales: (1) coping with the condition (8 items); knowledge of the condition (10 items); and skills for independent hospital visits (11 items). General independence during consultations was scored on a visual analogue scale (range 1-10) and independent behavior during last consultation was assessed with 7 dichotomous questions.[26]

Four quotes, taken from adolescents' interviews,[22] were presented to assess transitional attitudes on a 5-point Likert scale. Adolescents' scored their perception of transfer readiness by a single question with 4 response categories.[26]

2.3 Statistical analysis

Data were analyzed using SPSS 18.0 (SPSS Inc., Chicago, IL.). Descriptive statistics with means, standard deviations, and proportions are presented. Reliability (Cronbach's alpha) of scales was calculated. All tests were two-tailed, and *P* values < .05 were considered statistically significant.

The degree of fit to each QCP was checked by calculating sum-scores. Then, bivariate associations between all measures and the QCP-narratives were calculated by Spearman's Rho ρ to test hypotheses about the construct validity of the QCPs.

In order to explore in separate multivariate logistic regression analyses which variables contributed to the degree of fit to each QCP, the scores were dichotomized. Scores 1 (does not fit at all), 2 (does not fit), and 3 (fits a little) were recoded into 0 (no good fit); and the scores 4 (fits well) and 5 (fits very well) were recoded into 1 (good fit). Odds Ratios (OR) and 95% confidence intervals (CI) were calculated. Nagelkerke R^2 was used to express the substantive significance of each model.

Only socio-demographic and disease-related variables were included into the initial multivariate logistic regression analyses; psychological measures were excluded to avoid possible bias with the narratives. Multicollinearity was checked by calculating the Variance Inflation Factor (VIF). Finally, for each profile, a reduced model was calculated using the significant variables from the initial analysis (*P* < .05).

3 Results

3.1 Study population

An initial response was received from 1318 of the 3648 invitees (36.1%). In 231 cases the response was in the form of returning the response card and explaining why they would not participate: 144 (62.3%) claimed they were not chronically ill. Analyses revealed that nonresponders were more frequently males and had non Dutch surnames; they were older and less frequent visitors to the hospital compared to responders ($P < .05$).

Of the 1087 questionnaires received (29.8%), 94 were excluded as the four QCP-narratives were not scored. In 49 questionnaires (4.9%) the same score was assigned to all four QCPs. Subsequently, these were scrutinized. In half of these cases, adolescents endorsed "fits a little" to all QCPs. Three questionnaires were excluded as equal scores were assigned to the next 13 questions as well. The remaining 990 questionnaires were used in the analysis.

Table 1 presents participants' characteristics. Table 2 provides adolescents' responses to the psychological measures. Except for the scale assessing independent behavior during last consultation ($\alpha = .56$), reliability of all scales was good ($\alpha > .70$).

Table 1   *Respondents' characteristics (n = 990)*

Age, *mean (SD), y*	*15.0 (1.9)*
12-15 y, *n* (%)	608 (61.4)
16-19 y, *n* (%)	382 (38.6)
Gender, *n* (%)	
male	430 (43.4)
female	560 (56.6)
Ethnicity, *n* (%)	
Dutch surname	851 (86.0)
non Dutch surname	139 (14.0)
Educational level, *n* (%)[a]	
lower / middle	525 (55.7)
higher	417 (44.3)
Type of education, *n* (%)[b]	
mainstream	852 (90.1)
special education for the disabled	94 (9.9)
Diagnosis after age of six, *n* (% yes)[c]	304 (30.7)
Hospital admissions in past 3 years (yes), n (%)	238 (24.0)
Number of outpatient visits in past 3 years, *mean (SD)*	*17.2 (16.3)*
≤ 12	501 (50.6)
> 13	489 (49.4)

Table 1   *(continued)*

Number of different outpatient departments in past 3 years, *mean (SD)*	*3.1 (2.3)*
1	265 (26.8)
>1	725 (73.2)
Prescribed medications, diet or exercise (yes), *n* (%)	623 (62.9)
Presence of physical limitations (yes), *n* (%)	285 (28.8)
General health score (range: 1-5), *mean (SD)*	*3.6 (1.0)*
School / work absenteeism due to illness (range: 1-5), *mean (SD)*	*1.9 (0.9)*

[a] *missing data: n = 48*

[b] *missing data: n = 44*

[c] *Taken from the International Statistical Classification of Diseases and Related Health Problems (ICD-9 CM), the five largest diagnostic categories in the sample were: congenital anomalies and conditions originating in the perinatal period (26.7%); neoplasms (12.7%), endocrine, nutritional and metabolic diseases including immunity disorders (11.8%); diseases of the nervous system and sense organs (11.4%); and diseases of the musculoskeletal system and connective tissue (10.2%).*

Table 2   *Survey responses[a]*

	n	*mean (SD)*	Cronbach's α
Health-Related Quality of Life			
KIDSCREEN-10 (10 items; range: 10-100)	990	77.0 (16.4)	.83
DCGM-10 (DISABKIDS) (10 items; range: 10-100)	989	80.2 (16.0)	.80
Self-management			
Self-efficacy coping with condition (8 items; range: 8-32)	975	26.7 (4.1)	.78
Self-efficacy knowledge about condition (10 items; range: 10-40)	966	32.5 (4.9)	.78
Self-efficacy skills for independent hospital visits (11 items; range: 11-44)	958	35.0 (6.0)	.87
Independent behavior during last consultation (7 items; range: 0-7)	957	2.5 (1.5)	.56
General score of independence during consultations (1 item; range: 1-10)	957	7.1 (2.0)	
Attitude toward transition			
Attitude toward transition scale (4 items; range: 4-20)	951	12.0 (3.5)	.70
Transfer readiness (1 item; range: 1-4)	951	2.6 (0.9)	

[a] *For each measure, a higher score indicates a better outcome (higher self-efficacy; more positive attitude) or higher degree of absenteeism, etc.*

3.2   Distribution of QCPs

Four out of every five (79.4%) adolescents felt that at least one of the four profiles fitted them well or very well; the others (20.6%) did not score fit to any profile as well or very well, implying that no profile description fitted them well. Only 4.2% assigned a good or very good fit to more than two QCPs.

Figure 1 shows that fit to the 'Conscious & Compliant' profile was most prevailing: 56.1% of adolescents indicated this profile fitted them (very) well. Only 16.4% indicated a (very) good degree of fit to the 'Backseat Patient' profile, while 26.4% recognized themselves (very) much in the 'Self-confident & Autonomous' profile and 24.6% in the 'Worried & Insecure' profile.

Three QCPs were inter-correlated, but only weakly (Table 3). Profile 'Conscious & Compliant' was positively correlated with profile 'Self-confident & Autonomous' (ρ = .27, P < .001), but negatively with the 'Backseat Patient' and 'Worried & Insecure' profiles.

Figure 1   *Distribution of degree of fit to Q-Care Profiles (n = 990)*
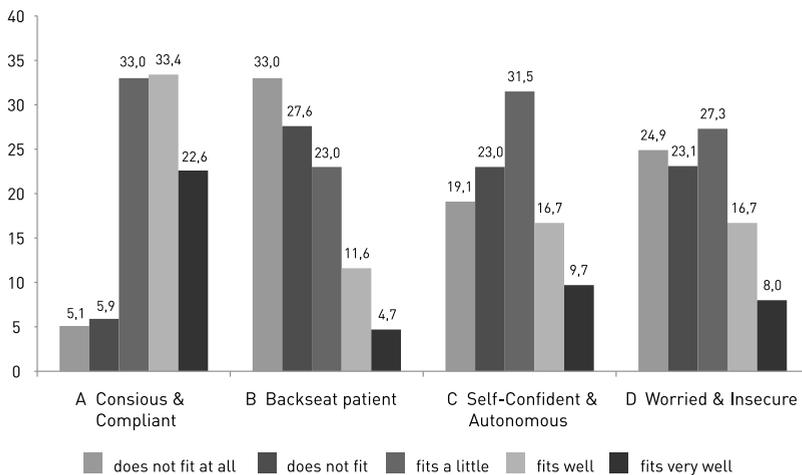
Table 3   *Correlations between degree of fit to Q-Care Profiles[a] (n = 990)*

	Conscious & Compliant		Backseat Patient		Self-confident & Autonomous	
	ρ	*P*	ρ	*P*	ρ	*P*
Backseat Patient	-.07	.03	-			
Self-confident & Autonomous	.27	< .001	.03	.34	-	
Worried & Insecure	-.12	< .001	.14	< .001	-.05	.13

[a] *Spearman's Rho ρ; P = two-tailed.*

3.3 Convergent validity of QCPs with other measures

Table 4 presents bivariate associations between fit to each QCP and all measures. In this way, convergent correlations were explored to determine whether the QCP-assessments are related to concepts that they should theoretically be related with including self-efficacy, HRQoL, attitude toward transition and transfer readiness.

'Conscious & Compliant' Profile. Feeling a good fit with this profile was associated with higher age, having a Dutch surname, and a higher educational level. These adolescents also attended regular education more often. They had lower number of outpatient visits in the past three years, fewer hospital admissions, and were less likely to have a therapeutic regimen. The 'Conscious & Compliant' adolescents also presented with fewer physical limitations, a higher general health score, and less work / school absenteeism indicating they suffered less impact of the chronic condition. This was confirmed by their higher scores on HRQoL (DCGM-10). The adolescents in this profile also scored higher self-efficacy in all domains, demonstrated more independent behaviors during consultations and rated their independence higher. They were also more ready for readiness and displayed a positive attitude toward transition.

'Backseat Patient' Profile. This profile was associated with lower age, male gender, and Dutch ethnicity. They had a lower educational level and more often frequented special education. 'Backseat patient' adolescents experienced more hospital admissions and suffered more often from physical limitations and absenteeism. Their HRQoL (KIDSCREEN) score is also lower. They are less self-efficacious, demonstrate fewer independent behaviors during consultations, and feel less ready for the transfer to adult care. Also, their attitude toward transition is less positive.

'Self-confident & Autonomous' Profile. Few correlations tested significant for this profile. These adolescents were older and less likely to have a therapeutic regimen. They experienced higher HRQoL (as measured with DCGM-10), as well as higher self-efficacy for skills for hospital visits. They scored their own independence during consultations higher than others. Like the 'Conscious & Compliant' profile, they had a higher transfer readiness, and a more positive attitude toward transition.

'Worried & Insecure' Profile. This profile appealed more to girls with non Dutch surnames who attended lower educational levels and special education more often. They had more outpatient visits and more often had a therapeutic regimen. Like in the 'Backseat Patient', having physical limitations and school absenteeism was associated with this profile. Also, their HRQoL as measured with KIDSCREEN and DCGM-10 was significantly lower, as well as their self-efficacy with respect to knowledge the condition and skills for independent hospital visits. In contrast, they feel more self-efficacious in coping with their condition. Their attitude toward transition and transfer readiness were less positive and they had lower rated independency during consultations.

Table 4 *Bivariate associations with Q-Care Profile scores*[a,b]

	n	Conscious & Compliant		Backseat Patient		Self-confident & Autonomous		Worried & Insecure	
		ρ	P	ρ	P	ρ	P	ρ	P
Socio-demographic characteristics									
Age	990	.14	<.001	-.26	<.001	.08	.02	-.06	.06
Gender (male)	990	.04	.19	.07	.03	.01	.78	-.11	<.001
Ethnicity (non Dutch surname)	990	-.11	<.001	-.10	.002	-.02	.50	.08	.01
Educational level (higher)	942	.14	<.001	-.13	<.001	.03	.37	-.16	<.001
Type of education (special education)	946	-.10	.002	.14	<.001	-.07	.05	.11	.001
Disease-related characteristics									
Diagnosis after age of six	988	.01	.75	-.06	.07	-.01	.78	.03	.43
Number of outpatient visits in past 3 years	990	-.07	.04	.06	.06	-.04	.17	.07	.03
Number of different outpatient departments	990	-.06	.05	.07	.03	-.06	.08	.09	.004
Hospital admissions in past 3 years	990	-.09	.004	.10	.002	-.05	.15	.02	.63
Prescribed medications, diet or exercise	990	-.10	.002	.03	.35	-.09	.007	.22	<.001
Presence of physical limitations	990	-.11	<.001	.10	.001	.09	.79	.16	<.001
General health score	990	.13	<.001	-.01	.72	.04	.26	-.25	<.001
School / work absenteeism due to illness	990	-.10	.002	.08	.02	-.03	.38	.15	<.001
Health-Related Quality of life									
KIDSCREEN-10	990	.06	.07	.07	.04	-.02	.45	-.19	<.001
DCGM-10 (DISABKIDS)	990	.18	<.001	-.04	.19	.10	.003	-.37	<.001

Table 4 *(continued)*

	n	Conscious & Compliant		Backseat Patient		Self-confident & Autonomous		Worried & Insecure	
		ρ	P	ρ	P	ρ	P	ρ	P
Self-management									
Self-efficacy coping with condition	975	.19	<.001	-.05	.14	.05	.15	.23	<.001
Self-efficacy knowledge about condition	966	.20	<.001	-.17	<.001	.04	.20	-.13	<.001
Self-efficacy skills for independent hospital visits	958	.25	<.001	-.32	<.001	.18	<.001	-.19	<.001
Independent behavior during last consultation	957	.17	<.001	-.27	<.001	.05	.16	.01	.85
General score of independence during consultations	957	.20	<.001	-.32	<.001	.09	.004	-.14	<.001
Attitude toward transition									
Attitude toward transition scale	951	.16	<.001	-.10	.002	.11	.001	-.14	<.001
Transfer readiness	951	.18	<.001	-.28	<.001	.14	<.001	-.14	<.001

a *Spearman's Rho ρ; P = two-tailed.*
b *Using 5-point Likert scale: 1 = does not fit at all; 2 = does not fit very well; 3 = fits a little; 4 = fits well; 5 = fits very well.*

4

3.4   Multivariate associations

Separate logistic regression analyses served to test the contribution to adolescents' fit to each QCP of all independent socio-demographic and disease-related variables listed in Table 4 that were significantly associated with any one QCP (Table 5). As age at diagnosis was not significantly correlated with any of the profiles, this variable was not included in the multivariate model. In the multivariate model, average VIF was 1.30 while no individual VIFs exceeded 2.20, indicating that multicollinearity was not a problem.

In the final reduced models (Table 6), only significant variables in the original multivariate analyses ($P < .05$) were included. Those adhering to the 'Conscious & Compliant' profile ($R^2 = .08$) are typified by being older, of Dutch descent, and having better general health. Explained variance was highest in the 'Backseat Patient' profile ($R^2 = .15$). This fitted better when adolescents were younger and of Dutch ethnicity, had a lower educational level, followed special education, and experienced physical limitations. No variables contributed significantly to a good fit with the profile 'Self-confident & Autonomous' ($R^2 = .01$). The 'Worried & Insecure' description ($R^2 = .11$) more often attracted girls, those who were younger, of non Dutch ethnicity, lower educated and with poorer general health than those not attracted to this profile.

4   Discussion

This study is the first to research self-management styles representing different attitudes and roles in a large sample of chronically ill adolescents. Four out of every five adolescents could identify well with at least one of the profile descriptions. The four Q-Care Profiles distinguished well with only limited overlap between them. They differed in proportions of respondents attracted to them and in their associations with other variables. Previous hypotheses about the correlations with the profiles were confirmed; except for the influence of disease duration.[20] The bivariate correlations tested fitted the expected pattern, thus contributing evidence of construct validity. Most respondents felt that the 'Conscious & Compliant' profile fitted them well, indicating that it would be possible to motivate chronically ill adolescents to take (more) responsibility for self-management and to be concordant patients. However, these respondents were generally older and higher educated than others. They also had better general health and experienced less impact of the disease. 'Backseat Patients' typically displayed lack of independence and self-efficacy, but tended to be younger and may be expected to still develop these competencies. Also, physically disabled and lower educated youth attending special education were overrepresented in this group, indicating they need more encouragement to take up self-management responsibilities. Most at risk for nonadherence seems to be the 'Self-confident & Autonomous' adolescent representing another one-quarter of respondents. This profile could not be predicted from socio-demographic or disease-related variables. Although feeling self-efficacious in handling their own affairs, they seem critical of professional advice. The one-quarter of respondents who best fitted into the 'Worried & Insecure' profile were typically younger girls, of non Dutch background and lower educated. They

Table 5 *Multivariable logistic regression analyses with degree of fit to Q-Care Profile scores[a,b] (n = 941)*

	Conscious & Compliant		Backseat Patient		Self-confident & Autonomous		Worried & Insecure	
R^2 (Nagelkerke)	.10		.16		.03		.13	
	OR (95% CI)	P	OR (95% CI)	P	OR (95% CI)	P	OR (95% CI)	P
Age	1.77(1.33-2.35)	<.001	0.35 (0.23-0.55)	<.001	1.35 (0.99-1.83)	.06	0.68 (0.48-0.95)	.02
Gender (male)	1.26 (0.95-1.66)	.11	1.25 (0.85-1.83)	.25	1.22 (0.91-1.66)	.19	0.67 (0.48-0.94)	.02
Ethnicity (non Dutch surname)	0.51 (0.34-0.76)	.001	0.32 (0.15-0.66)	.002	0.75 (0.47-1.20)	.23	1.60 (1.04-2.46)	.03
Educational level (higher)	1.64 (1.24-2.17)	.001	0.46 (0.30-0.70)	<.001	0.96 (0.71-1.31)	.81	0.48 (0.34-0.68)	<.001
Type of education (special education)	0.67 (0.41-1.08)	.10	3.26 (1.91-5.55)	<.001	0.70 (0.39-1.24)	.22	1.19 (0.71-1.98)	.52
Number of outpatient visits in past 3 years	1.01 (1.00-1.02)	.22	1.00 (0.98-1.02)	.83	1.01 (1.00-1.03)	.07	1.01 (0.99-1.02)	.44
Number of different outpatient departments	0.93 (0.86-1.02)	.13	0.99 (0.88-1.12)	.92	0.89 (0.80-0.98)	.02	1.00 (0.91-1.11)	.98
Hospital admissions in past 3 years	0.99 (0.97-1.00)	.11	1.00 (0.97-1.02)	.86	0.99 (0.97-1.01)	.31	0.98 (0.96-1.01)	.20
Prescribed medications, diet or exercise	0.88 (0.65-1.18)	.39	1.11 (0.73-1.69)	.64	0.83 (0.60-1.14)	.25	1.33 (0.93-1.91)	.12
Presence of physical limitations	0.92 (0.67-1.26)	.60	1.59 (1.05-2.43)	.03	1.27 (0.89-1.80)	.19	1.30 (0.91-1.85)	.15
General health score	1.22 (1.04-1.42)	.01	1.13 (0.91-1.40)	.26	1.11 (0.94-1.32)	.21	0.79 (0.66-0.95)	.01
School / work absenteeism due to illness	0.95 (0.81-1.12)	.52	1.23 (0.98-1.53)	.07	1.02 (0.85-1.22)	.88	1.25 (1.05-1.50)	.01

[a] The multivariate analysis for each profile adjusted for all variables.
[b] Compiling the scores of the profiles 1 = does not fit at all; 2 = does not fit very well; and 3 = fits a little into NO GOOD FIT; and 4 = fits well; 5 = fits very well into GOOD FIT.

4

Table 6 *Multivariable logistic regression analyses with degree of fit to Q-Care Profile scores (final reduced models)[a, b]*

	n	OR (95% *CI*)	*P*
Conscious & Compliant; R^2 = .08	**942**		
Age		1.70 (1.29-2.25)	<.001
Non Dutch surname		0.50 (0.34-0.74)	.001
Educational level (higher)		1.77 (1.35-2.32)	<.001
Hospital admissions in past 3 years		0.99 (0.97-1.00)	.09
General health score		1.30 (1.13-1.49)	<.001
Backseat Patient; R^2 = .15	**941**		
Age		0.34 (0.22-0.53)	<.001
Non Dutch surname		0.33 (0.16-0.67)	.002
Educational level (higher)		0.46 (0.30-0.70)	<.001
Type of education (special)		3.35 (2.00-5.62)	<.001
Presence of physical limitations		1.57 (1.06-2.32)	.03
Self-confident & Autonomous; R^2 = .01	**990**		
Number of different outpatient departments		0.94 (0.88-1.01)	.08
Worried & Insecure; R^2 = .11	**942**		
Age		0.68 (0.49-0.94)	.02
Gender (male)		0.67 (0.48-0.93)	.02
Non Dutch surname		1.60 (1.04-2.46)	.03
Educational level (higher)		0.46 (0.33-0.64)	<.001
General health score		0.74 (0.63-0.88)	.001
School / work absenteeism due to illness		1.30 (1.10-1.54)	.002

[a] *The multivariate analysis for each profile adjusted for all variables.*
[b] *Compiling the scores of the profiles 1 = does not fit at all; 2 = does not fit very well; and 3 = fits a little into NO GOOD FIT; and 4 = fits well; 5 = fits very well into GOOD FIT.*

had lower HRQoL and general health scores, and reported more school absenteeism – indicating they are vulnerable for depression, social isolation and psychosocial problems.

Self-management encompasses the ability to affect the cognitive, behavioral and emotional responses necessary to maintain a positive quality of life.[7] The QCPs compile these varying responses into comprehensive narratives that reflect the challenges of growing up with a chronic condition. The low explained variance indicates that self-management styles cannot be predicted easily and that pediatricians should inquire directly into them. Establishing adolescents' fit with these four QCP-descriptions and discussing the outcomes could help identify risky attitudes and support adolescents' emerging capacity for self-management.[27]

We found only two comparable studies taking a profile-based approach assessing self-management styles in youth with diabetes.[28, 29] Cluster analysis revealed three distinct styles to achieve

glycemic control: inadequate, moderate and high adaptive style. However, these studies were restricted to adherence to diabetic treatment and did not include adolescents' opinions on barriers and facilitators for self-management.[30-32] In contrast, the QCPs reflect adolescents' own perspectives and not those of clinicians.

4.1   Strengths and limitations of the study

A large heterogeneous sample of adolescents originating from a university hospital in the Netherlands assessed fit to the QCPs. However, the nonresponse rate was fairly high (64%), and a nonresponse analysis revealed that older adolescents, boys, and those with non Dutch surnames were underrepresented in this self-selected sample. Because the QCP-distribution proved associated with gender, age and ethnicity, this skewed representation might have affected the outcomes.

The QCP-narratives transcended specific conditions and demonstrated the value of a generic approach.[8] Consequently, however, (differences in) the distribution of QCPs in specific diseases and relations with clinical outcomes or disease severity could not be explored, so it remains unknown whether the QCPs accurately predict treatment adherence.

An advantage of presenting the four previously constructed narratives was that the coherence of the QCPs as a package was preserved.[17] The QCPs reflect complex self-management attitudes, preferences and roles that are not restricted to adherence. There is only limed overlap between them. Construct validity could be established and relations were in the expected direction. However, as one in five adolescents could not identify well with any of the QCPs, we could have missed other profiles. Also, adolescents could assign themselves to multiple profiles. Based on their scores, we have tried to assign them to one profile only. This proved possible in about two thirds of all cases (68%; data not presented here), indicating that not all adolescents can be classified into one of the QCPs. Also, the lack of a gold standard and of other studies distinguishing self-management styles limited exploration of the predictive validity of the QCPs.

Furthermore, the explained variance in the multivariate regression analyses was quite low, suggesting that there are other variables – not included in our data set – that could explain adolescents' self-management attitudes and preferences. This deserves further study, as well as the question whether adolescents' identification with the QCPs changes over time.

4.2   Clinical implications

Clinicians should be aware of the existence of distinct self-management styles among chronically ill adolescents. Since educational interventions alone are insufficient to promote adherence, adding a behavioral component to adherence interventions could increase potential efficacy.[33] The QCPs may provide this. Inviting adolescents to score their fit to the QCPs during consultations could be a first step towards self-exploration of their attitudes and self-management roles. Only one profile, 'Conscious & Compliant', sets a positive example for the others because it scores well on independence, self-efficacy, quality of life, and on intention to treatment adherence. The three other QCPs involve potential risks for self-management failure. While all adolescents should be encouraged to learn new skills and would benefit from action plans, transition readiness assessments,[26, 34, 35] and

independent hospital consultations,[21, 36, 37] this is particularly relevant for 'Backseat Patients' and 'Worried & Insecure' adolescents. They seem not ready for self-management or transfer to adult care, and 'Worried & Insecure' adolescents need extra attention to their psychosocial needs. Most challenging is the 'Self-confident & Autonomous' type as they may be inclined to ignore providers' advice. A careful exploration of adolescents' views, for example through Motivational Interviewing,[38] may contribute to positive outcomes.

5   Conclusion

Considerable variation between adolescents with chronic conditions with respect to their self-management styles was established. Associations were found with age, gender, ethnicity, education, and general health but not with disease duration. Use of QCPs in clinical practice may be promising as these seem to catch several important concepts in one short description and invite adolescents to discuss their opinions. Further research into the predictive value of the QCP-descriptions is warranted.

Acknowledgements

References

1. Viner RM, Barker M. Young people's health: the need for action. *BMJ*. 2005;330(7496):901-903.
2. McDonagh JE, Viner RM. Lost in transition? Between paediatric and adult services. *BMJ*. 2006;332(7539):435-436.
3. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child*. 2008;93(2):160-163.
4. van Staa A, Jedeloo S, van der Stege H, On Your Own Feet Research Group. "What we want": chronically ill adolescents' preferences and priorities for improving health care. *Patient Preference and Adherence*. 2011;5(1):291-305.
5. American Academy of Pediatrics, American Academy of Family Physicians and American College of Physicians, Transitions Clinical Reporting Group. Supporting the health care transition from adolescence to adulthood in the medical home. *Pediatrics*. 2011;128(1):182-200.
6. Lorig KR, Holman H. Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med*. 2003;26(1):1-7.
7. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J. Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns*. 2002;48(2):177-187.
8. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet*. 2007;369(9571):1481-1489.
9. Kyngas H. Compliance of adolescents with chronic disease. *J Clin Nurs*. 2000;9(4):549-556.
10. Britto MT, Slap GB, DeVellis RF, Hornung RW, Atherton HD, Knopf JM, et al. Specialists understanding of the health care preferences of chronically ill adolescents. *J Adolesc Health*. 2007;40(4):334-341.
11. Wray J, Maynard L. Specialist cardiac services: what do young people want? *Cardiol Young*. 2008;18(6):569-574.
12. La Greca AM. Issues in adherence with pediatric regimens. *J Pediatr Psychol*. 1990;15(4):423-436.
13. Cross RM. Exploring attitudes: the case for Q methodology. *Health Educ Res*. 2005;20(2):206-213.
14. Watts S, Stenner P. Doing Q methodology: theory, method and interpretation. *Qual Res Psychol* 2005;2(2):67-91.
15. Akhtar-Danesh N, Baumann A, Cordingley L. Q-methodology in nursing research: a promising method for the study of subjectivity. *West J Nurs Res*. 2008;30(6):759-773.
16. Brown SR. *Political subjectivity: applications of Q methodology in political science*. New Haven, CT: Yale University Press; 1980.
17. Baker RM, van Exel J, Mason H, Stricklin M. Connecting Q & surveys: a test of three methods to explore factor membership in a large sample. *Operant Subjectivity*. 2010;34(1):38–58.
18. Tielen M, van Staa AL, Jedeloo S, van Exel NJ, Weimar W. Q-methodology to identify young adult renal transplant recipients at risk for nonadherence. *Transplantation*. 2008;85(5):700-706.
19. van Exel NJ, de Graaf G, Brouwer WB. "Everyone dies, so you might as well have fun!" Attitudes of Dutch youths about their health lifestyle. *Soc Sci Med*. 2006;63(10):2628-2639.
20. Jedeloo S, van Staa A, Latour JM, van Exel NJ. Preferences for health care and self-management among Dutch adolescents with chronic conditions: a Q-methodological investigation. *Int J Nurs Stud*. 2010;47(5):593-603.
21. van Staa A, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: the added value of mixed methods research. *Patient Educ Couns*. 2011;82(3):455–464.
22. van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam [Rotterdam University]; 2007. ISBN 978-90-8017-769-7.
23. de Klerk M, Iedema J, van Campen C. *SCP-maat voor lichamelijke beperkingen op basis van AVO 2003. [Measure for physical impairments based on AVO-2003].* SCP-werkdocument 121. Den Haag: Sociaal Cultureel Planbureau; 2006. ISBN 90-377-0268-6.
24. Ravens-Sieberer U, Erhart M, Rajmil L, Herdman M, Auquier P, Bruil J, et al. Reliability, construct and criterion validity of the KIDSCREEN-10 score: a short measure for children and adolescents' well-being and health-related quality of life. *Qual Life Res*. 2010;19(10):1487-1500.

25. Schmidt S, Petersen C, Mühlan H, Simeoni MC, Debensason D, Thyen U, et al. *The DISABKIDS Questionnaires Handbook*. Lengerich: Pabst Science Publishers; 2006. ISBN 3-89967-166-X.

26. van Staa AL, van der Stege HA, Jedeloo S, Moll HA, Hilberink S. Readiness to transfer to adult care of adolescents with chronic conditions: Exploration of associated factors. *J Adolesc Health*. 2011;48(3):295–302.

27. Sawyer S, Drew S, Duncan R. Adolescents with chronic disease--the double whammy. *Aust Fam Physician*. 2007;36(8):622-627.

28. Rohan JM, Delamater A, Shroff Pendley J, Dolan L, Reeves G, Drotar D. Identification of self-management patterns in pediatric type 1 diabetes using cluster analysis. *Pediatr Diabetes*. 2011;12(7):611-618.

29. Schneider S, Iannotti RJ, Nansel TR, Haynie DL, Simons-Morton B, Sobel DO, et al. Identification of distinct self-management styles of adolescents with type 1 diabetes. *Diabetes Care*. 2007;30(5):1107-1112.

30. George M, Rand-Giovannetti D, Eakin MN, Borrelli B, Zettler M, Riekert KA. Perceptions of barriers and facilitators: self-management decisions by older adolescents and adults with CF. *J Cyst Fibros*. 2010;9(6):425-432.

31. Rhee H, Belyea MJ, Ciurzynski S, Brasch J. Barriers to asthma self-management in adolescents: Relationships to psychosocial factors. *Pediatr Pulmonol*. 2009;44(2):183-191.

32. Hanna KM, Guthrie D. Adolescents' perceived benefits and barriers related to diabetes self-management--Part 1. *Issues Compr Pediatr Nurs*. 2000;23(3):165-174.

33. Dean AJ, Walters J, Hall A. A systematic review of interventions to enhance medication adherence in children and adolescents with chronic illness. *Arch Dis Child*. 2010;95(9):717-723.

34. Fredericks EM, Dore-Stites D, Well A, Magee JC, Freed GL, Shieck V, et al. Assessment of transition readiness skills and adherence in pediatric liver transplant recipients. *Pediatr Transplant*. 2010;14(8):944-953.

35. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, et al. Measuring the transition readiness of youth with special healthcare needs: validation of the TRAQ--Transition Readiness Assessment Questionnaire. *J Pediatr Psychol*. 2011;36(2):160–171.

36. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: a multicentre cohort of adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2005;44(6):806-812.

37. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2007;46(1):161-168.

38. Naar-King S, Suarez M. *Motivational Interviewing with adolescents and young adults*. New York: The Guilford Press; 2011.

4

Part 2

Competencies for care

5

Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: the added value of mixed methods research

AnneLoes van Staa, On Your Own Feet Research Group

Abstract

OBJECTIVE: To integrate findings of a mixed methods research into adolescents' preferences and competencies for communication during consultations, in order to demonstrate the added value of MMR for health communication research.

METHODS: Sequential mixed methods research with adolescents (12-19 yrs) with various chronic conditions in a university hospital. Methods comprised: (1) 31 face-to-face interviews, (2) Q-methodology; (3) 39 observations of outpatient consultations, (4) three focus groups with 27 health care providers, and (5) web-based questionnaire in 960 adolescents.

RESULTS: Adolescents had different preferences regarding health communication, but all wished to be involved as partners. Yet, their actual participation during consultations was low. They often acted as bystanders rather than main characters because their participation was neither requested nor encouraged. Parents filled the gap, to health care providers' frustration. The questionnaire confirmed the discrepancy between self-efficacy and self-reported independent behavior during consultations.

CONCLUSION: Triadic communication was all but multi-party-talk and adolescents did not act and were not considered as main partners. Mixed methods research was of pivotal importance for our understanding.

PRACTICE IMPLICATIONS: As chronically ill adolescents need to prepare themselves for transition to adult care, health care providers should encourage them to take the lead in communication by initiating independent visits and changing the parents' roles.

1   Introduction

Medical interaction should be studied in the context of time, setting and participants.[1, 2] This plea is particularly relevant in pediatric consultations, where at least one parent is likely to be present next to the child and the doctor. Triadic communication and opportunity for partnership is framed, first and foremost, by the 'ceremonial order of the clinic'[2, 3]: the organizational and legal setting of pediatric clinics.[4]

Research into triadic communication in the past decades has shown that the child's contribution is rather limited, seeing that doctors control the turn-taking and parents control their child's participation.[4-11] Doctors,[5] nurses,[12, 13] or dieticians[5, 9] appear to address the parent more than the child; parents frequently take over when the doctor turns to the child,[11, 14] whereas children have very little say.[5] Parental speaking for the child is institutionally co-constructed: it is hardly ever questioned by children and ratified by doctors.[15] So, children's communication competence is not only dependent upon their own capacity, but also on parents' and health care professionals' attitudes.[16, 17]

Most studies on triadic communication were conducted in general practice[14, 15, 17-19]; some in outpatient settings,[4, 6, 10, 11] or inpatient wards.[20, 21] Improving doctor-patient communication and establishing patient partnership is especially relevant in the context of chronic illness[22]. Youth with chronic conditions are rarely consulted, however, about their views on and preferences for involvement in communication.[23, 24] Most research has focused on school-aged children (6-12 years),[5, 6, 11, 14, 15, 17, 19, 25, 26] while relatively few observational studies involved adolescents (eg, children over 12 years of age).[10, 11, 20, 23, 27] As children mature, they achieve greater competence for making independent decisions[16] and increasingly want to be involved in decision making.[28, 29] Also, the Dutch Medical Treatment Act (WGBO; 1995) grants adolescents aged 12-15 the right to codecide with parents in treatment decisions and to decide for themselves when they are over 16. Adolescents with chronic conditions on the way to adult care should be trained and empowered to become effective partners in their own health care communication.[30] This does not only improve interaction, but is also enhances diagnostic and therapeutic processes.[1]

In a project entitled 'On Your Own Feet' we studied these adolescents' preferences and competencies for health care communication. We chose a flexible, multi-method design to account for the complexity of a multi-party context. Mixed Methods Research (MMR) has the potential to access knowledge or insights unavailable to a qualitative study or a quantitative study undertaken independently. MMR is defined as a single study in which qualitative data collection and/or analysis is combined with quantitative data collection and/or analysis either in a concurrent or sequential design.[31] MMR has become popular,[32] but the basic requirements for a good MMR study are much debated.[33] MMR has even been designated the third methodological paradigm: an intellectual and practical synthesis based on qualitative and quantitative research.[34] Integration of different strands of research is the biggest methodological challenge.[35] The key issue is whether the end product is really more than the sum of the individual quantitative and qualitative parts.

We chose MMR for several reasons. First, *comprehensiveness*: using different methods to address different aspects of the overall research question. Second, *improvement of validity and generalizability* of findings: by combining qualitative and quantitative methods, inherent weak-

nesses of each methodology could be compensated for.[31, 32] Third, as our ultimate aim was to give adolescents a voice, *patient-centeredness* was another justification for MMR.[32]

The objective of this paper is twofold: (1) to integrate findings of a MMR study into preferences and competencies for hospital consultations of adolescents with chronic conditions; (2) to demonstrate whether the mixed methods approach has added value in triadic health communication research in pediatric settings. We followed the guidelines for Good Reporting of A Mixed Methods Study (Box 1).[33]

Box 1   *Quality criteria for Good Reporting of A Mixed Methods Study (GRAMMS) (taken from O'Cathain et al 2008)[33]*

---

1   Describe the justification for using a mixed methods approach to the research question
2   Describe the design in terms of the purpose, priority and sequence of methods
3   Describe each method in terms of sampling, data collection and analysis
4   Describe where integration has occurred, how it has occurred and who has participated in it
5   Describe any limitation of one method associated with the presence of the other method
6   Describe any insights gained from mixing or integrating methods

---

2   Methods

2.1   General design and setting

The research project 'On Your Own Feet' employed a sequential strategy of inquiry.[31] All studies were performed in the Erasmus MC-Sophia Children's Hospital, the largest tertiary referral centre in the Netherlands. The project was originally designed as a participatory, multi-method qualitative study, but later extended with a follow-up a questionnaire to strengthen the outcomes. Overall aim of 'On Your Own Feet' was to map preferences for health care delivery and competencies required to enable adolescents with chronic conditions to independently direct their own treatment (self-management).

In the study sample we aimed to include all patients between 12-19 years of age with somatic chronic conditions who had visited this hospital at least once in the past six months and had been under continuous treatment for the past three years. However, those with intellectual disabilities, and those already transferred to adult care, were excluded. The study was conducted in five consecutive steps, presented in Figure 1 and further described in sections 2.2-2.6.

Figure 1 *Mixed methods research 'On Your Own Feet'*

Qualitative research	**I**	Semi-structured *interviews* with youth with chronic conditions (*n* = 31; 12-19 yrs) [2004-2005]
	II	*Q-methodology* with same interviewees (*n* = 31; 12-19 yrs) [2004-2005]
	III	*Observations* of medical and nursing consultations in 8 outpatient clinics (*n* = 39; 12-22 yrs) [2006]
	IV	*Focus group discussions* (*n* = 3) with 27 health care providers [2006]
Quantitative research	**V**	*Web-based questionnaire* in 1087 adolescents with chronic conditions (12-19 yrs) [2006-2007]

2.2   Interviews

We distinguished between two age groups: younger adolescents (12 to 15-year-olds, *n* = 1191) and older adolescents (16 to 19-year-olds, *n* = 1011). To facilitate purposive sampling, equal numbers of random cases were drawn from both groups and then we aimed at equal numbers of participants, and even distributions of sex, hospital experience, and nature of the condition (ie, congenital or acquired in past five years, physically disabling or not) within both age groups.

Semistructured interviews were conducted either by pairs of purpose-trained nursing and paramedical students or by a researcher. Adolescents were interviewed alone at their homes. They were asked to describe their most recent consultation in the hospital and to explain their preferences for their own and their parents' roles in communication. Interviews were audiotape recorded and lasted between 45-90 minutes.

Interviews were transcribed verbatim and imported into the qualitative software package ATLAS. ti 5.0 (www.atlasti.com) using a method of constant comparison, which involved comparing and contrasting incidents in the data to develop themes. A first coding frame, developed on the basis of the interview guide, was continuously modified and expanded as new themes emerged during thematic analysis.[36] The coding frame was flexible – a combination of induction and deduction.

Thirty-one of the 66 invited adolescents (47%) consented to an interview. Data on responders and nonresponders have been published elsewhere, revealing no significant differences between participants and nonparticipants.[37] Socio-demographic characteristics are summarized in Table 1. Five adolescents presented with surgical conditions, two were chronically disabled and the others had a variety of chronic illnesses.

Table 1 *Adolescents' socio-demographic and disease-related characteristics in MMR On Your Own Feet[a]*

	Interviews / Q-study *n* = 31		Observations *n* = 30		Questionnaire *n* = 960	
	n	%	*n*	%	*n*	%
Gender						
girls	15	48.4	10	33.3	539	56.1
boys	16	51.6	20	66.7	421	43.9
Age						
12-15 yrs	17	54.8	17	56.7	588	61.3
16-19 yrs[b]	14	45.2	13	43.4	372	38.8
mean (SD)	*15.3 (2.1)*		*15.6 (2.1)*		*15.0 (1.9)*	
Ethnicity						
Dutch surname	26	83.9	21	70.0	832	86.7
non Dutch surname	5	16.1	9	30.0	128	13.3
No. of outpatient visits in past three years						
≤ 12	19	61.3	13	43.3	488	50.8
≥ 13	12	38.7	17	56.7	472	49.2
Hospital admissions in past three years						
yes	12	38.7			233	24.3
no	19	61.3			727	75.7
missing data			30	100.0		
Diagnosis after age of six[c]						
yes	9	29.0	5	16.7	293	30.5
no (at birth & first 5 yrs)	22	71.0	25	83.3	666	69.4
missing data					1	.1
Therapeutic regimen (ie, medication, diet or exercise)						
yes	24	77.4	30	100.0	605	63.0
no	7	22.6	-		355	37.0
Presence of physical limitations						
yes	11	35.5	5	16.7	273	28.4
no	20	64.5	25	83.3	687	71.6

[a] *All data of interview and observation participants were collected directly. For the questionnaire, gender, age, ethnicity, and hospital visits were retrieved from the electronic hospital database; the other data were self-reported.*
[b] *One observation included a 22-year old man with CF; all others were ≤ 19 years of age*
[c] *Diagnoses represented in the interview study: scoliosis/kyphosis, facial schisis, benign intracranial hypertension, congenital bladder disorder, congenital heart disorders, rheumatoid arthritis, hemophilia, diabetes mellitus (DM), epilepsy, inflammatory bowel disease (IBD), cystic*

fibrosis (CF), various metabolic disorders, HIV, nephrotic syndrome, immune and hormone deficiencies, lung insufficiency, progressive kidney failure, congenital skin diseases, asthma, and neuromuscular diseases. Observations included adolescents with DM, hemophilia, HIV, IBD, CF, neuromuscular diseases, nephrology and metabolic diseases. In the questionnaire, the five largest diagnostic categories (ICD-classification) were: congenital anomalies and conditions originating in the perinatal period (26.6%); neoplasm (12.9%); endocrine, nutritional, metabolic diseases, and immunity disorders (11.6%); diseases of the nervous system and sense organs (11.4%); and diseases of the musculoskeletal system and connective tissue (10.2%).

2.3   Q-methodology

At the end of each interview (2.2), Q-methodology was employed. This is a small-sample, yet robust technique for the measurement of attitudes, combining qualitative and quantitative approaches to explore patterns in individual preferences.[37-40] Participants rank-ordered 37 statements about preferences for health care delivery and self-management on a score sheet using a quasi-normal distribution. Examples of statements related to the organization of consultations and health communication are given in Box 2. After having rank-ordered the statements into "most agree", "neutral", and "least agree", participants were asked to explain their ranking. By-person factor analysis (centroid factor extraction with varimax rotation), using PQMethod 2.11, was conducted to uncover patterns in the rankings of statements, described as Q-Care profiles.[37]

Box 2   *Examples of statements related to health communication and consultations used in Q-methodological study (Jedeloo et al 2010)[37]*

#2   It would be nice if you could also talk to the doctor or nurse in private, without your parents being present
#4   Health care professionals should not ask me personal questions in front of my parents
#6   It's important for me to have my parents present during consultations
#12 I would like health care professionals to treat me like an adult
#16 During consultations, I find it convenient if my parents do the talking for me
#22 Health care professionals should not be overprotective

2.4   Observations

Over 12-year-olds who visited a selected outpatient clinic on specific days were invited if they met the inclusion criteria stated in 2.1. Nonparticipant observations were conducted by six trained nurse specialists and four researchers. The nurse specialists did not observe consultations in their own subspecialties, nor observed doctors or nurses they worked with on a regular basis. Immediately after each observation they were debriefed by a researcher. Afterwards, all observers filled out structured forms about the adolescents' level of involvement in communication and demonstrated competencies. Conversations were audiotape recorded, transcribed and thematically

analyzed. Goffman's analysis of frames and interaction rituals, employing the theatre as a natural metaphor, was used.[3, 41]

Thirty adolescents were observed during 39 consultations in 8 outpatient clinics (diabetes, hemophilia, HIV, IBD, CF, neuromuscular diseases, nephrology and metabolic diseases). Socio-demographic characteristics are summarized in Table 1.

Twenty-seven consultations were with doctors; 8 with nurse specialists; 4 with a dietician.

2.5   Focus groups

Preliminary findings of the observations were discussed in focus groups with a self-selected sample of professional staff, recruited through e-mail messages and postings to bulletin boards. After the preliminary results of the observations and interviews were presented, three patient case histories related to self-management, communication and organization of consultations were discussed. The case histories were fictitious ones, composed on the basis of the observations. Conversations were audiotape recorded and analyzed (see 2.2).

Twenty-seven staff members participated in three two-hour group discussions; 16 nurses (including all nurse specialists working with youth); 5 medical doctors, 5 psychologists and one social worker. All major subspecialties of the hospital were represented.

2.6   Questionnaire

All adolescents who met the inclusion criteria stated in 2.1 on 1st July 2006 were invited to complete an online questionnaire that was accessible from October-December 2006 with a unique code on a secured Internet site. It measured socio-demographic characteristics, disease- and health care-related variables, and self-management with respect to consultations. The questionnaire was built on findings from a literature review, extensive data-analysis of the previously held interviews, and pilot tests of a draft questionnaire in face-to-face interviews with five adolescents and four parents. Information on the measures is published elsewhere. [42]

Capability for self-management was measured through assessing self-efficacy and related independent behaviors. As no validated instruments measuring adolescents' self-efficacy were available, a new instrument was developed using Bandura's Guide for the framing of self-efficacy questions (*How confident are you that you could successfully perform this task?*).[43] One scale measured self-efficacy in skills for independent hospital visits (11 items; Table 2) on a 4-point Likert scale (1 = no, certainly not; 2 = no, probably not; 3 = yes, probably; 4 = yes, certainly). Furthermore, adolescents rated their general independence during hospital consultations on a visual analogue scale (range 1-10) and they assessed independent behaviors during their most recent consultation on a dichotomous 7-item scale (Table 2).

Questionnaire data were analyzed using SPSS 17.0 (SPSS Inc., Chicago Ill.). Descriptive statistics were applied. Univariate correlations were calculated by Spearman's rho. Chi-square tests served to compare the means of self-reported behavior and self-efficacy for consultations. All tests were two-tailed, and *p* values < .05 were considered statistically significant. A multivariate logistic regression analysis was performed to examine the variables associated with the adolescents' actual

behavior during the most recent consultation. Predictors were included in the regression analysis on the basis of theoretical assumptions emerging from the qualitative studies. The percentage of explained variance (Nagelkerke's R^2) was calculated to give an indication of the fit of the regression model.

The study population consisted of 3648 adolescents. Of the 1087 questionnaires received (29.8%), 127 were excluded as they were incomplete, leading to a total of 960 valid questionnaires. An analysis of response and nonresponse is published elsewhere.[42] Nonresponders were more frequently males and had non Dutch surnames; in addition, they were older and less frequently came to the hospital for consultations as compared with responders ($P < .05$). Table 1 presents participants' characteristics. All major chronic conditions were represented.

Table 2 *Self-efficacy and self-reported independent behavior during last consultation (questionnaire; n = 960)*

	mean (SD)	% 'yes'[a]
Self-Efficacy in skills for independent hospital visits	range 1-4[a]	
I am convinced that…		
I could explain the doctor how I feel and what my needs are	3.48 (.66)	93.4
I would dare ask the doctor explain things until I understand it all	3.39 (.72)	88.8
I could answer all questions of the doctor myself	3.38 (.69)	90.4
I could explain to others what we discussed in the consultation room	3.30 (.77)	87.1
I could talk with the doctor on my own	3.27 (.81)	83.1
I would dare tell the doctor if I should disagree with her or him	3.23 (.76)	83.6
I would dare confess to the doctor that I did not stick to my regimen or hospital appointments	3.20 (.74)	83.8
I could deal with it if the doctor should criticize me	3.19 (.77)	85.6
I would dare ask the doctor anything, even about my private matters	3.06 (.86)	74.6
I could make hospital appointments on my own	2.99 (.95)	70.2
I could manage to travel to the hospital on my own	2.59 (1.14)	51.5
Self-reported independent behavior during last consultation	range 1-2[b]	% 'yes'[b]
During my last consultation…		
I went into the consultation room on my own		12.8
I made the appointment myself		13.0
I asked the doctor a question about a private matter		23.2
I prepared the consultation		24.1
I asked most questions to the doctor myself		41.3
I participated in a decision about my treatment		55.4
I answered most questions of the doctor myself		81.3

Table 2 *(Continued)*

	mean (SD)	% 'yes'[a]
Paired differences between self-efficacy and behavior[c]	**χ2 (df)**	**P**
I asked most questions to the doctor myself vs. I would dare ask the doctor explain things until I understand it all	30.2 (1)	< .001
I answered most questions of the doctor myself vs. I could answer all questions of the doctor myself	75.0 (1)	< .001
I went into the consultation room on my own vs. I could talk with the doctor on my own	10.7 (1)	.001
I asked the doctor a question about a private matter vs. I would dare ask the doctor anything, even about my private matters	34.4 (1)	< .001
I made the appointment myself vs. I could make hospital appointments on my own	48.3 (1)	< .001

[a] *1 = no, certainly not; 2 = no, probably not; 3 = yes, probably; 4 = yes, certainly. The last two scores were compiled into one category YES*

[b] *Here, a score of 1 indicates NO; while 2 indicates YES*

[c] *In Chi-square tests (χ2), differences in the means between two dichotomous statements were compared. For example, the statement from the Self-reported independent behavior scale "I answered most questions of the doctor myself" (dependent variable; YES / NO) was compared with a similar statement from the Self-efficacy scale "I could answer all questions of the doctor myself" (independent variable; YES / NO)*

2.7 Validation and integration of findings

Validation for the qualitative findings was primarily realized by method triangulation and peer-review. Preliminary analyses of interviews and observations were discussed within the research group and with the nurse coresearchers. Respondent validation was also employed: participants of the focus group sessions were invited to reflect upon the interpretations of the hospital observations. Interview findings were discussed with young patients who acted as coresearchers in a participative study conducted in 2006.[44]

Integration occurred at two stages. First, the research group thematically summarized all qualitative findings in a popularly published book.[45] These qualitative findings provided direct input for questionnaire development: by taking quotes from the interviews, by testing self-efficacy instruments, by presenting the Q-Care profile descriptions, and by determining which independent variables should be included in the regression analyses. Then, while some results from substudies have been published separately,[37, 42, 44] the drafting of this manuscript stimulated further integration by exploring and explaining adolescents' participation during consultations. For this, interpretations from the qualitative studies were compared with the quantitative data-set and discussed in the research group.

2.8   Ethical concerns

All study procedures were approved by the Institutional Review Board of the Erasmus Medical Center. The researchers had no access to participants' medical records. Participants were assured of confidentiality and data were processed anonymously. Eligible adolescents and their parents received written information about the study and gave informed consent, as well as health care providers involved.

3   Results

3.1   Interviews: adolescents' views of health care communication

Young people found routine hospital consultations little interesting and often boring ("*always the same*"). The pediatrician takes the lead and sets the agenda, the adolescent follows, answers questions if asked to do so and only rarely poses questions. The younger adolescents (under 16) reported that parents and doctors do most of the talking. Yet, most interviewees maintained they are very well capable of playing a more active role. There are two reasons why they do not do this:
  - it is not necessary: *"my parents do this for me"* and *"the doctor doesn't tell me to do so";*
  - it is not interesting or profitable: *"I don't really care"*, *"it's easier this way".*

Only few adolescents (mainly the younger ones) did not feel capable to represent themselves *("that is too difficult for me"*). On the contrary, most of the older ones said they wished to be the main partner in communication and some, all over 17, claimed an active role during consultations. They did not accept being marginalized in communication. Daphne (19; CF) said: *"I prefer to represent myself. If I forget something, it's OK for my mum to step in, but I used to get so irritated when doctors addressed my parents instead of me, with me just sitting there!".* Adolescents' attitudes toward communication represented their transitional status: in-between typical child and adult roles.

Their parents' roles were described in ambivalent terms. On the one hand, parents' presence and interference was perceived as *"helpful"* (by the younger ones) or *"supplementary"* (by the older ones). Adolescents claimed to be *"forgetful"* and parents' support was needed to refresh their memory or answer difficult questions. Also, the parents' presence was judged indispensable in critical situations. Many youth appreciated the expert knowledge, active involvement and support of their parents. They generally share the same goals when in consultation.

On the other hand, adolescents described their parents' interference as annoying *("overdone"*) and superfluous; it unnecessarily lengthened the consultation and embarrassed them in front of the doctor's. Parents' presence also inhibited them in discussing sensitive topics or asking questions themselves.

3.2   Q-Care profiles: adolescents' communication preferences

The Q-methodological analyses identified four distinct preference profiles for health care delivery and self-management: 'Conscious & Compliant'; 'Backseat Patient'; 'Self-confident & Autonomous'; and 'Worried & Insecure'.[37] The differences lie in various aspects: for example preferences for physician-patient interaction and view of parents' role during consultations.

   With respect to communication, adolescents in profiles 'Conscious & Compliant' and especially 'Self-confident & Autonomous' feel more independent and display a higher degree of self-efficacy for hospital visits than those in the other two. 'Backseat Patient' adolescents feel less capable of self-management and strongly agree with the statement "*It's important for me to have my parents present during consultations*" whereas those in the 'Self-confident & Autonomous' profile disagree. 'Backseat Patients' lean on their parents because they feel uninvolved and incompetent. 'Worried & Insecure' adolescents lack confidence because they worry about their health prospects. They need their parents to support them in coping with insecurities.

   Adolescents also share preferences. They all want to have a say in important treatment-related decisions. They are not opposed to doctors asking personal questions in front of their parents. Nevertheless, when sensitive issues such as sexuality and heredity are raised, parental presence "*may sometimes be inconvenient*". All adolescents would like to talk to doctors and nurses alone, even 'Backseat Patients'.

3.3   Observations: role play

The observations confirmed the general picture of nonparticipation and marginalization, with exceptions in older adolescents. Adolescents' most displayed attitude was to watch and wait, providing short and general answers to nonspecific questions. Parents were present in 70% of all consultations and played an important role. Only when adolescents came alone (in 9 consultations), they were more involved and talkative. Only in the HIV-department it was standard practice that the nurse specialist sees adolescents alone. Apart from this, no other differences between nursing and medical consultations or between the various subspecialties were detected with respect to adolescent involvement and parental roles.

   Using Goffman's dramaturgical metaphor,[41] we envisaged the hospital consultation as a theatre play with the pediatrician/nurse as stage director, with less fixed roles and sometimes interchangeable parts for the other parties. The youngest adolescents often acted as bystanders or played minor parts. Parents acted not only as stagehands or prompters, but often performed the main character role, sometimes presenting monologues and reducing their children to stage extras. At times, parents also acted as the advocate of the child's best interests by critically reviewing doctor's proposals. The parents' role is less prominent when children were older. However, even then there were many instances in which parents participated more actively.

   Staff did not ask adolescents' permission for their parents' presence, nor asked to see adolescents alone for some time. The dominant presence of parents during hospital consultations was facilitated by their children's nonparticipation, and tacitly accepted and endorsed by health care

providers. Though doctors almost always tried to direct the communication toward the adolescent, in due course discussions were frequently 'taken over' by parents.

This silencing is sometimes protested against by adolescents through nonverbal communication of discontent: Jim (16; end-stage renal disease) turned away in protest to his mother's ventilated frustration about Jim's noncompliant behavior. Or by rebelling verbally like Richard (14; muscular disease) who protested when his mother started discussing his habits.

3.4   Focus groups: health care providers' impotence

The focus group session revealed that health care providers find communicating with adolescents often challenging and sometimes frustrating. Teenagers are not motivated to be involved; this is explained as an inevitable consequence of puberty; *"it's just a phase"*. Several tactics were employed to involve them: making jokes, asking about hobbies, steering the conversation towards them or asking directly for their opinions – with varied success. At the same time, parents have been partners in communication for so long that this has become "*a fixed pattern"* that is hard to change: *"parents are so used to playing the first fiddle"*. Although staff was very dedicated to adolescent patients and did not question their competence, they also sympathized with parents' frustration with adolescent nonadherence and noninvolvement. Still, some professionals found dealing with parents during consultations even more difficult than dealing with patients. Yet, interference in the relationship between parents and adolescents was seen as undesirable and *"tricky"*. Adolescents were seldom seen independently, because staff was afraid parents *"would not accept this"*.

Hospital staff experienced managing triadic communication as balancing on a slack rope. Many felt incompetent to restrict parents' involvement and to activate adolescents. Though professionals set the stage in hospital consultations, they seemed unaware of their power to turn the tables.

3.5   First integration of qualitative findings

The most important roles and preferences of all parties involved in triadic consultations, as emerged from the qualitative studies were compiled in Box 3.[45] Several possible factors related to adolescents' involvement during consultations were identified: age (older adolescents participated more), gender (girls seemed more active), and ethnicity (those who came alone were often of non Dutch background). Furthermore, physical disability and poor health appeared to enhance parents' dominance, while adolescents actively involved in daily self-management appeared more involved during consultations.

Box 3 *Integration of interview and observational data: triadic communication during hospital consultations*

Youth (12-15 yrs)
• act as bystander
• pose very few questions
• do not feel involved

Youth (16-19 yrs)
• want to be the main partner in communication
• seem more involved
• do not wish to discuss private matters in front of parents

Parents
• report / keep the overview
• pose questions
• provide answers / supplement child's contribution
• arrange all practical affairs

Health care providers
• think adolescents are passive and sometimes unapproachable
• see parents as main partners in communication
• wish to involve young people more

3.6 Questionnaire: gap between capability and behavior

These factors were further explored in the quantitative analysis. Questionnaire participants assigned a mean score of 7.1 (*SD* = 2.0) on a scale from 1-10 to their general independence during consultations; only 21.9% saw their independence as insufficient (ie, below 6); 29.1% scored 9 or 10. Their mean rating on the scale 'Self-efficacy in skills for independent hospital visits' (Cronbach's α = .88; min. 11 − max. 44) was 35.1 (*SD* = 6.0). The majority of adolescents were convinced of their own capability of managing consultations (Table 2). For example, 90.4% answered *"yes"* (*"probably"* or *"definitely"*) when asked whether they could answer all questions of the doctor themselves. Eighty-three percent was confident to talk with the doctor alone. Nevertheless, their self-reported behavior during the most recent consultation told a different story. Few (12.8%) reported to have gone into the consultation room alone and less than half (41.3%) said they had asked most questions to the health care provider themselves. Chi-square tests, comparing the means of the self-reported behavior and the stated self-efficacy for the alleged competency demonstrated a gap between capability and behavior in all pairs (*P* < .001).

In a multivariate logistic regression analysis, factors associated with two independent behaviors were explored (Table 3). Those who had consulted the doctor alone more often had a non Dutch surname, were older, visited the outpatient department more frequently (indicating a more active disease process) and had a higher self-efficacy than those who came with their parent(s). Those who asked most questions themselves were older, more often female, and had a higher educational level and a higher self-efficacy than those who asked fewer questions.

Table 3 *Correlations with and multivariate logistic regression analysis with (a) going into consultation room independently and (b) asking most questions myself during most recent hospital visit (n = 941)*

(a) Going into the consultation room independently	Bivariate analysis	Multivariate logistic regression analysis		
	ρ^a	OR^b	95% *CI*	R^2
				.17
Gender (male)	.019	1.17	.76-1.79	
Age	.130***	1.64*	1.06-2.54	
Educational level (high)	-.039	.76	.50-1.17	
Type of education (special)	.022	1.90	.91-3.97	
Non Dutch surname	.152***	3.12***	1.89-5.16	
Hospital admissions in past three years	-.023	.63	.38-1.02	
Outpatient visits in past three years	.097**	2.03**	1.31-3.15	
Prescribed medications, diet or exercise	-.029	.76	.49-1.20	
Diagnosis after age of six years	.019	.90	.57-1.40	
Presence of physical limitations	-.048	.69	.41-1.16	
General health score	.001	.85	.68-1.06	
General score of independence during consultations	.189***	1.20**	1.05-1.38	
Self-Efficacy Scale in skills for independent hospital visits	.211***	1.08**	1.03-1.13	
(b) Asking most questions to the health care provider myself	Bivariate analysis	Multivariate logistic regression analysis		
	ρ^a	OR^b	95% *CI*	R^2
				.28
Gender (male)	-.071*	.70*	.52-.95	
Age	.215***	1.60**	1.17-2.20	
Educational level (high)	.120***	1.48*	1.09-2.00	
Type of education (special)	-.097**	.92	.51-1.66	
Non Dutch surname	.051	1.34	1.87-2.06	
Hospital admissions in past three years	-.024	.96	.67-1.37	
Outpatient visits in past three years	.039	1.23	.91-1.68	
Prescribed medications, diet or exercise	-.077*	.84	.61-1.15	
Diagnosis after age of six years	.066*	1.04	.76-1.44	
Presence of physical limitations	-.045	1.03	.72-1.47	

Table 3   *(continued)*

(b) Asking most questions to the health care provider myself	Bivariate analysis	Multivariate logistic regression analysis		
	ρ^a	OR[b]	95% *CI*	R²
				.28
General health score	.021	.92	.78-1.09	
General score of independence during consultations	.342***	1.24***	1.13-1.36	
Self-Efficacy Scale in skills for independent hospital visits	.406***	1.13***	1.09-1.17	

** P < .05; ** P < .01; *** P < .001*
ᵃ The bivariate correlations were calculated with Spearman's Rho ρ.
ᵃ The multivariate analysis adjusted for all variables. OR: Odds Ratio; CI: Confidence Interval; R²: explained variance by Nagelkerke R-square test

3.7   Final integration: ambivalence toward independent communication

We conclude there is a good deal of ambivalence toward triadic relations in communications with chronically ill adolescents. Adolescents' lack of involvement is co-constructed by all parties. Most adolescents felt competent to manage their own affairs during consultation and wanted to be involved in their own care, yet failed to demonstrate independence and let their parents do most of the talking. Adolescents differed in their preferences for an executive role in communication, and in their demonstrated competencies. The older ones, the girls, the higher-educated, the more experienced in terms of hospital visits, and those who feel more self-efficacious, reported to be more active during consultations.

Most explained their marginalized position as a result of their own indifference or as a conse-quence of *"not being asked to participate".* A minority saw themselves as incompetent as they were *"still a child",* while others were not happy with being left out of communication and *"treated in a childish way"*. Parents filled the gap. Although adolescents realized that one day they will have to manage communication themselves, many chose a comfortable position leaving the hard work to parents. At the same they were ambivalent about the parents' role: while they needed their parents and often appreciated their support, they also felt not at ease when parents interfered.

Pediatric staff is equally ambivalent: they tried to involve adolescents, while not restricting par-ents' presence or dominance of the communication. Although they expressed frustration with *"overbearing"* parents, they shared parental concern toward adolescents' noncompliance or lack of involvement. In conclusion, triadic communication with adolescents was all but multi-party-talk and adolescents did not act and were not considered as main partners.

4   Discussion

This study confirmed that adolescents with chronic conditions desire to participate in their own care and have their viewpoints taken seriously.[24, 28] A higher age was associated with a preference for communication directly to the adolescent.[24] Several qualitative studies found that preferences with regard to parents' role differ between adolescents.[20, 23, 27] The four Q-Care Profiles we described show that such preferences are closely related to (perceived) health care competencies.[37]

We established that a higher age was linked with more self-reported independent behaviors during consultation and with higher self-efficacy. Girls seemed more active asking questions. Williams also reported that mothers were more likely to act as mediator for their sons, as they saw their daughters as more responsible.[46]

Our study confirms that adolescents often remain inactive during triadic consultations[6, 11] despite good interactional competence. Multi-party-talk, as sometimes advocated,[17, 25] is difficult to accomplish. In any encounter involving three or more actors there is a tendency for two of them to enter a coalition in order to advance a personal agenda or achieve an agreed outcome.[4] In adolescent care, the health care provider and the parents tend to enter a bipartite coalition. Pyörälä found that adolescents with diabetes had an active patient role in two-party encounters with dieticians, whereas in triadic encounters they often turned into withdrawn bystanders.[11] Parents' presence and executive roles seem to be the key issues here. By acting as an 'alert assistant',[46] parents could, unwillingly, delay the development of adolescent self-management skills.

Pediatric providers in the present study felt ambivalent about asking parents to step aside, even though the Dutch Medical Treatment Act gave them a legal basis to involve adolescent patients actively. This may be related to lack of awareness or of ways to handle this complex situation.

Promoting independence in young people with chronic illness can be difficult for parents as several aspects of the parenting role compete: maintaining a supervisory role while supporting the child's emerging ability to independently manage their health.[47] Parents' presence during consultations is regarded as inhibiting or as supportive.[20, 23, 27] It may be also experienced as a threat to confidentiality, a communication aspect known to be important for adolescents in general.[18] Adolescents with chronic conditions, however, are perhaps more comfortable involving parents than are healthy peers.[48] Still, parents' presence also raises tension especially when personal or sensitive topics are discussed.[23] Nevertheless, risky behaviors and development of self-management are crucial for developmentally appropriate adolescent care.[49] Effective communication has the potential to improve adherence with the treatment regimen and improve young people's wellbeing and disease outcomes.[50]

4.1   Limitations of the study

One important limitation of our approach was that parents' voices were not studied and that only a limited number of pediatricians participated in the focus groups.

Also, wide ranges of practices, health conditions, and preferences were researched. We did so because chronic patients share many common tasks and challenges.[49] As a logical consequence,

differences in adolescents' and staffs' experiences with health communication related to the chronic conditions themselves cannot be accounted for by the used study design.

This research was carried out in one university hospital in the Netherlands and results may thus not apply to other settings and countries. Although employing different interviewers and observers may be seen as threatening the quality of data collection, being a coresearcher stimulated health care providers to change their practices. We tried minimizing the potential threat by intensive training, debriefing, and use of semistructured interview and observation guides.

The nonresponse rate was fairly high for both the interviews and questionnaire, implying that the results may not be applicable to all adolescents with chronic conditions. Also, more structured observations could be useful to test hypotheses about the importance of age, gender and ethnicity for adolescents' competencies.

4.2   Relevance and application of mixed methods in health communication research

Most studies on health communication in the context of pediatric consultations have used a quantitative approach, measuring participation, turn-taking and rating the communication type from video tapes,[5, 14, 15, 17, 19, 25] or audio tapes.[6] Other studies have relied on qualitative methods, using participant observation,[5, 9] interviews and/or focus groups.[20, 23, 27] Both approaches seem valuable, as they shed light on different aspects and actors. *Preferences* for communication and attitudes toward participation during consultations are best studied through in-depth interviews outside the hospital. The observations revealed that often adolescents demonstrated fewer *competencies* than they said they possessed. The strength of qualitative observations was that actual behavior could be studied, for which the interviews and focus group sessions parties offered explanations and justifications. Although a qualitative approach has many advantages and allows for richness and contextual data, the small numbers of participants do not allow us to make inferences about the prevalence and importance of certain characteristics for communication skills. Studying communication in context would ideally require a mix of qualitative and quantitative approaches.

Our study can be classified as a sequential, exploratory design, in which both methods were given equal priority and weight and in which the second phase partly developed from the first.[51] Qualitative studies helped us to generate hypotheses to be tested in the questionnaire, but vice-versa, findings from the questionnaire (for example the discrepancy between self-efficacy and self-reported independent behaviors), could be explained through insights obtained from interviews and observations. Full integration is difficult to achieve in MMR, however, and even more complicated to publish.[35] A review of 75 MMR studies in health care in the UK found that researchers often ignored their mixed methods design and described only the separate components.[32] Considerable experience and expertise – both in qualitative and quantitative research traditions – would seem to be needed to achieve good integration and good study outcomes.[52] In our case, the project leader, originally purely schooled in qualitative methods, learned to handle quantitative methods as well. This seems a prerequisite to integration. As few researchers possess this 'double expertise', qualitative and quantitative studies tend to be split up in MMR, instead of being mixed. This conflicts with the purpose of MMR: to gain a more complete understanding of

social phenomena.[51] In this study, MMR had an added value over a single method approach as it helped to unravel complex processes, allowed contextual data to be included and gave voice to multiple parties.

5 Conclusion

Adolescents desired to be involved, but their actual level of involvement in health care communication during hospital consultations was low. Many adolescents felt they were competent to become partner in communication, thus demonstrating another gap between capability and actual behavior. While the legal context grants adolescents every right to participate and (co)decide and they are supposed taking over control from their parents as an essential step in transition to adult care, the current structure of consultations, the communication style employed and the presence of parents hindered adolescent involvement in communication.

The mixed methods approach was of pivotal importance for our understanding of triadic consultations.

5.1 Practice implications

As patient partnership improves health outcomes, a key issue in adolescent health communication is the true partnering between adolescents, parents and doctors. In the practice of pediatric chronic care, parents' rights to participation seem better protected than those of their children, even when these have reached adolescence. The legal context provides a firm basis for including adolescents as main partners in communication. The organizational setting, however, does not. Triadic encounters are likely to generate tensions and differences of opinion, and in the presence of two or more adults, adolescents' voices are likely to be muffled. Therefore, doctors and nurses should be trained in age-appropriate communication skills, such as asking direct questions, discussing psychosocial and treatment-related subjects relevant to adolescents, and maintaining confidentiality.[45, 48, 50, 53] Health care providers should listen to parents and support their roles, but should see young people alone for part of the consultation.

Acknowledgements

References

1. Bensing J, van Dulmen S, Tates K. Communication in context: new directions in communication research. *Patient Educ Couns*. 2003;50(1):27-32.
2. Silverman D. *Communication and medical practice: social relations in the clinic*. London: Sage Publication; 1987.
3. Strong PM. *The ceremonial order of the clinic: parents, doctors and medical bureaucracies*. London: Routledge and Kegan Paul; 1979.
4. Gabe J, Olumide G, Bury M. 'It takes three to tango': a framework for understanding patient partnership in paediatric clinics. *Soc Sci Med*. 2004;59(5):1071-1079.
5. van Dulmen AM. Children's contributions to pediatric outpatient encounters. *Pediatrics*. 1998;102(3 Pt 1):563-568.
6. Wassmer E, Minnaar G, Abdel Aal N, Atkinson M, Gupta E, Yuen S, *et al.* How do paediatricians communicate with children and parents? *Acta Paediatr*. 2004;93(11):1501-1506.
7. Tates K, Meeuwesen L. Doctor-parent-child communication. A (re)view of the literature. *Soc Sci Med*. 2001;52(6):839-851.
8. Coyne I. Children's participation in consultations and decision-making at health service level: a review of the literature. *Int J Nurs Stud*. 2008;45(11):1682-1689.
9. Cahill P, Papageorgiou A. Triadic communication in the primary care paediatric consultation: a review of the literature. *Br J Gen Pract*. 2007;57(544):904-911.
10. Savage E, Callery P. Clinic consultations with children and parents on the dietary management of cystic fibrosis. *Soc Sci Med*. 2007;64(2):363-374.
11. Pyörälä E. The participation roles of children and adolescents in the dietary counseling of diabetics. *Patient Educ Couns*. 2004;55(3):385-395.
12. Runeson I, Enskar K, Elander G, Hermeren G. Professionals' perceptions of children's participation in decision making in health care. *J Clin Nurs*. 2001;10(1):70-78.
13. Martenson EK, Fagerskiold AM. Information exchange in paediatric settings: an observational study. *Paediatr Nurs*. 2007;19(7):40-43.
14. Tates K, Meeuwesen L. 'Let mum have her say': turntaking in doctor-parent-child communication. *Patient Educ Couns*. 2000;40(2):151-162.
15. Tates K, Meeuwesen L, Elbers E, Bensing J. 'I've come for his throat': roles and identities in doctor-parent-child communication. *Child Care Health Dev*. 2002;28(1):109-116.
16. Martenson EK, Fagerskiold AM. A review of children's decision-making competence in health care. *J Clin Nurs*. 2007;17(23):3131-3141.
17. Tates K, Elbers E, Meeuwesen L, Bensing J. Doctor-parent-child relationships: a 'pas de trois'. *Patient Educ Couns*. 2002;48(1):5-14.
18. Rutishauser C, Esslinger A, Bond L, Sennhauser FH. Consultations with adolescents: the gap between their expectations and their experiences. *Acta Paediatr*. 2003;92(11):1322-1326.
19. Cahill P, Papageorgiou A. Video analysis of communication in paediatric consultations in primary care. *Br J Gen Pract*. 2007;57(544):866-871.
20. Young B, Dixon-Woods M, Windridge KC, Heney D. Managing communication with young people who have a potentially life threatening chronic illness: qualitative study of patients and parents. *Br Med J*. 2003;326(7384):305.
21. Coyne I. Consultation with children in hospital: children, parents' and nurses' perspectives. *J Clin Nurs*. 2006;15(1):61-71.
22. Thorne SE, Harris SR, Mahoney K, Con A, McGuinness L. The context of health care communication in chronic illness. *Patient Educ Couns*. 2004;54(3):299-306.
23. Beresford BA, Sloper P. Chronically ill adolescents' experiences of communicating with doctors: a qualitative study. *J Adolesc Health*. 2003;33(3):172-179.
24. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics*. 2004;114(5):1272-1280.
25. van Dulmen S. Pediatrician-parent-child communication: problem-related or not? *Patient Educ Couns*. 2004;52(1):61-68.
26. Alderson P, Sutcliffe K, Curtis K. Children as partners with adults in their medical care. *Arch Dis Child*. 2006;91(4):300-303.

27. Zwaanswijk M, Tates K, Dulmen Sv, Hoogerbrugge PM, Kamps WA, Bensing JM. Young patients', parents', and survivors' communication preferences in paediatric oncology: Results of online focus groups. *BMC Pediatr*. 2007;7(35).

28. Dovey-Pearce G, Hurrell R, May C, Walker C, Doherty Y. Young adults' (16-25 years) suggestions for providing developmentally appropriate diabetes services: a qualitative study. *Health Soc Care Community*. 2005;13(5):409-419.

29. Alderson P. Competent children? Minors' consent to health care treatment and research. *Soc Sci Med*. 2007;65(11):2272-2283.

30. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child*. 2008;93(2):160-163.

31. Creswell JW. *Research Design: Qualitative, Quantitative, and Mixed Methods Approaches*. 2nd Edition ed. London: Sage Publications; 2003.

32. O'Cathain A, Murphy E, Nicholl J. Why, and how, mixed methods research is undertaken in health services research in England: a mixed methods study. *BMC Health Serv Res*. 2007;7:85.

33. O'Cathain A, Murphy E, Nicholl J. The quality of mixed methods studies in health services research. *J Health Serv Res Policy*. 2008;13(2):92-98.

34. Burke Johnson R, Onwuegbuzie AJ, Turner LA. Toward a Definition of Mixed Methods Research. *J Mix Methods Res*. 2007;1(2):112-133.

35. Bryman A. Barriers to Integrating Quantitative and Qualitative Research. *J Mix Methods Res*. 2007;1(1):8-22.

36. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. 2006;3(2):77-101.

37. Jedeloo S, van Staa A, Latour JM, van Exel NJ. Preferences for health care and self-management among Dutch adolescents with chronic conditions: A Q-methodological investigation. *Int J Nurs Stud*. 2010;47(5):593-603.

38. Akhtar-Danesh N, Baumann A, Cordingley L. Q-Methodology in Nursing Research: A Promising Method for the Study of Subjectivity. *West J Nurs Res*. 2008;30(6):759-773.

39. Morecroft C, Cantrill J, Tully MP. Individual patient's preferences for hypertension management: a Q-methodological approach. *Patient Educ Couns*. 2006;61(3):354-362.

40. Cross RM. Exploring attitudes: the case for Q methodology. *Health Educ Res*. 2005;20(2):206-213.

41. Goffman E. *The presentation of self in everyday life*. Garden City, NY: Doubleday Anchor; 1959.

42. van Staa AL, van der Stege HA, Jedeloo S, Moll HA, Hilberink S. Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors. *J Adolesc Health*. 2011;48(3):295–302.

43. Bandura A. Guide for constructing self-efficacy scales. In: Pajares F, Urdan T, editors. *Self-efficacy beliefs of edolescents*. Greenwich, CT: Information Age Publishing; 2006.

44. van Staa A, Jedeloo S, Latour JM, Trappenburg MJ. Exciting but exhausting: experiences with participatory research with chronically ill adolescents. *Health Expect*. 2010;13(1):95–107.

45. van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-8017-769-7.

46. Williams C. Alert assistants in managing chronic illness: the case of mothers and teenage sons. *Sociol Health Illn*. 2000;22(2):254-272.

47. Sawyer SM, Aroni RA. Self-management in adolescents with chronic illness. What does it mean and how can it be achieved? *Med J Aust*. 2005;183(8):405-409.

48. Klostermann BK, Slap GB, Nebrig DM, Tivorsak TL, Britto MT. Earning trust and losing it: adolescents' views on trusting physicians. *J Fam Pract*. 2005;54(8):679-687.

49. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet*. 2007;369(9571):1481-1489.

50. Rutishauser C. Communicating with young people. *Paediatr Respir Rev*. 2003;4(4):319-324.

51. Creswell JW, Plano Clark VL. *Designing and conducting mixed methods research*. Thousand Oaks CA: Sage; 2007.

52. Doyle L, Brady AM, Byrne G. An overview of mixed methods research. *J Res Nurs.* 2009;14(2):175-185.

53. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2007;46(1):161-168.

6

Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors

AnneLoes van Staa, Heleen A. van der Stege, Susan Jedeloo, Henriëtte A. Moll, Sander R. Hilberink

Abstract

PURPOSE: To explore associations between adolescents' perception of their readiness to transfer to adult care and socio-demographic and disease-related characteristics, effect of the condition, self-management ability, and attitude toward transition.

METHODS: A cross-sectional study was conducted in a Dutch university hospital where all adolescents (12-19 yrs) with somatic chronic conditions (n = 3648) were invited to participate in a web-based questionnaire. Invitations were issued to those without an intellectual disability and who had been under treatment for at least 3 years.

RESULTS: In all, 30% (n = 1087) of the adolescents responded; 954 assessed their perception of readiness for transfer. The majority (56%) felt that they were ready for transfer. Logistic regression analyses showed that 48% of the total variance in transfer readiness could be explained. Feeling more self-efficacious in skills for independent hospital visits and a greater perceived independence during consultations were most strongly associated with being ready to transfer. Higher transfer readiness was associated with older age, but age did not prove to be the most important explaining variable. Adolescents with a more positive attitude toward transition and those who reported more discussions about future transfer also felt more ready. Disease-related factors and effect of the condition including quality of life were only weakly associated with transfer readiness.

CONCLUSIONS: Adolescents' attitude to transition and their level of self-efficacy in managing self-care seem to be the keystones to transfer readiness. This study suggests that individual transition plans and readiness assessments might prove to be beneficial. Strengthening adolescents' independence and self-management competencies, combined with early preparation and repeated discussions on transition, seem to be useful strategies to increase adolescents' readiness for transfer to adult care.

1   Introduction

Transition of care for adolescents with chronic conditions has been much discussed, but rarely studied.[1] Blum *et al* define transition as "a multi-faceted, active process that attends to the medical, psychosocial and educational and vocational needs of adolescents as they move from child-focused to the adult-focused health-care system".[2] Ideally, it is a purposeful, planned process – as advocated repeatedly in policy documents,[3, 4] clinical guidelines, and journal editorial.[5] However, daily clinical practice is somewhat relentless, and Viner therefore aptly concluded that a major cultural shift in staff attitudes is needed as well as training.[6] Some authors concluded the evidence on which to build appropriate interventions is weak.[7, 8] Others, however, identified essential elements for a transition program,[9, 10] such as having reached a certain age and the availability of developmentally appropriate services, early start of preparation, coordination of care and appointment of a specific key worker.[10] One of the most important elements is that young patients need to be trained and empowered to become effective partners in their own care.[6]

The term "transition" refers to the process prior to and after the event of "transfer", that is, the actual shift from pediatric to adult health care.[11] For transfer to be successful, all partners involved need to be "ready". The process of "getting ready" then encompasses the specific decisions made and actions taken for building the capacity of the adolescent, parents and providers to prepare for, to begin, to continue, and to finish the process of transition. There is no consensus on age limits for these stages. Chronological age, however, is not a sufficient criterion for transfer because physical and psychological criteria should be met as well.[7, 12] A developmental perspective on adolescence provides a conceptual framework to better understand the young adult's readiness to engage in medical therapy.[13] However, concepts such as developmental readiness and social maturity are difficult to operationalize. Only a few empirical studies have explored the factors which are indicative of readiness for transfer and the method by which readiness could be assessed.[14-16]

The Pediatrics Consensus statement proposes that the timing of the transfer should depend on developmental readiness, complexity of health problems, characteristics of the adolescent and family, and the availability of skilled adult health providers.[3] Practitioners have enlarged on this, suggesting that patients should be able to self-manage care as well as understand their disease process.[11, 17] Self-care responsibility may be a better predictor for readiness to transfer as compared with level of knowledge or age.[12] Therefore, one's socio-demographic characteristics, severity and effect of the chronic condition, ability to self-manage the condition and attitude toward the transition process are potentially relevant for transfer readiness (TR). Good self-management in chronic care requires a high level of self-efficacy.[18] Bandura defines self-efficacy as one's belief in one's capabilities to organize and execute the courses of action required to produce given attainments.[19]

Most studies aimed at improving transitional services focus on specific diseases, disregarding the common challenges that adolescents with all kinds of chronic conditions face.[20, 21] Moreover, the prevailing perspective is that of health care providers[7]; young people's voices on what facilitates successful transition are largely absent, with some exceptions.[22-24]

The present study was concerned with the adolescents' personal views on their own self-management competencies and their readiness to transfer. In a large sample of adolescents with all kinds of chronic conditions, we explored the associations between adolescents' perception

of their transfer readiness and the following factors: (a) socio-demographic characteristics, (b) disease-related factors; (c) effect of the condition, (d) self-management ability, and (e) attitude toward transition.

2 Methods

2.1 Participants

The target group was selected from the hospital database, which consisted of all adolescents who were aged 12-19 years and who mid-2006 were under active long-term treatment for a somatic chronic condition in the Department of Pediatrics or the Department of Pediatric Surgery at the Erasmus MC - Sophia Children's Hospital. Inclusion criteria included the following:

- Before July 1, 2006, the adolescent should have been under treatment for > 3 years;
- The adolescent should have made outpatient visits and/or should have been hospitalized at least three times in these 3 years.

Adolescents were excluded in cases where transfer to adult care had already been performed or when they had a documented diagnosis of intellectual impairment.

At the time of the research (between October and December, 2006) no transitional programs were conducted in the hospital, except for hemophilia patients. Preparation for transition and collaboration with adult health care was virtually nonexistent. Transfer usually took place around the age of 18.

Ethical approval for the study was obtained from the Erasmus MC Medical Ethics Review Board. Participants were assured of confidentiality and the data were processed anonymously. The researchers had no access to the medical records of the participants. Eligible adolescents and their parents received written information regarding the study and the adolescents were invited to complete a web-based questionnaire which was accessible for 3 months with a unique code on a secured Internet site.

Response postcards were included to encourage adolescents to state the reasons because of which they did not qualify for the study, if this should be the case, or to provide an explanation as why they did not wish to participate. All adolescents received a reminder after 3 weeks. There was no financial remuneration; however, the participants were involved in a lottery for 2 iPods and a cell phone.

2.2 Measures

The questionnaire measured the following five domains: (a) socio-demographic characteristics, (b) variables related to disease and health care, (c) effect of the chronic condition (including quality of life), (d) self-management, and (e) attitude toward transition. Table 1 shows variable descriptions, data sources and numbers of items. The questionnaire was built on findings from a literature

Table 1   *Overview of measurement instruments per variable and domain (n = 938)*

Variable	Source[a]	No. of items	Cronbach's α	Theoretical range[b]	Actual range	Mean (SD)
Disease-related factors						
Hospital admissions in past 3 years	DB	1			0-138	4.5 (9.2)
Outpatient visits in past 3 years	DB	1		≥ 3	3-146	16.9 (15.7)
Number of different outpatient departments visited in past three years	DB	1			1-15	3.1 (2.2)
Prescribed medications, diet or exercise	Q	1		0-1	0-1	0.6 (.5)
Diagnosis after age of 6	Q	1		0-1	0-1	0.3 (.5)
Presence of physical limitations (AVO-99)	Q	10	.90	10-40	10-34	11.0 (3.2)
Effect of condition						
School / work absenteeism due to illness	Q	1		1-5	1-5	1.9 (.9)
Burden of visibility of condition	Q	2	.81	2-10	2-10	4.6 (2.1)
General Health score	Q	1		1-5	1-5	3.6 (1.0)
KIDSCREEN-10 (General health-related quality of life)	Q	10	.83	0-100	15-100	77.0 (16.3)
DCGM-10 (Impact of disease on well-being)	Q	10	.80	0-100	16-100	80.3 (15.9)
Self-management						
Self-Efficacy in coping with condition	Q	8	.77	8-32	8-32	26.8 (4.0)
Self-Efficacy in knowledge about condition	Q	10	.77	10-40	13-40	32.5 (4.8)
Self-Efficacy in skills for independent hospital visits	Q	11	.88	11-44	11-44	35.1 (6.0)
Independent behavior during last consultation	Q	7	.56	0-7	0-7	2.5 (1.5)
General score of independence during consultations	Q	1		1-10	1-10	7.1 (2.0)
Attitude toward transition						
Attitude toward transition	Q	4	.70	4-20	4-20	12.0 (3.5)
Transition of care often discussed during consultations	Q	1		1-5	1-5	1.6 (1.0)
Importance of discussing transitional care during consultations	Q	1		1-5	1-5	3.2 (1.2)

[a] *DB = electronic hospital database; Q = questionnaire.*
[b] *Scores are arranged so that a higher score on each variable indicates a better outcome or more absenteeism, self-efficacy, quality of life etc.*

6

review, extensive data-analysis of our previously conducted interviews with chronically ill youth,[25] and pilot tests of the draft questionnaire in face-to-face interviews with five adolescents and four parents.

2.1.1   Outcome variable

Adolescents' perception of transfer readiness was assessed by a single question *"Do you think that you are ready to transfer to adult care?"* (range: 1 = no, definitely not; 2 = no, probably not; 3 = yes, probably; 4 = yes, definitely).

2.1.2   Socio-demographic characteristics

Data of the participants' age, gender, and medical diagnosis (International Statistical Classification of Diseases and Related Health Problems [ICD-9 CM]) were retrieved from the hospital database. Because ethnicity is not recorded there, two researchers [AvS;HvdS] classified the family names into Dutch versus non Dutch using the Dutch Databank of Surnames. Educational level and type of education (regular vs special education for the physically-disabled) were obtained through the questionnaire.

2.1.3   Disease-related factors

Numbers of outpatient visits, hospital admissions, and the various outpatient departments visited between July 1, 2003 and June 30, 2006 were retrieved from the hospital database. Adolescents provided information on any prescribed medication, diet, or exercises. They were also asked to assess the presence of physical limitations in mobility including independence in activities of daily living. The original 10-item scale (AVO-99)[26] was dichotomized and in cases where any physical limitations were present, it was recoded as one.

2.1.4   Effect of the chronic condition

General health status, absenteeism from school or work because of illness in the past year, and the burden of the visibility of their condition were rated using 5-point Likert scales. Health-related quality of life was assessed by using the self-report versions of KIDSCREEN[27] and DISABKIDS.[28] The KIDSCREEN-10 Index provides a one-dimensional global health-related quality of life index with good internal consistency (α = .82) and good test-retest reliability (r = .73; intraclass cor-relation coefficient (ICC) = .72).[27] The short form DISABKIDS Chronic Generic Module (DCGM-10) measures general quality of life and the level of distress caused by a chronic disease (impact on well-being). It consists of twelve 5-point Likert-scaled items which were assigned to the following three domains: mental, social, and physical. Two items related to the impact of medication use were excluded in the present analysis because not all respondents had a medication regimen. The remaining 10 items combined produce a total score; this scale was found to have good internal consistency (α = .84).[28]

2.1.5   Ability for self-management

Ability for self-management was measured through assessing self-efficacy and independent be-
haviors. Validated instruments measuring self-efficacy in adolescents with a variety of chronic con-
ditions were not available. Instead, items from available self-efficacy instruments for diabetes,[29]
arthritis,[30] and sickle cell disease[31] were selected. These items were supplemented with items from
readiness assessments available on the Internet in 2006, such as the California Healthy & Ready To
Work Transition Assessment Tool.[32] Bandura's Guide was used for the framing of self-efficacy ques-
tions (*How confident are you that you could successfully perform this task?*).[33] After preliminary
testing of the formulations and response categories in face-to-face interviews, a final self-efficacy
instrument was constructed to measure the following three nondisease-specific domains (Table
2): (1) coping with the condition (8 items); (2) knowledge of the condition (10 items); and (3) skills
for independent hospital visits (11 items). Adolescents rated their self-efficacy on a 4-point Likert
scale (1 = no, certainly not; 2 = no, probably not; 3 = yes, probably; 4 = yes, certainly).

Adolescents rated their general independence during hospital consultations using a visual ana-
logue scale (range: 1 - 10). They assessed independent behaviors (such as actively participating
in treatment decisions, talking to the doctor on your own) during their last consultation on a
dichotomous 7-item scale.

Table 2   *Self-efficacy scales used in the study (n = 954)*

	mean (SD)	Cronbach's α
Self-efficacy in coping with condition		**.77**
I am convinced that		
I could manage to tell new classmates or friends about my condition	3.36 (.87)	
I could manage to explain my teacher or boss about my condition	3.49 (.76)	
I could manage to tell my parents or friends if I should be bullied	3.37 (.76)	
I could manage to tell my teacher or boss if I should be bullied	3.23 (.81)	
I could handle the fact that people may treat me "differently"	2.86 (.96)	
I can live with the knowledge that I will have my illness or disability for the rest of my life	3.31 (.88)	
I can accomplish as much as other people with the same illness or disability	3.62 (.68)	
I can accomplish as much as other people without an illness or disability	3.51 (.77)	
Self-efficacy in knowledge of condition		**.77**
I am convinced that		
I can clearly describe the future consequences of my illness or disability	3.16 (.81)	
I know how to reach the doctor or nurse, if necessary	3.20 (.85)	
I can tell which doctor or nurse at the hospital treats me	3.36 (.87)	

Table 2   *(Continued)*

	mean (SD)	Cronbach's α
I know which regimen the doctor has prescribed me (e.g., medications, diet or other lifestyle advice)	3.47 (.79)	
I could explain precisely what my regimen is for	3.39 (.75)	
I realize the future and present consequences of not adhering to my regimen now	3.31 (.85)	
I can find information about my illness or disability myself (books, Internet, leaflets)	3.43 (.84)	
I would recognize signs of health decline or of complications of my illness or disability	3.21 (.86)	
I know how often I must go back to the hospital for check-ups	3.51 (.75)	
I know what happens when I will transfer to adult care	2.46 (1.03)	
Self-efficacy in skills for independent hospital visits		**.88**
I am convinced that		
I could talk with the doctor on my own	3.27 (.81)	
I could make hospital appointments on my own	2.98 (.95)	
I could manage to travel to the hospital on my own	2.58 (1.14)	
I could explain the doctor how I feel and what my needs are	3.48 (.66)	
I could answer all questions of the doctor myself	3.38 (.69)	
I would dare ask the doctor anything, even about my personal issues	3.06 (.86)	
I would dare ask the doctor to explain things until I understand it all	3.39 (.72)	
I would dare confess to the doctor that I did not stick to my regimen or hospital appointments	3.20 (.74)	
I could deal with it if the doctor should criticize me	3.19 (.77)	
I would dare tell the doctor if I should disagree with her or him	3.23 (.76)	
I could explain to others what we discussed in the consultation room	3.29 (.77)	

2.1.6   Attitude toward transition

Four quotes, taken from adolescents' interviews,[25] were presented to assess attitude toward transition: "*It is a normal part of growing up*"; "*I am very concerned about having to leave Sophia Children's Hospital*"; "*I am rather looking forward to leave the Children's Hospital*" and "*I do not really care where I am being treated*". Responses were scored on a 5-point Likert scale (1 = totally disagree; 5 = totally agree); a higher score indicating a more positive attitude. Adolescents also stated on a 5-point Likert scale how often transition of care was being discussed during consultations and how important they considered it to be.

2.2   Data analysis

Data were analyzed using SPSS 17.0 (SPSS Inc., Chicago IL). Descriptive statistics were applied. Reliability (Cronbach's alpha) of scales was calculated. All tests were two-tailed, and P values < .05 were considered statistically significant. Next, univariate odds ratios were calculated to explore which measures in the domains were significantly associated with the outcome variable: an adolescent's transfer readiness (0 = definitively or probably not ready; 1 = definitively or probably ready). All variables were then entered into multivariate logistic regression analyses. Odds ratios and 95% confidence intervals were calculated for all models. Nagelkerke R^2 was used to express the explained variance in each domain. The regression models analyzed the five domains separately. In the final combined model, only significant variables ($P < .05$) were included. Finally, multi-collinearity was checked calculating the Variance Inflation Factor by means of multivariate analysis.

3   Results

3.1   Study population

Of the original sample of 3861, a total of 213 subjects were not included in the study because they either met the exclusion criteria (179 intellectual disabilities; four already transferred) or inadvertently did not receive the letter (n = 30). Therefore, a total of 3648 formed the study population (Figure 1). A response was received from 1318 adolescents (36.1%). However, 231 of them returned the response card explaining why they would not participate: of these, 144 (62.3%) claimed they were not chronically ill. Of the 1087 questionnaires received from the adolescents (29.8%), 133 were excluded because they were incomplete. leading to a total of 954 valid questionnaires. The number of nonresponders was 2330 (63.9%). Analyses revealed that nonresponders were more frequently males and had non Dutch surnames; in addition, they were older and were less frequent visitors at the hospital (outpatient department and hospital admissions) as compared with responders ($P < .05$).

Table 3 presents participants' characteristics. Mean age was 15.5 years ($SD:$ 1.9); females predominated in this study; 13.3% of the participants had a non Dutch surname. Over 90% of them lived with their parents or guardians. In all, 45% attended higher levels of education and 9.9% received special education for the physically disabled.

Congenital anomalies and conditions originating in the perinatal period (26.7%); neoplasms (12.7%), endocrine, nutritional, metabolic diseases, and immunity disorders (11.8%); diseases of the nervous system and sense organs (11.4%); and diseases of the musculoskeletal system and connective tissue (10.2%) were the five largest diagnostic categories (ICD-classification) in the sample.

Figure 1 *Study population*

```
                    ┌─────────────────────────────────┐
                    │  3648 study population (100%)    │
                    └─────────────────────────────────┘
                                    │
             ┌──────────────────────┼──────────────────────┐
             ▼                                              ▼
┌─────────────────────────────────┐        ┌─────────────────────────────┐
│ 1318 response (36.1%)           │        │ 2330 nonresponse (63.9%)    │
│ - 231 response cards            │        └─────────────────────────────┘
│ stating that adolescent does not│
│ feel chronically ill or without │
│ explanation                     │
└─────────────────────────────────┘
             │
             ▼
┌─────────────────────────────────┐
│ 1087 response on questionnaire  │
│ (29.8%)                         │
│ - 133 invalid (did not complete │
│ questionnaire)                  │
└─────────────────────────────────┘
             │
             ▼
┌─────────────────────────────────┐
│ 954 assessed transfer readiness │
│ (26.2%)                         │
└─────────────────────────────────┘
```
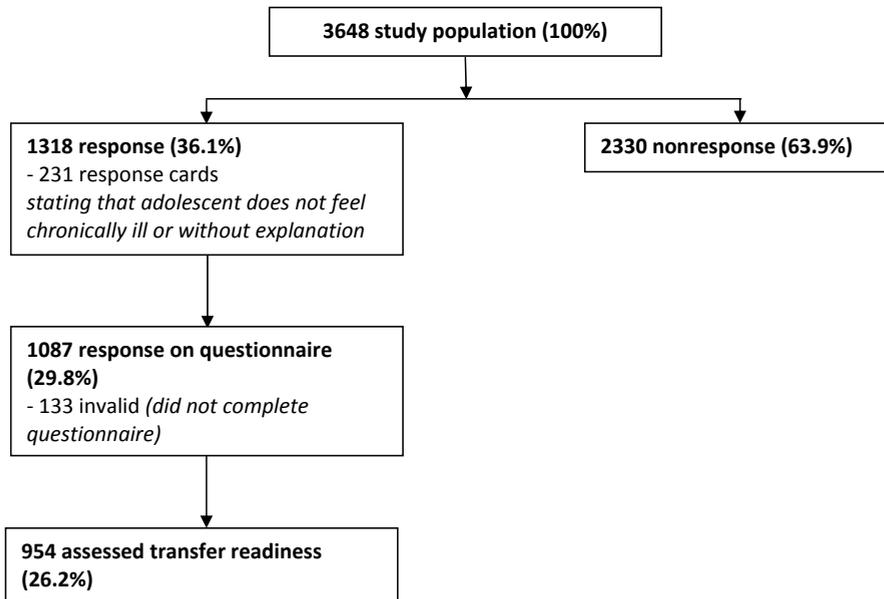
Table 3 *Adolescents' socio-demographic and disease-related characteristics (n = 954, unless indicated)*

	source[a]	*n*	%
Socio-demographic characteristics			
Gender	DB		
girls		536	56.2
boys		418	43.8
Age	DB		
12-15 yrs		585	61.3
16-19 yrs		369	38.7
Ethnicity	DB		
Dutch surname		827	86.7
Non Dutch surname		127	13.3
Level of education (*n* = 942)	Quest		
lower / middle		526	55.8
higher		416	44.2
Type of education (*n* = 946)	Quest		
mainstream		852	90.1

Table 3   *(Continued)*

	source[a]	*n*	%
special		94	9.9
Disease-related factors			
Hospital admissions in past 3 years	DB		
0		233	24.4
1 – 5		510	53.5
>6		211	22.1
Outpatient visits in past 3 years	DB		
<12		484	50.7
13 -24		275	28.8
>25		195	20.4
Number of different outpatient departments visited in past 3 years	DB		
1		254	26.6
2 – 5		569	59.6
>6		131	13.8
Prescribed medications, diet or exercise	Quest		
yes		602	63.1
no		352	36.9
Diagnosis after age of 6	Quest		
yes (ie, diagnosed between age 6-16)		290	30.4
no (ie, diagnosed at birth / during first 5 years)		664	69.6
Presence of physical limitations (AVO-99)	Quest		
yes		271	28.4
no		683	71.6

[a] *DB = electronic hospital database; Quest = questionnaire.*

3.1.1   Transfer readiness

About 56% of the 954 respondents perceived themselves to be "probably" (38.1%) or "definitely" (18.1%) ready to transfer to adult care (mean 2.6; *SD* = .9). Univariate associations between transfer readiness and variables in de five domains are presented in Table 4. With respect to *socio-demographic variables*, transfer readiness was positively associated with age: the older the adolescent, the higher the transfer readiness, and regarding gender, boys exhibited more readiness than girls. Twelve-year-olds scored transfer readiness with a mean of 2.0 (*SD:* .9); the 18-year-olds

with 3.2 (*SD* = .8). However, 25.7% of those aged 16-19 years felt not yet ready to transfer as compared with 55.2% in those aged 12-15 years.

Associations between transfer readiness and *disease-related variables* were relatively weak, however, having a physical limitation or a therapeutic regimen, reporting a higher absenteeism from school, and a higher burden of the visibility of the condition were all associated with lower transfer readiness. A higher general health score and a higher quality of life (as measured with DCGM-10) were associated with higher transfer readiness. Transfer readiness scores did not differ between the five major ICD-9 groups.

Table 4 *Univariate and multivariatea associations of variables in five domains with Transfer Readiness (n = 938)*

	Univariate analysis		Multivariate logistic regression analysis		
	OR	95% *CI*	OR[b]	95% *CI*	R²
Socio-demographic characteristics					.19***
Gender (male)	1.25	0.96-1.62	1.46**	1.10-1.94	
Age	1.49***	1.38-1.61	1.53***	1.42-1.66	
Educational level (high)	1.22	0.94-1.58	1.27	0.95-1.70	
Type of education (special)	0.56**	0.36-0.86	0.51**	0.31-0.83	
Non Dutch surname	1.35	0.91-2.00	1.72*	1.12-2.62	
Disease-related factors					.03*
Hospital admissions in past 3 years	0.99	0.97-1.00	0.99	0.98-1.01	
Outpatient visits in past 3 years	0.99	0.99-1.00	1.00	0.98-1.01	
Number of different outpatient departments visited in past 3 years	0.97	0.92-1.03	1.02	0.94-1.11	
Prescribed medications, diet or exercise	0.63**	0.48-0.82	0.67**	0.51-0.82	
Diagnosis after age of 6	1.16	0.88-1.54	1.21	0.91-1.61	
Presence of physical limitations (AVO-99)	0.68**	0.51-0.90	0.76†	0.56-1.02	
Effect of condition					.04***
School / work absenteeism due to illness	0.74***	0.64-0.86	0.81**	0.69-0.94	
Burden of visibility of condition	0.88***	0.83-0.94	0.92*	0.85-0.98	
General health score	1.25**	1.09-1.43	1.12	0.96-1.31	
KIDSCREEN-10 (general Health-related Quality of Life)	1.01	1.00-1.01	1.00	0.99-1.00	
DCGM-10 (impact of disease on well-being)	1.02**	1.01-1.02	1.01	0.99-1.02	

Table 4   *(Continued)*

	Univariate analysis		Multivariate logistic regression analysis		
	OR	95% *CI*	OR[b]	95% *CI*	R²
Self-management					.24***
Self-efficacy in coping with condition	1.07***	1.04-1.11	1.00	0.96-1.04	
Self-efficacy in knowledge of condition	1.10***	1.07-1.13	1.01	0.97-1.04	
Self-efficacy in skills for independent hospital visits	1.16***	1.13-1.20	1.11***	1.08-1.15	
Independent behavior during last consultation	1.39***	1.27-1.53	1.05	0.94-1.18	
General score of independence during consultations	1.49***	1.38-1.61	1.27***	1.16-1.38	
Attitude toward transition					.30***
Attitude toward transition	1.32***	1.26-1.39	1.35***	1.28-1.42	
Transition of care often discussed during consultations	1.78***	1.51-2.11	1.92***	1.60-2.30	
Importance of discussing transitional care during consultations	1.01	0.91-1.13	1.03	0.91-1.17	

OR = Odds Ratio; CI = Confidence Interval; R² = explained variance by Nagelkerke R-square test.

****** P < .05*

******* P < .01*

******** P < .001*

[a] *The multivariate analyses were conducted for each domain separately. The multivariate models only included the variables listed in each domain.*
[b] *Adjusted for the variables in the domain only.*

3.1.2   Self-management

The 29-item self-efficacy questionnaire consisted of three factors explaining 41.8% of the variance, and showed good sampling adequacy in confirmative factor analyses (Kaiser-Meyer-Olkin .90; *P* < .001). The reliability of the three scales was good, α ranging from .77 to .88 (Table 2). Among the different scales used in this study, only the scale "Independent behavior during last consultation" had a moderate reliability (α = .56) (Table 1). All self-management measures were strongly associated with transfer readiness (Table 4).

Adolescents' general *attitude toward transition* was found to be moderately positive. Three quarters (75.5%) of the study group responded positively to the statement that transition is "*a normal part of growing up*". Nonetheless, 70.2% indicated they were not looking forward to the prospect and admitted to being concerned about transition to a little or even a large extent. Only 28.6% stated they were not (at all) concerned about transition. The attitude toward transition scale showed reasonable internal consistency ($\alpha = .70$) and was positively associated with transfer readiness. The issue of the future transfer to adult care was found to be not discussed much during consultations: 65.3% stated it is never discussed; 20.3% sometimes, 6.9% regularly, and only 7.4% claim transfer is discussed (very) often. This factor was found to be associated with age: the older the participant, the more frequently the issue was discussed. Nevertheless, 45.8% of the participants considered discussing this issue as (very) important; 29.0% reported it as not being important (at all). There was a positive association between frequency of discussion and transfer readiness. This was further investigated in the logistic regression analysis.

3.2   Multivariate associations

Logistic regression analyses (*n* = 938) explored the influences of all variables in the five domains considered potentially relevant to transfer readiness (Table 4). Most of the variance was explained by the socio-demographic characteristics, self-management measures and the attitude toward transition. Disease-related factors and effect of the condition (including quality of life) contributed minimally to explained variance.

A final combined model that included all statistically significant variables (*P* < .05) explained 48% of all variance (Table 5). Age was an important factor contributing toward "feeling more ready". Of the other socio-demographic variables, only non Dutch ethnicity contributed significantly to higher transfer readiness. Both the presence of a therapeutic regimen as well as a higher absenteeism because of illness contributed toward lower transfer readiness; other disease-related variables did not contribute toward transfer readiness. More self-efficacy skills for independent behavior in the hospital and a higher score of general independence during consultations contributed toward a higher transfer readiness. Having a more positive attitude toward transition and reporting more discussions about transfer during consultations were also significantly associated with higher transfer readiness.

As our analyses involved concepts that may be interrelated, we checked for multi-collinearity. In all models of the multivariate analysis, the Variance Inflation Factor (VIF) was never higher than 2.24. In the combined model, VIF ranged from 1.00 to 1.59, confirming that collinearity is not a problem.

Table 5 *Combined model in logistic regression analysis with transfer readiness; including all variables significant at P < .05 in Table 3 (n = 938)*

	Adjusted[a] OR	95% *CI*	R²
Combined model			**.48***
Gender (male)	.78	.55-1.11	
Age	1.43***	1.29-1.59	
Type of education (special)	.84	.46-1.52	
Non Dutch surname	1.66*	1.01-2.73	
Prescribed medications, diet or exercise	.70*	.49-1.00	
School / work absenteeism due to illness	.80*	.66-0.96	
Burden of visibility of condition	.95	.88-1.03	
Self-efficacy in skills for independent hospital visits	1.07***	1.03-1.10	
General score of independence during consultations	1.25***	1.14-1.38	
Attitude toward transition	1.36***	1.28-1.44	
Transition of care often discussed during consultations	1.56***	1.26-1.92	

OR = Odds Ratio; CI = Confidence Interval; R2 = explained variance by Nagelkerke R2 test.
* P < .05
** P < .01
*** P < .001
[a] Adjusted for all variables listed in the table.

4   Discussion

This study investigated factors that might be associated with perception of being ready to transfer to adult care. Age proved to be an important factor in this sense, whereas other socio-demographic variables and disease-related factors (including quality of life) did not prove to be important. Moreover, adolescents' attitude toward transition and their level of self-efficacy in managing day-to-day self-care and in hospital consultations were strongly associated with transfer readiness.

Similar to our study, other studies have also reported that transfer readiness is positively influenced after reaching a certain age.[3, 12] Moreover, as seen in our study, Lutz Stehl also reported no significant association between other demographic and disease variables and adolescent-rated readiness.[34] McPherson *et al,* however, found that severity of disease had a negative impact on patients' interest in transition and anticipated difficulty of transfer.[16]

The fact that *disease-related variables* are not related to transfer readiness in our study may support the idea that transition issues are not unique to any disease process, but are instead universal among all those growing up with special health care needs.[21] Similar to other studies, our adolescents were moderately concerned about the forthcoming transition.[24, 35] This study also supports the idea that health care professionals may facilitate transfer readiness by paying more attention to transition preparation.[6, 10]

The pivotal importance of adolescents being self-efficacious in managing consultations for transfer readiness is in line with other studies[36-38] and recommendations.[4] Because we used general measures to assess self-efficacy, we could not establish correlations between transfer readiness and self-efficacy related to treatment regimen, but a study on diabetes found self-efficacy this to be an important predictor of self-care behavior and clinical outcomes.[13] Moreover, in a 3-year follow-up study of adolescents with sickle cell disease, self-efficacy in disease management predicted whether the individual would remain in care after transfer to adult care.[39] We expected that self-efficacy related to competencies in everyday self-management would have a strong association with transfer readiness.

4.1   Strengths and limitations

This study gave voice to a large sample of adolescents with a wide range of chronic conditions and yielded valuable information regarding their perceived readiness to transfer of care. The sample was heterogeneous with respect to congenital and acquired conditions, and age. The sample had its origin from the largest university hospital in the Netherlands, which includes all major pediatric subspecialties. However, the very fact that the study sample included adolescents with a wide range of chronic conditions, made it impossible to explore relations between transfer readiness and specific conditions or disease severity.

The nonresponse rate in this broad sample was fairly high (64%), probably because candidates were approached through an impersonal letter and were asked to access the questionnaire on the Internet. From the returned response postcards it appeared many adolescents did not consider themselves as chronically ill. This may be because of knowledge deficits regarding the nature of their condition, but it may also reflect lay views on when you are considered to be "ill" and on the importance of "being normal". The fact that nonresponders were less frequent hospital visitors than responders may imply that they represent a healthier population, although it may also indicate no-show. The nonresponse analysis revealed that, notably, older adolescents, boys, and adolescents with non Dutch surnames were underrepresented in the sample. This might have affected the outcomes, considering that transfer readiness was associated with age and ethnicity. It is impossible, however, to tell in what way.

Most of the measurement scales were newly designed for the purpose of this study. The internal reliability of each of the new scales was within the range of reasonable to good, but further validation is warranted. Several concepts, including the main outcome variable, were based on single questions, thereby limiting reliability.

Choosing a nondisease-specific sample limited the inclusion of more specific self-management measures related to self-efficacy and actual self-management of day-to-day therapy, as more than one third of the total sample did not have a therapeutic regimen. A generic and a disease-specific approach, each have their own value.[20]

Although self-report is useful for collecting information on perceived readiness, this method cannot fully capture the complex range of actual behaviors that characterize self-management and transition readiness. A partial solution could be found in comparing adolescent self-report of readiness with the perspectives of parents or health care providers, and to assess adolescents'

actual knowledge of the transitional process. McPherson *et al* found that adolescents with sickle cell disease knew little about the basic steps in the transition to adult-oriented care.[16]

4.2   Recommendations for further research

The final multivariate model has indicated relevant factors explaining the readiness felt by the adolescents, but it requires further investigation. Future studies should include measures on adolescent development and self-management related to treatment regimen. Also, use of transition readiness assessment instruments deserves scientific evaluation,[15] as it is not known whether these have any predictive value for a successful transition.

4.3   Clinical implications

To enhance transfer readiness, health care providers should focus on improving adolescents' competency and behavioral skills,[12] without failing to enhance and assess adolescents' knowledge of their condition and treatment. Introducing independent visits, that is, without the parents present, when the time is ripe, is a powerful strategy to prepare adolescents for transition and to assess self-management.[1, 10, 37-38] A timely start and greater attention to transitional care issues during consultations may help to instigate more positive attitudes in youth and parents toward transition and may increase transfer readiness.[11, 40] Adolescents may overrate their readiness for transfer, but might later be confident of being ready to transfer after receiving appropriate instruction and support. Formulating self-management assessments and individual transition plans could really make a difference.[4, 38]

This study also demonstrates that young people with any chronic illness have many aspects in common,[21] supporting the implementation of generic transition readiness assessments such as the new Transition Readiness Assessment Questionnaire (TRAQ).[15]

5   Conclusion

Apart from age, adolescents' attitude to transition, and their level of self-efficacy in managing day-to-day self-care and hospital consultations seem to be the keystones to transfer readiness. Strengthening adolescents' independence and self-management competencies, combined with early preparation and repeated discussions about the forthcoming transition, seems to be a useful strategy in increasing adolescents' readiness to transfer.

Acknowledgements

References

1. Reid GJ, Irvine MJ, McCrindle BW, Sananes R, Ritvo PG, Siu SC, *et al.* Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics.* 2004;113(3 Pt 1):e197-205.

2. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, *et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. *J Adolesc Health*. 1993;14(7):570-576.

3. American Academy of Pediatrics, American Academy of Family Physicians and American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics*. 2002;110(6 Pt 2):1304-1306:1304-1306.

4. Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM. Transition to adult health care for adolescents and young adults with chronic conditions: position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 2003;33(4):309-311.

5. McDonagh JE, Viner RM. Lost in transition? Between paediatric and adult services. *Brit Med J.* 2006;332(7539):435-436.

6. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child.* 2008;93(2):160-163.

7. While A, Forbes A, Ullman R, Lewis S, Mathes L, Griffiths P. Good practices that address continuity during transition from child to adult care: synthesis of the evidence. *Child Care Health Dev.* 2004;30(5):439-452.

8. Binks JA, Barden WS, Burke TA, Young NL. What do we really know about the transition to adult-centered health care? A focus on cerebral palsy and spina bifida. *Arch Phys Med Rehabil.* 2007;88(8):1064-1073.

9. Betz CL. Transition of adolescents with special health care needs: review and analysis of the literature. *Issues Compr Pediatr Nurs.* 2004;27(3):179-241.

10. McDonagh JE. Growing up ready for emerging adulthood. An evidence base for professionals involved in transitional care for young people with chronic illness and/or disabilities. London: Department of Health. Paper read at Conference; 26th March 2006. http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4137427

11. Kennedy A, Sawyer S. Transition from pediatric to adult services: are we getting it right? *Curr Opin Pediatr*. 2008;20(4):403-409

12. Cappelli M, MacDonald NE, McGrath PJ. Assessment of readiness to transfer to adult care for adolescents with cystic fibrosis. *Child Health Care*. 1989;18(4):218-224.

13. Anderson BJ, Wolpert HA. A developmental perspective on the challenges of diabetes education and care during the young adult period. *Patient Educ Couns*. 2004;53(3):347-352.

14. Betz CL, Redcay G, Tan S. Self-reported health care self-care needs of transition-age youth: a pilot study. *Issues Compr Pediatr Nurs*. 2003;26(3):159-181.

15. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, *et al.* Measuring the transition readiness of youth with special healthcare needs: validation of the TRAQ--Transition Readiness Assessment Questionnaire. *J Pediatr Psychol.* 2011;36(2):160–171.

16. McPherson M, Thaniel L, Minniti CP. Transition of patients with sickle cell disease from pediatric to adult care: Assessing patient readiness. *Pediatr Blood Cancer.* 2009;52(7):838-841.

17. Por J, Golberg B, Lennox V, Burr P, Barrow J, Dennard L. Transition of care: health care professionals' view. *J Nurs Manag*. 2004;12(5):354-361.

18. Marks R, Allegrante JP, Lorig K. A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part I). *Health Promot Pract.* 2005;6(1):37-43.

19. Bandura A. *Self-efficacy: the exercise of control*. New York: W.H. Freeman and Company; 1997.

20. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.

21. Okumura MJ. Growing up and getting old(er) with childhood-onset chronic diseases: paving the way to better chronic illness care worldwide. *J Adolesc Health*. 2009;45(6):541-542.

22. Shaw KL, Southwood TR, McDonagh JE. User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2004;43(6):770-778.

23. Reiss JG, Gibson RW, Walker LR. Health care transition: youth, family, and provider perspectives. *Pediatrics.* 2005;115(1):112-120.

24. Moons P, Pinxten S, Dedroog D, Van Deyk K, Gewillig M, Hilderson D, *et al.* Expectations and experiences of adolescents with congenital heart disease on being transferred from pediatric cardiology to an adult congenital heart disease program. *J Adolesc Health.* 2009;44(4):316-322.

25. van Staa AL, Jedeloo S, Kuijper M, Latour JM. Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-8017-769-7.

26. de Klerk M, Iedema J, van Campen C. *SCP-maat voor lichamelijke beperkingen op basis van AVO 2003. [Measure for physical limitations based on AVO-2003].* SCP-werkdocument 121. Den Haag: Sociaal Cultureel Planbureau; 2006. ISBN 90-377-0268-6.

27. Ravens-Sieberer U, Auquier P, Erhart M, Gosch A, Rajmil L, Bruil J, *et al.* The KIDSCREEN-27 quality of life measure for children and adolescents: psychometric results from a cross-cultural survey in 13 European countries. *Qual Life Res.* 2007;16(8):1347-1356.

28. Simeoni MC, Schmidt S, Mühlan H, Debensason D, Bullinger M. Field testing of a European quality of life instrument for children and adolescents with chronic conditions: the 37-item DISABKIDS Chronic Generic Module. *Qual Life Res.* 2007;16(5):881-893.

29. Moens A, Grypdonck MH, van der Bijl JJ. The development and psychometric testing of an instrument to measure diabetes management self-efficacy in adolescents with type 1 diabetes. *Sch Inq Nurs Pract.* 2001;15(3):223-233.

30. Barlow JH, Shaw KL, Wright CC. Development and preliminary validation of a children's arthritis self-efficacy scale. *Arthritis Rheum.* 2001;45(2):159-166.

31. Edwards R, Telfair J, Cecil H, Lenoci C. Reliability and validity of a self-efficacy instrument specific to sickle cell disease. *Behav Res Ther.* 2000;38(9):951-963.

32. Betz CL. California Healthy and Ready To Work transition health care guide: developmental guidelines for teaching health care self-care skills to children. *Issues Compr Pediatr Nurs.* 2000;23(4):203-244.

33. Bandura A. Guide for constructing self-efficacy scales. In: Pajares F, Urdan T, eds. *Self-efficacy beliefs of adolescents.* Greenwich, CT: Information Age Publishing; 2006.

34. Lutz Stehl MJ. *Transition readiness in adolescents with blood disorders: the importance of family functioning and parent-adolescent communication style.* Philadelphia, PA: Drexel University; 2005.

35. Anderson DL, Flume PA, Hardy KK, Gray S. Transition programs in cystic fibrosis centers: perceptions of patients. *Pediatr Pulmonol*. 2002;33(5):327-331.

36. Betz CL, Redcay G. Creating Healthy Futures: an innovative nurse-managed transition clinic for adolescents and young adults with special health care needs. *Pediatr Nurs*. 2003;29(1):25-30.

37. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: a multicentre cohort of adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2005;44(6):806-812.

38. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2007;46(1):161-168.

39. Wojciechowski EA, Hurtig A, Dorn L. A natural history study of adolescents and young adults with sickle cell disease as they transfer to adult care: a need for case management services. *J Pediatr Nurs*. 2002;17(1):18-27.

40. Lotstein DS, Ghandour R, Cash A, McGuire E, Strickland B, Newacheck P. Planning for health care transitions: results from the 2005-2006 National Survey of Children With Special Health Care Needs. *Pediatrics*. 2009;123(1):e145-152.

7

On Your Own Feet Self-Efficacy Scale: development and validation of a generic instrument for chronically ill adolescents

AnneLoes van Staa, Mathilde M.H. Strating, On Your Own Feet Research Group

Abstract

BACKGROUND: Nursing interventions may support self-management in adolescents with chronic conditions. It is important to measure self-efficacy as this is one of the mechanisms to produce effective self-management. For adolescents, only disease-specific self-efficacy instruments are available.

OBJECTIVES: To develop a short, reliable and valid instrument measuring self-efficacy in adolescents with chronic conditions.

DESIGN: Sequential mixed methods design consisting of semi-structured, face-to-face interviews followed by a cross-sectional survey.

SETTING: University children's hospital in the Netherlands.

PARTICIPANTS: Adolescents (12–19 years) with somatic chronic conditions. Thirty-one from a purposive sample of 66 were interviewed (16 boys and 15 girls; response 47%). All adolescents in long-term treatment (n = 3648) were invited to complete a web-based questionnaire; 30% (n = 1087) responded.

METHODS: Based on theory, existing disease-specific instruments, experts' opinion, and adolescent interviews, a 29-item instrument was constructed measuring self-efficacy in three nondisease-specific domains: coping with the condition; knowledge of the condition; skills for hospital visits. This scale was tested and shortened to 17 items by means of structural equation modeling. Convergent validity was tested by a correlational approach; predictive validity was examined with multiple regression analyses in which measures of self-management and health-related quality of life were regressed on socio-demographic variables and the final scale.

RESULTS: 958 adolescents with different chronic conditions completed the self-efficacy instrument. Explanatory factor analysis with varimax rotation confirmed the three-factor structure, explaining 41.7% of the variance. Confirmatory factor analysis revealed good fit indices. Item-reduction analysis resulted in the On Your Own Feet Self-Efficacy Scale (OYOF-SES); Cronbach's alpha was .85. Younger adolescents, girls, those with lower education and physical limitations had lower self-efficacy. Correlations with measures for managing the therapeutic regimen and independence in health care tasks were significant and in the expected direction. Multivariable regression analyses demonstrated predictive validity of the OYOF-SES for health-related quality of life and self-management during hospital consultations.

CONCLUSIONS: The newly constructed 17-item OYOF-SES is a valid, reliable, and discriminative instrument for assessing self-efficacy in adolescents with chronic conditions. Further testing of responsiveness to change should indicate whether it could guide nursing interventions aimed at enhancing adolescents' self-efficacy for self-management.

What is already known about the topic?
- Adolescence is a pivotal period for the acquisition of independent self-management skills and attitudes.
- Self-efficacy is both an important condition for and a predictor of self-management behavior.
- Self-efficacy should be measured by carefully developed and validated instruments. For chronically ill adolescents only disease-specific instruments are available.

What this paper adds
- The new, generic self-efficacy scale measures three domains related to coping, disease knowledge, and skills for independent hospital consultations of adolescents with various chronic conditions.
- The On Your Own Feet Self-Efficacy Scale has good reliability estimates while concurrent as well as predictive validity for independent behavior during consultations could be established.
- The instrument's responsiveness to change and predictive value for adherence to treatment needs further examination.

1   Background

A growing number of children in the Netherlands is living with a chronic condition. The most recent estimation was 500.000 children, ie, 14% of all Dutch children below 18 years of age. [1] Chronic illness affects adolescents in many different ways. In addition to the demands of complex treatment regimens, chronic illness can affect their growth and development, their appearance, identity, mental health, relationships with peers, and engagement with education and employment. [2] Therefore, a major developmental task is assuming adolescents' responsibility for self-care through expanding the knowledge, attitudes and life skills that underpin engagement with self-management. [3, 4] This requires a balance between their own efforts at self-management and parental support. [5] Also, as adolescents gradually grow out of the pediatric environment - new working partnerships with physicians and nurses must be formed, [6] reflecting adolescents' transition to self-management.

Self-management is defined as "the individual's ability to manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes inherent in living with a chronic condition". [7] Efficacious self-management encompasses the ability to monitor one's condition and to affect the cognitive, behavioral and emotional responses. The ultimate goal is maintaining a satisfactory quality of life, which requires hard 'work' on the part of the patient and their family. They are faced with three major tasks: medical management of the condition and prescribed therapy, dealing with the impact of illness on functioning and relationships (role management), and the management of emotions. [8]

Self-efficacy is the key mediating variable in a wide range of self-management behaviors. [9] It "refers to the beliefs in one's capabilities to organize and execute the courses of action required to produce given attainments" (Bandura, 1997:3). [10] Good self-efficacy can improve clinical and social outcomes, including quality of life. [11] Self-efficacy should be assessed in terms of particularized judgments of capability that may vary across realms of activity, different levels of task demands within a given activity domain, and different situational circumstances. [12] Self-efficacy is particularly

important for complex and life-long changes of behavior and therefore a critical feature in chronic disease management.[7, 8] As in chronically ill adults, adolescents' self-efficacy is reported to be a predictor for positive health outcomes in, for example, diabetes,[13-15] and sickle cell disease.[16, 17]

Self-management support goes beyond traditional knowledge-based patient education to include processes that develop patient problem solving skills, improve confidence (ie, self-efficacy), and support application of knowledge in real-life situations that matter to patients.[18] Therefore, nurses are well suited to strengthen chronically ill adolescents' self-management skills,[19] by applying interventions that promote self-efficacy. Therefore, being able to identify those with low self-efficacy and those who might be less likely to perform self-management tasks would be important for them.

The burden of chronic illness is not necessarily disease-specific as different health problems encompass many comparable adaptive tasks.[20, 21] A review of the lived experience of living with a chronic illness during adolescence identified seven common themes: developing and maintaining friendships, being normal and getting on with life, the importance of family, attitude to treatment, experiences of school, relationship with health care professionals, and the future.[22]

A good measurement instrument should be able to detect individual differences, to identify lower levels of self-efficacy in certain areas, and be useful to evaluate the impact of interventions.[23] At the moment, only disease-specific instruments are available for measuring self-efficacy in adolescents with a chronic disease, disregarding the common challenges that adolescents with all kinds of chronic conditions face and also limiting comparison across conditions.[24] According to Frei *et al*,[23] the development and validation process of most of these self-efficacy instruments had major limitations. The aim of our study was to develop a reliable and valid measurement instrument for generic self-efficacy for management of a chronic condition at home, in social life and during hospital consultations.

2 Methods

2.1 Design, setting, and participants

This study is part of the mixed methods research project "On Your Own Feet" [2004-2007] that employed a sequential strategy of inquiry.[25] Overall aim of the "On Your Own Feet" program was to map preferences for health care delivery and competencies required to enable adolescents with chronic conditions to independently direct their own treatment and life (ie, self-management).[26, 27] All studies were performed in the Erasmus University Medical Center, Sophia Children's Hospital - a large tertiary referral centre in the Netherlands, targeting adolescents aged 12-19 years who were in active long-term treatment (over three years prior to July 1st 2006) for a somatic chronic condition in the departments of Pediatrics or Pediatric Surgery in this hospital.

The aim of the substudy reported here was to develop and validate a generic self-efficacy scale in four consecutive stages. A systematic approach for the development and validation of self-efficacy instruments was followed by defining the aim and *a priori* considerations for the instrument (face

validity); describing the process of identification and selection of items (content validity) (stage 1 and 2); and finally, determining the concurrent and predictive validity (stage 3 and 4).[23]

2.2   Stage 1: developing a draft instrument

Selection of items. We searched the literature in 2005 for validated instruments to measure adolescents' competencies for self-management. From available self-efficacy scales for adolescents with diabetes,[28] rheumatoid arthritis,[29] and sickle cell disease,[30] we identified generic items and issues that reflected the three major tasks in self-management (medical management, role management, and the management of emotions). We also selected items from online available transition readiness assessments at that time: the California Healthy and Ready to Work Transition (CHRWT) Assessment Tool[31] and the ICI Boston Health Care Skills Checklist.[32] Since these tools were developed for diagnostic purposes to use in clinical nursing practice in the United States, we first explored to what extent they would be useful for our purpose.

We deemed the CHRWT Assessment Tool dichotomous response format (yes / no) not useful in research. The Boston list used the following categories to rate independent performance of skills; needs practice, plans to start practicing, or already accomplished the skill. This response format was adapted into: "not independent at all", "partially independent" and "fully independent". We also added an extra category: "I would like to learn this task" (yes / no). The draft instrument consisted of 48 items in 4 domains: knowledge of the condition and of health care providers (16 items); independence in treatment (5 items); independent behavior during consultations (13 items); and independence in daily life activities (14 items).

Interview study. The draft instrument was presented to a purposive sample of 31 adolescents (response 47%; 16 boys and 15 girls; 12-19 years) who participated in semi-structured interviews in their homes. The interviewers were instructed to inform after the adolescents' understanding of their condition; its impact on their lives; their perceived independency with respect to self-management; adherence to treatment; and their experiences during hospital consultations. Adolescents were asked what the concept 'self-management' meant to them; what illness-related tasks they considered important; and how they assessed their own competencies. The sampling procedures and analytical strategy are described in other papers.[26, 27, 33] Twenty-nine (out of 31) respondents completed the draft instrument. Descriptive statistics were used and respondents' comments on the response format and the phrasing of the questions were analyzed.

2.3   Stage 2: content validation of the instrument

In this stage the self-efficacy scale was developed based on the experiences with the draft instrument. A research team, including two experienced pediatric nurse researchers, defined the aims of the instrument and assessed the *content validity*, ie, the degree to which elements of the instrument are relevant to and representative of the targeted construct for a particular assessment purpose.[34]

Analysis of adolescents' responses. The results from the qualitative interviews and the draft instruments were integrated to compile a final list of the necessary generic competencies – knowl-

edge, attitudes, and skills – for successful self-management as a prerequisite for transfer to adult care. Younger respondents associated independence and self-management with *"doing things yourself, without help from your parents"*, while older adolescents mentioned more often *"being autonomous in taking care of yourself and in decision-making"*. Both aspects were included in the self-efficacy scale. Items related to the more general autonomy in daily life activities (for example 'being able to cook a meal') were marked as less relevant. We excluded these, also in view of the fact that the respondents considered the 48-item list too long.

Response format and pre-test. Adolescents criticized the proposed response format as well. First, the distinction between performing tasks independently, or with help from others, did not fit with all questions. Second, several adolescents pointed at the discrepancy between their actual independent behavior and their perceived competence: *"Most things I can do without any help from others – only I do not do them very often"; "I am capable of talking to the doctor, but my parents do this for me".* Also, hardly anyone ticked the box "I would like to learn this task". Therefore, we reverted to the usual format for the framing of self-efficacy questions ("How confident are you that you could successfully perform this task?"), as suggested by Bandura.[35] Self-efficacy can be assessed at three levels of generality: general, intermediate or specific. Our format aims to measure the *intermediate level* of self-efficacy, ie, for a class of performances within the same activity domain under a class of conditions sharing common properties.[10] Lastly, the adapted scale (29 items) and the entire questionnaire were pre-tested with five adolescents, four parents and six nurse specialists who worked with the target group. Several items were rephrased following respondents' suggestions.

Aims: the generic self-efficacy instrument was aimed to be both discriminative and evaluative, implying that it should be able to detect differences between adolescents and changes over time.[23] The first aim was tested in a cross-sectional study with a large number of adolescents with different chronic conditions (stage 3; 2.4.1). Additionally, we wanted to develop a short, but nevertheless valid and reliable instrument in order to reduce measurement burden for future respondents. The second goal could not be tested with this study design.

2.4   Stage 3: validation of the 29-item self-efficacy scale

2.4.1   Participants and procedure

The On Your Own Feet Self-Efficacy Scale's psychometric properties were tested in a cross-sectional study inviting all adolescents who met the inclusion criteria stated in 2.1 on 1st July 2006 (*n*=3,648).

Procedures and ethical considerations: Approval for the study was obtained from the Erasmus MC Institutional Review Board. Participants were assured of confidentiality and the data were processed anonymously. The researchers had no access to medical records. Eligible adolescents received written information and were invited to complete a web-based questionnaire accessible for three months (end 2006) with a unique code on a secured internet site. All received a reminder after three weeks. Response postcards were included to encourage adolescents to state they did not qualify for the study, if this should be the case, or to explain why they did not wish to partici-

pate. There was no financial remuneration, although participants were entered in a lottery for two iPods and a cell phone.

2.4.2 Measures

Self-efficacy: The tested self-efficacy instrument consisted of 29 items (OYOF-29). Appendix 1 presents the full instrument in the original language version (Dutch) and in the English translation. The instrument measured the following three nondisease-specific domains: (1) coping with the chronic condition (8 items), (2) knowledge of the chronic condition (10 items), and (3) skills for independent hospital visits (11 items). Adolescents were to rate their self-efficacy on a 4-point Likert scale (1 = no, certainly not; 2 = no, probably not; 3 = yes, probably; 4 = yes, certainly). The same format was used in three additional 4-item self-efficacy scales related to the therapeutic regimen that were presented to those who indicated they had been prescribed medications, diet and/or exercises.

Socio-demographic characteristics: Participants' age, gender, and medical diagnosis were retrieved from the hospital database. As ethnicity is not recorded in the hospital database, two researchers independently classified the family names into Dutch versus non Dutch using the Dutch Databank of Surnames. Educational level and type of education (mainstream versus special education for the physically disabled) were obtained through the questionnaire.

Disease-related characteristics: Numbers of outpatient visits, hospital admissions and different outpatient departments visited between July 1st 2003 and June 30th 2006 were retrieved from the hospital database. Adolescents provided information on when their chronic condition was diagnosed (at birth; before the age of six; between 6 to 12 years; after the age of 12) and on any prescribed therapeutic regimen. They were also asked to assess physical performance in mobility on a 4-point Likert scale ranging from 'without any difficulty' to 'only with help from other'. This original 10-item AVO-99 scale[36] was subsequently dichotomized: the presence of any physical limitations was coded as 1.

Health-related Quality of Life (HRQoL) was assessed by the self-report short versions of the cross-culturally validated short form KIDSCREEN and DISABKIDS questionnaires. All items are answered on a 5-point response scale ranging from 'never/not at all' to 'always'. KIDSCREEN-10 provides a one-dimensional global HRQoL index with good internal consistency (Cronbach's α = .81).[37] The 10-item DISABKIDS chronic generic measure (DCGM-10) measures general quality of life and the level of distress caused by a chronic disease (α = .84).[38]

Self-management. Several self-management measures were newly constructed for the questionnaire:

a) Visual analogue scales (VAS; range 1 - 10) were presented to assess adolescents' rating of their general independence during consultations; and their independence in managing their therapeutic regimen (medication, diet, and/or exercises).

b) Adolescents also assessed actual independent behavior (such as actively participating in treatment decisions, talking to the doctor on your own) during their most recent consultation using a dichotomous 7-item scale.

c) Two index-scores were calculated for those adolescents who indicated they had a prescribed therapeutic regimen: one measuring independent behavior and the other adherence to the therapeutic regimen. Adolescents indicated on a 4-point Likert scale how they had performed the stated behavior and how consistently they had adhered to the regimen: none, some days, most days, or all days in the past week. The scores for managing medication, diet, and exercises were averaged to one single score for independent behavior and one for adherence in managing the therapeutic regimen for each adolescent.

2.5   Stage 4: testing of psychometric properties of the final scale

Data were analyzed using SPSS 18.0 (SPSS Inc., Chicago, IL.) and LISREL 8.80 (SSI, Lincolnwood, IL.). The statistical analyses involved nine subsequent steps:

1. The *sample characteristics* were analyzed using descriptive statistics.
2. All items of the OYOF-29 scale were *data-screened* by examining number of missing, mean and standard deviation of each item.
3. *Exploratory factor analysis with varimax rotation* was conducted to explore to what extent a one-dimensional or multidimensional structure was apparent.
4. *Confirmatory factor analysis* using the LISREL program[39] served to verify the factor structure of the OYOF-29 scale and to test whether observed variables are related with their underlying latent constructs exists. No correlation errors either within or across sets of items were allowed in the model. Four indices of model fit were tested. The cut-off criteria for these four indices were those proposed by Hu and Bentler.[40] First, the overall test of goodness-of-fit assesses the discrepancy between the model implied and the sample covariance matrix by means of a normal-theory weighted least squares test. A plausible model has low, preferably nonsignificant Chi-square (χ^2) values.[41] However, Chi-square is greatly influenced by sample size, leading to difficulty in obtaining desired nonsignificant level.[42] For this reason, we used the relative χ^2, the ratio of χ^2 to degrees of freedom (df), as an informal measure of fit. Generally, a relative χ^2 less than 2 is preferred.[43] Secondly, the Root Means Square Error of Approximation (RMSEA) reflects the estimation error divided by the degrees of freedom as a penalty function. Values on RMSEA below 0.06 indicate small differences between the estimated and observed model. Thirdly, we used the Standardized Root Means square Residual (SRMR), which is a scale invariant index for global fit that ranges between 0 and 1. Values on SRMR lower than 0.08 indicate a good fit. As a fourth index of model fit the Incremental Fit Index (IFI) was calculated. This index compares the independence model (ie, observed variables are unrelated) to the estimated model. Preferably, values on IFI should be larger than 0.95.[40]
5. Then, *item reduction analysis* was performed to develop a short version: the final On Your Own Feet Self-Efficacy Scale (OYOF-SES). Items were removed from the original pool following several criteria: 1) items were excluded one by one following modification indices provided by LISREL and the strength of the factor loadings; 2) eliminating items was stopped when the estimate reliability (measured by Cronbach's α) of each subscale drops below .70; and 3) there should be as few items as possible with a minimum of three, without loss of content and

psychometric quality. Listwise deletion of cases with missing data on these 29 items resulted in *n*=958.

6. *Internal consistency* of the original and final OYOF-SES and subscales was assessed by calculating Cronbach's alphas (α), inter-item correlations within each subscale and correlations between subscales.

7. *Discriminative value*: to evaluate whether the OYOF-SES detects individual differences and helps to identify adolescents with lower levels of self-efficacy in certain areas, one-way ANOVA and T-tests were used to test differences between different age groups, boys and girls, those with lower and higher education, with and without physical limitations / a Dutch surname.

8. In order to investigate *concurrent validity* of the OYOF-SES, Pearson's correlations (*r*) for interval/ratio variables and Spearman's Rho (*ρ*) were calculated of each subscale and the total scale with three 4-item scales measuring self-efficacy in managing the therapeutic regimen, and with two visual analogue scales (range 1 - 10) where adolescents had scored their perceived general independence during consultations (1 item) and in managing therapeutic regimen (average of 3 items).

9. To test the *predictive validity* of the OYOF-SES, multivariable regression analyses were conducted. HRQoL and three self-management measures (independent behavior during the last hospital consultation; independent behavior and adherence with respect to the therapeutic regimen) were regressed on the OYOF-SES, controlling for relevant socio-demographic and disease-related variables. Beta's (β) were calculated and R^2 was used to express the explained variance for each multivariate analysis.

3   Results

Here, we present the analyses to validate the final 'On Your Own Feet' Self-efficacy Scale (OYOF-SES) as described in Stage 4 (2.5).

3.1   Sample characteristics

1318 adolescents (36.1%) responded to the invitation. However, 231 of them reacted by returning the response card explaining why they would not participate in the study: of these, 144 (62.3%) claimed they were not chronically ill. A total of 1087 questionnaires were received (response rate 29.8%) and 958 adolescents assessed their self-efficacy on all 29 items.

Participants' characteristics are provided in Table 1. All major chronic conditions were represented. The five largest diagnostic categories in the sample (taken from the International Statistical Classification of Diseases and Related Health Problems, ICD-9 CM) were: congenital anomalies and conditions originating in the perinatal period (26.3%); neoplasms (12.9%); endocrine, nutritional and metabolic diseases including immunity disorders (11.7%); diseases of the nervous system and sense organs (11.5%); and diseases of the musculoskeletal system and connective tissue (10.2%).

Nonresponse. Analyses revealed that nonresponders were more frequently males and more often had non Dutch surnames, in addition, they were older and were less frequent visitors at the hospital as compared with responders (*P* < .05).

Table 1 *Respondents' characteristics (n = 958)*

	n (%)
Age, *mean (SD), y*	*15.5 (1.9)*
12-15 y, *n* (%)	588 (61)
16-19 y, *n* (%)	370 (39)
Gender, *n* (%)	
male	417 (43)
female	541 (57)
Ethnicity, *n* (%)	
Dutch surname	830 (87)
non Dutch surname	128 (13)
Educational level, *n* (%)[a]	
lower & middle level	524 (56)
higher level	417 (44)
Type of education, *n* (%)[b]	
mainstream	851 (90)
special education for the disabled	93 (10)
Diagnosis after age of six, *n* (% yes)	394 (31)
Prescribed therapeutic regimen (i.e. medications, diet and/or exercise) (yes), *n* (%)	603 (63)
Presence of physical limitations (yes), *n* (%)	274 (29)
Hospital admissions in past 3 years (yes), *n* (%)	232 (24)
	mean (SD)
Number of outpatient visits in past 3 years	16.8 (15.6)
Number of different outpatient departments in past 3 years	3.1 (2.2)
HRQoL KIDSCREEN-10 (10 items; range: 10-100)	76.9 (16.5)
HRQoL DCGM-10 (DISABKIDS) (10 items; range: 10-100)	80.2 (15.9)
Independent behavior during last consultation (7 items; range: 0-7)	2.5 (1.5)
General independence during consultations (1 item; range: 1-10)	7.1 (2.0)
General independence in managing therapeutic regimen (sumscore of average of 3 items; range: 1-10)[c]	8.0 (2.0)

[a] *n = 941*
[b] *n = 944*
[c] *n = 596*

3.2 Data-screening

The item means and standard deviations (*SD*) of the OYOF-29 are presented in Appendix B. All items were screened for univariate and bivariate normality, and to detect outliers. Percentage of missing values on items ranged from 5.8-7.2%, which is within the acceptable range (Table 2).

3.3 Explanatory factor analysis

Explanatory factor analysis using varimax rotation demonstrated that three factors explained 41.7% of the variance. It showed good sampling adequacy in confirmative factor analyses (Kaiser-Meyer-Olkin .90; *P* < .001). One item loaded on two factors. The three factors were similar to the originally proposed subscales (Appendix I).

3.4 Confirmatory factor analysis with 29-item version

All items had factor loadings above 0.46 on the intended factor. Standardized loadings of the items are shown in Table 2. The significant Normal Theory Weighted Least Square χ^2 statistic is not surprising given its sensitivity to sample size; it was 4666.020 (*P* < .01); and the relative χ^2 was 4.9. The RMSEA was just above cut-off value, but acceptable, according to the criteria of Browne and Cudeck.[44] IFI was above cut-off value of .95 and SRMR was equal to the cut-off value of 0.08. All indices indicated that the model fit was acceptable, but left room for improvement and shortening.

3.5 Item reduction analysis

Following the factor loadings, modification indices and checking the internal consistency of each subscale, the stepwise procedure resulted in the subsequent elimination of items 8, 9, 18, 20, 21, 19, 22, 27, 29, 5, 6, 7, 16, 17, 15. Three items (9, 18, and 19) were retained, however, because we considered these of vital importance for adolescents' self-efficacy in preparing for the transition to adult care. This did not affect the order of elimination of items.

The final, short version of the OYOF-SES consisted of 17 items with four, seven and six items for the respective subscales. The overall fit of this final model was improved as the Normal Theory Weighted Least Square χ^2 significantly decreased to 1602,500. The relative χ^2 was 1.5, indicating good fit. RMSEA was equal to the cut-off point of 0.06, and the value of IFI was 0.97 indicating that the specified relations between variables are well supported by the data. The SRMR index was equal to the cut-off point of 0.08, indicating a good fit of the overall model.

Table 2 *Item characteristics and factor loadings of the first full model[a]*

#		valid n	mean	SD	% missing	λ[b]
Subscale Self-efficacy in coping with condition						
	I am convinced that…					
1.	I could manage to tell new classmates or friends about my condition	979	3.35	.88	5.8	.84
2.	I could manage to explain my teacher or boss about my condition	979	3.49	.76	5.8	.88
3.	I could manage to tell my parents or friends if I should be bullied	978	3.38	.75	5.9	.59
4.	I could manage to tell my teacher or boss if I should be bullied	979	3.23	.81	5.8	.59
5.	I could handle the fact that people may treat me "differently"	979	2.86	.97	5.8	.51
6.	I can live with the knowledge that I will have my illness or disability for the rest of my life	979	3.31	.88	5.8	.55
7.	I can accomplish as much as other people with the same illness or disability	979	3.62	.68	5.8	.54
8.	I can accomplish as much as other people without an illness or disability	979	3.51	.76	5.8	.46
Subscale Self-efficacy in knowledge of condition						
	I am convinced that…					
9.	I can clearly describe the future consequences of my illness or disability*	971	3.15	.81	6.5	.49
10.	I know how to reach the doctor or nurse, if necessary	971	3.20	.85	6.5	.60
11.	I can tell which doctor or nurse at the hospital treats me	971	3.36	.87	6.5	.61
12.	I know which regimen the doctor has prescribed me (eg, medications, diet or other lifestyle advice)	971	3.47	.80	6.5	.69
13.	I could explain precisely what my regimen is for	971	3.39	.75	6.5	.69
14.	I realize the future and present consequences of not adhering to my regimen now	970	3.31	.85	6.6	.66
15.	I can find information about my illness or disability myself (books, Internet, leaflets)	971	3.43	.84	6.5	.58

Table 2 *(Continued)*

#		valid n	mean	SD	% missing	λᵇ
16.	I would recognize signs of health decline or of complications of my illness or disability	971	3.21	.86	6.5	.53
17.	I know how often I must go back to the hospital for check-ups	971	3.51	.75	6.5	.59
18.	**I know what happens when I will transfer to adult care***	**971**	**2.47**	**1.03**	**6.5**	**.46**
Subscale Self-efficacy in skills for independent hospital visits						
	I am convinced that…					
19.	**I could talk with the doctor on my own***	**964**	**3.27**	**.81**	**7.2**	**.72**
20.	I could make hospital appointments on my own	964	2.98	.95	7.2	.65
21.	I could manage to travel to the hospital on my own	962	2.58	1.14	7.4	.54
22.	I could explain the doctor how I feel and what my needs are	963	3.48	.66	7.3	.84
23.	**I could answer all questions of the doctor myself**	**964**	**3.38**	**.69**	**7.2**	**.79**
24.	**I would dare ask the doctor anything, even about my personal issues**	**964**	**3.06**	**.86**	**7.2**	**.79**
25.	**I would dare ask the doctor to explain things until I understand it all**	**964**	**3.39**	**.72**	**7.2**	**.80**
26.	**I would dare confess to the doctor that I did not stick to my regimen or hospital appointments**	**964**	**3.20**	**.74**	**7.2**	**.65**
27.	I could deal with it if the doctor should criticize me	964	3.19	.77	7.2	.66
28.	**I would dare tell the doctor if I should disagree with her or him**	**964**	**3.23**	**.76**	**7.2**	**.69**
29.	I could explain to others what we discussed in the consultation room	964	3.29	.77	7.2	.68

ᵃ *Items printed in bold are included in the short version; * items 9, 18, and 19 were retained in the short version on substantive grounds.*
ᵇ *λ is factor loading based on full model.*

Note:
Model fit indices for full (29-item) version: χ^2 (p) = 4666.02 (p <.01); relative χ^2 (χ^2/ df) = 4.9; RMSEA = .06; IFI = .96; SRMR = .08.
Model fit indices for short (17-item) version: χ^2 (p) = 1602.50 (p <.01); relative χ^2 (χ^2/ df) = 1.5; RMSEA = .07; IFI = .97; SRMR = .08.

3.6 Internal consistency and inter-correlations

The reliability of the three sub-scales was good, Cronbach's alpha (α) ranging from .78 - .87 in the original version to .73 - .83 in the final OYOF-SES (Table 3). The Cronbach's α of the 29-item scale was .89 and .85 for the 17-item version. Table 3 shows that the Pearson correlations between the original subscales and their short versions were good (ranging from .88 to .96), indicating acceptable coverage of the sub-dimensions of self-efficacy. The three subscales and the total OYOF-SES were also significantly and positively inter-correlated, indicating that the subscales are conceptually related but also distinct.

3.7 Discriminative value

The mean score for the total OYOF-SES was 55.30 (SD = 7.43) (Table 3). There was a slight, but statistically significant, difference between the mean scores for boys and girls: mean 55.86 (SD = 7.50) versus 54.87 (SD = 7.35); t = -2.03; df = 956; P = .04). In general, younger adolescents are less self-efficacious than older ones (F = 4.28, df1 = 7, df2 = 950; P < .001), but age is not the only relevant factor as is demonstrated by the relatively wide standard deviations: mean at age 12 = 53.55 (SD = 7.91) and at age 18, mean is 57.18 (SD = 7.35). Higher educated adolescents have higher self-efficacy scores than those who attend lower educational levels (t = -2.64; df = 939; P = .009), as well as those without physical limitations (t = 2.55; df = 956; P = .01). No differences were found between adolescents with and without a Dutch surname (t = .26; df = 956; P = .8).

3.8 Concurrent validity

For the investigation of concurrent validity, bivariate correlations were calculated between the final OYOF-SES and its subscales and related constructs assessed in the survey (self-efficacy related to managing the therapeutic regimen and visual analogue scorings of independence during consultations and in managing their regimen). Table 4 demonstrates that the OYOF-SES is positively correlated with similar concepts such as self-reported independence and self-efficacy in managing the therapeutic regimen (P < .001). With the exception of the 'Coping with condition' subscale, the other two subscales also present statistically significant positive correlations. For example, the scoring of general independence during consultations is strongly correlated (r = .50; P < .001) with the 'Skills for independent hospital visits' subscale.

3.9 Predictive validity

Predictive validity was assessed by computing correlations between the OYOF-SES and the KID-SCREEN-10 and DCGM-10 HRQoL measures. Both of these variables were significantly positively associated with the total OYOF-SES and its subscales (P < .001; Table 4), indicating that higher self-efficacy is associated with higher quality of life.

Correlations were also computed with the 7-item scale of independent behaviors during consultations, and – for those who indicated they had a therapeutic regimen – with a 1-item index score

Table 3   *Scale characteristics and inter-correlations of the On Your Own Feet 29-item and final 17-item Self-Efficacy Scales (n = 958)*

	items OYOF-SES 17-items	Cronbach's alpha 29-item version	Cronbach's alpha OYOF-SES 17-items	correlation with 29-item scale[a]	theoretical / actual range	mean (SD)	inter-item correlations range[a]	1	2	3
Subscale Self-efficacy in coping with condition	1, 2, 3, 4	.78	.77	.88	4-16 / 4-16	13.46 (2.45)	.31-72	-	-	-
Subscale Self-efficacy in knowledge of condition	9, 10, 11, 12, 13, 14, 18	.78	.73	.95	7-28 / 7-28	22.34 (3.67)	.13-46	.36**	-	-
Subscale Self-efficacy in skills for independent hospital visits	19, 23, 24, 25, 26, 28	.87	.83	.94	6-24 / 6-24	19.50 (3.37)	.35-61	.40**	.45**	-
On Your Own Feet-SES	1, 2, 3, 4, 9, 10, 11, 12, 13, 14, 18, 19, 23, 24, 25, 26, 28	.89	.85	.96	17-68 / 17-68	55.30 (7.43)	.13-72	.69**	.85**	.81**

** P < .01 (1-tailed)

[a] *Pearson's correlation*

7

Table 4 *Correlations of On Your Own Feet Self-Efficacy Scale (OYOF-SES) and subscales with other measures[a]*

	n	Subscale Self-efficacy in coping with condition (4 items)		Subscale Self-efficacy in knowledge about condition (7 items)		Subscale Self-efficacy in skills for independent hospital visits (6 items)		Total OYOF-SES (17 items)	
		r	p	r	p	r	p	r	p
Concurrent validity									
Self-efficacy therapeutic regimen (scales of 4 items each; 4-point Likert)									
Self-efficacy medication use (α = .75)	464	.10	.03	.23	<.001	.21	<.001	.25	<.001
Self-efficacy diet (α = .84)	185	.14	.05	.26	<.001	.17	.02	.25	<.001
Self-efficacy exercise (α = .77)	283	.15	.01	.34	<.001	.25	<.001	.34	<.001
Scoring of own independency (visual analogue scale; range 1-10)									
General independence during consultations (1 item)	954	.11	.001	.28	<.001	.50	<.001	.40	<.001
General independence in managing therapeutic regimen (average of 3 items)	596	.05	.19	.26	<.001	.27	<.001	.27	<.001

Table 4 *(Continued)*

	n	Subscale Self-efficacy in coping with condition (4 items)		Subscale Self-efficacy in knowledge about condition (7 items)		Subscale Self-efficacy in skills for independent hospital visits (6 items)		Total OYOF-SES (17 items)	
		r	p	r	p	r	p	r	p
Predictive validity									
Health-Related Quality of life									
KIDSCREEN-10 (α = .83)	958	.30	<.001	.22	<.001	.20	<.001	.29	<.001
DCGM-10 (DISABKIDS) (α = .80)	958	.33	<.001	.22	<.001	.25	<.001	.33	<.001
Independent behavior / adherence to therapeutic regimen									
Independent behavior during consultation (7 items; dichotomous; α = .56)	954	.07	.04	.26	<.001	.43	<.001	.34	<.001
Independent behavior in managing therapeutic regimen (sumscore of (average of) 3 items; 4-point Likert)	572	-.05	.25	.14	.001	.10	.02	.10	.02
Adherence to therapeutic regimen (sumscore of (average of) 3 items; 4-point Likert)	572	.04	.39	.15	<.001	.04	.37	.10	.01

[a] *Pearson's correlation r for interval/ratio variables; otherwise Spearman's Rho ρ. P = two-tailed.*

7

of independent behavior in managing therapeutic regimen, and the 1-item index score of adherence (Table 4). Higher self-efficacy is strongly correlated with more independent behaviors during consultations ($P < .001$). This scale was most strongly associated with the 'Skills for independent hospital visits' subscale ($r = .43$; $P < .001$). Total self-efficacy is less strongly, but still significantly, correlated with independent self-management of the regimen ($P = .02$) and adherence ($P = .01$). These measures are most strongly associated with the 'Knowledge of condition' subscale ($P < .001$).

The predictive validity of the OYOF-SES for these outcome measures was further tested in multiple multivariate regression analyses, presented in Table 5. After controlling for age, gender, ethnicity, education (level and type), age at diagnosis, and presence of physical limitations, self-efficacy was positively associated with both HRQoL ($\beta = .31$; $P < .001$). The explained variance for KIDSCREEN was 17%; for DCGM-10 (DISABKIDS) 22%. HRQoL is significantly higher in males and in younger adolescents and those without physical limitations ($P < .001$). In DCGM-10, which measures the experienced impact of the disease, Dutch ethnicity ($P < .001$), higher education ($P = .02$) and attending regular schools (ie, not special education) ($P < .01$) also significantly contributed to higher quality of life.

The presence of more independent behaviors during consultations was strongly predicted by higher self-efficacy ($\beta = .29$; $P < .001$) and by higher age ($\beta = .28$; $P < .001$). Non Dutch adolescents ($\beta = .12$; $P = .000$) and girls ($\beta = .10$; $P = .001$) displayed more independency. The explained variance for this outcome measure was 22%. However, for the other two outcome measures (independent behavior in managing therapeutic regimen and adherence), explained variance was very low. Self-efficacy did not significantly contribute to the prediction of independent behavior in managing the regimen ($R^2 = 5\%$), unlike attending regular education ($\beta = .18$; $P = .001$) and a higher age ($\beta = .09$; $P < .05$). For adherence, self-efficacy contributed somewhat to the total explained variance ($R^2 = 4\%$) with $\beta = .09$ ($P < .05$), just like having a Dutch surname ($\beta = .13$; $P < .01$) and being a boy ($\beta = .12$; $P < .01$).

4   Discussion

Self-efficacy means that persons believe they can exercise control over their chronic condition and perform a specific action in a particular situation. The stronger the self-efficacy, the more successful these adolescents will be in assuming responsibility for their own self-management and maintaining these behaviors over time. Because self-efficacy expectations predict behavior,[45] measuring self-efficacy will help plan and evaluate patient education programs and other interventions.[46] Self-efficacy measurement could also detect variations in adjustment between patients and serve as an indicator of health outcomes. However, many instruments suffer from major limitations.[23]

For lack of a generic instrument, we developed the On Your Own Feet Self-Efficacy Scale (OYOF-SES) and tested this in a large sample of Dutch adolescents with a wide range of chronic conditions. The 17-item scale resulted from item reduction analysis of a 29-item scale generated on the basis of a literature review, expert opinions, and a qualitative study. The OYOF-SES was shown to

Table 5 Multivariable regression analyses with On Your Own Feet Self-Efficacy Scale (OYOF-SES)

	KIDSCREEN-10		DCGM-10 (DISABKIDS condition-generic module)		Independent behavior during consultation		Independent behavior in managing therapeutic regimen		Adherence to therapeutic regimen	
R^2	.17		.25		.22		.05		.04	
F (df); P	25.78 (8, 931); <.001		41.07 (8, 931); <.001		33.22 (8, 931); <.001		4.46 (8, 560); <.001		3.80 (8, 560); <.001	
n	939		939		939		568		568	
	β	P	β	P	β	P	β	P	β	P
Age	-.22	<.001	-.13	<.001	.28	<.001	.09	.03	-.03	.54
Gender (male)	.13	<.001	.12	<.001	-.10	.001	.02	.60	.12	.003
Ethnicity (non-Dutch surname)	-.01	.64	-.12	<.001	.12	<.001	.00	.92	-.13	.002
Educational level (higher)	-.03	.37	.07	.017	.04	.15	.04	.41	-.03	.57
Type of education (special education)	.02	.45	-.09	.004	-.07	.03	-.18	<.001	.02	.64
Age at diagnosis	-.03	.26	.01	.78	-.02	.53	-.03	.48	-.02	.67
Presence of physical limitations (dummy)	-.16	<.001	-.27	<.001	.01	.76	-.07	.09	-.08	.07
OYOF-SES scale	.31	<.001	.31	<.001	.29	<.001	.04	.30	.09	.04

7

be reliable and valid. Confirmatory factor analyses revealed good indices of fit, and concurrent validity with related constructs could be established.

Differences in self-efficacy between adolescents could be established. Younger adolescents, those with physical limitations and with a lower level of education are at risk for lower self-efficacy. In adults, age is a well-known determinant of self-efficacy as a person's experiences and sense of responsibility will grow over time.[47] The age relationship was also found in other studies in adolescents with diabetes,[13,15] but not in a study on asthma.[48] In our study, boys were more self-efficacious than girls, but the difference was small and probably not clinically relevant because the individual differences among boys and girls were much bigger. Our finding coincides with the conclusion from a review stating that, in general, males rate their self-efficacy higher than females, but that the difference is quite small.[47]

Existing self-efficacy scales tend to emphasize disease management aspects rather than social and emotional issues.[29] The final OYOF-SES consists of three subscales representing participants' perceptions of their ability to (1) function on a day-to-day basis; (2) manage symptoms; and (3) manage relations with health care providers. In contrast to all existing disease-specific self-efficacy scales, the OYOF-SES does not only measure regimen-specific tasks but also skills for managing hospital consultations. Our findings support that self-efficacy in these adolescents has a multi-dimensional structure, encompassing core self-management skills related to problem-solving, decision-making, resource utilization, forming of a patient/health care provider partnership, and taking action.[8]

Predictive validity of the OYOF-SES was established for HRQoL and adolescents' independent behaviors during consultations, but was not convincing for adherence to and independence in relation to the therapeutic regimen. The latter may be explained perhaps by the fact that we did not use validated self-report measures for (non)-adherence, but it could also reflect the existence of diverse pathways from self-efficacy to behavior. For example, we did not investigate the role of outcome expectations, the beliefs regarding positive and negative consequences of perform-ing the desired behavior. According to the social cognitive theory,[10] the combined effect of self-efficacy and outcome expectations is much greater than of self-efficacy alone. This was confirmed in a study by Iannotti *et al*[13] who found that self-efficacy and the interaction of self-efficacy with expectations of positive outcomes were significantly associated with diabetes self-management adherence and glycemic control in older adolescents. The effect of self-efficacy was greatest when adolescents had stronger beliefs in the beneficial outcomes of adherence. [13] However, Zebracki and Drotar found that a high outcome expectancy was associated with greater asthma morbidity, but unrelated to self-management or treatment adherence.[48] Also, other theoretical models, such the self-regulation model, emphasize the importance of emotional processes in influencing illness perceptions and coping strategies. Illness perceptions, beliefs and concerns about the necessity of prescribed medications are known to be important predictors of adherence.[49]

According to Bandura,[10] self-efficacy can be enhanced in several ways, including through mastery experiences in which the person gains confidence, observation of similar others (modeling), and verbal persuasion. The organizing focus of such enhancing interventions should be on improving coping, communication, and patient control.[46] This provides a clear direction for nurses, who are to support adolescents with chronic conditions in taking more responsibility for self-care.[4, 19] Examples of promising self-efficacy enhancing interventions in adolescent health care include motivational interviewing,[50] individual transition plans and transition readiness assessments,[51, 52] independent consultations without the parents present,[27, 53] and shared medical appointments (group consultations).[54]

4.1   Strengths and limitations

A major strength of this validation study was the large sample size and the heterogeneity of the sample. However, the very fact that the sample was heterogeneous with respect to congenital and acquired conditions made it impossible to explore relations between self-efficacy and specific clinical outcomes such as glycemic control, pain, specific somatic symptoms, and hospitalizations or specific health care utilization measures. It would be important to investigate whether higher self-efficacy on the OYOF-SES indeed predicts fewer somatic symptoms, better clinical outcomes and less health care utilization. Such positive effects were demonstrated in studies using disease-specific self-efficacy instruments. For example, higher levels of self-efficacy in adolescents with asthma [55], diabetes [13], rheumatoid arthritis [56], and sickle cell disease [16,17] were related to fewer physical and psychological symptoms.

With respect to our sample, the nonresponse rate was fairly high (64%), probably because candidates were approached through an impersonal letter and were asked to access the questionnaire on the Internet. From the returned response postcards it appeared that many adolescents did not consider themselves as chronically ill. The nonresponse analysis revealed that, notably, older adolescents, boys, and adolescents with non Dutch surnames were underrepresented in the sample. This might have affected the outcomes, considering that level of self-efficacy was associated with age and gender. It is impossible, however, to tell in what way.

With respect to the instruments used, most were newly designed for the purpose of this study. Although the internal reliability of the OYOF-SES was good, further validation is warranted especially with respect to its value for evaluation purposes (responsiveness to change). Since this validation study was cross-sectional, issues of causality and predictive value over time still need to be examined. Also, we failed to establish the predictive value for behaviors in the day-to-day management of the therapeutic regimen, likely because independent behaviors and adherence to therapeutic regimen were measured through self-constructed, single questions, thereby limiting reliability. Also, though self-report is useful for collecting information, this method cannot fully capture the complex range of actual behaviors that characterize self-management.

5 Conclusion

Nurses are in an excellent position to enhance adolescents' self-efficacy through supporting independent self-management behaviors which will in turn increase mastery experience. The OYOF-SES proved a useful measure of generic self-efficacy in managing psychological and social functioning, and relations with health care providers in Dutch adolescents with chronic conditions. Self-reported self-efficacy could well serve as an outcome measure in clinical and research settings to evaluate nursing interventions, and as a diagnostic tool to identify need of self-management support. Further research should therefore focus on its responsiveness to change and predictive value for adherence and actual independent behaviors. Also, applicability of the OYOF-SES in other settings and countries needs to be established.

Acknowledgements

#	Ik ben ervan overtuigd dat … [b]	I am convinced that…	mean (SD)	1	2	3
				component		
	Self-efficacy in coping with condition					
1.	het mij lukt om in een nieuwe klas of aan nieuwe vrienden te vertellen wat ik heb	I could manage to tell new classmates or friends about my condition	3.36 (.87)	.09	.66	.19
2.	het mij lukt om zelf aan mijn leraar of baas te vertellen wat ik heb	I could manage to explain my teacher or boss about my condition	3.49 (.76)	.15	.70	.14
3.	het mij lukt om aan mijn ouders of vrienden te vertellen als ik word gepest	I could manage to tell my parents or friends if I should be bullied	3.37 (.76)	.12	.68	.01
4.	het mij lukt om het aan mijn leraar of baas te vertellen als ik word gepest	I could manage to tell my teacher or boss if I should be bullied	3.23 (.81)	.18	.68	.01
5.	het mij lukt ermee om te gaan dat sommige mensen mij 'anders' behandelen	I could handle the fact that people may treat me "differently"	2.86 (.96)	.11	.54	.03
6.	ik kan leven met de gedachte dat ik mijn ziekte of handicap de rest van mijn leven heb	I can live with the knowledge that I will have my illness or disability for the rest of my life	3.31 (.88)	-.04	.55	.16
7.	ik net zoveel kan bereiken in mijn leven als andere mensen met dezelfde ziekte of handicap	I can accomplish as much as other people with the same illness or disability	3.62 (.68)	.04	.47	.23
8.	ik net zoveel kan bereiken in mijn leven als andere mensen zonder ziekte of handicap	I can accomplish as much as other people without an illness or disability	3.51 (.77)	.12	.41	.18
	Self-efficacy in knowledge of condition					
9.	ik de gevolgen van mijn ziekte of handicap voor de toekomst goed kan omschrijven*	I can clearly describe the future consequences of my illness or disability*	3.16 (.81)	.19	.34	.34
10.	ik weet hoe ik de dokter of verpleegkundige moet bereiken, als dat nodig is	I know how to reach the doctor or nurse, if necessary	3.20 (.85)	.36	.10	.44

Appendix B   *(Continued)*

#	*Ik ben ervan overtuigd dat … [b]*	*I am convinced that…*	mean (SD)	1	2	3
				component		
11.	ik kan vertellen bij wie (welke dokter of verpleegkundige) ik onder behandeling ben in het ziekenhuis	I can tell which doctor or nurse at the hospital treats me	3.36 (.87)	.17	.02	.58
12.	ik weet welke behandeling de dokter mij heeft voorgeschreven (bv. medicijnen, dieet, of andere leefregels)	I know which regimen the doctor has prescribed me (eg, medications, diet or other lifestyle advice)	3.47 (.79)	.00	.09	.73
13.	ik kan uitleggen waar mijn behandeling precies voor dient	I could explain precisely what my regimen is for	3.39 (.75)	.11	.18	.63
14.	ik weet wat (later of nu) de gevolgen zijn, als ik me NU niet aan de behandeling houd	I realize the future and present consequences of not adhering to my regimen now	3.31 (.85)	.04	.24	.62
15.	ik zelf informatie kan opzoeken over mijn ziekte of handicap (boeken, internet, folders)	I can find information about my illness or disability myself (books, Internet, leaflets)	3.43 (.84)	.16	.18	.49
16.	ik de signalen van een verslechtering van mijn gezondheid of van een complicatie van mijn ziekte of handicap kan herkennen	I would recognize signs of health decline or of complications of my illness or disability	3.21 (.86)	.15	.21	.46
17.	ik weet hoe vaak ik voor controle moet komen in het ziekenhuis	I know how often I must go back to the hospital for check-ups	3.51 (.75)	.23	.00	.52
18.	ik weet wat er gebeurt als ik later overstap naar de zorg voor volwassenen*	I know what happens when I will transfer to adult care*	2.46 (1.03)	.29	.03	.39
	Self-efficacy in skills for independent hospital visits					
19.	ik best alleen met de dokter kan praten*	I could talk with the doctor on my own*	3.27 (.81)	.73	.01	.14
20.	ik zelf afspraken kan maken in het ziekenhuis	I could make hospital appointments on my own	2.98 (.95)	.66	-.10	.28

7

Appendix B *(Continued)*

#	Ik ben ervan overtuigd dat … [b]	I am convinced that…	mean (SD)	1	2	3
				component		
21.	ik zelf alleen naar het ziekenhuis kan komen	I could manage to travel to the hospital on my own	2.58 (1.14)	.59	-.13	.18
22.	ik bij het spreekuur aan de dokter kan uitleggen hoe het met me gaat en wat mijn behoeftes zijn	I could explain the doctor how I feel and what my needs are	3.48 (.66)	.71	.12	.27
23.	ik bij het spreekuur zelf antwoord kan geven op alle vragen van de dokter	I could answer all questions of the doctor myself	3.38 (.69)	.69	.10	.22
24.	ik bij het spreekuur alles durf te vragen aan de dokter, ook over persoonlijke onderwerpen	I would dare ask the doctor anything, even about my personal issues	3.06 (.86)	.71	.29	.09
25.	ik uitleg durf te vragen aan de dokter tot ik het begrijp	I would dare ask the doctor to explain things until I understand it all	3.39 (.72)	.67	.32	.11
26.	ik het eerlijk aan de dokter durf te vertellen als ik me niet aan de behandeling of aan de afspraken heb gehouden	I would dare confess to the doctor that I did not stick to my regimen or hospital appointments	3.20 (.74)	.57	.31	.03
27.	ik er mee om kan gaan als de dokter kritiek heeft op mij	I could deal with it if the doctor should criticize me	3.19 (.77)	.51	.38	.13
28.	ik het tegen de dokter durf te zeggen als ik het ergens niet mee eens ben	I would dare tell the doctor if I should disagree with her or him	3.23 (.76)	.61	.27	.06
29.	ik aan anderen kan uitleggen wat er op het spreekuur is besproken	I could explain to others what we discussed in the consultation room	3.29 (.77)	.48	.25	.36

[a] *The English translation was performed by a certified English translator, and the back-translation was done by an independent native speaker of both Dutch and English.*
[b] *Items printed in bold are included in the final version.*
* *Items 9, 18, and 19 were retained in the final version on substantive grounds.*

References

1. Mokkink LB, van der Lee JH, Grootenhuis MA, Offringa M, van Praag BMS, Heymans HSA. Omvang en gevolgen van chronische aandoeningen bij kinderen. [Extent and consequences of chronic conditions in children]. *Tijdschr Kindergeneeskd.* 2007;75(4):138-142.
2. Yeo M, Sawyer S. Chronic illness and disability. *Brit Med J.* 2005;330(7493):721-723.
3. Karlsson A, Arman M, Wikblad K. Teenagers with type 1 diabetes--a phenomenological study of the transition towards autonomy in self-management. *Int J Nurs Stud.* 2008;45(4):562-570.
4. Hanna KM, Decker CL. A concept analysis: assuming responsibility for self-care among adolescents with type 1 diabetes. *J Spec Pediatr Nurs.* 2010;15(2):99-110.
5. Giarelli E, Bernhardt BA, Mack R, Pyeritz RE. Adolescents' transition to self-management of a chronic genetic disorder. *Qual Health Res.* 2008;18(4):441-457.
6. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child.* 2008;93(2):160-163.
7. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J. Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns.* 2002;48(2):177-187.
8. Lorig KR, Holman H. Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med.* 2003;26(1):1-7.
9. Holloway A, Watson HE. Role of self-efficacy and behavior change. *Int J Nurs Pract.* 2002;8(2):106-115.
10. Bandura A. *Self-efficacy: the exercise of control*. New York: W.H. Freeman and Company; 1997.
11. Marks R, Allegrante JP, Lorig K. A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part II). *Health Promot Pract.* 2005;6(2):148-156.
12. van der Bijl JJ, Shortridge-Baggett LM. The theory and measurement of the self-efficacy construct. *Sch Inq Nurs Pract.* 2001;15(3):189-207.
13. Iannotti RJ, Schneider S, Nansel TR, Haynie DL, Plotnick LP, Clark LM, *et al.* Self-efficacy, outcome expectations, and diabetes self-management in adolescents with type 1 diabetes. *J Dev Behav Pediatr.* 2006;27(2):98-105.
14. Griva K, Myers LB, Newman S. Illness perceptions and self efficacy beliefs in adolescents and young adults with insulin dependent diabetes mellitus. *Psychol. Health.* 2000;15(6):733-750.
15. Ott J, Greening L, Palardy N, Holderby A, DeBell WK. Self-efficacy as a mediator variable for adolescents' adherence to treatment for insulin-dependent diabetes mellitus. *Child. Health Care.* 2000;29(1):47-63.
16. Clay OJ, Telfair J. Evaluation of a disease-specific self-efficacy instrument in adolescents with sickle cell disease and its relationship to adjustment. *Child Neuropsychol.* 2007;13(2):188-203.
17. Edwards R, Telfair J, Cecil H, Lenoci J. Self-efficacy as a predictor of adult adjustment to sickle cell disease: one-year outcomes. *Psychosom Med.* 2001;63(5):850-858.
18. Coleman MT, Newton KS. Supporting self-management in patients with chronic illness. *Am Fam Physician.* 2005;72(8):1503-1510.
19. Betz CL. Facilitating the transition of adolescents with developmental disabilities: nursing practice issues and care. *J Pediatr Nurs.* 2007;22(2):103-115.
20. Heijmans M, Rijken M, Foets M, de Ridder D, Schreurs K, Bensing J. The stress of being chronically ill: from disease-specific to task-specific aspects. *J Behav Med.* 2004;27(3):255-271.
21. Moos RH, Holahan CJ. Adaptive tasks and methods of coping with illness and disability. In: Martz E, Livneh H, eds. *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. New York: Springer; 2007.
22. Taylor RM, Gibson F, Franck LS. The experience of living with a chronic illness during adolescence: a critical review of the literature. *J Clin Nurs.* 2008;17(23):3083-3091.
23. Frei A, Svarin A, Steurer-Stey C, Puhan MA. Self-efficacy instruments for patients with chronic diseases suffer from methodological limitations--a systematic review. *Health Qual Life Outcomes.* 2009;7(86):1-10.
24. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.
25. Creswell JW. *Research design: qualitative, quantitative, and mixed methods approaches*. 2nd Edition. London: Sage Publications; 2003.

26.   van Staa A, Jedeloo S, van der Stege H, On Your Own Feet Research Group. "What we want": chronically ill adolescents' preferences and priorities for improving health care. *Patient Prefer Adherence.* 21 June 2011 2011;5(1):291-305.

27.   van Staa A, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: The added value of mixed methods research. *Patient Educ Couns.* 2011;82(3):455–464.

28.   Moens A, Grypdonck MH, van der Bijl JJ. The development and psychometric testing of an instrument to measure diabetes management self-efficacy in adolescents with type 1 diabetes. *Sch Inq Nurs Pract.* 2001;15(3):223-233.

29.   Barlow JH, Shaw KL, Wright CC. Development and preliminary validation of a children's arthritis self-efficacy scale. *Arthritis Rheum.* 2001;45(2):159-166.

30.   Edwards R, Telfair J, Cecil H, Lenoci J. Reliability and validity of a self-efficacy instrument specific to sickle cell disease. *Behav Res Ther.* 2000;38(9):951-963.

31.   Betz CL. California Healthy and Ready To Work transition health care guide: developmental guidelines for teaching health care self-care skills to children. *Issues Compr Pediatr Nurs.* 2000;23(4):203-244.

32.   Porter S, Freeman L, Reeves Griffin L. *Transition planning for adolescents with Special Health Care Needs and disabilities: a guide for health care providers.* Boston: Institute for Community Inclusion & UAP at Children's Hospital, Boston; 2000.

33.   Jedeloo S, van Staa A, Latour JM, van Exel NJ. Preferences for health care and self-management among Dutch adolescents with chronic conditions: A Q-methodological investigation. *Int J Nurs Stud.* 2010;47(5):593-603.

34.   Haynes SN, Richard DCS, Kubany ES. Content validity in psychological assessment: A functional approach to concepts and methods. *Psychol Assess.* 1995;7(3):238-247.

35.   Bandura A. Guide for constructing self-efficacy scales. In: Pajares F, Urdan T, eds. *Self-efficacy beliefs of adolescents.* Vol V. Greenwich, CT: Information Age Publishing; 2006:307-338.

36.   de Klerk M, Iedema J, van Campen C. *SCP-maat voor lichamelijke beperkingen op basis van AVO 2003. [Measure for physical limitations based on AVO-2003].* SCP-werkdocument 121. Den Haag: Sociaal Cultureel Planbureau; 2006. ISBN 90-377-0268-6.

37.   Ravens-Sieberer U, Auquier P, Erhart M, Gosch A, Rajmil L, Bruil J, *et al.* The KIDSCREEN-27 quality of life measure for children and adolescents: psychometric results from a cross-cultural survey in 13 European countries. *Qual Life Res.* 2007;16(8):1347-1356.

38.   Schmidt S, Petersen C, Mühlan H, Simeoni MC, Debensason D, Thyen U, *et al. The DISABKIDS Questionnaires Handbook*. Lengerich: Pabst Science Publishers; 2006.

39.   Jöreskog KG, Sörbom D. *LISREL 8: User's Reference Guide* Chicago: Scientific Software International; 1996.

40.   Hu L, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. *Struct Equ Modeling.* 1999;6(1):1-55.

41.   Hayduk LA. *Structural Equation Modeling with LISREL: essentials and advances*. Baltimore: Johns Hopkins University Press; 1987.

42.   Bagozzi RP, Yi Y, Phillips LW. Assessing construct validity in organizational research. *Adm Sci Q.* 1991;36(3):421-458.

43.   Wang L, Fan X, Willson VL. Effects of nonnormal data on parameter estimates and fit indices for a model with latent and manifest variables: an empirical study. *Struct Equ Modeling.* 996;3(3):228-247.

44.   Browne MW, Cudeck R. Alternative ways of assessing model fit. In: Bollen KA, Long JS, eds. *Testing Structural Equation Models*. Beverly Hills, CA: Sage Publishers; 1993:136–162.

45.   Schwartzer R, Luszczynska A. Self-efficacy, adolescents' risk-taking behaviors, and health. In: Pajares F, Urdan T, eds. *Self-efficacy beliefs of adolescents.* Vol V. Greenwich, CT: Information Age Publishing; 2006:139-159.

46.   Marks R, Allegrante JP, Lorig K. A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part I). *Health Promot Pract.* 2005;6(1):37-43.

47.   Gecas V. The social psychology of self-efficacy. *Annu Rev Sociol.* 1989;15(1):291-316.

48.   Zebracki K, Drotar D. Outcome expectancy and self-efficacy in adolescent asthma self-management. *Child Health Care.* 2004;33(2):133-149.

49. Horne R, Weinman J. Self-regulation and self-management in asthma: Exploring the role of illness perceptions and treatment beliefs in explaining non-adherence to preventer medication. *Psychol. Health.* 2002;17(1):17-32.

50. Naar-King S, Suarez M. *Motivational Interviewing with adolescents and young adults.* 1st Edition. New York: The Guilford Press; 2011.

51. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2007;46(1):161-168.

52. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, *et al.* Measuring the transition readiness of youth with Special Healthcare Needs: validation of the TRAQ--Transition Readiness Assessment Questionnaire. *J Pediatr Psychol.* 2011;36(2):160–171.

53. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: a multicentre cohort of adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2005;44(6):806-812.

54. Rijswijk C, Zantinge E, Seesing F, Raats I, van Dulmen S. Shared and individual medical appointments for children and adolescents with type 1 diabetes; differences in topics discussed? *Patient Educ Couns.* 2010;79(3):351-355.

55. Bursch B, Tsao JC, Meldrum M, Zeltzer LK. Preliminary validation of a self-efficacy scale for child functioning despite chronic pain (child and parent versions). *Pain.* 2006;125(1-2):35-42.

56. Vuorimaa H, Honkanen V, Konttinen YT, Komulainen E, Santavirta N. Improved factor structure for self-efficacy scales for children with JIA (CASE) and their parents (PASE). *Clin Exp Rheumatol.* 2007;25(3):494-501.

8

The proxy problem anatomized: child-parent disagreement in health-related quality of life reports of chronically ill adolescents

Jane N. T. Sattoe, AnneLoes van Staa, Henriëtte A. Moll, On Your Own Feet Research Group

Abstract

BACKGROUND: Discrepancy between self-reports and parent-proxy reports of adolescent health-related quality of life has been repeatedly acknowledged in the literature as the proxy problem. However, little is known about the extent and direction of this discrepancy. The purpose of this study is to explore to what extent and in what direction quality of life self-reports of adolescents with chronic conditions and those of their parents differ.

METHODS: A cross-sectional survey was conducted among adolescents suffering from chronic conditions and their parents. Socio-demographic and disease-related characteristics were collected and information about consequences of the chronic condition was assessed. Health-related quality of life was measured with KIDSCREEN-10 and DISABKIDS condition generic measure (DCGM-10). Agreement was analyzed through defining a threshold of agreement based on half of the standard deviation of the score with the highest variance. Agreement occurred if the difference between adolescent and parent scores was less than or equal to half of the standard deviation. Intra-class correlation coefficients and Bland-Altman plots were also computed. The characteristics associated with direction of disagreement were statistically tested with one-way ANOVA and Chi-square tests.

RESULTS: 584 paired health-related quality of life scores were obtained. Ratings from both adolescents and parents were high, compared to European norm data. Differences between adolescents and parents were statistically significant, yet relatively small. Disagreement existed in both directions: in 24.5% (KIDSCREEN-10) and 16.8% (DCGM-10) of the cases adolescents rated their HRQoL lower than did their parent, while in 32.2% (KIDSCREEN-10) and 31.7% (DCGM-10) of the cases the opposite was true. Adolescent's age, educational level and type of education, parent's educational level, number of hospital admissions and several other disease-related factors influenced direction of disagreement.

CONCLUSIONS: In a reasonable proportion of cases the adolescent and parent agreed on the adolescent's health-related quality of life (43-51% of the cases) and most disagreement tended to be minor. Thus, the proxy problem may be smaller than presented in the literature and its extent may differ per population. As adolescents are expected to become partners in their own health care, it is recommended to focus on adolescents' own perceptions of health-related quality of life.

1   Background

Pediatric care professionals have been debating whether parent proxy-reports of their children's Health-Related Quality of Life (HRQoL) are reliable enough.[1, 2] Since both patient and (parent) proxy-reports are often used in pediatric and adolescent care, discrepancies between the two may complicate the use of HRQoL information in clinical practice – for instance, when determining if complementary interventions are needed.[3]

Discrepancies between child HRQoL reports and parent proxy-reports have repeatedly been acknowledged in the literature as 'the proxy problem',[1, 2, 4, 5] but little is known about influencing factors[2, 6-9] and the direction of discrepancy.[10, 11] A systematic review about child-parent agreement in HRQoL reports that agreement is influenced by the child's age, gender and health status. However, no consistent conclusions about the direction and extent of influence of these factors could be derived.[1]

White-Koning *et al*[12] evaluated Quality of Life (QoL) reports of children with cerebral palsy and their parents and found that the following factors influenced agreement: disease severity, the family's socioeconomic status, parental characteristics, and the absence of behavioral problems. They also found that the child's gender did not independently seem to affect child-parent agreement, a finding confirmed by various other studies.[13-17] Most studies on child-parent (dis)agreement, however, focus on specific diagnoses and younger children. The question arises to what extent these results hold for chronically ill adolescents and their parents more generally.

Gaining more insight into child-parent disagreement is particularly valuable in the field of adolescent care. An important goal for care for chronically ill adolescents is preparing the transition from pediatric to adult care. Transition requires good self-management competencies and skills.[18] A first step in enhancing these adolescents' self-reliance is to explore how they evaluate their chronic condition. It also seems important to find out how parents think about their children's health, because parental perception can influence the child's use of health care services[4] and parents are expected gradually to relinquish their care giving responsibilities to their child.[7, 18]

The aim of this study is to explore to what extent and in what direction HRQoL self-reports of adolescents with somatic chronic conditions and those of their parents differ, and to study associated factors.

2   Methods

2.1   Population

The data in this study are derived from a study among adolescents with chronic conditions and their parents recruited from a university children's hospital in the Netherlands, focusing on adolescents' preferences and competencies for health care and self-management (reported elsewhere).[19, 20] This substudy focused on the comparison of adolescent and parent ratings of HRQoL.

The target group consisted of all adolescents aged 12-19 years suffering from a somatic chronic condition or physical impairment, who were treated in the departments of Pediatrics or Pediatric

Surgery at Erasmus MC-Sophia Children's Hospital, Rotterdam, the Netherlands. More specifically: they must have consulted the outpatient clinic at least three times or must have been hospitalized at least once in the three years prior to July 1st 2006. Exclusion criteria were the following: transfer to adult care already effected or documented diagnosis of intellectual impairment.

Eligible adolescents and their parents received written information about the study and were invited to complete a web-based questionnaire accessible for three months (October – December 2006) with a unique code on a secured Internet site.

Response cards were included to encourage adolescents to state, if this should be the case, that they did not qualify for the study, or to explain why they did not wish to participate. All potential participants received a reminder after three weeks. There was no financial remuneration, although participants were entered in a lottery for two iPods and a cell phone.

Approval was obtained from the Erasmus MC Institutional Review Board. Participants were assured of confidentiality and data were processed anonymously. The researchers had no access to participants' medical records.

2.2 Measures

The parent version of the questionnaire was constructed as a mirror version of the adolescent version (ie, parents were asked to rate presumed adolescents' perceptions).

2.2.1 Main outcome variables

Respondents completed the generic short forms of the European KIDSCREEN questionnaire (KID-SCREEN-10)[21] and the European DISABKIDS condition generic measure (DCGM-10).[22, 23] We chose the short versions to reduce the time respondents needed to fill in the questionnaires. Proxy versions are available for both questionnaires. The KIDSCREEN-10 questionnaire is validated to assess HRQoL in both healthy and chronically ill adolescents and children and provides a singular index of global HRQoL.[21, 24] Its 10 items are all scored on a 5-point scale ranging from 'never/not at all' to 'always'. The item scores are combined into a final score on a scale from 0 to 100.[21]

The DISABKIDS condition generic measure was designed to document the HRQoL of children and adolescents and to describe the impact of a disease on their wellbeing.[22, 23, 25] The chronic generic short version assesses HRQoL aspects related to being ill in general. It consists of 12 Likert-scaled items assigned to mental, social and physical domains of HRQoL. The items are scored on a 5-point scale ranging from 'never' to 'always'. Ten items produce a score on a scale from 0 to 100.[25] Two items are related to the use of medication and are not included in the final score.

The availability of both an adolescent and a parent version and the good psychometric properties of the questionnaires were important reasons for choosing the KIDSCREEN-10 and the DCGM-10 questionnaires. The developers report a good internal consistency: Cronbach's alpha is .82 for the child version of the KIDSCREEN-10 and .82 for the parent version. The reported concordance between the parent and child version is also good, with a Pearson coefficient r = .73.[21] For the DCGM-10 the reported Cronbach's alpha is .84 for the child version and .86 for the parent version, with a Pearson coefficient r = .82.[25]

2.2.2 Socio-demographic characteristics, disease-related characteristics and consequences of the condition

Adolescents' age and gender were retrieved from the hospital database. Educational level (higher, indicating preparation for higher education, versus lower) and type of education (regular education versus special education for the physically disabled) of adolescents and parents were informed after in the questionnaire. Because ethnicity is not recorded in the hospital database, the family names were manually classified by two independent researchers into Dutch versus non Dutch, using the Dutch Databank of Surnames. This method has shown good reliability in other studies.[26, 27]

Health care-related characteristics such as the number of outpatient consultations, hospital admissions and the different outpatient departments visited between July 1st 2003 and June 30st 2006 were retrieved from the hospital database. Age at diagnosis (0-5 years, or after the age of 5) and absenteeism from school or work due to illness in the past year were assessed in the questionnaire, by asking how often a day at school or work had been missed (1-item question on a 5-point Likert scale; range: 1 = never, 2 = sometimes, 3 = regularly, 4 = often, 5 = very often). Adolescents and parents also provided information on any therapeutic regimen (ie, medication, diet or exercises) prescribed to the adolescent. Adolescents' limitations in mobility and independence were measured with the Activities of Daily Living Tool (AVO-99).[28] The original 10-item scale was dichotomized: if any physical limitation was present, this was recorded as 1.

The experienced burden of the visibility of the condition was measured through a combination of two questions in each questionnaire. These questions were *"Can other people see that you are / your child is disabled?"* (range: 1 = never, 2 = sometimes, 3 = regularly, 4 = often, 5 = very often/ always) and *"How annoying is this for you / your child?"* (range: 1 = not annoying at all, 2 = not annoying, 3 = a little annoying, 4 = annoying, 5 = very annoying). The sum score of these questions in both versions of the questionnaire was computed by adding up the two ratings. This led to a variable with a theoretical range between 2 and 10.[19]

2.3 Statistical analysis

SPSS 17.0 (SPSS Inc, Chicago, IL) was used for all the statistical analyses. Means, standard deviations and proportions were used for descriptive analyses. McNemar tests were used to test for differences between adolescent and parent reports of dichotomous disease-related variables. Paired-Samples t-tests were performed to test whether the means of the continuous disease-related factors differ significantly between adolescents and parents. Paired Sample t-tests were also performed to test differences in means of HRQoL between adolescents and parents.

To study the direction of agreement between adolescent self-reports and parent proxy-reports, agreement was established according to the definition of clinically meaningful difference in quality of life.[29] Agreement was assumed to occur when the absolute difference between the scores of adolescents and their parents was less than or equal to 0.5 *SD* of the score with the largest variability (this group is referred to as AGREE). Disagreement was also based on computing difference scores and was defined to occur if adolescents rated their HRQoL lower (this group is referred to as ADOL LOW) or higher (this group is referred to as ADOL HIGH) than did their parents – indicated by

a difference in rating that is higher than the threshold for agreement. The extent of disagreement was classified into four levels: from 0.5 to 1 *SD* (minor), from 1 to 1.5 *SD* (intermediate), from 1.5 to 2 *SD* (major), and higher than 2 *SD* (substantial). Alternatively, Bland-Altman plots[30] were computed to study the extent of disagreement and intraclass correlation coefficients (ICCs) were computed to identify any disagreement between adolescents and their parents.

One-way ANOVA and Chi-square tests served to study the demographic, health care- and disease-related factors associated with the direction of agreement. In addition, Tukey post-hoc tests and Chi-square post-hoc tests with Bonferonni correction were applied. Variables were considered significant predictors at *P* < .05 and all the statistical tests were two-tailed.

3   Results

3.1   Studied population

We obtained 584 paired adolescent-parent responses for the HRQoL questionnaires (53.7% of the net adolescent response and 68.1% of the net parent response). Analyses revealed that nonresponders were more frequently males and had non Dutch surnames; they were older and less frequent visitors to the hospital compared to responders (*P* < .05). In the study sample, the five largest diagnostic categories (ICD-9 classification) were: congenital anomalies and conditions originating in the perinatal period (31%); neoplasm (13%); endocrine, nutritional, metabolic diseases, and immunity disorders (12%); diseases of the nervous system and sense organs (11%); and diseases of the musculoskeletal system and connective tissue (33%). Table 1 presents the socio-demographic characteristics of the adolescents and their parents, the disease-related characteristics of the adolescents measured through both the adolescent and parent questionnaire, and the health care-related characteristics, retrieved from the hospital database (including the five largest ICD-9 diagnostic groups). The differences in adolescent and parent perceptions turned out to be significant for two of the four disease-related factors (Table 1).

Since our analysis concerned a selection of all adolescents and parents that participated in the study, we performed additional independent samples Mann-Whitney U tests and t-tests to compare the study sample with the excluded sample. The mean HRQoL did not significantly differ between adolescents for whom parent-proxy reports were available and the other adolescents. The same was true for mean age, gender and educational level. The excluded sample contained a higher proportion of adolescents with non Dutch surnames and of adolescents who were six years or older when their condition was diagnosed. More details of this analysis are presented in an additional file [see Additional file 1]. The tests were repeated between parents for whom adolescent self-reports were available and other parents. The only significant difference here was that the former group contained a higher proportion of mothers.

Table 1 *Description of the study sample according to respondent; No. (%), n = 584 (unless indicated)*

	Adolescents	Parents	P^a
Socio-demographic characteristics			
Gender			
female	322 (55.1)	303 (54.7)	-
male	262 (44.9)	251 (45.3)	
Age			
12 – 15	371 (63.5)	-	-
16 – 19	213 (36.5)		
mean (SD)	*14.9 (1.9)*		
Educational level			
lower	326 (56.2)	338 (59.7)	-
higher	254 (43.8)	228 (40.3)	
Education type[b]			
regular	526 (90.7)	-	-
special	54 (9.3)		
Ethnicity			
Dutch surname	526 (90.7)	-	-
non Dutch surname	54 (9.3)		
Disease-related characteristics			
Age at diagnosis			
0-5 yrs	428 (73.3)	-	-
≥6 yrs	156 (26.7)		
Number of visits of outpatient department			
range	1-111	-	-
mean (SD)	*16.9 (15.4)*		
Number of hospital admissions			
range	0-138	-	-
mean (SD)	*4.9 (9.8)*		
Number of different outpatient departments			
range	1-15	-	-
mean (SD)	*3.1 (2.2)*		
Consequences of chronic condition			
Presence therapeutic regimen			
yes	378 (64.7)	386 (66.1)	ns
Presence physical limitations			
yes	165 (28.3)	133 (22.9)	< .01
School/work absenteeism			
range	1-5	1-5	ns
mean (SD)	*1.9 (.90)*	*1.9 (.85)*	
Experienced burden			
range	2-10	2-10	< .01
mean (SD)	*4.6 (2.1)*	*5.2 (2.1)*	

[a] *McNemar test or Paired-Sample t-test to test if the means differ significantly between adolescent reports and parent reports.*

[b] *n = 580*

3.2   Health-Related Quality of Life

Table 2 provides ranges, means, standard deviations, medians, and interquartile ranges of scores on the KIDSCREEN-10 and DCGM-10 scales.

On average, adolescents scored their HRQoL higher than did their parents. The mean scores of adolescents were respectively 78.3 (SD = 15.6) and 80.2 (SD = 16.3) for KIDSCREEN-10 and DCGM-10. The mean scores of parents were respectively 76.8 (SD = 16.1) and 76.4 (SD = 17.7) for KIDSCREEN-10 and DCGM-10.

The adolescents' median scores were 80.0 and 83.3 for KIDSCREEN-10 and DCGM-10, respectively. These are similar to the Dutch norm data. The medians in the norm data were 77.5 for KIDSCREEN-10 and between 82.5 and 85.0 for DCGM-10. Compared to the European norm data, our mean KIDSCREEN-10 score was higher, but the standard deviation in our sample was similar. The norm score was 71.9 (SD = 15.0).

Table 2   *Main outcome variables; n = 584*

	KIDSCREEN-10	DCGM-10
range	17.5 – 100	16.7 – 100
No. of items	10	10
mean (SD) Adolescents	*78.2 (15.6)**	*80.2 (16.3)***
median Adolescents	80.0	83.3
interquartile range Adolescents	22.5	19.4
mean (SD) Parents	*76.9 (16.0)**	*76.5 (17.6)***
median Parents	80.0	77.8
interquartile range Parents	22.5	27.8

* P < .05 in Related-Samples Wilcoxon Signed Ranks test to test if the means differ significantly between adolescent and parent reports.

** P < .01 in Related-Samples Wilcoxon Signed Ranks test to test if the means differ significantly between adolescent and parent reports.

The Cronbach's alpha was .64 for the child version of the KIDSCREEN-10 self-report and .71 for the parent version. The Cronbach's alphas of the DCGM-10 questionnaire were satisfactory values (child version: .82 and parent version: .87). The degree of correlation between the KIDSCREEN-10 HRQoL score and the DCGM-10 HRQoL score was considerable. The Pearson correlation coefficient was .57 for adolescents and .68 for parents (both P < .01).

The Paired Samples t-tests showed statistically significant differences between the adolescents' and parents' scores (P < .05; Table 2). For KIDSCREEN-10 the mean difference was 1.3 (SD = 17.1); for DCGM-10 the mean difference was 3.7 (SD = 15.1). The threshold for agreement was around 8 points for the KIDSCREEN-10 HRQoL scores and around 9 points for the DCGM-10 HRQoL scores.

Figure 1 represents the distribution of agreement between adolescent and parent reports. For KIDSCREEN-10, 43% of the adolescent-parent pairs agreed with each other. For DCGM-10 this was 51%. Disagreement occurred in either direction. The ICC (using an absolute agreement definition) for the KIDSCREEN-10 measure was .42; for the DCGM-10 measure it was .59. Both were significant ($P < .01$), indicating that there is agreement about adolescent HRQoL between adolescents and their parents.

Figure 1   *Distribution of agreement between adolescent and parent reports (percentage of complete pairs)*

■ Adolescent < Parent   ■ Agreement   ■ Adolescent > Parent

KIDSCREEN-10	DCGM-10
32%	32%
43%	51%
25%	17%
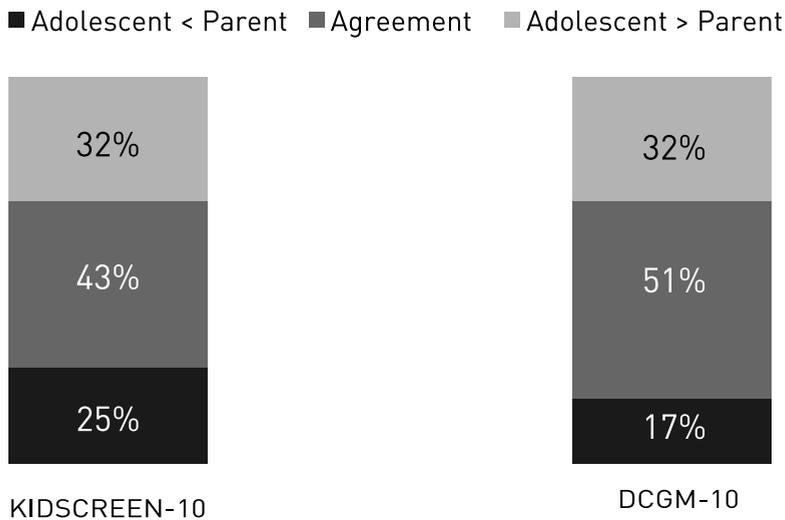
Note: Agreement = adolescent – parent score ≤ .5 greatest SD of scores, ie, the threshold for respectively KIDSCREEN-10 and DCGM-10: 8 points, 9 points.

3.3   Extent of disagreement

Taking the threshold of agreement for KIDSCREEN-10 as 8 points, four levels to explore the extent of disagreement were defined: minor: 8-15 points (0.5 - 1 *SD*); intermediate: 16 - 23 points (1 - 1.5 *SD*); major: 24-31 points (1.5 - 2 *SD*); and substantial: 32 or more points (2 *SD* or higher). Almost half of the disagreement in KIDSCREEN-10 reports was minor; 28% was intermediate; 13% was major; and 13% was substantial (Figure 2). The mean difference between adolescent and parent reports was 1.3 (*SD* = 17.1); most adolescent-parent pairs fell within the agreement limits in the Bland-Altman plot (Figure 3).

Figure 2 *Distribution of disagreement in KIDSCREEN-10 reports (percentage of complete pairs)*
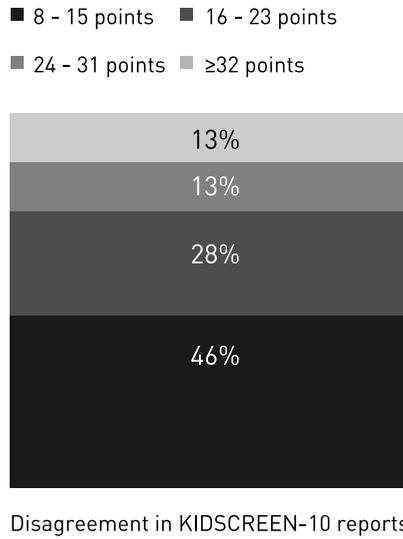

Disagreement in KIDSCREEN-10 reports

Note: Disagreement = adolescent – parent score > respectively 0.5, 1.0, 1.5, and 2.0 times the SD of the HRQoL score with the highest variability.

**Figure 3** *Adolescent-parent agreement in KIDSCREEN-10 reports*
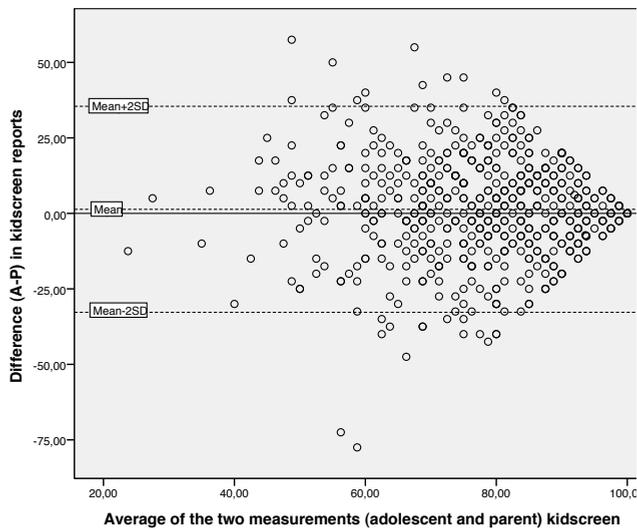
Note: Bland-Altman analysis: mean difference (SD) = 1.3 (17.1)

Figure 4 *Distribution of disagreement in DCGM-10 reports (percentage of complete pairs)*
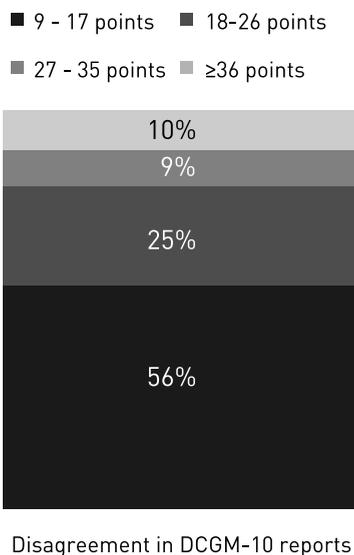
■ 9 – 17 points ■ 18–26 points
■ 27 – 35 points ■ ≥36 points

10%

9%

25%

56%

Disagreement in DCGM–10 reports

Note: Disagreement = adolescent – parent score > respectively 0.5, 1.0, 1.5, and 2.0 times the SD of the HRQoL score with the highest variability.

Figure 5 *Adolescent-parent agreement in DCGM-10 reports*
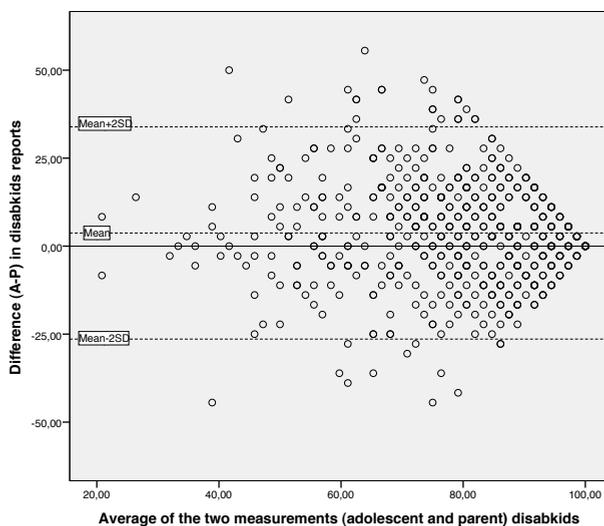
Note: Bland-Altman analysis: mean difference (SD) = 3.7 (15.1)

The threshold of agreement for DCGM-10 was 9 points. The levels of disagreement were respectively: minor: 9-17 points (0.5 - 1 SD); intermediate: 18 - 26 points (1 - 1.5 SD); major: 27-35 points (1.5 - 2 SD); and substantial: 36 or more points (2 SD or higher). Fifty-six percent of the disagreement in DCGM-10 reports was minor; 25% was intermediate; 9% was major; and 10% was substantial (Figure 4). The mean difference between adolescent and parent reports was 3.7 (SD = 15.1); most adolescent-parent pairs fell within the agreement limits in the Bland-Altman plot (Figure 5).

3.4 Direction of disagreement

Three groups of (dis)agreement were defined: ADOL LOW, AGREE, and ADOL HIGH; differences between these groups were tested with one-way ANOVA tests and Chi-square tests.

3.4.1 KIDSCREEN-10

With respect to the rating of global HRQoL, the three groups significantly differed on several demographic characteristics of the adolescent: age, educational level and type of education; and on adolescents' disease-related characteristics as perceived by their parents: physical limitations, school/work absenteeism and experienced disease burden. The results are presented in Table 3.

Post-hoc tests revealed that adolescents in the ADOL LOW group (15.3, SD = 1.9, P < .05) were significantly older than those in the AGREE group (14.8, SD = 1.9, P < .05) and that a lower educational level was more common in the ADOL HIGH group (65.2%) than in the AGREE group (51.4%; P < .017). Furthermore, special education was more common in the ADOL HIGH group (15.0%) than in the AGREE group (6.4%) and in the ADOL LOW group (6.9%; P < .017). The presence of a physical limitation, as perceived by the parent, was more likely in the ADOL HIGH group (33.2%) versus both the AGREE group (20.6%; P < .017) and the ADOL LOW group (13.4%; P < .017). School/work absenteeism as perceived by parents was significantly higher in de ADOL HIGH group (2.1, SD = .94, P < .01) than in the AGREE group (1.8, SD = .80, P < .01) and in the ADOL LOW group (1.8, SD = .78, P < .01). Finally, the experienced disease burden (as perceived by parents) in the ADOL HIGH group (5.5, SD = 1.9) was higher than that in the AGREE group (5.0, SD = 2.1, P < .05).

Table 3 *KIDSCREEN-10 results, mean (SD) or No. (%); n = 584 (unless indicated)*

	ADOL LOW	AGREE	ADOL HIGH	dfM	dfR	F or H	*P*
Socio-demographic characteristics*							
Gender (A) female	91 (63.2)	132 (52.4)	99 (52.7)	2	-	4.73	ns
Gender (P) female	85 (61.6)	124 (52.3)	94 (52.5)	2	-	3.37	ns
Age (A)	15.3 (1.9)a	14.8 (1.9)	14.8 (1.9)	2	581	3.63	< .05
Educational level (A) lower	76 (52.8)	128 (51.4)	122 (65.2)b	2	-	9.25	< .05

Table 3   *(Continued)*

	ADOL LOW	AGREE	ADOL HIGH	dfM	dfR	F or H	*P*
Educational level (P)							
lower	86 (61.9)	146 (60.1)	106 (57.6)	2	-	.552	ns
Education type (A)							
regular	134 (93.1)c	233 (93.6)	159 (85.0)b	2	-	9.83	< .01
Ethnicity (A)							
Dutch surname	134 (93.1)	225 (89.3)	177 (94.1)	2	-	4.33	ns
Disease-related characteristics*							
Age at diagnosis (A)							
before age of six	111 (77.1)	173 (68.7)	144 (76.6)	2	-	5.64	ns
No. of outpatient visits	15.8 (12.5)	16.4 (14.6)	18.4 (18.1)	2	581	1.31	ns
No. hospital admissions	4.3 (6.1)	4.9 (10.7)	5.4 (10.8)	2	581	.494	ns
No. different outpatient departments	2.9 (1.9)	3.0 (2.1)	3.5 (2.6)	2	581	3.36	ns
Consequences of chronic condition*							
Therapeutic regimen (A)							
yes	87 (60.4)	163 (64.7)	128 (68.1)	2	-	1.92	ns
Therapeutic regimen (P)							
yes	88 (61.1)	166 (65.9)	132 (70.2)	2	-	3.15	ns
Physical limitations (A)							
yes	42 (29.2)	60 (23.8)	63 (33.5)	2	-	5.32	ns
Physical limitations (P)							
yes	19 (13.4)c	52 (20.6)	62 (33.2)b	2	-	19.02	< .01
School/work absenteeism (A)	1.9 (.87)	1.8 (.87)	2.0 (.97)	2	581	2.03	ns
School/work absenteeism (P)	1.8 (.78)d	1.8 (.80)	2.1 (.94)a	2	577	7.01	< .01
Experienced burden (A)	4.7 (2.2)	4.5 (2.1)	4.6 (2.1)	2	580	.733	ns
Experienced burden (P)	5.2 (2.3)	5.0 (2.1)	5.5 (1.9)a	2	581	3.07	< .05

[a] *A Tukey post-hoc test revealed that this group differed significantly from the agreement group AGREE on a P < .05 level.*

[b] *A Chi-square post-hoc test with Bonferonni correction revealed that this group differed significantly from the agreement group AGREE on a P < .017 level.*

[c] *A Chi-square post-hoc test with Bonferonni correction revealed that this group differed significantly from the disagreement group ADOL HIGH on a P < .017 level.*

[d] *A Tukey post-hoc test revealed that this group differed significantly from the disagreement group ADOL HIGH on a P < .01 level.*

* (A) stands for information assessed in the adolescent questionnaire, while (P) stands for information coming from the parent questionnaire.

3.4.2   DCGM-10

With respect to the impact of the chronic condition on the adolescent's HRQoL, the (dis)agreement groups differed on educational level of both the adolescent and the parent, the number of hospital admissions and on disease-related characteristics as perceived by parents: presence of physical limitations and experienced burden of the condition. Results are presented in Table 4.

Post-hoc tests revealed that a lower educational level of the adolescent was more common in the ADOL LOW group (67.0%) than in the AGREE group (51.3%; $P < .017$). A lower parent educational level was also more common in the ADOL LOW group (72.5%) versus both the AGREE group (57.5%) and the ADOL HIGH group (56.8%; $P < .017$).

The number of hospital admissions was higher in the ADOL HIGH group (6.6, $SD = 15.0$, $P < .05$) versus the AGREE group (4.2, $SD = 5.9$, $P < .05$). A physical limitation, as perceived by the parent, was more likely in the ADOL HIGH group (30.3%) versus the ADOL LOW group (14.4%; $P < .017$). Finally, the disease burden (as perceived by the parent) was significantly higher in the ADOL HIGH group (5.8, $SD = 2.0$) versus both the AGREE group (4.9, $SD = 2.1$, $P < .01$) and the ADOL LOW group (4.9, $SD = 2.1$, $P < .01$).

Table 4   *DCGM-10 results, mean (SD) or No. (%); n = 584 (unless indicated)*

	ADOL LOW	AGREE	ADOL HIGH	dfM	dfR	F or H	P
Socio-demographic characteristics*							
Gender (A) female	55 (56.1)	163 (54.2)	104 (56.2)	2	-	.243	ns
Gender (P) female	51 (54.8)	155 (54.2)	97 (55.4)	2	-	.068	ns
Age (A)	14.8 (1.9)	14.9 (1.9)	15.0 (1.9)	2	581	.192	ns
Educational level (A) lower	65 (67.0)b	153 (51.3)	108 (58.4)	2	-	7.82	< .05
Educational level (P) lower	66 (72.5)b, c	168 (57.5)	104 (56.8)	2	-	7.42	< .05
Education type (A) regular	89 (91.8)	272 (91.3)	165 (89.2)	2	-	.744	ns
Ethnicity (A) Dutch surname	88 (83.7)	274 (91.0)	165 (94.1)	2	-	2.00	ns
Disease-related characteristics*							
Age at diagnosis (A) before age of six	82 (83.7)	212 (70.4)	134 (72.4)	2	-	3.14	ns
No. of outpatient visits	16.6 (12.8)	16.2 (14.4)	18.3 (18.0)	2	581	1.09	ns
No. hospital admissions	3.9 (5.8)	4.2 (5.9)	6.6 (15.0)a	2	581	4.04	< .05
No. different outpatient departments	3.0 (2.0)	3.1 (2.3)	3.4 (2.3)	2	581	1.38	ns
Consequences of chronic condition*							

Table 4   *(Continued)*

	ADOL LOW	AGREE	ADOL HIGH	dfM	dfR	F or H	*P*
Therapeutic regimen (A) yes	62 (63.3)	188 (62.5)	57 (30.8)	2	-	2.38	ns
Therapeutic regimen (P) yes	65 (66.3)	190 (63.1)	131 (70.8)	2	-	3.03	ns
Physical limitations (A) yes	28 (28.6)	88 (29.2)	49 (26.5)	2	-	.433	ns
Physical limitations (P) yes	14 (14.4)c	63 (21.1)	56 (30.3)	2	-	10.20	< .01
School/work absenteeism (A)	1.8 (.79)	1.9 (.97)	1.9 (.85)	2	581	.801	ns
School/work absenteeism (P)	1.7 (.72)	1.9 (.89)	2.0 (.84)	2	577	2.83	ns
Experienced burden (A)	4.8 (2.3)	4.6 (2.1)	4.6 (2.1)	2	580	.640	ns
Experienced burden (P)	4.9 (2.1)d	4.9 (2.1)	5.8 (2.0)a	2	581	12.27	< .01

[a] *A Tukey post-hoc test revealed that this group differed significantly from the agreement group AGREE on a P < .05 level.*

[b] *A Chi-square post-hoc test with Bonferonni correction revealed that this group differed significantly from the agreement group AGREE on a P < .017 level.*

[c] *A Chi-square post-hoc test with Bonferonni correction revealed that this group differed significantly from the disagreement group ADOL HIGH on a P < .017 level.*

[d] *A Tukey post-hoc test revealed that this group differed significantly from the disagreement group ADOL HIGH on a P < .01 level.*

* (A) stands for information assessed in the adolescent questionnaire, while (P) stands for information coming from the parent questionnaire.

4   Discussion

This study investigated the extent and direction of disagreement between HRQoL reports of adolescents with a variety of somatic chronic conditions and their parents in a sample of 584 pairs. About half of the pairs agreed on adolescents' HRQoL. For the other pairs, statistically significant disagreement in either direction was found. Yet, the differences were relatively small (respectively 74% (KIDSCREEN-10) and 81% (DCGM-10) of the adolescent-parent disagreement was minor or intermediate). The ICCs and Bland-Altman plots also indicated reasonable agreement between adolescents and parents.

Our results would suggest that the 'proxy problem' of child-parent disagreement in HRQoL evaluations is perhaps not as meaningful as is often assumed in the literature. For example, White-Koning et al found a higher rate of disagreement (64%) than we did (respectively 57% and 48% for KIDSCREEN-10 and DCGM-10).[12] They also defined agreement in terms of a clinically meaningful difference in quality of life. HRQoL was measured with the KIDSCREEN-52 questionnaire and their population size was comparable to ours.[12] However, White-Koning et al studied 8-12-year-old children with cerebral palsy, whereas we studied 12-19-year-old adolescents with a variety of chronic conditions. So it seems plausible that the size of the proxy problem may depend on disease category and age group. Shaw et al,[11] for instance, found a rate of disagreement in a population of adolescents with juvenile rheumatic arthritis consistent with our findings, while Ylimainen et al[31] found poor agreement between parent and child reports of the child's HRQoL in young persons with limb reduction deficiency. Next to this, the small thresholds of agreement in our study, 8 and 9 points respectively for KIDSCREEN-10 and DCGM-10, are additional arguments to question the size of the proxy problem, because they indicate little variance in HRQoL. Most of the disagreement we found was minor.

Yet, a considerable proportion of adolescents and parents disagreed with each other on HRQoL. In these cases, the adolescent usually reported a higher HRQoL. This is consistent with previous studies in children with chronic conditions.[4, 10, 16, 17, 32, 33] Conversely, a minority of parents rated their child's HRQoL higher than did the adolescents themselves, which has not often been reported in the literature on chronically ill adolescents.[4]

In our study, adolescents who disagreed with their parents on both global HRQoL and HRQoL related to the impact of a chronic condition were more likely to have a lower educational level than those who agreed with their parents. An explanation could perhaps be found in social status differences, which are seen to be related to the differential ways that parents and children rate health.[32] The same explanation could hold for our finding that parents with a lower educational level are more likely to overestimate their child's HRQoL instead of agreeing with their child or underestimating the HRQoL.

Regarding age, Cremeens et al[33] and Majnemer et al[34] found that agreement increased with increasing age of the adolescent. In our study, however, adolescents agreeing with their parents were more often younger than the ones who rated their global HRQoL lower than did their parents. This conflicting finding may perhaps be explained by the fact that the aforementioned studies did not correct for direction of disagreement. Previous findings on direction of disagreement mostly focused on the ADOL HIGH group.[4, 32, 34] The discrepancy between findings is plausible since

the effect of age was evident only when comparing the ADOL LOW group with the AGREE group. Parents were more likely to overestimate HRQoL of older adolescents. The differences in age of adolescents were minute, indicating that even a few months in this crucial period of adolescence make a difference. Perhaps parents saw older adolescents as more capable when it comes to living with a chronic condition. As another explanation, parents may be less well informed about their child's wellbeing at adolescent age, implicating that health care providers would do well to focus on the opinions of the adolescents themselves.

Adolescents who rated their HRQoL higher than their parents did, scored less well on the health care-related and disease-related factors (interpreted by the parents) than did all other adolescents. Parents seem to attach greater value to these factors. Our finding is consistent with literature findings indicating that disease-severity factors are associated with child-parent disagreement.[10, 35] This is also seen in the cases of adolescents who rated their HRQoL lower than did their parents. These adolescents' parents perceived fewer physical limitations, lower school absenteeism, and lower experienced burden than the parents that underestimated their child's HRQoL. Perhaps the differences in HRQoL perception could in part be explained by the discrepancy in adolescents' perception and parents' perception of the impact of the condition on quality of life. As an additional argument, the correlation between the proxy versions of KIDSCREEN-10 and DCGM-10 is higher than that between the child versions, indicating that parents perceive a stronger relation between general HRQoL and the HRQoL related to impact of the condition than adolescents do. Gates *et al* also found that parents focus more on functional aspects than adolescents do.[36] While adolescents tend to focus on their abilities, the parent's perspective is more likely one of disability.[37] Therefore, adolescent self-reports and parent proxy-reports of HRQoL are not interchangeable. Furthermore, given that parents of chronically ill children themselves report seriously lower HRQoL compared to controls,[38] and parental wellbeing is known to influence (proxy) measurement of HRQoL,[12] assessing parents' own HRQoL is perhaps more meaningful than asking them for a proxy-report of their child.

Finally, the adolescents' mean HRQoL score was higher than the European norm score for KIDSCREEN-10 [21] – despite the fact that all adolescents were chronically ill.[21] The descriptive statistics indicate a ceiling effect, which may be ascribed to the so-called 'disability-paradox' explaining "why many people with serious and persistent disabilities report that they experience a good or excellent quality of life when to most external observers these individuals seen to live an undesirable daily existence".[39] This paradox implies that HRQoL for persons with disabilities is broader than just health, encompassing the person's social context and environment too. Perhaps our population benefited from a positive and supportive social environment. Next to this, adaptation (a phenomenon referred to as 'response shift'[40]) cannot be ruled out. The majority of our adolescent population has lived with their condition for almost all of their conscious life.

4.1   Strengths and limitations

Our study included a large sample of adolescents with a wide range of chronic conditions. The sample was heterogeneous in terms of congenital and acquired conditions, and in age. It originates from the largest university hospital in the Netherlands, which comprises all major pediatric

subspecialties. Yet the wide range of chronic conditions made it impossible to explore the impact of nature of the disease and that of disease severity. This diversity in chronic conditions may also be responsible for the wide standard deviations in both adolescent and parent reports. However, since chronically ill adolescents all face the same adaptive challenges,[41] studying chronic conditions in general is not considered a flaw. Disease severity, however, is a broad concept that can be operationalized in different ways. In this study, we included only health care- and disease-related variables into the models but no psychological measures. Certain psychological factors, such as child-parent conflict, could have had an effect on the extent and direction of disagreement.[42] Also, the short forms of the used HRQoL questionnaires did not allow for analyses at the level of the different HRQoL domains. There are indications that child-parent (dis)agreement is dissimilar in these domains[43] – for example, one study established more disagreement for the mental (psychological) domain compared to the physical and social domains.[12] Analyses of (dis)agreement at the level of specific domains could have provided further insight in the spread and nature of (dis)agreement in our study population. For further research, we recommend using the longer versions to be able to test for differences between the HRQoL domains.

Furthermore, the nonresponse rate was fairly high (63%). More information on the sample and the nonresponse is reported elsewhere.[19] Candidates received an impersonal letter and were required to access the questionnaire on the Internet. The returned response postcards made clear that many candidates did not feel 'chronically ill'. Apart from this, lay views on 'being ill' and the importance of 'being normal' may have played a role here. Nonresponders consulted the hospital less frequently than did responders, which may imply that they represent a healthier population, although it may also indicate no-show. The nonresponse analysis revealed that notably older adolescents, boys and adolescents with non Dutch surnames were underrepresented. This might have affected the outcomes. It is impossible, however, to tell in what way. Adolescents excluded from analysis because there was no proxy questionnaire available, more often had a non Dutch surname. An explanation for this finding might be that non Dutch parents were facing more language and cultural barriers than Dutch parents when asked for participation in (HRQoL) research. This has been reported before in Turkish and Moroccan ethnic minority patients in the Netherlands.[44] However, little is known about child-parent disagreement in ethnic minorities. Therefore it is impossible to tell if, and how, this finding affected the outcomes of the study. The same is true for our finding that excluded adolescents more often reported having received a diagnosis after the age of six. There were no significant differences between the total sample of parents and our sub-sample of parents, with the exception of parent gender: the sub-sample included more mothers. This is the case in most of the comparable studies.[1] The effect of parent gender on HRQoL assessment, however, is unknown.[1]

5   Conclusion

In this sample of chronically ill adolescents and their parents any disagreement was predominantly minor, which raises questions about the size of the proxy problem. However, in around 20% of all the cases adolescents and parents disagreed to a greater extent. Parents tended to underestimate

their child's HRQoL, but still a reasonable number overestimated it. Parents' and adolescents' educational level and adolescent's age should be taken into account when interpreting HRQoL-reports. Parents seem to weigh the impact of the condition more heavily than their child does, indicating that self-reports and parent-proxy reports are not interchangeable. However, since adolescents are expected become partners in their own health care and HRQoL measures provide relevant clinical information about psychosocial functioning, it is recommended to focus on the adolescent's own perceptions of HRQoL.

Acknowledgements

Additional file 1 *Comparison of the study sample with the sample excluded from analysis; No. (%) or mean (SD)*

	Adolescents included in study sample n = 584	Adolescents excluded from analysis n = 455	P*
Socio-demographic characteristics			
Gender			
female	322 (55.1)	263 (57.8)	ns
male	262 (44.9)	192 (42.2)	
Age			
12 – 15	371 (63.5)	268 (58.9)	ns
16 – 19	213 (36.5)	187 (41.1)	
mean (SD)	*14.9 (1.9)*	*15.1 (2.0)*	
Educational level[a,b]			
lower	326 (56.2)	202 (55.2)	ns
higher	254 (43.8)	164 (44.8)	
Ethnicity			
Dutch surname	526 (90.7)	351 (77.1)	<.001
non Dutch surname	54 (9.3)	104 (22.9)	
Disease-related characteristics			
Age at diagnosis[c]			
0-5 yrs	428 (73.3)	282 (62.8)	<.001
≥ 6 yrs	156 (26.7)	167 (37.2)	
Number of visits of outpatient department			
range	1-111	1-146	ns
mean (SD)	*16.9 (15.4)*	*17.6 (17.9)*	
Number of hospital admissions			
range	0-138	0-139	ns
mean (SD)	*4.9 (9.8)*	*4.4 (10.1)*	
Number of different outpatient departments			
range	1-15	1-15	ns
mean (SD)	*3.1 (2.2)*	*3.1 (2.3)*	
Consequences of chronic condition			
Presence therapeutic regimen[d]			
yes	378 (64.7)	259 (60.0)	ns
Presence physical limitations[e]			
yes	165 (28.3)	129 (28.9)	ns
School/work absenteeism			
range	1-5	1-5	ns
mean (SD)	*1.9 (.90)*	*2.0 (.93)*	
Experienced burden			
range	2-10	2-10	ns
mean (SD)	*4.6 (2.1)*	*4.8 (2.3)*	

* Independent samples Mann-Whitney U test to test if distributions or Paired-Sample t-test to test if the means differ significantly between study sample and excluded sample.

[a] *n = 580 for study sample*

[b] *n = 366 for excluded sample*

[c] *n = 449 for excluded sample*

[d] *n = 432 for excluded sample*

[e] *n = 447 for excluded sample*

References

1. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. *Qual Life Res.* 2001;10(4):347-357.
2. Theunissen, NCM, Vogels AG, Verrips GHW, Verloove-Vanhorick SP, Kamphuis RP, *et al.* The proxy problem: child report versus parent report in health-related quality of life research. *Qual Life Res.* 1998;7:387-39.
3. Ravelli A, Viola S, Migliavacca D, Pistorio A, Ruperto N, Martini A. Discordance between proxy-reported and observed assessment of functional ability of children with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2001;40(8):914-919.
4. Upton P, Lawford J, Eiser C. Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Qual Life Res.* 2008;17(6):895-913.
5. Gordijn MS, Cremers EM, Kaspers GJ, Gemke RJ. Fatigue in children: reliability and validity of the Dutch PedsQL Multidimensional Fatigue Scale. *Qual Life Res.* 2011;20(7):1103-1108.
6. Vrijmoet-Wiersma CM, Kooloos VM, Koopman HM, Kolk AM, van der Laan I, Grootenhuis MA, *et al.* Health-related quality of life, cognitive functioning and behaviour problems in children with Langerhans cell histiocytosis. *Pediatr Blood Cancer.* 2009;52(1):116-122.
7. Sawyer MG, Reynolds KE, Couper JJ, French DJ, Kennedy D, Martin J, *et al.* A two-year prospective study of the health-related quality of life of children with chronic illness--the parents' perspective. *Qual Life Res.* 2005;14(2):395-405.
8. Klassen AF, Miller A, Fine S. Health-related quality of life in children and adolescents who have a diagnosis of attention-deficit/hyperactivity disorder. *Pediatrics.* 2004;114(5):e541-547.
9. Solans M, Pane S, Estrada MD, Serra-Sutton V, Berra S, Herdman M, *et al.* Health-related quality of life measurement in children and adolescents: a systematic review of generic and disease-specific instruments. *Value Health.* 2008;11(4):742-764.
10. White-Koning M, Grandjean H, Colver A, Arnaud C. Parent and professional reports of the quality of life of children with cerebral palsy and associated intellectual impairment. *Dev Med Child Neurol.* 2008;50:618-624.
11. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: parents as proxies of adolescents with juvenile idiopathic arthritis. *Arthritis Rheum.* 2006;55(2):189-198.
12. White-Koning M, Arnaud C, Dickinson HO, Thyen U, Beckung E, Fauconnier J, *et al.* Determinants of child-parent agreement in quality-of-life reports: a European study of children with cerebral palsy. *Pediatrics.* 2007;120(4):e804-814.
13. Janse AJ: *Quality of life of chronically ill children. Perception of patients, parents and physicians.* Enschede: Febodruk Enschede; 2005.
14. Janse AJ, Uiterwaal CS, Gemke RJ, Kimpen JL, Sinnema G. A difference in perception of quality of life in chronically ill children was found between parents and pediatricians. *J Clin Epidemiol.* 2005;58(5):495-502.
15. Arrington-Sanders R, Yi MS, Tsevat J, Wilmott RW, Mrus JM, Britto MT. Gender differences in health-related quality of life of adolescents with cystic fibrosis. *Health Qual Life Outcomes.* 2006;4:5.
16. Britto MT, Kotagal UR, Chenier T, Tsevat J, Atherton HD, Wilmott RW. Differences between adolescents' and parents' reports of health-related quality of life in cystic fibrosis. *Pediatr Pulmonol.* 2004;37(2):165-171.
17. Youngblade LM, Shenkman EA. Congruence between parents' and adolescents' reports of special health care needs in a Title XXI program. *J Pediatr Psychol.* 2003;28(6):393-401.
18. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child.* 2008;93(2):160-163.
19. van Staa A, van der Stege HA, Jedeloo S, Moll HA, Hilberink SR. Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors. *J Adolesc Health.* 2011;48(3):295-302.
20. van Staa A, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: the added value of mixed methods research. *Patient Educ Couns.* 2011;82(3):455-464.
21. The KIDSCREEN Group Europe. *The KIDSCREEN Questionnaires - Quality of life Questionnaires for children and adolescents. Handbook.* Lengerich: Pabst Science Publishers; 2006. ISBN 3-89967-334-4.

22. Bullinger M, Schmidt S, Petersen C. Assessing quality of life of children with chronic health conditions and disabilities: a European approach. *Int J Rehabil Res.* 2002;25(3):197-206.

23. Petersen C, Schmidt S, Power M, Bullinger M. Development and pilot-testing of a health-related quality of life chronic generic module for children and adolescents with chronic health conditions: a European perspective. *Qual Life Res.* 2005;14(4):1065-1077.

24. Ravens-Sieberer U, Gosch A, Rajmil L, Erhart M, Bruil J, Duer W, *et al.* KIDSCREEN-52 quality-of-life measure for children and adolescents. *Expert Rev Pharmacoecon Outcomes Res.* 2005;5(3):353-364.

25. Schmidt S, Petersen C, Mühlan H, Simeoni MC, Debensason D, Thyen U, *et al. The DISABKIDS Questionnaires - Handbook.* Lengerich: Pabst Science Publishers; 2006. ISBN 3-89967-166-X.

26. Fiscella K, Fremont AM. Use of geocoding and surname analysis to estimate race and ethnicity. *Health Serv Res.* 2006;41(4 Pt 1):1482-1500.

27. Bouwhuis CB, Moll HA. Determination of ethnicity in children in The Netherlands: two methods compared. *Eur J Epidemiol.* 2003;18(5):385-388.

28. de Klerk M, Iedema J, van Campen C. *SCP-maat voor lichamelijke beperkingen op basis van AVO 2003 [SPC measure for physical limitations based on AVO 2003].* SCP-werkdocument 121. Den Haag: Sociaal Cultureel Planbureau; 2006. ISBN 90-377-0268-6.

29. Norman GR, Sloan JA, Wyrwich KW. Interpretation of changes in health-related quality of life: the remarkable universality of half a standard deviation. *Med Care.* 2003;41(5):582-592.

30. Bland JM, Altman DG. Statistical methods for assessing agreement between two methods of clinical measurement. *Lancet.* 1986;327:307-310.

31. Ylimainen K, Nachemson A, Sommerstein K, Stockselius A, Norling Hermansson L. Health-related quality of life in Swedish children and adolescents with limb reduction deficiency. *Acta Paediatr.* 2010;99(10):1550-1555.

32. Verrips GH, Vogels AG, den Ouden AL, Paneth N, Verloove-Vanhorick SP. Measuring health-related quality of life in adolescents: agreement between raters and between methods of administration. *Child Care Health Dev.* 2000;26(6):457-469.

33. Cremeens J, Eiser C, Blades M. Factors influencing agreement between child self-report and parent proxy-reports on the Pediatric Quality of Life Inventory 4.0 (PedsQL) generic core scales. *Health Qual Life Outcomes.* 2006;4:58.

34. Majnemer A, Shevell M, Law M, Poulin C, Rosenbaum P. Reliability in the ratings of quality of life between parents and their children of school age with cerebral palsy. *Qual Life Res.* 2008;17(9):1163-1171.

35. Waters E, Stewart-Brown S, Fitzpatrick R. Agreement between adolescent self-report and parent reports of health and well-being: results of an epidemiological study. *Child Care Health Dev.* 2003;29(6):501-509.

36. Gates P, Otsuka N, Sanders J, McGee-Brown J. Functioning and health-related quality of life of adolescents with cerebral palsy: self versus parent perspectives. *Dev Med Child Neurol.* 2010;52(9):843-849.

37. Oeffinger D, Gorton G, Bagley A, Nicholson D, Barnes D, Calmes J, *et al.* Outcome assessments in children with cerebral palsy, part I: descriptive characteristics of GMFCS Levels I to III. *Dev Med Child Neurol.* 2007;49(3):172-180.

38. Hatzmann J, Heymans HS, Ferrer-i-Carbonell A, van Praag BM, Grootenhuis MA. Hidden consequences of success in pediatrics: parental health-related quality of life--results from the Care Project. *Pediatrics.* 2008;122(5):e1030-1038.

39. Albrecht GL, Devlieger PJ. The disability paradox: high quality of life against all odds. *Soc Sci Med.* 1999;48(8):977-988.

40. Sprangers MA, Schwartz CE. Integrating response shift into health-related quality of life research: a theoretical model. *Soc Sci Med.* 1999;48(11):1507-1515.

41. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.

42. Giannakopoulos G, Dimitrakaki C, Pedeli X, Kolaitis G, Rotsika V, Ravens-Sieberer U, *et al.* Adolescents' wellbeing and functioning: relationships with parents' subjective general physical and mental health. *Health Qual Life Outcomes.* 2009;7:100.

43. van Dijk J, Huisman J, Moll CM, Schouten-van Meeteren AYN, Bezemer PD, Ringens PJ, *et al.* Health-related quality of life of child and adolescent retinoblastoma survivors in the Netherlands. *Health Qual Life Outcomes.* 2007;5:65.

44. Hoopman R, Terwee CB, Muller MJ, Ory FG, Aaronson NK. Methodological challenges in quality of life research among Turkish and Moroccan ethnic minority cancer patients: translation, recruitment and ethical issues. *Ethn Health*. 2009;14:237-253.

Part 3

Transitional care:
the need for action

9

Crossing the transition chasm: experiences and recommendations for improving transitional care of young adults, parents and providers

AnneLoes van Staa, Susan Jedeloo, Jetty van Meeteren, Jos M. Latour

Abstract

BACKGROUND: Transition from pediatric to adult health care has received little attention in the Netherlands. This study aimed to (i) map experiences with the transfer to adult care of young adults with chronic conditions, (ii) identify recommendations for transitional care from the perspectives of young adults, their parents and health care providers.

METHODS: Semi-structured interviews with 24 young adults after transfer (aged 15-22 years; diagnosed with hemophilia, diabetes mellitus, spina bifida, congenital heart disorders, cystic fibrosis, juvenile rheumatoid arthritis, or sickle cell disease), 24 parents, and 17 health care providers. Thematic analysis was performed.

RESULTS: Only the hemophilia department offered a structured transition program, most patients had not been prepared for transition. Experiences and views of patients, parents and professionals mainly overlapped and were condensed into four core themes. Two are related to moving to adult care: (1) 'leaving pediatric care is a logical step'. Leaving familiar surroundings was harder for parents than for young adults who displayed a positive 'wait-and-see' attitude; and (2) 'transition is complicated by cultural gaps between pediatric and adult services'. Young adults and parents felt lost after transfer and recommended their peers 'to be alert and involved'. Providers also recognized the cultural chasm between both services and worried about noncompliance, lost to follow-up, and lack of independence. Two other themes indicated priorities for improvement: (3) 'better patient and parent preparation' for differences between health care settings and for new roles and responsibilities with respect to self-management; and (4) 'more collaboration and personal links' between pediatric and adult care providers.

CONCLUSIONS: Action is required to cross the chasm between pediatric and adult-oriented care. Preparation for transition should start early and focus on strengthening adolescents' independency without undermining parental involvement. Building bridges between services, gaining trust and investing in new personal relations is a challenge for all parties involved: transition is about responding and bonding.

1   Introduction

Moving from pediatric to adult health care is an essential process in the lives of all young people with chronic conditions. It is one of the many and often concurrent transitions in their lives. In addition to becoming socially independent, young people must move from parental control of their health care needs to self-care.[1] Thus, parents are important partners in transition as well.

We have gradually begun to understand the challenges of realizing a successful transition to adult life where optimal social participation is the ultimate goal. There is more to it than just transfer to adult health care services: major changes in both the organization and content of adolescent health care are required. Three elements must be pursued: a cultural shift in staff's attitudes, effective transition programs, and teaching adolescents to become active partners in their own care.[2]

Ideally, transition of care is a purposeful, planned process – as advocated in policy documents, professional guidelines and expert opinion articles.[3, 4, 5] The term "transition" refers to the process prior to and after the "transfer" event, that is, the actual shift from pediatric to adult health care.[6] Only few experimental studies have evaluated transition programs and services,[7] mostly conducted in the UK.[8, 9] There is no evidence that particular models of transition are more effective than others.[10, 11] Nevertheless, there is a growing evidence base on key elements of transitional care.[12-14] Especially the need to improve care in different chronic conditions has been documented well in various countries with different health care systems. Several empirical, mostly qualitative, studies explored user expectations and experiences around their transfer to adult services.[1, 15-25]

There is much communality in the themes described in these studies – across issues and conditions. Recurrent themes are: the challenges facing patients and parents alike while *moving to adult services* (going into the unknown; going into a different world; disrupted relationships and ways of working); recognition of the *opportunities for personal growth* (a shift in roles and responsibilities between adolescent and parents), and *recommendations* for improving both process and outcomes.

Unfortunately, daily clinical practice has not kept up with the current evidence and recommendations. In the Netherlands there are no national standards or policy documents advocating transitional care, and few professional guidelines address this issue. A recent survey showed that only a minority of Dutch institutions offered any transitional services to their adolescent patients,[26] while the experiences and effects of transition to adult services on patients and their parents have hardly been studied.[27-29]

Therefore, we designed an explorative study with a twofold aim: (1) to map experiences with the recent transfer to adult care of young adults with chronic conditions receiving care in one university hospital in the Netherlands; and (2) to identify recommendations to improve the transition process from the perspectives of young adults, their parents, and health care providers.

2   Methods

2.1   Design and setting

A qualitative study was conducted between 2004-2007 in the Erasmus University Medical Center - Sophia Children's Hospital, a tertiary referral centre, among young adults diagnosed with hemophilia (HP), diabetes mellitus (DM), spina bifida (SB), congenital heart disorders (CHD), cystic fibrosis (CF), juvenile rheumatoid arthritis (JRA), or sickle cell disease / thalassemia (SCD). They were eligible for participation if they had no record of intellectual disabilities and had been transferred to adult care in the past two years. There was one exception, however: at the time of the research (2004), 25 CF-patients over 18 had not been transferred yet. Hence, those to be transferred within six months were listed. The term 'young adult' refers to those already or about to be transferred to adult care (aged over 16), to be distinguished from 'adolescents' still receiving pediatric care.

The study consisted of semistructured interviews conducted with young adults, parents and health care providers from pediatric and adult care.

2.2   Sampling

In each diagnostic group, three young adults were randomly selected from a list of patients officially discharged in the previous two years (and in CF from the waiting list). Parents were approached after the young adult had given consent. When young adults did not reply within two weeks, they were sent a reminder letter and a few days later, they were called by telephone. When no consent was given or the young adult could not be reached, new patients were approached – until three in each group had consented in an interview. During the interviews it appeared that three SB-patients had not visited adult care yet, so we invited three extra participants.

For the health care provider interviews, pediatric providers were first interviewed and asked to suggest participants from adult care.

2.3   Data collection

All interviews were carried out by a trained nursing or physiotherapy student after extensive training by the research team (authors of this paper). The patient and parent interviews were conducted at home and lasted 45 to 120 minutes. Parents and young adults were interviewed separately along the lines of an interview guide developed by the researchers. The interviews focused on expectations and experiences with transfer and perceived quality of care in pediatric and adult services (Box 1). Disease-specific questions were added to gain more in depth information on specific health care needs.

In the health care provider interviews, attitudes toward transition and current transfer practices were explored. They were interviewed at their workplaces and interviews lasted from 25 to 60 minutes.

Box 1   *Interview guide: young adults' and parents' experiences with care provision in the transitional period*

How did the transfer from pediatric to adult care turn out?
Positive and negative experiences and feelings about transfer
Timing of the transfer
Preparation of the transfer
Felt readiness at time of transfer
Experiences with care coordination
Suggestions to improve transitional care

How did you experience and value pediatric care?
Positive and negative aspects of pediatric care
Roles of parents, young persons, and health care providers during consultations
Experiences with different professionals in the health care team
Last visit, leaving pediatric care
Experiences with inpatient facilities
Age-appropriate care: attention paid to psychosocial aspects, coping with condition, career, future
Suggestions to improve pediatric care

How do you experience and evaluate adult care?
Positive and negative aspects of adult care
Perceived differences with pediatric care
Roles of parents, young people, and health care providers during consultations
Experiences with different professionals in the health care team
First visit, reception
Experiences with inpatient facilities
Age-appropriate care: attention paid to psychosocial aspects, coping with condition, career, future
Suggestions to improve adult care

What advice would you give to others in the same circumstances?

2.4   Data analysis

Interviews were digitally recorded, transcribed verbatim and then imported into the qualitative software package ATLAS.ti 5.0 (www.atlasti.com). Thematic analysis was chosen for its flexibility and theoretical freedom, and applied in several phases.[30]

   All interviews were reviewed and coded by AvS who read them repeatedly to familiarize herself with the data. Initial codes (subthemes) were formulated on the basis of the interview guide. Subsequently, these were modified, expanded or merged as new issues emerged during the analysis. The third step was collating subthemes to identify potential themes; emerging themes were checked iteratively in other interviews. Possible relations between respondents' experiences and relevant (demographic) characteristics were identified. The research team examined the coding process and the emerging themes were discussed continually until consensus was reached.

2.5   Validity and reliability

To enhance credibility we used both peer debriefing in the research team as well as respondent validation. The recommendations for transitional care were presented to 27 pediatric health care providers from the same hospital in three focus groups (data not reported here).[31]

2.6   Ethical aspects

The study protocol was approved by the Institutional Review Board of the Erasmus University Medical Center. All study participants gave written informed consent. Researchers had no access to hospital charts and all participants were assured of confidentiality and anonymity.

3   Results

3.1   Study population

Table 1 presents the characteristics of the 65 participants: 24 young adults (mean age 18.7 years; range 15-22), 24 parents, and 17 health care providers. Six young adults (25%) were about to transfer, 18 had already been transferred to adult care. One-third of them was now being treated in nonacademic hospitals. Twenty young adults gave permission to invite their parents for an interview; all parents consented, so 20 pairs were interviewed. Of four young adults, parents were not interviewed. Three SB-patients and one CHD-patient who did not wish to participate, gave permission to interview their parents.

Response rates varied between the various conditions. The three approached CF-patients all consented, but in SB, RA, and CHD the initial response rates were 30% or less. Nonparticipation was mostly related to lack of interest, as "*I do not go to the hospital very often*", and *"my disease does not bother me".* Those under 18 and those with limited disease activity were less inclined to participate. Also, 60% of all selected SCD-patients, and 38% of DM-patients, could not be reached through mail or phone.

3.2   Transfer practices in seven chronic conditions

Table 2 gives an overview of transfer practices as reported by health care providers. At the time, only the HP department offered a structured transition program in which patients learned about their medication and were taught to make appointments and to take responsibility for self-management of their medical condition. HP patients also had the opportunity to meet their new providers during holiday camps.

The other departments started discussing the impending transfer no earlier than 6 to 12 months in advance. Timing of the transfer differed between the chronic conditions: SB-patients were discharged at 15 - 16 years; other young adults were usually transferred between 16 and 18 years, by the time they graduated from secondary education. Although many claimed that timing depended

Table 1 *Background characteristics of 65 study participants*

	Response rate[a]	Total	Young adults	Parents	Health care providers
Number of participants		65	24	24	17
Male / female			13 / 11	3 / 21	6 / 11
Treatment setting: pediatric care / adult care			6 / 18	9 / 15	11 / 6
Chronic condition					
Diabetes Mellitus	60%	8	3	3	2
Hemophilia	60%	7	3	2	2
Spina Bifida	30%	17	6	9	2
Congenital Heart Disorders	23%	7	3	3	1
Cystic Fibrosis	100%	8	3	3	2
Juvenile Rheumatoid Arthritis	27%	12	3	3	6
Sickle Cell Disease	60%	6	3	1	2
Young adults' characteristics					
Age					
15-18 years			13		
19-22 years			11		
Non Dutch ethnic background			3		
Living independently			5		
Studying / working / unemployed			19 / 2 / 3		
Educational level: higher / lower			10 / 14		
Health care providers characteristics					
Medical specialist					10
Nurse specialist / Nurse Practitioner					6
Physiotherapist					1

[a] *Percentage of young adults that consented to participate after initial approach.*

upon adolescents' developmental readiness and not on a fixed age, only the HP department tested knowledge and self-management skills.

Transfer usually was to specialists of the same university hospital; only patients with DM were transferred to other hospitals for organizational reasons. Those with JRA and SB were offered the option of a specialist closer to home. For CF, SCD, and JRA centralization of care was still at a preliminary stage and joint treatment protocols were not yet in place. The CF Center was the first to formulate a joint mission statement, but protocols and procedures had not been aligned yet and many young adults were still on the waiting list for transfer.

Even though the children's hospital and the adult facilities are located at the same premises, most professionals in pediatric and adult care of the same specialty did not know each other. Joint consultations were not organized and there was no formalized consultation between pediatric

Table 2 *Overview of transfer practices in seven subspecialties at time of research*

	HP (2004)	DM (2004)	CHD (2004)	CF (2004)	SB (2004)	JRA (2006)	SCD (2007)
Written medical transfer document	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Personal links between pediatric and adult care	Yes	No	Yes	No	Yes	No	No
Dedicated professional for treating young persons in adult care	Yes	No	Yes	No	Yes	No	No
Joint mission statement / written policy	No	No	No	Yes	No[a]	No	No
Flexible moment of transfer possible	Yes	No	No	Yes	No	Yes	Yes
Transfer always within same institution (including transfer of medical dossier)	Yes	No	Yes	No	No	No	Yes
Meeting new health care providers in advance	Yes	No	No	No	Yes	No	No
Joint medical treatment protocol with adult team	Yes	No	Yes	No[a]	No	No	No
Alignment of procedures and approach of young persons / parents	Yes	No	Some	No	No	No	No
Multidisciplinary team approach in both settings	Yes	No	No	Yes	Yes	Yes	Limited
Structural consultation between pediatric and adult care	Yes	No	No	No	No	No	No
Structured transition program, including early preparation	Yes	No	No	No	No	No	No
Joint consultations, transition clinic	No	No	No	No	No	No	No

[a] *In preparation at time of research.*

Note: HP, hemophilia; DM, diabetes mellitus; CHD, congenital heart disease; CF, cystic fibrosis; SB, spina bifida; JRA, juvenile rheumatoid arthritis; SCD, sickle cell disease.

and adult health care. The HP department offered a farewell meeting and personal hand-over of patients after crossing the bridge that connects the children's and adult hospital, a ritual some other health care providers thought of as *"perhaps a bit over the top"*.

Both settings differed largely in treatment protocols and working methods. The multidisciplinary team approach, providing more holistic care, was standard in pediatric care. Most adult care facilities had higher patient load, less consultation time and fewer supporting staff. For example, in CHD, the adult specialist was dedicated to congenital heart diseases, but he worked alone, and did

not consult with the pediatric team. In the SCD-department, psychosocial support was not always continued after transfer.

3.3 Moving on to adult services

Two core themes related to the process of 'moving on to adult care' emerged from the interviews with young adults, parents, and health care providers:

1. Leaving pediatric care is a logical step.
2. Transition is complicated by cultural gaps between pediatric and adult services.

3.3.1 Leaving pediatric care is a logical step

Parents and young adults shared many common views. Moving on to adult care is not only *"inevitable"* but also appropriate for grownups. Even (parents of) young adults with serious, life-threatening conditions recognized the necessity. Many young adults said they had *"grown out of the children's hospital",* whereas few did not feel ready yet.

Before transfer, young adults did not know what to expect, but most did not seem too worried about it (displaying a positive, wait-and-see attitude) – in contrast to their parents. Several parents said they had been sorry to leave, as they felt *"safe"* in the children's hospital:

I didn't want to leave the trusted environment where everything is familiar and where you're in charge. […] Still, raising kids implies that you have to let them go and accept that they make their own choices. I didn't like it that he had to go, but I saw the necessity. And now I see it's good. (Parent of 18-year-old male, HP)

Another parent was opposed to transfer because her son did not adhere to treatment; besides, in pediatric care they knew their situation very well. The son himself had a different attitude:

I'll need to get used to it. I've known my doctor awfully long, for 18 years. But I'll just see what's going to happen. […] Actually, I'm getting too old now for a children's hospital. Seems to be the right age [for transfer] because I'm an adult now, aren't I? (19-year-old male, CF)

Health care providers recognized transfer as *"a natural process"* that is *"age-appropriate".* They had different views on the proper age to transfer. Some considered 18 years as *"the upper limit".* Pediatric providers tended to stress that age boundaries should be flexible, depending upon the adolescent and his parents. Adult providers felt that *"most young people are ready"* to be more involved in their health care. Young adults need to be *"pushed"* a little, as leaning back comes naturally to them. This may create tension with *"overprotective"* parents. A pediatric rheumatologist felt that parents were *"being sidetracked in adult care"* as consultants "*are not used to conducting triadic consultations"*, but she also acknowledged that *"we are pampering those children too much".* All health care providers were convinced that parents have more difficulty in leaving behind the trusted pediatric environment than young adults themselves, and that the young adults are tired of being *"patronized"* by pediatric staff and parents.

3.3.2 Transition is complicated by cultural gaps between pediatric and adult services

Some parents and young adults looked back at transfer as *"no big deal"* and even as *"peanuts"*, when the process had been smooth or *"seamless".* But most young adults and especially parents said it had been more stressful and difficult than anticipated. Those who had had frequent contact with pediatric providers found it hard to establish trust and familiarity with the new staff, as reflected by metaphors like *"being lost", "falling into a deep hole", "feeling abandoned"* and even *"waking up in a horror movie".* However, this was seen as temporary; transition was perceived as a rite of passage: *"you have to get used to it, that's all".*

Parents and young adults described pediatric surroundings and relationships with the staff as warm, familiar, cozy and trusted *("feels like a second home", "they are family")*. They were mostly negative about the look-and-feel of the adult-oriented surroundings *("treated like a number", "sterile environment"*).

All young adults and parents, except those in the HP department, said they had been uninvolved in transfer decisions and had not been prepared for the differences. Two of the three young adults with HP and their parents were positive about the transition program, one felt unprepared. However, also HP patients noticed differences in way of working. The farewell ceremony was seen as a clear demarcation of 'bridging services', but one young man thought this was *"more for parents than for us boys".*

All participants could easily identify upsides and downsides of both settings (Table 3). Advantages of pediatric care (*"it's familiar, home-like")* contrasted with disadvantages of adult care (*"everything is new and feels different")*. At the same time, perceived disadvantages of the children's hospital (*"some treat you as if you're still a child")* were compensated for in the new setting ("*you take more control of your own affairs"). Young adults liked it that they were *"more involved as an adult"* and that consultations were more business-like. Still, all had to get used to new staff, procedures and protocols, and a different care culture.

Most challenging for parents and young adults is the role shift with respect to self-management and responsibility. Parents found it difficult to step aside, even though they agreed it was necessary. They wondered whether their children could take up the full responsibility for their treatment. Young adults, too, had noted that more independence and self-reliance was expected of them. They were positive about their potential to achieve this, even though they found it hard *"to be fully responsible now"*. Gaining trust in your child was the major challenge for parents; gaining trust in your own capacities and developing trusted relations with new health care providers that for young adults.

All health care providers recognized cultural differences between the pediatric and adult-oriented specialties that complicated transfer. These are summarized in Table 4. The adult care *"business-like approach"* was often contrasted with the pediatric *"holistic, system-oriented approach".* Health care providers in both settings felt there is truth in the stereotypes about pediatrics being a "*pampering"* environment where "*everything is arranged for"*, and that parents and patients in adult services *"fall into a deep hole when they have to do things by themselves"*. The hematologist said that SCD-patients were *"somewhat spoilt in pediatrics"*, while his pediatric colleague stressed that intensive surveillance is needed because of poor adherence, high no-show rates, and (psycho)

Table 3 *Advantages and disadvantages of pediatric and adult care, as perceived by young adults and their parents*

Advantages of pediatric care	Disadvantages of adult care
• familiar • cozy, relaxed atmosphere • child-friendly • parents involved • respect for parent's and patient's expertise • trusted providers' expertise • good collaboration between care providers • multidisciplinary teamwork • holistic approach • excellent conditions for inpatient care • everything is arranged for you	• unfamiliar • formal, stand-offish; strict • not focused on young people • parents less welcome • lack of respect for patient's and parent's expertise • expertise not always trusted • poor coordination with pediatric care / with other specialist providers • team approach is not self-evident • less attention paid to psychosocial issues • poor conditions for inpatient care • confronted with older patients • different methods and treatment procedures than in pediatric care • you have to arrange everything yourself
Advantages of adult care	**Disadvantages of pediatric care**
• age-appropriate (adult-like) • business-like, matter-of-fact atmosphere • more focus on responsibility & self-management • young adult more involved in decision making • exciting to build new relationships; make a fresh start • information relevant to adult issues • possibility to chose hospital closer to home • new treatment options possible	• childish, not age-appropriate • confronted with young children • less encouragement of independence & self-management • adolescent less involved in decision making; presence of parents limits freedom to speak / youth participation • "fixed" relationships • lack of information on adult issues • doctors reluctant to treat aggressively and to try new options

social problems. In turn, this was accredited for by the hematologist, who felt that social work should be continued after transfer. Pediatric providers worried that their long-standing bonds would be severed after transfer, and those in adult services saw failure to adhere to treatment and loss to follow-up as the major risks of transition.

Generally, health care providers saw the large cultural gaps as unwanted. Differences in treatment protocols and procedures should be smoothened, but on the other hand, a pediatric rheumatologist felt: "*We should not pamper transition as well! Patients could handle this very well themselves*".

Table 4 *Cultures of care: typical differences between pediatrics and adult specialist medicine, according to interviewed health care professionals*

Pediatrics	Adult care
Typical patient is healthy, only a minority is chronically or terminally ill	Typical patient has complex, chronic and often progressive condition
Patient seen as fragile, vulnerable, dependent	Patient seen as coresponsible, self-reliant
Family-centered care: parents always involved	Individual-based care
Shared decision making and education focuses on parents rather than on patients	Empowerment of patient by means of with information and expectations of self-reliance
Informal, relaxed communication style; empathic but also more paternalistic	Formal and direct communication style; more distant and 'business-like'
Holistic care: attention to developmental and learning issues, social functioning	Disease-oriented care: strong focus on treatment complications and adherence
Interdisciplinary team approach	Specialist orientation, less team work and care coordination

3.4   Recommendations for better transition

Table 5 summarizes recommendations from the study participants. Two core themes emerged:
1.   Better patient and parent preparation.
2.   Better organization and communication between pediatric and adult care.

3.4.1   Better patient and parent preparation

Young adults and parents would have appreciated more information, at an earlier stage, and more time to make choices: *"give young people more time to decide when they want to leave. Do not tell them: now you're 16, you have to go."* Both wished to be involved in the decision making. Several young adults suggested it would be nice to meet the new health care provider before transfer and all wanted to be prepared for differences in ways of working.

Health care provides generally supported these recommendations. Almost all felt that the present process of transition should be improved; only the HP-nurses were content because they already worked in that way. Transfer now often is too abrupt, with patients and parents not being well prepared. However, transitional care goes beyond the mere transfer of information:

It is a pathway in which patients are ready to take on full responsibility for their health care at the moment they transfer, while the parental role is declining. (Pediatric pulmonologist)

Health care providers mentioned that young adults therefore needed to know more about their condition, and should improve self-management skills. Involvement during consultations should be encouraged, as "*we deal with patients, not parents"* (adult rheumatologist). Since parents are almost always present in pediatric consultations, this poses an enormous challenge.

Health care providers proposed concrete interventions such as seeing adolescents independently (without parents), using checklists and individual transition planning, developing a tran-

Table 5 *Recommendations from young adults, parents and health care providers to improve transitional care*

Better patient and parent preparation	
Young adults	**Health care providers**
• Start preparation earlier • Allow more time and more choice • Give more information enabling informed choices • Prepare in advance for differences in care • Try to become more independent • Prepare yourself and be more involved in your own care *Be alert and involved: do it yourself* **Parents** • Start preparation earlier • Allow more time and more choice • Adjust transfer to other life transitions • Give more information, also on paper • Involve parents • Make young people responsible, they are the experts *Don't leave this to professionals, do it yourself, be alert and hang on there*	• Preparation should start early and transition should be gradual • Timing of transfer should be flexible, adjusted to other life transitions and to patient readiness • Set up transition clinics / young adult teams; introducing new providers early • Involve adolescents more in their own care and listen to their opinions • Have parents stay involved, but in a different role • Prepare patients and parents for differences between pediatric and adult care • Work systematically on the fostering of adolescents' independence; use checklists and transition readiness assessments • Encourage more independent behaviors during consultations; see adolescents without parents • Less is more: less pampering, but more self-management of young people • Pay attention to adult issues such as career, sexuality and intimate relations, etc. *Do not pamper; prepare patients to take care in their own hands*

Better organization of transition, more communication between providers	
Young adults	**Health care providers**
• Arrange a meeting with adult health care providers before transfer; organize a period of joint care • Improve logistics of the transfer process • Improve communication and alignment between pediatric-adult care • Make young adults feel welcome in adult services • Give patients access to their own dossier • Respect patient expertise *Make transfer safe, smooth and simple; respect patient expertise* **Parents** • Appoint someone who coordinates care • Organize transition clinic / period of joint care • Procedures in adult care should be the same as in pediatric services • Improve logistics of the transfer process • Improve communication and alignment between pediatric-adult care • Improve communication with parents in adult services • Make parents feel welcome in adult services *Make transfer safe, smooth and simple; keep parents involved*	• Invest in personal relationships between providers in pediatric and adult care • Exchange knowledge and experiences through clinical lectures, patient rounds, internships, and staff exchange • Formulate a joint mission statement • Smoothen differences in working ways and treatment protocols as best as possible • Appoint as a go-between a professional in adult health care who is trained / interested in treating young adults • Involve doctors and consultants, do not leave transition to nurses and social workers • Organize joint medical consultations / transition clinics • Organize regular consultation (transition meetings) between pediatric and adult care about patients to be transferred • Design a structured transition program • Pediatric care should provide multi-disciplinary referral notes timely; adult care should provide feedback on transferred patients • Invest in building good communication and relations with young adults: it pays back *Invest in relations between pediatric and adult services and with patients and parents*

sition protocol, and organizing joint consultations. Differences in care should be smoothened, whenever possible.

Parents and young adults indicated that preparation for transfer requires action from all actors involved, not only from health care providers. Young adults advised their peers to be involved and more alert: *"make sure you set the facts straight"; "get familiar with your medication, prepare a to-discuss list before you see your doctor".* Parents acknowledged the expert role of their children, but also stressed that parents' involvement during transition remains crucial. Their advice to other parents was: *"be alert, don't make yourself dependent upon professionals"*, *"hang on there"*.

3.4.2 Better organization and communication between pediatric and adult care

A common view was that the logistics and organization of the transfer itself would benefit from resources to develop transition clinics or joint clinical pathways. Nevertheless, enhanced communication is most needed. Not only between doctors and nurses of pediatric and adult services, but also with social workers and other allied professionals. Young adults and parents stressed that adult care should be more accessible and responsive to their needs: *"make us feel welcome".*

Almost all health care providers regretted that they did not know their counterparts personally but only through 'paper'. Closer personal bonds and enhanced integration between adult and pediatric services were seen as key conditions for better communication and collaboration. Professionals recommended having more staff exchange, holding consultation meetings about patients before and after transition, and setting up joint clinics.

4   Discussion

This was the first study in the Netherlands to explore the lived experiences of young adults transferring to adult care, their parents, and pediatric and adult-oriented providers. Although a multi-actor perspective is recommended,[25] few qualitative studies have included adult-oriented health care providers' views.[17, 19, 32] Transitional care should not be confined to a pediatric paradigm and be disconnected from the principles and practice of adolescent medicine.[6] Our study showed that health care providers were well aware that transition poses challenges to patients and parents, and were motivated to initiate change. This is exemplified by the recently improved collaboration between pediatric and adult providers in the Erasmus University Medical Center, where transition programs are now being implemented for adolescents with CF, JRA, SB, and SCD.

Adolescents with any kind of chronic condition are facing the same challenges with respect to their transition to adulthood.[17, 33] We studied seven patient populations and the type of condition hardly seemed to influence practices, attitudes, and concerns. This supports generic, inter-specialty developments in transitional care.[13]

All actors in our study considered moving to adult services as 'normal' and even desirable for young people with chronic conditions, as reported in other studies as well.[19, 20, 25] Nevertheless, parents and adolescents may have different perceptions before transfer: parents will typically be anxious, whereas adolescents display a wait-and-see attitude.[21, 25] Still, the prevailing belief of

our young adults and parents after transfer was that transition was desirable and well-timed. In another study, we found that a majority of adolescents still in pediatric care already felt 'ready for transfer'.[34]

Time is an essential element in transition,[17] and therefore longitudinal studies are required to explore the initial phase, midcourse experience, and outcome of the transition experience.[35] The only longitudinal study performed so far established that feelings about the desirability of transfer, the appreciation of medical care, the relationships with health care providers, and the parent's role changed over time during the stages of transition.[20] Adolescents anticipating transfer to adult care had ambivalent feelings, but after transfer they acknowledged benefits of the adult-oriented system. Pediatric providers may therefore overestimate reluctance to transfer or anticipated difficulties.

Our study confirmed that young adults and parents often experience the transfer period as challenging, because moving to adult services implies going into a different world and adjusting to the new environment while leaving behind familiar surroundings and trusted health care providers.[1, 15, 22] Especially sudden and unprepared transfer will affect young adults and parents.[16] Pediatric providers and their patients are often assumed to be strongly attached,[36] but this is not always the case. It seems that the strength of the relationship depends on the duration and intensity of contact. Transition is not only a time of losses, but also of gains[35]: new relations offer new opportunities and it is a period for disease-related learning and personal growth for adolescents and parents.[24] Transition should therefore be incorporated in a comprehensive, lifespan perspective on health care for young people with chronic conditions.[37]

According to health care providers in our study, cultural and organizational differences between pediatric and adult-oriented services should be smoothened out, as they inhibited transition. For young adults, however, it seemed most important to anticipate on these differences so they could adapt to them. Also, the change from family-oriented to patient-oriented care was not unwelcome for them. Young adults preferred health care staff addressing just them rather than their parents, being spoken to in an adult manner, and being responsible for their own care.[18, 20] The shift from parental care to self-care in roles between adolescents and parents is the most decisive element for successful transition.[1, 16, 25]

With respect to transitional care, all actors in our study agreed that the move from pediatric to adult services should be better prepared for, that young people's views should be listened to and that they should be taught self-management skills. Meeting providers alone during consultations is often seen as a useful intervention to encourage self-efficacy and self-reliance in adolescents.[12, 23, 38] Also, earlier discussions about transition, opportunities to meet new providers and visits to adult-oriented venues prior to transition might aid in the transition process.[12, 16, 17, 19] Positive attitudes toward transition and more discussions have a positive effect on follow-up,[39] and on transfer readiness.[34]

The key challenge for health services relates to bridging the differing cultures of pediatric and adult health care. More communication and collaboration between health care providers is essential.[21, 24] These issues seem universal features of the Western biomedical system characterized by high specialization and fragmentation of services. According to Rosen,[5] physicians in all

specialties develop unique styles of care, reinforced by stereotypes, socialization, and the working environment. However, cultural differences between pediatric and adult care are also functional: the clear role for parents, the supportive practice style and the awareness of developmental issues in pediatrics are geared to the care of young children, but these attributes become dysfunctional and self-limiting for older adolescents and young adults. This is confirmed by our young adults' assertion that they *"grew out"* of pediatric care. As adolescents must become self-efficacious partners in their own care, self-reliance should be encouraged and parents' roles must be redefined.[5]

The experiences and recommendations we collected from Dutch young adults, parents and health care providers were quite similar to those reported in international studies. An example are the similarities between the (dis)advantages of pediatric and adult care reported in Table 3 and the findings of Wray & Maynard on specialist cardiac services in the UK.[36] There is also close agreement between the experiences and challenges in transition, and solutions for improving the process – as suggested by patients and parents in our study and in studies from the UK, the USA, Canada, and Australia – despite the large differences in health care systems. We think it unlikely that international parent-to-parent or youth-to-youth communication influenced or shaped our respondents' experiences. At least, it was never mentioned. Since less than 10% of interviewed parents or young adults were members of a patient organization, we do not feel that they were influenced by experiences of others, especially not from abroad.

Nevertheless, we encountered some differences in experiences cross-culturally. For example, our respondents never reported problems related to accessibility of health care services because adult-oriented medical care is fully covered by insurance in the Netherlands – in contrast to US-based studies such as Reiss *et al*[17] and Tuchman *et al.*[20] This demonstrates that system-related variables are indeed important in transition, although they did not raise barriers in the Netherlands.

Our results firmly support the key elements for transitional care,[13] as well as the need for action.[14] These Dutch professionals had little awareness of and designated attention to adolescent health issues. Most had only just begun to think about transition of care and did not use protocols or other interventions to smoothen the process.

With the findings of this study, it is now possible to direct pediatric and adult health care providers in the Netherlands toward better organization of multidimensional and multidisciplinary health care transition.

4.1   Limitations of the study

A limitation of this study is that in some chronic conditions, nonresponse and refusal rates were high, implying that there could be a selection bias. Young adults with mild health complaints may have been less inclined to participate, which may have resulted in an overrepresentation of more severe conditions and an overestimation of the reported difficulties during transfer. On the other hand, many patients with SCD and DM could not be reached. We do not know whether they have dropped out of care.

Furthermore, the small numbers of participants in each diagnostic group did not allow for detecting differences between the conditions. Also, possible benefits of a structured transition program could not be established, as only one department offered this at the time. This study was

conducted at one university hospital with short communication lines because the pediatric and adult services are located on the same premises. Most young adults were transferred within this hospital. However, they complained of the same lack of coordination and differences in care as those transferred to other hospitals.

5   Conclusion

Preventing adolescents becoming lost in the transfer between pediatric and adult health services is a major challenge in view of the cultural chasm between pediatric and adult-oriented services. Until recently, the specific needs of young adults and their parents during health care transition have been largely ignored in the Netherlands. The directions emerging from this study are clear: better preparation for transition, early start, and involvement of adolescents and parents. The focus should be on strengthening adolescents' independency and changing parents' roles. Health care providers' first priority is building bridges through enhanced communication between pediatric and adult-oriented care. Gaining trust and investing in new personal relations is the way forward for all parties involved: transition is about responding and bonding.

Key messages
- Moving on to adult care is a welcome and positive challenge for young adults with chronic conditions and their parents, despite inherent insecurities.
- Young adults and parents wish to be involved in the transition process and be better informed about the changes.
- The key to successful transition is strengthening independency and encouraging self-management right from childhood.
- Gaining trust in each other and building new personal relations is essential for successful transition, not only for patients and their parents, but also for pediatric and adult-oriented health care providers.
- From a lifespan care perspective, providing comprehensive transitional care is a necessity: so let's just do it.

Acknowledgements

References

1. Kirk S. Transitions in the lives of young people with complex health care needs. *Child Care Health Dev.* 2008;34(5):567-575.
2. Viner RM. Transition of care from paediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child.* 2008;93(2):160-163.
3. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, *et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. *J Adol Health*. 1993;14(7):570-576.
4. American Academy of Pediatrics, American Academy of Family Physicians & American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics.* 2002;110(6 Pt 2):1304-1306.
5. Rosen D. Between two worlds: bridging the cultures of child health and adult medicine. *J Adolesc Health*. 1995;17(1):10-16.
6. Kennedy A, Sawyer S. Transition from pediatric to adult services: are we getting it right? *Curr Opin Pediatr.* 2008;20(4):403-409.
7. Hilderson D, Westhovens R, Wouters C, Moons P. Transitional care for adolescents with rheumatic diseases: urgent need for more research. *Child Care Health Dev.* 2008;34(3):401-402.
8. McDonagh JE, Shaw KL, Southwood TR. Growing up and moving on in rheumatology: development and preliminary evaluation of a transitional care program for a multicentre cohort of adolescents with juvenile idiopathic arthritis. *J Child Health Care*. 2006;10(1):22-42.
9. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care program on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2007;46(1):161-168.
10. While A, Forbes A, Ullman R, Lewis S, Mathes L, Griffiths P. Good practices that address continuity during transition from child to adult care: synthesis of the evidence. *Child Care Health Dev.* 2004;30(5):439-452.
11. Binks JA, Barden WS, Burke TA, Young NL. What do we really know about the transition to adult-centered health care? A focus on cerebral palsy and spina bifida. *Arch Phys Med Rehabil.* 2007;88(8):1064-1073.
12. McDonagh JE. *Growing up ready for emerging adulthood. An evidence base for professionals involved in transitional care for young people with chronic illness and/or disabilities*. Paper read at Conference 26th March 2006. Department of Health Publications, London, UK; 2006. Available at: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4137427
13. McDonagh JE, Viner RM. Lost in transition? Between paediatric and adult services. *Brit Med J*. 2006;332(7539):435-436.
14. Christie D, Viner R. Chronic illness and transition: time for action. *Adolesc Med State Art Rev.* 2009;20(3):981-987,xi.
15. Stewart DA, Law MC, Rosenbaum P, Willms DG. A qualitative study of the transition to adulthood for youth with physical disabilities. *Phys Occup Ther Pediatr.* 2001;21(4):3-21.
16. Shaw KL, Southwood TR, McDonagh JE. User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2004;43(6):770-778.
17. Reiss JG, Gibson RW, Walker LR. Health care transition: youth, family, and provider perspectives. *Pediatrics.* 2005;115(1):112-120.
18. Stabile L, Rosser L, Porterfield KM, *et al.* Transfer versus transition: success in pediatric transplantation brings the welcome challenge of transition. *Prog Transplant.* 2005;15(4):363-370.
19. Östlie IL, Dale O, Moller A. From childhood to adult life with juvenile idiopathic arthritis (JIA): a pilot study. *Disabil Rehabil*. 2007;29(6):445-452.
20. Tuchman LK, Slap GB, Britto MT. Transition to adult care: experiences and expectations of adolescents with a chronic illness. *Child Care Health Dev.* 2008;34(5):557-563.
21. Anthony SJ, Kaufman M, Drabble A, Seifert-Hansen M, Dipchand AI, Martin K. Perceptions of transitional care needs and experiences in pediatric heart transplant recipients. *American Journal of Transplantation.* 2009;9(3):614-619.
22. Young NL, Barden WS, Mills WA, Burke TA, Law M, Boydell K. Transition to adult-oriented health care: perspectives of youth and adults with complex physical disabilities. *Phys Occup Ther Pediatr.* 2009;29(4):345-361.

23.   Clarizia NA, Chahal N, Manlhiot C, Kilburn J, Redington AN, McCrindle BW. Transition to adult health care for adolescents and young adults with congenital heart disease: perspectives of the patient, parent and health care provider. *Can J Cardiol.* 2009;25(9):e317-322.

24.   Valenzuela JM, Buchanan CL, Radcliffe J, Ambrose C, Hawkins LA, Tanney M, *et al.* Transition to Adult Services among Behaviorally Infected Adolescents with HIV--A Qualitative Study. *J Pediatr Psychol*. 2011;36(2):134-140.

25.   Moons P, Pinxten S, Dedroog D, Van Deyk K, Gewillig M, Hilderson D, *et al.* Expectations and experiences of adolescents with congenital heart disease on being transferred from pediatric cardiology to an adult congenital heart disease program. *J Adolesc Health.* 2009;44(4):316-322.

26.   van Staa AL, Eysink Smeets-van de Burgt AE, van der Stege HA, Hilberink SR. Transitie in zorg van jongeren met chronische aandoeningen in Nederland nog onder de maat. [Transition in care for young people with chronic conditions: substandard in the Netherlands]. *Tijdschr Kindergeneeskd.* 2010;78(6):227-236.

27.   Verhoeven L, van Staa AL. ''Vanzelfsprekend, maar niet vanzelf''. Transitie in zorg: ervaringen van adolescenten met Cystic Fibrosis en hun ouders [Transition of care: a matter of course, but no plain sailing. Experiences of adolescents with Cystic Fibrosis and their parents]. *Verpleegkunde.* 2006;3:186-195.

28.   Hilberink SR, Roebroeck ME, Nieuwstraten W, Jalink L, Verheijden JM, Stam HJ. Health issues in young adults with cerebral palsy: towards a life-span perspective. *J Rehabil Med.* 2007;39(8):605-611.

29.   Geerts E, van de Wiel H, Tamminga R. A pilot study on the effects of the transition of paediatric to adult health care in patients with haemophilia and in their parents: patient and parent worries, parental illness-related distress and health-related Quality of Life. *Haemophilia.* 2008;14(5):1007-1013.

30.   Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res in Psychol.* 2006;3(2):77-101.

31.   van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-8017-769-7.

32.   Shaw KL, Southwood TR, McDonagh JE. Transitional care for adolescents with juvenile idiopathic arthritis: a Delphi study. *Rheumatology (Oxford).* 2004;43(8):1000-1006.

33.   Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet.* 2007;369(9571):1481-1489.

34.   van Staa AL, van der Stege HA, Jedeloo S, Moll HA, Hilberink S. Readiness to transfer to adult care of adolescents with chronic conditions: exploration of associated factors. *J Adolesc Health.* 2011;48(3):295–302.

35.   Kralik D, Visentin K, van Loon A. Transition: a literature review. *J Adv Nurs.* 2006;55(3):320-329.

36.   Wray J, Maynard L. Specialist cardiac services: what do young people want? *Cardiol Young.* 2008;18(6):569-574.

37.   Roebroeck ME, Jahnsen R, Carona C, Kent RM, Chamberlain MA. Adult outcomes and lifespan issues for people with childhood-onset physical disability. *Dev Med Child Neurol.* 2009;51(8):670-678.

38.   van Staa A, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: the added value of mixed methods research. *Patient Educ Couns.* 2011;82(3):455–464.

39.   Reid GJ, Irvine MJ, McCrindle BW, Sananes R, Ritvo PG, Siu SC, *et al.* Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics*. 2004;113(3 Pt 1):e197-205.

Transition of care for young people with chronic conditions: substandard in the Netherlands

AnneLoes van Staa, Annelies E. Eysink Smeets-van de Burgt, Heleen A. van der Stege, Sander R. Hilberink

Abstract

OBJECTIVE: Transition of care is a big challenge for pediatrics. In the Netherlands, not much is known about preparation for transition, transfer policies and collaboration with adult care. Objective of this research was to gather insight into current transitional practices and into the felt necessity for improvement of adolescent care.

METHODS: Cross-sectional, descriptive research in a convenience sample of health care professionals involved with adolescents (12-25 years old) with chronic somatic conditions. 115 professionals (48% pediatricians) from (pediatric) hospitals filled out the web-questionnaire and 8 additional telephone interviews were conducted.

RESULTS: Apart from medical issues, care professionals saw the importance of addressing the social position of these adolescents during consultations. Preparation for transition of care and the promotion of more independence of young people got the attention of most of the teams, but more structural interventions were rare. Essential elements of a transition program such as transition protocols, individual action plans, and the use of a transition coordinator or transition clinic were only present in a minority of the teams. Over 85% of the respondents gave high priority to improving self-management, adherence and self-reliance in their population.

CONCLUSIONS: There is a discrepancy between the priorities of health care professionals and the way in which transition of care is being practiced at the moment in the Netherlands. Promotion of self-management and social participation are important areas for improvement. The growing number of young people with chronic conditions necessitates better organization of transition of care and more attention toward self-management.

1   Introduction

A growing number of children in the Netherlands are living with a (serious) chronic condition. The most recent estimation was 500.000 children, ie, 14% of the total number.[1] More than 90% of these children will reach adult age, due to better medical care.[2-4] Therefore, young people with chronic conditions as well as their parents need to prepare for transfer to specific care services for adults. This process of preparing, transfer and follow-up is referred to as *transition of care*. The pediatricians Donckerwolcke & Van Zeben[3] were one of the first in the Netherlands to draw attention to this issue. On the basis of a survey among departmental heads in the university children's hospitals they concluded that transfer often proceeds *ad hoc*, that arrangements with the adult care services usually are not protocolized, and that management typically does not include a structured transition process. Hardly any empirical research into the transition process has been performed in the Netherlands since then,[5-7] although several articles of an opiniating nature have been published, either on general pediatric care[8] or specific chronic conditions.[4, 9-12]

Why is special attention to (the medical management of) young people with chronic conditions so important? Apart from their medical needs, these young people are particularly disadvantaged in becoming independent and fulfilling adult roles in society.[13] As documented in Dutch research, they lag behind age peers in becoming independent and in social participation.[14-16] Nevertheless, the care for these young adults is usually restricted to medical and functional aspects. Although more and more studies on quality of life of Dutch young people with chronic conditions are being published, social consequences of chronic conditions are little researched.[17] Consequently, the views of these young people themselves have remained obscure, which was the rationale for setting up the research program *Op Eigen Benen* (On Your Own Feet), conducted in the Erasmus Medical Center - Sophia Children's Hospital.[18, 19] The program sought to uncover how young people with different chronic conditions handle the task of combining daily management of the condition with leading a life as ordinary and pleasant as possible.

They are facing big challenges: apart from actively and independently applying life style rules and treatment regimens (self-management),[13, 20] they also need to maintain quality of life and to tackle barriers in social functioning.[17] These young people may well be inclined to turn their backs to the entire health care circuit, with the risk of dropping out from health care and developing complications.[21] Problems with acquiring independence, therapy adherence and risk behavior are frequent in adolescence and require a specific treatment strategy. The health care system, however, is not well geared to the age-specific needs,[19, 22-25] or to the challenge to make young people partners in care.[18, 19, 26]

Most of these young people will transition from the pediatric to the adult care system at the age of 18 years, which often is a momentous event for them and their parents.[7] Having fewer contacts with their familiar providers, however, and the lack of an age-specific approach contribute to worse clinical outcomes and possible drop-out from health care.[21] In addition, specialists in the adult care services may be unfamiliar with congenital conditions and with the long-term consequences of chronic conditions developed in childhood,[4, 27] and multidisciplinary treatment teams may be lacking in the adult setting.[11, 28]

In the Dutch pediatric setting, hardly any research has explored the extent of *loss-to-follow-up*, the advancement of self-management, or the effects of bad therapy adherence in puberty. Still, there are indications that the transfer to adult care often is organized in a less-than-optimal way.[3, 7, 9] In rehabilitation care, it appeared that half of the young adults with cerebral paresis had not consulted with a rehabilitation physician in the past year, even though they had care needs.[22, 23, 29]

Achieving a safe and effective transition is therefore a core indicator of quality of adolescent care and one of the major challenges in pediatrics.[26, 30, 31] An effective transition is more than just organizing a 'smooth transfer': the point is that the young person should become partner in his/her care and be enabled to effectively 'manage' his/her condition and life.[26] Even though empirical studies of the effectiveness of transition programs are still scarce,[32] there is a growing evidence base for beneficial interventions.[21] The need for special guidelines and standards with regard to the support of young people with chronic conditions is widely recognized internationally; and, increasingly, professional organizations of pediatricians incorporate these in their policies.[26, 31, 33, 34] In the Netherlands, however, such a policy does not exist.

A first step in the direction of policy-making in the Netherlands is to map the current transition care experiences and to identify interventions applied by health care professionals. Therefore, a web-based survey was conducted among pediatric providers aimed at gaining insight into (1) the state of affairs regarding transition of care within Dutch pediatrics, and (2) the perceived need of improving the current care to adolescents.

2   Methods

This is a cross-sectional, descriptive study in the shape of a web-based survey among professionals involved in the care for young people with chronic somatic conditions (12 - 25 years old). Because there is no register of pediatricians or other providers who (mainly) dedicate themselves to chronic care, a convenience sample was used.

2.1   Recruitment

Recruitment for participation in the study was stepwise. Early 2008, a personal invitation by email with a link to the survey was sent to some 300 pediatricians, physicians, nurses, paramedics and other providers who treat young people with chronic somatic conditions. First, candidate participants were identified from the relations database of the Rotterdam University Expertise Center Transitions of Care, which included many providers from the target group of this survey. Next, other pediatricians and nurses were identified from the websites of all 92 Dutch hospitals with a children's department. Their e-mail addresses were obtained from *the Geneeskundige Adresgids* (2007) or telephone inquiries. Furthermore, the heads of the children's (outpatient) departments of all Dutch hospitals were sent a request to complete the survey completed. In addition, a call was published in various newsletters and on websites of professional organizations such as the Dutch Pediatrics Association (NVK) and the Association of Pediatric Nurses (VVKV). Per treatment team, one person was invited to participate, and instructed to describe the views of and about the

treatment team in question. To increase the response several iPod Shuffles and cakes were raffled among the participants. A reminder was sent by e-mail to those who after two working weeks had not yet responded.

Moreover, we performed a document study of existing transition protocols. Semistructured interviews by telephone were held with eight respondents who had stated their teams were running a transition clinic or used a transition protocol.

2.2   Measurement instrument and analysis

A questionnaire was composed on the basis of literature research and international examples,[35-37] and was tested with a nurse who is a transition coordinator and with three pediatric rehabilitation physicians, who were not again invited to participate. The final questionnaire was made available on the website www.opeigenbenen.NU. The questionnaire encompassed an inventory of:

• Present procedure around transition of care
• Use of interventions promoting self-management
• Existing services and supply for young people
• Current practices during consultation hours
• Importance of themes in consultation
• Perceived necessity of improvement projects

We included current practices of consultation because the literature shows that not only the young person's self-management of daily care, but also the relationship with the health care provider is an important requirement for successful transition.[21] Responses to all questions on opinions (eg, the priorities for improvement of one's own care practice) were made on 5-point Likert scales (1 = not at all important / necessary; 5 = very important / necessary). Descriptive statistics were used for the analysis. Differences between respondents (pediatrician or not) and between university and general hospitals were tested with the Chi^2 test.

3   Results

3.1   Response

In total, 115 persons in (children's) hospitals have completed the web-based questionnaire, next to 49 rehabilitation providers, about whom we reported elsewhere.[38] These 115 persons represented 115 different treatment teams. Table 1 shows characteristics of the respondents. Almost half (47.8%) were pediatricians. Some 49.6% worked in general hospitals; 47.8% work in university hospitals. Of all university medical centers, at least one team responded; of the 92 general hospitals with a children's department 30% participated, typically the larger regional hospitals. All teams were multidisciplinary in nature, with the pediatrician as the 'spider in the web'.

The largest patient category covered is that of general pediatrics (64 teams); followed by 24 diabetes teams, 15 pulmonology teams (treating patients with asthma and Cystic Fibrosis), 7 pediatric surgery teams, and 5 teams treating young people with Inflammatory Bowel Diseases.

Table 1   *Respondent characteristics (n = 115)*

|	*n*	*%*
Type of the institution		
University hospital	55	47.8
General hospital	57	49.6
Other	3	2.6
Education / profession		
Pediatrician	55	47.8
Specialist working in adult care	5	4.3
Nurse (specialist)	48	41.7
Psychologist	2	1.7
Paramedical professional	4	3.5
Other	1	0.9
Patient group		
General pediatrics	64	55.7
Diabetes type 1	24	20.9
Pulmonology	15	13.0
Pediatric surgery	7	6.1
Inflammatory Bowel Disease	5	4.3

3.2   Present procedures

3.2.1   Transfer to adult care

Yearly, 50.5% of the teams transferred 1 to 10 patients to adult care; 24.3% transferred 11 to 50 young people and 6.9% even more than 50. Half of the teams (49.6%) transferred young people at a fixed moment, ie, when they reached the age of 18 years or when they leave secondary education. In 46.1% of cases there was no fixed moment of transfer; the moment was rather determined in consultation with the young people and their parents or with providers of the adult services.

Figure 1 lists the interventions that treatment teams were using to prepare young people and parents for transition. Discussing the process during consultations was the most reported intervention (83.5%). Usually medical transferral notes were prepared. The *timing* of transfer and information on *the person* to whom the care is transferred, however, were not always discussed

Figure 1 *Percentages of teams that perform this activity to prepare adolescents for the transfer to adult care or for self-management (n = 115)*
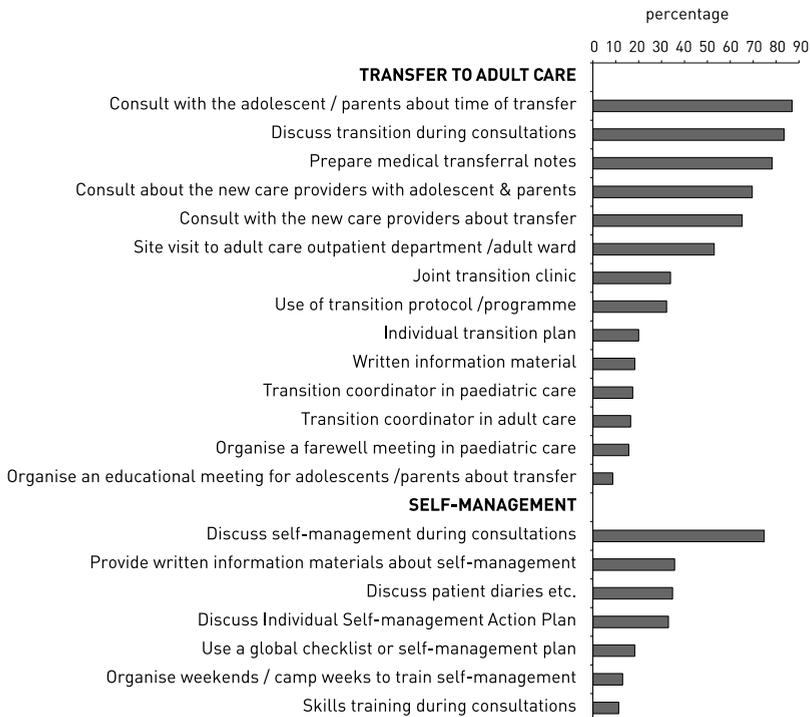
with parents and the young person: in university hospitals this happened less often than in general hospitals (*P* < .05). The new care providers were usually consulted, and half of the teams arranged a visit to the new ward prior to the actual transfer. This happened more often in the university hospitals (*P* < .05). Specific interventions to systematically prepare the young people were still less common: a joint transition clinic (33.9%), transition protocol (32.2%), use of a checklist or individual transition plan (20.0%), transition coordinator in pediatric- and / or adult care (17.4% and 16.5%, respectively). Transition programs, transition coordinators and transition clinics were more common in university hospitals than in general hospitals (*P*'s < .05). Teams that transferred more young people more often run a transition clinic (*P* < .05).

A minority of the teams started preparing the future transfer at an early moment: 37.5% of the teams did so before the young person's 16th birthday, 35.6% around age 16 years (mean 15.2 years; *SD* = 1.6). In a considerable proportion of cases, issues surrounding transition were not discussed until a later age (Figure 2).

Figure 2 *Current age versus ideal age at which transfer to adult care should be first discussed (n = 104 / 111 respectively)*
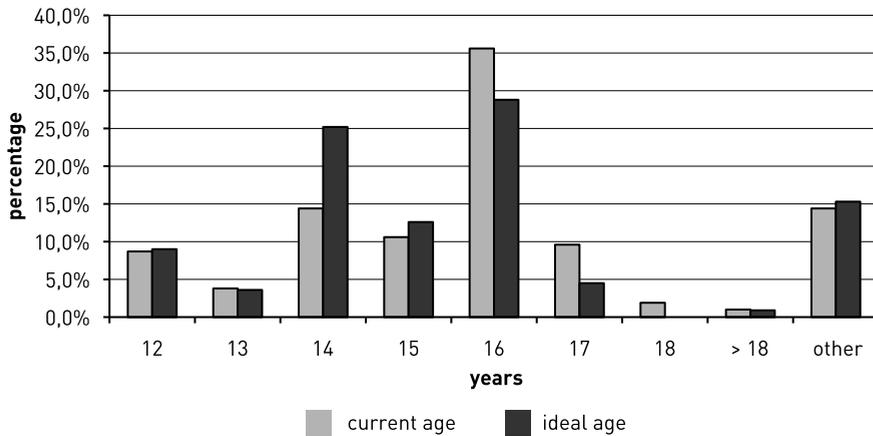

3.2.2   Self-management

Targeted interventions aimed at providing more independence and responsibility in the daily self-care were not common (Figure 1). Three quarters of the treatment teams (74.8%) provided only oral information during consultations; 35.7% also used printed material, one third of the teams encouraged the young person to keep diaries or registrations at home, or discussed individual self-management action plans. Global action plans, skills training during consultations, or peer activities were less often used. In this respect there were no differences between university hospitals and general hospitals, except that the general hospitals made more use of diaries ($P < .05$).

Providers considered it essential to start promoting the young person's independence at an early age. More than half of the teams started discussing self-management with the young person and parents even before the child's 12th birthday (Figure 3).

Figure 3 *Current age versus ideal age at which self-management should be discussed (n = 107 / 111 respectively)*
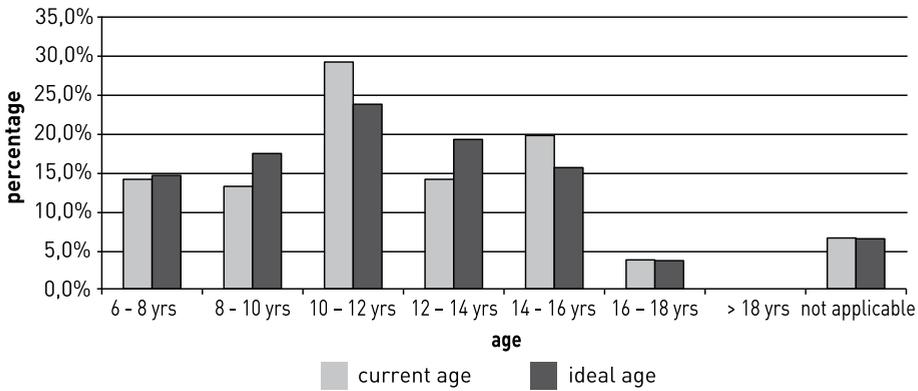
3.2.3   Medical consultations

Of all respondents, 41.7% reported that parents as a rule are present during *all* consultations. Still, physicians or nurses occasionally would see the young people on their own, without parents present, mostly when the young people were between 14 to 16 years old (38.3%); but 27.0% would not see the young people without parents present until the age of 16 to 18 years.

Almost all respondents attached (great) importance to discussing the young people's views on coping with the disease and themes such as disclosure of the condition (Table 2). Apart from discussing the young person's independence and therapy adherence, over 90% of respondents also attached (great) importance to participation issues such as school, employment, leisure time activities, going out and the home situation; followed by transition to adult care, career planning, and risk behavior (alcohol and drugs). At the bottom of the list we find issues such as independent living, social benefits and health insurances, but still over half of respondents considered these matters important. Several differences between respondents from general hospitals and those from university hospitals were seen: themes such as physical exercise, diet, going out, alcohol, drugs and disclosure about the condition were considered more important in general hospitals than in university hospitals, while respondents from university hospitals ranked therapy adherence as even more important (*P*'s < .05).

Table 2 *Important themes for hospital consultations with adolescents; ranked in order of the mean scores[a] (n = 115)*

Themes	mean (SD)	% (very) important
Feelings / coping with the disease, depression	4.6 (0.5)	99.1
Disclosure	4.6 (0.5)	98.2
Independence / self-management	4.6 (0.5)	97.3
Adherence	4.6 (0.6)	92.9
Physical exercise / Sports	4.5 (0.6)	97.3
Education	4.4 (0.5)	97.3
Home situation	4.4 (0.6)	93.8
Going out / activities with peers	4.4 (0.7)	92.9
Vocational issues / (side) job	4.3 (0.6)	92.9
Diet / nutrition	4.3 (0.7)	87.5
Transition / transfer to adult care	4.3 (0.7)	86.5
Career planning	4.1 (0.7)	82.7
Alcohol	4.1 (0.8)	80.2
Sexuality / intimate relations	4.0 (0.8)	79.3
Balance between activity and rest	4.0 (0.8)	73.0
Drugs	4.0 (0.9)	75.9
Life expectancy / prognosis	3.9 (0.8)	70.5
Safe sex / contraception	3.9 (1.0)	67.6
Independent living	3.7 (0.8)	59.8
Social benefits / health insurance	3.6 (0.8)	50.5

[a] *Likert scale: 1 = not important at all; 2 = not important; 3 = neutral; 4 = important; 5 = very important.*

3.3 Priorities for improvement

The questionnaire included twelve suggestions for improvement, to be ranked according to the necessity of being taken up, considering the treatment team's own situation. Table 3 presents the priorities for improvement ranked by the means of the response categories (1 - 5 scale). The necessity of improving the care for adolescents with chronic conditions was strongly felt. The highest priority was assigned to interventions to encourage self-management and therapy adherence: more than 90% of respondents judged improvement of self-management, of therapy adherence and of self-sufficiency (social participation) of the young people (quite) necessary for the own team. Also the development of a comprehensive transition program was highly desired. The teams from the university hospitals and those from general hospitals did not differ with regard to priorities for improvement, and neither did pediatricians and the other respondents.

Table 3 *Interventions considered necessary for the respondents' own team of health care providers; ranked in order of mean scores[a] (n=115)*

Priorities for improvement	mean (SD)	% (very) necessary
Interventions to enhance adherence	4.5 (0.7)	93.6
Interventions to encourage self-management	4.4 (0.7)	94.7
Interventions to enhance social participation	4.4 (0.7)	91.2
Transition program	4.3 (0.8)	84.8
Structural collaboration between pediatric and adult care	4.3 (0.8)	87.5
Screening for psychosocial problems	4.3 (0.8)	89.2
Interventions to address psychosocial problems	4.3 (0.8)	86.5
Screening for lack of adherence	4.3 (0.8)	88.2
Staff training in adolescent health issues	4.0 (0.9)	79.6
Interventions to encourage contact with fellow patients	3.9 (0.9)	73.5
Change in style of consultations	3.9 (0.9)	72.6
Adolescent-friendly facilities	3.8 (1.0)	64.3

[a] *Likert scale: 1 = not necessary at all; 2 = not necessary; 3 = neutral; 4 = necessary; 5 = very necessary.*

Providers were of the opinion that the future transition ideally should be discussed at an earlier moment than is the case now. The mean ideal age to start preparations was 14.8 years (*SD* = 1.5), which differed significantly from the age at which currently transition is now first discussed (mean 15.2 years; *SD* = 1.6) (t = 2.43; df = 74, *P* < .05; Figure 2). Regarding the encouragement of self-management and independence, the difference between current age and ideal age was not significant (Figure 3).

There was room for improvement as well in stimulating these young people's independent behavior in the consultation room. Half of the respondents (50.5%) stated that the first consultation with the young person alone should take place when they are between 14 to 16 years of age; 15.6% stated this could be even before their 14th birthday, whereas 31.2% preferred to start at an age between 16 to 18 years. Hardly anyone (0.9%) saw this as undesirable or as something that should be done after actual transfer.

3.4   Additional data collection

The 57 teams who stated they used a transition protocol or were running a transition clinic were invited by email to send documentation; however, only a few responded to this request. Eight providers were interviewed by telephone. These interviews revealed that protocols often have not yet been documented or that the transition clinic is still under development. Two teams had prepared a disease-specific transition protocol. Most of the teams had little contact with other providers in their own institutions and there was hardly any exchange of best practices.

4   Discussion

This survey mapped current practices of transition, encouraging self-management and self-reliance, upheld by 115 treatment teams in the Netherlands who are treating young people with chronic conditions. Practices mainly consisted of discussing the upcoming transition during consultations and discussing the right moment of transfer with these young people and their parents. No more than one in five teams stated they pursued (some) structured activities recommended in the literature, such as adhering to a transition protocol, using individual plans of action, appointing a transition coordinator, or setting up a transition clinic.[21, 34] It is not clear, however, if these activities in fact were taking place or were still under development. From the interviews by telephone it appeared that teams often had progressed less far than stated in response to the questionnaire.

According to the literature, the essential elements of a transition program are: a joint policy on transition, coordination, continuity of care, individual transition plans, extensive information for the young people and their parents, and an early start of preparation.[21] To date, only very few publications have reported on such programs in place in Dutch health care.[39, 40] From the questionnaire, it appeared that hardly any of such comprehensive transition programs were in place in the Netherlands. The same held true for self-management: systematic attention to promoting independence was still limited. Transition interventions were more frequent in university hospitals than in general hospitals, but, on the other hand, fewer teams in the academic setting discussed moment and place of transition with the young persons and their parents. There were no differences between both settings regarding current practices to promote the young persons' self-management.

It appeared that the providers largely agreed on the necessity of improving their care of adolescents with chronic conditions. The list of priorities for improvement was based on the desired content of a comprehensive transition program.[21] Over 95% stated it is (highly) necessary to stimulate self-management and self-reliance of these young people even more. In addition, ideally such themes should be paid attention to at a much younger age than currently is the case; and the process of transfer to adult care should be better organized. It cannot be excluded that respondents were mainly the health care providers who already were convinced of the necessity of improvement. On the other hand, the unanimity may also indicate a widely felt necessity among pediatricians and pediatric nurses to improve current practices.

The results were unanimous, too, with regard to themes considered important in consultations with young people with chronic conditions: acceptance of the disease, independence, and therapy adherence, but also nonmedical issues such as physical exercise, education, employment and going out. In order to stimulate these young people to do the talking themselves and to be able to address more personal, sensitive subjects, consultations should be more regularly held with the young person alone, without the parents. This is also an important element in preparing them for what is customary in adult care.[21, 23] Parents now usually accompanied their children, which is not conducive to promoting these young people's independent behavior.[41] Over 70% of the respondents indeed were convinced of the necessity to change consultation practices. Nevertheless, in practice pediatricians report having difficulty in changing the parents' role in the consultation room.[42]

10

A limitation of this study is that it was not clear whether the respondents form a good representation of all treatment teams for young people with chronic conditions in somatic care and whether they indeed were voicing the opinion of their own treatment team. Bias may have occurred because this was a convenience sample (in part drawn from the On Your Own Feet relations date base); it is well possible that respondents were mainly those with an interest in the issue of transition and the ones who already had been implementing interventions or planning to do so in the near future.

It is not possible to pronounce on representativity of the sample. However, there was a good spread of respondents from different hospitals all over the Netherlands and from different sub-specialties. As inquiry by telephone revealed that in practice many interventions had far less progressed than was stated in the questionnaire, this survey perhaps presents a too rosy picture of the state of affairs in the Netherlands. Alas, mere good intentions do not suffice to achieve improvements in adolescent care.

Still, the broad backing of all suggested priorities for improvement shows, anyhow, acceptance among the Dutch pediatricians of innovations aimed both at achieving better self-management and social participation of young people with chronic conditions and at improving organizational aspects of the transition from pediatric care to adult care. These are the two goals of the Quality Improvement Collaborative *On Your Own Feet Ahead!,*[43] which since 2008 is being run by the Rotterdam University (Expertise Center Transitions of Care) and the Dutch Institute for Health care Improvement CBO.* This program includes the testing of interventions and the dissemination of best practices among hospitals and rehabilitation centres. Experts, such as the UMC Utrecht Multidisciplinary teams from pediatric- and adult care themselves are formulating aims to achieve content and process improvements. They do so using the Breakthrough method,[44] in consultation with patients and under the supervision of advisers. The program also provides for scientific evaluation research.

5   Conclusion

Since the most recent survey of the state of affairs with regard to transition and transfer in Dutch somatic pediatric care,[3] we have seen growing attention to this subject and raised awareness of its importance. Ever more initiatives are being reported that aim to structure the care of young people with chronic conditions in a better way – also outside the university medical centers. At the same time, we are only just at the beginning in the Netherlands, because many treatment teams are not yet running a comprehensive transition program; and mission statements and policy-making in the professional organizations are still largely lacking. The increase in the number of young people with chronic conditions – who are the adults of the future – also in the Netherlands signals that transition of care should be better organized and more attention should be paid to promoting self-management and social participation.

Acknowledgements

References

1. Mokkink LB, van der Lee JH, Grootenhuis MA, Offringa M, van Praag BMS, Heymans HSA. Omvang en gevolgen van chronische aandoeningen bij kinderen. [Extent and consequences of chronic conditions in children]. *Tijdschr Kindergeneeskd*. 2007;75(4):138-142.

2. Scal P. Transition for youth with chronic conditions: primary care physicians' approaches. *Pediatrics*. 2002;110(6 Pt 2):1315-1321.

3. Donckerwolcke RAMG, van Zeben-van der Aa DMCB. Overdracht van de zorg voor adolescenten met chronische ziekten: van kindergeneeskunde naar specialismen voor volwassenen. [Transfer of care for adolescents with chronic illnesses: from pediatrics to specialists for adults]. *Ned Tijdschr Geneeskd*. 2002;146(14):675-678.

4. Meijboom FJ, Roos-Hesselink JW, Bogers AJJC. Aangeboren hartafwijkingen op volwassen leeftijd. [Congenital heart disorders in adulthood]. *Tijdschr Kindergeneeskd*. 2006;74(Dec):236-241.

5. Geerts E, van de Wiel H, Tamminga R. A pilot study on the effects of the transition of pediatric to adult health care in patients with haemophilia and in their parents: patient and parent worries, parental illness-related distress and health-related Quality of Life. *Haemophilia*. 2008;14(5):1007-1013.

6. Verhoeven-Wind L, van Staa AL. ''Vanzelfsprekend, maar niet vanzelf''. Transitie in zorg: ervaringen van adolescenten met Cystic Fibrosis en hun ouders. [Transition of care: a matter of course, but no plain sailing. Experiences of adolescents with Cystic Fibrosis and their parents]. *Verpleegkunde*. 2006;3:186-195.

7. van Staa AL, Jedeloo S, van Meeteren J, Latour JM. Crossing the transition chasm: Experiences and recommendations for improving transitional care of young adults, parents and providers. *Child Care Health Dev*. 2011;37(6):821-832.

8. Maassen H. Een soepele overgang. Van kinderarts naar 'gewone' dokter. [A smooth transition. From pediatrician to 'ordinary' doctor]. *Med Contact*. 2005;60(17):700-703.

9. Escher JC, van der Woude CJ. Transitie van de adolescent met een chronische inflammatoire darmziekte. [Transition of the adolescent with a chronic inflammatory bowel disease]. *Tijdschr Kindergeneeskd*. 2005;73(Feb):36-41. // Escher JC. Transition from pediatric to adult health care in inflammatory bowel disease. *Dig Dis*. 2009;27(3):382-386.

10. van der Ent CK, Drubbel I, de Vrankrijker AMM, Arets HGM, Heijerman HGM. Cystic fibrosis; een vergrijzend ziektebeeld? [Cystic fibrosis: an ageing disorder?]. *Tijdschr Kindergeneeskd*. 2006;74(Dec):242-248.

11. de Muinck Keizer-Schrama SMPF. Het belang van zorgvuldige transitie van zorg voor patienten met het Syndroom van Turner. [The importance of a careful transition of care for patients with Turner's Syndrome]. *Ned Tijdschr Geneeskd*. 2007;151:1612-1615.

12. Schrander-Stumpel CT, Sinnema M, van der Hout L, Maaskant MA, van Schrojenstein Lantman-de Valk HM, Wagemans A, *et al*. Health care transition in persons with intellectual disabilities: general issues, the Maastricht model, and Prader-Willi syndrome. *Am J Med Genet C Semin Med Genet*. 2007;145c(3):241-247.

13. Sawyer SM, Drew S, Yeo MS. Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet*. 2007;369(9571):1481-1489.

14. van Mechelen MC, Verhoef M, van Asbeck FW, Post MW. Work participation among young adults with spina bifida in the Netherlands. *Dev Med Child Neurol*. 2008;50(10):772-777.

15. Stam H, Hartman EE, Deurloo JA, Groothoff J, Grootenhuis MA. Young adult patients with a history of pediatric disease: impact on course of life and transition into adulthood. *J Adolesc Health*. 2006;39(1):4-13.

16. Calsbeek H, Rijken M, Bekkers MJ, Kerssens JJ, Dekker J, van Berge Henegouwen GP. Social position of adolescents with chronic digestive disorders. *Eur J Gastroenterol Hepatol*. 2002;14(5):543-549.

17. Mokkink LB, van der Lee JH, Offringra M, van Praag BMS, Heymans HS, Grootenhuis MA. Sociaal-maatschappelijke gevolgen en kwaliteit van leven in Nederland bij kinderen en jongvolwassenen opgegroeid met een chronische aandoening. [Social consequences and quality of life in Dutch children and young adults with a childhood-onset chronic condition]. *Tijdschr Kindergeneeskd*. 2007;75(6):229-235.

18. van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-8017-769-7.

19. van Staa AL, van der Stege HA, Jedeloo S. *Op Eigen Benen Verder. Jongeren met chronische aandoeningen op weg naar zelfstandigheid in de zorg. [On Your Own Feet Ahead. Young people with chronic conditions on their way to independence in health care]*. Rotterdam: Hogeschool Rotterdam; 2008. ISBN 978-90-7905-902-7.

20. Colland VT. Zelfmanagement en therapietrouw: menage à deux van dokter en patiënt. Self-management and adherence: 'menage à deux' of doctor and patient]. *Tijdschr Kindergeneeskd*. 2008;76(4):205-213.

21. McDonagh JE. *Growing up ready for emerging adulthood. An evidence base for professionals involved in transitional care for young people with chronic illness and/or disabilities*. Paper read at Conference 26th March 2006. Department of Health Publications, London, UK; 2006. Available at: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4137427

22. Nieuwenhuijsen C, van der Laar Y, Donkervoort M, Nieuwstraten W, Roebroeck ME, Stam HJ. Unmet needs and health care utilization in young adults with cerebral palsy. *Disabil Rehabil*. 2008;30(17):1254-1262.

23. Roebroeck ME, van Meeteren J, Wiegerink D, Nieuwenhuijsen C, Donkervoort M, Spijkerman D, *et al*. Lost in Transition? Ontwikkeling en implementatie van Polikliniek Jongvolwassenen. [Lost in Transition? Development and implementation of the Young Adult Clinic]. *Revalidata*. 2006;131(Jun):35.

24. Burger CA, Warmenhoven NJC, Rol M, Roebroeck ME. Transitie naar volwassenheid. De kinderrevalidatie, en dan? [Transition to adulthood. What comes after pediatric rehabilitation?]. *Revalidata*. 2005;126:12-18.

25. Nieuwenhuijsen C, Donkervoort M, Nieuwstraten W, Stam HJ, Roebroeck ME. Experienced problems of young adults with cerebral palsy: targets for rehabilitation care. *Arch Phys Med Rehabil*. 2009;90(11):1891-1897.

26. Viner RM. Transition of care from pediatric to adult services: one part of improved health services for adolescents. *Arch Dis Child*. 2008;93(2):160-163.

27. Postma A, Schouten-van Meeteren AYN, Hakvoort-Cammel FGAJ, Bresters D, Versluys AB, Bökkerink JPM, *et al.* Gezondheidsproblemen na de behandeling van kinderkanker. [Health problems after the treatment for cancer in childhood]. *Tijdschr Kindergeneeskd*. 2006;74(Dec):209-214.

28. Prevo A, Post M, van Asbeck FW, Barf HA, Gooskens RHMJ, Jennekes-Schinkel A, *et al.* Jongvolwassenen met spina bifida. [Young adults with spina bifida]. *Med Contact*. 2004;59(19):789-791.

29. Hilberink SR, Roebroeck ME, Nieuwstraten W, Jalink L, Verheijden JM, Stam HJ. Health issues in young adults with cerebral palsy: towards a life-span perspective. *J Rehabil Med*. 2007;39(8):605-611.

30. Olsen DG, Swigonski NL. Transition to adulthood: the important role of the pediatrician. *Pediatrics*. 2004;113(3 Pt 1):e159-162.

31. American Academy of Pediatrics; American Academy of Family Physicians; American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics*. 2002;110(6 Pt 2):1304-1306.

32. Shaw KL, Southwood TR, McDonagh JE. Young people's satisfaction of transitional care in adolescent rheumatology in the UK. *Child Care Health Dev*. 2007;33(4):368-379.

33. American Academy of Pediatrics Committee on Children with Disabilities. The role of the pediatrician in transitioning children and adolescents with developmental disabilities and chronic illnesses from school to work or college. *Pediatrics*. 2000;106(4):854-856.

34. Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM. Transition to adult health care for adolescents and young adults with chronic conditions: position paper of the Society for Adolescent Medicine. *J Adolesc Health*. 2003;33(4):309-311.

35. McDonagh JE, Shaw KL, Southwood TR. Growing up and moving on in rheumatology: development and preliminary evaluation of a transitional care program for a multicentre cohort of adolescents with juvenile idiopathic arthritis. *J Child Health Care*. 2006;10(1):22-42.

36. Betz CL. Transition of adolescents with special health care needs: review and analysis of the literature. *Issues Compr Pediatr Nurs*. 2004;27(3):179-241.

37. Shaw KL, Southwood TR, McDonagh JE. Developing a programme of transitional care for adolescents with juvenile idiopathic arthritis: results of a postal survey. *Rheumatology (Oxford)*. 2004;43(2):211-219.

38. van Staa AL, Hilberink SR, Eysink Smeets-van de Burgt A, van der Stege HA, Jedeloo S, Roebroeck ME. Transitie van kinderzorg naar volwassenenzorg: revalidatie in actie. [Transition of care to adult care in

10

the Netherlands: results from a survey among 159 healthcare providers in hospitals and rehabilitation centers]. *Revalidata.* 2008;30(146):3-10.

39. Ammerlaan JW, van Pelt PA, Masselink IM, Prakken ABJ, Kruize AA. De jongvolwassene met juveniele idiopathische artritis (JIA): Transitie binnen het UMC Utrecht van WKZ naar AZU. [The young adult with JIA: Transition within the UMC Utrecht from WKZ to AZU]. *Ned Tijdschr Reumatol.* 2007;(3):7-12.

40. Scholten M, Kooijman R. Jongeren met hemofilie in stappen over de brug. [Young people with haemophilia are crossing the bridge]. *Kind en Ziekenhuis.* 2006;29(1):11-13.

41. van Staa AL, On Your Own Feet Research Group. Unraveling triadic communication in hospital consultations with adolescents with chronic conditions: The added value of mixed methods research. *Patient Educ Couns.* 2011;82(3):455–464.

42. van Staa AL, Kuijper M, Jedeloo S, Latour JM. Wat doen we met de ouders? [What to do with the parents?]. *Kind en Ziekenhuis.* 2007;30(2):49-51.

43. Jedeloo S, Janssen I, van Staa AL. Doorbraak in transitiezorg: Actieprogramma op Eigen Benen Vooruit! Van onderzoek naar zorgvernieuwing voor jongeren met chronische aandoeningen. [Breakthrough in transitional care for young people with chronic conditions: From research to action in the innovation program On Your Own Feet Ahead!]. *Kwaliteit in Zorg.* 2009(2):10-13.

44. Schouten L, Minkman M, de Moel J, van Everdingen J. *Doorbreken met resultaten in de gezondheidszorg. [Breakthrough with results in health care].* Assen: Van Gorcum; 2007.

Conclusion

general discussion and
future directions

Introduction

Adolescents' own perspectives on health care in their transition to adulthood and adult care were the central focus of our studies. This final chapter highlights the main findings of the studies by addressing the four major research questions described in the Introduction.

I) What are the preferences of adolescents with chronic conditions for (participation in) health care and self-management?

II) What are the competencies for self-management, transfer readiness and self-efficacy of adolescents with chronic conditions?

III) What are the experiences and recommendations of young adults with chronic conditions, their parents and health care providers with regard to the transition from pediatric to adult health care services?

IV) What is the added value of a participatory approach, Q-methodology, and a mixed methods design?

In four paragraphs, the subquestions associated with these major research questions will be answered, followed by a critical reflection on the conclusions by discussing their contribution to the body of knowledge in adolescent health. Methodological strengths and weaknesses of our approach are discussed in paragraph 4. This chapter ends (paragraph 5) with the presentation of implications for clinical practice.

1   Adolescents' preferences for health care and self-management

1.1   Conclusions

1.1.1   Health care preferences

Ia.   What are the preferences of chronically ill adolescents for health care professionals, outpatient and inpatient service delivery, and what improvements are recommended for adolescent health care provision in Erasmus MC – Sophia?

The adolescents who participated in the studies are able and willing to express their perceptions of the quality of health care services provided to them. They have a strong preference for health care providers who are competent, honest and straightforward. Most importantly, these should possess technical competence as well as good communication skills and attitudes. All wish to have a say in all matters that concern them and wish to be taken seriously. The older they are, the keener they are that the focus is on themselves rather than on their parents and that they are

treated like adults. Notably the younger ones acknowledge that their parents' support in dealing with professionals is still important to them.

Although adolescents are very positive about the current care they receive in the Sophia Children's Hospital, they would like to be treated less childish and more age-appropriate as they get older and gradually grow out of the pediatric environment. Central themes are *having a feeling of trust* and *having voice and choice.* In outpatient consultations they appreciate being seen on time and having their questions and needs attended to. Having as little pain or discomfort as possible, as well as maintaining contact with family and friends, are considered most important during hospitalization. In general, provider-related issues are rated as more important than environmental aspects.

1.1.2   Q-Care Profiles

Ib.   Which profiles for self-management and preferences for care (Q-Care Profiles) can be elicited in adolescents with chronic conditions?

Our Q-methodological study revealed that not all adolescents share the same ideas about illness and treatment. Instead, their ideas fit into four distinct so-called Q-Care Profiles (QCPs), which we named: 'Conscious & Compliant'; 'Backseat Patient'; 'Self-confident & Autonomous'; and 'Worried & Insecure'(Figure 1). All profiles share a desire to participate in decision making, but differ in attitudes toward independence, appreciation of the parents' role, self-management, treatment adherence, adjustment to illness or disability, and worries about their health.

Adolescents in profiles 'Conscious & Compliant' and 'Self-confident & Autonomous' feel more independent and display a higher degree of self-efficacy than those in the other two. However, these profiles are characterized by different attitudes toward treatment adherence. While 'Conscious & Compliant' adolescents prefer to adhere to treatment to avoid future health problems, 'Self-confident & Autonomous' adolescents consciously decide to be nonadherent. The 'Backseat Patient' and the 'Worried & Insecure' adolescent share a common feeling of dependency and lack of self-confidence, but for different reasons. 'Backseat Patient' adolescents lean on their parents because they feel uninvolved and incompetent. Leaving responsibility to their parents and having parents do the talking is easier for them. 'Worried & Insecure' adolescents lack confidence because they worry about their future health. They need their parents to support them in coping with insecurities.

All but 'Worried & Insecure' adolescents want to have a say in when to transfer to adult care and only the 'Backseat Patients' agree that they are not looking forward to this transition. Contact with fellow patients is desirable for 'Self-confident & Autonomous' adolescents only but not for those constituting the 'Conscious & Compliant' profile.

10

Figure 1   *The four Q-Care Profiles*

"I take my own decisions, I will not have my illness limit me"

Self-confident & Autonomous

"Just imagine that things will go wrong later"

Worried & Insecure

"As long as I just follow doctor's orders, I don't have to worry"

Conscious & Compliant

"If my parents know what to do, it's OK"

Backseat Patiënt

1.1.3   Q-Care Profiles: distribution and associations

Ic.   What is the distribution of the Q-Care Profiles in adolescents with chronic conditions and what are the associations between fit to the Q-Care Profiles and relevant variables?

A Q-methodological study neither informs us about the proportions of people adhering to the identified viewpoints, nor about associations with relevant socio-demographic and disease-related variables. Therefore, we presented the narrative descriptions of the Q-Care Profiles (QCPs) without their names to a large population of chronically ill adolescents and asked them their fit to each QCP.

   Four out of every five adolescents could identify well with at least one of the profile descriptions; fit to the QCPs little overlaps between them. The 'Conscious & Compliant' profile fits 56%, appealing more to older and higher educated youth. These experience better health and feel confident to manage their condition. The 'Backseat Patient' profile fits least (16%) and is associated with being younger, physically disabled, and lower educated. They experience lower quality of life and feel less self-efficacious. The 'Self-confident & Autonomous' profile fits 26%, but none of the variables considered is significantly associated with fit to this profile. The 'Worried & Insecure' profile fits 25%, mostly younger, lower educated girls of non Dutch ethnicity. They experience poor health, lower quality of life, and feel less self-efficacious.

   The four QCPs distinguish well between different self-management styles and may help identify risky behaviors. 'Backseat Patients' demonstrate lack of independence, 'Worried & Insecure' persons risk depression, while 'Self-confident & Autonomous' adolescents may fail to adhere to prescribed treatment.

1.2   Discussion

I)   Adolescents desire to be involved in their health care and their perspectives need to be incorporated into adolescent health care provision

For young people with chronic conditions, being heard and listened to by the hospital staff is important, as was reported before.[1] They welcome being treated as an equal partner in care and want more attention to the needs of older children.[2] Nevertheless, several studies have made clear that children's opinions and views are underused that they have mixed experiences of being consulted about wishes for disease management.[1, 3] In the Netherlands, children or adolescents are rarely involved in the evaluation of health care services.[4, 5]

Our adolescents' preferences for health care are in line with international studies among adolescents with various chronic conditions.[6-11] Professional expertise, honesty and trust are seen as core concepts in the relation between adolescents and health professionals.[2, 10] Our study adds that professional expertise is most important, while honesty and kindness of the provider came in second and third place, respectively. Younger adolescents are more concerned about staff kindness, pets' visits and the discomfort of painful procedures, like in a study in another Dutch hospital.[4] In contrast to some other studies, participants in our study did not express concerns about patient safety, hygiene, or preferred gender of providers.[6, 8, 9]

Provider-related issues seem more important than aspects of the physical environment or process issues, suggesting that adolescents understand and value both scientific and interpersonal aspects of care.[2, 12] Those who frequent visit the hospital are more concerned about staff listening to them and seem more interested in meeting fellow patients. In contrast to some other studies, our participants do not voice a strong need for a dedicated adolescent unit[13] or more age-appropriate waiting areas.

Inquiring into adolescents' preferences contributes to making adolescent health more age-appropriate and patient-centered.

II)   Differences in attitudes and preferences in adolescents can be seized in comprehensive taxonomies representing different self-management styles

There is no "one size fits all" approach to stimulate taking responsibility for self-management. The more so because preferences for health care provision differ. It may thus be practical to identify sizeable and meaningful subgroups.[14] Since self-management encompasses the ability to affect the cognitive, behavioral and emotional responses necessary to maintain a positive quality of life,[15] integrative taxonomies are needed that transcend 'medical' issues and incorporate social and emotional attitudes as well. The QCPs we identified provide these.

Most adolescents empathically try to see themselves as normal individuals,[16] but their strategies for normalization may vary. The QCPs compile these varying responses into comprehensive narratives that reflect the challenges of growing up with a chronic condition. This is a new approach. We found only two comparable studies, in youth with diabetes, which revealed three distinct styles to achieve glycemic control: inadequate, moderate and high adaptive style.[17, 18] However, these stud-

ies only considered adherence to diabetic treatment and did not include adolescents' opinions. The QCPs reflect adolescents' own perspectives and not those of clinicians.

Previous studies on adolescents' health care preferences and priorities have focused on major-ity views of adolescents,[2] or on specific disease groups. In the literature, different preferences were reported with regard to communication, being treated as an adult,[6, 19] level of involvement in decision making,[19-21] and parental presence during consultations.[2, 10, 22] Knopf *et al* found that half of the adolescents studied favored a passive decision making style followed by one third preferring shared decision making.[23] Our study confirms these differences in preferred level of involvement, yet all adolescents appreciate being able to have a say in important matters about their health or treatment. This is related to the feeling that they are the very experts in self-management and decision making, while parental encouragement makes them even more confident.[24] Also, adolescents with chronic illnesses seem more comfortable involving parents in health care than are healthy adolescents.[10] Still, adolescents have different preferences on parents' involvement. Those in the 'Conscious & Compliant' and 'Self-confident & Autonomous' profiles prefer to be treated as adults and to have communication directed to them rather than to their parents.[2, 8] These adolescents feel competent to handle their own affairs, in contrast to the passive 'Backseat Patients', who do not mind (and need) their parents doing the talking for them. So, preferences for health care and self-management are inter-related and may be expected to correlate with competencies as well.

Inspired by "Leary's Rose", a model developed by Timothy Leary in 1957 to map communication in-teractions, two basic dimensions (axes) may be distinguished positioning the four QCPs (Figure 2):

Figure 2 *Dimensions of the QCPs*

279
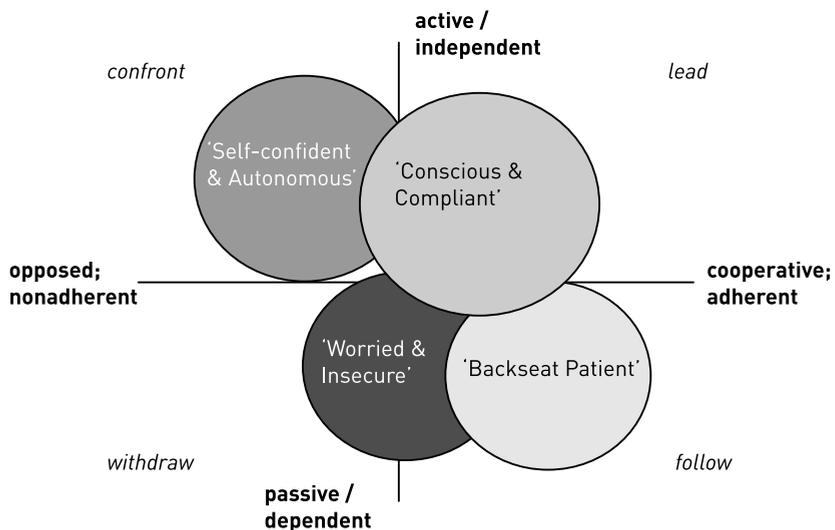
1. one axe representing the attitude toward self-determination: active (independent) behavior versus passive (dependent) behavior (Leary's 'above' and 'below' dimension, representing power and influence or lack thereof)
2. the other axe representing the attitude toward self-management and adherence: cooperative, adherent behavior versus opposed, nonadherent (Leary's dimension of personal distance or closeness: 'opposed' or 'together')

The overlap between the QCPs and the established correlations with HRQoL, socio-demographic variables, and self-efficacy seem to corroborate this model. A qualitative study into self-care coping strategies in adults with diabetes found three different types of patients: the proactive manager, the passive follower, and the nonconformist.[25] Three of our QCPs are recognizable in these vignettes, but not the 'Worried & Insecure' type – a group particularly at risk for low HRQoL and social isolation.

In our study, however, predictive validity of fitting into a particular QCP for self-management measures or clinical outcomes could not be established. Also, we do not know whether the QCPs represent a stable self-management typology, a character trait or a 'temporary state'. For example, as 'Backseat Patients' tend to be younger, they may be expected to further develop their competencies and become more pro-active self-managers. In due time 'Worried and Insecure' adolescents could also develop more self-confidence for coping with their disease. However, time since diagnosis was not related to any profile preference. These matters should be further explored in longitudinal studies and studies on correlations with psychological measures.

2   Adolescents' health care competencies

2.1   Conclusions

2.1.1   Competencies for managing hospital consultations

IIa.   What are the perceived and observed competencies for hospital consultations of adolescents with chronic conditions?

Adolescents desire to be involved in health care communication, but their actual, observed level of involvement during hospital consultations is low. Most explain their marginalized position as a result of their own indifference or as a consequence of *"not being asked to participate"* while a minority sees themselves as incompetent due to being *"still a child".* They indeed differ not only in their preferences for an executive role in communication, but also in their competencies. The older ones, the girls, the higher-educated, the more experienced in terms of hospital visits, and those who feel more self-efficacious, are the ones who are most active during consultations. Although realizing that one day they will have to manage communication themselves, many adolescents choose to leave the hard work to parents. At the same time, they are ambivalent about

the parents' role: while they need their parents and often appreciate their support, they also feel not at ease when parents interfere.

We found that pediatric staff do not restrict parents' presence or dominance of the communication but still try to involve adolescents. They are frustrated by *"overbearing"* parents, but just as the parents, they are anxious if the adolescents do not adhere to treatment or show lack of involvement. Triadic communication with adolescents is all but multi-party-talk and adolescents do not act and are not considered as equal partners. While the Dutch legal context grants adolescents every right to participate and (co)decide, and are supposed to take over control from their parents as an essential step in transition to adult care, the current structure of consultations, the communication style employed and the presence of parents hindered adolescent involvement in communication.

2.1.2   Transfer readiness

IIb.   *What constitutes adolescents' readiness to transfer to adult care?*

For transfer from pediatric to adult care to be successful, all partners involved need to be 'ready'. The process of 'getting ready' encompasses building the capacity of the adolescent, parents and providers to prepare for and to begin the process of transition. The majority of adolescents feels that they are ready for transfer. Feeling more self-efficacious in skills for independent hospital visits and a greater perceived independence during consultations are most strongly associated with being ready to transfer. Higher readiness is associated with older age, but age is not the most important explaining variable. Apart from age, adolescents' attitude to transition, and their level of self-efficacy in managing day-to-day self-care and hospital consultations seem to be the keystones to transfer readiness. Disease-related factors and effect of the condition including quality of life are only weakly associated with transfer readiness.

2.1.3   Self-efficacy

IIc.   *What are the essential, generic tasks for self-management and how can self-efficacy for*
           these be measured in adolescents with chronic conditions?

Successful self-management includes three major tasks for any person with a chronic illness: managing the medical aspects of the illness, dealing with the psychological consequences and managing social roles and relations. As we wanted to develop a scale measuring the required competencies for self-management, we asked adolescents for their ideas on self-efficacy. Younger respondents came up with *"doing things yourself, without help from your parents"*, while older respondents more often mentioned *"being autonomous in taking care of yourself and in decision-making"*. Both aspects were covered in the new On Your Own Feet Self-Efficacy Scale rating adolescents' confidence for performing tasks in three nondisease-specific domains: (1) coping with the chronic condition (corresponding with dealing with emotional consequences and social relations),

(2) knowledge of the chronic condition (managing the medical aspects), and (3) skills for independent hospital visits (dealing with the patient role).

Our survey revealed a high mean score for the total scale and some differences between adolescents. Younger adolescents are less self-efficacious than older ones, but age is not the only relevant factor for self-efficacy. Although the difference is small, boys rate their self-efficacy higher than girls. Higher educated adolescents as well as those without physical limitations have higher self-efficacy scores than those who attend lower educational levels and experience disabilities.

2.1.4   Health-Related Quality of Life in adolescents, comparing adolescent self-reports and proxy reports

IId.   What is the extent and direction of agreement between Health-Related Quality of Life adolescent self-reports and proxy reports of their parents and which factors are associated with child-parent agreement?

Pairs of adolescents and their parents were studied to investigate agreement on adolescents' Health-Related Quality of Life (HRQoL) measured by KIDSCREEN-10 and DISABKIDS condition generic measure (DCGM-10). On average, adolescents score their HRQoL higher than do their parents. In almost half of the cases the adolescent and parent agreed on the adolescent's HRQoL. For the other pairs, statistically significant disagreement in either direction existed, albeit this tended to be relatively minor (defined as 0.5 - 1 *SD*). Substantial differences (> 2 *SD*) were found in no more than 13% of KIDSCREEN-10 pairs and 10% of DCGM-10 pairs.

Parents seem to weigh the impact of the condition more heavily than their child does, which means that self-reports and parent proxy-reports are not interchangeable. Since adolescents are expected become partners in their own health care and HRQoL measures provide relevant clinical information about psychosocial functioning, it is recommended to focus on the adolescent's own perceptions of HRQoL.

2.2   Discussion

III)   Triadic communication during consultations marginalizes adolescents and does not strengthen their competencies to become partners in health care

In the Netherlands, it is common practice that parents are present during adolescent health care consultations. Our survey revealed that only 12.8% had gone into the consultation room alone during their most recent consultation. Our study confirmed sparse international research establishing that adolescents often remain inactive during triadic consultations despite good interactional competence,[26, 27] and that this situation does not correspond to adolescents' desire to be actively involved in their care.[8] Triadic encounters are likely to generate tensions and differences of opinion, and in the presence of two or more adults, adolescents' voices are likely to be muffled. In any encounter involving three or more actors there is a tendency for two of them to enter a coalition in order to advance a personal agenda or achieve an agreed outcome.[28] Pyörälä found

that adolescents with diabetes had an active patient role in two-party encounters with dieticians, whereas in triadic encounters they often turned into withdrawn bystanders.[27]

Parents' presence during consultations is regarded as both inhibiting and supportive.[8, 21, 29] Parents' presence could, unwillingly, delay the development of adolescent self-management skills.[30] It raises tension especially when personal or sensitive topics are discussed,[8] but discussing risky behaviors and the development of independence is crucial for developmentally appropriate adolescent care.[31] From our observational study we learned that adolescents spoke 15.6% the time (SD = 7.4) when parents were present, and 27.5% (SD = 9.4) without parents (P < .01). These percentages were highest in adolescents over 16 years of age and in girls (Gianotten 2011 - unpublished Master's thesis).

Pediatric providers in our study felt ambivalent about asking parents to step aside, even though the Dutch Medical Treatment Act (WGBO) gives them a firm legal basis to do so. This ambivalence may be related to insufficient communication skills to handle this complex situation. Therefore, doctors and nurses should be trained in age-appropriate communication skills, such as asking direct questions, discussing psychosocial and treatment-related subjects relevant to adolescents, and maintaining confidentiality.[10, 32] Health care providers should listen to parents and support their roles, but should see young people alone for part of the consultation.[33-35]

IV) An integrated developmental perspective, including the strengthening of adolescents' independence and self-management competencies should guide adolescent care

Adolescence is a 'stage not an age', implying there are no clear age limits to indicate when one is ready for transfer to adult care, although higher age is a positive predictor of transfer readiness.[36] Nevertheless, as we also found, physical and psychological criteria should be met as well.[36, 37] A developmental perspective on adolescence provides a conceptual framework to better understand the young adult's readiness to engage in self-management.[38] Only a few empirical studies have explored predictive factors for transfer readiness and the method by which readiness could be assessed.[39, 40] McPherson *et al* found that severity of disease had a negative impact on patients' interest in transition and anticipated difficulty of transfer.[40] Similar to other studies, our adolescents are moderately concerned about the forthcoming transition.[41] Also, disease-related variables are not related to transfer readiness, but adolescents' attitude to transition and their level of self-efficacy in managing self-care are the keystones to transfer readiness. This indicates that a medical model, only concerned with clinical outcomes, is not sufficient to successfully guide adolescents with chronic conditions to adulthood and adult care.

Strengthening adolescents' independence and self-management competencies is a challenge for health care professionals,[34, 35, 42] as there is some evidence that self-efficacy in disease management is beneficial for health outcomes,[43-46] social participation,[47] and to adolescents remaining in care after transfer.[48] Formulating self-management assessments and individual transition plans could really make a difference.[35, 49] These assessments are a form of Patient Reported Outcomes (PROs). Discussing PROs such as HRQoL outcomes during hospital consultations with adolescents, is reported to facilitate discussion about psychosocial wellbeing.[50, 51]

V)   *Agreement between adolescent self-report and parent proxy reports of patients*
       reported outcomes is considerable; adolescents are well capable of judging their
       preferences and competencies

In our survey we collected data from parents and adolescents - allowing the exploration of the (dis)agreement between both. Preliminary analyses revealed significant differences between parent and adolescent ratings of health care preferences, self-efficacy and self-management, Q-profiles, and HRQoL.[52] Not all outcomes of these comparisons are reported in this thesis, with the exception of direction and extent of agreement surrounding HRQoL measurements. Discrepancies between child HRQoL reports and parent proxy-reports are referred to as 'the proxy problem',[53, 54] but little is known about influencing factors and the direction of discrepancy.[55] What is known is that parents tend to rate their child's HRQoL lower than their children do,[56] but our study shows parents may also overestimate their child's HRQoL. In addition, we found considerable agreement between adolescent-parent pairs (defined as within the range of + 2.0 *SD*). Most disagreement tended to be minor, suggesting that the proxy problem in adolescents with chronic conditions may be overestimated. Unexplained is the finding that a lower educational level of the adolescent or the parent is associated with more disagreement.

Parent - adolescent ratings with respect to preferences for care, transfer readiness and attitude toward transitions were also quite similar (van Oijen 2010 - unpublished Master's thesis). Most pairs agreed on the scores of the preferences for health care provision and the priorities for change in the Sophia Children's Hospital. Also, parents and adolescents often agree on the adolescents' and parents' readiness for transfer. Seventy percent of the pairs agreed with one another. We have not yet explored (dis)agreement between adolescents and parents on self-efficacy, self-management and fit to QCPs. The overriding principle is that there are two different, equally important, perspectives to consider.[57]

The fairly high agreement between adolescent self-reports and parent proxy reports could be interpreted as a sign of growing adolescent capability to judge their preferences and competencies. On the other hand, differences in Patient Reported Outcomes could stimulate discussion between parents and adolescents about shifting responsibilities for self-care. Giving them appropriate instruction and ensuring parental support will help the adolescents gain positive experiences and self-confidence.

3   Experiences with transitional care

3.1   Conclusions

3.1.1   *Experiences with transfer to adult care*

IIIa.   *What are the experiences with the transfer to adult care of young adults with chronic*
          conditions, their parents and health care providers and what are their recommendations
          for improvement of transitional care?

Seeing that there is a cultural chasm between pediatric and adult-oriented services, we must prevent that these chronically ill adolescents get caught in this chasm. At the time of the study, only the hemophilia department offered a structured transition program in the Sophia Children's Hospital. As a consequence, most patients and parents are not being prepared for transition. Patients, parents and professionals consider leaving pediatric care a logical step towards adulthood. Leaving familiar surroundings is harder for parents than for young adults who display a positive 'wait-and-see' attitude. All parties agree that currently, transition of care is complicated by cultural gaps between pediatric and adult services. Young adults and parents feel lost after transfer and recommend their peers to be alert and involved. Providers, on the other hand, worry about non-compliance, lost to follow-up, and lack of independence.

Improvement priorities seem to be better patient and parent preparation for differences between health care settings and for new roles and responsibilities, as well as more collaboration and personal links between pediatric and adult care providers.

3.1.2   Transitional care practices and priorities

IIIb.   What are the current practices regarding transitional care and which priorities for change are identified by teams of health care providers?

Our research reveals that most of the treatment teams surveyed pay some attention to preparation for transition of care, but mostly fail to use more structural interventions. No more than one in five teams employ some form of transitional care: using transition protocols, individual action plans, or having a transition coordinator or transition clinic in place. At the same time, over 85% of the professionals give high priority to improving self-management, adherence and self-reliance in their population. This discrepancy indicates that promotion of self-management and social participation are indeed important areas for improvement for adolescent health care in the Netherlands.

3.2   Discussion

VI)   Transition to adult care is a logical and positive process, but the adolescents and their parents need to be better prepared

All actors in our study considered moving to adult services as "logical" and even desirable for young people with chronic conditions, as reported in other studies as well.[41, 58, 59] Nevertheless, parents and adolescents may have different perceptions before transfer: parents will typically be anxious, whereas adolescents display a 'wait-and-see' attitude.[41, 60] Still, the prevailing belief of our young adults and parents after transfer is that it was welcome and well-timed. Still, transfer is not easy because it implies adjusting to the new environment while leaving behind familiar surroundings and trusted health care providers.[61-63] Especially sudden and unprepared transfer will be challenging,[64] and may lead to drop-out or poor adherence to treatment.

In 1909, the French anthropologist Arnold van Gennep described three stages marking the transition to adulthood in primitive societies: *segregation stage*, *liminal stage*, and *(re)incorporation*

stage.[65] These are recognized during the transition to adult care as well. Adolescents describe a process of segregation in which they gradually "grow out" of their old pediatric environment. In the next stage, they are most vulnerable as they are literally *in limbo* between two worlds. Liminality implies being on a threshold. Young adults and their parents describe this stage as *"feeling lost"*. In the (re)incorporation stage the passage has been completed and the child now has a new, adult identity. During transition, *rites de passage* (public ceremonies) support the social system and protect the young persons against mischief. These help people in transition to feel connected and guide them through the crisis. However, in current transitional care practices, these *rites de passage* are often absent. All actors in our study agreed that the move from pediatric to adult services should be better prepared for and rituals such as farewell ceremonies may support this.

Transition does not only imply loss, but also gain[66]: new relations offer new opportunities and it is a period for disease-related learning and personal growth for adolescents and parents.[67] Being situated – having found your place in a new situation -, mastery of new skills, and a new identity are the typical features of having (successfully) passed through a transition.[68] For the young adults we interviewed, the change from family-oriented to patient-oriented care was not unwelcome. Yet the shift from parental care to self-care is most critical for achieving successful transition.[41, 61, 64]

The key challenge for health services lies in bridging the differing cultures of pediatric and adult health care by more communication and collaboration. Worldwide, the cultural differences between pediatric and adult care are considerable.[69] Nevertheless, these differences are also functional. In pediatric care there is a the clear role for parents, supportive practice style and awareness of developmental issues, but these attributes become dysfunctional and self-limiting for older adolescents and young adults. This implies that young people's views should be listened to and that they should be taught self-management skills. Meeting providers alone during consultations and earlier discussions about transition, opportunities to meet new providers and visits to adult-oriented venues prior to transition might aid adolescents in the transition process.[58, 64, 70]

VII) Current transitional practices are inadequate in Dutch hospitals and there is a necessity to improve adolescent health care

Since 2002, when the most recent survey of the state of affairs with regard to transition and transfer in Dutch somatic pediatric care was published,[71] attention to this subject and awareness of its importance has grown. However, to date very few publications have reported on such programs in Dutch hospitals[72, 73] and the same holds true for self-management. During our research (2008) few comprehensive transition programs were in place and systematic attention to promoting independence was limited despite the international evidence for essential elements of transitional care: adhering to a transition protocol, using individual transition action plans, appointing a transition coordinator, or setting up a transition clinic.[74] This lack of focus to adolescent health needs is also apparent in the existing professional guidelines, protocols or consensus statements. Our 2010 review of all 25 Dutch pediatric treatment guidelines revealed that only six paid (some) attention to issues related to transition of care and self-management; no more than half addressed HRQoL issues.[75]

Still, national consensus guidelines do not guarantee that transition programs are in place. In the US, the American Association of Pediatrics, the American Academy of Family Physicians and the American College of Physicians-American Society of Internal Medicine signed such a consensus statement in 2002.[76] From the US National Survey of Children with Special Health Care Needs 2005-2006 it appeared that only two in five adolescents received transitional care. Adolescents should have access to a comprehensive health care system that avoids preventable problems and manages chronic problems. The present US health care system falls short of accomplishing these demands and many gaps exist.[77]

The Dutch providers we surveyed agree that the care of adolescents with chronic conditions must be improved, implying better organization of transitional care and better support for self-management.

VIII) *A generic, noncategorical approach to adolescent health care has advantages of scale and acknowledges the existence of common adaptive tasks across conditions*

Rather than opting for a disease-specific approach, we opted for a noncategorical approach, as advocated by Stein and Jessop who concluded that diagnosis is not a helpful categorization when examining the psychological and social variables related to chronic illness.[78] People with chronic conditions face common challenges regardless of their diagnosis: the need to reconcile the demands and limitations of their condition with the desire to have a meaningful life and maintain a sense of wellbeing.[31, 79] Our studies confirm this. The diagnostic categories differ little in preferences and competencies for health care, but show large within variations . For example, in seven patient populations, the type of condition hardly seems to influence attitudes and concerns surrounding transition of care. This finding would support the implementation of generic self-management action plans and transition readiness assessments,[39] although it does not mean that disease-related factors do not matter at all. Rather consistently, our adolescents with physical disabilities display less self-efficacy, transfer readiness, and independent behaviors and more often feel drawn to the 'Backseat Patient' profile. Attention to self-reliance is extra important in these vulnerable groups. The QCPs may help to identify them.

In the biomedical sciences, this generic noncategorical approach is still quite exceptional, but feasible. It provides insights into the communalities and differences between adolescents within and across diagnostic groups. By providing clear directions for effective solutions, the research also contributed to efficiencies of scale.[31] Especially in the study of the low prevalence chronic conditions in childhood, small numbers of participants may limit the generalizability of findings.[57] On the other hand, choosing a generic, nondisease-specific sample limits the inclusion of more specific self-management measures related to self-efficacy and actual self-management of day-to-day therapy, and of measures directly related to disease severity. Therefore, a generic and a disease-specific approach each have their own value.[31]

The research was carried out in one university hospital in the Netherlands; similar research in other settings and countries may well yield different findings. However, compared to international studies, many of our findings resemble those established in other health care systems and settings. Although national health care systems differ in accessibility and affordability of chronic care,

Western biomedicine has penetrated most corners of the world with its specific features such as a predominance of the medical gaze, the distinction between 'care' and 'cure', a high degree of specialization combined with a lack of a holistic integral disease management, and the existence of fragmented services.

4 Methodological contributions

4.1 Conclusions

There is a relative lack of robust data on the specific adolescent health issues and on the perspectives of young people.[31, 57] When researching this age group, one should consider matters such participation in the research process of the subjects themselves, cognitive and psychosocial development, and effective recruitment.[57] Developmentally appropriate methods are key to ensure success. Therefore, we tested several promising research strategies, including participatory research (involving adolescents as coresearchers), Q-methodology, and a mixed methods strategy. Also, we developed a new generic scale to measure self-efficacy in adolescents with chronic conditions.

4.1.1 Participatory research

IVa. What are the feasibility, the advantages and the limitations of collaborating with chronically ill adolescents in participatory research?

Participation of young people from the onset of the research process is thought to enhance research quality.[57, 80, 81] In the Netherlands, participatory research with young children with Type 1 diabetes worked out well and gave deeper insight into the worldviews and competencies of children and into facilitators and barriers of child-adult collaboration.[5] In one of the 'On Your Own Feet' projects, we therefore asked a number of chronically ill adolescents to interview their fellow patients about experiences and satisfaction with several aspects of hospital care. Adolescents are able to participate to a certain extent and researchers find it exciting to work with them. Yet, participatory research is no easy job. The project implied a lot of work for the research team, hospital staff, volunteers and coresearchers alike. The research benefitted less than we had hoped for, both in terms of recruitment and of research quality.

Representativeness might have been a methodological issue here. The coresearchers differed from their fellow patients in being more outspoken about preferences, more critical of care received and more self-confident in talking to staff and peers. Also, despite our efforts, coresearchers' involvement was fairly limited. In our experience, participatory research demands high commitment, extensive resources, and hard work while not always succeeding in providing meaningful results. Most participatory research initiatives report satisfaction with both the process and the results, while labeling downsides as 'challenges'.[82-85] We feel that finding a balance between the benefits and costs of participation is necessary to ensure sustainability of efforts, commitment and credibility of results.

4.1.2 Q-methodology

IVb. What does Q-methodology add to our understanding of adolescents' preferences related to self-management and health care?

Q-methodology provides a scientific foundation for the systematic study of people's opinions, attitudes and preferences.[86-89] The research subjects represent their viewpoints by ranking a set of statements, after which by-person factor analysis reduces these many viewpoints to a few shared perspectives. Q-methodology clusters respondents according to their ranking of the statements presented, whereas conventional factor analysis clusters statements according to respondents' ratings. A population of viewpoints is described rather than a population of people. The focus on similarities and differences elicits the diversity of viewpoints and helps avoid concentrating on commonalities between participants.[89] Q-methodology may be seen as a hybrid research methodology, combining elements of qualitative and quantitative data collection and analysis.[90]

As shown before, applying Q-methodology among adolescents can be quite successful in triggering adolescents to speak freely and extensively about their own views and preferences.[91-92] In the course of the study we learned that health care professionals, too, appreciated the Q-typology. We believe that Q-narratives are potentially useful for clinical practice – a novelty in the world of Q.

The results of a Q-methodological study can only be generalized to the study topic, not to the wider population of respondents. This form of representativeness plays no role in Q-methodology.[87] Ours is one of the first studies to connect Q-Care Profiles with a survey exploring factor membership in a large sample.[93] So, the presentation of the Q-narratives to a broad population of adolescents with chronic conditions and the exploration of their associations may be seen as a contribution to Q-methodology.

4.1.3 Mixed methods

IVc. What is the added value of a Mixed Methods approach integrating qualitative and quantitative data in studying adolescents' preferences and competencies for care?

The mixed methods approach is a major asset of our research. We employed a sequential, exploratory design in which qualitative and quantitative methods are given equal priority and weight, and in which the second phase partly develops from the first. Approaching the topics and the population from different angles and through different research strategies (triangulation) enhances both the internal and external validity of the studies.[94]

Especially in the area of preferences, self-management styles, and triadic consultations, we feel this analytical approach had added value. By comparing the results of qualitative and quantitative studies (concurrent data analysis), inconsistencies and gaps could be explained, for example between 'potentially being able' and actual demonstration of abilities. Furthermore, qualitative studies helped us to generate hypotheses to be tested in the questionnaire, but vice-versa, findings from the questionnaire (for example, the discrepancy between self-efficacy and self-reported

independent behaviors) could be explained through insights obtained from interviews and obser-vations (sequential data analysis). However, integration of results is difficult to achieve in mixed methods research.[95] It is also quite rare: a recent review of 168 mixed methods studies in health care research states that parallel data analysis without integration is still the trend.[96]

Trying to combine two different worlds will inevitably meet with criticism from firm believers in one specific tradition only who probably feel that 'a bit of both' is never good enough: *"Oh, East is East, and West is West, and never the twain shall meet."* (Rudyard Kipling, 1892). But is boundary work not the most exciting academic enterprise of all?

4.1.4   Development of a self-efficacy instrument

IVd.   How can a new generic self-efficacy instrument for adolescents with chronic conditions be developed and validated?

Self-efficacy is both a condition for and a predictor of self-management behavior. We developed and validated a new, shortened 17-item generic self-efficacy scale measuring three domains related to coping, disease knowledge, and skills for independent hospital consultations. Psycho-metric properties of the new scale were good. The three factor structure of the original scale was retained and the subscales had good internal consistency, rendering the On Your Own Feet Self-Efficacy Scale a valid and reliable instrument for assessing self-efficacy in adolescents with chronic conditions. Concurrent validity with similar constructs as well as predictive validity for independent behavior during consultations could be established.

As self-reported self-efficacy could well serve as an outcome measure in clinical and research settings to evaluate interventions and as a diagnostic tool to identify need of self-management support, the new instrument is ready for further testing of its responsiveness to change and its predictive value for adherence to treatment.

4.2   Discussion of the research strategies

4.2.1   A broad scope of issues and actors

In the studies described in this thesis we explored many different areas of adolescent daily life, self-management abilities, views on being chronically ill and experiences with health care in both interviews and a questionnaire survey. A wide range of 187 socio-demographic, disease-related and psychological variables were collected in this survey. Nevertheless, in-depth exploration of all areas proved impossible. As the questionnaire was already very long, we refrained from including other relevant psychological measures, such as validated scales related to coping, general self-efficacy, depression or anxiety, youth-parent interaction or conflict. Use of the index versions of the HRQoL limited exploration of the various domains. Also, the parent questionnaire was a mirror version of the adolescent questionnaire (asking parents how they viewed their child's HRQoL for example), so we could not fully explore parents' own views, experiences, HRQoL, and perspectives.

A multi-actor perspective is often recommended, but less frequently employed. The actors in the survey were adolescents and parents; complemented with pediatric and adult health care providers in the study into experiences with transition. So far, few studies in transitional care have included adult-oriented health care providers' views.[22, 58, 70]

4.5.2 Sampling & response: a critical review

The survey was aimed at a total population sample: all adolescents treated on a regular basis over the past 3 years in the Sophia Children's Hospital. Although the final sample was large, the nonresponse rate was fairly high (64%). Moreover, 145 of the respondents (13.3% of the total number of invitees) did not finish the questionnaire until the end and were excluded from analysis. This implies a high risk for selection bias. Several factors may have negatively affected the response rate:

- Participation rates in social and epidemiologic studies conducted with postal invitations / questionnaires have steadily declined since 1970s. It may be hard to involve adolescents in research when the subject is one they do not have a particular interest in or when the study is related to issues that set them apart from others (such as being ill or disabled).[57]
- Our sample consisted of the total population of long-term patients. Since no specific group or recent event was targeted, it was not possible to address them personally or more directly. The letter inviting them to participate therefore was not signed by their own doctor or someone they knew personally. As only indirect measures could be used to target chronic conditions (the hospital does not record ICD-codes any more), it is possible we unintentionally approached nonchronically ill patients.
- The setting: compared to questionnaires administered to children and adolescents in the hospital setting, response rates in postal or computer-assisted questionnaires are much lower, as was shown in a Danish study: the response rate in a paper questionnaire 3–6 weeks after a hospital visit was 54%, compared to 73% in a questionnaire administered at the outpatient clinic directly after consultation.[97]
- The mode of administration (Internet based survey) affected the response rate, although people in the Netherlands (and youth in particular) have a high Internet access (over 85%). After receiving an invitational letter by postal mail, one had to log in at a secured Internet page. A meta-analysis of 39 Internet-based survey studies found an average response rate of 34%,[98] comparable to ours, while paper mail surveys yielded an unweighted average response rate that was about 10% higher.
- The length of the questionnaire: it comprised of 187 questions requiring at least 30 minutes to fill out.
- Our resources were only sufficient to send one postal reminder only; telephone reminders were not performed.

Consequently, the results may not be representative for the total population of the chronically ill adolescents seen in the hospital. Nonresponse analyses indicated that younger adolescents, girls and those with more extensive hospital experience were over-represented in the survey and

among the peer-research participants. Those who did not feel "chronically ill" were less inclined to fill out the questionnaire. Attempts to recruit young people for disease-related research may be unsuccessful simply because young people do not want to be reminded of their condition.[57] Since we also do not know whether our sample criteria were effective in selecting the 'truly' chronically ill, we could also see this as re-assuring: at least our respondents were more frequent hospital users than the nonrespondents. Finally, in all studies it was hard to catch adolescents with a non Dutch ethnic background. Because health care preferences and competencies proved associated with gender, age and ethnicity, this skewed representation might have affected the outcomes - although it is impossible to predict how.

In the On Your Own Feet qualitative interview study (including the Q-methodological study), purposive sampling was applied in order to achieve a heterogeneous, maximum variation sample.[99] Participants were sampled on (theoretical) potentially relevant variables that could influence coping with the chronic condition, preferences and competencies for self-management. These variables included age, gender, ethnicity, nature of the condition and extent of hospital experience. However, other potentially relevant characteristics, such as severity of the condition and impact on daily life could not be assessed beforehand. Here, too, nonresponse was fairly large (53%) and often adolescents provided the rationale that their disease *"did not bother them"*.

For the participatory research study and the focus groups with health care providers, we had no other option but to use convenience samples although this may have come at the expense of information and credibility.[99] In the focus groups, several pediatric nurse specialists but only few pediatricians participated; while the number of participant of the disco party was rather low. The observations of health care consultations were made in a variety of different outpatient departments representing different patient populations, but we were dependent upon the collaboration of professionals. In six clinics we observed between 3-6 different patients, but in the other two clinics (IBD and metabolic diseases) only one patient each, limiting the representativeness. Still, we observed more similarities than differences in the way consultations were performed across the various outpatient departments and the professionals involved. However, the number of consultations in general was too low to detect different professional styles, as there were few consultations involving only nurse specialists ($n = 8$) or a dietician ($n = 4$).

In the study examining experiences with transitional care, we combined a combination of random and stratified sampling strategies to invite those who had recently transferred to adult care with seven different conditions. Snowball sampling was used to identify adult health care professionals. Here, it may be regarded as a strength that we were able to compare young adults', parents', and professionals' perspectives. There were some differences between parents and adolescents, but we also noted a remarkable resemblance of parent' and patients' recommendations to improve transitional care. A limitation of this study is that in some chronic conditions, adolescent nonresponse and refusal rates were high, implying that there could be a selection bias. Young adults with mild health complaints may have been less inclined to participate, resulting in an overrepresentation of more severe conditions and an overestimation of the reported difficulties during transfer. On the other hand, many young adults could not be reached and we do not know whether they have dropped out of care. Furthermore, the small numbers of participants in each diagnostic group did not allow for detecting differences between the conditions. Finally, pos-

sible benefits of a structured transition program could not be established, as only one department offered this at the time.

In the national survey among teams of health care providers, selection bias may have occurred because this too was a convenience sample. We do not know whether the respondents form a good representation of all treatment teams for young people with chronic conditions in somatic care and whether they indeed were voicing the opinion of their own treatment team. Nevertheless, there was a good spread of respondents from different hospitals (and rehabilitation centers) all over the Netherlands and from different subspecialties.

4.5.3 The need for a developmental perspective in research

Our research projects were all cross-sectional, providing a momentary snap shot of adolescents' preferences, competencies and experiences with care. Since time is an essential element in transition,[69] longitudinal studies are required to explore the initial phase, midcourse experience, and outcome of the transition experience.[67] The only longitudinal study performed so far established that feelings about the desirability of transfer, the appreciation of medical care, the relationships with health care providers, and the parent's role changed over time.[59] Very few studies have prospectively studied the changes in self-efficacy, self-management and self-advocacy, physical and psychological symptoms in adolescents during transition to adult care.[46, 74]

5 Implications for clinical practice

1) Inquire into young people's preferences, respond to their needs

Since young people with chronic conditions are able and willing to express their views on the perceived quality of health care services provided to them, health care professionals should inquire after these views and adjust their communication style accordingly. Health care professionals should try to involve them in their own treatment as much as possible. This helps to build positive, trusting relations which is a prerequisite of shared responsibility for treatment.

Parents must be involved in adolescent health care, but since adolescents are expected and desire to become partners in their own health care, it is recommended to focus on their own judgments. Health care providers should listen to parents and support their roles, but adolescents' perspectives should be leading.

2) Adolescents should be seen alone for part of the consultation

A key issue in adolescent health communication is the true partnering between adolescents, parents and doctors. At the same time, in their transition to adulthood, adolescents need to start managing their own affairs. One key opportunity in this regard is feeling confident to see the health professional alone. The Dutch Medical Treatment Act (WGBO) provides a firm basis for including adolescents as main partners in communication. The organizational setting, however, usu-

ally does not. Therefore, doctors and nurses should be trained in skills for effective communication with adolescents. While they should listen to parents' concerns and support parental roles, they should see young patients alone for part of the consultation and encourage independent hospital communication.[33-35] This helps to strengthen adolescents' competencies for shared responsibility in treatment.

3) Advice and approaches to adolescents with chronic conditions should be adjusted to different self-management styles

Use of Q-Care Profiles (QCPs) in clinical practice may be promising as these seem to catch several important concepts in one short description and incite adolescents to talk about their opinions. Presenting the QCPs during consultations could thus stimulate a much needed discussion about adolescents' attitudes toward self-management roles. Clinicians should be aware that self-management styles may differ among chronically ill adolescents and adjust their advice accordingly. Since educational interventions alone are insufficient to promote adherence, adding a behavioral component could increase efficacy.[100] The QCPs may serve as such a component.

Only one QCP, 'Conscious & Compliant', sets a positive example for the others, scoring better on independence, self-efficacy, quality of life, and on intention to treatment adherence. The three other QCPs involve (different) potential health risks and introducing the profiles may help identify such risks and select appropriate interventions. While all adolescents should be encouraged to learn new skills, this is particularly relevant for 'Backseat Patients' and 'Worried & Insecure' adolescents. They seem not ready for self-management or transfer to adult care. Furthermore, 'Worried & Insecure' adolescents need extra attention because they may be at risk for clinical depression. And then, the 'Self-confident & Autonomous' type may be inclined to ignore providers' advice. Carefully exploring adolescents' motivations and barriers related to adherence, for example through Motivational Interviewing,[101] may contribute to positive outcomes.

4) Comprehensive adolescent health care addresses not only medical needs, but also psychosocial, self-management and educational issues

Optimal adolescent health care includes both medical, psychosocial and educational / vocational aspects.[76, 102] To enhance transfer readiness, health care providers should focus on improving adolescents' competency and behavioral skills,[36] without failing to assess adolescents' knowledge of their condition and treatment. Repeated discussions on transition could further enhance adolescents' readiness for transfer to adult care.

Health care providers should pay systematic attention to the psychosocial developmental trajectory of their adolescent patients.[103] Assessment of Patient Reported Outcomes such as HRQoL,[50, 51, 104] self-management and independent behaviors (in action plans such as individual transition plans),[35, 105, 106] and of transition readiness [39, 107] seems promising to this end.

5)   Better preparation for transition in care is needed

Transitional care should not be confined to a pediatric paradigm and be disconnected from the principles and practice of adolescent medicine.[108] Transition should therefore be incorporated in a comprehensive, lifespan perspective on health care for young people with chronic conditions.[109] Our research findings suggest that transition should be better prepared by starting earlier and strengthening adolescents' independency and changing parents' roles. Health care providers' first priority is building bridges between pediatric and adult-oriented care. Responsibility for a safe and smooth transition extends beyond pediatrics: transitional care should therefore not be restricted to the child-oriented services. Gaining trust and investing in new personal relations is the way forward for all parties involved: transition is about responding and bonding.

6)   A generic nonconditional approach when feasible, a disease-specific approach when necessary

Adolescents with any kind of chronic condition are facing the same challenges related to coping with their condition and their transition to adulthood,[31, 70] which call for generic, inter-specialty developments in transitional care.[110] Besides, adopting a generic approach is more efficient than developing new interventions and instruments for each disease group.

The road ahead....

The increase in the number of young people with chronic conditions in the Netherlands – who are the adults of the future – signals the importance of smooth transition to adulthood and to adult care, implying that self-management and social participation should be promoted. The conclusions from the On Your Own Feet research program guided the development of a framework for the national 'On Your Own Feet Ahead' Quality Improvement Collaborative (Figure 3). However, it would require another PhD thesis to go into our experiences with this action program and I happily leave this job to someone else....


Figure 3   *Framework Quality Improvement Collaborative 'On Your Own Feet Ahead'.*

References

1. Coyne I. Consultation with children in hospital: children, parents' and nurses' perspectives. *J Clin Nurs.* 2006;15(1):61-71.
2. Britto MT, DeVellis RF, Hornung RW, DeFriese GH, Atherton HD, Slap GB. Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics.* 2004;114(5):1272-1280.
3. Coad JE, Shaw KL. Is children's choice in health care rhetoric or reality? A scoping review. *J Adv Nurs.* 2008;64(4):318-327.
4. Snel MC. Dokter, dit vind ik! Verbeterpunten voor een megacool ziekenhuis [Doctor, this is my opinion! Ideas for a megacool hospital]. *Kind en Ziekenhuis.* 2006;29(1):18-22.
5. Dedding CWM. *Delen in macht en onmacht : Kindparticipatie in de (alledaagse) diabeteszorg*. [Sharing power and impotence: Child participation in (every day) diabetes care]. PhD Thesis. Amsterdam, Universiteit van Amsterdam; 2009.
6. Freake H, Barley V, Kent G. Adolescents' views of helping professionals: a review of the literature. *J Adolesc.* 2007;30(4):639-653.
7. Woodgate RL. Health professionals caring for chronically ill adolescents: adolescents' perspectives. *J Soc Pediatr Nurs.* 1998;3(2):57-68.
8. Beresford BA, Sloper P. Chronically ill adolescents' experiences of communicating with doctors: a qualitative study. *J Adolesc Health.* 2003;33(3):172-179.
9. Farrant B, Watson PD. Health care delivery: perspectives of young people with chronic illness and their parents. *J Paediatr Child Health.* 2004;40(4):175-179.
10. Klostermann BK, Slap GB, Nebrig DM, Tivorsak TL, Britto MT. Earning trust and losing it: adolescents' views on trusting physicians. *J Fam Pract.* 2005;54(8):679-687.
11. Moules T. 'They wouldn't know how it feels...': characteristics of quality care from young people's perspectives: a participatory research project. *J Child Health Care.* 2009;13(4):322-332.
12. Shaw KL, Southwood TR, McDonagh JE. Young people's satisfaction of transitional care in adolescent rheumatology in the UK. *Child Care Health Dev.* 2007;33(4):368-379.
13. Smith S. Adolescent units—an evidence-based approach to quality nursing in adolescent care. *European J Oncol Nurs.* 2004;8(1):20-29.
14. La Greca AM. Issues in adherence with pediatric regimens. *J Pediatr Psychol.* 1990;15(4):423-436.
15. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J. Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns.* 2002;48(2):177-187.
16. Taylor RM, Gibson F, Franck LS. A concept analysis of health-related quality of life in young people with chronic illness. *J Clin Nurs.* 2008;17(14):1823-1833.
17. Rohan JM, Delamater A, Shroff Pendley J, Dolan L, Reeves G, Drotar D. Identification of self-management patterns in pediatric type 1 diabetes using cluster analysis. *Pediatr Diabetes*. 2011;12(7):611-618.
18. Schneider S, Iannotti RJ, Nansel TR, Haynie DL, Simons-Morton B, Sobel DO, *et al.* Identification of distinct self-management styles of adolescents with type 1 diabetes. *Diabetes Care*. 2007;30(5):1107-1112.
19. Wray J, Maynard L. Specialist cardiac services: what do young people want? *Cardiol Young.* 2008;18(6):569-574.
20. Dovey-Pearce G, Hurrell R, May C, Walker C, Doherty Y. Young adults' (16-25 years) suggestions for providing developmentally appropriate diabetes services: a qualitative study. *Health Soc Care Community.* 2005;13(5):409-419.
21. Zwaanswijk M, Tates K, van Dulmen S, Hoogerbrugge PM, Kamps WA, Bensing JM. Young patients', parents', and survivors' communication preferences in paediatric oncology: results of online focus groups. *BMC Pediatrics.* 2007;7:35.
22. Shaw KL, Southwood TR, McDonagh JE. Transitional care for adolescents with juvenile idiopathic arthritis: a Delphi study. *Rheumatology (Oxford).* 2004;43(8):1000-1006.
23. Knopf JM, Hornung RW, Slap GB, DeVellis RF, Britto MT. Views of treatment decision making from adolescents with chronic illnesses and their parents: a pilot study. *Health Expect.* 2008;11(4):343-354
24. Karlsson A, Arman M, Wikblad K. Teenagers with type 1 diabetes--a phenomenological study of the transition towards autonomy in self-management. *Int J Nurs Stud.* 2008;45(4):562-570.
25. Collins MM, Bradley CP, O'Sullivan T, Perry IJ. Self-care coping strategies in people with diabetes: a qualitative exploratory study. *BMC Endocr Disord.* 2009;9:6.

26. Wassmer E, Minnaar G, Abdel Aal N, Atkinson M, Gupta E, Yuen S, *et al.* How do paediatricians communicate with children and parents? *Acta Paediatr*. 2004;93(11):1501-1506.

27. Pyörälä E. The participation roles of children and adolescents in the dietary counseling of diabetics. *Patient Educ Couns*. 2004;55(3):385-395.

28. Gabe J, Olumide G, Bury M. 'It takes three to tango': a framework for understanding patient partnership in paediatric clinics. *Soc Sci Med*. 2004;59(5):1071-1079.

29. Young B, Dixon-Woods M, Windridge KC, Heney D. Managing communication with young people who have a potentially life threatening chronic illness: qualitative study of patients and parents. *Br Med J*. 2003;326(7384):305.

30. Williams C. Alert assistants in managing chronic illness: the case of mothers and teenage sons. *Sociol Health Illn*. 2000;22(2):254-272.

31. Sawyer SM, Drew S, Yeo MS, Britto MT. Adolescents with a chronic condition: challenges living, challenges treating. *Lancet*. 2007;369(9571):1481-1489.

32. Rutishauser C, Esslinger A, Bond L, Sennhauser FH. Consultations with adolescents: the gap between their expectations and their experiences. *Acta Paediatr*. 2003;92(11):1322-1326.

33. Reid GJ, Irvine MJ, McCrindle BW, Sananes R, Ritvo PG, Siu SC, *et al.* Prevalence and correlates of successful transfer from pediatric to adult health care among a cohort of young adults with complex congenital heart defects. *Pediatrics*. 2004;113(3 Pt 1):e197-205.

34. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: a multicentre cohort of adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2005;44(6):806-812.

35. McDonagh JE, Southwood TR, Shaw KL. The impact of a coordinated transitional care program on adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford)*. 2007;46(1):161-168.

36. Cappelli M, MacDonald NE, McGrath PJ. Assessment of readiness to transfer to adult care for adolescents with cystic fibrosis. *Child Health Care*. 1989;18(4):218-224.

37. While A, Forbes A, Ullman R, Lewis S, Mathes L, Griffiths P. Good practices that address continuity during transition from child to adult care: synthesis of the evidence. *Child Care Health Dev.* 2004;30(5):439-452.

38. Anderson BJ, Wolpert HA. A developmental perspective on the challenges of diabetes education and care during the young adult period. *Patient Educ Couns.* 2004;53(3):347-352.

39. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, *et al.* Measuring the Transition Readiness of Youth with Special Healthcare Needs: Validation of the TRAQ--Transition Readiness Assessment Questionnaire. *J Pediatr Psychol.* 2011;36(2):160–171.

40. McPherson M, Thaniel L, Minniti CP. Transition of patients with sickle cell disease from pediatric to adult care: Assessing patient readiness. *Pediatr Blood Cancer*. 2009;52(7):838-841.

41. Moons P, Pinxten S, Dedroog D, Van Deyk K, Gewillig M, Hilderson D, *et al.* Expectations and experiences of adolescents with congenital heart disease on being transferred from pediatric cardiology to an adult congenital heart disease program. *J Adolesc Health.* 2009;44(4):316-322.

42. Betz CL, Redcay G. Creating Healthy Futures: an innovative nurse-managed transition clinic for adolescents and young adults with special health care needs. *Pediatr Nurs.* 2003;29(1):25-30.

43. Iannotti RJ, Schneider S, Nansel TR, Haynie DL, Plotnick LP, Clark LM, *et al.* Self-efficacy, outcome expectations, and diabetes self-management in adolescents with type 1 diabetes. *J Dev Behav Pediatr.* 2006;27(2):98-105.

44. Griva K, Myers LB, Newman S. Illness perceptions and self efficacy beliefs in adolescents and young adults with insulin dependent diabetes mellitus. *Psychol. Health.* 2000;15(6):733-750.

45. Ott J, Greening L, Palardy N, Holderby A, DeBell WK. Self-efficacy as a mediator variable for adolescents' adherence to treatment for insulin-dependent diabetes mellitus. *Child. Health Care.* 2000;29(1):47-63.

46. Edwards R, Telfair J, Cecil H, Lenoci J. Self-efficacy as a predictor of adult adjustment to sickle cell disease: one-year outcomes. Psychosom Med. 2001;63(5):850-858.

47. van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, Wensink-Boonstra AE, Stam HJ, Roebroeck ME. Participation and health-related quality of life in adults with spastic bilateral cerebral palsy and the role of self-efficacy. *J Rehabil Med.* 2010;42(6):528-535.

48. Wojciechowski EA, Hurtig A, Dorn L. A natural history study of adolescents and young adults with sickle cell disease as they transfer to adult care: a need for case management services. *J Pediatr Nurs.* 2002;17(1):18-27.

49. Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM. Transition to adult health care for adolescents and young adults with chronic conditions: position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 2003;33(4):309-311.

50. de Wit M, Delemarre-van de Waal HA, Bokma JA, Haasnoot K, Houdijk MC, Gemke RJ, *et al.* Monitoring and discussing health-related quality of life in adolescents with type 1 diabetes improve psychosocial well-being: a randomized controlled trial. *Diabetes Care.* 2008;31(8):1521-1526.

51. Haverman L, Engelen V, van Rossum MA, Heymans HS, Grootenhuis MA. Monitoring health-related quality of life in paediatric practice: development of an innovative web-based application. *BMC Pediatr.* 2011;11:3.

52. van Staa AL, van der Stege HA, Jedeloo S. *Op Eigen Benen Verder. Jongeren met chronische aandoeningen op weg naar zelfstandigheid in de zorg. [On Your Own Feet Ahead. Young people with chronic conditions on their way to independence in health care].* Rotterdam: Hogeschool Rotterdam; 2008. ISBN 978-90-7905-902-7.

53. Eiser C, Morse R. Can parents rate their child's health-related quality of life? Results of a systematic review. *Qual Life Res.* 2001;10(4):347-357.

54. Theunissen, NCM, Vogels AG, Verrips GHW, Verloove-Vanhorick SP, Kamphuis RP, *et al.* The proxy problem: child report versus parent report in health-related quality of life research. *Qual Life Res.* 1998;7:387-39.

55. Shaw KL, Southwood TR, McDonagh JE. Growing up and moving on in rheumatology: parents as proxies of adolescents with juvenile idiopathic arthritis. *Arthritis Rheum.* 2006;55(2):189-198.

56. Upton P, Lawford J, Eiser C. Parent-child agreement across child health-related quality of life instruments: a review of the literature. *Qual Life Res.* 2008;17(6):895-913.

57. McDonagh JE, Bateman B. 'Nothing about us without us': considerations for research involving young people. *Arch Dis Child Educ Pract Ed* 2011 Jul 29 [Epub ahead of print] doi:10.1136/adc.2010.197947

58. Östlie IL, Dale O, Moller A. From childhood to adult life with juvenile idiopathic arthritis (JIA): a pilot study. *Disabil Rehabil*. 2007;29(6):445-452.

59. Tuchman LK, Slap GB, Britto MT. Transition to adult care: experiences and expectations of adolescents with a chronic illness. *Child Care Health Dev.* 2008;34(5):557-563.

60. Anthony SJ, Kaufman M, Drabble A, Seifert-Hansen M, Dipchand AI, Martin K. Perceptions of transitional care needs and experiences in pediatric heart transplant recipients. *American Journal of Transplantation.* 2009;9(3):614-619.

61. Kirk S. Transitions in the lives of young people with complex health care needs. *Child Care Health Dev.* 2008;34(5):567-575.

62. Stewart DA, Law MC, Rosenbaum P, Willms DG. A qualitative study of the transition to adulthood for youth with physical disabilities. *Phys Occup Ther Pediatr.* 2001;21(4):3-21.

63. Young NL, Barden WS, Mills WA, Burke TA, Law M, Boydell K. Transition to adult-oriented health care: perspectives of youth and adults with complex physical disabilities. *Phys Occup Ther Pediatr.* 2009;29(4):345-361.

64. Shaw KL, Southwood TR, McDonagh JE. User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. *Rheumatology (Oxford).* 2004;43(6):770-778.

65. van Gennep A. *The rites of passage*. London: Routledge & Kegan; 1906 [1960].

66. Kralik D, Visentin K, van Loon A. Transition: a literature review. *J Adv Nurs*. 2006;55(3):320-329.

67. Valenzuela JM, Buchanan CL, Radcliffe J, Ambrose C, Hawkins LA, Tanney M, *et al.* Transition to adult services among behaviorally infected adolescents with HIV—a qualitative study. *J Pediatr Psychol*. 2011;36(2):134-140.

68. Meleis AI, Sawyer LM, Im EO, Hilfinger Messias DK, Schumacher K. Experiencing transitions: an emerging middle-range theory. *Adv Nurs Sci.* 2000;23(1):12-28.

69. Rosen D. Between two worlds: bridging the cultures of child health and adult medicine. *J Adolesc Health*. 1995;17(1):10-16.

70. Reiss JG, Gibson RW, Walker LR. Health care transition: youth, family, and provider perspectives. *Pediatrics.* 2005;115(1):112-120.

71. Donckerwolcke RAMG, van Zeben-van der Aa DMCB. Overdracht van de zorg voor adolescenten met chronische ziekten: van kindergeneeskunde naar specialismen voor volwassenen. [Transfer of care for adolescents with chronic illnesses: from pediatrics to specialists for adults]. *Ned Tijdschr Geneeskd*. 2002;146(14):675-678.

72. Ammerlaan JW, van Pelt PA, Masselink IM, Prakken ABJ, Kruize AA. De jongvolwassene met juveniele idiopathische artritis (JIA): Transitie binnen het UMC Utrecht van WKZ naar AZU. [The young adult with JIA: Transition within the UMC Utrecht from WKZ to AZU]. *Ned Tijdschr Reumatol*. 2007;(3):7-12.

73. Scholten M, Kooijman R. Jongeren met hemofilie in stappen over de brug. [Young people with haemophilia are crossing the bridge]. *Kind en Ziekenhuis.* 2006;29(1):11-13.

74. Crowley R, Wolfe I, Lock K, McKee M. Improving the transition between paediatric and adult healthcare: a systematic review. *Arch Dis Child.* 2011;96(6):548-553.

75. van den Berg M, van Staa AL, Havers J. *De kloof tussen wat we weten en wat we doen in transitiezorg voor jongeren en adolescenten. Een Gap-analyse van de richtlijnen en standaarden.* [The gap between what we know and what we do in transitional care for adolescents. A gap analysis of guidelines and standards.] Rotterdam: XX Science / Hogeschool Rotterdam; 2010. www.opeigenbenen.nu

76. American Academy of Pediatrics; American Academy of Family Physicians; American College of Physicians-American Society of Internal Medicine. A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics*. 2002;110(6 Pt 2):1304-1306.

77. Park MJ, Adams SH, Irwin CE Jr. Health care services and the transition to young adulthood: challenges and opportunities. *Acad Pediatr.* 2011;11(2):115-122.

78. Stein RE, Jessop DJ. What diagnosis does not tell: the case for a noncategorical approach to chronic illness in childhood. *Soc Sci Med.* 1989;29(6):769-778.

79. Moos RH, Holahan CJ. Adaptive tasks and methods of coping with illness and disability. In: Martz E, Livneh H, eds. *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. New York: Springer; 2007.

80. Litt IF. Research with, not on, adolescents: community-based participatory research. *J Adolesc Health*. 2003;33(5):315-316.

81. Santelli JS, Smith Rogers A, Rosenfeld WD, DuRant RH, Dubler N, Morreale M, *et al.* Guidelines for adolescent health research. A position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 2003;33(5):396-409.

82. Cargo M, Mercer SL. The value and challenges of participatory research: strengthening its practice. *Annu Rev Public Health*. 2008;29:325-350.

83. Viswanathan M, Ammerman A, Eng E, Garlehner G, Lohr KN, Griffith D, *et al. Community-based participatory research: assessing the evidence.* Summary, Evidence Report/Technology Assessment: No. 99. AHRQ Publication No. 04-E022-1. Rockville (MD): Agency for Health care Research and Quality; 2004. www.ahrq.gov/clinic/epcsums/cbprsum.htm

84. Flicker S. Who benefits from community-based participatory research? A case study of the Positive Youth Project. *Health Educ Behav*. 2008;35(1):70-86.

85. Harper GW, Carver LJ. "Out-of-the-mainstream" youth as partners in collaborative research: exploring the benefits and challenges. *Health Educ Behav.* 1999;26(2):250-265.

86. Cross RM. Exploring attitudes: the case for Q methodology. *Health Educ Res*. 2005;20(2):206-213.

87. Watts S, Stenner P. Doing Q methodology: theory, method and interpretation. *Qual Res Psychol.* 2005;2(2):67-91.

88. Brown SR. *Political subjectivity: applications of Q methodology in political science*. New Haven, CT: Yale University Press; 1980.

89. Akhtar-Danesh N, Baumann A, Cordingley L. Q-Methodology in Nursing Research: A Promising Method for the Study of Subjectivity. *West J Nurs Res*. 2008;30(6):759-773.

90. Jedeloo S, van Staa AL. Q-methodologie, een werkelijke mix van kwalitatief en kwantitatief onderzoek? [Q-methodology: a true mix of qualitative and quantitative research?]. *KWALON*. 2009;14(2):5-15.

91. van Exel NJ, de Graaf G, Brouwer WB. "Everyone dies, so you might as well have fun!" Attitudes of Dutch youths about their health lifestyle. *Soc Sci Med*. 2006;63(10):2628-2639.

92. Tielen M, van Staa AL, Jedeloo S, van Exel NJ, Weimar W. Q-methodology to identify young adult renal transplant recipients at risk for nonadherence. *Transplantation*. 2008;85(5):700-706.

93. Baker RM, Van Exel J, Mason H, Stricklin M. Connecting Q & surveys: a test of three methods to explore factor membership in a large sample. *Operant Subjectivity*. 2010;34(1):38–58.

94. Creswell JW, Plano Clark VL. *Designing and conducting Mixed Methods Research*. Thousand Oaks CA: Sage; 2007.

95. Doyle L, Brady AM, Byrne G. An overview of mixed methods research. *J Res Nurs.* 009;14(2):175-185.

96.   Östlund U, Kidd L, Wengström Y, Rowa-Dewar N. Combining qualitative and quantitative research within mixed method research designs: A methodological review. *Int J Nurs Stud*. 2011;48(3):369-383.

97.   Jensen HI, Ammentorp J, Kofoed PE. Assessment of health care by children and adolescents depends on when they respond to the questionnaire. *Int J Qual Health Care.* 2010;22(4):259-265.

98.   Shih TH, Fan X. Comparing response rates from web and mail surveys: A meta-analysis. *Field Methods.* 2008;20:249-271.

99.   Creswell JW. *Qualitative inquiry and research design; choosing among five traditions*. 1st ed. London: Sage Publications; 1998.

100. Dean AJ, Walters J, Hall A. A systematic review of interventions to enhance medication adherence in children and adolescents with chronic illness. *Arch Dis Child.* 2010;95(9):717-723.

101. Naar-King S, Suarez M. *Motivational Interviewing with adolescents and young adults*. 1st ed. New York: The Guilford Press; 2011.

102. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, *et al.* Transition from child-centered to adult health-care systems for adolescents with chronic conditions. A position paper of the Society for Adolescent Medicine. *J Adolesc Health.* 1993;14(7):570-576.

103. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis MA. Growing into disability benefits? Psychosocial course of life of young adults with a chronic somatic disease or disability. *Acta Paediatr.* 2012;101(1):e19-26.

104. Engelen V, Haverman L, Koopman H, Schouten-van Meeteren N, Meijer-van den Bergh E, Vrijmoet-Wiersma J, *et al.* Development and implementation of a patient reported outcome intervention (QLIC-ON PROfile) in clinical paediatric oncology practice. *Patient Educ Couns.* 2010;81(2):235-244.

105. Sawyer SM. Action plans, self-monitoring and adherence: changing behaviour to promote better self-management. *Med J Aust.* 2002;177 Suppl:S72-74.

106. Lorig KR, Holman H. Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med.* 2003;26(1):1-7.

107. Fredericks EM, Dore-Stites D, Well A, Magee JC, Freed GL, Shieck V, *et al.* Assessment of transition readiness skills and adherence in pediatric liver transplant recipients. *Pediatr Transplant.* 2010;14(8):944-953.

108. Kennedy A, Sawyer S. Transition from pediatric to adult services: are we getting it right? *Curr Opin Pediatr.* 2008;20(4):403-409.

109. Roebroeck ME, Jahnsen R, Carona C, Kent RM, Chamberlain MA. Adult outcomes and lifespan issues for people with childhood-onset physical disability. *Dev Med Child Neurol.* 2009;51(8):670-678.

110. McDonagh JE, Viner RM. Lost in transition? Between paediatric and adult services. *Br Med J.* 2006;332(7539):435-436.

Summary

Summary

In the Netherlands, over 90% of all children suffering from congenital and acquired chronic conditions and (physical) limitations now survive and reach adulthood. Chronic conditions often limit children and adolescents in their activities and may restrict their social participation. For all young people, adolescence is a period of (sometimes tumultuous) biological, social and emotional changes, in which they have to complete four major developmental tasks: to consolidate their identity, to achieve independence from their parents, to establish adult relationships outside their families, and to find a vocation.

   Becoming an adult can be particularly challenging for those who have a chronic health condition, as the specific adaptive tasks that their disease, disability, or treatment demands may conflict with their developmental tasks. Preparing adolescents with chronic conditions for adulthood (in terms of self-management, social participation, and empowerment) should therefore be in key with optimal support in tackling problems such as insufficient adolescent independence, parental overprotection (or absence), poor adherence to treatment, and loss to follow-up.

   Apart from building self-sufficiency and independence, youth with special health care needs are also facing transition from pediatric care to adult care. Indications exist that adolescent health care is not well tuned to adolescents' needs and that transfer to adult care is not optimally organized or prepared. Although there are some 500.000 children in the Netherlands growing up with chronic conditions (14% of the total) and they are major users of pediatric hospital services, little is known about their knowledge, attitudes, and skills (competencies) regarding self-management of their condition. Furthermore, their experiences with transfer to adult care have hardly been explored before in the Netherlands. These knowledge gaps formed the motivation to conduct this PhD research.

The studies in this PhD thesis were all part of the 'On Your Own Feet' research program that aimed to map preferences and competencies for health care of adolescents with chronic conditions in the transitional period. In addition, their experiences with the transition to adult care were collected, as well as those of parents and health care providers. All studies were conducted with adolescents and young adults treated in the Erasmus MC – Sophia Children's Hospital in Rotterdam. We took a generic, ie, a nondisease-specific approach because the psychosocial challenges (eg, frequent hospitalizations, school absenteeism, and restriction of social participation) are generally similar between diverse chronic conditions. We therefore focused on the similarities between adolescents with chronic conditions rather than on the differences between diagnosis groups.

   The results of four different research projects in the 'On Your Own Feet' program have been integrated in this thesis. One multi-method qualitative project concerned face-to-face interviews with 31 adolescents (aged 12-19) at home, 34 peer interviews conducted by 9 adolescent coresearchers (aged 15-17) during a disco party, 39 observations of consultations in 8 outpatient clinics, and 3 focus group sessions with 27 health care providers. In addition, through a quantitative web-based questionnaire we collected data on preferences, attitudes toward self-management and transition in care, self-efficacy, and Health-Related Quality of Life (HRQoL). *Self-efficacy* is one's belief in one's ability to succeed in specific situations. 1087 adolescents (aged 12-19) and 830 parents

responded (including 593 adolescent-parent pairs). In another project, we interviewed 24 young adults (aged 15-22) with 7 different chronic conditions, their parents (*n*=24), and 17 health care providers about their experiences with the transfer from pediatric to adult care. Finally, as part of the Quality Improvement Collaborative 'On Your Own Feet Ahead!', set up by our research team, 115 teams of health care providers in Dutch hospitals were surveyed on their current transitional care practices, and their priorities for improvement.

The thesis consists of three parts, reporting respectively on the results related to adolescents' preferences, their competencies, and the need for action to improve transitional care in the Netherlands. The various research projects also dealt with methodological issues, such as the supposed added value of a mixed methods approach (the combination of qualitative and quantitative research strategies), of participatory research (involving adolescents as coresearchers), and of Q-methodology (a specific methodology that combines elements of qualitative and quantitative research).

Part I   Preferences for care

The first part of the thesis explores adolescents' experiences and preferences for health care, and their attitudes toward self-management. *Preferences for health care* reflect what patients think are the most important elements of particular health care services and what they expect from health care providers. *Self-management* refers to the ability to manage the chronic condition (in terms of symptoms, treatment, physical and psychosocial consequences, and life style changes) in day-to-day life – with the aim of achieving optimal quality of life.

Chapter 1 evaluates the feasibility, benefits and limitations of a participatory research project featuring chronically ill adolescents as co-researchers. To learn more about the usefulness of the participatory approach in adolescent health research, nine adolescents with chronic conditions, aged 15–17 years, were invited to become coresearchers. During a disco party held for this purpose, they interviewed each other and 25 fellow patients (12-19 years) about their experiences in the Sophia Children's Hospital. Involving adolescents as coresearchers is not often done, but it proved feasible and was appreciated by research team and the coresearchers alike. Participatory research also produces positive publicity. Nevertheless, it had its drawbacks too. The disco party attracted few participants; the interviews lacked depth and did not yield substantial new insights, while the costs and time investments of the research team and coresearchers were high. Maintaining a high level of participation of the chronically ill coresearchers also proved difficult. We conclude that adolescents with chronic conditions like to have a say in the design and evaluation of hospital services, but that their involvement as coresearchers is not necessarily the most effective and efficient way to achieve more responsive services. Applying other creative and more sustainable ways for involving youth in health care service development and innovation are therefore recommended, because it is important that their voices are being heard.

Chapter 2 highlights chronically ill adolescents' preferences regarding providers' qualities, outpatient and inpatient care. Their suggestions for improvement of service delivery were also collected. The qualitative results of peer interviews during the disco party and face-to-face interviews of adolescents at home were compared with the Top 5 rankings of preferences in order of perceived importance, obtained through a questionnaire (n = 990). Adolescents found it most important to have *'a feeling of trust'* and *'voice and choice'* in the hospital. As they gradually grow out of the pediatric environment, they appreciated staff attitudes to become less childish and more age-appropriate. They welcomed being treated as an equal partner in care. With respect to providers' qualities, they preferred technically competent providers, who are honest, trustworthy and attend to their needs. For outpatient consultations, adolescents were most concerned with getting answers to all their questions and having clear communication, while limited waiting times and attractive outpatient surroundings were less important to them. Regarding hospitalization, adolescents most preferred to *'avoid pain and discomfort'*; *'keep in touch with home'*; *'be entertained'*; while *'being hospitalized with peers'* and *'being heard'* were rated least important. Regarding priorities for improvement, 52% of the respondents felt that more attention should be paid to older children, followed by enabling more contact with family and friends (45%), shorter waiting times (43%), and more activities to meet fellow patients (35%). Health care professionals should inquire into preferences and adjust their communication style accordingly.

Chapter 3 investigates adolescents' preferences and attitudes toward self-management and hospital care delivery. Adolescents with chronic conditions have to learn to self-manage their health in preparation for transitioning to adult care. This aspect was explored in a Q-methodological study that was part of the interview study. The respondents rank-ordered 37 opinion statements and motivated their ranking. Factor analysis served to uncover patterns in the rankings of statements. Four distinct factors were described as so-termed Q-Care Profiles: 'Conscious & Compliant'; 'Backseat Patient'; 'Self-confident & Autonomous'; and 'Worried & Insecure'. The Q-Care Profiles differed in the desired level of independence, involvement with self-management, adherence to therapeutic regimen, and appreciation of the parents' and health care providers' roles. There were also similarities: the desire to participate in treatment-related decisions is important to all preference profiles. Thus, there is no 'one size fits all' approach to adolescent health care but rather a limited number of distinct preference profiles.

Chapter 4: As Q-methodology allows no inferences with respect to the relative distribution of these profiles and the associations with socio-demographic and disease-related variables, the Q-Care Profiles were presented without their names to a large sample of adolescents in the questionnaire. The 990 respondents considerably varied in opinion on their self-management roles (Q-Care Profiles). The 'Conscious & Compliant' profile fitted 56%, appealing more to older and higher educated youth. These experienced better health and felt more confident to manage their condition. Adolescents felt that the 'Backseat Patient' profile fitted least (16%). This was associated with being younger, physically disabled, and lower educated. These adolescents experienced lower quality of life and felt less self-efficacious. The 'Self-confident & Autonomous' profile fitted 26%, but none of the variables considered was significantly associated with fit to this profile. The

'Worried & Insecure' profile fitted 25%, mostly younger, lower educated girls of non Dutch ethnicity. They experienced poor health, lower quality of life, and felt less self-efficacious.

Use of Q-Care Profiles in clinical care seems promising, as they catch several important concepts in one short description and they distinguish well between different self-management attitudes. The Q-Care Profiles may help identify risky behaviors. 'Backseat Patients' demonstrate lack of independence, 'Worried & Insecure' persons risk depression, while 'Self-confident & Autonomous' adolescents may be fail to adhere to prescribed treatment. Further research should elaborate on the predictive value of Q-Care Profiles to assess nonadherence or lack of independence.

Part II   Competencies for care

On the road to adulthood adolescents are expected to develop the knowledge, attitude, and skills (*competencies*) required to successfully perform developmental tasks or to take up new challenges, such as self-management of their condition. Planning for future health care will involve preparing for the transition from pediatric care to adult care, as well as learning new skills for an independent role as health care consumer.

Chapter 5 discusses adolescents' preferences and competencies for health communication during triadic hospital consultations (consultations jointly with the patient, the health care professional, and one or both parents). We integrated data from the qualitative interviews at home, the Q-Care Profiles, the observations of outpatient consultations and of the focus groups with health care providers, and from the questionnaire. Adolescents' preferences regarding (independent) health communication varied, but all adolescents wished to be involved as partners in care. Yet, their actual participation during consultations was low. They often acted as bystanders rather than main characters because their participation was neither requested nor encouraged. Parents filled the gap, to healthcare providers' frustration. Adolescents' lack of involvement seemed co-constructed by all parties and was surrounded with ambivalence. Most explained their marginalized position as a result of their own indifference or as a consequence of *"not being asked to participate".* A minority saw themselves as incompetent as they were *"still a child"*, while others were not happy with being left out of communication and being *"treated in a childish way"*. Adolescents were ambivalent about the parents' role: while they needed their parents and often appreciated their support, they also felt not at ease when parents interfered. Pediatric staff was equally ambivalent: they tried to involve adolescents, while not restricting parents' presence or dominance of the communication.

Findings from the questionnaire (*n* = 960) confirmed the gap between feeling competent to manage one's own affairs during consultation and the actual presence of independent behaviors (such as asking questions, being involved in treatment decisions, going into the consultation room on your own). The older ones, the girls, the higher-educated, the more experienced in terms of hospital visits, and those who felt more self-efficacious, reported to be more active during consultations. We conclude that triadic communication appears to be all but multi-party-talk. As chronically ill adolescents need to prepare themselves for transition to adult care, health care

providers should encourage them to take the lead in communication by initiating independent (unaccompanied) visits and changing the parents' roles.

Chapter 6 explores the associations between adolescents' perception of their readiness to transfer to adult care and socio-demographic and disease-related characteristics, effect of the condition, self-management ability, and attitude toward transition. Of the 954 adolescents participating in this study, a slight majority (56%) felt ready for transfer. Feeling more self-efficacious in skills for independent hospital visits and a greater perceived independence during consultations were most strongly associated with being ready to transfer. Higher transfer readiness was associated with older age, but age did not prove to be the most important explanation. Adolescents with a more positive attitude toward transition and those who reported more discussions about future transfer also felt more ready. Disease-related factors and effect of the condition, including quality of life, were only weakly associated with transfer readiness. Since attitude to transition and level of self-efficacy in managing self-care seem to be the keystones to feeling ready for transfer, strengthening adolescents' independence and self-management competencies, combined with early preparation and repeated discussions on transition during consultations, seem to be useful strategies to increase adolescents' readiness for transfer to adult care.

Chapter 7 describes the development, testing, and validation of the On Your Own Feet Self-Efficacy Scale (OYOF-SES). This newly constructed generic instrument measures self-efficacy using a 4-point Likert response format on three generic domains: (1) coping with the condition; (2) knowledge of the condition; and (3) skills for communication during hospital consultations. The instrument was constructed from existing disease-specific self-efficacy instruments, the results of the thematic analysis of the interview data, and the experiences with the test version. It was presented to a sample of 958 adolescents with a variety of chronic conditions, and its validity and reliability were examined. On the basis of item-reduction analysis, a short 17-item version of the OYOF-SES was developed. The psychometric properties of the final scale were satisfactory, rendering the OYOF-SES a valid and reliable instrument for assessing self-efficacy in adolescents with chronic conditions. Statistical analyses demonstrated the predictive validity of the instrument for health-related quality of life and independent behaviors during hospital consultations. Adolescents give high ratings to their self-efficacy. Self-efficacy increases with age but age is not the only important factor. Although the differences are small, boys score their self-efficacy higher than girls. Also those with a higher educational level and those without physical disabilities have higher OYOF-SES scores. Further testing of responsiveness to change should indicate whether this is a useful tool for guiding nursing interventions to enhance adolescents' self-efficacy in order to support self-management.

Chapter 8 compares adolescent self-reports of their Health-Related Quality of Life with parent (proxy) reports. This is measured with two validated instruments, KIDSCREEN-10 and Disabkids (DCGM-10). This comparison is thought to give an indication of adolescents' capacity to judge their quality of life. Both the extent and the direction of (dis)agreement on quality of life were studied in 584 adolescent-parent pairs. Both parents and the adolescents themselves rated the adolescents'

quality of life as high, even compared to European norm data (healthy peers) and other chronically ill populations. Differences between adolescents and parents were statistically significant, yet relatively small. In 43% (KIDSCREEN-10) and 51% (DCGM-10) of the adolescent and parent pairs, both agreed on the adolescent's HRQoL. Most disagreement tended to be minor. Disagreement existed in both directions: in 25% (KIDSCREEN-10) and 17% DCGM-10) of cases, adolescents rated their HRQoL lower than did their parents, while in 32% (KIDSCREEN-10) and 32% (DCGM-10) of the cases the opposite was true. Adolescent's age, educational level and type of education, parent's educational level, number of hospital admissions and several other disease-related factors influenced direction of disagreement. We concluded that the *proxy problem* (implying that parents and children strongly differ in their opinions about the child's HRQoL) may be smaller than is sometimes presumed in the literature. As adolescents are expected to become partners in their own health care, it is recommended to focus on the adolescent's own perceptions of HRQoL.

Part III   Transitional care: the need for action

Chapter 9 reports on a qualitative study that aimed to map the experiences with the transfer to adult care of young adults with chronic conditions. We also collected recommendations for improving transitional care from the perspectives of young adults, their parents and health care providers. This study was conducted (2004-2007) in seven different diagnostic groups: hemophilia, diabetes mellitus, spina bifida, congenital heart disorders, cystic fibrosis, juvenile rheumatoid arthritis, or sickle cell disease. Semi-structured interviews were performed with 24 young adults after transfer (aged 15-22), 24 parents, and 17 health care providers. Most patients had not been prepared for transition. Experiences and views of patients, parents and professionals mainly overlapped and were condensed into four core themes. The first theme is *'leaving pediatric care is a logical step'*. Although all parties agree about this, leaving familiar surroundings frequently produces insecurity and feelings of loss. Leaving pediatric care was harder for parents than for young adults, who more often displayed a positive *'wait-and-see'* attitude. The second theme is *'transition is complicated by cultural gaps between pediatric and adult services'*: young adults and parents felt lost after transfer and recommended their peers *'to be alert and involved'*. Providers also recognized the cultural chasm between both services and worried about nonadherence, loss to follow-up, and lack of independence. The two other themes indicated priorities for improvement: *'better patient and parent preparation'* for differences between health care settings and for new roles and responsibilities with respect to self-management; and *'more collaboration and personal links'* between pediatric and adult care providers.

To cross the chasm between pediatric and adult-oriented care, preparation for transition should start early and focus on strengthening adolescents' independency without undermining parental involvement. Building bridges between services, gaining trust and investing in new personal relations is a challenge for all parties involved: transition is about responding and bonding.

Chapter 10 looks at the challenge to ameliorate transitional care in the Netherlands in a sample of health care professionals involved with adolescents (aged 12 - 25) with chronic somatic condi-

tions. In 2008, 115 professionals (48% pediatricians) from (pediatric) hospitals filled out a web-questionnaire and 8 additional telephone interviews were conducted. This research aimed at gaining insight into current transitional practices and into the felt necessity for improvement of adolescent care. Pediatric health care professionals saw the importance of not only addressing medical issues during consultations, but also the social position of these adolescents. Preparation for transition of care and the promotion of more independence of young people was taken up by most of the teams, but more structural interventions were rare. Essential elements of a transition program, such as transition protocols, individual action plans, and the use of a transition coordinator or transition clinic, were covered by only a minority of the teams. Over 85% of the respondents gave high priority to improving self-management, adherence and self-reliance in their population. We noticed a discrepancy between the priorities of health care professionals and the way in which transition of care is being practiced at the moment in the Netherlands. Promotion of self-management and social participation are important areas for improvement. The growing number of young people with chronic conditions necessitates better organization of transition of care and more attention toward self-management.

To conclude...

Adolescents with different chronic conditions all experience the same challenges in coping with the condition and in their transition to adulthood. This justifies a generic approach to transitional care program development. The rising number of chronically ill youth in the Netherlands underpins the importance of a smooth transition to adult care and a broader perspective on adolescent health care. Adolescents with chronic conditions should be trained in self-management and efforts should be made to enhance their social participation. To provide a safe transition is no luxury, but a necessity!

Seven recommendations for the health care practice

1. Enquire into young people's preferences, involve them in their own care and respond to their needs and concerns;
2. Make sure that parents stay involved while focusing on the young person's perspective;
3. See adolescents alone for (part of) the consultation as this increases their competencies and enhances their involvement in care;
4. Adapt health communication and education to the different self-management styles represented in the Q-Care Profiles;
5. Good adolescent health care does not only include medical issues, but also addresses the psychological, social and educational needs of adolescents;
6. Timely prepare adolescents and their parents for the transfer, and bridge the cultural gap between pediatric and adult care: a smooth transition is a joint responsibility;
7. Apply a generic approach when feasible, a disease-specific approach when necessary.

Appendices

List of abbreviations

ADOL HIGH	group of cases in which parents underestimate their child's HRQoL compared with the adolescent's rating
ADOL LOW	group of cases in which parents overestimate their child's HRQoL compared with the adolescent's rating
AGREE	group of cases in which adolescents and their parents agree about the adolescent's HRQoL
ALL	Acute Lymphoblastic Leukemia
CBO	Kwaliteitsinstituut voor de gezondheidszorg; CBO bv *(Dutch Institute for Healthcare Improvement CBO)*
CBS	Centraal Bureau voor de Statistiek *(Statistics Netherlands)*
CF	Cystic Fibrosis *or* mucoviscidosis
CHD	Congenital Heart Disease
CHRTW	California Healthy & Ready To Work (transition assessment tool)
CI	95% Confidence Interval (in statistical significance testing)
CP	Cerebral Palsy
CSHCN	Children with Special Health Care Needs
DB	electronic hospital database
DCGM-10	DISABKIDS Chronic Generic Module (short form)
df	degrees of freedom (in statistics)
DM1	Diabetes Mellitus type 1
DMD	Duchenne Muscular Dystrophy
eg,	*exempli gratia (L);* meaning "for example"
et al	*et alii (L);* meaning "and others"
HIV	Human Immunodeficiency Virus
HP	hemophilia
HRQoL	Health Related Quality of Life
IBD	Inflammatory Bowel Disease
ICC	Intraclass Correlation Coefficient (in statistics)
ICF	International Classification of Functioning
ICD-9 / -10	International Statistical Classification of Diseases and Related Health Problems – 9th or 10th revision
ie,	*id est (L);* "that is"
JRA	Juvenile Rheumatoid Arthritis
MMR	mixed methods research; ie, the combination of qualitative and quantitative research approaches in one single study
Nivel	Nederlands instituut onderzoek van de gezondheidszorg *(Netherlands Institute for Health Services Research)*
n	number
No.	number
ns	nonsignificant (in statistical significance testing)

NVK	Nederlandse Vereniging voor Kinderartsen *(Dutch Pediatrics Association)*
OR	Odds Ratio
OYOF	On Your Own Feet
P	*p*-value (in statistical significance testing)
PedsQL	Pediatric Quality of Life Inventory
PR	Participatory Research
PROs	Patient Reported Outcomes
QCPs	Q-Care Profiles
QoL	Quality of Life
Quest	questionnaire
SB	Spina Bifida *or* meningomyelocele
SCD	Sickle Cell Disease
SCP	Sociaal en Cultureel Planbureau *(The Netherlands Institute for Social Research)*
SD	Standard Deviation (in statistics)
SES	Socio-Economic Status
TR	Transfer Readiness
TS	Turner Syndrome
VIF	Variance Inflation Factor (in statistics)
vs.	versus
WGBO	Wet Geneeskundige Behandel Overeenkomst (1995) (*Dutch Medical Treatment Act*)
WHO	World Health Organization

List of co-authors and affiliations PhD Thesis AL van Staa

Co-author	Affiliation
N. Job A. van Exel PhD	Erasmus University Rotterdam Institute of Health Policy & Management
Annelies E. Eysink Smeets-van de Burgt MSc RN	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
Sander R. Hilberink MSc	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
	Erasmus MC University Medical Center; Rotterdam Department of Rehabilitation Medicine & Physiotherapy
Susan Jedeloo PhD	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
Jos M. Latour PhD RN	Erasmus MC University Medical Center; Rotterdam Department of Pediatrics, Sophia Children's Hospital
Jetty van Meeteren PhD MD	Erasmus MC University Medical Center; Rotterdam Department of Rehabilitation Medicine & Physiotherapy
Henriëtte A. Moll PhD MD	Erasmus MC University Medical Center; Rotterdam Department of Pediatrics, Sophia Children's Hospital
On Your Own Feet Research Group	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
	Erasmus MC University Medical Center; Rotterdam Department of Pediatrics, Sophia Children's Hospital
Jane N.T. Sattoe MSc	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
	Erasmus University Rotterdam Institute of Health Policy & Management
Heleen A. van der Stege PhD	Rotterdam University, university of applied sciences Expertise Center Transitions of Care
Mathilde M.H. Strating PhD	Erasmus University Rotterdam Institute of Health Policy & Management
Margo J. Trappenburg PhD	Erasmus University Rotterdam Institute of Health Policy & Management
	Utrecht University Utrecht School of Governance

List of Publications AL van Staa 2004-2012

Articles marked with an asterix (*) relate to the work described in this thesis, but are not included here.

Peer reviewed journal or book publications

Helder O, Verweij JCM, van Staa AL. Transition from Neonatal Intensive Care Unit to Special Care Nurseries: Experiences of parents and nurses. *Pediatric Critical Care Medicine*. In press.

* Sonneveld HM, Strating MMH, van Staa AL, Nieboer AP. Gaps in transitional care: What are the perceptions of adolescents, parents and providers? *Child: Care Health Development.* In press.

* van Huijzen, SS, van Staa AL. Social participation and chronic ventilation: experiences of adults with neuromuscular disorders. *Scandinavian Journal of Occupational Therapy.* Accepted.

* Bindels-de Heus GCB, van Staa AL, van Vliet I, Ewals FVPM, Hilberink SR. Transferring young persons with profound intellectual and multiple disabilities from paediatric to adult medical care: Parents' experiences and recommendations. Submitted.

2011

* van der Stege HA, van Staa AL, Hilberink SR, Visser APh. Het gebruik van het nieuwe bordspel SeCZ TaLK om de communicatie over seksualiteit te stimuleren bij jongeren met een chronische aandoening. *Wetenschappelijk Tijdschrift voor Ergotherapie.* 2011;4(1):21-37.

2010

* van Staa AL, Eysink Smeets-van de Burgt AE, van der Stege HA, Hilberink SR. Transitie in zorg van jongeren met chronische aandoeningen in Nederland nog onder de maat. *Tijdschrift voor Kindergeneeskunde.* 2010;78(6):227-236.

* van der Stege HA, van Staa AL, Hilberink SR, Visser APh. Using the new board game SeCZ TaLK to stimulate the communication on sexual health for adolescents with chronic conditions. *Patient Education Counseling. Special Issue Reproductive Health.* 2010;81(3):324-331.

* van Staa AL. [Book review]. Managing transition. Support for individuals at key point of change. *International Journal of Integrated Care.* 2010;10(March). http://www.ijic.org/index.php/ijic/article/view/515/1027.

Evers JC, van Staa AL. Qualitative Analysis in Case Study. In: Albert Mills, Gabrielle Durepos, Elden Wiebe (eds.): *Encyclopedia of Case Study Research;* Part 2, pp.749-757. Thousand Oaks (CA): Sage Publications; 2010. ISBN: 978 14 1295 670 3.

2009

Tielen M, Jedeloo S, van Staa AL, Weimar W. Immunosuppressive Drugs and Young Adults: A difficult combination. In: Weimar W, Bos MA, van Busschbach JJ (eds.): *Organ Transplantation: Ethical, Legal and Psychosocial Aspects*. *Towards a Common European Policy;* pp. 563-569*. Lengerich (Germany): PABST Science Publishers; 2009. IBSN 978-3-89967-415-6.

2008

Tielen M, van Staa AL, Jedeloo S, van Exel NJA, Weimar W. Q-methodology to identify young adult renal transplant recipients at risk for non-adherence. *Transplantation,* 2008;85(5):700-706.

2006

Faber E, Burdorf A, van Staa AL, Miedema HS, Verhaar JAN. Qualitative evaluation of a form for standardized information exchange between orthopedic surgeons and occupational physicians. *BMC Health Services Research*. 2006;6:144:1-8

2004

* Verhoeven-Wind L, van Staa AL. Vanzelfsprekend, maar niet vanzelf. Transitie in zorg: ervaringen van adolescenten met Cystic Fibrosis en hun ouders. *Verpleegkunde* 2006;20(3):186-195.
de Lange J, van Staa AL. Transities in ziekte en zorg; op zoek naar een nieuw evenwicht; lectorale rede. *Verpleegkunde.* 2004;19:42-50.

2000

van Staa AL, Visser APh, van der Zouwe N. Caring for caregivers: experiences and evaluation of interventions for a palliative care team. *Patient Education Counseling.* 2000;41(1):93-105.

Professional journal or book publications

2010

van Staa AL, Evers JC. 'Thick analysis': strategie om de kwaliteit van kwalitatieve data-analyse te verhogen. *KWALON.* 2010;15(1):5-12.
Evers JC, van Staa AL. Repliek: Niet eenvoudiger, wel creatiever en efficiënter. *KWALON.* 2010;15(1):17-19.

Adams S, van Staa AL. Ten geleide. Internet & kwalitatief onderzoek. *KWALON.* 2010;15(2):3-11.

2009

Jedeloo S, van Staa AL. Q-methodologie, een werkelijke mix van kwalitatief en kwantitatief onderzoek? *KWALON.* 2009;14(2):5-15. Dupliek: 19-20.

Jedeloo S, Janssen I, van Staa AL. Doorbraak in transitiezorg: Actieprogramma op Eigen Benen Vooruit! Van onderzoek naar zorgvernieuwing voor jongeren met chronische aandoeningen. *Kwaliteit in Zorg.* 2009;12(2):10-13.

* van Staa AL. Snakes & Ladders: Hoe meer participatie, hoe beter? *KWALON.* 2009;14(1): 15-21.

* van Staa AL. Van zorgstandaard naar zorgpraktijk: goede adolescentenzorg in de transitiefase. *DiabeteSpecialist.* 2009;32:24-25.

2008

van Staa AL, Steultjens E. Systematische Reviews. In: K Cox, D de Louw, J Verhoef & CHZ Kuiper. *Evidence-based practice voor verpleegkundigen. Methodiek en implementatie*; pp. 175-193. 2e herz. druk. Utrecht: Lemma; 2008. ISBN 978 90 5931 226 5.

Steultjens E, van Staa AL. Systematische Reviews. In: CHZ Kuiper, J Verhoef, D de Louw & K Cox. *Evidence-based practice voor paramedici. Methodiek en implementatie*; pp. 169-187. 2e herz. druk. Utrecht: Lemma; 2008. ISBN 978 90 5931 221 0.

* van Staa AL, Hilberink SR, Eysink Smeets-van de Burgt AE, van der Stege HA, Jedeloo S, Roebroeck ME. Transitie van kinderzorg naar volwassenenzorg: Revalidatie in actie. *Revalidata.* 2008;146: 3-10.

2007

Borgdorff H, van Staa AL, Vos J. Kennis in context. Onderzoek aan hogescholen. *Th&ma, Tijdschrift voor Hoger Onderwijs & Management.* 2007;14(5):10-17.

* van Staa AL, Kuijper M, Jedeloo S, Latour JM. Wat doen we met de ouders? *Kind en Ziekenhuis.* 2007;30(2): 49-51.

* van Staa AL, Kuijper M, Jedeloo S, Latour JM. Participatie van jongeren in de spreekkamer. Hoe kunnen kinderverpleegkundigen jongeren met chronische aandoeningen activeren? *Tijdschrift Kinderverpleging.* 2007;13(1):10-13.

van Staa AL. Onderzoek hoort óók bij hbo. *Transfer.* 2007;juli:15.

* van Staa AL. Jongeren doen zorgonderzoek in de disco. *Mediator. ZonMw bericht over onderzoek en ontwikkeling.* 2007;18(8):30-32.

2006

* van Staa AL. Gewoon als ieder ander: levensverhalen met jongeren met chronische aandoeningen. In: E. Bohlmeijer, & L. Mies. *De betekenis van levensverhalen*. Bohn Stafleu Van Loghum, Houten; 2006. ISBN: 978 90 3134 874 9.

Martina S, van Staa AL. Een volwassen leven met Congenitale Anorectale Misvorming. *ErVaring van de Vereniging Anusatresie (VA).* 2006;21(juni).

ter Maten-Speksnijder A, van Staa AL. Met de patiënt in gesprek over vermoeidheid. Een onderzoek door studenten HBO-V binnen de Kenniskring Transities in Zorg samen met afdelingsverpleegkundigen in Erasmus MC. *Tijdschrift voor Verpleegkundigen TvZ.* 2006;116(1):45-48.

de Pree C, van Staa AL. Segregatie van kinderen met cystic fibrosis in het ziekenhuis. Markeren of niet. *Tijdschrift Kinderverpleegkunde.* 2006;12(3):4-7.

* van Staa AL. Transitie in zorg. Een hele overstap. *Kind & Ziekenhuis.* 2006;28(1):4-10.

* van Staa AL Transitie in zorg. Een hele overstap. *Nieuwsbrief sectie Revalidatie NIP.* 2006;20(49):7-8.

2005

van Staa AL. De toekomstverwachting van… *Verpleegkunde.* 2005;20(4):249-250.

van Staa AL. Actueel: "De komst van hoogleraren en lectoren heeft wel bijgedragen aan de professionalisering van verpleegkunde, maar niet aan de stand van de wetenschap". *Verpleegkunde.* 2005;20(4):220.

2004

de Lange J & van Staa AL. *Transities in ziekte en zorg; op zoek naar een nieuw evenwicht*. Lectorale Rede. Rotterdam, Hogeschool Rotterdam; 2004 2nd. Ed. ISBN: 90 8017 762 8.

Steultjens E, van Staa AL. Systematische Reviews. In: CHZ Kuiper, J Verhoef, D de Louw & K Cox *Evidence-based practice voor paramedici. Methodiek en implementatie*; pp. 153-170. Utrecht: Uitgeverij Lemma; 2004. ISBN 90 5931 210 4.

van Staa AL, Steultjens E. Systematische Reviews. In: K Cox, D de Louw, J Verhoef & CHZ Kuiper *Evidence-based practice voor verpleegkundigen. Methodiek en implementatie*; pp. 153-170. Utrecht: Uitgeverij Lemma; 2004. ISBN 90 5931 211 2.

* van Staa AL. Transitiezorg is meer dan een goede transfer. *Tijdschrift voor Verpleegkundigen TvZ.* 2004; 114(6):18-23.

* van Staa AL. Te groot voor het servet, te klein voor het tafellaken? De uitdaging voor kinderverpleegkundigen bij transitie in zorg voor jongeren met chronische aandoeningen. *Tijdschrift Kinderverpleegkunde.* 2004;8(4):4-6.

van Staa AL, de Lange J. Onderzoek in de Bachelor: Niet laten maar doen! *Tijdschrift voor Verpleegkundigen TvZ.* 2004;114(10):27-29.

Popular journal or book publications

2009

* van Lieshout A, van Staa AL. Volwassen worden met de ziekte van Hirschsprung: sluit de zorg aan op de behoeften? *Drukwerk (Tijdschrift Vereniging Ziekte van Hirschsprung).* 2009;21(3):7-9.

2008

* van Staa AL, van der Stege HA, Jedeloo S. *Op Eigen Benen Verder. Jongeren met chronische aandoeningen op weg naar zelfstandigheid in de zorg. [On Your Own Feet Ahead. Young people with chronic conditions on their way to independence in health care]*. Rotterdam: Hogeschool Rotterdam; 2008. ISBN 978 90 79059 02 7.
* van Staa AL, van Huijzen SS. Transitie in zorg bij anusatresiepatiënten. Aan de overgang van kinder- naar volwassenenzorg valt nog veel te verbeteren. *ErVaring, Tijdschrift Vereniging Anusatresie.* 2008;(December):7-8.

2007

* van Staa AL, Jedeloo S, Kuijper M, Latour JM. *Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? [On Your Own Feet. Young people with chronic conditions: what are their preferences and competencies for health care?]*. Rotterdam: Hogeschool Rotterdam; 2007. ISBN 978-90-801776-9-7.
* van Huijzen SS, van Staa AL, Langemeijer RATM. Een volwassen leven met CARM (anusatresie). Het vervolg… *ErVaring, Tijdschrift Vereniging Anusatresie.* 2007;(Maart):16-19.
* van Staa AL, Kuijper M, Jedeloo S, Latour JM. Voorbereiding op de volwassenenzorg: wat willen en kunnen jongeren. *BOSK magazine.* 2007;2:34.
* van Staa AL. "Jij en het Sophia": hoe kan de zorg in het Erasmus MC – Sophia nog beter worden?. *ZieSo, blad voor patiënten en ouders in het Erasmus MC – Sophia.* 2007;23(1):16-17.

List of Presentations AL van Staa 2004-2012

Only presentations that relate to work described in this thesis and that were performed by the author, are listed here.

2012

Jan 13   **International Pediatric Advanced Nursing Practice (ANP) Conference** «Challenges in Pediatric Care: Innovations through Advanced Nursing Practice»; Basel (Switzerland) - 'On Your Own Feet Ahead: Nurse-led innovations in Dutch transitional care for adolescents with chronic conditions'. *Poster presentation.*

Mar 8   **Periodieke Conferentie Sociale Pediatrie West Nederland**; Rotterdam. - 'Op Eigen Benen Vooruit! Naar een betere transitie voor jongeren met chronische aandoeningen zoals astma, diabetes en ADHD'. *Keynote speaker.*

Mar 23   **Update@kempenhaeghe.nl. 14th Annual Clinical Symposium Kempenhaeghe**; Heeze. - 'Transition into adulthood for youth with disabilities'. *Keynote speaker.*

Mar 30   **Nederlandse Diabetes Dagen**; Den Haag. - 'Op Eigen Benen Vooruit Diabetes'. *Poster presentation*

2011

Jan 19   **Kwaliteitsconferentie Regionaal Genootschap Fysiotherapie**; Maassluis. - 'Jongeren met chronische aandoeningen'. *Oral presentation.*

Jan 24   **Periodieke Conferentie Kindergeneeskunde Oost Nederland**; Nijmegen. - 'Het belang van transitie: een nieuw aspect in de zorg'. *Keynote speaker.*

Mar 30   **Nederlandse Nefrologie Dagen**; Veldhoven. - 'Op Eigen Benen Vooruit! nieuwe interventies voor ondersteuning zelfmanagement van jongeren met chronische nierziekten'. *Oral presentation*

Apr 12   **Symposium 'Uit de kinderschoenen' Nierstichting**; Zeist. - 'Op Eigen Benen Vooruit! Naar een betere transitie voor jongeren met nierziekten'. *Keynote speaker.*

May 10   **Erasmus University Rotterdam; HSMO meeting. iBMG**; Rotterdam. - 'Q-Care Profiles. Development and distribution'. *Oral presentation*

Jun 20-21   **16th European International Association Adolescent Health meeting**; Copenhagen. - 'Parents of adolescents and young adults with severe motor and intellectual disability - experiences with (the preparation for) the transfer to adult care'. *Oral presentation* - 'On Your Own Feet Ahead - participation in transition'. *Keynote speaker.* - 'On Your Own Feet Ahead: Innovations in Dutch Transitional Care for Adolescents with Chronic Conditions'. *Poster presentation.*

Oct 11	**Projectleidersdag Programma Jeugd: Participatie ZonMw**; Utrecht.
	- 'Snakes & Ladders: ervaringen met participatief onderzoek in Op Eigen Benen'. *Keynote speaker.*
Oct 11	**Symposium Proud2Be a Nurse, VIP² Erasmus MC**; Rotterdam.
	- 'Proud To Be A Nurse: de verpleegkundige als SPIL voor zelfmanagement'. *Keynote speaker.*
Nov 14	**Naar verankering van actie. Congres Nationaal Actieprogramma Diabetes**: Den Haag.
	- 'Op Eigen Benen Vooruit Diabetes'. *Poster presentation.*
Nov 17	**Symposium Op Eigen Kracht. Vereniging Spierziekten Nederland**; Baarn.
	- 'Afhankelijk én autonoom: transitie bij jongeren met NMA'. *Keynote speaker.*

2010

Mar 10	**Q & coffee meeting**; Rotterdam.
	- 'Combining Q and R, Dilemmas and challenges'. *Oral presentation.*
Apr 22	**"Samen nog beter" Congres voor kinderartsen & kinderpsychologen**; Zeist.
	- 'Jongeren met chronische aandoeningen: Weeskinderen van de zorg? Volwassenen van de toekomst!' *Keynote speaker.*
May 18	**Sigma Theta Tau - Rho Chi Chapter Meeting**; Rotterdam
	- 'Jongeren met chronische aandoeningen: Weeskinderen van de zorg? Volwassenen van de toekomst!' *Keynote speaker.*
May 19	**Expertmeeting Transitienet**; Eindhoven.
	-'Een praktische kijk op zorginnovatie'. *Oral presentation.*
Jun 4	**Lustrumcongres Nederlandse Vereniging Kinderfysiotherapie**; Zwolle.
	- 'Jongeren met chronische aandoeningen: Weeskinderen van de zorg? Volwassenen van de toekomst!' *Keynote speaker.*
Sep 7-10	**International Conference on Communication in Health Care EACH 2010**; Verona.
	- 'Activate! Tools and tricks to increase adolescents' participation during medical consultations'. *Workshop.*
	- 'From passive bystander to active partner in communication: skills and roles of adolescents with chronic conditions during medical consultations'. *Poster presentation.*
	- 'Let's talk about sex: research and development of the new board game for adolescents with chronic conditions: SeCZ Talk**'.** *Poster presentation.*
Sep 25	**Congres Zeehospitium**; de Haan België.
	- 'Jongeren met chronische aandoeningen: Weeskinderen van de zorg? Volwassenen van de toekomst!' *Keynote speaker.*
	- 'Jongeren Activeren'. *2 Workshops.*
Oct 5	**Refereerbijeenkomst Psychosociale afdeling; Emma Kinderziekenhuis Amsterdam**. *Oral Presentation.*

Oct 23-26   **EAPS Congress** (The European Society for Paediatric Research (ESPR), the European Academy of Paediatrics (EAP), the European Society of Paediatric and Neonatal Intensive Care (ESPNIC), PNAE (Paediatric Nurses). Copenhagen.

- 'Let's talk about sex: Rationale for a new board game for adolescents with chronic conditions'. *Oral presentation.*

- 'On Your Own Feet Ahead: Innovations in Dutch Transitional Care for Adolescents with Chronic Conditions'. *Poster presentation.*

- Let's talk about sex: Rationale for a new board game for adolescents with chronic conditions. *Poster presentation.*

- 'Q-Care profiles: Development of a new instrument to stimulate communication with adolescents with chronic conditions during consultations'. *Poster presentation.*

Nov 1   **Kennis Delen Kindernefrologie**; Utrecht.

- 'Op Eigen Benen Vooruit: Maak kennis met nieuwe interventies voor betere adolescentenzorg'. *Oral presentation.*

Nov 3-5   **Congres Nederlandse Vereniging Kinderartsen**; Veldhoven.

- 'Overstappen'. *Symposium* Stap voor stap opgroeien met een chronische ziekte: de arts als change agent'. *Oral presentation*.

- 'Let's talk about sex – ontwikkeling en eerste ervaringen met SeCZ TaLK, bordspel voor chronisch zieke jongeren'. *Poster presentation.*

Nov 17   **Symposium "Voor alle leeftijden";** Rijndam Revalidatiecentrum Rotterdam.

-'Let's SeCZ Talk. Aanleiding en ontwikkeling van een nieuw bordspel'. *Keynote speaker.*

Dec 10   **Audit-bijeenkomst Adolescenten**; CF-Centrale (CF-centra Utrecht, Den Haag & Rotterdam) Bodegraven.

- 'Transitie bij CF: Veel gedoe om niets? Of tijd voor actie?' *Keynote speaker.*

2009

Jan 21   **NCFS-Symposium Van Hielprik tot Donorlong: 40 jaar NCFS**; Amersfoort (NL).

- 'Transitie bij CF'. *2 Workshops.*

Jan 22   **Congres Hoofdzaken; Tien Jaar Innovatiefonds Zorgverzekeraars**; Utrecht (NL)

- 'Op Eigen Benen Vooruit'. *Workshop*

- 'Praten met pubers in de spreekkamer'. *Workshop.*

Nov 5-6   **9th Conference International Network Integrated Care (INIC 09)**; Vienna.

- 'Bridging the gap between paediatric and adult care: experiences with transition'. *Oral Presentation.*

- 'From research to innovation: A Breakthrough in transitional care for adolescents with chronic conditions'. *Oral Presentation.*

2008

Mar 17   **Werkgroep Kinderdiabetesverpleegkundigen Bijscholing**; Utrecht.

- 'Een stap in de toekomst' *Keynote speaker.*

Oct 2   **Werkgroep Transitie Erasmus MC**; Rotterdam 'Op Eigen Benen'. *Keynote speaker.*

Dec 11   **Symposium 25-jarig bestaan Tienerafdeling AMC-Emma Zorg voor tieners: kopzorg of topzorg?** *Keynote speaker.*

Dec 12   **5ᵉ KWALON Najaarsconferentie**: Utrecht.

- 'Snakes & Ladders. Een kritische kijk op de participatieladder en participatief onderzoek'. *Keynote speaker.*

Sept 2-5   **13th Research Conference of the Workgroup of European Nurse Researchers (WENR);** Vienna.

- 'Participation of nursing specialist in observations of adolescents with chronic disorders in the outpatient department'. *Oral presentation.*

- 'Participatory research: Adolescents with chronic disorders interviewing fellow patients about health care preferences'. *Oral presentation.*

2007

Apr 19   **Conferentie Klaar? … Over!**; Rotterdam.

- 'Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg?' *Keynote speaker.*

- 'Participatief onderzoek binnen het project Op Eigen Benen'. *Workshop.*

Jun 8   **Grand Round Erasmus MC – Sophia, Rotterdam**.

- 'Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg?' *Key-note speaker.*

Aug 25   **International Congress of Pediatric Nursing**; Athens.

- 'Transition of adolescents with chronic disorders from paediatric care tot adult care: not just a simple transfer'. *Oral presentation.*

- 'Participatory research: Adolescents with chronic disorders interviewing fellow patients about health care preferences'. *Oral presentation.*

Aug 25-30   **25th International Congress of Pediatrics**; Athens.

- 'A normal life with an unhealthy body: self-identity in adolescents growing up with chronic illness'. *Oral presentation.*

- 'Using Q-methodology to explore preferences for care of adolescents with chronic disorders: Four profiles'. *Oral presentation.*

- 'Observations of adolescents with chronic disorders in the surgery bystanders or main characters?' *Poster presentation.*

Sep 29   **Familieweekend Nierpatiëntenvereniging Nederland**; Baarlo.

- 'Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? ' *2 Workshops.*

Nov 9	**Openingscongres Week Chronisch Zieken**; Rotterdam.
	- 'Op Eigen Benen. Jongeren met chronische aandoeningen: wat willen en kunnen zij in de zorg? ' *Oral presentation & Workshop.*
Nov 30	**Vlaams Nederlands Wetenschappelijk Congres Verpleging & Verzorging**; Amsterdam.
	- 'Observaties van jongeren met een chronische aandoening op de polikliniek: van toeschouwer naar hoofdpersoon'. *Oral presentation.*

2006

Jun 7	**Studiemiddag Transitie in de Revalidatie, Vereniging Spierziekten Nederland**; Baarn.
	- 'Te klein voor tafellaken, te groot voor servet? Naar een soepele transitie in zorg'. *Keynote speaker.*
Oct 4	**Minisymposium IBD Transitiepoli**; Erasmus MC – Rotterdam.
	- 'Tien gouden tips bij transitie'. *Keynote speaker.*
Nov 14	**Symposium Week Chronisch Zieken**; Hogeschool Rotterdam.
	- 'Jongeren interviewen jongeren over Erasmus MC – Sophia'. *Keynote speaker.*
Nov 16	**Congres De Ziekte van je Leven**, Vereniging van Kinderverpleegkundigen; Ede.
	- 'Op Eigen Benen'. *Workshop.*

2005

May 12	**11th Qualitative Health Research Conference**; Utrecht.
	- 'Participatory research & evidence: researching adolescents with special health care needs'. *Oral presentation.*
Nov 7	**Symposium Kiezen voor …. Participatie van jongeren met chronische beperkingen. Week Chronisch Zieken**, Hogeschool Rotterdam. *Keynote speaker.*
Dec 2	**Vlaams Nederlands Wetenschappelijk Congres Verpleging & Verzorging**; Leuven.
	- 'Geen moment maar proces: Ervaringen van jongeren bij hun transitie in zorg van Erasmus MC – Sophia naar de volwassenenzorg'. *Oral presentation.*

2004

Nov 4	**Landelijk Congres Vereniging van Kinderverpleegkundigen**; Ede.
	- 'Te klein voor het tafellaken, te groot voor het servet? De uitdaging voor kinderverpleegkundigen bij transitie in zorg'. *Keynote speaker.*
	- 'Transitie van adolescenten naar de volwassenenzorg: geen moment maar proces. Pilotstudy in Erasmus MC – Sophia'. *Workshop / Oral presentation.*

PhD Portfolio Summary

Summary of PhD training and teaching activities

Name PhD student: AnneLoes van Staa	PhD period: 2008-2012
Erasmus University Rotterdam	Promotor(s):
institute Health Policy & Management	Prof.dr. M.T. Trappenburg / Prof.dr. H.A. Moll
Research school: none	Supervisors: none

1. PhD training

	Year	Workload (Hours)
Research skills		
- KWALON: Cursus Kwalitatief Interviewen; Focus Groep Discussies; Kwaliteit van kwalitatief onderzoek; Introductie Atlas.Ti	2004	48
- KWALON Advanced Course Atlas.Ti	2005	24
- Applied Multivariate Statistics – Utrecht University Summer Course	2010	80
Research group		
- Research meetings On Your Own Feet Group	2008 - 2011	250
Presentations on national & international conferences		
Seminars and workshops		
- see List of Publications / Presentations		
Other		
- Peer reviewer for international academic journals (*Journal Adolescent Health* 4x; *Archives Diseases Childhood* 1x; *Child Care Health & Development* 1x; *Pediatric Pulmonology* 1x; BMC Family Practice 1x)	2010 -	48
- Reviewer for (inter)national research bodies (ZonMw 8x)	2009 -	64
- Expert activities on nursing / chronic care / adolescent health care / transitional care	2006 -	100

2. Teaching activities

	Year	Workload (Hours)
Lecturing		
- KWALON: Cursus Kwalitatieve analyse (post graduate)	2007 - 2009	216
- Bachelor & schakelprogramma Gezondheidswetenschappen (iBMG): Sociaal Medische Wetenschappen / Kwalitatief onderzoek	2008 - 2010 - 2011	250
- Erasmus International Master program *Health Information Management* (Socio-cultural context of health and health care)	2004 - 2008	20
- *Master Health Economics, Policy, and Law* (iBMG): Socio-cultural aspects of the use of pharmaceuticals	2010 - 2011	16
- Bachelor Instituut voor Gezondheidszorg (Hogeschool Rotterdam): diverse bijdragen aan minoronderwijs	2009 – 2012	120
- *Master Kinderfysiotherapie*: Kwalitatief onderzoek	2009 - 2011	24
Supervising Master's theses		
- *Master Advanced Nursing Practice* (Rotterdam University)	2005 - 2012	240
- *Master Health Economics, Policy, and Law* (iBMG)	2009 - 2012	240
- *Master Verplegingswetenschappen* (Universiteit Utrecht)	2006 - 2010	120
Other		
- Member of the Internal Audit team Bachelor Gezondheidswetenschappen (iBMG)	2010	24
- Postgraduate education lectures for specialist nurses (diabetes, asthma , CF)	2006 - 2010	60

Dankwoord

Wie wat bewaart die heeft wat (*dit proefschrift*). Promoveren in het midden van je carrière in plaats van aan het begin heeft vele voordelen. Het eerste voordeel was dat ik kon profiteren van een overdaad aan goede voorbeelden. Talloze collega's gingen mij voor en ik heb schaamteloos gebruik gemaakt van al hun goede en slechte ervaringen. Dank dus aan (oud) collega's (in alfabetische volgorde): *Jos Aarts, Luuk Arends, Hester van de Bovenkamp, Werner Brouwer, Jolanda Dwarswaard, Job van Exel, Isabelle Fabbricotti, Jan Kees Helderman, Xander Koolman, Chris Kuiper, Jacomine de Lange, Arjen Stoop, Jeroen van Wijngaarden*, en nog vele anderen. Van hen heb ik geleerd wat te doen en te vermijden.

Het tweede voordeel was, dat ik het promoveren kon beschouwen als een feestje in plaats van als noeste, vaak frustrerende arbeid. Immers, mijn zelfvertrouwen noch mijn twee vaste aanstellingen hingen er van af. Toch had ik niet verwacht dat ik het zo leuk zou vinden en dat ik zo weinig dieptepunten zou beleven. Een zeker talent voor verdringing, een overdaad aan zelfverzekerdheid en een fantastisch team hebben mij daarbij zeker geholpen.

Het derde voordeel was dat ik kon putten uit jarenlange ervaring als docent en onderzoeker. Ondanks mijn niet-gepromoveerde status, bij aanvang van mijn promotietraject al een informele status als 'senior' en als formele leider van een onderzoeksteam had veroverd. Dit promotietraject heb ik, ironisch genoeg, niet aan de universiteit, maar op de eerste plaats aan het hoger beroepsonderwijs te danken. Het feit dat ik in 2003 samen met *Jacomine de Lange* lector Transities in Zorg aan de Hogeschool Rotterdam werd, heeft niet alleen vleugels gegeven aan mijn carrière, maar ook dit proefschrift mogelijk gemaakt. In die beginjaren kregen wij als lectoren alle ruimte om een eigenwijze invulling aan onderzoek in het hbo te geven. Wij hebben die uitdaging voortvarend opgepakt en laten zien dat praktijkgericht onderzoek productief en succesvol kan zijn. Jammer dat de pioniersjaren voorbij zijn en het vertrouwen in professionals plaats lijkt te maken voor controle en beheersing. *Jasper Tuytel*, scheidend voorzitter van het College van Bestuur van de Hogeschool Rotterdam, dank ik voor het feit dat hij mij als lastige lector heeft gedoogd en mij meer ruimte gaf dan hij eigenlijk verantwoord vond.

Het vierde voordeel was dat ik in mijn promotietraject zélf nooit last heb gehad van inperking van mijn professionele ruimte: ik heb altijd het gevoel gehad dat ik zelf de richting en het tempo mocht bepalen. Waarschijnlijk is dit op de eerste plaats te danken aan de intelligente evenwichtskunst van mijn promotoren *Margo Trappenburg* en *Henriëtte Moll* die mij niet alleen veel vrijheid hebben gegund, maar ook op het juiste moment wisten af te remmen, aan te moedigen en bij te sturen. Dankzij hen heb ik mij tijdens mijn promotieonderzoek echt "op eigen benen" gevoeld. Ook mijn leidinggevenden *Roland Bal* (bij iBMG), eerst *Clazina Pool* en later *Marleen Goumans* (bij de HR) gaven mij naast ruimte ook aanmoediging om te werken aan dit proefschrift. Ik hoop dat ik jullie vertrouwen (en geduld) niet alleen op de proef heb gesteld, maar ook heb verdiend.

Toch kleefden er ook nadelen aan promoveren op middelbare leeftijd. Sommigen zagen in een jarenlange ongedoctoreerde staat het bewijs van mijn beperkte academische kwaliteiten. Mijn voormalige baas *Marc Berg* uitte zijn oprechte twijfels aan mijn promoveerbaarheid. Dit heeft mij zeker geprikkeld nog harder mijn best te doen. Ook het feit dat mijn proefschrift de afgelopen jaren voortdurend moest concurreren met andere taken, verantwoordelijkheden en plichten die

je als lector, projectleider, docent, vriendin, echtgenote én moeder nu eenmaal hebt, maakte dat ik er meestal minder tijd aan kon besteden dan gewenst. Dat chronisch tijdgebrek bleek overigens een probaat middel voor hoge productiviteit en tegen verveling. Leven zonder televisie blijkt bovendien de noodzakelijke innerlijke rust en distantie tot de opgewonden buitenwereld te geven.

Tijdens de vijf jaar die ik aan dit proefschrift heb besteed, stond ik niet alleen, maar was ik omringd door velen. Voor de morele steun gaat dank uit naar mijn familie en persoonlijke vrienden, die intens meeleefden en voor lief hebben genomen dat ik niet alleen 's avonds en in het weekend, maar ook tijdens de heilige zomervakantie achter het apparaat kroop of slechts een enkel weekje bij de tent kwam zitten. Gelukkig heb ik een geweldige (schoon)familie, een zeer zorgzame en zelfredzame echtgenoot en hele zelfstandige en verstandige kinderen. Lieve allemaal, we hebben heel wat in te halen. Jammer dat mijn schoonvader *Jan van der Dussen*, die zo betrokken was bij dit alles, mij niet meer op het promotiefeest kan toespreken.

Met goede raad, maar vooral ook door vele praktische daden ben ik verwend door mijn Op Eigen Benen onderzoeksteam. Zonder de inspanningen van mijn paranimfen *Susan Jedeloo en Heleen van der Stege,* naast *Sander Hilberink, Jane Sattoe, Jos Latour, Marijn Kuijper* en *Lausanne Mies*, was dit proefschrift er vast en zeker niet gekomen. In het Sophia Kinderziekenhuis waren *Bert van der Heijden, Wil de Groot* en *Jos Latour* direct te porren voor 'Op Eigen Benen' en hun enthousiasme is overgenomen door veel artsen en verpleegkundigen. Ook jongeren en ouders hebben op allerlei manieren meegewerkt aan het 'Op Eigen Benen' programma en ook de leden van diverse begeleidingscommissies en andere onderzoekspartners droegen hun steentje bij. Samen met diverse collega's van het CBO, in het bijzonder *Jeroen Havers, Ingrid Janssen* en *Hanke Timmermans*, hebben we de afgelopen jaren waarlijk 'iets groots' verricht met het Actieprogramma Op Eigen Benen Vooruit! Als ik ergens trots op ben, dan is het wel daarop! De dertig behandelteams die hebben meegedaan dank ik daarom voor hun enthousiasme en inspiratie. Ook noem ik hier het iBMG-evaluatieteam van het Actieprogramma (*Mathilde Strating, Anna Nieboer, Henk Sonneveld*) waar ook *Marij Roebroeck* (Erasmus MC-Revalidatie) deel van uitmaakt. Tot slot hebben de subsidiegevers, waaronder ZonMw, Kinderpostzegels, Nationaal Actieprogramma Diabetes, Nierstichting, maar vooral het Innovatiefonds Zorgverzekeraars in de persoon van *Ine van Hest*, onze projecten altijd met veel meer dan geld alleen gesteund.

Voor de ondersteuning rond de Op Eigen Benen projecten dank ik de drie *Mariekes* (respectievelijk *van den Heuvel, Vonk* en *Wildenbeest*), *Sanne Wesseling, Ilona van der Lee* en *Kim van Ierssel. Ko Hagoort* en *Mariël Lam* ben ik dankbaar voor het feit dat ik hen op de meest onmogelijke uren en over de hele wereld kon lastig vallen voor respectievelijk Engelse taalcorrectie en de vormgeving van de Op Eigen Benen projecten en dit proefschrift. Daarnaast hebben talloze oudstudenten van de Hogeschool Rotterdam en het instituut Beleid & Management Gezondheidszorg een bijdrage geleverd aan de dataverzameling rondom de Op Eigen Benen projecten en gaven vele collega's van de (voormalige) Kenniskring Transities in Zorg en de sectie Health Care Governance kritische feedback op de analyses, interpretaties en concept artikelen. Tot slot kan de bijdrage van ons secretariaat, *Christi Bakker, Olivia Man Li, Hanneke van der Ploeg* - maar vooral - *Elly Katoen* niet licht worden overschat. *Elly* is zoveel meer dan mijn secretaresse: mijn buurvrouw én mijn steun en toeverlaat tijdens alle Dwaze Dagen die mijn leven nu eenmaal kent.

In de vijf en twintig jaar van mijn carrière, en ruim vijftig jaar van mijn leven zijn er talloze mensen aan wie ik schatplichtig ben, omdat zij mij hebben gevormd, uitgedaagd of geïnspireerd. Ik zou nooit zover in mijn (academische) leven gekomen zijn zonder de bijdrage van vele mensen – docenten, studenten, collega's en samenwerkingspartners - die ik hier in willekeurige volgorde wil noemen - zonder de pretentie volledig te zijn. *Sjaak van der Geest* beschouw ik als mijn mentor in de medische antropologie; *Jacomine de Lange* is mijn voorbeeld én vertrouweling in het hoge-schoolgewoel; *Adriaan Visser* was eerst mijn onderzoeksbegeleider en werd later mijn eigenzinni-ge assistent; *Jos Aarts* is niet alleen mijn kamergenoot maar ook mijn vaste concertmaatje – dank voor al die Triple A ervaringen; *Marij Roebroeck* was tien jaar geleden de enige in Nederland die ook 'iets met transitie en onderzoek deed' maar onze samenwerking (o.a. in TransitieNet en SPIL) heeft er voor gezorgd dat we (allang) niet meer alleen staan. *Hans Nijeboer* zette mij op het spoor van een lectoraat; *Liesbeth Verhoeven-Wind* zorgde ervoor dat ik in 1996 het onderwerp transitie niet los liet na een mislukte NWO aanvraag; *Jeanine Evers* deed mij inzien dat ik beter pas in de empirisch-analytische dan in de hermeneutisch-interpretatieve wetenschapstraditie; *Cees Smit* waardeer ik omdat hij niet alleen zijn ervaringsdeskundigheid tot wapen heeft geslepen, maar ook een bijzonder hartelijk mens is. Bijzondere dank aan de *(oud) collega's* van de Kenniskring Transities in Zorg (toen) en het Kenniscentrum Zorginnovatie (nu) van de Hogeschool Rotterdam voor de uitdagende 'ontwikkelingssamenwerking' die we bedrijven in het hbo. Toch ben ik blij dat ik na al die jaren óók nog steeds een rustige werkplek heb in de academie bij het iBMG (Sociaal Medische Sectie en later de sectie Health Care Governance) waar het nieuwe werken gelukkig nog niet is ingevoerd.

In mijn persoonlijk leven dank ik allereerst *Hans Schoots*, mijn beste en trouwste vriend. In de af-gelopen 35 jaar van mijn woelig persoonlijk leven is hij de meest constante factor gebleken en hij is ook nooit te beroerd bij te springen als oppas, schrijver, toehoorder, steunpilaar en mantelzorger. Ik ben ook blij met de langdurige vriendschap met *Rob Oostvogels, Jan van Duppen, Irene Cornet, Richard Weyndling, Marjolijn Mercx* en *Dick de Korte*. Onmisbaar zijn mijn 'brussen' door hun aan-moediging en steun in barre tijden: mijn immer scherpe en slimme zus *Arian*, mijn hooggeleerde broer *Tjeerd Pieter* met zijn jaloersmakende H-Index, en mijn dappere, vrijgevochten zusje *Jetske* (†). Bijzonder dankbaar ben ik ook voor de aanwezigheid in mijn leven van mijn schoonmoeder *Lida van der Dussen - Quist*, mijn (stief)kinderen *Ruben* en *Stefan, Tommy* en *Pien*: jullie zijn niet alleen de leukste en liefste kinderen van de wereld, maar ook een niet aflatende inspiratiebron voor mijn fascinatie met pubers en jongvolwassenen. *Last but not least* bedank ik *Arjun* voor alles wat hij mij heeft gegeven - van onvoorwaardelijke liefde, de huwelijkse staat, de noodzakelijke persoonlijke ruimte en huishoudelijke ondersteuning, tot en met de genoegens van een heus 18[e] eeuws grachtenpand midden in de mooiste stad van ons land.

Van alle mensen ben ik het meest dank verschuldigd aan mijn strijdbare en knappe moeder *Loes Bolderheij* († 1983), die al een tegenstander van deeltijdwerk was voordat het was uitgevonden. Haar sociale betrokkenheid, toewijding, werklust en kritische liefde voor de geneeskunst én voor haar vier kinderen hebben mij gevormd en ik ben haar daar eeuwig dankbaar voor. Aan haar innige nagedachtenis, en aan die van mijn veel te jong gestorven jongste zusje *Jetske van Staa* († 1998), draag ik dit werk op.

About the author

Born in the South-African explosive town of Modderfontein near Johannesburg on August 28th 1957, AnneLoes van Staa first set foot on Dutch soil two years later. In her youth, she never lived anywhere much longer than for a few years – attending schools in Rijswijk, Veldhoven, Eindhoven, and Arnhem, where she finished her pre-university education in the Stedelijk Gymnasium in 1975. She then moved to Amsterdam and studied sociology at the University of Amsterdam. Her radical beliefs made her move to Rotterdam in 1977, where she worked in factories and offices before starting her training as a general nurse in the then Academic Hospital Rotterdam (Dijkzigt), now the Erasmus University Medical Centre.

Roy Borghouts

After her graduation, she trained as a pediatric nurse in the Sophia Children's Hospital (1984) and worked in numerous health care settings in the Netherlands. She also served the people in Nicaragua, where she worked in 1985-1986 (Lenin Fonseca Hospital - Managua) and in 1988-1989 (Artsen zonder Grenzen MSF, Puerto Cabezas). Living in Sandinist Nicaragua eventually convinced her that she disliked Marxist politics after all and she decided to return to her a progressive humanist roots instead. From 1986 to 1993, she studied medicine and cultural anthropology at the University of Amsterdam and specialized in medical anthropology (cum laude) and medicine (*vrij doctoraal* - cum laude). The subject of her double Master thesis (1993) was *'Myth and Metronidazole in Metro Manila'* in which she researched beliefs and prescribing practices of doctors in the treatment of acute childhood diarrhea in the Philippines.

From 1994 onwards, she works as assistant professor in the Institute of Health Policy and Management, Erasmus University Rotterdam, where she is involved in research, education and educational management. In 2003, she was also appointed professor of 'Transitions of Care' in the Centre of Expertise Innovations in Care of Rotterdam University, university of applied sciences. There, she heads a research and development program into adolescent health care, self-management and social participation in young people with chronic conditions.

AnneLoes is a chronic patient too. She lives with a lifelong allergic asthma and was successfully treated for breast cancer in 2006. After an eventful but rather unsuccessful love life, AnneLoes finally found happiness in 2001 with her present husband Arjun van der Dussen, father of two children of his own (Ruben and Stefan) and step-father of her two children, Tommy Maaiveld (1994) and Pien Tamboer (1998). She lives in the small village of Hekelingen near Spijkenisse, and in the most beautiful city of the world, Amsterdam. When not working (which occurs now and then), she loves to read (crime) novels; to listen to classical music; to visit concerts, operas, art house films and theatre plays; to cook and enjoy good food and wine with her family and friends – while dreaming of all the travelling she will do after finishing this thesis!