



GROWING UP WITH A CHRONIC CONDITION:

Challenges for self-management and self-management support

Jane N.T. Sattoe

**Growing up with a Chronic Condition:
Challenges for Self-management and Self-management Support**

Jane N.T. Sattoe

All studies in this thesis were part of the *Self-management & Participation Innovation Lab*, a collaborative research program of Rotterdam University of Applied Sciences (Research Centre Innovations in Care), Erasmus Medical Centre (Sophia Children's Hospital and Rehabilitation/TransitieNet), and the Department of Health Policy and Management of Erasmus University Rotterdam.

The work in this thesis was financially supported by grants, provided by SIA-RAAK (the Foundation Innovation Alliance) with funding from the Dutch ministry of Education, Culture and Science (OCW), and by the Dutch Kidney Foundation.

Rotterdam University of Applied Sciences awarded a *Promotievoucher* (PhD grant) to the author in 2011, and financially supported the printing of this thesis.



ISBN: 978-94-6169-684-7

Cover design: Diederik Corvers, Ogentroost

Lay-out and printing: Optima Grafische Communicatie, Rotterdam, The Netherlands

© Jane N.T. Sattoe, 2015

All rights reserved. No part of this book may be reproduced, stored in a retrieval system of any nature, or transmitted, in any form or by any means, electronic, mechanical, photocopying, recording or otherwise, without written permission from the author.

**Growing up with a Chronic Condition:
Challenges for Self-management and Self-management Support**

**Opgroeien met een chronische aandoening:
uitdagingen voor zelfmanagement en zelfmanagementondersteuning**

Proefschrift

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

De openbare verdediging zal plaatsvinden op
donderdag 25 juni 2015 om 13.30 uur

door

Jane Natascha Triedevi Sattoe
geboren te Rotterdam

PROMOTIECOMMISSIE

Promotor: Prof.dr. R. Bal

Copromotor: Dr. A.L. van Staa

Overige leden: Prof.dr. P. Moons
Prof.dr. H.A. Moll
Prof.dr. M.A. Grootenhuis

TABLE OF CONTENTS

1	Introduction	7
---	--------------	---

PART I Self-management and self-management support for young people: Explorations and recommendations

2	Unraveling self-management: A Delphi study exploring an ambiguous concept	27
3	'Skills for Growing Up': Supporting autonomy in young people with kidney disease	47
4	Self-management interventions for young people with chronic conditions: A systematic overview	65

PART II Transitions and self-management of young people with chronic conditions

5	Lagging behind or not? Four distinctive social participation patterns among young adults with chronic conditions	121
6	Young adults' experiences and satisfaction with the transfer of care: A follow-up study in a large hospital sample	139
7	How is self-management related to health-related quality of life in young adults with chronic conditions?	159

PART III Self-management support for young people with chronic conditions: Intervention effectiveness

8	Exploring effectiveness and effective components of self-management interventions for young people with chronic conditions: A systematic review	179
9	Effective peer-to-peer support for young people with end-stage renal disease: A mixed methods evaluation of Camp COOL	217
10	Conclusion: Main findings, general discussion & future directions	245
	Summary	273
	Appendices	289
	Dankwoord	291
	PhD portfolio	295
	List of publications	297
	Curriculum Vitae	299

1

Introduction



SPENDING TIME WITH SEM

John's eyes flashed with anger. It took him a great deal of self-discipline not to physically attack Sem. He clenched his fists and avoided his father's steady gaze. He could not deal with the clear condemnation in his father's eyes, not at this moment. All kinds of thoughts raced through his mind. How could he explain his feelings to his father? How to tell him that spending all these hours with Sem deprived him of... of living his own life. He sighed and sat down on the big white chair in the middle of the room, right before Sem. He stared at the floor, and his eyes fell on the black cable. He followed it with his eyes, over the floor, to the giant plug in the wall-socket. John frowned as another thought struck him. Why would he still name 'it'? He was seventeen now, not seven anymore! He was supposed to go to the movies tonight, with his classmates. Like all others in his class, and with... Julia. Why did he have to spend hours with a human-named machine?! This was not the first time it stood in his way. He shook his head, and glanced at the machine that made it possible to do hemodialysis at home. The device that he had started using again after his donor kidney failed him three years ago. The device that he had named Sem. A memory ran through his mind. They had completed his dialysis training in the hospital, and finally the machine had come. The nurse, Sophie, was setting it up for the first time. In this very same room. John recalled the intense joy he had felt at that moment. He saw the hopeful faces of his parents before him. He remembered how happy he felt telling them that this was not a machine, this was Sem, his new friend. Because Sem... Sem would be good for him. Sem would save him long, very long boring hours in the hospital. Sem would give him breathing space and time. He would be able to do more fun things, to be more 'normal'. He would have more control, more freedom. John relaxed his fingers and sat straight. He looked at his father. A little smile broke through John's face. The expression on his fathers' face softened. 'Alright, let's get this done', said John. Thirty minutes later, his mobile phone beeped. John reached for it with his free hand. 'I'm on dialysis again... Boring! Ready for a new game? This time I'll beat you', read a message from Ron, his buddy from camp. John grinned. 'Bring it on!' He texted back, after turning on his gaming device. He just scored his second point when his mobile phone beeped again. 'This movie is a joke! Want to join me for a fun one later this week? X Julia.'

GROWING UP WITH A CHRONIC CONDITION

There are thousands of young people like John in the story. Young people who grow up with a childhood-onset chronic condition. In the Netherlands, the most recent estimations indicate that 14% of all under 18-year-olds [1] and 11% of all under 25-year-olds [2] are living with a chronic condition or otherwise have special health care needs. World-

1. This is not a true story, but rather a compilation of impressions from field research.

wide, this number is growing, and these young people are therefore an increasingly important group of health care users with their own specific needs.

Young people go through different developmental stages and reach various developmental milestones. They are expected to become autonomous adults, who, eventually, leave their parents' (or caregivers') home, reach educational and/or vocational goals, start their own families, and participate in society. This multifaceted life-stage transition is already challenging, but is extra demanding for those with (childhood-onset) chronic conditions [3]. They have to balance the usual developmental tasks of adolescence and young adulthood with the adaptive tasks presented by their chronic condition. Fulfillment of these tasks is important for adjustment to adult life. According to Moos & Holahan these adaptive tasks are the following: managing symptoms, managing treatment, forming relationships with health care providers, managing emotions, maintaining a positive self-image, relating to family members and friends and preparing for an uncertain future [4]. Balancing between different tasks is complex, because a chronic illness and its treatment can have manifold effects on different areas of daily life and development, while developmental changes during the transition to adulthood reciprocally affect both illness and treatment [5-7].

Having a chronic condition may influence physical appearance, development and growth, but also mental health, emotional wellbeing, relationships with relatives and peers, and educational and vocational participation [5, 6, 8, 9]. Studies from the Netherlands have reported problems with psychosocial development, social participation or social functioning in young people with juvenile idiopathic arthritis [10], myelomeningocele [11], cerebral palsy [12-15], end-stage renal disease [16, 17], anorectal malformations, Hirschsprung's disease [17], physical limitations in general [18], and in survivors of childhood cancer [17]. Although there are differences between studies and between conditions, these young people in general reach developmental milestones later [17], and are at risk for poorer psychosocial development than their healthy peers [7, 17].

Furthermore, a review of qualitative studies on adolescents' experiences of living with a chronic condition made clear that having a chronic condition indeed complicates the development of friendships and school participation, as for instance hospitalizations and disclosure issues may stand in the way [19]. Moreover, it was found that the desire of these young people to 'be normal' influences how they deal with their chronic condition [3, 19]. Striving for normality might help young people develop resilience [20], but it is also reported as a common barrier to adherence, possibly leading to no-show at medical consultations and not complying with treatment regimens [21]. Low physical well-being, lack of support from peers or disclosure issues, and conflicts with parents also form barriers to treatment adherence [21]. Developmental changes, such as onset of puberty, might affect illness symptoms and disease patterns [5]. Poor disease control and associated health risk behaviors have been reported [5, 22]. Day-to-day manage-

ment of a chronic illness furthermore involves participation in health care, and for young people this includes the transition from pediatric to adult care [3]. If this is suboptimal, it could also lead to no-show at medical consultations or poor treatment adherence – with risk of medical complications and deterioration of illness [23].

Thus, balancing between different developmental and adaptive tasks is indeed complicated, and young people growing up with chronic conditions often need tailored support to successfully do so or, in other words, to take up self-management during transition to adulthood and adult care. Self-management support is therefore considered an integral part of today's chronic care, both for young people and adults.

TODAY'S CHRONIC CARE AND THE EMPHASIS ON SELF-MANAGEMENT SUPPORT

In the past decades, the changing course of diseases has posed a challenge to health systems that traditionally deal with acute health care needs [24]. The profound and increasing burden of chronic disease has led to the development of new service delivery models of care. In the Netherlands, the Council for Public Health and Health Care advised the government to use the Chronic Care Model as a framework to organize Dutch health care and improve its quality [25]. This model was developed as a guide for quality improvement in chronic care [26]. It endorses patient-centeredness and evidence based practice, and proposes six interrelated system changes to improve patient outcomes in chronic care. Four of these are directly related to the health system and organization of health care, and include the implementation of: self-management support, a coherent design of the care process (with preferably one coordinator), adequate decision support, and an adequate clinical information system. The two other changes relate to community and policy, and resources [26].

Self-management support (often combined with delivery system design) has been most often associated with improvements in health outcomes, functional status and quality of life, is [27], and both self-management and self-management support are top priorities in current health care for the chronically ill [26, 28-30]. The Dutch government emphasizes that self-management support is essential for good and structured care for people with chronic conditions [31]. This increasing policy attention is also reflected in research activities. A recent bibliometric analysis of chronic disease self-management studies found a fourfold increase in the number of publications in ten years [32]. Nevertheless, systematic attention to and integration of self-management (support) in daily care is still lacking [33, 34]. Also, little is clear about the effectiveness of self-management interventions [35, 36], and this is even more pertinent to self-management interventions in pediatric care [37, 38]. An important reason for the scarcity of evidence is the existence

of different conceptions of self-management, leading to very heterogeneous evaluation studies, and a lack of overview of what may be aims and outcomes of self-management interventions.

CONCEPTIONS OF SELF-MANAGEMENT

Notwithstanding the current emphasis on self-management and self-management support, the original notion of self-management was introduced in the 1960s and 1970s and stems from more general emancipation movements [36, 39]. In health care, self-management was promoted to challenge the notion of the passive patient, and to call for patient emancipation. Patients as consumers of health care wanted to be more involved in their own treatments and decisions around treatments, and self-management was a means to a more active role. Shared decision-making and equal partnerships between professionals and patients were called for [36, 39-41].

However, after advances in technology in the 1980s and 1990s had created more possibilities for self-monitoring and behavioral change, the focus shifted and self-management was introduced as a strategy that could ease the financial burden of increased chronic disease [39]. This conception of self-management emphasized a responsibility shift from professionals to patients. Patients were encouraged to monitor and take care of their own health, or in other words to 'self-care'. Self-care in this case refers to the "performance of activities or tasks [by patients] traditionally performed by professional health care providers" [42]. This was expected to lower patients' health service usage and consequently to decrease health care costs.

In the following twenty years, a modified notion of self-management arose. The emphasis was still on patients' own responsibilities, but the aim was different. In this notion of self-management, patients were expected to do as specified by professionals, with the aim to improve their health outcomes and quality of life as viewed by professionals [36, 39]. Self-management as such became associated with concepts as 'therapeutic adherence' and 'patient compliance'. However, in the past ten years, patients' lived experiences and psychological adjustment [32] were underlined as an opposing force to the focus on health outcomes and the professional as regulatory expert. Self-management in this view is more patient-centered and refers to the ability of patients to integrate the chronic condition in their daily lives with the best possible quality of life from their point of view (cf. [43]). Again, the necessity of shared decision-making and equal partnerships with health care professionals was emphasized.

Thus, over time, self-management has been reverted from an emancipation strategy of patients to gain control over their lives, a cost-cutting strategy of policy makers to reduce the financial burden of chronic care, a regulatory strategy of health care profes-

sionals to improve patient outcomes, back to the original notion of active patientship – with the caveat that self-management is not only a means to give patients a voice in a medical setting, but also a way to use patients' expertise in dealing with a chronic condition to optimize their living as they seem fit. Currently, there is no straightforward prevailing notion of self-management in health care practice. In fact, self-management has become a confusing term, which the different parties use to reflect their own beliefs and ideas about good chronic care, the roles of involved parties, and good self-management support [44, 45].

The different meanings attached to self-management, might enable the further development of self-management practices and research, as different actors can align with the concept. In a way, precisely the ambiguity and multiplicity of the term might allow it to become a 'boundary object' [46] that got different actors to pursue a self-management agenda. On the other hand, this ambiguity might also hamper its further development, or could lead to the dominance of specific definitions over others. This thesis contributes to the establishment of self-management support as an integral part of chronic care for young people with chronic conditions, by providing a more clear view on what self-management entails, what the different roles and abilities of involved parties are, as well as insight in influencing factors, and effective approaches or interventions for tailored self-management support.

SELF-MANAGEMENT IN DUTCH HEALTH CARE

Despite the co-existence of different notions of self-management, in recent years there seems to be a tendency towards a broader definition of self-management in the Netherlands [47, 48]. In 2008, the four-year 'National Action Program Self-management' (NAPS) was launched, financed by the Dutch government. It aimed to improve the implementation of self-management and self-management support in Dutch health care [49], and employed the broad definition of self-management as proposed by Barlow and colleagues: "the ability to manage the symptoms, treatment, physical and psychosocial consequences and life style changes inherent in living with a chronic condition" [43].

Through the NAPS, this broad view on self-management was introduced in Dutch health care, as well as a model named the Generic Model Self-management [49]. After conclusion of the program, a working group continued to implement its results, for instance by publishing a framework for self-management support in Dutch health care, entitled *Zorgmodule Zelfmanagement 1.0* [50]. It is not yet clear how these national initiatives have worked out in practice. Moreover, current policy reports and initiatives predominantly focus on adults with chronic conditions or on self-management in general, mostly neglecting the specific needs of young people growing up with chronic condi-

tions [3]. In pediatric care we thus find even a more pertinent lack of clarity about the concept of self-management [51]. Nevertheless, the notion that adult self-management models are not directly applicable to young people growing up with chronic conditions is more and more acknowledged [52-54].

The NAPS refers to the 'On Your Own Feet' (OYOF) research program when it comes to the subtheme of self-management of young people with chronic conditions. The OYOF program dealt with important topics for these young people, and their preferences and competencies in health care. It addressed the views of young people, their parents and health care professionals [3]. As such, it provided insights into important elements of self-management for young people with chronic conditions, and the different roles and abilities of involved parties. The projects included in this thesis continued where the OYOF research program stopped, and aimed to further conceptualize self-management in pediatric care, and to research influencing factors and effective approaches or interventions for tailored self-management support.

SELF-MANAGEMENT OF YOUNG PEOPLE WITH CHRONIC CONDITIONS

In 2011, a redefinition of the WHO definition of health was proposed in which health is defined as "the ability to adapt and self-manage in the face of social, physical, and emotional challenges" [55]. Although some professionals criticized this new definition [56, 57] - mostly questioning whether or not it is appropriate to leave out the disease part - it does reflect the increased attention for self-management and acknowledges the adaptive tasks of people with illnesses. Moreover, it emphasizes that health and self-management include more than physical or medical aspects. This is especially true for young people growing up with chronic conditions, given the reciprocal relationship between illness and development and the extra challenges it brings for them.

Considering this, it seems appropriate to adopt the broad definition of self-management mentioned earlier in the case of young people. It refers to dealing with "symptoms, treatment, physical and psychosocial consequences, and life style changes", and defines the ultimate goal of self-management as maintaining "a satisfactory quality of life" [43]. By doing so, it fits with the more patient-centered notion of self-management and provides a holistic view on self-management, allowing for consideration of developmental trajectories from childhood into adolescence and young adulthood.

Recently, Modi and colleagues introduced the Pediatric Self-management Model (PSM) [54]. The PSM underlines that people with childhood-onset chronic conditions have a wider range of self-management tasks compared to adults with chronic conditions. It promotes a system-based approach allowing for consideration of the social and physical environmental world of young people [54]. Still, it appears to more narrowly

focus on developing health and health care related skills like “determining health needs” and “communication with the medical team” [54]. As such, it does not consider other relevant tasks more related to integrating the consequences of the chronic condition in daily life activities. This is also the case in other recent studies and reviews, which mostly acknowledge the physical, emotional and social challenges of young people, but still speak of “chronic illness self-management” and focus on self-care and medical tasks [37, 58-60].

Consequently – and despite the recent acknowledgement that self-management is more than just the management of a medical condition – very little is known about how self-management is conceptualized and operationalized in health care services for young persons. Also, there is a lack of insight into influencing factors, and effective approaches or interventions for tailored self-management support for young people growing up with chronic conditions.

THIS THESIS

Research and aims

PART I

As has become clear above, the concept of self-management is multi-faceted and contested, and it is not clear how self-management is currently conceptualized by the different actors involved, e.g. policy-makers, health professionals, patients and researchers. The first part of this thesis therefore set out to explore the concept of self-management and self-management support. The research aim and questions were:

- I. **To explore how the concept of self-management and self-management support is embedded in current health care for young people with chronic conditions.**
 - A. How do Dutch researchers and policy-makers conceptualize self-management? (*chapter 2*)
 - B. What are the first experiences with the development and delivery of a self-management intervention for young people with chronic kidney disease? (*chapter 3*)
 - C. What are characteristics, contents, underlying theories, and expected outcomes of self-management interventions offered to young people with chronic conditions? (*chapter 4*)

PART II

It has also become clear that little is known about how young people growing up with chronic conditions develop self-management skills, and what may influence this pro-

cess. The second part of this thesis therefore explored these aspects in a cohort of young people with various chronic conditions. The research aim and questions were:

II. To research the development of self-management skills, and what factors may influence this process in young people with chronic conditions.

- D. What are the different patterns of autonomy in social participation (i.e. transition to adulthood) of young people with chronic conditions, and how do they differ? (*chapter 5*)
- E. How do young people with chronic conditions experience transfer to adult care and what are the associated characteristics? (*chapter 6*)
- F. How is self-management related to health-related quality of life in young people with chronic conditions? (*chapter 7*)

PART III

Lastly, it was noticed that there is no insight into outcomes of self-management support for young people with chronic conditions. The third part of this thesis hence studied the effectiveness of self-management interventions for this group. The research aim and questions were:

III. To study the effectiveness of self-management interventions for young people with chronic conditions.

- G. What can be said about effectiveness and effective intervention components of self-management interventions for young people with chronic conditions from a non-categorical approach to self-management? (*chapter 8*)
- H. What are the effects of a recreational camp as a self-management intervention for young people with chronic kidney disease? (*chapter 9*)

Study population

The study participants per research project are presented in Figure 1, and included researchers and policy advisors, young people² with chronic kidney disease (CKD) and their parents, pediatric nephrology professionals, initiators of a recreational camp for these young people, and a cohort of young people with a variety of chronic conditions that had been included in the On Your Own Feet research program [3].

2. In this thesis, the term ‘**child**’ refers to a person in the age range of 6 to 12 years, ‘**adolescent**’ is used for those in the age range of 13 to 18 years, and ‘**young adults**’ are those within the age range of 19 to 25 years. Whenever reference is made to groups that include two or more age ranges, the term ‘**young people**’ is used. The Medical Subject Headings (MeSH) were used to define these age ranges.

PART I Self-management and self-management support for young people: Explorations and recommendations	Chapter 2	<i>Delphi study</i> with researchers and policy advisors (n=20)
	Chapter 3	<i>Delphi study</i> with adolescents with chronic kidney disease (n=9), their parents (n=6), pediatric nephrology professionals (n=20), and experts (n=3)
		<i>Semi-structured interviews</i> with pediatric nephrology professionals (n=6)
		<i>Focus group discussions</i> with pediatric nephrology healthcare teams (n=4)
	Chapter 4	<i>Systematic overview</i> of self-management interventions for young people with chronic conditions (n=71)
PART II Transitions and self-management of young people with chronic conditions	Chapters 5, 6, & 7	<i>Follow-up questionnaire</i> (2012) among a cohort of young adults with chronic conditions (n=513) that participated in the 'On Your Own Feet questionnaire' in 2006
PART III Self-management support for young people with chronic conditions: Intervention effectiveness	Chapter 8	<i>Systematic literature review</i> of studies evaluating self-management interventions for young people with chronic conditions (n=31)
	Chapter 9	<i>Semi-structured interviews</i> with initiators/staff (n=4), participants of Camp COOL (young people with chronic kidney disease, n=12), and pediatric nephrology professionals (n=3)
		<i>Retrospective questionnaire</i> among former participants of Camp COOL (n=24)
		<i>Pre-post questionnaires</i> among current participants of Camp COOL (n=36)

Figure 1 Overview of studies and participants

The two intervention studies involved samples of young people with CKD, because these are known to be a vulnerable group in need of support [61]. They seem to have a harder time balancing their developmental tasks and the adaptive tasks associated with their chronic condition. Young people with CKD, for instance, often achieve fewer developmental milestones and lag behind in development compared to both healthy peers and to peers with other chronic conditions [17]. Also, they are at risk for cognitive impairments, low educational attainment, and psychosocial and psychiatric problems [62, 63].

The cohort study included young adults with a variety of chronic conditions³. This non-categorical approach seems appropriate, because young people face comparable challenges and similar adaptive tasks irrespective of type of condition [3, 4, 7].

Study context and overview

The research for this thesis was conducted within the framework of the research program ‘Self-management & Participation Innovation Lab’ (SPIL). SPIL is a four-year program that started in 2011 and aims to improve and support self-management of young people with chronic conditions. It is a collaborative research program of Rotterdam University of Applied Sciences (Research Centre Innovations in Care), Erasmus Medical Center, and the Department of Health Policy and Management of Erasmus University Rotterdam, and is financed by the incentive program RAAK-PRO of the Foundation Innovation Alliance (SIA - Stichting Innovatie Alliantie). Within SPIL, projects are being carried out in the Erasmus MC-Sophia Children’s Hospital, and at the Erasmus MC Rehabilitation department. The projects included in this thesis were carried out within the context of the Erasmus MC-Sophia Children’s Hospital and/or on national level, and build on the results of the OYOF research program [3]. More information about this program and the different projects within SPIL is available at: www.oepenbenen.nu. An overview of the research projects included in this thesis is presented in Figure 1.

Thesis outline

Part I of this thesis consists of three chapters that explore the concept of self-management and self-management support of young people. **Chapter 2** presents the results of a Delphi study with researchers and policy advisors in Dutch health care. It involves a critical exploration of different views on self-management. In **chapter 3**, the development and testing of a self-management intervention for young people with chronic kidney disease (The ‘Skills for Growing Up in pediatric Nephrology’ program) is described. By providing a systematic overview of existing self-management interventions for young people with chronic conditions, **chapter 4** considers the content, intervention formats,

3. The study samples in this research included young people growing up with chronic conditions – either congenital or acquired in childhood. A **chronic condition** is therefore defined according to the comprehensive definition proposed by Mokkink and colleagues after reaching national consensus in the Netherlands: “A disease or condition is considered to be a chronic condition in childhood if: (1) it occurs in children aged 0 up to 18 years; (2) the diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods or instruments according to professional standards; (3) it is not (yet) curable or, for mental health conditions, if it is highly resistant to treatment and (4) it has been present for longer than three months or it will, very probably, last longer than three months, or it has occurred three times or more during the past year and will probably reoccur” [64].

underlying theories, and expected outcomes of self-management support for these young people.

Part II elaborates on transitions and self-management of young people with chronic conditions. It provides insights into the development of self-management and into possibly influencing factors. **Chapter 5** identifies different patterns of autonomy in social participation of young adults with chronic conditions, and explores the nature of the differences. Then, in **chapter 6**, young adults' experiences and satisfaction with the transfer from pediatric to adult care are presented in relation to associated factors. Part II ends with **chapter 7** that explores the relationship between self-management and health-related quality of life in young adults with chronic conditions.

Part III contains two chapters that address intervention effectiveness. In **chapter 8**, the results of a systematic literature review into effective interventions and effective intervention components are described. **Chapter 9** includes a mixed-methods evaluation of a recreational camp as self-management intervention for young people with chronic kidney disease (Camp COOL).

Lastly, the overall results of this thesis are reviewed and discussed in **chapter 10**. This chapter also includes practice implications and recommendations for further research.

REFERENCES

1. Mokkink LB, van der Lee JH, Grootenhuys MA, Offringa M, van Praag BMS, Heymans HSA: Omvang en gevolgen van chronische aandoeningen bij kinderen [Extent and consequences of chronic conditions in children]. *Nederlands Tijdschrift voor Kindergeneeskunde [Dutch Journal of Pediatrics]* 2007, 75(4):138-342.
2. Gijzen R, van Oostrom SH, Schellevis F: Hoeveel mensen hebben één of meer chronische ziekten? [How many people have one or more chronic diseases?]. In: *Volksgezondheid Toekomst Verkenning, Nationaal Kompas Volksgezondheid*. Bilthoven: RIVM; 2013.
3. van Staa AL: On Your Own Feet. Preferences and competencies for care of adolescents with chronic conditions. Rotterdam: Hogeschool Rotterdam; 2012.
4. Moos RH, Holahan CJ: Adaptive tasks and methods of coping with illness and disability. In: *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. Edited by Martz E, Livneh H. New York: Springer; 2007.
5. Suris JC, Michaud PA, Viner R: The adolescent with a chronic condition. Part I: developmental issues. *Arch Dis Child* 2004, 89(10):938-942.
6. Yeo M, Sawyer S: Chronic illness and disability. *BMJ* 2005, 330(7493):721-723.
7. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.
8. Gorter JW, Stewart D, Woodbury Smith M, King G, Wright M, Nguyen T, Freeman M, Swinton M: pathways toward Positive Psychosocial Outcomes and Mental Health for Youth with Disabilities: A Knowledge Synthesis of Developmental Trajectories. *Can J Commun Ment Health* 2014, 33(1):45-61.
9. Ferro MA: Adolescents and young adults with physical illness: a comparative study of psychological distress. *Acta Paediatr* 2014, 103(1):e32-37.
10. Haverman L, Verhoof EJ, Maurice-Stam H, Heymans HS, Gerlag DM, van Rossum MA, Grootenhuys MA: Health-related quality of life and psychosocial developmental trajectory in young female beneficiaries with JIA. *Rheumatology (Oxford, England)* 2012, 51(2):368-374.
11. Buffart LM, van den Berg-Emons RJ, van Meeteren J, Stam HJ, Roebroek ME: Lifestyle, participation, and health-related quality of life in adolescents and young adults with myelomeningocele. *Dev Med Child Neurol* 2009, 51(11):886-894.
12. Verhoef JA, Bramsen I, Miedema HS, Stam HJ, Roebroek ME: Development of work participation in young adults with cerebral palsy: A longitudinal study. *J Rehabil Med* 2014, 46(7):648-655.
13. Tan SS, van Meeteren J, Ketelaar M, Schuengel C, Reinders-Messelink HA, Raat H, Dallmeijer AJ, Roebroek ME: Long-Term Trajectories of Health-Related Quality of Life in Individuals With Cerebral Palsy: A Multicenter Longitudinal Study. *Arch Phys Med Rehabil* 2014, 95(11):2029-2039.
14. Wiegerink DJ, Stam HJ, Gorter JW, Cohen-Kettenis PT, Roebroek ME: Development of romantic relationships and sexual activity in young adults with cerebral palsy: a longitudinal study. *Arch Phys Med Rehabil* 2010, 91(9):1423-1428.
15. van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, Wensink-Boonstra AE, Stam HJ, Roebroek 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.
16. Tjaden LA, Vogelzang J, Jager KJ, van Stralen KJ, Maurice-Stam H, Grootenhuys MA, Groothoff JW: Long-term quality of life and social outcome of childhood end-stage renal disease. *J Pediatr* 2014, 165(2):336-342.e331.

17. 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.
18. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis M: 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.
19. 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.
20. Ferguson P, Walker H: 'Getting on with life': resilience and normalcy in adolescents living with chronic illness. *Int J Inclusiv Educ* 2012, 18(3):227-240.
21. Hanghoj S, Boisen KA: Self-reported barriers to medication adherence among chronically ill adolescents: a systematic review. *J Adolesc Health* 2014, 54(2):121-138.
22. Michaud PA, Suris JC, Viner R: The adolescent with a chronic condition. Part II: healthcare provision. *Arch Dis Child* 2004, 89(10):943-949.
23. Lugasi T, Achille M, Stevenson M: Patients' perspective on factors that facilitate transition from child-centered to adult-centered health care: a theory integrated metasummary of quantitative and qualitative studies. *J Adolesc Health* 2011, 48(5):429-440.
24. World Health Organization: Innovative Care for Chronic Conditions. Building Blocks for Action. In. Geneva; 2002.
25. Huijben MEM: Het Chronic Care Model in Nederland [The Chronic Care model in the Netherlands]. In. Den Haag [The Hague]: Raad voor Volksgezondheid en Zorg [Council for Public Health and Health Care]; 2011.
26. Wagner EH, Austin BT, Davis C, Hindmarsh M, Schaefer J, Bonomi A: Improving chronic illness care: translating evidence into action. *Health Aff* 2001, 20(6):64-78.
27. van der Vlegel-Brouwer W: Integrated healthcare for chronically ill. Reflections on the gap between science and practice and how to bridge the gap. *Int J Integr Care* 2013, 13:e019.
28. Harvey PW, Petkov JN, Misan G, Fuller J, Battersby MW, Cayetano TN, Warren K, Holmes P: Self-management support and training for patients with chronic and complex conditions improves health-related behaviour and health outcomes. *Aust Health Rev* 2008, 32(2):330-338.
29. Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, Hoes A, Schuurmans M: Self-management: one size does not fit all. *Patient Educ Couns* 2013, 92(1):134-137.
30. Nolte E, Knai C, McKee M: Managing chronic conditions. Experience in eight countries. In. Copenhagen, Denmark: World Health Organization, on behalf of the European Observatory on Health Systems and Policies; 2008.
31. Zorginstituut Nederland: Toetsingskader kwaliteitsstandaarden en meetinstrumenten. Diemen; 2014.
32. Lu Y, Li Z, Arthur D: Mapping publication status and exploring hotspots in a research field: chronic disease self-management. *J Adv Nurs* 2014, 70(8):1937-1844.
33. Ursum J, Rijken M, Heijmans M, Cardol M, Schellevis F: Zorg voor chronisch zieken. Organisatie van zorg, zelfmanagement, zelfredzaamheid en participatie [Care for chronically ill. Organization of care, self-management, self-reliance and participation]. In. Utrecht: NIVEL; 2011.
34. Elissen A, Nolte E, Knai C, Brunn M, Chevreul K, Conklin A, Durand-Zaleski I, Erler A, Flamm M, Frolich A *et al*: Is Europe putting theory into practice? A qualitative study of the level of self-management support in chronic care management approaches. *BMC Health Serv Res* 2013, 13:117.
35. Newman S, Steed L, Mulligan K: Chronic physical illness: Self-management and behavioural interventions. New York: Open University Press; 2009.

36. Jonsdottir H: Self-management programmes for people living with chronic obstructive pulmonary disease: a call for a reconceptualisation. *J Clin Nurs* 2013, 22(5-6):621-637.
37. Kirk S, Beatty S, Callery P, Gellatly J, Milnes L, Prymachuk S: The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child Care Health Dev* 2012, 39(3):305-324.
38. Lindsay S, Kingsnorth S, McDougall C, Keating H: A systematic review of self-management interventions for children and youth with physical disabilities. *Disabil Rehabil* 2014, 36(4):276-288.
39. Kendall E, Ehrlich C, Sunderland N, Muenchberger H, Rushton C: Self-managing versus self-management: reinvigorating the socio-political dimensions of self-management. *Chronic illn* 2011, 7(1):87-98.
40. Rothman DJ: The origins and consequences of patient autonomy: a 25-year retrospective. *Health Care Anal* 2001, 9(3):255-264.
41. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26(1):1-7.
42. Medical Subject Headings [<http://www.nlm.nih.gov/mesh/>]
43. 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.
44. Rijken M, Jones M, Heijmans M, Dixon A: Supporting self-management. In: *Caring for people with chronic conditions A health system perspective*. Edited by Nolte E, McKee M. Berkshire: Open University Press; 2008.
45. van Hooft SM, Dwarswaard J, Jedeloo S, Bal R, van Staa A: Four perspectives on self-management support by nurses for people with chronic conditions: A Q-methodological study. *Int J Nurs Studies* 2014.
46. Star SL, Griesemer JR: Institutional Ecology, 'Translations' and Boundary Objects: Amateurs and Professionals in Berkeley's Museum of Vertebrate Zoology, 1907-39. *Soc Stud Sci* 1989, 19(3):387-420.
47. van den Brink R, Timmermans H, Havers J, Veenendaal Hv (eds.): Ruimte voor regie. Pioniers over zelfmanagement in de zorg [Room for agency. Pioneers about self-management in health care]. Utrecht: Kluwer; 2013.
48. Ouwens M, van der Burg S, Faber M, van der Weijden T: Shared Decision Making & Zelfmanagement [Shared Decision Making & Self-management]. In: Nijmegen: Scientific Institute for Quality of Healthcare (IQ healthcare); 2012.
49. www.zelfmanagement.com
50. CBO: Zorgmodule Zelfmanagement 1.0. Het ondersteunen van eigen regie bij mensen met één of meerdere chronische ziekten [Zorgmodule Self-management 1.0. Supporting self-management of people with one or more chronic diseases]. 2014.
51. Grypdonck MHF, de Jonge R, van der Bijl J, Kappen M, Duijnstee MSH: Zelfmanagement van adolescenten met een chronische ziekte: een kritische analyse van de literatuur [Self-management of adolescents with chronic conditions: a critical review of the literature]. In: *State-of-the-Art Studie Verpleging en Verzorging*. Edited by Jongerden I, Heynen-Kaales Y. Utrecht: LEVV; 2003.
52. 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.
53. Kirk S: Transitions in the lives of young people with complex healthcare needs. *Child Care Health Dev* 2008, 34(5):567-575.

54. Modi AC, Pai AL, Hommel KA, Hood KK, Cortina S, Hilliard ME, Guilfoyle SM, Gray WN, Drotar D: Pediatric self-management: a framework for research, practice, and policy. *Pediatrics* 2012, 129(2): e473-485.
55. Huber M, Knottnerus JA, Green L, van der Horst H, Jadad AR, Kromhout D, Leonard B, Lorig K, Loureiro MI, van der Meer JW *et al*: How should we define health? *BMJ* 2011, 343:d4163.
56. Tallini A: Health is state of physical, mental, and social wellbeing. *BMJ* 2011, 343:d5358.
57. Lewis DM: WHO definition of health remains fit for purpose. *BMJ* 2011, 343:d5357.
58. Lindsay S, Kingsnorth S, Hamdani Y: Barriers and facilitators of chronic illness self-management among adolescents: a review and future directions. *J Nurs Healthc Chronic Illn* 2011, 3(3):186-208.
59. Kirk S, Beatty S, Callery P, Milnes L, Prymachuk S: Perceptions of effective self-care support for children and young people with long-term conditions. *J Clin Nurs* 2012, 21(13-14):1974-1987.
60. Law GU, Tolgyesi CS, Howard RA: Illness beliefs and self-management in children and young people with chronic illness: a systematic review. *Health Psychol Rev* 2014, 8(3):362-380.
61. Kiberd JA, Acott P, Kiberd BA: Kidney transplant survival in pediatric and young adults. *BMC Nephrol* 2011, 12:54.
62. Icard PF, Hower SJ, Kuchenreuther AR, Hooper SR, Gipson DS: The transition from childhood to adulthood with ESRD: educational and social challenges. *Clin Nephrol* 2008, 69(1):1-7.
63. Grootenhuys 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.
64. Mokkink LB, van der Lee JH, Grootenhuys MA, Offringa M, Heymans HS: Defining chronic diseases and health conditions in childhood (0-18 years of age): national consensus in the Netherlands. *Eur J Pediatr* 2008, 167(12):1441-1447.

PART I

Self-management and self-management
support for young people:
Explorations and recommendations



2

Unraveling self-management: A Delphi study exploring an ambiguous concept



Jane N.T. Sattoe, Marjolijn I. Bal, AnneLoes van Staa, Roland Bal

Submitted

ABSTRACT

Background

Self-management support is advocated as a key element for chronic care, but the conceptualization of self-management is unclear, complicating research and policy decisions around self-management support.

Purpose and setting

Results of a Delphi study are used to explore the concept within a research and policy context. Experts were Dutch researchers and policy advisors in the field of chronic care. The Delphi study primarily aimed to reach consensus about synonyms of self-management. The argumentation given to in- or exclude a term was subjected to qualitative content analysis.

Principal findings

No consensus was reached for many terms, indicating disagreement amongst experts on whether specific terms were part of self-management. There seemed to be a tendency to link self-management with person-centered concepts and less with medical tasks. Furthermore, self-management was not seen as part of cost-cutting strategies.

Conclusions

Experts disagreed on what defines self-management. While medical professionals should be challenged not to limit self-management to medical management, researchers and policy advisors should be discouraged to overlook the importance of this domain. Patients' needs should determine the focus and content of self-management support. Researchers and policy advisors should be explicit about these needs and the aims of self-management interventions.

INTRODUCTION

The global epidemic of chronic conditions has confronted healthcare systems worldwide with a great challenge. The focus of care is shifting, e.g., between treatment and prevention, between hospitals and other care settings, in degree of professional and patient involvement, and between intuition and evidence in clinical practice [1]. The growing complexity of healthcare has led to the development of new service delivery models of care. One well-known model, endorsed by the World Health Organization, is the Chronic Care Model (CCM) that provides a comprehensive framework for the organization of healthcare systems that adequately respond to the profound and increasing burden of chronic disease. The CCM's key components of good chronic care are: self-management support, delivery system design, decision support and clinical information systems [2]. In 2008, the Council for Public Health and Health Care advised the Dutch government to use the CCM as a framework to organize Dutch healthcare and improve its quality [3].

Particular attention has been given to the establishment of self-management support. In 2009, a national program on self-management was started, financed by the Dutch Ministry of Health, Welfare and Sport. It aimed to stimulate self-management of people with chronic conditions [4]. Both nationally and internationally, self-management and support thereof from health care professionals are seen as important priorities in today's healthcare for chronically ill [2, 5-8]. However, a recent study found that self-management support is relatively underdeveloped [9]. Also, solid evidence regarding the effectiveness of self-management interventions and self-management support is lacking, complicating policy decisions around the use of self-management interventions. One explanation for this may be the difficulties in evaluating these heterogeneous interventions [7, 10, 11], which are also present in evaluation of comprehensive care programs [12] and chronic care in general [13]. Another explanation could be that self-management behavior is difficult to measure and to compare across studies because it is conceptualized in different ways [14], which points to a more fundamental challenge caused by a lack of common definitions.

Whereas some understand self-management to serve the purpose of improving clinical outcomes, as in therapy adherence [15, 16], others advocate a broader view that accounts for the dynamic life context of the chronically ill [17-22]. The first view reflects the medical viewpoint that considers healthcare professionals experts, and focuses on the chronic condition of the patient. It is based on outsider notions of how people should care for themselves. The second represents a patient-centered view on self-management based on lived experiences, shared decision-making, and a supportive role for healthcare professionals in helping patients integrate their chronic conditions in their daily lives [17, 21]. Also, predominantly in health policy documents, self-management

has been defined as a means to reduce the burden of increased disease incidence and prevalence on today's healthcare and healthcare costs [23].

However, despite differing views on self-management, there is increased attention for the patient-centered view on self-management in scientific literature [24], and even a call for a paradigm shift in healthcare [17]. Lorig and Holman (2003) subscribe to this view, and emphasize that even non-compliance with therapy can reflect a self-management style. Referring to Corbin and Strauss (1988), they defined self-management as a combination of three tasks: medical management (considering symptoms and treatment), role management (considering participation in society), and emotion management (considering emotional consequences of being ill). Consequently, self-management support seems to be located on a continuum of strategies and interventions aimed at different tasks [20], requiring different skills.

At the same time, other authors claim that the more narrow medical view on self-management is prevailing, considering that self-management support in clinical practice mostly focuses on the healthcare professional's perspective and patient compliance [17, 19, 21, 25]. Besides, little is known about the prevailing conceptualizations of self-management among researchers and policy advisors in healthcare. Therefore, this paper presents a conceptual exploration of self-management. It aims to gain insight into the conceptualization of self-management by Dutch researchers and policy advisors.

MATERIALS AND METHODS

Study design

This study was designed as a three-round online Delphi study among Dutch researchers and policy advisors, based on anonymity, iteration (subsequent rounds), and controlled feedback. The Delphi methodology is considered useful to measure and obtain group consensus in case of uncertainty or a lack of empirical evidence [26, 27]. It identifies the collective view of a group of respondents on a certain subject. The methodology is often used in health policy to develop healthcare quality indicators [28] or to build consensus, but it can also be used to study underlying factors preventing consensus, i.e. as a communication tool to generate debate [27]. In a Delphi study, experts are invited to provide opinions on a subject in three subsequent rounds. After the first round, the experts are sent an overview of the results of the first round, and are instructed to consider this overview when answering the questions in the second round. This procedure is repeated for the second round, leading to the third (and often final) round [26, 27].

The Delphi study primarily aimed to provide information for the development of a search strategy for a review of self-management interventions. Within the study, we searched for relevant terms that describe self-management or are part of or related

to this concept. The question presented to the experts was: ‘what proxy terms should researchers use in search for self-management interventions?’ Based on Wittgenstein’s theory of family resemblance [29], this question can be translated to: What defines the concept of self-management? Wittgenstein argued in his *Philosophical Investigations* that certain terms used in context with another term define this other term. Terms such as ‘games’ or ‘numbers’, he argues, cannot be defined absolutely, but by looking at the different ways in which the terms are used, we are able to bound them for all practical purposes. With this as a starting point, this paper reports on both the quantitative results of the Delphi study and the qualitative content analysis on the argumentation used by the experts in the Delphi study.

The experts

Dutch experts in self-management research were identified and selected from the project members’ network and from an invitation list of an earlier nationwide self-management expert meeting [30]. The only selection criterion was involvement in policy making or research on self-management (support) of people with chronic conditions. To obtain a broad range of views on self-management, 39 experts (34 researchers and 5 policy advisors) from 24 different organizations in the Netherlands were approached.

The Delphi study

The Delphi study took place between September and November 2012. The selected experts were provided with information about the study’s aims and procedure via e-mail, and were invited to fill out the first Delphi form online. They were asked to inform us if they did not wish to participate. The response period was set at two weeks, and a reminder was sent after one week. The final results of the Delphi study were provided to the participating experts via blind carbon copied emails.

The Delphi rounds

In the first round, we presented the conceptual model in which self-management encompasses medical management, role management and emotion management (i.e. the classification presented by Lorig and Holman [25]). The experts were first asked to reflect on the model and to comment on whether it would be useful to map self-management interventions. Then they were presented with eleven potential synonyms or proxies for the term ‘self-management’, namely: self-care, coping, self-control, self-monitoring, autonomy, goal setting, adherence, problem solving, self-determination, independence, and empowerment, which we had identified in a literature search. For each term they were asked to state whether it should be included in a search strategy on self-management interventions, and they were asked to motivate their decisions. Experts were also invited to propose alternative or additional terms.

Round two focused on the terms presented in the first round on which no consensus was reached and on newly proposed terms. The Delphi form for round two presented summary information about the qualitative responses to the first round. Taking this information into account, the experts were asked once again to decide on inclusion and exclusion of terms and to motivate their answers.

This procedure was repeated in round three regarding the remaining terms on which no consensus had been reached. The third Delphi form thus presented information about responses in the second round. Again, the question was to rate terms for inclusion and exclusion from the search strategy, while taking the argumentation of round two into account.

Analyses

Descriptive statistics were used to analyze consensus, which the research team defined as 70% or more of the experts agreeing on in- or excluding specific terms. Argumentations given to in- or exclude terms were categorized per term, and subjected to qualitative content analysis. The experts' ideas about associations or relations between terms and the concept of self-management were identified and summarized per term, regardless of whether these terms had to be in- or excluded according to the quantitative results. Argumentations that were more technical about a search strategy for interventions or that were unclear in their focus were not taken into account, because for this part of the study, we were primarily interested in the experts' ideas about the concept of self-management.

RESULTS

Response

Twenty-eight of the invited 39 experts responded to the invitation (72%) and 20 actually participated in the first Delphi round (51%): 3 policy advisors, and 17 researchers from 13 reference or research institutes. The eight respondents who did not participate in the study claimed to have no time or to be on vacation. In the second and third Delphi rounds, respectively 17 (44%) and 16 (41%) experts participated. Four researchers dropped out due to time constraints. Participants had various backgrounds: biology, health sciences, psychology, occupational therapy, cultural and social anthropology, human movement sciences, and medicine; but all were engaged in improving or researching self-management from a health services perspective.

Consensus and non-consensus: quantitative results

After the first Delphi round, the experts reached consensus on the inclusion of four terms: self-monitoring (94.7%), self-care (89.5%), empowerment (84.2%), and self-control (73.7%). No consensus was reached for seven other terms. A wide array of additional terms was proposed: confidence, self-regulation, together-management, self-diagnosis, self-efficacy, e-Health, telemedicine, active patient involvement, self-development, participation, chronic care management, disease management, motivation, shared decision making, compliance, education, learning skills, communication skills, competencies, knowledge, social support, lifestyle changes, self-reflection, personal health maintenance, and self-medication. These were presented in the second round.

After the second Delphi round there was still no consensus on the seven non-consensus terms from round 1. However, for all these terms, proportions of respondents who stated it should be included had decreased: problem-solving (50.0%), autonomy (43.8%), adherence (43.8%), self-determination (43.8%), independence (43.8%), goal-setting (31.3%), and coping (31.3%). The experts reached consensus on the inclusion of the following newly-proposed terms: self-regulation (86.7%), self-efficacy (80.0%), and shared decision-making (73.3%). Furthermore, consensus was reached on the exclusion of: confidence (93.3%), self-development (86.7%), learning skills (73.3%), and telemedicine (73.3%). No consensus was reached for the other 18 terms.

After the third Delphi round, consensus was reached for exclusion of 2 of the 7 original terms proposed by the project group on which so far no consensus had been reached: coping (80.0%), and independence (73.3%). Furthermore, the experts agreed on the exclusion of 6 of the 18 newly-proposed terms on which so far no consensus had been reached: self-diagnosis (93.8%), together-management (81.3%), e-health (81.3%), communication skills (81.3%), self-reflection (80.0%), knowledge (75.0%), and on inclusion of personal health maintenance (81.3%). In total, no consensus was reached for 16 out of 36 terms (Table 1).

Associations with the concept of self-management: qualitative results

During the first Delphi round, respondents provided feedback on the usefulness of the conceptual model of Lorig and Holman (2003) in which self-management encompasses medical management, role management and emotion management. It was generally found to be useful to map self-management interventions, but some experts argued that (provider) communication skills should be added, since these are essential for good self-management support. Also, it was pointed out that the model did not take into account the (social) environment of the person with a chronic condition, the attending health care professional, and the organizational structure within healthcare institutions.

All experts explained a large part of their answers by considering associations between the terms presented and the concept of self-management, rather than thinking

Table 1 Consensus and non-consensus after the last Delphi round

Consensus terms for inclusion (level of consensus in %)	Consensus terms for exclusion (level of consensus in %)	Non-consensus terms (level of consensus for inclusion in %)
Self-monitoring (94.7)	Confidence (93.3)	Active patient involvement (68.8)
Self-care (89.5)	Self-diagnosis (93.8)	Disease management (68.8)
Self-regulation (86.7)	Self-development (86.7)	Lifestyle changes (66.7)
Empowerment (84.2)	Communication skills (81.3)	Chronic care management (62.5)
Personal health maintenance (81.3)	Together-management (81.3)	Self-determination (56.3)
Self-efficacy (80.0)	E-health (81.3)	Self-medication (53.3)
Self-control (73.7)	Coping (80.0)	Goal-setting (50.0)
Shared decision making (73.3)	Self-reflection (80.0)	Autonomy (50.0)
	Knowledge (75.0)	Social support (43.8)
	Independence (73.3)	Participation (40.0)
	Learning skills (73.3)	Education (40.0)
	Telemedicine (73.3)	Compliance (40.0)
		Problem solving (37.5)
		Competencies (37.5)
		Motivation (37.5)
		Adherence (31.3)

about search terms for a review about self-management interventions. The most used argumentation considered if the term represented the whole concept of self-management, or merely an (important) part or element of it (Table 2). A term was positively associated with self-management if a term could be related to one of the domains of self-management, e.g., coping to emotion management. Furthermore, terms were seen as the same as or related to self-management if they were about persons dealing with their chronic condition, deciding for themselves, and knowing when to ask for help from professionals, as was the case for terms such as coping, self-determination, autonomy and independence. Professional help was linked to self-management if it was directed at activating patients and enabling them to manage their conditions. On the other hand, terms were believed not to be related to self-management (negative associations with self-management) if a term or concept was thought to be about: doing all ‘caring’ by yourself, dealing with ‘everything’ (not necessarily related to the chronic condition or treatment), diagnosing the disease instead of coping with the disease (and reacting to changes in symptoms, lifestyle or quality of life), and professional jargon and the organization of healthcare; as was for instance the case for concepts such as chronic care management, disease management, and self-diagnosis (Table 2).

Different positive and negative associations between terms and the concept of self-management overlapped. This was for instance the case for self-care. Self-care was positively associated with self-management by some experts, because it was thought to be related to medical management, but also negatively associated with self-management by others, because it was thought to be about ‘doing all caring by yourself’ and thus neglected persons’ (social) environment. More specifically, experts disagreed on whether

Table 2 Terms' associations with self-management according to respondents*

Terms	Associations with self-management (SM)	
	The term...	
Self-care	+	is an important part of SM
	+	refers to medical management
	–	is not necessarily related to SM, because SM is not about doing all 'caring' by yourself
Coping	+	is a synonym for SM, it is a way of self-managing
	+	is not similar to SM, but it is an important determinant of dealing with your condition
	+	refers to emotion management and role management
	–	is too broad and refers to dealing with 'everything', it is not necessarily related to dealing with a chronic condition
Self-control	+	is an aspect of SM
	+	is not similar to SM, but refers to a trait, determinant or outcome of SM
	–	is too broad, self-controlling is not necessarily self-managing a chronic condition
Self-monitoring	+	is an aspect of SM
	+	refers to medical management
Autonomy	+	is similar to SM, because it is about deciding for yourself based on good knowledge
	+	is an outcome or determinant of SM
Empowerment	+	is a relevant part of SM, because it is about enabling patients to manage their conditions
	+	refers to emotion management
Adherence	+	is an aspect, outcome or goal of SM, because monitoring adherence is part of managing your condition
	+	refers to medical management
	–	is not a part of SM
Problem-solving	+	represents SM, which can be seen as a problem-solving cycle
	+	is an aspect or determinant of SM, is needed for SM
	–	is too broad and it is not necessarily related to dealing with a chronic condition
Self-determination	+	is a synonym to SM
	+	reflecting on your own competencies is important to decide whether or not help from a professional is needed
	+	is not a synonym, but is an aspect or determinant of SM
	+	is one of the theoretical perspectives underlying SM
Independence	+	is a part of SM
	+	is a determinant or outcome of SM
	+	is related to SM, but being dependent and deciding on the help you get is also SM
	–	is too general, you may feel independent but do nothing about SM of your condition
	–	is not necessarily related to SM
Goal-setting	+	is an essential element of effective SM
	+	refers to role and emotion management
	–	is not necessarily related to SM, because it is not about being forced to set goals if you don't want to
Confidence	+	is an outcome of SM
	–	does not strictly relate to SM
Self-regulation	+	is an important part of SM
	+	is close to SM when it means that a person or group governs itself without outside assistance or influence
Together-management	+	may be a better term than SM
	–	is not a very commonly used term, and not related to SM

Table 2 Terms' associations with self-management according to respondents*(continued)

Terms	Associations with self-management (SM)	
	The term...	
Self-diagnosis	+	may be part of SM and is important
	–	is about diagnosing the disease, while SM is about coping with the disease and reacting to changes in symptoms, lifestyle or quality of life
	–	diagnosing yourself is not related to SM, it is not an aim of SM and seems to come earlier in the process than SM
Self-efficacy	+	is an essential part of SM, and strongly related to the concept of SM
	+	is an outcome of SM
e-Health	+	represents tools for SM, but is not always related to SM
Telemedicine	+	represents tools for SM, but is not always related to SM
	–	is not related to SM, because telemedicine tools often employ healthcare professionals' perspectives, instead those of patients
Active patient involvement	+	is an important part of SM, because activating patients is the prerequisite for SM and an important part of SM support
	+	directly relates to SM, because SM is always patient involvement
	–	is only relevant for SM if you relate it to 'treatment', because involvement could point at many activities
Self-development	+	may be an outcome of SM
	–	is completely different, has nothing to do with SM
Participation	+	is a goal or an outcome of SM
	+	is a very important part of SM that relates to social participation (role management)
Chronic care management	+	is related to SM, because SM is part of chronic care management
	–	is not related to SM, because from the patients' perspective it is about their conditions, and not about healthcare (organization)
Disease management	+	is related to SM, because SM is part of disease management
	–	is not related to SM, it is an organizational concept
Motivation	+	is a part or determinant of SM
	+	is one of the pillars for successful SM, a patient may have all the knowledge and skills for SM but without the motivation SM will fail or won't work
Shared decision making	+	is an important part of SM
	–	it is different from SM and requires other methods and approaches
Compliance	+	is part of SM
	+	is a synonym to adherence and medical management
	–	is not related to SM, another aspect
Education	+	is needed for SM (patient education or SM education)
Learning skills	•	no associations found
Communication skills	+	is important for performing SM activities
Competencies	+	is related to SM skills
Knowledge	+	is not exactly the same as SM, but an important element
	+	influences the way SM is done, is necessary for SM
Social support	+	an essential part of SM, because SM does not happen alone or in vacuum; social support often plays a role

Table 2 Terms' associations with self-management according to respondents*(continued)

Terms	Associations with self-management (SM)	
	The term...	
Lifestyle changes	+	an outcome of SM, because SM eventually is about patients adapting their lifestyles in order to manage their chronic conditions
	–	is not always related to SM, because SM is not always about changing lifestyles
Self-reflection	+	might be necessary for SM
	–	is not per se related to SM
Personal health maintenance	+	is a goal of SM
	+	refers to medical management
Self-medication	+	is part of SM
	+	refers to medical management, because it might also be related to taking non-prescribed drugs (e.g. for pain)

* a '+' sign indicates a positive association, while a '-' sign indicates a negative association

or not the following terms were related to or represented (a part of) self-management: lifestyle changes, compliance, adherence, disease management, chronic care management, and goal-setting. Other terms experts disagreed about for the same reason were: self-reflection, shared decision-making, self-development, self-diagnosis, self-control, coping, and self-care. However, they eventually reached consensus on these terms, because of technical argumentations related to the search (e.g., excluding a term because it would yield too many irrelevant hits).

DISCUSSION

The self in self-management

Since there is no MeSH-term or specific thesaurus term for the concept of self-management, we conducted a Delphi study to reach consensus about indispensable terms in a search strategy for self-management interventions. However, the results did not produce a straightforward search strategy because the terms on which consensus for inclusion was reached disproportionally represented the various domains of self-management. Also, for many terms, no consensus was reached, and experts seemed to disagree on what self-management holds.

When reviewing the associations between terms and the concept of self-management, the results seemed to reflect the contrasting narrow and broad views on self-management found in the international literature, while the view of 'reducing the burden on healthcare' prevailing in health care policy was remarkably absent. The results indicate that groups of experts had different 'family resemblances' linked to self-management. While some experts judged terms to be too broad to be related to

self-management because these did not focus on the chronic condition and its treatment, others advocated they were indeed part of self-management, for the very reason that they were broad and did not only focus on the medical domain. The narrow view on self-management employs a professional-centered approach [17, 21] and is in line with the dominant medical viewpoint that perceives the chronic condition to be one's master status [31]. But, is self-management nothing more than a way to improve medical outcomes? And is 'the self' just another term for the chronic condition? Strauss and Glaser (1975) described a chronic illness as a "negotiated reality" [32], emphasizing that people with chronic conditions are not always sick. One's illness behavior depends on one's perception of symptoms and on the extent to which symptoms interfere with daily life [33], i.e. their lived experiences [34-36]. In this light, the self is shaped by these lived experiences, and self-management and self-management support should therefore include more than medical management, i.e. role management and emotion management [25]. Still, if the ultimate goal of self-management is to maintain patient-defined wellbeing or satisfactory quality of life, then is this concept not just a catch-all for an array of patient-centeredness strategies to explore patients' lived experiences in daily life? If so, self-management could consequently lose its specific focus and become part of a more general concern with person-centeredness. Also, even though the holistic view seems to gain more and more popularity, and several authors have pointed out that the self is more than medical management [17, 19, 21, 25, 37, 38], little is known about what this self in self-management entails [39].

We asked a variety of Dutch experts to share their views on self-management in order to shed some light on the self in self-management. Indeed, at first glance, the collective view emerging from the present study seems to reflect the broad view on the self in self-management. Experts eventually agreed on including *empowerment* and *self-regulation*, terms that go beyond medical management. Yet they seem to disagree on assigning equal importance to medical management as well. They endorsed the term *self-care* as part of self-management, but failed to reach consensus on inclusion or exclusion of *adherence* and *compliance*, and the level of consensus decreased after the first two rounds, indicating a shift away from medical management. Moreover, they explicitly disagreed on whether *adherence* and *compliance* were even associated with self-management. However, although adherence with a treatment regimen may not be the primary goal of self-management, adaptive tasks to manage symptoms and treatment cannot be excluded from the self-management agenda as they are essential for everyone with a chronic condition [25, 40]. Also, many evaluation studies on self-management interventions target adherence or compliance. The tendency of our participants to underrate medical management within the context of self-management and to focus on the emotion and role domains is perhaps understandable given the strong focus on medical management in the literature and healthcare practice. Still, it also seems problematic,

because it could imply that researchers shift away from the very core of health care, i.e. health and illness. This underestimation of (the importance of) medical management may lead to a disconnection between research and healthcare practice, thereby nullifying the value of research for clinical practice and healthcare innovation and policy.

The broad view on self-management thus should include all three domains proposed by Lorig and Holman (2003). The self then seems to refer to: the person with the chronic condition (medical management), the manner of relating to others in society (role management), and unique experiences (emotion or identity management). This conceptualization is in line with other researchers' conceptualizations employing a holistic view on self-management (cf. [17, 19, 20, 22, 38, 41]). Moos and Holahan (2007) specifically mention the following adaptive tasks for people with chronic conditions: managing symptoms, managing treatment, forming relationships with healthcare providers, managing emotions, maintaining a positive self-image, relating to family members and friends, and preparing for an uncertain future. The first three seem to fall in the medical domain, the fourth and fifth reflect the emotion domain, and the latter two encompass the role domain. Specific competencies are needed for the ongoing dynamic interaction between these tasks. Self-management support consequently can be viewed as a twofold task of healthcare professionals: first, gaining insight into the person's needs considering self-management tasks (i.e., dealing with a medical condition (and its treatment), lived experiences, and societal roles); and second, facilitating the development of the required competencies – what some experts defined as 'enabling persons to self-manage'. This view is in line with the Chronic Care Model [2] that presents self-management support as a crucial component of healthcare for chronically ill. It includes professionals acknowledging for individual preferences that are important for self-management [42], and allows for a more general task of a healthcare professional: the individualization of care [43].

Nevertheless, placing an emphasis on the 'self' and on 'individualized care' does not mean that the focus of self-management support should be on the individual patient *per se*. The experts we consulted considered social and material contexts to be of utmost importance. What comes out as the 'self' is in itself a negotiated reality, and what is being managed is not just the individual person, but rather the person as part of a social and material environment. This also means that others than the patient him- or herself will take part in the 'care for the self' (cf. [44]), and that efforts should be directed at this broader understanding of the 'self' in context.

The management in self-management

Apart from the question of *what* is managed when talking about self-management (the *self*), the *management* part in self-management also raises questions. Management in general refers to the coordination of certain activities to achieve certain goals, moni-

tored by a manager. The question then becomes, whose goals are monitored in the case of self-management and by whom? Kendall et al. (2011) found that self-management is differently conceptualized by different actors: as a cost-cutting strategy for policy makers, as a quality improvement strategy for health professionals, and as an emancipation strategy for patients. The authors strongly favor the latter, and argue that “self-management approaches currently privilege professional ways of knowing over [patient] ways of knowing, thus limiting opportunities to exercise choice” [23].

Their reasoning is similar to that implicitly seen in a growing body of literature, and is related to the call for shared decision-making. In terms of chess, a change from ‘pawns’ into ‘queens’ is necessary for patients to equal the healthcare professional in decision-making [45]. In this respect, people with chronic conditions and professionals share the same self-management goals, and engage in a partnership in which the professional has a more facilitating and supporting role. In the present study this view is reflected in the final consensus on inclusion of the terms *shared decision making* and *empowerment*, and is part of a bigger paradigm shift in modern healthcare to neo-liberal methods of governance that emphasize individuality and freedom to choose [46]. The present-day tendency to replace the term ‘self-care’ with ‘self-management’ is illustrative for this shift.

Still, consensus on a more specific management term such as *goal-setting* was not reached, and consensus even decreased through the rounds. While some experts considered goal-setting essential to self-management, others claimed that it is up to the individual person to set goals or not. This is rather surprising, because goal-setting is inherent to management. *Management* as such implicitly requires action, while *self-management* in the broad view implies that “one cannot not [self-]manage” [25]. Even the *informed* decision not to act is self-management, yet it holds no action, and certainly no explicit goal-setting.

Critical management studies have shown the often illusory nature of goal-setting in management [47], and favor more experimental modes in which goals are emergent properties of management processes. Such a different view on management can explain some of the confusion about the meaning of ‘management’ in self-management, and point into other directions. Shared decision-making, for example, would not be focused on patients *choosing* but would be rather more explorative in trying to carve out options and possibilities for experimentations with preferred life paths, cf. [48], thus enabling patients to make their own informed choices considering their own lives. The management in self-management then seems to refer to informed choice, and goal-setting might be one of several – not necessarily effective – means to substantiate this choice.

Management may not be the right term to refer to the activities undertaken (or not) and decisions made to deal with the dynamic interaction between the medical, role, and emotion domains of life with a chronic condition. Moos and Holahan’s (2007) use of the

term ‘adaptive tasks’ which allows for *non-adaption* (i.e. the decision not to act) and as such seems more appropriate in the context of people with chronic conditions, might be an alternative. Anyhow, if management refers to informed choice, self-management support – and more specifically facilitating the development of required competencies – is about informing persons, and handing them the means to make their own informed choices (within their contexts).

Strengths and limitations

This study was a first attempt to include stakeholders in defining a search strategy considering self-management interventions. Recruited nationwide, participating experts came from a variety of organizations. However, the Delphi study did not solve the problem of heterogeneity, but rather reflected it. As such, it did not answer our primary research question, meaning that no straightforward search strategy with proxy terms for self-management could be derived from the results. Apparently, the Delphi method is not a useful approach to reach consensus in case of ‘conceptual confusion’ but we believe the methodology is still useful for conceptual explorations (as presented in this paper) as it can also structure group communication [27].

The gist of the Delphi study turned out to be the question of ‘what is (part of) self-management’ rather than ‘what (proxy) search terms would be effective to identify self-management interventions’. This shift may be related to features of the Delphi method or more specifically to the composition of the expert panel. Because the issue was research- and policy-oriented, we invited only policy advisors and researchers to participate in this Delphi study, and thus missed out on the opportunity to learn the unique views healthcare professionals and patients may have on self-management. As another limitation, the response period for each round was set at two weeks on account of which response rates were moderate, and several experts dropped out over the course of the study. Despite these limitations, our study offers new insight into the concept of self-management, and is among the first, to our knowledge, to include researchers and policy-advisors as stakeholders, and to explicitly discuss the difficulties of underlying assumptions for research into self-management support.

CONCLUSIONS

Although experts disagreed on what self-management holds, there seemed to be a tendency to link self-management with person-centered concepts and less with medical management. Yet, the lack of clarity around the conceptualization of self-management that is found between disciplines is also present within the group of healthcare researchers and policy advisors. While medical professionals should indeed be discouraged to

limit self-management to medical management, researchers and policy advisors should be discouraged to overlook the importance of medical management in daily living with a chronic condition. Self-management support starts with getting insights into persons' needs considering their adaptive tasks (at the medical, role and emotion domains). Consequently, persons' needs determine the focus of self-management support. To enhance the development of an evidence base for self-management interventions and to inform related health policy, we recommend future researchers as well as policy advisors to be more explicit about patients' needs self-management support responses to, i.e. the aims of self-management interventions.

ACKNOWLEDGEMENTS

The authors thank the experts who participated. Harald Miedema is thanked for having suggested this approach and his suggestions considering the study design. Finally, we thank the members of our Self-management & Participation Research Group for their comments on an earlier draft of this paper.

REFERENCES

1. Nolte E, McKee M: Caring for people with chronic conditions: an introduction. In: *Caring for people with chronic conditions. A health system perspective*. Edited by Nolte E, McKee M. Berkshire: Open University Press; 2008.
2. Wagner EH, Austin BT, Davis C, Hindmarsh M, Schaefer J, Bonomi A: Improving chronic illness care: translating evidence into action. *Health Aff (Millwood)* 2001, 20(6):64-78.
3. Huijben, MEM: Het Chronic Care Model in Nederland [The Chronic Care model in the Netherlands]. Den Haag [The Hague]: Raad voor Volksgezondheid en Zorg [Council for Public Health and Health Care]; 2011.
4. Landelijk Actieprogramma Zelfmanagement [www.zelfmanagement.com]
5. Harvey PW, Petkov JN, Misan G, Fuller J, Battersby MW, Cayetano TN, Warren K, Holmes P: Self-management support and training for patients with chronic and complex conditions improves health-related behaviour and health outcomes. *Aust Health Rev* 2008, 32(2):330-338.
6. Wagner EH: Chronic disease management: what will it take to improve care for chronic illness? *Eff Clin Pract* 1998, 1(1):2-4.
7. Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, Hoes A, Schuurmans M: Self-management: One size does not fit all. *Patient Educ Couns* 2013, 92(1):134-137.
8. Nolte E, Knai C, McKee M: Managing Chronic conditions: Experience in Eight Countries. Copenhagen: European Observatory on Health Systems and Policies; 2008.
9. Elissen A, Nolte E, Knai C, Brunn M, Chevreur K, Conklin A, Durand-Zaleski I, Erler A, Flamm M, Frølich A, Fullerton B, Jacobsen R, Saz-Parkinson Z, Sarria-Santamera A, Sönnichsen A, Vrijhoef H: Is Europe putting theory into practice? A qualitative study of the level of self-management support in chronic care management approaches. *BMC Health Serv Res* 2013, 13:117.
10. Newman S, Steed L, Mulligan K: Chronic physical illness: Self-management and behavioural interventions. New York: Open University Press; 2004.
11. Nolte S, Elsworth GR, Newman S, Osborne RH: Measurement issues in the evaluation of chronic disease self-management programs. *Qual Life Res* 2013, 22(7):1655-1664.
12. de Bruin SR, Versnel N, Lemmens LC, Molema CC, Schellevis FG, Nijpels G, Baan CA: Comprehensive care programs for patients with multiple chronic conditions: a systematic literature review. *Health Policy* 2012, 107(2-3):108-145.
13. Knai C, Nolte E, Brunn M, Elissen A, Conklin A, Pedersen JP, Brereton L, Erler A, Frølich A, Flamm M, Fullerton B, Jacobsen R, Krohn R, Saz-Parkinson Z, Vrijhoef B, Chevreur K, Durand-Zaleski I, Farsi F, Sarria-Santamera A, Soennichsen A: Reported barriers to evaluation in chronic care: Experiences in six European countries. *Health Policy* 2013, 110(2-3):220-228.
14. Rijken M, Jones M, Heijmans M, Dixon A: Supporting self-management. In: *Caring for people with chronic conditions. A health system perspective*. edn. Edited by Nolte E, McKee M. Berkshire: Open University Press; 2008.
15. Coleman MT, Newton KS: Supporting self-management in patients with chronic illness. *Am Fam Physician* 2005, 72(8):1503-1510.
16. Bourbeau J: Disease-specific self-management programs in patients with advanced chronic obstructive pulmonary disease: a comprehensive and critical evaluation. *Dis Manag Health Outc* 2003, 11(5):311-319.
17. Jonsdottir H: Self-management programmes for people living with chronic obstructive pulmonary disease: a call for a reconceptualisation. *J Clin Nurs* 2013, 22(5-6):621-637.

18. Schulman-Green D, Jaser S, Martin F, Alonzo A, Grey M, McCorkle R, Redeker NS, Reynolds N, Whittemore R: Processes of self-management in chronic illness. *J Nurs Scholarsh* 2012, 44(2):136-144.
19. Udlis KA: Self-management in chronic illness: concept and dimensional analysis. *J Nurs Healthc Chronic Illn* 2011, 3(2):130-139.
20. Jones MC, MacGillivray S, Kroll T, Zohoor AR, Connaghan J: A thematic analysis of the conceptualisation of self-care, self-management and self-management support in the long-term conditions management literature. *J Nurs Healthc Chronic Illn* 2011, 3(3):174-185.
21. Koch T, Jenkin P, Kralik D: Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2004, 48(5):484-492.
22. 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.
23. Kendall E, Ehrlich C, Sunderland N, Muenchberger H, Rushton C: Self-managing versus self-management: reinvigorating the socio-political dimensions of self-management. *Chronic Illn* 2011, 7(1):87-98.
24. Novak M, Costantini L, Schneider S, Beanlands H: Approaches to self-management in chronic illness. *Semin Dial* 2013, 26(2):188-194.
25. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26(1):1-7.
26. Jones J, Hunter D: Consensus methods for medical and health services research. *BMJ* 1995, 311(7001):376-380.
27. Powell C: The Delphi technique: myths and realities. *J Adv Nurs* 2003, 41(4):376-382.
28. Boulkedid R, Abdoul H, Loustau M, Sibony O, Alberti C: Using and reporting the Delphi method for selecting healthcare quality indicators: a systematic review. *PLoS One* 2011, 6(6):e20476.
29. Wittgenstein L: *Philosophical Investigations*. Oxford: Blackwell; 1958.
30. CBO, TNO, and Vilans: Report on researchers' expert meeting on self-management, 28 June 2012. Utrecht; 2012.
31. Goffman E: *Stigma: Notes on the management of spoiled identity*. Englewood Cliffs, NJ: Prentice Hall; 1963.
32. Strauss A, Glaser B: *Chronic Illness and the Quality of Life*. St. Louis, MO: Mosby; 1975.
33. Mechanic D, Volkart EH: Illness behaviour and medical diagnosis. *J Health Hum Behav* 1960, 1: 86-94.
34. Allen D, Wainwright M, Hutchinson T: 'Non-compliance' as illness management: Hemodialysis patients' descriptions of adversarial patient-clinician interactions. *Soc Sci Med* 2011; 73(1):129-134.
35. Atkin K, Stapley S, Easton A: No one listens to me, nobody believes me: self management and the experience of living with encephalitis. *Soc Sci Med* 2010; 71(2):386-393.
36. Gately C, Rogers A, Sanders C: Re-thinking the relationship between long-term condition self-management education and the utilisation of health services. *Soc Sci Med* 2007; 65(5):934-945.
37. Richard AA, Shea K: Delineation of self-care and associated concepts. *J Nurs Scholarsh* 2011; 43(3): 255-264.
38. Audulv A, Asplund K, Norbergh KG: The integration of chronic illness self-management. *Qual Health Res* 2012; 22(3):332-345.
39. Ratanachadawan Y: A response to T. Koch, P. Jenkin & D. Kralik (2004) Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2005; 52(5):571-572.
40. Moos RH, Holahan CJ: Adaptive tasks and methods of coping with illness and disability. In: *Coping with chronic disease and disability: theoretical, empirical, and clinical aspects*. Edited by Martz E, Livneh H. Springer: New York; 2007.

41. Kralik D, Koch T, Price K, Howard N: Chronic illness self-management: taking action to create order. *J Clin Nurs* 2004; 13(2):259-267.
42. Audulv A, Asplund K, Norbergh KG: Who's in charge? The role of responsibility attribution in self-management among people with chronic illness. *Patient Educ Couns* 2010; 81(1):94-100.
43. Gray JA: Self-management in chronic illness. *Lancet* 2004; 364(9444):1467-1468.
44. Corbin JM, Strauss AL: Unending work and care: managing chronic illness at home. San Francisco: Jossey-Bass; 1988.
45. Le Grand J: Motivation, Agency and Public Policy: Of Knights & Knaves, Pawns & Queens. Oxford: Oxford University Press; 2003.
46. Crawshaw P: Governing at a distance: social marketing and the (bio) politics of responsibility. *Soc Sci Med* 2012; 75(1):200-207.
47. Thomas AB: Controversies in management: Issues, debates, answers. 2 ed. London and New York: Routledge; 2003.
48. Mol A: Proving or improving: on health care research as a form of self-reflection. *Qual Health Res* 2006; 16(3):405-414.

3

'Skills for Growing Up': Supporting autonomy in young people with kidney disease



Jane N.T. Sattoe, Sander R. Hilberink, Mariëlle A.C. Peeters, AnneLoes van Staa

Journal of Renal Care. 2014; 40(2):131-139

ABSTRACT

Background and Objectives

End-stage kidney disease (ESKD) may interfere with children's developmental task of acquiring autonomy and participation. The *Skills for Growing Up* tool encourages normal development towards independence and autonomy during pediatric rehabilitation. This study aimed to adapt this self-management tool for use in pediatric nephrology, and to test whether its use is feasible in daily practice.

Design and Participants

A Delphi study was conducted among patients, their parents, professionals, and experts to adjust the tool for use in nephrology. Feasibility was studied through individual and group interviews with professionals in all Dutch pediatric nephrology centers.

Results

Agreement was reached on the areas of social participation and medical management of ESKD. Compared with the original, the new instrument holds considerable more attention for autonomy in the renal healthcare area; for example, dealing with medication and transplantation. Professionals used and appreciated the tool, but the paper form was seen to limit feasibility.

Conclusions

Making the tool available online is important. The challenge for professionals is to move beyond the focus on medical management and to consider developmental tasks when coaching their patients into adulthood.

Application to Practice

The *Skills for Growing Up - Nephrology* (SGU-N) is a promising instrument for use by professionals in pediatric nephrology. Its use can help young people achieving autonomy and may contribute to their successful transition to adulthood and adult care.

INTRODUCTION

A chronic medical condition may interfere with the main developmental task of acquiring autonomy and participation [1, 2]. Young people with end-stage kidney disease (ESKD) often achieve fewer developmental milestones or lag behind in development, compared with healthy peers and peers with other chronic conditions [3]. They are at risk for cognitive impairment, low educational attainment, and psychosocial and psychiatric problems [4, 5]. Young adults who reached fewer developmental milestones in adolescence experience greater impact of their condition on their daily lives [5]. As sound psychosocial development in early life relates to successful participation [6, 7], it seems valuable to help young people to achieve psychosocial milestones [8].

The Holland Bloorview Kids Rehabilitation Hospital in Toronto developed the *Skills for Growing Up* (SGU) tool [9]. An authorized Dutch adaptation appeared feasible in rehabilitation practice [10]. The SGU encourages typical development towards independence and autonomy in nine life areas: 'me', 'health care', 'relationships', 'education', 'work', 'living and ADL (activities of daily living)', 'transportation', 'leisure activities', and 'sports'. It consists of three age-appropriate checklists (7-11 years: 'Getting started'; 12-16 years: 'On my way'; 17 years and older: 'Almost there').

Apart from the tasks young people with chronic conditions have in common [11], those with ESKD face disease-specific challenges (e.g. medication/diet adherence, reductions in fluid allowance, dialyses or transplantation), stressing the need for an adaption of the SGU for pediatric nephrology. Also, as it is most likely to be used in a hospital setting, its feasibility for the hospital staff is important. This study aimed to adapt the SGU for use in pediatric nephrology, and to test whether the use of the tool is feasible in pediatric kidney care.

PATIENTS AND METHODS

The Skills for Growing Up tool

The SGU is built on four key principles:

- Universality, meaning that the content encourages family interaction about normal development (i.e. making choices/participating in life).
- Family centeredness reflected by the way in which young people and their families identify items they want to work on and set goals.
- Shared management [12, 13]: a conceptual approach for the transition to adulthood.
- Developmental approach: independence increases by developmentally appropriate steps in knowledge and competencies regarding self-reliance and autonomy in the nine life areas. Therefore, developmental age determines which list is appropriate to use.

Young people and their parents score the SGU's items in the appropriate checklist with 'yes' or 'no', depending on whether they perceive to have already acquired the skills or knowledge referred to. Next, they choose three items to work on for the next months, set goals to be achieved, and draft an action plan on how to achieve these goals. The forms are completed at home, and the instruction for parents of children of 12 years or older is to let their children fill out the form and make an action plan by themselves. In this way, young people would be allowed to address key adolescent health issues they might not want to share with their parents (e.g. substance use). For children younger than 12 years, parents fill out the form together with the child. This has to do with their ability to read and understand the items. Professionals may assist by addressing young people's independence during medical consultation (i.e. skills training or referral to other professionals). Examples of items and the action plan format are presented in Box 1.

Box 1 Item examples and action plan format

ME
'I can tell others what my condition is and what it practically means for my daily life' (12-16 yrs)*

HEALTHCARE
'I know what to do when I forget to take my medication' (12-16 yrs)

RELATIONSHIPS
'I spend time with my friends outside school' (12-16 yrs)

EDUCATION
'I know what to do to get an internship' (17+ yrs)

WORK
'I know the influence of my condition on work' (17+ yrs)

LIVING AND ADL
'I sometimes do chores at home' (7-11 yrs)

TRANSPORTATION
'I travel by myself by public transportation' (17+ yrs)

LEISURE ACTIVITIES
'I attend a camp, like school camp or soccer camp' (7-11 yrs)

SPORTS
'I can swim' (7-11 yrs)

ACTION PLAN
Step 1: I want to work on the following items:
[items scored with no]
Step 2: I will take the following steps to work on these items:
[description of steps to take]
Step 3: I will work on these items on:
[description of step] [date]

*yrs = years

Study samples

This study was conducted in three phases:

- 1. adaptation of the SGU;
- 2. pilot testing of the SGU for use in pediatric nephrology (SGU-N); and

3. finalizing the SGU-N.

All pediatric nephrology centers in the Netherlands participated. The Medical Ethics Committee of the Erasmus MC University Medical Center approved all study procedures. Participants received information letters and informed consent forms through the contact persons in the centers. They were assured of confidentiality and data were processed anonymously. All participants gave informed consent. If children were younger than 16 years, parents gave informed consent. In case of adolescents of 16 years and older, both adolescents and parents filled out and signed the informed consent forms.

In the first phase, professionals were invited to participate in a Delphi study [14]. Each center invited two parents and two young people with ESKD to participate. These young people and parents were unrelated, and received an information letter and were subsequently contacted via telephone. Four experts in the areas of nephrology and the development of autonomy and self-management in chronically ill were invited to participate in adapting and finalizing the SGU-N (phases 1 and 3).

In the second phase, each center selected nine young people without severe learning disabilities, equally distributed over the three age groups, and their parents to participate in the pilot. These young people with ESRD all had an appointment scheduled for consultation within two months after the start of the pilot. In each center, one professional who used the tool was interviewed.

Study procedure

Phase 1: Adaptation of the SGU. The Delphi study consisted of three rounds. Participants checked the relevance and phrasing of the original items and added nephrology-specific items (response categories: yes/no) (Round 1). To assess the extent of consensus participants assessed items' relevance on a seven-point Likert scale (Round 2). The experts reviewed the non-consensus items and decided on the definitive item-pool (Round 3).

Phase 2: Pilot of the SGU-N. The teams were trained in application of the tool. Next, the SGU-N was pilot tested in all centers for two months and individual and focus group interviews with professionals were conducted to assess feasibility. For both, interview guides were drafted. In the individual interviews, professionals reflected on their experiences with the SGU-N, its item-pool, and its potential impact on young people with ESKD and their families. The results were used to structure the focus group interviews, which primarily aimed at item reduction but also dealt with the format of the SGU-N and its value for nephrology practice.

Phase 3: Finalizing the SGU-N. The results of the group interviews and an additional expert meeting were used to finalize the SGU-N, to reduce the number of items, and to underpin recommendations for its use in clinical practice. Professionals decided on the final healthcare item pool, and experts determined the final participation item pool.

Analyses

After Delphi Round 1, items were maintained for re-assessment in Round 2 if:

1. $\geq 75\%$ of the respondents marked the item as relevant without need for reformulation or
2. between 30% and 75% of the respondents marked the item as relevant, but with need for reformulation.

Proposals for reformulations were reviewed by two researchers (JS and SH) independently and discussed and reformulated together. After Round 2, item-relevance and consensus among respondents was determined by a median item score (≥ 6) and an interquartile range (IQR ≤ 2.0) respectively. SPSS 17.0 was used for the statistical analyses. The pilot version of the SGU-N was then drafted. J.N.T.S. and S.R.H. independently scored the items of the pilot SGU-N with 'aimed at knowledge' or 'aimed at skills'. Cohen's kappa coefficient (κ) determined inter-rater agreement.

The individual and group interviews were digitally recorded, transcribed ad verbatim, and then imported into the qualitative software package Atlas.ti 6.2.27 (www.atlasti.com). Thematic analysis was applied [15]. In Atlas.ti initial codes (subthemes) were formulated on the basis of the interview guide and these were complemented with newly formed codes.

After the group interviews and expert meeting, the final SGU-N was developed. Again, J.N.T.S. and S.R.H. independently scored the nature of the items and Cohen's kappa coefficient was computed.

RESULTS

Participants

Twenty-six professionals, that is (specialized) nurses, social workers, nephrologists, psychologists, pedagogic workers and a dietitian, 10 adolescents with ESKD, 10 parents, and 4 experts were invited to participate in the Delphi study. Thirty-eight of these (22 professionals, 7 adolescents, 6 parents, 3 experts) (76%) participated in Round 1. Thirty-seven (20 professionals, 9 adolescents, 6 parents, 2 experts) (74%) participated in Round 2 (Table 1). Finally, three experts (75% of all invited) participated in Round 3. One had expertise in care for chronically ill adolescents, one was involved in the development of the original SGU, and one was experienced in research in ESKD.

Five professionals (one from each center) were individually interviewed - a psychologist and four (specialized) nurses. Focus group interviews took place in four of the centers with all the professionals who participated in the Delphi study. In the fifth

Table 1 Participants' characteristics

Pediatric nephrology centre	Role	Gender
1	Pediatric nurse*	Female
	Social worker	Male
	Nephrologist	Female
	Psychologist	Female
	Educational consultant	Female
	Pedagogue	Female
	Child aged 12-16 years	Male
	Child aged 17+ years	Male
	Parent of child aged ≤ 12 years	Female
	Parent of child aged ≥ 13 years	Female
2	Nurse specialist*	Female
	Nephrologist (adult care)	Male
	Nephrologist	Female
	Psychologist	Female
	Social worker	Male
	Child aged 12-16 years	Male
	Child aged 17+ years	Female
	Parent of child aged ≤ 12 years	Female
	Parent of child aged ≥ 13 years	Female
3	Nurse specialist*	Female
	Specialized nurse	Female
	Dietitian	Female
	Psychologist	Female
	Nephrologist	Male
	Child aged 12-16 years	Male
	Child aged 17+ years	Female
	Parent of child aged ≤ 12 years	Female
	Parent of child aged ≥ 13 years	Female
4	Nurse practitioner*	Female
	Nephrologist	Female
	Social worker	Female
	Psychologist	Female
	Nurse	Female
	Child aged 12-16 years	Male
	Child aged 17+ years	Female
	Parent of child aged ≤ 12 years	Female
	Parent of child aged ≥ 13 years	Female

Table 1 Participants' characteristics (continued)

Pediatric nephrology centre	Role	Gender
5	Nurse specialist*	Female
	Nephrologist	Male
	Educationalist	Female
	Social worker (adult care)	Female
	Social worker	Female
	Nurse (dialysis)	Female
	Child aged 12-16 years	Male
	Child aged 17+ years	Female
	Parent of child aged ≤12 years	Female
	Parent of child aged ≥13 years	Female

*Contact persons that helped with recruiting children and parents

center one individual interview with a specialized nurse was held instead, because of time constraints.

Adapting and finalizing the SGU

'Getting started' consisted of 41 original items, 36 were maintained and 18 were added (Round 1). Of the 54, 41 items were maintained ($IQR \leq 2$; median ≥ 6) (76%) (Round 2). Finally, the experts added 7 items (Round 3); 37% of the items addressed knowledge, while 65% covered skills ($\kappa = .94$). Of the 60 items of 'On my way', 53 were judged eligible and 35 items were added (Round 1); 5 of the 88 'On my way' items (6%) were removed ($IQR > 2$; median < 6) (Round 2). Finally, the experts added 8 items (Round 3); 47% of the items addressed knowledge and 43% skills ($\kappa = .96$). 'Almost there' contained 50 items of which 48 were judged eligible (Round 1); 31 items were added. None of the 79 items was removed (Round 2). Finally, the experts added 4 items (Round 3); 47% of the items addressed knowledge, while 53% addressed skills ($\kappa = .88$).

Since the pilot version of the SGU-N was considered to be too long for daily clinical practice, shortening was deemed necessary. The experts determined the core items in the participation areas, and group interviews with the teams were held to reduce the number of items in the healthcare area. The distribution of items within the SGU, the pilot SGU-N and the final SGU-N are presented in Table 2. The healthcare items in the SGU-N were divided into five categories: nutrition, symptoms and medication, visiting the hospital, dialysis and transplantation. In 'Getting started' 31% of the items addressed knowledge, while 69% covered skills ($\kappa = 1.0$). In 'On my way' 42% considered knowledge and 58% skills ($\kappa = 1.0$). In 'Almost there' 45% of the items covered knowledge, while 55% addressed skills ($\kappa = .91$).

Table 2 Comparison of the distribution of items within the SGU, pilot SGU-N, and final SGU-N

Life areas	Getting started <i>n</i> (%)*			On my way <i>n</i> (%)*			Almost there <i>n</i> (%)*		
	SGU	pilot SGU-N	final SGU-N	SGU	pilot SGU-N	final SGU-N	SGU	pilot SGU-N	final SGU-N
Me	6 (15%)	5 (10%)	6 (14%)	6 (10%)	10 (11%)	10 (13%)	5 (10%)	10 (12%)	10 (14%)
Living	15 (37%)	11 (23%)	9 (21%)	12 (20%)	8 (9%)	7 (9%)	7 (14%)	7 (8%)	7 (10%)
Relationships	3 (7%)	2 (4%)	4 (10%)	10 (17%)	11 (12%)	10 (13%)	10 (20%)	14 (17%)	15 (21%)
Education	3 (7%)	3 (6%)	3 (7%)	8 (14%)	9 (10%)	8 (10%)	3 (6%)	4 (5%)	4 (6%)
Transportation	2 (5%)	2 (4%)	2 (5%)	3 (5%)	2 (2%)	2 (3%)	2 (4%)	2 (2%)	2 (3%)
Sports	3 (7%)	3 (6%)	3 (7%)	2 (3%)	2 (2%)	2 (3%)	2 (4%)	2 (2%)	2 (3%)
Leisure	2 (5%)	3 (6%)	3 (7%)	5 (8%)	8 (9%)	9 (11%)	4 (8%)	5 (6%)	4 (6%)
Work	-	-	-	1 (2%)	1 (1%)	1 (1%)	6 (12%)	5 (6%)	5 (7%)
Healthcare	7 (17%)	19 (40%)	12 (29%)	12 (20%)	40 (44%)	29 (37%)	11 (22%)	34 (41%)	21 (30%)

*Number of items (% items compared to total in all life areas)

Pilot testing

SGU-N in clinical practice

Nurses primarily introduced the SGU-N to young people with ESRD and their parents and worked with the tool in daily clinical practice. In one center, a psychologist worked with the SGU-N during the pilot. The checklists and action plans were discussed with the young people and their parents and the outcomes were reported during multidisciplinary team meetings. If necessary, young people and their parents would be supported by other professionals in achieving their goals. *"We think that [working with the SGU-N] fits our job, because we form the pivot of the team. We [...] can ask other professionals for their input or support"* (Nurse). However, some items raised questions about the healthcare professional's role. They wondered if their supporting role indeed had to extend as far as the areas of living, transportation, and me. *"This boy formulated a goal in the area of living. He came up with it himself and his mother was very happy with it, so they are going to work on it now. However, I as a psychologist have nothing to do with that. Neither do our nurses or doctors"* (Psychologist).

Most participants filled out the checklists, but found it hard to formulate goals and working plans. The professionals thus ended up “*screening the whole list*”, while paying attention to the items scored with ‘no’. They tried to support the formulation of goals and plans. Professionals did not find this working method problematic, although it did cost extra time. *“I don’t expect [people not formulating goals and plans] to change in the future. It is something we have to support them in, which is absolutely no problem.”* (Nurse). Yet, it also worked this way if young people and their parents did make an action plan with goals. *“What I eventually saw was the action plan, but I got curious and I asked if I could see the whole list. The mother and child were okay with this”* (Psychologist).

Most of the professionals reported that working with the SGU-N was time consuming for two reasons. Firstly, the SGU-N was too long. *“The items. The areas. These are good. However, the list is too long.”* (Nurse). The second reason was that professionals received the lists and plans at consultation and thus needed extra time during consultation to review these. *“I think it could [save us time] if we would get an electronic version of the SGU-N.”* (Nurse). An electronic version of the SGU-N was highly preferred, also because it would give the possibility to send people an automatic reminder. *“People often forget to bring [the list] with them. [...] If it’s electronic, they could receive an automatic reminder”* (Nurse). Furthermore, professionals thought an electronic version would help them to fully embed the SGU-N in their working routines, and that it could provide the additional benefit of integration with electronic patient files – leading to a better overview of the patient for the whole team. They shared the view that the SGU-N has potential for fitting well to their daily routine, but at the end of the pilot this had not yet been achieved.

The value of the SGU-N

All nine life areas were considered relevant. The SGU-N gave professionals the opportunity to “*systematically*” address important issues, which was hardly done before. As such it provides healthcare professionals with “*guidance to support young people and their parents*”. *“[It helps us to] concretize the attention for transition [to adult care and adulthood]”* (Nurse). *“It can act as a guideline. If you have to deal with a non-adherent patient, it helps you realize what steps you can take to withdraw the focus from the non-adherence, while at the same time reaching the subject”* (Nurse).

Furthermore, professionals valued that the tool enabled them to focus on autonomy and self-management of patients at young age, and that it allowed for small steps in the development of independence. The family interaction that was stimulated by the tool was appreciated. Yet, they had to get used to the shift in focus of giving input to getting input from adolescents and their parents. Most professionals welcomed it, but some regretted that the SGU-N is not designed for “*testing knowledge*”. *“An important difference in view was that we wanted to see the SGU-N as a checklist to determine patients’ progress. [...] We think this is a missed chance”* (Nurse).

Professionals received predominantly positive reactions from participants. Many parents said it was an “eye-opener” and supported child-parent interaction. *“Using the instrument creates an opportunity for parents and children to start a conversation”* (Nurse). Professionals mentioned that the SGU-N could be particularly useful for those who have a difficult home situation (less social support), who find it difficult to start conversations on certain topics or who are overprotected by their family.

DISCUSSION

The SGU-N’s feasibility: thwarting logistical problems

The SGU was adapted for use in five pediatric nephrology centers in the Netherlands and the tool’s feasibility was evaluated. All teams had implemented the SGU-N and appreciated the four key principles. The SGU-N was viewed as a valuable addition in care for young people with ESKD, as is the original SGU in rehabilitation care [10]. Nevertheless, the logistical problems associated with the paper version of the SGU-N formed a major interfering factor for good embedment in daily practice. This barrier might be overcome by integrating the SGU-N into the KLIK PROfile, which is an effective web-based application for the use of patient reported outcomes [15, 17]. Electronic availability of the tool will add to user friendliness and facilitate nurses to incorporate the SGU-N into their consultations, contributing to the quality of holistic care for young people with ESKD.

Healthcare professionals: moving beyond medical management

The need for a tool to support young people to develop self-management skills and become an autonomous individual is widely recognized [18, 19]. Interventions to enhance psychosocial functioning and social participation of young people with a chronic condition are considered an integral component of the comprehensive care [2, 20]. Yet, professionals working in pediatric nephrology seem to have a strong focus on supporting patients’ knowledge and skills in the healthcare area. Numbers of healthcare items in the SGU-N increased 1.5 to 2-fold compared to the original tool, and professionals wondered if their supporting role had to extend to specific topics in the participation areas. Self-management is often used as a synonym for self-care or therapeutic adherence [21] and is incorrectly assumed to serve the goal of maximizing clinical outcomes and treatment efficacy in pediatric care [22, 23].

Self-management refers to “the individual’s ability to manage the symptoms and the consequences of living with a chronic condition, including treatment, physical, social, and lifestyle changes” [24]. In this broad view, it encompasses three elements: medical management (treatment/healthcare), role management (participation in society), and emotion management (emotional consequences of being ill) [22]. Research has shown

that young people and adults with ESRD, who have to deal with dialysis or transplantation, need support on all three elements [20, 25-27]. In a recent study, adults with a chronic kidney disease prioritized research themes as “mastery”, “partnership”, “responsibility”, “sexuality”, “work”, and “social contacts” for practically oriented research that induce support in dealing with their condition in daily life [28]. This contrasts to the dominant medical viewpoint that refers to the chronic condition as one’s ‘master status’ [29]. The ambiguity surrounding the term ‘self-management’ calls for more precise definitions of what is important for people with chronic conditions. Although this encompasses medical management of symptoms and a burdened body, the ‘self’ should not be reduced to a ‘disease manager’. More consideration of non-medical issues as self-reliance, autonomy and social participation in clinical practice is needed to improve the quality of pediatric care, and the quality of life of young people with chronic kidney failure [18]. The SGU-N that translates the broad definition of self-management into nine specific life areas, seems to be a promising tool allowing professionals in pediatric nephrology to widen their focus.

Self-management: weighing up costs and benefits

Goal-setting by participants appeared to be limited. They might find it hard to actually formulate goals to work with, or to change their role during consultation. The tool demands an active role of young people and parents and assigns a more passive demand-driven role to professionals. Young people and parents now need to explicitly set (a part of) the agenda; they may have to adjust to this new responsibility.

Professionals did not perceive the lack of action plans as problematic, it allowed them to pro-actively support adolescents and their parents in their goal-setting, which they viewed as an important task in using the SGU-N. This should not be a problem as long as professionals do not entirely take over the agenda-setting. Yet, some professionals wanted to see the whole list, even if an action plan was presented, and explicitly wished to use it to test patients’ knowledge and competencies. These professionals might have a natural tendency to take over responsibility of their patients. However, telling young people what they should do is not a viable option [30]. Moreover, developing self-management and becoming autonomous does not imply that adolescents will do what is considered right from a medical perspective.

Young people with ESRD are known to weigh medical advantages against social disadvantages [31, 32]. This weighing is a normal task of adolescence that should be acknowledged [33]. Non-compliance is often viewed as “indirect self-destructive behavior” or “disease-sustaining behavior” in clinical practice and negatively attributed to youths, while in fact they feel the need to talk about their motivation for their behavior [34, 35]. Themes as ‘living a normal life’ and ‘independence’ strongly relate to young people’s decision making [36]. Training in knowledge and competencies is necessary, but not

enough to understand and support self-management. Professionals should gain insight into young people's lived experiences in acquiring autonomy and social participation and identify challenging areas – as these will affect their self-management [30, 37-40]. The SGU-N allows professionals to address different life areas, and its use might for instance be complemented with motivational interviewing [41] to deepen the insight in these life areas, and to support adolescents in formulating their goals and action plans. In this way, professionals guide adolescents in their transition to independence.

Strengths and limitations

Our study included all pediatric nephrology centers in the Netherlands, and the sample of patients was heterogeneous in terms of age. The response rates were fairly good. Furthermore, the KLIK PROfile allows for good monitoring of patient reported outcomes [17], and integration of the SGU-N in this system seems promising. However, the effectiveness of the instrument is not considered in this study. Neither are the participants' views on the SGU-N. Hilberink *et al.* (2014) conducted a pre-post evaluation [10]. Unfortunately, our small pilot sample did not allow for such approach. Since the instrument is for young people with ESRD and their parents, and patients' view are important considerations in research with young people [42, 43], an essential recommendation for future research is to evaluate its effectiveness.

Implications for clinical practice

The SGU-N is a promising tool for professionals in pediatric nephrology. A web-based application is considered to increase the feasibility within daily practice. The tool can help young people achieving autonomy and hence may contribute to their successful transition to adulthood and adult care.

CONCLUSIONS

The SGU-N was developed in a participative way to encourage normal development towards autonomy. Young people, parents, professionals, and experts reached consensus on the broad scope life areas, including both social participation and medical management of ESKD. Professionals applied and appreciated the instrument, but having it available on-line is important for the instrument's feasibility. Furthermore, they have to get used to this new working method, and seem hesitant about a more passive role transferring responsibility to young people and parents. The challenge for professionals in pediatric nephrology is to move beyond the focus on medical management and to consider young people's developmental tasks when coaching them into adulthood. Insight into their lived experiences is essential for good self-management support.

ACKNOWLEDGEMENTS

The authors want to thank the young people and their parents, and the professionals of the participating centers. Furthermore, we thank Andrea Blok, Daniella Kramp, Lisette van Sintruije, and Evelien Polderman (Rotterdam University for Applied Sciences) for their help in data collection. Also, we are grateful to the expert group, especially the people who were involved in the development of the original Dutch tool: Karel Maathuis (University Medical Centers Groningen), Marij Roebroek (Erasmus Medical Center Rotterdam), and Ieteke Vos (Rehabilitation Center Blixembosch Eindhoven). Next, we thank the Holland Bloorview Kids Rehabilitation Hospital in Toronto for allowing us to use the original Skills for Growing Up (SGU) tool. Finally, we thank Lotte Haverman and Hedy van Oers (Emma Children's Hospital AMC) for their efforts in exploring the integration of the SGU-N into the KLIK PROfile. This study was funded by the Dutch Kidney Foundation.

REFERENCES

1. Turkel S, Pao M: Late consequences of chronic pediatric illness. *Psychiatr Clin North Am* 2007, 30: 819-835.
2. 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, 54:1453-1461.
3. Stam H, Hartman EE, Deurloo JA, Groothoff JW, Grootenhuys MA: Young adult patients with a history of pediatric disease: impact on course of life and transition into adulthood. *J Adolesc Health* 2006, 39:4-13.
4. Icard PF, Hower SJ, Kuchenreuther AR, Hooper SR, Gipson DS: The transition from childhood to adulthood with ESRD: educational and social challenges. *Clin Nephrol* 2008, 69:1-7.
5. Grootenhuys 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, 21:538-44.
6. Maurice-Stam H, Verhoof EJ, Caron HN, Grootenhuys MA: Are survivors of childhood cancer with an unfavourable psychosocial developmental trajectory more likely to apply for disability benefits? *Psychooncology* 2011, 22:708-714.
7. Haverman L, Verhoof EJ, Maurice-Stam H, Heymans HS, Gerlag DM, van Rossum MA, Grootenhuys MA: Health-related quality of life and psychosocial developmental trajectory in young female beneficiaries with JIA. *Rheumatology (Oxford)* 2012, 51:368-74.
8. Verhoof E, Maurice-Stam H, Heymans HS, Grootenhuys MA: Growing into disability benefits? Psychosocial course of life of young adults with a chronic somatic disease or disability. *Acta Paediatr* 2011, 101:e19-26.
9. Guidelines for service providers Supporting Youth & Families in Using the Skills for Growing Up Checklists. Guidelines for Service Providers - Bloorview Kids Rehab, 2007 [<https://secure1.securewebexchange.com/bloorview.ca/resourcecentre/familyresourfam/documents/serviceproviderguidelines.pdf>]
10. Hilberink SR, Vos I, Roebroek ME, Maathuis CGB: Skills for growing up for young persons with a physical disability. A feasibility and effectiveness study. *Submitted* 2014.
11. Van Staa AL, 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, 48:295-302.
12. Gall C, Kingsnorth S, Healy H: Growing up ready: a shared management approach. *Phys Occup Ther Pediatr* 2006, 26:47-62.
13. Kieckhefer GM: A shared management approach to transition. Paper presented at the Hospital for Sick Children, 2002 Toronto, ON.
14. Jones J, Hunter D: Consensus methods for medical and health services research. *BMJ* 1995, 311: 376-380.
15. Creswell JW: Research design: Qualitative, quantitative and mixed methods. London: SAGE; 2003.
16. Haverman L, Engelen V, van Rossum MA, Heymans HS, Grootenhuys MA: Monitoring health-related quality of life in paediatric practice: development of an innovative web-based application. *BMC Pediatr* 2011, 11:3.
17. Haverman L, van Rossum MA, van Veenendaal M, van den Berg JM, Dolman KM, Swart J, Kuijpers TW, Grootenhuys MA: Effectiveness of a web-based application to monitor health-related quality of life. *Pediatrics* 2013, 131:e533-543.

18. Annunziato RA, Jerson B, Seidel J, Glenwick DS: The psychosocial challenges of solid organ transplant recipients during childhood. *Pediatr Transplant* 2012, 16:803-811.
19. Newman S, Steed L, Mulligan K: Chronic physical illness: Self-management and behavioural interventions. New York, Open University Press; 2009.
20. Martinez W, Carter JS, Legato LJ: Social competence in children with chronic illness: a meta-analytic review. *J Pediatr Psychol* 2011, 36:878-890.
21. Koch T, Jenkin P, Kralik D: Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2004, 48: 484-492.
22. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26:1-7.
23. Modi AC, Pai AL, Hommel KA, Hood KK, Cortina S, Hilliard ME, Guilfoyle SM, Gray WN, Drotar D: Pediatric self-management: a framework for research, practice, and policy. *Pediatrics* 2012, 129: e473-485.
24. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J: Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns* 2002, 48:177-187.
25. Groothoff JW, Grootenhuys MA, Offringa M, Stronks K, Hutten GJ, Heymans HS: Social consequences in adult life of end-stage renal disease in childhood. *J Pediatr* 2005, 146:512-517.
26. Jansen DL, Heijmans M, Rijken M, Kaptein AA: The development of and first experiences with a behavioural self-regulation intervention for end-stage renal disease patients and their partners. *J Health Psychol* 2010, 16:274-283.
27. McDonagh JE: Child-adult interface. The adolescent challenge. *Nephrol Dial Transplant* 2000, 15: 1761-1765.
28. Schipper K, Abma TA: Coping, family and mastery: top priorities for social science research by patients with chronic kidney disease. *Nephrol Dial Transplant* 2011, 26:3189-3195.
29. Goffman E: Stigma: Notes on the management of spoiled identity. Englewood Cliffs, NJ Prentice Hall; 1963.
30. Glasgow RE, Davis CL, Funnell MM, Beck A: Implementing practical interventions to support chronic illness self-management. *Jt Comm J Qual Saf* 2003, 29:563-574.
31. Wells F, Ritchie D, McPherson AC: 'It is life threatening but I don't mind'. A qualitative study using photo elicitation interviews to explore adolescents' experiences of renal replacement therapies. *Child Care Health Dev* 2013, 39:602-12.
32. Nicholas DB, Picone G, Selkirk EK: The lived experiences of children and adolescents with end-stage renal disease. *Qual Health Res* 2011, 21:162-173.
33. Curtis-Tyler K: Levers and barriers to patient-centred care with children: findings from a synthesis of studies of the experiences of children living with type 1 diabetes or asthma. *Child Care Health Dev* 2010, 37:540-550.
34. Wolff G, Strecker K, Vester U, Latta K, Ehrich JH: Non-compliance following renal transplantation in children and adolescents. *Pediatr Nephrol* 1998, 12:703-708.
35. Waters AL: An ethnography of a children's renal unit: experiences of children and young people with long-term renal illness. *J Clin Nurs* 2008, 17:3103-3114.
36. Mitchell WA: Making choices about medical interventions: the experience of disabled young people with degenerative conditions. *Health Expect* 2014, 17:254-266.
37. Snethen JA, Broome ME, Kelber S, Warady BA: Coping strategies utilized by adolescents with end stage renal disease. *Nephrol Nurs J* 2004, 31:41-49.
38. Schulman-Green D, Jaser S, Martin F, Alonzo A, Grey M, McCorkle R, Redeker NS, Reynolds N, Whittemore R: Processes of self-management in chronic illness. *J Nurs Scholarsh* 2012, 44:136-144.

39. Walker RG, Buchbinder R: Fostering the ability of patients to self-manage their CKD. *Nephrol Dial Transplant* 2012, 27:3680-3682.
40. Reid J, Noble HR, Porter S, Shields JS, Maxwell AP: A literature review of end-stage renal disease and cachexia: understanding experience to inform evidence-based healthcare. *J Ren Care* 2013, 39:47-51.
41. Naar-King S, Suarez M: *Motivational Interviewing with Adolescents and Young Adults*. New York, The Guilford Press; 2010.
42. 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, 37:821-832.
43. Darbyshire P, Oster C, Hennings P: Children's and young people's experiences of chronic renal disease: a review of the literature, methodological commentary and a alternative proposal. *Journal of Clinical Nursing* 2005, 15:751-760.

4

Self-management interventions for young people with chronic conditions: A systematic overview



Jane N.T. Sattoe, Marjolijn I. Bal, Pepijn D.D.M. Roelofs, Roland Bal,
Harald S. Miedema, AnneLoes van Staa

Patient Education and Counseling. 2015; 98(6):704-715

ABSTRACT

Objective

To provide a systematic overview of self-management interventions (SMI) for young people with chronic conditions with respect to content, formats, theories, and evaluated outcomes.

Methods

Embase, Medline, PsycINFO, Web-of-Science, CINAHL, and Cochrane were searched. Reviews' reference lists were scrutinized. Selected studies were: Original research articles in English published between 2003-March 2014; about the evaluation of SMI for 7 to 25-year-olds with somatic chronic conditions/physical disabilities; with clear outcomes and intervention descriptions. The classification of medical, role and emotion management served to review content. Formats, theories, and evaluated outcomes were summarized.

Results

86 studies were reviewed. Most aimed at medical management and were unclear about theoretical bases. Although a variety of outcomes was evaluated and the distribution over self-management domains was quite unpredictable, outcomes conceptually related to specific content. A content-based framework for the evaluation of self-management interventions is presented.

Conclusions and practice implications

SMI relate to self-management tasks and skill-building. Yet, conceptualizations of self-management support often remained unclear and content focuses predominantly on the medical domain, neglecting psycho-social challenges for chronically ill young people. Future evaluations should match outcomes/themes to content and characteristics. Our framework and overview of SMI characteristics and outcomes may assist clinicians in providing self-management support.

INTRODUCTION

Worldwide, the number of young people living with a chronic condition or with special health care needs is growing. In the USA, the 2009-2010 National Survey of Children with Special Health Care Needs showed that 15.1% of all under 17-years-olds fell in this category [1]. In the Netherlands, the most recent estimations are 14% of all under 18-year-olds [2] and 11% of all under 25-year-olds [3].

Chronic illness affects young people in many ways during their transition to adulthood and adult care [4,5]. Supporting them to develop independence and self-management skills is therefore a key task of healthcare professionals. For that matter, self-management support is considered an integral part of healthcare for all people with chronic conditions [6-8]. Recently, the WHO definition of health was even redefined as “the ability to adapt and self-manage in the face of social, physical, and emotional challenges” [9].

Living with a chronic condition is an “ongoing process of inner negotiation” between social and medical needs [10] or what is described as shifting between the illness-on-the-foreground and wellness-on-the-foreground perspective [11]. Self-management therefore has been defined as “the individual’s ability to manage the symptoms and the consequences of living with a chronic condition, including treatment, physical, social, and lifestyle changes” [12]. Note, however, that self-management is not restricted to one’s individual ability, especially not in pediatrics where parents tend to play a key role. Adding the phrase “ [...] in conjunction with family, community, and healthcare professionals [...]” [13] seems to present a more complete picture. This holistic view accounts for the three tasks involved in self-management: medical management (*re. treatment*), role management (*re. social participation*), and emotion or identity management (*re. emotional consequences of being ill*) [14]. Young people with chronic conditions have to learn these tasks, and in supporting them we must take their developmental transition into account [15].

Various self-management interventions (SMI) for the chronically ill are available, but their effectiveness is not clear [16,17]. This is even more pertinent to SMI in pediatric care [16,18,19]. Newman and colleagues (2009) emphasize that a theory-based approach is needed to evaluate complex SMI, and recommend a more systematic comparison of different types of SMI [20]. Recent studies on SMI for people with chronic conditions in general [17,21] and for young people with physical disabilities [19] endorse this view, and recommend to standardize SMI evaluation by using a core set of outcomes [19,22].

We reviewed and systematically compared the characteristics and content of offered SMI for young people (7-25 years) with chronic conditions, their theoretical foundations, if any, and the evaluated outcomes. Based on the results we present content-related outcome measures for the evaluation of different types of self-management interventions.

METHODS

Study design

A systematic overview, defined by Grant and Booth [23] as a “summary of the literature that attempts to survey the literature and describe its characteristics”, was applied. This approach allows for a systematic comparison of SMI and outcome measures used in evaluation studies. Methodological characteristics according to the ‘Search, Appraisal, Synthesis and Analysis’ (SALSA) framework [23] are: comprehensive searching, quality assessment, narrative synthesis with tabular features, and thematic analysis. The review process was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [24].

Search strategy

The search strategy employed variations and Boolean connections (AND, OR) of the following terms: self-management, children, adolescents, young adults, chronic illness, and intervention. Relevant variations were derived from database thesauruses and relevant review articles (i.e. childhood, youth, chronic disease, physical disability, program etc.). Six health-related databases were searched: Embase, Medline, PsycINFO, Web-of-Science, CINAHL, and Cochrane. An information specialist helped define the final search strategies, employing a combination of free-text and thesaurus terms. The strategy used in Embase is presented in Box 1. Two researchers (JS, MB) supplemented the database searches by scrutinizing relevant reviews’ references for additional relevant publications.

Box 1 Search strategy in Embase

```
((('self care'/de OR 'self medication'/de OR 'self help'/de OR 'drug self administration'/de OR (((self OR shared)
NEAR/3 (manag* OR care* OR medicat* OR efficac* OR help*)):ab,ti) OR (((('coping behavior'/exp OR 'health
education'/de OR 'patient education'/de OR emotion/de OR emotionality/de) AND ('intervention study'/de OR
psychotherapy/exp OR 'program development'/de)) OR (psychotherap* OR ((coping OR cope OR cognitiv* OR
behavio* OR emotion* OR education* OR psychologic*) NEAR/6 (therap* OR interven* OR program*)):ab,ti))
AND ('chronic disease'/de OR 'genetic and familial disorders'/exp OR 'congenital disorder'/exp OR 'disabled
person'/de OR 'handicapped child'/de OR disability/exp OR (((chronic* OR longterm OR 'long term' OR 'end
stage' OR endstage* OR degenerat* OR persisten* OR genetic* OR familial* OR congenit*) NEAR/3 (ill* OR
disease* OR condition* OR disorder*)) OR (physic* NEAR/3 (handicap* OR disab* OR challeng*)):de,ab,ti)
AND (child/exp OR adolescent/exp OR adolescence/exp OR 'child health care'/de OR 'child care'/de OR 'child
hospitalization'/de OR 'handicapped child'/de OR (young OR youth OR child* OR adolescen* OR teenage*
OR teen OR teens OR juvenile*):ab,ti) AND ('comparative effectiveness'/de OR 'clinical effectiveness'/de OR
evaluation/de OR 'self evaluation'/de OR (effectiv* OR evaluat*):ab,ti)
```

Inclusion criteria

- *Study types*: only original research articles in English language published from 2003 to March 2014. No restrictions were placed on study design.
- *Participants*: young people (aged 7–25 years) with somatic chronic conditions or physical disability.
- *Interventions*: studies focusing on the evaluation of an SMI and describing the SMI or referring to previous description(s) of the intervention.
- *Outcome measures*: No restrictions were placed on the type of outcome measures, as this was a main interest. However, outcome measures needed to be clearly defined.

Studies had to meet all inclusion criteria to be included for further analysis. Furthermore, the term ‘children’ is used for young people aged 7–12 years, the term ‘adolescents’ is used for the age group of 13–18 years, and the term ‘young adults’ is used for those aged 19–25 years.

Selection, quality assessment, and data extraction

Retrieved records ($n = 6,373$) were imported into Endnote®. Two reviewers (JS, MB) independently selected eligible studies from both title and abstract and categorized them into: include, exclude or not clear. Any discrepancies were resolved, and decisions were made on the ‘not clear’ category. Full texts of all agreed-upon articles ($n = 444$) were retrieved. The two reviewers decided on final inclusion of articles based on the full text, resulting in 103 publications. The selection process is presented in Figure 1. Three reviewers (JS, MB, PR) assessed methodological quality of randomized controlled trials and cohort studies with methodology checklists of the Scottish Intercollegiate Guidelines Network (SIGN) [25]. For qualitative studies, the ‘Consolidated criteria for reporting qualitative research’ (COREQ) checklist was used [26]. Any discrepancies were resolved by discussion. Seventeen studies were excluded because outcome measures were not clear, leaving 86 studies. Two reviewers (JS, MB) extracted data on study design, study sample, type, format and content of interventions, settings of interventions, interventionists, theoretical basis, and outcome measures. Data were recorded in an electronic extraction form.

Analysis

General study characteristics were summarized, i.e. study country, chronic conditions addressed and study designs, as well as SMI characteristics, i.e. the modes, formats, elements and settings of SMI and professionals involved. Lorig and Holman’s classification of domains of self-management [14] served as a framework to review the content of SMI. Interventions could aim at medical management, role management, emotion management or a combination thereof. Further analysis included comparisons of theories underlying SMI per self-management domain. Finally, evaluated outcome measures

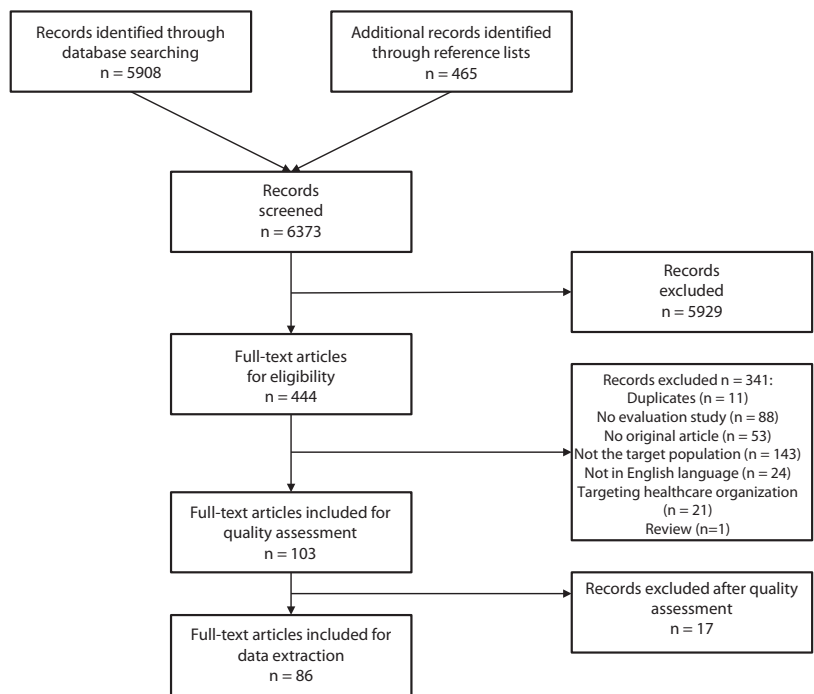


Figure 1 Selection process

were inventoried and linked to the content of SMI. On the premise that certain outcome measures logically relate to specific content of SMI, one reviewer (JS) linked all outcome measures to the content descriptions. Another reviewer (MB) checked this to enhance validity of the analysis.

RESULTS

General study characteristics (n=86)

- *Countries:* Most studies hailed from the USA (n=51), followed by the Netherlands (n=8), the UK (n=7), Australia (n=4), Canada (n=3), Germany (n=3), Hungary (n=2), Taiwan (n=2), Austria (n=1), China (n=1), Denmark (n=1), France (n=1), Haiti (n=1), and Norway (n=1).
- *Chronic conditions:* Most studies targeted asthma (n=18), followed by diabetes (n=16). Six studies targeted several chronic conditions (Table 1).
- *Study designs:* All but nine studies had fully quantitative study designs. Forty-five of them were randomized controlled trials, 29 were cohort studies and 3 were cross-sectional studies. Three studies had fully qualitative study designs, while five were

mixed-methods studies and one was a case study. Twenty-three studies (30.2%) were classified as pilot evaluations.

- *Interventions:* A total of 81 different interventions were reviewed, because different studies evaluated the same intervention with different outcome measures ([27] and [28]; [111] and [112]; [92] and [94] and [95]; [75] and [77]).

Table 1 Studies by chronic condition (n=86)

Chronic condition	References	No. (%)
Asthma	[26-43]	18 (20.9)
Diabetes	[44-59]	16 (18.6)
Cancer	[60-64]	5 (5.8)
Chronic fatigue syndrome	[65]	1 (1.2)
Chronic condition (various)	[66-71]	6 (7.0)
Chronic pain	[72-76]	5 (5.8)
Chronic respiratory condition	[77]	1 (1.2)
Cystic Fibrosis	[78-81]	4 (4.7)
Eczema (atopic dermatitis)	[82]	1 (1.2)
End-stage renal disease	[83-85]	3 (3.5)
Epilepsy	[86]	1 (1.2)
Heart disease	[87]	1 (1.2)
Hiv	[88,89]	2 (2.3)
Inflammatory bowel disease	[90]	1 (1.2)
Ichthyosis	[91]	1 (1.2)
Juvenile Fibromyalgia	[92-94]	3 (3.5)
Juvenile Idiopathic Arthritis	[95-98]	4 (4.7)
Migraine	[99]	1 (1.2)
Phenylketonuria	[100]	1 (1.2)
Physical disability	[101-103]	3 (3.5)
Sickle cell disease	[104-108]	5 (5.8)
Spina bifida	[109-111]	3 (3.5)

Intervention characteristics (n=81)

Interventions were either applied at individual level (n=39; 48.1%), at group level (n=34; 42.0%) or both (n=8; 9.9%). Most interventions included educational and/or skills training sessions (n=35; 43.2%) or telemedicine systems (n=14; 17.3%). Intervention formats and elements are summarized in Table 2. In 20 interventions (24.7%), parents were included as participants. These interventions often considered educational and/or skills training and most included both separate and joint sessions. Three interventions (3.7%) offered joint sessions only, while seven interventions (8.6%) offered separate but parallel

Table 2 Formats and elements of self-management interventions according to mode

Modes	Formats	Elements
Individual	Educational sessions (with or without parents) or written materials	<ul style="list-style-type: none"> - Informational (comic) books and videos - Daily diaries or notebooks (with or without rewards) - Homework assignments (written or skills practice) or workbook - Check-in or booster telephone calls by interventionist - Role reversal (between educator and the one(s) being educated)
	Motivational interviewing sessions	<ul style="list-style-type: none"> - Awareness building - Problem solving - Goal setting
	(Skills) training sessions	<ul style="list-style-type: none"> - Symptom treatment (e.g. relaxation techniques or pain provocation technique)
	Cognitive behavioral therapy sessions (some of them with parents)	<ul style="list-style-type: none"> - Educational and skills training - Instructions for home practice
	Family sessions	<ul style="list-style-type: none"> - Written materials - Responsibility-sharing plan - Family discussions (with conflict resolution) - Problem solving training - Communication training - Homework assignments (behavior)
	Telemedicine system (e.g. through personal devices, text-messaging, websites, or web-based systems)	<ul style="list-style-type: none"> - Monitoring through daily diaries - Overview of (trends in) disease-specific outcomes - Individualized feedback - Reminders or cueing - Social media communication or online discussion board - 'Gamification' (with feedback or rewards), role-playing or knowledge quizzes - Goal-setting or action plans - Information messages, animated lessons or tips - Skills training - Modules with homework - Possibility to contact healthcare provider
	CD-ROM	<ul style="list-style-type: none"> - Educational modules - Active coping plan - 'Gamification' with feedback
	Peer-support (e.g. befriending program)	<ul style="list-style-type: none"> - Mentorship
	Individual (transition) plan	<ul style="list-style-type: none"> - Age and developmentally appropriate information resources - Goal-setting

Table 2 Formats and elements of self-management interventions according to mode (continued)

Modes	Formats	Elements
Group	Cognitive behavioral therapy sessions	<ul style="list-style-type: none"> - Fun activities and games or role-playing - Homework (skills practice) - Involvement of parents as coaches - Goal-setting
	Art therapy sessions	<ul style="list-style-type: none"> - Discussion of weekly topics - Art making - Discussing art and related feelings
	Camping programs	<ul style="list-style-type: none"> - Traditional camping activities (e.g. horse riding, boating, arts etc.) - Disease specific activities (e.g. educational sessions, support groups, discussions, problem solving, role-playing, knowledge-testing games)
	Skills training or workshop	<ul style="list-style-type: none"> - Goal assessment and goal-setting - Drafting action or transition plans - Practicing strategies for goal achievement (e.g. through role-playing, coaching, use of audio-visual aids, accessing the Internet etc.)
	Educational and/or support sessions	<ul style="list-style-type: none"> - Informational videos, (coloring) books, written information, educational stories - Didactic presentations - Question and answer sessions - Discussions and problem solving - Homework assignments, exercise books and skills practice - Self-monitoring with contingency management - Self-management plans - Devices for self-monitoring (e.g. peak flow meter) - Peer education - Sharing experiences
	Family sessions (parallel but separate groups for children and parents; in some cases one mixed session)	<ul style="list-style-type: none"> - Play therapy, narrative therapy or role play - Relaxation training - Group work - Social support - Training in coping strategies - Homework (practice skills)
	School program (with continued phone contact)	<ul style="list-style-type: none"> - Didactic presentation about the disease - Peer education

sessions for parents and their children. Intervention settings were camping sites (n=10; 12.4%), inpatient or outpatient clinics (n=35; 43.2%), home or public environments (n=13; 16.0%), school (n=9; 11.1%), or online (n=10; 12.4%). Settings were not exclusive for the formats of interventions. Four studies (4.9%) did not detail the settings.

Interventionists included pediatricians, nurses, physiotherapists, occupational therapists, psychologists, social workers, and pedagogues. Occasionally, the whole healthcare team was involved. Twenty-two studies (27.2%) lacked this information. See Appendix C4.1 for an overview of general study characteristics and intervention characteristics per study.

Medical, role and emotion management: content of self-management interventions

The content of interventions includes the actual themes, topics, issues or specific skills discussed, reviewed or practiced during the interventions. Content is categorized by the domains of self-management in Table 3. Many interventions (46.2%) were solely aimed at medical management; some considered role management (6.4%) or emotion management (2.6%) alone. Others addressed multiple domains, see Figure 2.

Medical management was either disease-specific or of a more general nature. The former refers to tasks or topics associated with or related to a specific diagnosis, e.g., self-monitoring of blood glucose values in diabetes. This type of content is not exchangeable between interventions, e.g., education on treatment of cystic fibrosis is not useful for renal transplant patients. General medical management refers to health and healthcare related tasks irrespective of diagnosis. For instance, accessing healthcare, but also child-parent sharing or teamwork related to medical management tasks.

Role management referred to tasks or topics on domains related to social participation, such as communicating, decision-making, assertiveness, and keeping up with peers. Domains are school, work, community, living, housing, recreation, sports and leisure, relationships and sexuality. A major focus is on peer relationships and disclosure of the condition in social environments.

Emotion (or identity) management referred to the young person's feelings and intrinsic characteristics. Topics covered are building self-confidence, developing a positive body image, self-appreciation, maintaining positive thinking, stress management, but also acceptance of the condition.

The content of interventions was not specifically linked to certain modes, formats, elements or settings of SMI. In general, interventionists were not exclusive for content of interventions, although occasionally specific interventionists were included, e.g., a sexologist. See Appendix C4.1 for the classifications of self-management domains per study.

Table 3 Content of interventions categorized by the domains of self-management^a

Domains ^b	Content of interventions	References
Medical management	Disease-specific:	[27, 30, 31-50,
	1. Understanding the disease	52-57, 59, 60,
	2. Understanding (the necessity of) medication and treatment regimen; understanding side effects; adherence	63, 64, 66, 73-76, 79, 82-85,
	3. About the use of specific treatment devices or techniques (e.g. peak flow meter for asthma)	87, 90-93, 96,
	4. Dealing with symptoms	100, 101, 105-109]
	5. Drafting an individualized care plan	
	6. Self-monitoring of clinical outcomes	
	General:	[33, 39, 44, 47,
	7. Accessing healthcare	50, 54, 65, 76,
	8. Communication with healthcare professionals	82, 84, 85, 89,
	9. Managing doctor visits	92, 96, 98, 103,
	10. Coping with hospitalizations	105, 110]
	11. Goals and dreams for the future related to health and healthcare (transition)	
	12. Child-parent sharing / teamwork related to disease-specific medical management	
	13. Knowing where to find specific information about the disease	
Role management	14. Knowing when to ask for (medical) help	
	15. Risk behavior (e.g. unsafe sex or drug and alcohol abuse)	
	1. Social initiation and friendship making; social networks; family and romantic relationships	[27, 29, 33, 39,
	2. Managing teasing and bullying; conflict resolution	47, 51, 57, 59,
	3. Participating in normal social activities; keeping up with peers; Internet and social media	61-63, 65-67,
	4. Goals and dreams for the future related to school, work, community, living, housing, recreation and leisure (looking ahead); school issues	69, 71, 72, 76,
	5. Romantic relationships and sexuality	81, 82, 86, 87,
	6. Explaining the condition to others (disclosure); educating peers	89, 92, 96-99,
	7. Setting (life) goals and becoming assertive; growing up	102-104, 107,
	8. Communication and social problem solving (sometimes within families); organizational skills	110, 111]
Emotion management	9. Independent living; traveling/staying abroad	
	10. Social rights and benefits	
	1. Self-confidence or self-esteem building; developing a positive body image; body esteem	[29, 40, 42,
	2. Self-appreciation; enhancing hope; enhancing self-efficacy	47, 49, 51, 59,
	3. Empathy; fear-related thinking;	61, 62, 65-67,
	4. Feelings related to condition; sharing of feelings and experiences	70, 71, 74, 76,
	5. Accepting condition; self-reflection	80, 83, 86, 87,
	6. Healthy expressions of anger and transforming or managing anger	96, 99, 100,
	7. Helpful / positive thoughts; stress management	103, 104, 111]
	8. Decreasing negative thoughts	
	9. Decreasing stress and boredom; decreasing social isolation	
	10. Spirituality	
	11. Emotions	

^aNumber of studies is 78, three studies were unclear about the content of the intervention: [58, 68, 88]^bAccording to the model of Lorig & Holman [14]

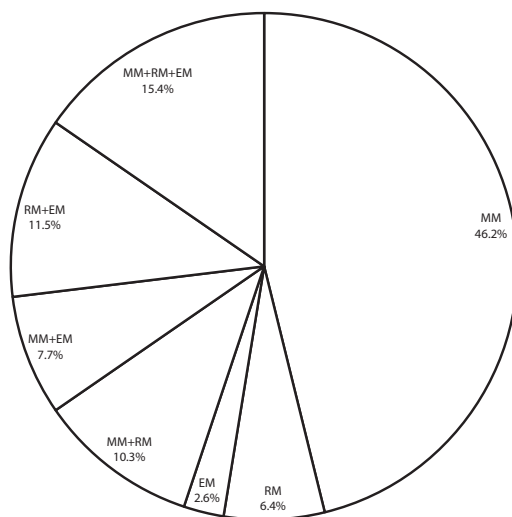


Figure 2 Distribution of interventions (n=68) over (combinations of) self-management domains
MM - medical management, RM - role management, EM - emotion management

Self-management interventions for different age groups

Most interventions targeted 12 to 17-year-olds (n=36; 44.4%) or 7 to 11-year-olds (n=23; 28.4%). Only five SMI (6.2%) targeted over 18-year-olds. For the rest, age groups overlapped. Formats and classification of self-management domains did not seem to be related to specific age groups, but content or themes obviously were not applicable to the whole age range. For example, an intervention classified as targeting both role and emotion management for children (mean age 10 years) targeted communication and social problem solving in general [51], while for young people (mean age 20 years) such an intervention targeted the social subtheme of romantic relationships [67]. Another theme specific for older age groups is vocational participation. Two interventions aimed at the whole age range (7 to 25 years) addressed medical management and self-monitoring through daily diaries, respectively.

Conceptualization of self-management: theoretical bases of self-management interventions

Fifty-five studies (67.9%) either failed to state whether the interventions were based on an existing theory (n=48) or, if they did so, did not specify the theoretical base (n=7). Of the other studies, most referred to learning theories like Bandura's (cognitive) social learning theory or cognitive behavioral theory (Table 4). A theoretical base was mostly mentioned in relation to interventions targeting medical management alone, while only one of the studies evaluating role management interventions mentioned a theoretical

Table 4 Theoretical bases of self-management interventions, no. (%)

Theoretical base	Number of interventions (n=26)	References
(Cognitive) social learning theory	10 (38.5)	[29, 31, 48, 51, 59, 65, 75, 76, 79, 89]
Cognitive behavioral theory	9 (34.6)	[64, 66, 70, 74, 75, 91, 93, 106, 109]
Health Belief Model	2 (7.7)	[35, 85]
Prochaska's transtheoretical model	1 (3.8)	[35]
Self-regulation model of health and illness	1 (3.8)	[65]
Transactional model of stress	1 (3.8)	[40]
Orem's self-care deficit theory of nursing	2 (7.7)	[39, 44]
Game-playing and health theory	1 (3.8)	[108]
Flirt Model	1 (3.8)	[67]
Self-confrontation	1 (3.8)	[99]
Model of Human Occupation	1 (3.8)	[104]

base. In general, neither the content of interventions nor intervention characteristics were specific for a certain theoretical base.

Evaluating self-management interventions: measured outcomes

Interventions were evaluated on a wide variety of outcomes, primarily health outcomes (61.5%), health-related quality of life (HRQoL) (35.9%), and knowledge about the disease and/or treatment (29.5%) (Table 5).

Interventions solely aimed at medical management (n=36) were evaluated on all outcome measures except coping, psychosocial functioning, and support by others. Of the five interventions solely aimed at role management, two were evaluated only on health outcomes and two on psychosocial functioning. One of the two emotion management intervention studies evaluated knowledge of disease and/or treatment, and the other social participation (Table 5). None of the outcomes or groups of outcomes could be related to one particular type of intervention and the distribution over self-management domains or combinations of self-management domains was quite unpredictable. Appendix C4.2 presents an overview of outcome measures per study (Table A.1), and of the groups of outcomes (Table A.2).

Linking content and outcomes: a content-based evaluation framework

Regarding the content of interventions (Table 3), certain content logically relates to groups of outcomes or themes. If, for example, 'understanding of the disease' and 'adherence' is addressed, it would seem logical to evaluate intervention effectiveness from improved knowledge, clinical outcomes and self-reported adherence rather than from psychological outcomes such as depressive symptoms or anxiety. Grounded on

Table 5 Outcomes used in the evaluation studies distributed over (combinations of) self-management domains

(Combined) domains of self-management ^a No. (% of total studies ^b)		MM	RM	EM	MM+RM	MM+EM	RM+EM	MM+RM+EM	Total ^b
Groups of outcome constructs or themes ^c		n=36 (46.2)	n=5 (6.4)	n=2 (2.6)	n=8 (10.3)	n=6 (7.7)	n=9 (11.5)	n=12 (15.4)	n=78 (61.5)
Health outcomes		27 (75.0)	2 (40.0)		4 (50.0)	5 (83.3)	3 (33.3)	7 (58.3)	48 (61.5)
Health-related quality of life		13 (36.1)			5 (62.5)	1 (16.7)	4 (44.4)	5 (41.7)	28 (35.9)
Knowledge of disease / treatment		12 (33.3)		1 (50.0)	6 (75.0)		2 (22.2)	2 (16.7)	23 (29.5)
Psychological outcomes		7 (19.4)			1 (12.5)	1 (16.7)	5 (55.6)	2 (16.7)	16 (20.5)
Self-efficacy		8 (22.2)			3 (37.5)		2 (22.2)	2 (16.7)	15 (19.2)
Vocational participation		5 (13.8)			2 (25.0)	1 (16.7)	2 (22.2)	2 (16.7)	12 (15.4)
Social participation		2 (5.6)	1 (20.0)	1 (50.0)	1 (12.5)		5 (55.6)	2 (16.7)	12 (15.4)
Coping		1 (2.8)			1 (12.5)	2 (33.3)	3 (33.3)	1 (8.3)	8 (10.3)
Self-care		2 (5.6)			3 (37.5)			2 (16.7)	7 (9.0)
Psychosocial functioning			2 (40.0)			1 (16.7)	1 (11.1)	2 (16.7)	6 (7.7)
Family involvement or conflict (related to disease-related management tasks)		4 (11.1)			1 (12.5)		2 (22.2)		7 (9.0)
Sense of control		1 (2.8)			2 (25.0)				3 (3.8)
Attitudes towards illness		2 (5.6)			1 (12.5)		1 (11.1)		3 (3.8)

Table 5 Outcomes used in the evaluation studies distributed over (combinations of) self-management domains (continued)

(Combined) domains of self-management ^a		MM	RM	EM	MM+RM	MM+EM	RM+EM	MM+RM+EM	Total ^b
No. (% of total studies ^b)		n=36 (46.2)	n=5 (6.4)	n=2 (2.6)	n=8 (10.3)	n=6 (9.9)	n=9 (11.5)	n=11 (15.4)	n=78
Groups of outcome constructs or themes ^c									
Self-perception of competencies		1 (3.1)			1 (14.3)				2 (2.6)
Problem solving		2 (6.3)							2 (2.6)
Support by others				1 (50.0)				1 (9.1)	2 (2.6)

^a According to the model of Lorig & Holman [14]: MM = medical management, RM = role management, EM = emotion management

^b Number of studies is 78, three studies were unclear about the content of the intervention: [58, 68, 88]

^c Only measured in young people (e.g., no parent proxy measures)

this premise, a conceptual content-based measurement framework for the selection of outcome measures in the evaluation of SMI is presented in Figure 3. The outcome measures correspond to the numbered content descriptions in Table 3. The only outcome related to all three domains was HRQoL.

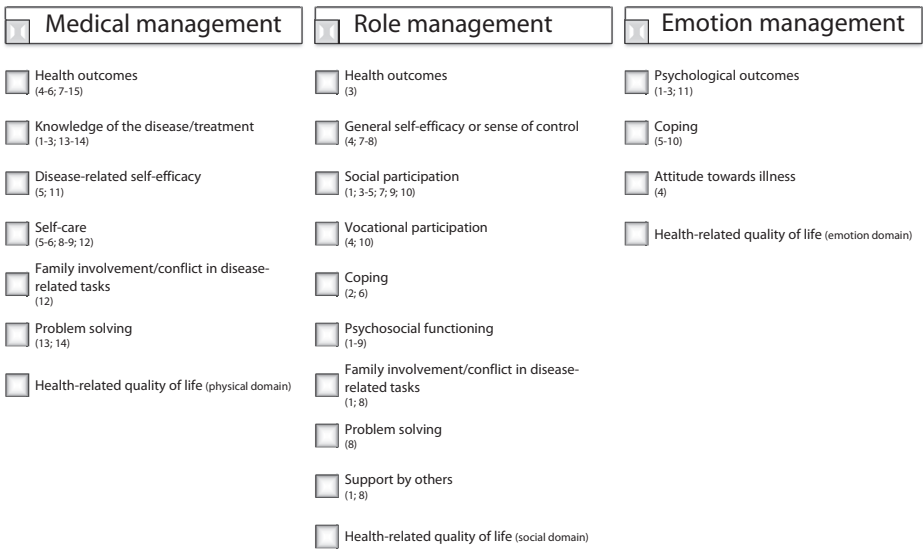


Figure 3 A content-based framework for the selection of (groups of) outcome measures
The numbers presented next to the outcomes correspond to specific content in Table 3.

DISCUSSION AND CONCLUSION

The focus of today’s self-management support

This review revealed that most interventions for young people represented in the literature solely aim at medical management, like interventions for adults [17,113,114]. This is not surprising, because medical tasks form the very core of healthcare. Moreover, these tasks represent common ground for healthcare professionals and people with chronic conditions, since medical consultations without fail will address symptoms and treatments. This may also explain why very few interventions address role management or emotion management alone. Still, the fact that 44% of interventions aim at multiple domains indicates a shift in focus of today’s self-management support for young people with chronic conditions. Healthcare professionals nevertheless are challenged to pay more attention to role management and emotion management.

Six self-management skills match the tasks of medical, role and emotion management: “problem solving, decision making, resource utilization, the formation of a

patient-provider partnership, action planning, and self-tailoring" [14]. Several SMI indeed were directed at developing such skills, e.g., drawing up an action plan. SMI content also seems to match self-management needs of people with chronic conditions, addressing the following processes: 'focusing on illness needs', 'activating resources', and 'living with a chronic illness' [21]. The first is addressed in, for example, SMI aiming to deal with symptoms, the second in SMI helping young people realize when and how to ask support.

However, the above-mentioned processes basically reflect experiences of adult patients. Additional developmental processes or factors will relate to young people's self-management processes as well [115], such as 'determining health needs' and 'communication with the medical team', processes that have been incorporated in the Pediatric Self-management Model [15]. Several SMI indeed target such processes, albeit the Pediatric Self-management Model seems to more narrowly focus on medical management. Young people have to learn to balance or "articulate" [116] self-management tasks, which their parents used to be responsible for. Parental involvement can either hinder or facilitate adolescents' development of self-management [117], and professionals and researchers should be aware of this [15,117]. Some SMI involved parents in the intervention or assessed family interaction or conflict. However, the notion that social context deserves attention when researching self-management, has only recently gained more attention [14,17,19,117-120].

The conceptualization of self-management support

For most of the interventions a theoretical base was not provided, which was also found in other reviews of SMI for both adults and young people [16-18]. The studies that did mention a theoretical base often referred to social learning and cognitive behavioral theories which were also found to underlie SMI for adults [16,17]. Social learning theory argues that people learn from others and in general aims at enhancing self-efficacy [121], while employing an "experiential" approach to self-management [17]. In this view, self-management refers to learning about and believing in yourself, and self-management support facilitates environments that allow to 'learn from others' and gain 'mastery experiences'. On the other hand, cognitive behavioral theory aims to change thoughts and attitudes and ultimately behavior [122], and from this point of view self-management support might be targeted at behavior thought to be beneficial from a medical perspective. In this light, it could represent a more "authoritative" approach to self-management [17]. The different theoretical bases thus represent different views on self-management. For young people, the experiential approach seems more appealing, as telling them what to do is less effective. Young people tend to weigh medical advantages against social disadvantages [4]. Moreover, self-assurance would form a firm basis for healthy behavior [115].

Evaluating self-management interventions: losing focus on what we wish to achieve

Outcome measures or themes varied greatly between studies and even within SMI aiming at a specific diagnostic group, as also reported by others [19]. Health outcomes predominated, which is not surprising given the focus on medical management. Remarkably, however, some studies that focused on a (partially) medical management intervention did not measure health outcomes. Likewise, some medical management interventions were evaluated with psychological outcomes, and an emotion management intervention was evaluated on knowledge of the disease. It seems that current evaluation studies tend to lose focus on what interventions are aimed at, which also hampers conclusions about their effectiveness. Others have recognized this, too, and recommend use of a core set of measurement outcomes to evaluate SMI [19,22,123].

A content-based framework for the selection of outcome measures or groups of outcome measures

The framework presented in Figure 3 proposes a start for a more standardized evaluation approach for SMI for young people with chronic conditions. The outcomes matched those in comparable reviews [18,19], which strengthens the validity of the framework. It may be used to select outcome measures on the basis of the specific content of interventions (as described and numbered per domain in Table 3). However, the classification is broad and measures must be selected based on the goal of the intervention and the measurement properties of the measure. Further sharpening requires more studies into outcomes and measurement instruments.

A fact worth mentioning is the lack of qualitative evaluation studies for SMI. Since qualitative research delves into the contexts of interventions, we recommend future studies to employ a mixed-methods design including a qualitative component. This would help identify ‘effective ingredients’ of SMI and answer the question of what works for whom [124]. The outcome measures in our framework may serve as themes for qualitative research, but themes related to the characteristics of interventions need to be included as well.

Strengths, limitations and other considerations

This study included a systematic and comprehensive search, and was the first to review content of pediatric SMI and classify interventions using a broad self-management framework. Other recent reviews in this field that focus particularly on children and/or adolescents (0-18 years), aimed at researching the effectiveness of SMI and included only RCT’s or studies with repeated measures designs [18,19]. In contrast, our study shed light on the broad content and range of today’s self-management support for young people with chronic conditions. As such, we dealt with the more fundamental question of what

exactly is meant by self-management and self-management support. Furthermore, by matching content of SMI and outcome measures used, a selection tool for future evaluation studies was presented. This also corresponds to the fundamental question of what might be expected from self-management support, and provides a first step towards a much-needed general evaluation framework for different types of interventions.

Using Lorig & Holman's model as a framework to classify SMI seems valid because it is regularly and increasingly referred to when researching self-management. Its' use seems also valid in the case of children, adolescents and young adults, because our results showed that SMI aimed at certain domains of self-management are not exclusive for age groups. This does not imply that certain content is applicable to all ages; for example, vocational participation is more relevant for older adolescents than for younger children. Differences between age groups should therefore be taken into account when evaluating SMI.

This study looked at many types of SMI across a range of chronic conditions. This may be a limitation, because our search terms did not include specific chronic conditions and we might have missed studies that did not include specific key words from our search. However, we feel this is always an issue when performing a systematic literature review which probably is more related to the way databases are organized than to the sensitivity of our search strategy. Furthermore, our non-categorical approach may also be a strength, because it enables a more general view on self-management irrespective of diagnosis. This is relevant because these young people face comparable challenges and similar adaptive tasks irrespective of type of condition [4,115]. Yet, they may need different support in view of individual socio-demographic and psychological factors [117]. In this respect young people within a specific diagnostic group may differ as much as those in different diagnostic groups [125]. Interestingly, only 7% of the SMI found in the present study were developed for chronic conditions in general. Since specific paediatric diagnostic groups are often small, achieving effectiveness and cost-effectiveness of disease-specific SMI would be problematic [20]. A more generic approach with a disease-specific component for different diagnostic groups may be more convenient [4], and should not be problematic since the core elements of self-management support are the same across different approaches [126]. An example is the 'Skills for Growing Up' tool developed in pediatric rehabilitation and adjusted on the disease-specific content for use in pediatric nephrology [127].

Gaining insight into effectiveness of different types of interventions was hindered by the heterogeneity in outcome measures. Most studies in this review were from Western countries, and interventions for young people with diabetes or asthma predominated. These conditions generally include a burdensome medical regimen, which may have added to the focus on medical management. Yet, a sub-analysis (not presented in this

paper) showed that even after removing diabetes and asthma studies, the focus still remained on medical management alone than on other self-management domains.

CONCLUSIONS

The content of different SMI relate to self-management tasks of people with chronic conditions, and self-management skills they should develop. Yet, healthcare professionals should be aware of the importance of role and emotion management in self-management. Also, in view of these young people's developmental challenges, an experiential approach focusing on learning (from others) and 'mastery experiences' might be more appropriate in pediatric care.

Future evaluations should provide details about theoretical bases of interventions, and should match evaluation outcomes and themes to intervention content and characteristics. The content-based evaluation framework presented in this study may assist in this, while further research might help identify valid outcome measurement instruments. Mixed-methods research is recommended to gain more insights in the contexts, including social context, and working mechanisms of SMI.

Practice implications

Self-management support is important for people with chronic conditions to help them deal with their condition in daily life. This is even more pertinent to young people growing up with chronic conditions, who have to face the normal tasks of development (e.g., acquiring autonomy) and have to engage in lifelong medical management of their condition. Therefore, it remains important to research the effects of SMI. Future evaluation studies should make sure that their evaluation outcomes match with the content and characteristics of the SMI, and may benefit from the use of more generic outcome measures in SMI evaluation. Our content-based evaluation framework and overview of SMI content, characteristics and outcomes may assist researchers in doing so. Furthermore, our overview may give clinicians and other healthcare professionals insight into the broad range of self-management and self-management support, and as such may assist them in determining the breadth and focus of the support they provide.

ACKNOWLEDGEMENTS

The study was part of the Self-management and Participation Innovation Lab, supported by SIA-RAAK, the Foundation Innovation Alliance with funding from the Dutch ministry of Education, Culture and Science (OCW) (PRO-02-025). The authors thank the

information specialist of the Erasmus MC medical library, Wichor Bramer, for his assistance in defining the search strategy. Ko Hagoort is thanked for his editorial assistance. Finally, the members of our Self-management & Participation Research Group (Rotterdam University of Applied Sciences) and Hester van de Bovenkamp (Erasmus University Rotterdam) are thanked for their comments on an earlier draft of this paper.

REFERENCES

1. The Child and Adolescent Health Measurement Initiative. National Survey of Children with Special Health Care Needs [<http://www.childhealthdata.org/learn/NS-CSHCN>]
2. 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]. *Tijdschrift voor Kindergeneeskunde [Dutch Journal of Pediatrics]* 2007, 75: 138-342.
3. Gijzen R, van Oostrom SH, Schellevis FC: Hoeveel mensen hebben één of meer chronische ziekten? [How many people have on or more chronic diseases?]. In: *Volksgezondheid Toekomst Verkenning, Nationaal Kompas Volksgezondheid*. Bilthoven: RIVM; 2013.
4. Van Staa AL: On Your Own Feet: Adolescents with chronic conditions and their preferences and competencies for care (Doctoral dissertation). Rotterdam: Rotterdam University; 2012.
5. Ratanachadawan Y: A response to T. Koch, P. Jenkin & D. Kralik (2004) Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2005, 52:571-572.
6. Harvey PW, Petkov JN, Misan G, Fuller J, Battersby MW, Cayetano TN, Warren K, Holmes P: Self-management support and training for patients with chronic and complex conditions improves health-related behaviour and health outcomes. *Aust Health Rev* 2008, 32:330-338.
7. Wagner EH: Chronic disease management: what will it take to improve care for chronic illness? *Eff Clin Pract* 1998, 1:2-4.
8. Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, Hoes A, Schuurmans M: Self-management: One size does not fit all. *Patient Educ Couns* 2013, 92:134-137.
9. Huber M, Knottnerus JA, Green L, van der Horst H, Jadad AR, Kromhout D, Leonard B, Lorig K, Loureiro MI, van der Meer JWM, Schnabel P, Smith R, van Weel C, Smid H: How should we define health? *BMJ* 2011, 343:d4163.
10. Audulv Å, Norbergh KG, Asplund K, Hörnsten Å: An ongoing process of inner negotiation – a Grounded Theory study of self-management among people living with chronic illness. *J Nurs Healthc Chronic Illn* 2009, 1:283-293.
11. Paterson BL: The shifting perspectives model of chronic illness. *J Nurs Scholarsh* 2001, 33:21-26.
12. Barlow J, Wright C, Sheasby J, Turner A, Hainsworth J: Self-management approaches for people with chronic conditions: a review. *Patient Educ Couns* 2002, 48:177-87.
13. Richard AA, Shea K: Delineation of self-care and associated concepts. *J Nurs Scholarsh* 2011, 43: 255-64.
14. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26:1-7.
15. Modi AC, Pai AL, Hommel KA, Hood KK, Cortina S, Hilliard ME, Guilfoyle SM, Gray WN, Drotar D: Pediatric self-management: a framework for research, practice, and policy. *Pediatr* 2012, 129: e473-485.
16. Newman S, Steed L, Mulligan K: Chronic physical illness: Self-management and behavioural interventions. New York: Open University Press; 2009.
17. Jonsdottir H: Self-management programmes for people living with chronic obstructive pulmonary disease: a call for a reconceptualisation. *J Clin Nurs* 2013, 22:621-637.
18. Kirk S, Beatty S, Callery P, Gellatly J, Milnes L, Prymachuk S: The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child Care Health Dev* 2012, 39:305-324.

19. Lindsay S, Kingsnorth S, McDougall C, Keating H: A systematic review of self-management interventions for children and youth with physical disabilities. *Disabil Rehabil* 2014, 36:276-288.
20. Newman S, Mulligan K, Steed L: What is meant by self-management and how can its efficacy be established? *Rheumatol* 2001, 40:1-4.
21. Schulman-Green D, Jaser S, Martin F, Alonzo A, Grey M, McCorkle R, Redeker NS, Reynolds N, Whittemore R: Processes of self-management in chronic illness. *J Nurs Scholarsh* 2012, 44(2):136-144.
22. Nolte S, Elsworth GR, Newman S, Osborne RH. Measurement issues in the evaluation of chronic disease self-management programs. *Qual Life Res* 2013, 22:1655-1664.
23. Grant MJ, Booth A. A typology of reviews: an analysis of 14 review types and associated methodologies. *Health Info Libr J* 2009, 26:91-108.
24. Moher D, Liberati A, Tetzlaff J, Altman DG, PRISMA Group: Preferred Reporting for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *Int J Surg* 2010, 8(5):336-341.
25. Scottish Intercollegiate Guidelines Network. Critical appraisal: notes and checklists [<http://www.sign.ac.uk/methodology/checklists.html#>]
26. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care* 2007, 19:349-357.
27. Rhee H, Belyea MJ, Hunt JF, Brasch J. Effects of a peer-led asthma self-management program for adolescents. *Arch Pediatr Adolesc Med* 2011, 165:513-519.
28. Rhee H, McQuillan BE, Belyea MJ: Evaluation of a peer-led asthma self-management program and benefits of the program for adolescent peer leaders. *Respir Care* 2012, 57:2082-2089.
29. Ng SM, Li AM, Lou VW, Tso IF, Wan PY, Chan DF: Incorporating family therapy into asthma group intervention: a randomized waitlist-controlled trial. *Fam process* 2008, 47:115-130.
30. Jan RL, Wang JY, Huang MC, Tseng SM, Su HJ, Liu LF: An internet-based interactive telemonitoring system for improving childhood asthma outcomes in Taiwan. *Telemed J E Health* 2007, 13:257-268.
31. Burkhart PV, Rayens MK, Oakley MG, Abshire DA, Zhang M: Testing an intervention to promote children's adherence to asthma self-management. *J Nurs Scholarsh* 2007, 39:133-140.
32. Butz A, Pham L, Lewis L, Lewis C, Hill K, Walker J, Winkelstein M: Rural children with asthma: Impact of a parent and child asthma education program. *J Asthma* 2005, 42:813-821.
33. Clark NM, Shah S, Dodge JA, Thomas LJ, Andridge RR, Little RJA: An evaluation of asthma interventions for preteen students. *J Sch Health* 2010, 80:80-87.
34. Huss K, Winkelstein M, Nanda J, Naumann PL, Sloand ED, Huss RW: Computer game for inner-city children does not improve asthma outcomes. *J Pediatr Health Care* 2003, 17:72-78.
35. Joseph CL, Peterson E, Havstad S, Johnson CC, Hoerauf S, Stringer S, Gibson-Scipio W, Ownby DR, Elston-Lafata J, Pallonen U, Strecher V, Asthma in Adolescents Research Team: A web-based, tailored asthma management program for urban African-American high school students. *Am J Respir Crit Care Med* 2007, 175:888-895.
36. Krishna S, Francisco BD, Balas EA, Konig P, Graff GR, Madsen RW: Internet-enabled interactive multimedia asthma education program: a randomized trial. *Pediatr* 2003, 111:503-510.
37. Pulgaron ER, Salamon KS, Patterson CA, Barakat LP: A problem-solving intervention for children with persistent asthma: A pilot of a randomized trial at a pediatric summer camp. *J Asthma* 2010, 47:1031-1039.
38. Shames RS, Sharek P, Mayer M, Robinson TN, Hoyte EG, Gonzalez-Hensley F, Bergman DA, Umetsu DT: Effectiveness of a multicomponent self-management program in at-risk, school-aged children with asthma. *Ann Allergy Asthma Immunol* 2004, 92:611-618.
39. Velsor-Friedrich B, Pigott T, Srof B: A practitioner-based asthma intervention program with African American inner-city school children. *J Pediatr Health Care* 2005, 19:163-171.

40. Hampel P, Rudolph H, Stachow R, Petermann F: Multimodal patient education program with stress management for childhood and adolescent asthma. *Patient Educ Couns* 2003, 49:59-66.
41. Chiang LC, Ma WF, Huang JL, Tseng LF, Hsueh KC: Effect of relaxation-breathing training on anxiety and asthma signs/symptoms of children with moderate-to-severe asthma: A randomized controlled trial. *Int J Nurs Stud* 2009, 46:1061-1070.
42. Beebe A, Gelfand EW, Bender B: A randomized trial to test the effectiveness of art therapy for children with asthma. *J Allergy Clin Immunol* 2010, 126:263-U14.
43. Clark NM, Brown R, Joseph CL, Anderson EW, Liu M, Valerio MA: Effects of a comprehensive school-based asthma program on symptoms, parent management, grades, and absenteeism. *Chest* 2004, 125:1674-1679.
44. Kouba J, Velsor-Friedrich B, Militello L, Harrison PR, Becklenberg A, White B, Surya S, Ahmed A: Efficacy of the I Can Control Asthma and Nutrition Now (ICAN) Pilot Program on Health Outcomes in High School Students With Asthma. *J School Nurs* 2013, 29:235-247.
45. Rami B, Popow C, Horn W, Waldhoer T, Schober E: Telemedical support to improve glycemic control in adolescents with type 1 diabetes mellitus. *Eur J Pediatr* 2006, 165:701-705.
46. Cafazzo JA, Casselman M, Hamming N, Katzman DK, Palmert MR: Design of an mHealth App for the Self-management of Adolescent Type 1 Diabetes: A Pilot Study. *J Med Internet Res* 2012, 14:171-183.
47. Løding RN, Wold JE, Skavhaug Å: Experiences with a group intervention for adolescents with type 1 diabetes and their parents. *Eur Diab Nursing* 2008, 5:9-14.
48. Franklin VL, Waller A, Pagliari C, Greene SA: A randomized controlled trial of Sweet Talk, a text-messaging system to support young people with diabetes. *Diabet Med* 2006, 23:1332-1338.
49. Martin C, Liveley K, Whitehead K: A health education group intervention for children with type 1 diabetes. *J Diabetes Nurs* 2009, 13:32-37.
50. Gerber BS, Solomon MC, Shaffer TL, Quinn MT, Lipton RB: Evaluation of an internet diabetes self-management training program for adolescents and young adults. *Diabetes Technol Ther* 2007, 9:60-67.
51. Grey M, Whittemore R, Jaser S, Ambrosino J, Lindemann E, Liberti L, Northrup V, Dziura J: Effects of coping skills training in school-age children with type 1 diabetes. *Res Nurs Health* 2009, 32:405-418.
52. Hanauer DA, Wentzell K, Laffel N, Laffel LM: Computerized Automated Reminder Diabetes System (CARDS): e-mail and SMS cell phone text messaging reminders to support diabetes management. *Diabetes Technol Ther* 2009, 11:99-106.
53. Kumar VS, Wentzell KJ, Mikkelsen T, Pentland A, Laffel LM: The DAILY (Daily Automated Intensive Log for Youth) trial: a wireless, portable system to improve adherence and glycemic control in youth with diabetes. *Diabetes Technol Ther* 2004, 6:445-453.
54. Laffel LM, Vangsness L, Connell A, Goebel-Fabbri A, Butler D, Anderson BJ: Impact of ambulatory, family-focused teamwork intervention on glycemic control in youth with type 1 diabetes. *J Pediatr*. 2003, 142:409-416.
55. Nansel TR, Iannotti RJ, Simons-Morton BG, Cox C, Plotnick LP, Clark LM, Zeitzorff L: Diabetes personal trainer outcomes: short-term and 1-year outcomes of a diabetes personal trainer intervention among youth with type 1 diabetes. *Diabetes Care* 2007, 30:2471-2477.
56. Wang YC, Stewart S, Tuli E, White P: Improved glycemic control in adolescents with type 1 diabetes mellitus who attend diabetes camp. *Pediatr Diabetes* 2008, 9:29-34.
57. Wysocki T, Harris MA, Buckloh LM, Mertlich D, Lochrie AS, Mauras N, White NH: Randomized trial of behavioral family systems therapy for diabetes: maintenance of effects on diabetes outcomes in adolescents. *Diabetes Care* 2007, 30:555-560.

58. Channon SJ, Huws-Thomas MV, Rollnick S, Hood K, Cannings-John RL, Rogers C, Gregory JW: A multicenter randomized controlled trial of motivational interviewing in teenagers with diabetes. *Diabetes Care* 2007, 30:1390-1395.
59. Newton KT, Ashley A. Pilot study of a web-based intervention for adolescents with type 1 diabetes: *J Telemed Telecare* 2013, 19:443-449.
60. Herbert LJ, Sweeney R, Kelly KP, Holmes C, Streisand R: Using Qualitative Methods to Evaluate a Family Behavioral Intervention for Type 1 Diabetes. *J Pediatr Health Care* 2014, 28:376-85.
61. Barrera M, Schulte F: A group social skills intervention program for survivors of childhood brain tumors. *J Pediatr Psychol* 2009, 34:1108-1118.
62. Canada AL, Schover LR, Li Y: A pilot intervention to enhance psychosexual development in adolescents and young adults with cancer. *Pediatr Blood Cancer* 2007, 49:824-828.
63. Jones JK, Kamani SA, Bush PJ, Hennessy KA, Marfatia A, Shad AT: Development and evaluation of an educational interactive CD-ROM for teens with cancer. *Pediatr Blood Cancer* 2010, 55:512-519.
64. Maurice-Stam H, Silberbusch LM, Last BF, Grootenhuis MA: Evaluation of a psycho-educational group intervention for children treated for cancer: a descriptive pilot study. *Psychooncology* 2009, 18:762-766.
65. Kato PM, Cole SW, Bradlyn AS, Pollock BH: A video game improves behavioral outcomes in adolescents and young adults with cancer: a randomized trial. *Pediatr* 2008, 122:e305-317.
66. Stulemeijer M, De Jong LWAM, Fiselier TJW, Hoogveld SWB, Bleijenberg G: Cognitive behaviour therapy for adolescents with chronic fatigue syndrome: Randomised controlled trial. *Br Med J* 2005, 330:14-17.
67. Hilberink SR, Kruijver E, Wiegerink DJHG, Vliet Vlieland TPM: A Pilot Implementation of an Intervention to Promote Sexual Health in Adolescents and Young Adults in Rehabilitation. *Sex Disabil* 2013:1-20.
68. Torok S, Kokonyei G, Karolyi L, Ittzes A, Tomcsanyi T: Outcome effectiveness of therapeutic recreation camping program for adolescents living with cancer and diabetes. *J Adolesc Health* 2006, 39: 445-7.
69. Bekesi A, Torok S, Kokonyei G, Bokretas I, Szentes A, Telepoczki G: Health-related quality of life changes of children and adolescents with chronic disease after participation in therapeutic recreation camping program. *Health Qual Life Outcomes* 2011, 9.
70. Curle C, Bradford J, Thompson J, Cawthron P: Users' views of a group therapy intervention for chronically ill or disabled children and their parents: Towards a meaningful assessment of therapeutic effectiveness. *Clin Child Psychol Psychiatry* 2005, 10:509-527.
71. Creedy D, Collis D, Ludlow T, Cosgrove S, Houston K, Irvine D, Fraser J, Moloney S: Development and evaluation of an intensive intervention program for children with a chronic health condition: a pilot study. *Contemp Nurse* 2004, 18:46-56.
72. Raghavendra P, Newman L, Grace E, Wood D: 'I could never do that before': Effectiveness of a tailored Internet support intervention to increase the social participation of youth with disabilities. *Child: Care, Health and Development* 2013, 39:552-561.
73. Hechler T, Dobe M, Damschen U, Blankenburg M, Schroeder S, Kosfelder J, Zernikow B: The Pain Provocation Technique for Adolescents with Chronic Pain: Preliminary Evidence for Its Effectiveness. *Pain Med (USA)* 2010, 11:897-910.
74. Eccleston C, Malleon PN, Clinch J, Connell H, Sourbut C: Chronic pain in adolescents: evaluation of a programme of interdisciplinary cognitive behaviour therapy. *Arch Dis Child* 2003, 88:881-885.

75. Palermo TM, Wilson AC, Peters M, Lewandowski A, Somhegyi H: Randomized controlled trial of an Internet-delivered family cognitive-behavioral therapy intervention for children and adolescents with chronic pain. *Pain* 2009,146:205-213.
76. Merlijn VPBM, Hunfeld JAM, van der Wouden JC, Hazebroek-Kampschreur AAJM, van Suijlekom-Smit LWA, Koes BW, Passchier J: A cognitive-behavioural program for adolescents with chronic pain-a pilot study. *Patient Educ Couns* 2005, 59:126-134.
77. Fales J, Palermo TM, Law EF, Wilson AC: Sleep Outcomes in Youth With Chronic Pain Participating in a Randomized Controlled Trial of Online Cognitive-Behavioral Therapy for Pain Management. *Behav Sleep Med* 2014, [epub ahead of print].
78. Newcombe PA, Dunn TL, Casey LM, Sheffield JK, Petsky H, Anderson-James S, Chang AB: Breathe Easier Online: Evaluation of a randomized controlled pilot trial of an Internet-based intervention to improve well-being in children and adolescents with a chronic respiratory condition. *J Med Internet Res* 2012, 14:115-126.
79. Downs JA, Roberts CM, Blackmore AM, Le Souef PN, Jenkins SC: Benefits of an education programme on the self-management of aerosol and airway clearance treatments for children with cystic fibrosis. *Chronic Respir Dis* 2006, 3:19-27.
80. MacDonald K, Greggans A: 'Cool friends': An evaluation of a community befriending programme for young people with cystic fibrosis. *J Clin Nurs* 2010, 19:2406-2414.
81. Christian BJ, D'Auria JP: Building life skills for children with cystic fibrosis: Effectiveness of an intervention. *Nurs Res* 2006, 55:300-307.
82. Davis MA, Quittner AL, Stack CM, Yang MCK: Controlled evaluation of the STARBRIGHT CD-ROM program for children and adolescents with cystic fibrosis. *J Pediatr Psychol* 2004, 29:259-267.
83. Staab D, Diepgen TL, Fartasch M, Kupfer J, Lob-Corzilius T, Ring J, Scheewe S, Scheidt R, Schmid-Ott G, Schnopp C, Szczepanski R, Werfel T, Wittenmeier M, Wahn U, Gieler U: Age related, structured educational programmes for the management of atopic dermatitis in children and adolescents: multicentre, randomised controlled trial. *BMJ* 2006, 332:933-8.
84. Meade MA, Creer TL, Mahan JD: A self-management program for adolescents and children with renal transplantation. *J Clin Psychol Med Settings* 2003, 10:165-171.
85. Payne ME, Eaton CK, Mee LL, Blount RL: Promoting medication adherence and regimen responsibility in two adolescents on hemodialysis for end-stage renal disease: A case study. *Clin Case Stud* 2013, 12:95-110.
86. Sattoe JNT, Jedeloo S, Van Staa A: Effective peer-to-peer support for young people with end-stage renal disease: A mixed methods evaluation of Camp COOL. *BMC Nephrol* 2013, 14:279.
87. Cushner-Weinstein S, Berl M, Salpekar JA, Johnson JL, Pearl PL, Conry JA, Kolodgie M, Scully A, Gaillard WD, Weinstein SL: The benefits of a camp designed for children with epilepsy: evaluating adaptive behaviors over 3 years. *Epilepsy Behav* 2007, 10:170-178.
88. Bultas MW, Budhathoki C, Balakas K: Evaluation of child and parent outcomes after a pediatric cardiac camp experience. *Journal for Specialists in Pediatric Nursing* 2013, 18:320-328.
89. Smith Fawzi MC, Eustache E, Oswald C, Louis E, Surkan PJ, Scanlan F, Hook S, Mancuso A, Mukherjee JS: Psychosocial support intervention for HIV-affected families in Haiti: Implications for programs and policies for orphans and vulnerable children. *Soc Sci Med* 2012, 74:1494-1503.
90. Berrien VM, Salazar JC, Reynolds E, McKay K, Group HIVMAI: Adherence to antiretroviral therapy in HIV-infected pediatric patients improves with home-based intensive nursing intervention. *AIDS Patient Care STDs* 2004, 18:355-363.
91. Hayutin LG, Blount RL, Lewis JD, Simons LE, McCormick ML: Skills-based group intervention for adolescent girls with inflammatory bowel disease. *Clin Case Stud* 2009, 8:355-365.

92. Dufresne H, Hady-Rabia S, Taieb C, Bodemer C: Importance of therapeutic patient education in ichthyosis: Results of a prospective single reference center study. *Orphanet J Rare Dis* 2013, 8:113.
93. Kashikar-Zuck S, Ting TV, Arnold LM, Bean J, Powers SW, Graham TB, Passo MH, Schikler KN, Hashkes PJ, Spalding S, Lynch-Jordan AM, Banez G, Richards MM, Lovell DJ: Cognitive behavioral therapy for the treatment of juvenile fibromyalgia: A multisite, single-blind, randomized, controlled clinical trial. *Arthritis Rheum* 2012, 64:297-305.
94. Kashikar-Zuck S, Flowers SR, Strotman D, Sil S, Ting TV, Schikler KN: Physical activity monitoring in adolescents with juvenile fibromyalgia: findings from a clinical trial of cognitive-behavioral therapy. *Arthritis Care Res (Hoboken)* 2013, 65:398-405.
95. Kashikar-Zuck S, Sil S, Lynch-Jordan AM, Ting TV, Peugh J, Schikler KN, Hashkes PJ, Arnold LM, Passo M, Richards-Mauze MM: Changes in pain coping, catastrophizing, and coping efficacy after cognitive-behavioral therapy in children and adolescents with juvenile fibromyalgia. *J Pain* 2013, 14:492-501.
96. Stinson JN, McGrath PJ, Hodnett ED, Feldman BM, Duffy CM, Huber AM, Tucker LB, Hetherington CR, Tse SM, Spiegel LR, Campillo S, Gill NK, White ME: An internet-based self-management program with telephone support for adolescents with arthritis: a pilot randomized controlled trial. *J Rheumatol* 2010, 37:1944-1952.
97. Hackett J, Johnson B, Shaw KL, McDonagh JE: Friends united: An evaluation of an innovative residential self-management programme in adolescent rheumatology. *Br J Occup Ther* 2005, 68: 567-573.
98. McDonagh JE, Southwood TR, Shaw KL: The impact of a coordinated transitional care programme on adolescents with juvenile idiopathic arthritis. *Rheumatology (UK)* 2007, 46:161-168.
99. Fuchs CE, Van Geelen SM, Hermans HJM, Van De Putte EM, Van Geel R, Sinnema G, Kuis W: Psychological intervention for adolescents with juvenile idiopathic arthritis: For whom and when? *J Rheumatol* 2013, 40:528-534.
100. Connelly M, Rapoff MA, Thompson N, Connelly W: Headstrong: A pilot study of a CD-ROM intervention for recurrent pediatric headache. *J Pediatr Psychol* 2006, 31:737-747.
101. Ten Hoedt AE, Hollak CE, Boelen CC, van der Herberg-van de Wetering NAP, Ter Horst NM, Jonkers CF, Wijburg FA, Bosch AM: "MY PKU": increasing self-management in patients with phenylketonuria. A randomized controlled trial. *Orphanet J Rare Dis* 2011, 6:48.
102. Hojberg AL, Steffensen BF: Developing and maintaining of user-defined personal competencies among young adults with congenital physical disability. *Dev Neurorehabil* 2010, 11:225-235.
103. Xenakis N, Goldberg J. The Young Women's Program: A health and wellness model to empower adolescents with physical disabilities. *Disabil Health J* 2010, 3:125-129.
104. Verhoef JAC, Roebroek ME, van Schaardenburgh N, Floothuis MCSG, Miedema HS: Improved occupational performance of young adults with a physical disability after a vocational rehabilitation intervention. *J Occup Rehabil* 2014, 24:42-51.
105. Barakat LP, Schwartz LA, Salamon KS, Radcliffe J: A family-based randomized controlled trial of pain intervention for adolescents with sickle cell disease. *J Pediatr Hematol Oncol* 2010, 32:540-547.
106. McClellan CB, Schatz JC, Puffer E, Sanchez CE, Stancil MT, Roberts CW: Use of handheld wireless technology for a home-based sickle cell pain management protocol. *J Pediatr Psychol* 2009, 34: 564-573.
107. Koontz K, Short AD, Kalinyak K, Noll RB: A randomized, controlled pilot trial of a school intervention for children with sickle cell anemia. *J Pediatr Psychol* 2004, 29:7-17.
108. Yoon SL, Godwin A: Enhancing self-management in children with sickle cell disease through playing a CD-ROM educational game: a pilot study. *Pediatr Nurs* 2007, 33:60-72.

109. Dobson CE, Byrne MW. Using Guided Imagery to Manage Pain in Young Children with Sickle Cell Disease. *Am J Nurs* 2014, 114:26-36.
110. Betz CL, Smith K, Macias K: Testing the transition preparation training program: A randomized controlled trial. *Int J Child Adolesc health* 2011, 3:595-607.
111. O'Mahar K, Holmbeck GN, Jandasek B, Zukerman J: A camp-based intervention targeting independence among individuals with spina bifida. *J Pediatr Psychol* 2010, 35:848-856.
112. Holbein CE, Murray CB, Psihogios AM, Wasserman RM, Essner BS, O'Hara LK, Holmbeck GN: A camp-based psychosocial intervention to promote independence and social function in individuals with spina bifida: Moderators of treatment effectiveness. *J Pediatr Psychol* 2013, 38:412-424.
113. Udlis KA. Self-management in chronic illness: concept and dimensional analysis. *J Nurs Healthc Chronic Illn* 2011, 3(2):130-139.
114. Koch T, Jenkin P, Kralik D: Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2004, 48: 484-492.
115. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369:1481-1489.
116. Corbin JM, Strauss AL: Unending work and care: managing chronic illness at home. San Francisco: Jossey-Bass, 1988.
117. Kirk S, Beatty S, Callery P, Milnes L, Prymachuk S: Perceptions of effective self-care support for children and young people with long-term conditions. *J Clin Nurs* 2012, 21:1974-1987.
118. Thirsk LM, Clark AM: What is the 'self' in chronic disease self-management? *Int J Nurs Stud* 2014, 51: 691-693.
119. Audulv A, Asplund K, Norbergh KG: The integration of chronic illness self-management. *Qual Health Res* 2012, 22:332-345.
120. Ong BN, Rogers A, Kennedy A, Bower P, Sanders T, Morden A, Cheraghi-Sohi S, Richardson JC, Stevenson F: Behaviour change and social blinkers? The role of sociology in trials of self-management behaviour in chronic conditions. *Sociol Health Illn* 2014, 36:226-238.
121. Bandura A: Social Learning Theory. New York: General Learning Press, 1977.
122. Hupp SDA, Reitman D, Jewell JD: Cognitive behavioral theory. New York: John Wiley & Sons, Inc., 2008.
123. Nolte S, Osborne RH. A systematic review of outcomes of chronic disease self-management interventions. *Qual Life Res* 2013, 22:1805-1816.
124. Coster S, Norman I: Cochrane reviews of educational and self-management interventions to guide nursing practice: a review. *Int J Nurs Stud* 2009, 46:508-528.
125. Stein RE, Jessop DJ: What diagnosis does not tell: the case for a noncategorical approach to chronic illness in childhood. *Soc Sci Med* 1989, 29:769-778.
126. Novak M, Costantini L, Schneider S, Beanlands H: Approaches to self-management in chronic illness. *Semin Dial* 2013, 26:188-194.
127. Sattoe JNT, Hilberink SR, Peeters MAC, van Staa A. 'Skills for Growing up': Supporting Autonomy in Young People with Kidney Disease. *J Ren Care* 2014, 40:131-139.

Appendix C4.1 Overview of study and intervention characteristics (n=86)*

Study characteristics		Participants					Intervention characteristics			Theoretical base		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	
Barakat et al. 2010	United States of America	RCT	Sickle cell disease	14.1	Individual	Home environment	Doctoral students in clinical psychology or psychologists	Educational sessions	Daily paper-and-pencil pain diaries, homework, review of homework, and biweekly check-in telephone calls	Sessions take 90 minutes. Intervention: 4 sessions (3 sessions, 2 weeks apart with a booster session 1 month later)	MM	NA**
Barrera & Schulte 2009	Canada	Cohort	Cancer	12.7	Group	Clinic (Pediatric center)	Psychologist, and clinical assistants	Skills training sessions	Fun activities and games guided by cognitive behavior strategies and expressive therapies such as music, art, and drama.	A 2-hr group session once a week for 8 weeks.	RM and EM	NA
Beebe et al. 2010	United States of America	RCT	Asthma	7-14	Group	School	NA	Art therapy sessions	Art making, and sharing feelings related to the art created.	A 1-hour session for 7 weeks.	MM and EM	NA
Bekeci et al. 2011	Hungary	Cohort	Oncology, diabetes, juvenile immune arthritis	13.3	Group	Camp, and clinic (medical centre)	Volunteers (selected and trained, profession not mentioned)	Camping program	Adventure-based program with activities and fun.	NA	RM	NA
Berrien et al. 2004	United States of America	RCT	HIV	10.0	Individual home visit	Home	Experienced registered nurse	Educational sessions	A comic book, a video, filling out notebooks with rewards (prizes) if they did, and eventually role reversal.	NA	MM	NA
Betz et al. 2011	United States of America	RCT	Spina Bifida	16.0	Group	Clinic	Trainer (not mentioned if this was a healthcare professional)	Skills training sessions	Making a transition plan, and practicing strategies to obtain goals, i.e. role playing, one-to-one consultation, coaching, reinforced learning, use of audio visual aids, accessing the Internet, and mentored learning.	2-day workshop	MM and RM	NA
Bultas et al. 2013	United States of America	Cohort	Heart disease	8-15	Group	Camp	Pediatric nurses and cardiologists	NA	NA	5 days and 4 nights	NA	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants			Intervention characteristics			Element(s)	Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)				
Burkhardt et al. 2007	United States of America	RCT	Asthma	7-12	Group sessions	Clinic (university center for nursing research)	Research associate	Educational sessions	Educational videos, and homework assignments	NA	MM	(Cognitive) social learning theory
Butz et al. 2005	United States of America	RCT	Asthma	8.0	Group sessions	School	Asthma educator (not mentioned if this was a healthcare professional)	Skills training sessions	Practiced and demonstrated specific skills (peak flow meter and inhaler technique). Discussions with the educator, and a coloring book, a peak flow meter, and a spacer device were given to children.	4 hours of instruction during two separate sessions	MM	NA
Cafazzo et al. 2012	Canada	Cohort	Diabetes	14.9	Individual	Online	Clinicians	Telemanagement system	Reminders, cueing, social media communication, and the gamification of routine management tasks.	NA	MM	NA
Canada et al. 2007	United States of America	Cohort	Cancer	21.3	Individual	Clinic	Doctoral level clinical psychologist	Educational (and support) sessions	Workbook, homework, and follow-up phone calls	NA	RM and EM	NA
Channon et al. 2007	United Kingdom	RCT	Diabetes	15.3	Individual	Home	Nurses, and a health psychologist	Motivational interviewing sessions	Awareness building, problem solving, and goal-setting.	The frequency and location of appointments was determined by the participants to fit with the patient- driven principles of motivational interviewing. Interviews lasts between 20 and 60 min.	NA	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics			Theoretical base		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM
Chiang et al. 2009	Taiwan	RCT	Asthma	6–14	Individual	Home, and clinic	Nursing graduate student	Skills training sessions	Relaxation training	During the 12-week intervention, participants practiced relaxation for 30 min at least three times per week.	MM
Christian & D'Auria 2006	United States of America	RCT	Cystic Fibrosis	8–12	Individual home visit, and group session	Home, and clinic	NA	Educational sessions and support sessions	A computer software program, a notebook with worksheets, and a journal (individual). Discussion and peer contact (group).	NA	RM
Clark et al. 2004	United States of America	RCT	Asthma	7–10	Group	School	NA	Educational sessions	Handouts and homework assignments, group discussions, asthma action plan.	NA	MM
Clark et al. 2010	United States of America	RCT	Asthma	11.9	Group lessons	School	Graduate students, and community leaders trained in the program (profession not mentioned)	School program	Interactive problem-solving activities, Peer education component (not necessarily Asthma patient) in which peers educated asthma awareness to participants through skits, creative dramas or music.	NA	MM, RM and EM
Connelly et al. 2006	United States of America	RCT	Migraine	7–12	Individual	Home	NA	CD-ROM	Educational modules, active coping plan, and gamification (with feedback).	NA	MM and EM
Creedy et al. 2004	Australia	Cohort	Various chronic conditions	10–14	Group sessions	NA	Graduates of a leadership training course co-facilitated the intervention with healthcare professionals	Family sessions (parallel but separate sessions for parents and children).	Peer support	8 weeks	RM and EM

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants		Intervention characteristics			Element(s)	Timing	Domain(s) of SM	Theoretical base	
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists					Format(s)
Curle et al. 2005	United States of America	Qualitative	Various chronic conditions	7-12	Group sessions	Clinic (specialized unit)	A clinical psychologist, occupational therapist, mental health nurses, and specialist pediatric nurses or social workers	Family sessions (parallel but separate sessions for parents and children).	Play therapy, narrative therapy, relaxation training and group work.	6-8 sessions	EM	Cognitive behavioral theory, and Systemic theory
Cushman-Weinstein et al. 2007	United States of America	Cohort	Epilepsy	7-17	Group sessions	Camp	Medical professionals, and counseling staff	Camping program	Traditional camp activities (rope course, swimming, arts, crafts etc.) combined with activities with condition-specific goals and relevance, and support groups (peer contacts).	7 days	MM, RM and EM	NA
Davis et al. 2004	United States of America	RCT	Cystic Fibrosis	7-17	Individual	Home	NA	CD-ROM	Educational modules	NA	MM, RM and EM	NA
Dobson 2014	United States of America	Cohort	Sickle cell disease	6-8	Individual	Clinic and home	Child life specialist	Diary and guided imagery, including one training session	Participants kept a diary, recording their daily activities and all pain episodes, including location and intensity, as well as strategies for management. The diaries included blank daily pages with the instruction, "Use one page a day to describe your activities and your pain, and one page to draw a picture".	The training sessions lasted from 15-45 minutes	MM	Cognitive behavioral therapy
Downs et al. 2006	Australia	RCT	Cystic Fibrosis	8.4	Individual	Clinic	Caregivers	Cognitive behavioral therapy sessions	NA	10-week period, with each of the 10 chapters taking approximately 20 minutes to complete	MM	(Cognitive) social learning theory

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants		Intervention characteristics			Theoretical base				
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	
Dufresne et al. 2013	France	Cohort	Ichthyosis	6 and older	Group	Reception centre	Physician and paramedic team member	Two sessions called 123 Tempeau sessions And a game with a set of multiple-choice questions was used.	Children and siblings: "What is ichthyosis? Why do I need the cream? Why am I sick? Is it normal to have pain? What about school and me? What about the hospital and me?" Parents, children and siblings > 12 years: "What is ichthyosis? What are the treatments? What is genetics? What is functional management? What are my social rights?" The game addressed various topics: therapy, genetics, care, pain, rehabilitation and social rights.	Two sessions of two hours each	MM+RM	NA
Eccleston et al. 2003	United Kingdom	Cohort	Chronic pain	14-3	Individual and family-centered	Clinic	A paediatric rheumatologist, clinical psychologist, physiotherapist, occupational therapist, and a nurse	Educational sessions, and cognitive behavioral therapy sessions.	Many sessions required evening or weekend written and skills practice homework. Patients received written information about all aspects of the programme which built into a patient manual.	Overall contact time was 110 hours (60 hours of physical and occupational activity, 35 hours of cognitive therapy, and 15 hours education). Each session lasted 50 minutes. The day was structured as a school day from 9:00 am to 3:45 pm.	MM and EM	(Cognitive) social learning theory, and Cognitive behavioral theory
Franklin et al. 2006	United Kingdom	RCT	Diabetes	11-16	Individualized	Clinic, and by phone	Diabetes healthcare team	Telemedicine system (text-messages on phone)	Individual goal-setting at clinic. Automated delivery of a series of messaging, including a weekly reminder of the goal set, and a daily message providing tips, information or reminders to reinforce this goal (by phone).	NA	MM	(Cognitive) social learning theory

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants				Intervention characteristics			Theoretical base		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Fuchs et al. 2013	Netherlands	Cohort	Juvenile idiopathic Arthritis	14-19	Individual	NA	Child psychologist and counselor/philosopher	Narrative self-reflections	Phase 1: self-investigation, about important life experiences; Phase 2: process-promoting, about daily situations and coping; Phase 3: second self-investigation, consistencies and changes in person narratives;	Phase 1 included one SMC session. Phase 2 consists of 6 weekly individual sessions of about 1 h each. Phase 3 consists of 3 weekly individual sessions of about 1 h each.	RM+EM	Self-confrontation method
Geber et al. 2007	United States of America	Cohort	Diabetes	22.3 yrs	Individualized, but also online discussion with peers possible	Online	Psychologist, patient advocacy expert and social worker	Telemedicine system (web-based)	Educational module, and goal-setting exercises with individualized feedback, role-playing, group discussion, empowerment activities, and communication skills training to improve interactions with health professionals. There was a discussion board available, and there were three 'ask the experts segments'.	NA	MM	NA
Grey et al. 2009	United States of America	RCT	Diabetes	9.9 yrs	Group sessions	Clinic	Mental health professional	Educational sessions	Role play, coaching, and practice at home.	6 weekly sessions	RM and EM	(Cognitive) social learning theory
Hackett et al. 2005	United Kingdom	Cross-sectional	Juvenile Idiopathic Arthritis	Median: 14 yrs	Group	Camp	Occupational therapist, Physiotherapist, Nurse / therapist, assistant	Camping program	Climbing, canoeing, abseiling and a trip to a theme park.	4 day annual event	RM	NA
Hampel et al. 2003	Germany	Cohort	Astma	11.6 yrs	Group	Clinic (inpatient)	Psychological, educational, and medical staff	Family sessions (separate for parents and children)	Educational group work	4 weeks stay, 10 1 h-long training sessions.	MM and EM	Stress theory

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants			Intervention characteristics			Intervention characteristics			Theoretical base	
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	
Hanauer et al. 2009	United States of America	RCT	Diabetes	17.9 yrs	Individualized	Online	NA	Telemedicine system (web-based)	BG diaries (blood glucose check) and two daily factoids of which one was related to diabetes and one related to unusual fun facts or trivia. System sends reminders to check BG, and gives (positive) feedback.	NA	MM	NA
Hayutin et al. 2009	United States of America	Cohort	Inflammatory Bowel Disease	13-17 yrs	Group sessions	NA	NA	Family sessions (separate for parents and children, except for relaxation session and conflict resolution and communication session)	Review of written homework, didactic presentations, discussion, problem solving and practice of the new skill, plans for application during the week, and assignment of homework related to the skills.	10 sessions	MM	Cognitive behavioral theory
Hechler et al. 2010	Germany	Cohort	Chronic pain	14.0 yrs	Individualized	Clinic (inpatient)	Therapists	Skills training sessions	Interoceptive exposure; bilateral stimulation in the form of tapping; and cognitive coping strategies to reduce pain intensity.	NA	MM	NA
Herbert et al. 2013	United States of America	Qualitative	Diabetes (type 1)	11-14	Group	Clinic	Study team counselors, but not mentioned who these were	TeamWork. Adolescent-parent type 1 diabetes (T1D) program developed to prevent deterioration in diabetes care among adolescents with T1D. Coping skills or education group with a study team counselor at four consecutive regularly scheduled diabetes clinic visits.	Coping skills session topics included communication and diabetes management, problem solving to improve blood glucose management, healthy food choices and avoiding arguments, and how attitudes affect behaviors and how these relate to physical activity. Each session started with an overview of the skill and was followed by a discussion about what was typical for the family and how they could use the skill in daily life.	NA	MM	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics				Participants		Intervention characteristics			Element(s)	Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)				
Hilberink et al. 2013	Netherlands	Mixed methods	Cerebral palsy, spina bifida, Neuromuscular disease	19.9 yrs	Group	School, and rehabilitation clinics	Pedagogues, psychologists, social workers, a sexologist, and a teacher	Educational/support sessions	Homework assignments	7 sessions (90 min each, scheduled over a 12-week period).	RM and EM	Flirt model
Højberg et al. 2010	Denmark	Cohort	Congenital physical disability	18-25 yrs	Group sessions	Rehabilitation clinic (and a trip to Lithuania)	Occupational therapist, and a socio-educational assistant (if necessary another assistant was hired)	Developmental instructional training course (skills training sessions)	NA	The group met 20 hours per week, 4 days a week. Approximately every sixth week out of house activities of 2-3 days duration. In addition: a 10-day study trip to Lithuania.	RM	NA
Huss et al. 2003	United States of America	RCT	Asthma	9.6 yrs	Individual	Home	NA	CD-ROM	Interactive game including levels and quizzes and feedback from an on-screen nurse.	NA	MM	NA
Jan et al. 2007	Taiwan	RCT	Asthma	10.5 yrs	Individual	Clinic (outpatient)	NA	Telemedicine system (Internet-based)	Educational modules, electronic diary, action plan, and monitoring system	NA	MM	NA
Jones et al. 2010	United States of America	RCT	Cancer	12-18 yrs	Individual	Home	Trained healthcare professionals	CD-ROM	Information videos, text, stories, and a game.	NA	MM and RM	NA
Joseph et al. 2007	United States of America	RCT	Asthma	15.3 yrs	Individual	School	NA	Telemedicine system (web-based)	Theory-based health messages and information on Asthma control. Normative (compared with other students) and ipsative (compared with your last session) feedback.	NA	MM	Trans theoretical, and health belief model

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants		Intervention characteristics				Theoretical base			
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Kashikar-Zuck et al. 2012; 2013	United States of America	RCT	Juvenile Fibromyalgia	15.0 yrs	Individual	Clinic	Therapists with postdoctoral training in pediatric psychology	Cognitive-behavioral therapy sessions (in 3 of the 8 parents were also present)	Education and skills training. Instructions for home practice were also reviewed with participants.	8 weekly individual sessions, and 2 booster sessions.	MM	Cognitive behavioral theory
Kato et al. 2008	United States of America	RCT	Cancer	13-29 yrs	Individual	NA	NA	CD-ROM (PC-game)	Game	NA	MM	Self-regulation theory, and (Cognitive) social learning theory
Koontz et al. 2004	United States of America	RCT	Sickle cell disease (anemia)	8-12 yrs	Group session	School	Teachers	School program	Peer education	1 hour	MM and RM	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics			Element(s)	Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)				
Kouba et al. 2013	United States of America	Cohort	Asthma	15.9	Group and individual	School	Registered nurses, dietitian and dietetic interns	The ICAN program is thus composed of four elements: (1) asthma education, (2) nutrition education synthesized with CST, targeting obesity prevention and management, (3) reinforcement visits with a registered nurse (RN) and dietetic intern, and (4) a family information meeting. Sessions included group problem-solving and role-play activities, which allowed opportunities to increase perceived competence and foster peer support for improved health choices. In addition electronic modules were developed and offered.	Quest for the Code, an interactive, three-dimensional video that combines asthma education with rich graphics and top celebrities, was viewed by the students in groups during lunch periods. The second session included a review, discussion about the group's asthma symptoms, medications, management, and additional content from the "Fight Asthma Now" program developed by the Respiratory Health Association. Each nutrition session included a focused nutrition lesson followed by introduction of a coping skill. Then interactive exercises were completed with students allowing them the opportunity to practice the coping skill with a nutrition scenario. The CST strategies were used to reinforce nutrition information. CST is a cognitive behavioral strategy that teaches students personal and social coping skills to assist in making health-related decisions. Electronic modules: These interactive modules encouraged students to seek resources and make decisions related to the target behaviors using existing Internet resources.	The ICAN educational session covered an 8-week period including the makeup session. This was followed by two reinforcement visits. The total program length spanned 14 weeks.	MM	Orem's Self-Care Deficit theory of Nursing

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants		Intervention characteristics			Theoretical base				
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Krishna et al. 2003	United States of America	RCT	Asthma	7-17 yrs old	Individual (clinical staff leaves when the patient is working on IMPACT)	Clinic	Multidisciplinary team involved in determining content of the program.	Telemedicine system (interactive multimedia program)	Animated lessons, each averaging a minute in length. Each template was designed to illustrate concepts, test comprehension and reinforce learning, develop decision-making skills or improve a child's ability to communicate their asthma.	During office visits	MM	NA
Kumar et al. 2004	United States of America	RCT	Diabetes	13.6 yrs	Individual	Online	NA	Telemedicine system (personal device)	Game with rewards	NA	MM	NA
Laffel et al. 2003	United States of America	RCT	Diabetes	12.1 yrs	Individualized, child with parent (during office visits)	Clinic	Research assistant	Family sessions (for child and parent together)	Written materials, discussion, making a responsibility-sharing plan	During office visits	MM	NA
Leding et al. 2008	Norway	Cohort	Diabetes	13-17 yrs	Group sessions	Clinic (outpatient)	A team of nurses, medical doctors, and a trained social worker	Separate support/ educational/skills sessions for adolescents and parents	Warm-up activity such as painting, movement/exercises, or a round of questions about topics that were not related to diabetes, followed by group discussion.	NA	MM, RM and EM	NA
MacDonald & Greggs 2010	United Kingdom	Qualitative	Cystic Fibrosis	8-18 yrs	Individual	Home/public environment	Volunteers (after following a training programme)	Peer-support	Mentorship	NA	EM	NA
Martin et al. 2009	United Kingdom	Cohort	Diabetes	9-11 yrs	Group sessions	Clinic	Pediatric diabetes nurse specialist, assistant psychologist under supervision of a consultant (pediatric clinical psychologist).	Educational sessions	Educational stories about diabetes, group discussions, coloring/drawing pictures, one-to-one computer session.	NA	MM and EM	(Cognitive) social learning theory

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Participants		Intervention characteristics			Element(s)			Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base	
Maurice-Stam et al. 2009	Netherlands	Cohort	Cancer	8-12 years	Group sessions	Clinic (outpatient)	Dermatologists or pediatricians, psychologists, and dietitians	Educational/skills/support sessions	Modelling, contingency management, exposure exercises and cognitive techniques.	NA	MM, RM and EM	Cognitive behavioral theory	
McClellan et al. 2009	United States of America	Cross-sectional	Sickle Cell Disease	8-20 yrs	Group session and individual device	Clinic, and by phone	NA	Skills training session, and telemedicine system (personal device)	Daily pain diary and coping skills practice (through audio files).	NA	MM	Cognitive behavioral theory	
McDonagh et al. 2007	United Kingdom	Cohort	Juvenile Idiopathic Arthritis	14.2 yrs	Individual	Clinic	Project coordinator from clinic assisted by local consultant rheumatologist	Individual (transition) plan	Age and developmentally appropriate information resources, and goal-setting	NA	MM and RM	NA	
Meade et al. 2003	United States of America	Cross-sectional	End-stage renal disease	13.7 yrs	Group sessions	Clinic	A nurse from the transplant team, and a nephrologist.	Educational/skills/support sessions	Problem solving, stress management/relaxation, talking with other teens and/or parents, communication role-plays, questions and answers with the nephrologist, medication presentation by the nurse.	2-4-hour sessions	MM	NA	
Merlijn et al. 2005	Netherlands	Cohort	Pain	14-18 yrs	Group sessions, and individual telephone contacts	Clinic	NA	Educational and skills training sessions (two sessions were for parents only: one at the beginning and one at the end). The others were for adolescents only.	Training and exercise book, peers, written material, exercise and homework assignments.	NA	MM, RM and EM	(Cognitive) social learning theory	

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics			Intervention characteristics				Theoretical base					
Identifier	Country	Design	Participants Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
O'Mahar et al. 2010	United States of America	Cohort	Spina bifida	16.6 yrs	Group and individual	Camp	Counselors, but not mentioned who these are.	Camping program with support sessions	NA	112-hour group sessions	RM and EM	NA
Holbein et al. 2013												
Palermo et al. 2009	United States of America	RCT	Chronic pain	14.8 yrs	Individualized	Online	Online therapist: a psychology postdoctoral fellow who had one year of specific experience in delivery of face-to- face CBT	Telemedicine system (web- based program).	Treatment modules with video files and audio files, and daily diary. Also queries (assignments).	NA	MM	(Cognitive) social learning theory, and Cognitive behavioral theory
Fales et al. 2014												
Payne et al. 2013	United States of America	Case study	End-stage renal disease	14-16	Individualized	Clinic	NA	Six modules: four individual and two joint (with parents). A quiz was given after each session to assess knowledge of skills before progressing to the next module.	Sessions were designed to identify and address knowledge, behavioral, and cognitive barriers to medication adherence. Session 1: knowledge and education about regimen, 2) sharing regimen responsibilities (joint session), 3) review of problem-solving skills and solutions (joint session), 4) role of cognitions as adherence barriers, 5) emotional regulation and coping skills to overcome barriers to adherence, 6) review of skills learned throughout intervention.	The intervention was completed in a minimum of 6 weeks. Including data collection, sessions lasted 60 to 90 minutes.	MM	Health Belief model

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics				Element(s)	Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Individualized	Delivery location	Interventionists	Format(s)				
Raghavendra et al. 2013	Australia	Mixed methods	CP, physical disability or acquired brain injury	10-18	Individualized		Home	Speech pathologist and research assistants	Educational and skills training sessions	Focused on: identifying and evaluating the family's current computer and Internet access; teaching the youth and family about cyber safety including the use of filtering and privacy settings, the development of house rules for expected online behaviour, and explanations/ discussion of age-appropriate social media; providing appropriate software and equipment based on the goals; providing training materials including visual supports or instructions as well as hands on training and practice to use the software, equipment or Internet; providing interface solutions, such as speech recognition software for text entry, word-prediction software or screen reading software. Training was framed to ensure youth accessed the identified web sites to engage in social networking with their peers, friends and families.	NA	RM	NA
Rami et al. 2006	Austria	RCT	Diabetes	10-19 yrs	Individualized		Online/Phone	NA	Telemedicine system (on phone)	Monitoring through daily diaries.	NA	MM	NA
Rhee et al. 2011	United States of America	RCT	Asthma	14.6 yrs	Group sessions		Camp	Peer leaders selected by school teachers/nurses or clinicians	Camping program	Peer leaders led group activities that involved discussion, strategic thinking, knowledge-testing games, and role playing.	Each session lasted approximately 45-60 minutes.	MM and RM	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics			Theoretical base		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM
Rhee et al. 2012	United States of America	RCT	Asthma	17.6 yrs	Training sessions	Camp	Peer leaders selected by school teachers/nurses or clinicians	Camping program	Disease-specific activities (sessions)	NA	NA
Sattoe et al. 2013	Netherlands	Mixed methods	End-stage renal disease	16-25	Group	Camp	Pediatric nephrology health care professionals	Camping program: with different elements: workshop present yourself, movie making workshop, dancing workshop, sports, cooking teams, free time, theater performance by professional artists, art workshop, drumming workshop, acting workshop. Also: buddy-attendant concept (peer-to-peer support).	Aimed at independent living with ESRD and developing self-management skills, but no focus on disease-specific issues; about transition to adulthood in general, i.e. aimed at general competencies like self-confidence, self-efficacy etc.	Five days	RM+EM
Shames et al. 2004	United States of America	RCT	Asthma	8.0 yrs	Individualized	Clinic, and home environment	Case manager, allergist / immunologist, and pediatric nurses	Educational and skills training sessions	Informational books, phone calls by interventionists, video game, asthma management plan	NA	MM
Smith Fawzi et al. 2012	Haiti	Mixed methods	HIV	10-17 yrs	Group sessions	Clinic	Social workers	Family sessions (child-parent pairs)	Sharing experiences	The sessions were implemented over a one year period (held bi-monthly).	MM, RM and EM
Staab et al. 2006	Germany	RCT	Atopic dermatitis (Eczema)	8-18 yrs	Group sessions	Clinic	Dermatologists, pediatricians, psychologists, and dietitians	Educational sessions and skills training	Sharing experiences, practicing skills	NA	MM and EM

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants			Intervention characteristics			Element(s)	Timing	Domain(s) of SM	Theoretical base
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists				
Stinson et al. 2010	Canada	RCT	Juvenile Idiopathic Arthritis	15.6 yrs	Individualized, but contact with peers possible	Online	Trained coach: non health care professional with an undergraduate degree in psychology	Telemedicine system (web-based)	NA	MM, RM and EM	NA
Stulemeijer et al. 2005	Netherlands	RCT	Chronic fatigue Syndrome	15.5 yrs	Individualized	Clinic	Child therapists who were trained and supervised by an experienced cognitive behavioral therapist	Cognitive-behavioral therapy sessions	10 sessions	MM, RM and EM	Cognitive behavioral theory
Ten Hoedt et al. 2011	Netherlands	RCT	Phenylketonuria	15 yrs	Individualized	Online	Dietician	Telemedicine system (secured website)	NA	MM	NA
Torok et al. 2006 ⁶⁵	Hungary	Cohort	Diabetes, oncologie	Oncologie 15.58 yrs; Diabetes 14.90 yrs	Group	Camp	NA	Camping program	2 separate 8-day sessions	NA	NA
Vekser-Friedrich et al. 2005	United States of America	RCT	Asthma	10.1 yrs	Group sessions, and individualized nurse visit	School	Nurse practitioner	Educational sessions	Six weekly sessions	MM and RM	Self-care theory

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics				Theoretical base		
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	
Verhoeef et al. 2014	Netherlands	Cohort	Physical disability	16-25	Group and individual	Clinic (outpatient)	Occupational therapist, psychologist, jobcoach, social worker	Educational and skills training sessions	Group sessions provided information and discussion about work-related topics and aimed to increase insight into personal (dis)abilities, addressing work objectives, coping strategies (dis)abilities, addressing work objectives, coping strategies, occupational balance, finding a (suitable) job, how to present oneself at a job interview, and employment regulations and social security. In addition, group sessions offered opportunities to develop communication and interaction skills, to share experiences, and to increase group members' self-efficacy. Homework assignments, for example preparing a self-presentation or preparation for a job interview, contributed to active participation and planning. Individual sessions: developing vocational skills and work routines, enhancing self-management skills, providing work placement opportunities and work experience, and advising on workplace modifications.	Group support programme consisting of six 2-hour sessions over 8 weeks.	RM and EM	Model of human occupation (MOHO)
Wang et al. 2008	United States of America	Cohort	Diabetes	14.0 yrs	Group sessions	Camp	Physician, medical students, and a dietitian	Camping program	Educational sessions	NA	MM	NA

Appendix C4.1 Overview of study and intervention characteristics (n=86)* (continued)

Study characteristics		Participants				Intervention characteristics						
Identifier	Country	Design	Condition	Mean age or age range (in years)	Mode	Delivery location	Interventionists	Format(s)	Element(s)	Timing	Domain(s) of SM	Theoretical base
Wysocki et al. 2007	United States of America	RCT	Diabetes	14.0 yrs	Family-centered	Clinic	Psychologists	Educational and skills training sessions	Problem solving training, communication training and cognitive restructuring and functional-structural family therapy. Therapists participated actively, frequently providing instructions, feedback, modeling and rehearsal. Also homework was given each session.	12 sessions over 6 months attended	MM and RM	NA
Xenakis et al. 2010	United States of America	Cohort	Physical disabilities, 93% congenital	14-21 yrs	Individualized and group sessions	Clinic / Hospital based center	A program coordinator, instructors and tutors, a care assistant, and volunteers. The instructors, experts in their chosen fields, have prior experience working with adolescents and/or persons with physical disabilities	Educational sessions	Discussions, expressive arts, goal-setting, field trips (community recreation and learning institutions).	12 consecutive 2½-hour weekly sessions	MM, RM and EM	NA
Yoon et al. 2007	United States of America	Cohort	Sickle Cell Disease	10.7 yrs	Individualized	Clinic	NA	CD-ROM	Game with feedback	NA	MM	Gaming theory

*Abbreviations: NA=Not available (meaning this information couldn't be derived from the original research article); RCT=Randomized Controlled Trial; SM=Self-management; MM=Medical management; RM=Role management; EM=Emotion management

Appendix C4.2 Overview of outcome measures used in the evaluation studies*

Groups of outcomes	Specific outcomes	References
Health outcomes	Clinical outcomes (e.g. blood glucose levels, peak expiratory flow rate or pulmonary functioning, BMI, fatigue)	[27, 30, 38, 39, 41, 44-47, 51-53, 55-57, 64, 66, 99, 101]
	Number of (outpatient) clinic visits or disease-related emergency department visits	[35, 39, 44, 64, 105]
	Hospitalizations (due to chronic condition)	[35]
	Frequency or intensity of disease-specific symptoms, i.e. morbidity (e.g. asthma episodes or pain)	[28-33, 35, 36, 38, 39, 41-45, 66, 73-77, 83, 91, 93, 96, 100, 105, 109]
	Activity limitations or functional or disease-related disability / perceived difficulty in performing daily activities	[73, 75, 77, 91, 93-95]
	Medication or treatment adherence or usage	[31, 34, 35, 38, 41, 44, 46, 55-57, 64, 79, 85, 90, 96, 101]
	Self-perceived health status	[41]
	Somatic awareness	[74]
	Disease-specific coping (e.g. coping with pain during SCD-related pain episodes)	[95, 105]
	Functional health status / Physiologic health status / Physical functioning or activity	[64, 66, 74, 81, 94, 99]
	Condition-related disability	[100]
	Goal status in individual health and wellness planning	[103]
Self-care	General satisfaction with health	[40]
	Condition-specific goals	[11]
	Risk behavior (e.g. smoking)	[35]
	Self-care behavior or practice	[39, 44, 46, 110]
	Disease-related skills	[65]
	Independent health behavior (e.g. self-medication or independent consultations)	[98]
	Motivation, knowledge, skills and abilities for self-care	[39]
	Disease-related self-regulation	[33]

Appendix C4.2 Overview of outcome measures used in the evaluation studies* (continued)

Groups of outcomes	Specific outcomes	References
Self-efficacy	Disease-related self-efficacy	[28, 32, 39, 44, 46, 51, 55, 59, 63, 64, 86, 96, 109]
	Self-efficacy related to managing interactions with healthcare providers	[105, 86]
	General self-efficacy	[67, 86]
	Outcome expectations of disease-related self-management	[59]
Self-perception	Confidence regarding disease-specific management	[108]
	Self-rated competence in the areas of (a) scholastic work, (b) social acceptance, (c) athletic ability, (d) physical appearance, (e) behavioral conduct, and (f) global self-worth	[107]
Sense of control	Sense or locus of control	[63, 64]
	Role mastery	[110]
Psychological outcomes	Depressive symptoms / depression	[51, 56, 61, 71, 74, 75, 78, 93-95]
	Anxiety or (dis)stress	[41, 62, 64, 71, 73, 74, 96]
	Catastrophic thinking about the condition	[74]
	Psychological symptoms	[89]
	Self-esteem	[39, 67, 71]
	Body image	[62]
Psychosocial functioning	Psychosocial adjustment	[29, 81]
	Developmental goals	[102]
	Psychosocial functioning	[49, 89, 99]
Coping	Coping or coping styles	[51, 63, 71]
	Adaptive and maladaptive behaviors and emotions	[42]
	Coping abilities/competencies and resourcefulness or coping styles	[40, 42, 44, 82]
	Internalizing and externalizing behavior	[61]

Appendix C4.2 Overview of outcome measures used in the evaluation studies* (continued)

Groups of outcomes	Specific outcomes	References
Health-related quality of life or subjective wellbeing	(Health-related) Quality of Life (generic)	[37, 54, 64, 69, 86, 93, 104]
	Disease-specific health-related quality of life	[27, 28, 30, 32, 33-36, 42-44, 46, 47, 51, 55, 56, 59, 61, 63, 76, 93, 96, 98]
	Subjective wellbeing	[110]
Vocational participation	School functioning (absence/attendance or grades)	[35, 43, 66, 73, 74, 105, 107, 109]
	Academic performance	[43]
	Occupational performance / work participation	[104]
	Pre-vocational experience	[98]
	Social and academic activities competencies	[61]
	Occupational problems	[102, 104]
Social participation	Challenges / problematic situations encountered in different life areas	[70, 82]
	Days of changed plans	[35]
	Physical, emotional and behavioral concerns related to certain life areas	[62]
	(Autonomy in) participation in several life areas (e.g., living and romantic relationships)	[67, 72, 109]
	(Influence on) daily life with the condition	[47, 86]
	Social skills	[61, 92, 112]
	Social goals / goal attainment	[111, 112]
	Social acceptance by peers	[112]
	Dating	[62]
	Independence	[112]
	Parent involvement in diabetes care	[54]
	Family functioning	[51]

Appendix C4.2 Overview of outcome measures used in the evaluation studies* (continued)

Groups of outcomes	Specific outcomes	References
Family involvement or conflict (related to disease-related management tasks)	Division/sharing of responsibility, conflict or interaction within families related to disease specific tasks (e.g. blood glucose monitoring or insulin injections)	[46, 54, 57, 60, 85, 111, 112]
Support by others	Perceived support	[80]
	Social support by family and friends	[89]
Others (not included in groups)	Knowledge of the disease and/or treatment	[28, 32, 34, 36-39, 44, 49, 53, 62-64, 79, 82, 90, 92, 96, 98, 105, 107, 108, 111]
	Attitudes towards illness	[27, 78, 79, 86]
	Problem solving	[37, 78]

*Outcomes considering satisfaction with the intervention and proxy outcomes were not included in the analysis of outcome measures

PART II

Transitions and self-management of young
people with chronic conditions



5

Lagging behind or not? Four distinctive social participation patterns among young adults with chronic conditions



Jane N.T. Sattoe, Sander R. Hilberink, AnneLoes van Staa, Roland Bal

Journal of Adolescent Health. 2014; 54(5):397-403

ABSTRACT

Purpose

Typical childhood and adolescent development and acquiring self-management skills are crucial for a satisfying adult life and autonomy in social participation. The aims of this study were to identify patterns of autonomy in social participation and to explore differences between these patterns.

Methods

Adolescents with various chronic conditions participating in a survey in 2006 (T₀) were re-invited for a follow-up study (T₁) in 2012. The young adults (18-25 years of age) assessed self-management skills, their condition's impact on school or work, health-related quality of life (HRQoL), and social participation in various domains. Patterns were identified through cluster analysis. Differences between patterns were analyzed in bivariate and multivariate analyses.

Results

Compared to healthy age-mates, our sample (n=483) generally lagged behind in social participation. Four patterns emerged: typical developers, financially secure laggards, slow developers, and outgoing laggards. The patterns differed regarding gender, educational level, attending special education, having disability benefits, and degree of physical limitations. Groups with a higher level of autonomy in social participation did not necessarily have higher HRQoL, but did report higher self-efficacy and independence at both measurements.

Conclusions

Autonomy in some participation domains can coincide with lack of autonomy in others. In addition, better social participation does not necessarily correlate with higher HRQoL, or vice versa. Yet, more social participation was associated with more self-efficacy and independence. Our results emphasize that there is no standardized approach. Clinicians should take care to address all life areas in clinical practice to screen patients' lived experiences and need for social and self-management support.

Implications and Contribution

Young adults with chronic conditions generally lag behind in social participation. However, distinct patterns exist, and some do even better than healthy peers. Better social participation does not necessarily correlate with higher HRQoL, but is associated with more self-efficacy and independence. Specialized support for these young adults is important.

INTRODUCTION

The vast majority of children with chronic conditions now live into adulthood [1]. The child reaching adulthood involves a role shift for both parents and healthcare professionals, who are expected to move from controlling the child's life to helping develop the young adult's self-management and social participation skills [2]. The importance of this new role is increasingly acknowledged, because a chronic condition may interfere with gaining autonomy, which is a main developmental task for young people [3]. Achieving developmental milestones and growing up into a self-efficacious individual – but also acquiring self-management skills – are crucial for a satisfying adult life and successful fulfilment of social roles [3, 4]. In turn, successful social participation positively affects one's health and wellbeing [4, 5].

The International Classification of Functioning, Disability and Health (ICF) defines social participation as a person's involvement in life-situations [6] in domains of social functioning such as education, employment, leisure and sexuality. Young adults (YAs) with chronic conditions often lag behind in social participation compared to their healthy peers [7]. In this regard, a variety of chronic conditions and physical disabilities have been associated with reduced work participation or school participation [8-10], as well as with delayed courtship, sexuality and independent living [11-13]. To strengthen self-management and social participation skills, several interventions aimed at children, adolescents and YAs with chronic conditions have been developed (e.g., [14-17]).

Becoming an adult is a transition that does not occur simultaneously in all domains of functioning [18]. This is why researchers often focus on separate domains, or measure multiple domains and provide sum scores of sub-domains (e.g., Life-H [19]) to describe participation and associative factors. This approach carries the risk, however, of missing important information on patterns of participation in several life areas that together constitute reality for YAs. The present study aims to identify different patterns of autonomy in social participation, and to explore how they differ with respect to self-management skills and health-related quality of life (HRQoL) of young adults now and during adolescence, as well as the impact of the chronic condition on current education or vocational participation.

METHODS

Participants

Participants of a web-based survey in 2006 (T₀) [20] were re-invited for a similar survey six years later (T₁). In 2006, they were adolescents aged 12 to 18 years, had been under treatment in the Erasmus Medical Center – Sophia's Children's Hospital Rotterdam for

more than 3 years. Contact information and death notices were retrieved from the hospital's electronic patient registry. Eligible YAs received an invitation letter providing relevant information and a unique password to log in on a secured website. Those who did not respond within a month were sent a reminder by mail. After another month, non-responders were reminded through a phone call. Respondents were entered in a lottery to win one of 25 cookbooks, two smart phones, or an iPad. The Medical Ethical Committee of the Erasmus Medical Centre approved the study.

Measures

The web-based follow-up questionnaire assessed four dimensions: 1) social participation, 2) background characteristics (including age, gender, and physical limitations), 3) aspects of self-managing the chronic condition (e.g., self-efficacy), and 4) HRQoL and the impact of the condition on school or work career. The questionnaire was based on a literature review and the previous questionnaire [21]. It was pilot tested in face-to-face (n=7) and telephone (n=3) interviews with young people with chronic conditions, who then were not included in the final sample.

Social participation

Level of autonomy in social participation was classified using the Rotterdam Transition Profile (RTP) [18]. The RTP describes participation in seven life areas defined in the International Classification of Functioning, Disability, and Health: (1) finances; (2) employment and education; (3) housing; (4) intimate relationships; (5) sexuality; (6) transportation; and (7) leisure. Furthermore it distinguishes four transition phases (0-3). Young persons in phases 0 and 1 are still fully dependent on adults (e.g., parents) or display typical child behaviour. Young persons in phase 2 experiment with adult behaviour or orient to it. Phase 3 refers to full autonomy in participation. Because we were interested in a successful transition to adulthood, we dichotomised the phases as follows: 0 = phases 0-2, and 1 = phase 3. Reference data from Dutch age-mates were extracted from the database of Statistics Netherlands (StatLine) [22], and from a report on sexual health of Dutch youths aged 15-25 years [23]. Reference data for transportation and leisure were not available, however.

Background characteristics

At T₀, physical limitations in mobility had been assessed through a 10-item scale (Cronbach $\alpha = .90$) [24]. Also, the codes from the International Classification of Diseases available in the hospital's database were used. At T₁ the following socio-demographic characteristics were addressed: gender (1 = female; 2 = male), age, ethnicity (1 = Dutch surname; 2 = non-Dutch surname), level of education (1 = low [junior vocational or secondary general low]; 2 = medium [secondary general high or senior vocational]; 3

= high [higher educational institutions or university], and type of education followed in the past (1 = mainstream education; 2 = special education). Respondents were also asked if they were benefit recipients within the framework of the *Wajong*, the Income Provision Act for Disabled Young People (1= yes; 2 = no). Young people who, because of their chronic condition, are (in part) unable to work may be eligible to receiving these benefits [8].

Self-efficacy, and independence during hospital consultations

Self-efficacy was measured by the On Your Own Feet Self-Efficacy Scale (OYOF-SES) using a 4-point Likert scale for every item (1 = no, definitely not; 2 = no, probably not; 3 = yes probably; 4 = yes certainly) [25]. It consists of three domains: coping with the condition (four items, Cronbach α = .82), knowledge about the condition (six items, Cronbach α = .78) and competencies during consultations (six items, Cronbach α = .85). The OYOF-SES is based on self-report and was included at both T0 and T1. The higher the sum score on the OYOF-SES, the higher the self-efficacy of the YAs. Finally, in both questionnaires, YAs rated their general independence during hospital consultations on a visual analog scale (1-10). The higher the score, the more independent the YA was.

Health-related quality of life, and impact of the condition on school or work career

HRQoL was assessed using the self-report versions of DISABKIDS questionnaire, originally designed for children and adolescents. The T0 questionnaire presented the short form measure (DCGM-10) [26], and scores had been transformed to a 0 - 100 scale. At T1, five domains of HRQoL were measured with the DCGM-37 [27] on a 5-point Likert scale (1 = often; 2 = quite often; 3 = sometimes; 4 = almost never; 5 = never): Independence (six items, Cronbach α = .86); Physical (six items, Cronbach α = .84); Emotion (seven items, Cronbach α = .91); Social exclusion (six items, Cronbach = .83); Social inclusion (six items, Cronbach α = .81). A general score was computed by combining all items (Cronbach α = .95). Scores were transformed to a 0 - 100 scale.

The perceived impact of the chronic condition on current education and vocational participation was only measured at T1 using a newly constructed 5-item scale whose items were derived from relevant literature and from interviews with YAs with chronic conditions or physical disabilities. Respondents rated the impact on a 5-point Likert scale (1 = totally disagree, 2 = disagree, 3 = neutral, 4 = agree, 5 = totally agree) (Cronbach α = .88).

Data analysis

Backward logistic analysis was used to detect selective response; determinants of study non-response were expressed in odds ratios (ORs) with the 95% confidence intervals (95% CI), and Nagelkerke R^2 indicated the proportion of explained variance. Model fit

was tested with the Hosmer and Lemeshow test. Patterns in social participation were identified with an exploratory hierarchical cluster analysis using the complete linkage method with simple matching coefficient to derive the number of patterns resulting from our data, followed by a two-step cluster analysis. The dichotomized domains of the RTP were used as input for the cluster analysis. The number of distinctive patterns was derived from the dendrogram resulting from the hierarchical cluster analysis [28]. Pearson correlation tests between the domains of social participation in the RTP supported our final cluster solution. Respondents' levels of autonomy in participation were compared with those of Dutch age-mates using binominal tests. Finally, exploratory multivariate analysis of covariance and chi-square tests, followed by Welch's analysis of variance and chi-square tests with post-hoc tests, identified inter-cluster differences.

RESULTS

Response

Of the 1,039 participants in the original study, 13 had died and 25 could not be traced. Consequently, 1,001 were invited, 88 of whom returned a postcard stating they declined to participate further. Eventually 518 YAs (net response 51.8%) submitted the survey. Backward logistic regression analysis showed that non-response was associated with male gender (OR = .57, 95%CI = .43 – .74), and non-Dutch ethnicity (OR = .49, 95%CI = .33 – .74) ($\chi^2 = 29.0$, $df = 2$, $p < .001$, $R^2 = .04$, Hosmer and Lemeshow test $p = .773$). Thirty-five YAs (6.8%) did not provide information on their social participation and were excluded. Hence, the final sample numbered 483 (Table 1).

With respect to their diagnosis, 29% of the YAs had congenital malformations, deformations and chromosomal abnormalities; 14% had neoplasms; 13% had endocrine, nutritional and metabolic diseases; 12% had diseases of the eye and adnexa or ear and mastoid process; 9% presented with diseases of the musculoskeletal system and connective tissue; 5% with diseases of the respiratory system; 5% had diseases of the digestive system; and the remaining 13% fell within different ICD chapters.

Patterns of social participation

The cluster analysis resulted in four distinctive patterns of autonomy in social participation (Table 2). These were characterized by, respectively, (1) typical autonomy development regarding all seven life areas (*'typical developers'*; $n=105$); (2) financial self-supporting, good chance of employment, but less autonomy in other life areas (*'financially secure laggards'*; $n=109$); (3) lagging behind in autonomy development on all life areas except transportation (*'slow developers'*; $n=96$); (4) less autonomy in the areas

Table 1 Description of the study sample, n=483 (unless indicated)

	Study sample at T1 n (%) or mean (±SD)	Study sample at T0 n (%) or mean (±SD)
Background characteristics		
Gender (male)	184 (38.1)	-
Age [18-25]^	20.6 (±1.9)	-
Educational level (high)	213 (44.7) ¹	-
Special education (yes)	83 (17.2)	-
Ethnicity (non-Dutch surname)	52 (10.8)	-
Physical limitation(s) (yes)	-	140 (29.1) ²
Disability benefit (Wajong*) (yes)	95 (19.7)	-
HRQoL and Impact of the condition		
HRQoL [1-100]^		
General HRQoL	77.9 (±16.0)	80.5 (±15.4) ³
Independence domain	83.7 (±16.2)	-
Emotion domain	76.0 (±21.3)	-
Social inclusion domain	76.2 (±16.6)	-
Social exclusion domain	83.1 (±18.2)	-
Physical domain	70.9 (±22.6)	-
Medication domain	72.9 (±21.9) ⁴	-
Perceived impact on school/work career [5-25]^	10.1 (±5.3)	-
Self-efficacy and independence		
Self-efficacy (OYOF-SES) [10-64]^	55.8 (± 6.6) ⁵	52.8 (± 6.9) ⁶
Independence during consultation [1-10]^	7.7 (±1.5) ⁵	7.0 (± 2.0) ⁷

^theoretical range

*the Income Provision Act for Disabled Young People

¹n=476, ²n=481, ³n= 479, ⁴n=216, ⁵n=374, ⁶n=456, ⁷n=457

of finances, employment, and living, but more often intimate and sexual relationships and socialization with peers ('*outgoing laggings*'; n=173).

Social participation compared to reference group

Compared to Dutch age-mates, YAs with chronic conditions in general lagged behind regarding employment (27.7% versus 63.3%; $p<.001$), living on their own (18.8% versus 44.9%; $p<.001$), having an intimate relationship (40.2% versus 64.0%; $p<.001$), and experience with sexual intercourse (52.8% versus 82.0%; $p<.001$) (Table 2). However, they were more often financially self-supporting (43.7% versus 30.2%; $p<.001$).

Typical developers were very similar to their healthy age-mates, and even more frequently were financially self-supporting and had a partner ($p<.001$). Financially secure laggings were more often financially self-supporting, but lagged behind in living independently, or having a partner. Also, they more frequently had not achieved autonomy

Table 2 Patterns regarding young adults' autonomy in social participation; % in phase 3 (n=483)

Life areas Patterns	Finances	Employ- ment	Living	Relation- ships	Sexuality	Transport- ation	Leisure
Typical developers (n=105)	100.0*	68.6	40.0	85.7*	78.1	96.2	55.2
Financially secure laggers (n=109)	97.2*	55.0	0.0*	0.0*	22.9*	80.7	41.3
Slow developers (n=96)	0.0*	0.0*	0.0*	0.0*	0.0*	93.8	49.0
Outgoing laggers (n=173)	0.0*	0.0*	28.3*	60.1	85.5	96.5	65.9
Total (n=483)	43.7*	27.3*	18.8*	40.2*	52.8*	92.3	54.7
Reference group	30.2 ¹	63.3 ²	44.9 ¹	64.0 ³	82.0 ³	-	-
Post hoc[^]	1,2>3,4	1>2>3,4	1>4>2,3	1>4>2,3	1,2,4>3 1,4>2	1,3,4>2	4>2,3 1>2

*p<.001 in binominal tests to test if there are significant differences between our sample and Dutch age-mates.

[^]A Chi² post hoc test revealed that these differences were significant on a p<.05 level

¹Reference group aged 20-25 years in 2011.

²Reference group aged 15-25 years in 2007.

³Data presented here are from a report on Dutch youths (15-25 yrs) in 2005.

in the life area sexuality than healthy age-mates (p<.001). Slow developers differed most from healthy age-mates, none of them were autonomous in the five life areas (p<.001). Compared to age-mates, outgoing laggers were less frequently living independently, and none of them were employed or financially self-supporting (p<.001) (Figure 1).

Differences between patterns

Ethnicity had no distinctive value for pattern classification (p= .086), but differences were found regarding gender, educational level, attending special education, having disability benefits, and degree of physical limitations (Table 3).

During adolescence (To), self-rated independence during consultation, and HRQoL were significantly different between patterns. During young adulthood (T1), there were no significant differences between the four patterns in the medical domain of HRQoL (p= .312). However, the patterns differed in: perceived impact on education/vocational participation, general HRQoL and all its subdomains, self-efficacy, and self-rated independence during consultation (Table 3).

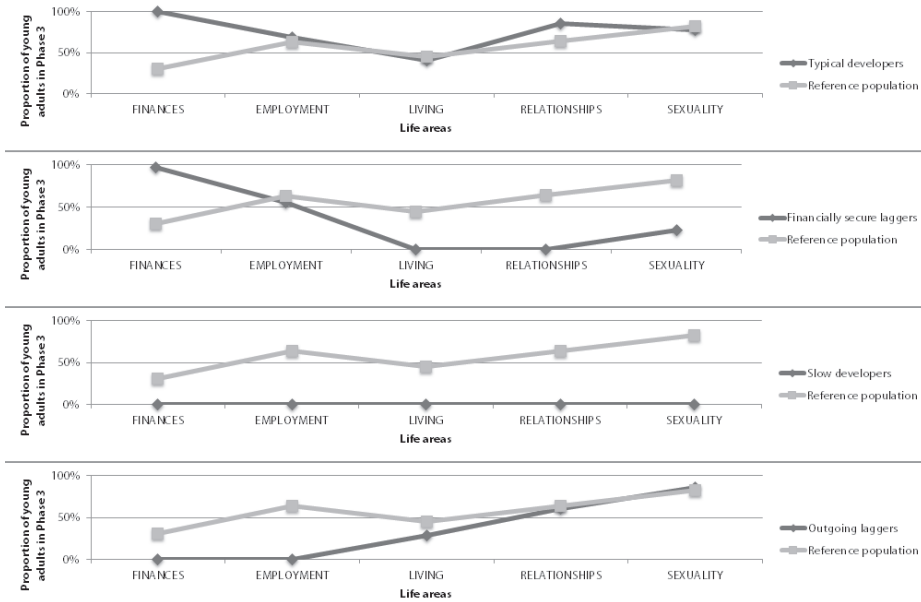


Figure 1 Comparison of social participation between patterns and reference population

To summarize Table 3, typical developers scored best on self-rated independence during consultations at T₀, and also did well on self-rated independence during hospital consultations at T₁. But they perceived higher impact of their condition on education/vocational participation, and experienced lower HRQoL at T₁. Financially secure laggards had more physical limitations, and more frequently had attended special education or currently received disability benefits. They reported lower HRQoL at both T₀ and T₁, had lower current self-efficacy and independence, and perceived higher impact of their condition on education/vocational participation. Slow developers were more frequently male and less frequently higher educated. They scored low on independence on both T₀ and T₁, but reported good HRQoL on both T₀ and T₁. Also, they perceived low impact of their condition on education/vocational participation. Last, outgoing laggards had higher HRQoL at both T₀ and T₁, scored best on self-efficacy and independence at T₁, and perceived lower impact of their condition on education/vocational participation.

Table 3 Inter-cluster differences in background, impact of the condition, and HRQoL; Mean(\pm SD) or frequency (%);

	1 Typical developers (n=105)	2 Financially secure laggards (n=109)	3 Slow developers (n=96)	4 Outgoing laggards (n=173)	df	X ² or F	p	Post hoc [^]
Background characteristics								
Age (covariate)	21.8 (\pm 1.8)	21.1 (\pm 1.8)	19.4 (\pm 1.5)	20.3 (\pm 1.7)	3, 479	36.7	<.001	1>2>4>3
Gender (male)	28 (26.7)	42 (38.5)	52 (54.2)	62 (35.8)	3	16.7	.001	3>1,2,4
Educational level (high)	33 (32.4)	35 (33.3)	46 (47.9)	99 (57.2)	3	23.2	<.001	3,4>1,2
Special education (yes)	24 (22.8)	40 (36.7)	10 (10.4)	9 (5.2)	3	52.1	<.001	2>1>3,4
Wajong* (yes)	39 (37.1)	56 (51.4)	0 (0.0)	0 (0.0)	3	155.5	<.001	2>1>3,4
Physical limitations (AVO)	10.9 (\pm 2.6)	13.4 (\pm 6.0)	10.3 (\pm .99)	10.3 (\pm .62)	3, 477	25.7	<.001	2>1,3,4
Self-efficacy, independence during hospital consultations, and HRQoL during adolescence (T0)								
Self-efficacy (OYOF-SES)	53.6 (\pm 7.7)	52.4 (\pm 6.4)	52.1 (\pm 7.3)	52.9 (\pm 6.4)	3, 452	.92	ns	ns
Self-rated independence during consultation	7.4 (\pm 1.9)	6.9 (\pm 2.1)	6.5 (\pm 2.0)	7.2 (\pm 1.9)	3, 453	4.4	.004	1>3
General HRQoL	78.6 (\pm 15.3)	76.8 (\pm 15.3)	83.5 (\pm 14.4)	82.3 (\pm 15.5)	3, 475	4.7	.003	3,4>2
Self-efficacy, and independence during hospital consultations (T1)								
Self-efficacy (OYOF-SES)	55.7 (\pm 6.7)	54.3 (\pm 7.2)	55.7 (\pm 6.0)	57.0 (\pm 6.1)	3, 370	3.1	.025	4>2
Self-rated independence during consultation	8.0 (\pm 1.6)	7.3 (\pm 1.7)	7.2 (\pm 1.4)	8.1 (\pm 1.3)	3, 370	9.5	<.001	1,4>2,3
Health-related quality of life, and impact of the condition on school/work career (T1)								
General HRQoL	73.8 (\pm 17.6)	73.1 (\pm 16.9)	81.6 (\pm 13.7)	81.4 (\pm 14.1)	3, 479	10.9	<.001	3,4>1,2
Physical HRQoL	66.2 (\pm 23.7)	64.3 (\pm 24.2)	78.6 (\pm 17.7)	73.8 (\pm 21.5)	3, 479	9.9	<.001	3,4>1,2
Emotion HRQoL	69.8 (\pm 23.5)	72.5 (\pm 20.6)	82.3 (\pm 18.1)	78.5 (\pm 20.7)	3, 479	7.9	<.001	3,4>1 3>2
Independence HRQoL	80.5 (\pm 17.8)	77.8 (\pm 18.7)	86.3 (\pm 13.5)	87.9 (\pm 13.1)	3, 479	11.8	<.001	4>1,2 3>2
Social exclusion HRQoL	79.1 (\pm 20.9)	77.3 (\pm 18.2)	85.5 (\pm 15.3)	88.0 (\pm 16.5)	3, 479	10.6	<.001	3,4>2 4>1

Table 3 Inter-cluster differences in background, impact of the condition, and HRQoL; Mean(\pm SD) or frequency (%); (continued)

	1 Typical developers (n=105)	2 Financially secure ladders (n=109)	3 Slow developers (n=96)	4 Outgoing ladders (n=173)	df	X ² or F	p	Post hoc [^]
Social inclusion HRQoL	74.7 (\pm 19.4)	70.6 (\pm 20.9)	75.7 (\pm 20.1)	80.9 (\pm 17.5)	3, 479	6.8	<.001	4>2
Impact on school/ work career	11.6 (\pm 5.5)	12.6 (\pm 5.9)	8.5 (\pm 4.2)	8.6 (\pm 4.4)	3, 479	20.9	<.001	1,2>3,4

*Disability benefits for disabled persons who are (partially) unable to work because of their condition

[^]A Chi² or Scheffe post hoc test revealed that these differences were significant on a p<.05 level (the groups before the '>' sign score significantly higher than the groups behind the sign)

DISCUSSION

We identified four distinctive patterns regarding autonomy in social participation among YAs with chronic conditions. Since this study is the first to our knowledge to explore such patterns, we cannot compare with pattern distribution in similar or other populations. However, evidence from previous studies supports our finding that these YAs generally lagged behind in social participation compared to healthy age-mates [7, 11, 29, 30]. We also found that full autonomy in one area of social participation can coincide with total lack of autonomy in other areas, endorsing the necessity to apply a holistic approach to support young people in their transition into adulthood, taking into account all of the important life areas. As Roebroek and colleagues [11] point out, there is a “need for incorporating a lifespan perspective throughout paediatric, transition, and adult healthcare”. Such an approach could include the deployment of Young Adult Teams (YATs) of healthcare professionals [31] that has been useful for supporting YAs’ social participation in England. Other options – introduced in the Netherlands – are group-based interventions aimed at single or multiple participation domains or discussing so-called patient reported outcomes during consultations [16, 17, 32–35].

In general, level of social participation appears to be positively associated with wellbeing [4, 5, 11]. Yet in our study, the typical developers reported lower general HRQoL whereas slow developers and outgoing ladders (with less developed autonomy) reported higher general HRQoL at both To and T1. Although the general thought is that more participation leads to better wellbeing, there are some studies that found the opposite to be true [36, 37]. The typical developers’ lower general HRQoL may be explained by more often having to face limitations when participating in society [16]. This suggests that full autonomy in all participation domains may be burdensome for YAs, and that they may need support to deal with this burden. The finding that those

with less developed autonomy reported higher HRQoL, might be attributed to the differentiation between having a disability and experienced health [31]. However, this was not true for the financially secure laggards who reported more physical limitations and more hospital admissions, what most likely explains their lower wellbeing, both now and during adolescence. The finding that they more often attended special education and received disability benefits supports this, and testifies to their being vulnerable despite their financial independence. The last also appears to apply to YAs who perceived quite some impact of their condition on their education/vocational participation (typical developers and financially secure laggards). They reported lower HRQoL on the emotional domain, irrespective of their level of autonomy in social participation.

Autonomy in social participation seemed to be positively associated with level of self-efficacy and level of independence. YAs with the least social participation (slow developers and financially secure laggards) reported lower self-efficacy and independence; YAs with more social participation (typical developers and outgoing laggards) reported higher self-efficacy and higher independence. This difference in independence between typical developers and slow developers was already present during adolescence. Healthcare professionals in adolescent care should do well, therefore, to help these patients develop good self-management skills. Our findings support the notion that identifying and paying attention to young persons' needs in the areas of social participation and lived experiences is crucial to have them achieve a successful and satisfying adult life [20, 38]. This is an integral part of transitional care for young persons [21], and professionals should use specific tools, e.g. the HEEADSSS tool for psychosocial screening [39], individual transition plans like the Skills for Growing up tool [33], and monitoring of HRQoL [34].

Strengths and limitations

Our study included a large sample of young persons 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. However, since all young people growing up with a chronic condition face comparable challenges and similar adaptive tasks [1], studying chronic conditions in general is not considered a flaw. Next to this, the correlations found between psychosocial variables were not high, indicating the absence of important psychosocial confounders.

Furthermore, the non-response analysis revealed that non-responders more frequently had a non-Dutch surname and were male. We found that there were significantly more males in the group of 'slow developers' compared to the others groups, suggesting that this group of YAs may be bigger than found in our study. As cultural

characteristics influence transition, underrepresentation of non-Dutch YAs might have affected the study outcomes. It is impossible, however, to tell in what direction. Another limitation is that the RTP originally was developed and validated for YAs with cerebral palsy [18] and was not designed to measure the level of participation, but rather to classify the phases of transition to adulthood (social participation). In the present study we were only interested in the final transition phase as indicator for having reached full autonomy in a certain domain. Therefore, we used the RTP as designed: to classify the level of autonomy in participation in important life areas.

Finally, we did not validate our cluster solutions in a second sample, and could not elaborate on their external validity. However, our study aim was to explore patterns of social participation, which can be validated in future research. Future studies should focus on studying patterns of social participation in an older age group, and on following a cohort of young people over a longer time to explore possible shifts in social participation. Also, alongside autonomy in or frequency of social participation, it is important to look at lived experiences in social participation (i.e., enjoyment) [40].

CONCLUSIONS

The variety of patterns of autonomy in social participation shows that achieving independence differs across life areas, and underscores the notion that these life areas together constitute reality for YAs. It appeared that more social participation does not necessarily lead to higher HRQoL, or vice versa, but seemed to be associated with more self-efficacy and independence. Clinicians should be aware of this and could make use of various tools to address all life areas to screen patients' lived experiences and need for social and self-management support. Our results emphasize that there is no 'one size fits all', and stress the importance of specialized support for these young persons in the healthcare setting.

ACKNOWLEDGEMENTS

The On Your Own Feet program [2004-2008] was funded by ZonMw, the Netherlands organization for Health Research and Development (Grant no. 32560005), and the Dutch charity foundation *Kinderpostzegels* (Grant no. 23764/1/4). The follow-up study was part of the Self-management and Participation Innovation Lab, supported by SIA-RAAK, the Foundation Innovation Alliance with funding from the Dutch ministry of Education, Culture and Science (OCW). Bert van der Heijden is thanked for having suggested this follow-up study. The authors thank the other members of the On Your Own Feet Research

Group Jos Latour, Susan Jedeloo, and Linda van der Knaap for their support in the design and execution of the study. Laila Oumansour, Wendy de Boer, Marjolijn Bal, and Mariëlle Peeters are thanked for their assistance in collecting data, while Kimberly Onderdijk, and Esmee Kuppen helped pre-testing the follow-up questionnaire.

REFERENCES

1. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.
2. Kieckhefer GM, Trahms CM: Supporting development of children with chronic conditions: from compliance toward shared management. *Paediatr Nurs* 2000, 26(4):354-363.
3. Turkel S, Pao M: Late consequences of chronic pediatric illness. *Psychiatr Clin North Am* 2007, 30(4):819-835.
4. 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.
5. Law M: Participation in the occupations of everyday life. *Am J Occup Ther* 2002, 56(6):640-649.
6. (WHO) WHO. International Classification of Functioning, Disability and Health: ICF. Geneva: World Health Organization; 2001.
7. Stam H, Hartman EE, Deurloo JA, Groothoff J, Grootenhuys 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.
8. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuys M: Growing into disability benefits? Psycho-social course of life of young adults with a chronic somatic disease or disability. *Acta paediatr* 2011, 101(1):e19-26.
9. Barf HA, Verhoef M, Post MW, Jennekens-Schinkel A, Gooskens RH, Mullaart RA, Prevo, AJH: Educational career and predictors of type of education in young adults with spina bifida. *Int J Rehabil Res* 2004, 27(1):45-52.
10. Maslow GR, Haydon A, McRee AL, Ford CA, Halpern CT: Growing up with a chronic illness: social success, educational/vocational distress. *J Adolesc Health* 2011, 49(2):206-212.
11. Roebroek 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.
12. Groothoff JW, Grootenhuys 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.
13. Plug I, Peters M, Mauser-Bunschoten EP, de Goede-Bolder A, Heijnen L, Smit C, Willemse J, Rosendaal FR, van der Bom JG: Social participation of patients with hemophilia in the Netherlands. *Blood* 2008, 111(4):1811-1815.
14. Punthakee Z, Gorter J: TRansition to Adulthood with Cyber Guide Evaluation (TRACE). 2011. Available at <http://canchild.ca/en/ourresearch/trace.asp> [accessed 10 april 2013].
15. Guidelines for Service Providers: Supporting Youth & Families in Using the Skills for Growing Up Checklists 2007. Available at <https://secure1.securewebexchange.com/bloorview.ca/resource-centre/familyresources/documents/serviceproviderguidelines.pdf> [accessed 11 april 2013]
16. Verhoef JAC, Roebroek ME, Van Meeteren J, Stam HJ, Miedema HS: A new intervention to improve work participation of young adults with physical disabilities: a feasibility study. *Dev Med Child Neurol* 2013, 55(8):722-728.
17. Hilberink SR, Kruijver E, Wiegerink DJHG, Vlietland TPM: A pilot implementation of an intervention to promote sexual health in adolescents and young adults in rehabilitation. *Sex Disabil* 2013, 31(4):373-392.
18. Donkervoort M, Wiegerink DJ, van Meeteren J, Stam HJ, Roebroek ME, Transition Research Group South West N: Transition to adulthood: validation of the Rotterdam Transition Profile for young adults with cerebral palsy and normal intelligence. *Dev Med Child Neurol* 2009, 51(1):53-62.

19. Fougeyrollas P, Noreau L, St-Michel G: Life Habits measure – shortened version (LIFE-H 3.0). Lac St Charles Quebec Canada: CQCIDIH; 2001.
20. 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.
21. Van Staa AL. On Your Own Feet: Adolescents with chronic conditions and their preferences and competencies for care (Doctoral dissertation). Rotterdam: Rotterdam University; 2012.
22. Centraal Bureau voor Statistiek [Statistics Netherlands] - StatLine. 2012. Available at <http://statline.cbs.nl/statweb/> [accessed 17 december 2012].
23. de Graaf H, Meijer S, Poelman J, Vanwesenbeeck I: Seks onder je 25^e. [Sex before the age of 25]. Rutgers Nisso Groep/Soa Aids Nederland; 2005.
24. de Klerk M, Iedema J, van Campen C: SCP-maat voor lichamelijke beperkingen op basis van AVO 2003 [SCP measure for physical limitations based on AVO 2003]. Den Haag: Sociaal Cultureel Planbureau; 2006.
25. van Staa AL, Strating MMH, On Your Own Feet Research Group: On Your Own Feet Self-Efficacy Scale: development and validation of a generic instrument for chronically ill adolescents. In: On Your Own Feet: Adolescents with chronic conditions and their preferences and competencies for care (Doctoral dissertation). Rotterdam: Rotterdam University; 2012.
26. Ravens-Sieberer U, Schmidt S, Gosch A, Erhart M, Petersen C, Bullinger M: Measuring subjective health in children and adolescents: results of the European KIDSCREEN/DISABKIDS Project. *Psychosoc Med* 2007, 4:Doc08.
27. Simeoni MC, Schmidt S, Muehlan H, Debensason D, Bullinger M, Group D. 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.
28. Mooi E, Sarstedt M: A Concise Guide to Market Research. Berlin Heidelberg: Springer-Verlag; 2011.
29. Martinez W, Carter JS, Legato LJ: Social competence in children with chronic illness: a meta-analytic review. *J Pediatr Psychol* 2011, 36(8):878-890.
30. Gorter JW, Stewart D, Woodbury-Smith M, Freeman M, Nguyen T, Wright M, Swinton M, Burke-Gaffney J, Missiuna C, Campbell W, Rosenbaum P, DeMatteo C, Leo CC: Developmental Trajectories of Youth with Disabilities (age 12-25 years of age): A Knowledge Synthesis. McMaster University CanChild Centre for Childhood Disability Research; 2011.
31. Bent N, Tennant A, Swift T, Posnett J, Scuffham P, Chamberlain MA: Team approach versus ad hoc health services for young people with physical disabilities: a retrospective cohort study. *Lancet* 2002, 360(9342):1280-1286.
32. Maathuis CGB, Vos I, Roebroek ME, Hilberink SR: Een instrument om vaardigheden voor zelfstandigheid te vergroten. De Groei-wijzer [An instrument to improve skills for independence. The Skills for Growing Up.]. *Nederlands Tijdschrift voor Revalidatiegeneeskunde* 2012, 3.
33. Sattoe JNT, Hilberink SR, Peeters MAC, van Staa A. 'Skills for Growing Up': supporting autonomy in young people with renal disease. *J Ren Care* 2014, 40(2):131-139.
34. Haverman L, van Rossum MA, van Veenendaal M, van den Berg JM, Dolman KM, Swart J, Kuijpers TW, Grootenhuys MA: Effectiveness of a web-based application to monitor health-related quality of life. *Pediatrics*, 2013;131(2):e533-543.
35. van der Stege HA, van Staa A, Hilberink SR, Visser AP: Using the new board game SeCZ TaLK to stimulate the communication on sexual health for adolescents with chronic conditions. *Patient Educ Couns* 2010, 81(3):324-331.

36. van Campen C, Iedema J: Are persons with physical disabilities who participate in society healthier and happier? Structural equation modelling of objective participation and subjective well-being. *Qual Life Res* 2007, 16:635-645.
37. van Campen C, Cardol M. When work and satisfaction with life do not go hand in hand: Health barriers and personal resources in the participation of people with chronic physical disabilities. *Soc Sci Med* 2009, 69:56-60.
38. Betz CL, Redcay G. An exploratory study of future plans and extracurricular activities of transition-age youth and young adults. *Issues Compr Pediatr Nurs* 2005, 28(1):33-61.
39. Goldenring JM, Rosen DS. Getting into adolescent heads: an essential update. *Contemp Pediatr* 2004, 21(1): 64-90
40. Granlund M. Participation – challenges in conceptualization, measurement and intervention. *Child Care Health Dev* 2013, 39(4):470-473.

6

Young adults' experiences and satisfaction with the transfer of care: A follow-up study in a large hospital sample



AnneLoes van Staa, Jane N.T. Sattoe

Journal of Adolescent Health. 2014; 55(6):796-803

ABSTRACT

Purpose

Transition of care of adolescents with chronic conditions is a critical area for clinicians. Patient-reported outcomes may provide important information on the quality of services. This cohort study examines young adults' experiences and satisfaction with the transfer to adult care and explores associations with patient characteristics.

Methods

Follow-up of 518 young adults (18-25 years) with various chronic conditions who completed a web-based survey in 2006 (response rate 52%). Outcome measures were the 18-item Transfer Experiences Scale ($\alpha=.93$), and satisfaction with the transfer process (visual analogue scale). Associations with demographic and healthcare-related variables, health-related quality of life, and self-management were explored with stepwise multivariate modelling, using past (2006-T0) and current (2012-T1) variables.

Results

Of the respondents, 315 (65%) had transferred, while 10% was still in pediatric care and 25% was not in treatment anymore. Twenty percent rated their transfer as unsatisfactory, 50% felt prepared at the time of transfer and 24% had met their adult-centered provider in advance. Men were more positive about their experiences and rated satisfaction higher than did women. Patient-centeredness of the adult healthcare provider was the most important determinant for experiences ($\beta=.29$). Higher self-efficacy at T1 was associated with more positive experiences, but not with higher satisfaction. The latter was higher for those transferred within the same hospital ($\beta=.28$).

Conclusions

The On Your Own Feet Transfer Experiences Scale is a useful instrument to measure transfer experiences. The importance young adults attach to good relations with their new provider, stresses the necessity of early involvement of and good collaboration with adult care.

Implications and Contribution

Transition of care is a critical area for adolescent health. This longitudinal study reported mixed experiences and satisfaction of young adults with moving to adult-centered care. Patient-centeredness of the provider was most strongly associated with positive experiences, indicating the importance of making adult services more responsive to young adults' needs.

INTRODUCTION

The past decades have seen a surge of interest in the challenges that young people with chronic conditions and special healthcare needs face when progressing into adulthood. The transition from child-centered to adult-oriented healthcare systems is part of a longer life course transition for adolescents with chronic conditions – a critical area for future health status and degree of social participation [1]. Since Blum and colleagues defined transition as a multi-faceted process in 1993 [2], dozens of journal articles and editorials have called for a better preparation of all parties involved and for a seamless and safe transition process [3]. Unfortunately, there has been a slow uptake of recommendations made [4-6]. A recent national US survey showed that the support needed for successful transition to adult care is mostly wanting [7]. Studies from different countries and in different diagnostic groups have consistently pointed out the lack of planning, communication and coordination surrounding the transfer of care. Recent reviews have also pointed out the paucity of evidence to inform best practice about both the process of and what constitutes effective transitional care programs [8-11], and the absence of longitudinal research into adult outcomes [9].

Moreover, young people's voices are often overlooked, even though they consistently ask to be involved in the process and want providers to listen to them and be sensitive to their needs [10,12,13]. A qualitative meta-synthesis of 18 studies of adolescents' and young adults' experiences with the transition from pediatric to adult hospital care found comparable experiences across diagnoses. Feelings of not belonging, of being redundant, and being unprepared during the transfer process were strikingly frequent [14]. Young people's experiences can be summarized into four major themes: facing changes in significant relationships, moving from a familiar to an unknown setting and culture, being prepared for transfer, and achieving responsibility [14]. Our previous qualitative study found that transition experiences and views of young people overlapped with those of their parents and professionals [13]. Leaving pediatric care was seen as a 'logical step' and participants noted specific positive and negative aspects of both settings. However, all desired better preparation, more collaboration and personal links between pediatric and adult providers [10,14].

As an important patient-reported outcome measure for the quality of the transition process, young adults' experiences and satisfaction with this process should be collected systematically, along with other relevant outcomes such as health status, quality of life, social participation, self-management skills, access to and quality of adult care [8,15]. There are very few reliable and valid measures that assess these outcomes [15]. A recent review of transfer satisfaction measures identified only 7 studies [16]. While some studies reported negative experiences [17] and significant worries [18], in another participants had no concerns and were satisfied [19]. Most studies were descriptive and used small

disease-specific samples with cross-sectional designs. Measures were usually developed *ad hoc* without following the established guidelines for patient reported outcomes [20]. Also, different concepts were measured at the same time (e.g., concern, attitudes, satisfaction) and all measures had poor quality according the COSMIN checklist criteria [16,20]. COSMIN stands for COnsensus-based Standards for the selection of health Measurement Instruments. The checklist can be used to determine whether a study on measurement properties meets the standards for good methodological quality [20]. Only one instrument (“Mind The Gap” [21]) has established reliability and validity, but it is not developed for measuring post-transfer experiences. Thus, there are no validated instruments to explore experiences during the transition from pediatric to adult care.

Longitudinal studies exploring young adults’ self-reported outcomes during transition are rare [10,22,23], and factors exerting a positive influence on adult outcomes are still unknown. In this study, a large sample of adolescents with various chronic conditions was followed into young adulthood. The aim of the study was to examine post-transfer experiences and satisfaction with transfer, as well as to link these with patient characteristics in adolescence and young adulthood.

METHODS

Participants and Study Design

The respondents of a web-based questionnaire in 2006 (T₀; n=1,039) [24] were invited for a similar survey in 2012 (T₁). In 2006, they were adolescents between 12-18 years of age who had been undergoing treatment in the Erasmus Medical Center – Sophia’s Children’s Hospital Rotterdam for over three years. The original sample presented with a broad range of chronic somatic conditions without intellectual disabilities. In 2012, current provider, healthcare utilization, recent contact addresses, and death notices were retrieved for the hospital’s electronic patient records. Eligible young adults (YA) were sent information and a unique password to log in on a secured website. Those who did not respond within a month were sent a reminder by post. After another month, non-responders were approached by phone. Respondents were entered in a lottery to win one out of twenty-five cookbooks, two smart phones, or an iPad.

Ethical concerns

The study was approved by the Medical Ethics Committee of the Erasmus MC and all data was processed anonymously. Participants provided assent for their participation in the study.

Variables

The new web-based questionnaire was partly based on the previous instrument [24] and was pilot-tested in seven face-to-face and three telephone interviews with young university students with chronic conditions who were not included in the final sample.

Outcome measures

Two new measures to assess experiences with transfer from pediatric (PC) to adult care (AC) were developed. The first was the newly constructed On Your Own Feet Transfer Experiences Scale (OYOF-TES) based on our previous qualitative research [13] and a review of existing measures. It was critically reviewed by nurses and pediatricians, and then pilot-tested with YA in different settings. The items focus on experiences with the transfer process because satisfaction scores often present a limited and optimistic picture. Questions about specific experiences seem more useful, since these can point to ways in which delivery of care can be improved [25].

The OYOF-TES consists of 18 items rated on 5-point Likert scales (1=strongly disagree; to 5=strongly agree). Exploratory factor analysis, followed by confirmatory factor analysis with Oblimin rotation resulted in two domains: A) perceived alignment and collaboration between pediatric and adult care (11 items, Cronbach's alpha (α)=.91); B) experienced preparation for transfer including readiness, timing and co-decision-making (7 items, α =.89). Correlation between the subscales was r =.56. A total of 57.3% of the variance was explained. Appendix C6.1 presents the means and scores of the OYOF-TES items and their factor loadings on the sub-scales.

The second outcome measure concerned satisfaction with the transfer process, using a visual analogue scale (VAS) by asking the YA "*How satisfied are you with the overall process of your transfer to adult care on a scale ranging from 1 to 10?*"

Independent variables

Demographic variables. Age and gender were recorded in both years, while data from the To questionnaire served to establish physical limitations in mobility (0=no; 1=yes) and age at onset of the chronic condition (before versus after the age of five) [24]. At T1, the present level of education was assessed (1=low [pre-vocational education or secondary vocational education; level 1-3]; 2=medium [pre-vocational education or secondary vocational education; level 4] 3=high [pre-university / senior general secondary education or higher educational institutions]), and type of education followed in the past (1=mainstream; 2=special education). Respondents were asked whether they were recipients of disability benefits under the Dutch Income Provision Act for Disabled Young People.

Healthcare-related variables were computed from the electronic patient records at T1. These included *age at transfer* and *timing of the transfer*, taken as the number of years

since the last consultation in pediatric care. This continuous variable was recoded to reflect how long ago transfer had taken place (0=in the past two years, 1=3 to 4 years ago, 2=5 to 6 years ago). The variable *adult healthcare setting* indicates whether transfer was within Erasmus MC or to another hospital. For those still treated in Erasmus MC, the *number of consultations* and of *hospitalizations* as well as the *number of missed appointments* in the past three years were also computed.

Self-management was operationalized through various self-reported measurements. *Disease-related self-efficacy* was measured at To and T1 with the On Your Own Feet Self-Efficacy Scale (OYOF-SES) using a 4-point Likert scale (from 1='no, definitely not' to 4='yes certainly') [24]. It consists of 17 items assigned to knowledge, coping, and skills for hospital consultations. One item ("*I know what will happen to me when I transfer to adult care*") was deleted, because it did not apply to those already transferred at T1. Hence, 16 items were offered ($\alpha=.87$). *General score of independence during consultations* was self-reported on a VAS (range 1-10). *Independent behavior during consultations* was self-reported at To with a dichotomous 7-items scale ($\alpha=.55$). To improve internal consistency, a 5-point Likert scale was used at T1 ($\alpha=.79$) (from 1='never'; to 5='always').

Attitude toward care and transition was measured at To in different ways. The *Attitude toward transition*-scale consisted of four quotes from adolescents. Responses were scored on a 5-point Likert scale from 1='totally disagree'; to 5='totally agree' ($\alpha=.74$) [13]. 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 [24]. *Transfer readiness* was assessed by a single question: "*Do you think that you are ready to transfer to adult care?*" (from 1='no, definitely not' to 4='yes, definitely') [24]. *Patient-centeredness of the current healthcare provider* was only measured at T1 with a 5-item 4-point Likert scale (from 1='never' to 4='always'; $\alpha=.90$). The scale is a validated Dutch adaptation of one scale from the American Consumer Assessment of Health Plan Surveys questionnaire [26].

Health-related quality of life (HRQoL) was assessed at To and T1 using the self-report short form measure of the DISABKIDS questionnaire [27] (DCGM-10; To $\alpha=.78$; T1 $\alpha=.90$); scores are transformed to a 0-100 scale.

Data analysis

Backward logistic analysis was used to detect selective response; determinants of study non-response were expressed in odds ratios (ORs) with 95% confidence intervals (95% CI), and Nagelkerke R^2 indicated the proportion of explained variance. Model fit was tested with the Hosmer and Lemeshow test. Descriptive statistics were used to describe the study sample. Bivariate correlations (Pearson's r or Spearman's ρ) between the outcome and other variables were established to determine which variables should be included in the regression analyses. Stepwise multivariate linear regression analyses were performed to identify factors associated with transfer experiences (TE) and transfer satisfaction (TS):

starting with background variables, then adding T₀ and subsequently T₁ variables. For HRQoL and self-efficacy at T₁ change scores (Δ) were used. Only variables significantly correlated ($P < .05$) with (one of) the outcome variables were included in the regression models. Multicollinearity was checked by calculating the Variance Inflation Factor (VIF).

RESULTS

Follow-up after six years: total sample

Of the 1,039 participants in the original study, 13 (1.25%) had died and 25 could not be traced. Consequently, 1,001 YA were included in the total study sample (Figure 1). Mean age was 20.59 years (SD ± 1.90 ; range 18-25 years) and 447 (44.7%) were male. According to the hospital's electronic patient records, 593 (59.3%) had been transferred to adult care ($n=466$ within the same university hospital and $n=127$ to other hospitals), while 139 (13.9%) were still treated in pediatric care. For 268 patients (26.8%), information about their current healthcare provider was unknown. Only in one record was it clearly stated that the patient had been officially discharged and transferred to primary care.

For those young adults who were still treated in the same university hospital ($n=605$), data on healthcare utilization in the past three years could be retrieved. Young adults treated in pediatric care were younger than those transferred to adult care ($P < .001$), while no-show for scheduled appointments was higher in the group transferred to adult care ($P < .01$); the mean number of missed consultations in the past three years being .78 (SD ± 1.86) in adult care versus .36 (SD $\pm .92$) in pediatric care.

Response and final study sample

Of the 1,001 invited, 606 YA responded (60.5%); 88 of whom returned a postcard stating they declined from participation (14.5%). Eventually, 518 YA (aged 18-25 years) submitted the survey (nett response rate 51.8%). Non-response was associated with male gender (OR, .57; 95% CI, .43-.74), and non-Dutch ethnicity (OR, .49; 95% CI, .33-.74) ($\chi^2=29.0$, $df=2$; $P < .001$; $R^2 = .04$; Hosmer and Lemeshow test $P = .77$). Compared to the responders, the non-responders more often belonged to the group for which no current healthcare provider had been indicated in the hospital's electronic patient records ($p < .01$).

Thirty respondents did not mention their current healthcare provision. Of the remaining 488, 48 (9.8%) were still in pediatric care and 125 (25.6%) were not in active treatment anymore. Ultimately, 315 (64.6%) had transferred to AC and could be included in the analyses. Forty percent had transferred in the past two years (39.9%), 30.6% 3-4 years ago, and 29.6% 5-6 years. Seventy-five percent of these ($n=236$) remained in Erasmus MC. Mean age at transfer was 17.6 years (SD=1.83); 55.5% transferred at 17 or 18 years. Characteristics are summarized in Table 1.

Table 1 Description of the study sample

Characteristics	n T0	Study sample at T0	α T0	n T1	Study sample at T1	α T1	P Value*
Demographic variables							
Age [T1=18-25]; [T0=12-19], mean (SD)	315	15.2 (1.9)		315	20.7 (1.9)		n.a.
Gender, male, No. (%)				315	118 (37.5)		
Presence of physical limitations, yes, No. (%)	312	109 (34.9)					
Age at onset of chronic condition, before age of five, No. (%)	315	225 (71.4)					
Educational level, low or medium, No. (%)				309	174 (56.3)		
Type of education followed in the past, mainstream, No. (%)				315	253 (80.3)		
Occupational status				315			
- still in school / education					199 (63.2)		
- paid employment					82 (26.0)		
- unemployed, not in school					34 (10.8)		
Entitlement for disability benefits, yes				315	81 (25.7)		
Healthcare-related variables							
Timing of the transfer, No. (%)				301			
- in the past two years					120 (39.9)		
- 3-4 years before					92 (30.6)		
- 5-6 years before					89 (29.6)		
Age at transfer to adult care [T1=12-24], mean (SD)				301	17.6 (1.8)		
Type of adult healthcare setting (Erasmus MC), T1, No. (%)				315	236 (74.9)		
Hospital admissions in past 3 years [0-16], mean (SD)				233	.70 (1.7)		
Planned consultations in past 3 years [0-97] ^a , mean (SD)				226	18.8 (17.8)		
Missed consultations in past 3 years (no show), mean (SD)				234	.5 (1.3)		
Self-management							
Self-efficacy (OYOF-SES) [10-64] [^] , mean (SD)	300	52.4 (7.1)	.87	307	55.7 (6.6)	.88	<.001
General score of independence during consultations [VAS; 1-10] [^] , mean (SD)	301	7.0 (1.9)		307	7.7 (1.4)		<.001
Independent behavior during consultations (7 items), mean (SD) ^b	301	2.5 (1.5) [range 0-7] [^]	.55	307	22.5 (6.2) [range 7-35] [^]	.79	n.a. ^b
Attitude towards care and transition / transfer							
Attitude toward transition [4-20] [^] , mean (SD)	300	11.7 (3.4)	.74				
Transition of care often discussed at consultations [1-5] [^] , mean (SD)	301	1.8 (1.1)					
Importance of discussing transition [1-5] [^] , mean (SD)	300	2.7 (1.2)					
Transfer readiness [1-4] [^] , mean (SD)	300	2.6 (.9)					
Patient-centeredness of the current healthcare provider [5-20] [^] , mean (SD)				310	17.1 (3.0)	.90	
Transfer experiences (OYOF-TES) [18-90] [^] , mean (SD)				315	61.8 (13.2)	.93	
Overall transfer satisfaction [1-10] [^] , mean (SD)				315	6.6 (1.8)	-	
Health-related quality of life							
HRQoL (DCGM-10) [1-100] [^] , mean (SD)	310	78.7 (14.7)	.78	312	74.9 (19.1)	.90	.001

* Paired samples t-test

[^] Theoretical range^a Excluding respondents with ≥ 100 hospital visits ($n=8$)^b Independent behavior during consultations was self-reported, at T0 with a dichotomous scale ($\alpha=.55$); while at T1 a 5-point Likert scale was used ($\alpha=.79$).

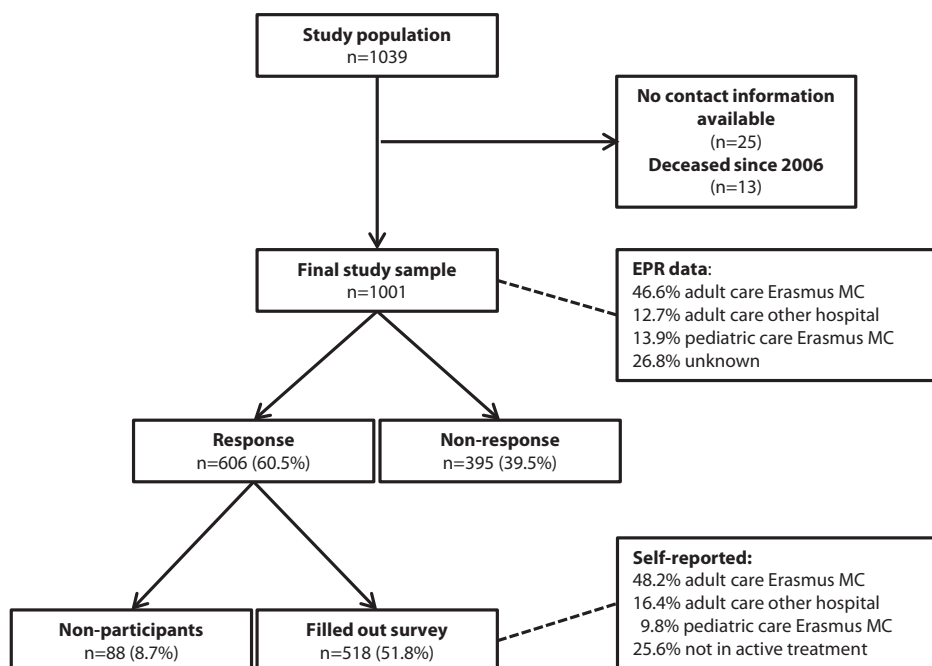


Figure 1 Study sample and response

Experiences and satisfaction with transfer

Self-efficacy and independence during consultation had increased between T₀ and T₁ (both $P < .001$), while HRQoL had decreased ($P = .001$).

The mean of the OYOF-TES was 61.83 (SD=13.22; range 18-90). The scale was normally distributed (skewness=-.47; SE=.14). The most appreciated items were: 'I have confidence in my adult healthcare providers' (80.9% agreed); 'I can manage well on my own during hospital consultations, also without my parents' (78.7% agreed), and 'The transfer to adult care was announced timely and did not come as a surprise' (77.2% agreed). The least appreciated items were: 'Before the transfer I had already met my new healthcare providers' (23.5% agreed); 'When I first met my adult care provider, I knew exactly what was expected of me and what I could expect from him / her' (38.1% agreed) and 'I had a say in the timing of the transfer' (39.7% agreed). Almost half (49.2%) felt they had been well prepared and 43.8% stated they received enough information. Appendix C6.1 lists all OYOF-TES items and means.

Mean TS was 6.63 (range 1-10; SD=1.79; 95% CI, 6.52-6.85); the median was 7 (IQR=2). One fifth (19.4%) scored their transfer process below six (i.e. unsatisfactory), while 34.4% was very satisfied (score ≥ 8) (Figure 2). YA who remained in Erasmus MC were more satisfied with the overall transfer process than those transferred to other hospitals ($P < .01$), while OYOF-TES scores and ratings of patient-centeredness of the current provider did

not significantly differ between these groups. The total OYOF-TES was highly correlated with TS ($r=.75$; $P<.001$). The correlation between TS and the OYOF-TES subscale A (alignment) was higher ($r=.73$; $P<.001$) than with subscale B (preparation) ($r=.62$; $P<.001$). The two OYOF-TES items that correlated best with satisfaction were '*I was taken care of very well in the adult care setting*' and '*There was good collaboration between pediatric and adult care*' ($r=.64$).

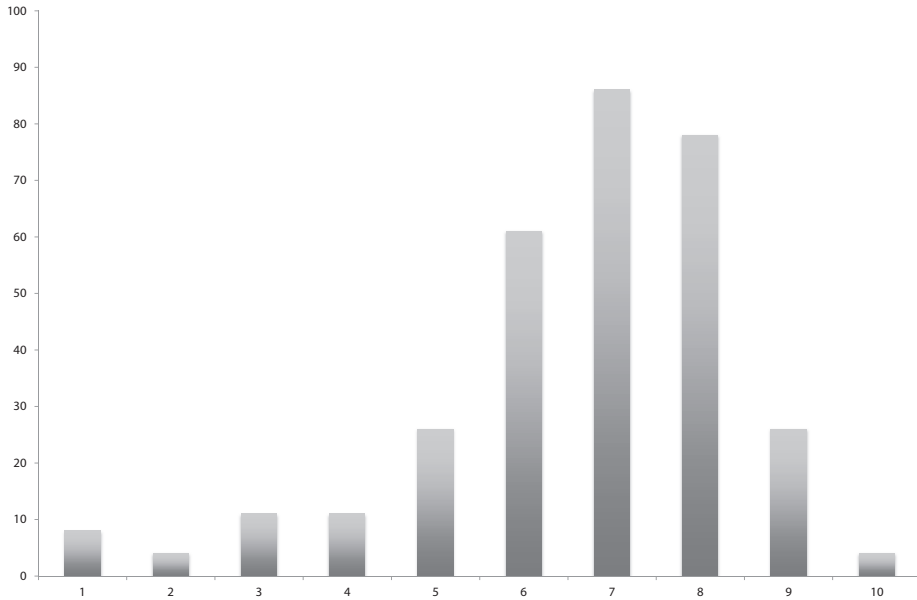


Figure 2 Distribution of transfer satisfaction (n=315)

Associations with transfer experiences and satisfaction

To determine which variables should be included in the regression analyses, bivariate correlations were tested (Table 2). The OYOF-TES multivariate model explained 30% of the total variance, for TS this was 21% (Table 3). VIF scores varied between 1 and 2; indicating the absence of multicollinearity.

Men were more positive about their TE ($\beta=.17$; $P<.001$) and scored their TS higher than women ($\beta=.12$; $P=.03$). Patient-centeredness of the adult healthcare provider was the most important determinant for TE ($\beta=.29$; $P<.001$), and was also strongly correlated with TS ($\beta=.16$; $P<.004$). Attitude toward transition at To ($\beta=.15$; $P=.005$) and higher self-efficacy change at T1 ($\beta=.16$; $P=.05$) were positively associated with TE, but not with TS. Satisfaction was higher when YA remained in Erasmus MC ($\beta=.28$; $P<.001$) and when YA thought discussing the transition was important at To ($\beta=.11$; $P=.04$).

Table 2 Bivariate correlations of variables with the total OYOF-TES score and transfer satisfaction (T0 and T1; Pearson's or Spearman's correlation coefficient r/p)

	n	OYOF-TES total	P Value	Overall transfer satisfaction	P Value
T0					
Presence of physical limitations	312	-.10	.09	-.06	.26
Age at onset of chronic condition	315	-.06	.27	-.04	.54
Self-efficacy (OYOF-SES), T0	300	.15	.01	.11	.05
General score of independence during consultations (VAS), T0	301	.07	.25	.01	.93
Independent behaviors during last consultation, T0	301	-.04	.45	-.06	.30
Attitude toward transition, T0	300	.26	<.001	.16	.01
Transition of care often discussed at consultations, T0	301	-.02	.75	.00	.97
Importance of discussing transition, T0	300	.12	.04	.15	.01
Transfer readiness, T0	300	.08	.18	.04	.51
HRQoL (DCGM-10), T0	310	.13	.02	.09	.10
T1					
Age	315	-.02	.70	-.04	.53
Gender	315	.27	<.001	.18	.002
Educational level	309	.00	.98	.03	.56
Type of education followed in the past	315	-.06	.31	-.07	.23
Entitlement for disability benefits	315	-.07	.22	-.07	.24
Timing of transfer	301	-.04	.53	-.10	.09
Age at transfer	301	-.79	.17	-.01	.87
Type of adult healthcare setting (Erasmus MC), T1	315	.08	.18	.14	.01
Self-efficacy change (OYOF-SES), T1-T0 (Δ)	294	.17	.004	.09	.122
General score of independence during consultations (VAS), T1	307	.18	.001	.16	.005
Independent behaviors during last consultation, T1	307	.15	.009	.10	.07
Patient-centeredness of the current healthcare provider, T1	310	.40	<.001	.39	<.001
HRQoL change (DCGM-10), T1-T0 (Δ)	308	.17	<.003	.16	.006

Table 3 Multivariate linear regression analyses of associations with transfer experiences (OYOF-TES) and transfer satisfaction (standardized betas β) (n=293)

	Step 1		Step 2		Step 3	
	β	P Value	β	P Value	β	P Value
OYOF-TES (TE)						
Gender (male)	.28	<.001	.23	<.001	.17	.001
Self-efficacy (OYOF-SES), T0			.09	.13	.13	.12
Attitude toward transition, T0			.16	.006	.15	.005
Importance of discussing transition, T0			.05	.34	.05	.36
HRQoL (DCGM-10), T0			.09	.13	.08	.18
Self-efficacy change T1-T0 (Δ)					.16	.05
HRQoL change T1-T0 (Δ)					.10	.08
Type of adult healthcare setting (Erasmus MC), T1					.08	.13
General score of independence during consultations (VAS), T1					.01	.85
Independent behaviors during last consultation, T1					.04	.53
Patient-centeredness of the current healthcare provider, T1					.29	<.001
Explained variance final model	R² = .08	<.001	R² = .14	<.001	R² = .30	<.001
F-value (df)	25.37 (1, 291)		9.16 (5, 287)		10.78 (11, 281)	
	β	P Value	β	P Value	β	P Value
Transfer satisfaction (TS)						
Gender (male)	.21	<.001	.18	.003	.12	.03
Self-efficacy (OYOF-SES), T0			.08	.20	.04	.66
Attitude toward transition, T0			.07	.25	.06	.26
Importance of discussing transition, T0			.11	.06	.11	.04
HRQoL (DCGM-10), T0			.07	.23	.08	.19
Self-efficacy change T1-T0 (Δ)					.01	.92
HRQoL change T1-T0 (Δ)					.10	.10
Type of adult healthcare setting (Erasmus MC), T1					.28	<.001
General score of independence during consultations (VAS), T1					.03	.66
Independent behaviors during last consultation, T1					.03	.69
Patient-centeredness of the current healthcare provider, T1					.16	.004
Explained variance final model	R² = .05	<.001	R² = .08	<.001	R² = .21	<.001
F-value (df)	14.00 (1, 291)		5.06 (5, 87)		6.92 (11, 281)	

DISCUSSION

After six years of follow-up, about 60% of the original sample had transferred to adult care, 13.9% were still seen in pediatric care, while in a quarter their destination was unknown. Young adults' satisfaction with transfer to adult care was not general: about one third was very satisfied, but one fifth rated their transfer as unsatisfactory. Men were more positive than women, mirroring the fact that boys reported higher readiness before transfer [24]. One other study found that girls anticipated more difficulties than boys [28]. More positive experiences were also predicted by attitude toward transition at baseline, confirming the importance of attitudes and expectations in transition [23,29,30]. No other baseline variables predicted TE or TS.

The main determinant of reporting positive TE was the degree of patient-centeredness of the adult-oriented provider. Receiving a warm welcome in AC and experiencing good alignment and collaboration between pediatric and adult services seem crucial factors for a positive experience. The importance of provider characteristics in adolescent care is well-known [31], but its relevance for young adult care has not been reported before. Concerns about transition mainly originate from pediatric care and few studies have included adult providers [32,33]. As a consequence, today's debate focusses on better preparation of adolescents, including (measuring) transition readiness [16,34,35], while making adult services more responsive to YA's needs receives less attention [36].

Transitional care should not be confined to a pediatric paradigm and be disconnected from the principles and practice of adolescent medicine [37]. The transition process continues into young adulthood and therefore AC. Still, different studies have indicated that up to 25% of YA became disconnected with care in the first year after transfer [19,38]. In our study, a quarter of the total sample did not receive follow-up in AC anymore, but reasons for this are not known. Promoting continuity of contact could reduce the risk of long-term disengagement with care [38]. In our total sample, the percentage of missed clinic appointments was higher in those treated in adult care compared to pediatric care, indicating a risk of disengagement.

Our sample had faith in their adult providers and was happy with the care received. Yet, less than half felt sufficiently prepared, confirming findings from other studies [19,29]. Timing is an essential element in transition and the transfer should be timed flexibly and decided jointly with young people [3] but in our sample only 40% felt that they had been involved, and about half thought the timing had been right. Transfer usually took place at 17 or 18 years of age, close to what is sometimes reported as 'the ideal age' [39], but unlike in another study [29], age at transfer did not influence experiences or satisfaction.

Our results indicate that the conditions for successful transition preparation and planning were not met [3,10]. About 64% of respondents felt they had been ready to

transfer, a lower percentage than reported before [19,40]. Preparing adolescents for future independent roles and assessing transition readiness deserves more attention [16,34,35]. For example, only 23.5% had met their adult providers in advance. Transition clinics offering this opportunity seem successful in increasing faith in their new providers [18,40] and there is some evidence for their effectiveness [8,9].

Strengths and limitations

Strength of the study was the longitudinal design, enabling to determine what happened to a wide sample of YA after six years of follow-up. Although the sample was fairly large, survey non-response was high (48.2%) especially among males and YA of non-Dutch ethnicity. This group may have included those who dropped out of care or distrusted their new doctor. Another limitation was that we only had healthcare utilization data from those that transferred within the same hospital. Future studies could adopt a more rigorous design involving all YA transferred from a clinic. Finally, we examined a large, heterogeneous sample because adolescents with any kind of chronic condition are facing the same challenges during their transition to adulthood [1]. This generic approach limited the inclusion of disease-specific outcomes including disease severity and the opportunity to research the effects of interventions employed by specialty clinics. However, transition issues are often not disease-specific, and the use of general tools like the TES to measure transition experiences might allow for transition research in less common diseases and comparisons between disease groups [35].

The newly developed OYOF-TES showed good reliability and internal consistency and met most of the COSMIN criteria [20]. The measurement covers all four themes distilled from a qualitative review into adolescents' experiences with transition [14]. The OYOF-TES is an improvement over existing measures [16], although further validation in other populations is needed. Also, to improve the specificity and usability of the measurement for quality improvement, it would be advisable to compare the OYOF-TES with the Health Care Transition Feedback Survey for Youth to determine whether the Six Core Elements of Transition are adequately covered [41].

CONCLUSIONS

Our research suggests that while adolescents should be better prepared for and involved in transition, it is healthcare providers' first priority to build 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.

ACKNOWLEDGEMENTS

The On Your Own Feet program (2004-2008) was funded by ZonMw, The Netherlands Organization for Health Research and Development (Grant 32560005), and the Dutch charity foundation Kinderpostzegels (Grant 23674/1/4). The follow-up study was part of the Self-management and Participation Innovation Lab, supported by SIA-RAAK, the Foundation Innovation Alliance with funding from the Dutch ministry of Education, Culture and Science (OCW) (PRO-02-025). The funding sources had no role in the design and conduct of the study; collection, management, analysis, and interpretation of the data; preparation, review, or approval of the manuscript; and decision to submit the manuscript for publication.

We thank the other members of the On Your Own Feet Research Group (Jos Latour PhD and Linda van der Knaap MANP from Erasmus University Medical Center Rotterdam; Susan Jedeloo PhD, Heleen van der Stege PhD, and Sander Hilberink PhD from Rotterdam University of Applied Sciences) for their contribution to the design, execution, and reporting of the study. The following students are thanked for their contribution: Patrick Polak BSc helped reviewing the existing transfer experiences measurements, while Kimberly Onderdijk and Esmee Kuppen (both Bachelor of speech language therapist) helped pre-testing the questionnaire. Finally, we thank Prof. Bert van der Heijden, then head of the Department of Pediatrics of Erasmus MC, for providing us with the original idea to perform this study.

REFERENCES

1. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.
2. Blum RW, Garell D, Hodgman CH, Jorissen TW, Okinow NA, Orr DP, Slap GB: 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, American College of Physicians, Transitions Clinical Report Authoring Group, Cooley WC, Sagerman PJ: Supporting the health care transition from adolescence to adulthood in the medical home. *Pediatrics* 2011, 128(1):182-200.
4. Rapley P, Davidson PM: Enough of the problem: a review of time for health care transition solutions for young adults with a chronic illness. *J Clin Nurs* 2010, 19(3-4):313-323.
5. McLaughlin SE, Diener-West M, Indurkha A, Rubin H, Heckmann R, Boyle MP: Improving transition from pediatric to adult cystic fibrosis care: lessons from a national survey of current practices. *Pediatrics* 2008, 121(5):e1160-1166.
6. Grant C, Pan J: A comparison of five transition programmes for youth with chronic illness in Canada. *Child Care Health Dev* 2011, 37(6):815-820.
7. McManus MA, Pollack LR, Cooley WC, McAllister JW, Lotstein D, Strickland B, Mann MY: Current status of transition preparation among youth with special needs in the United States. *Pediatrics* 2013, 131(6):1090-1097.
8. Bloom SR, Kuhlthau K, Van Cleave J, Knapp AA, Newacheck P, Perrin JM: Health care transition for youth with special health care needs. *J Adolesc Health* 2012, 51(3):213-219.
9. Crowley R, Wolfe I, Lock K, McKee M: Improving the transition between paediatric and adult healthcare: a systematic review. *Arch Dis Child* 2011, 96:548-553.
10. Lugasi T, Achille M, Stevenson M: Patients' Perspective on Factors That Facilitate Transition From Child-centered to Adult-centered Health Care: A Theory Integrated Metasummary of Quantitative and Qualitative Studies. *J Adolesc Health* 2011, 48(5):429-440.
11. Watson R, Parr JR, Joyce C, May C, Le Couteur AS: Models of transitional care for young people with complex health needs: a scoping review. *Child Care Health Dev* 2011, 37(6):780-791.
12. Betz CL, Lobo ML, Nehring WM, Bui K: Voices not heard: a systematic review of adolescents' and emerging adults' perspectives of health care transition. *Nurs Outlook* 2013, 61(5):311-336.
13. 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.
14. Fegran L, Hall EO, Uhrenfeldt L, Aagaard H, Ludvigsen MS: Adolescents' and young adults' transition experiences when transferring from paediatric to adult care: A qualitative metasynthesis. *Int J Nurs Stud* 2014, 51(1):123-135.
15. Betz CL, Smith K: Measuring health care transition planning outcomes: challenges and issues. *Int J Child Adolesc Health* 2010, 3:463-472.
16. Stinson J, Kohut SA, Spiegel L, White M, Gill N, Colbourne G, Sigurdson S, Duffy KW, Tucker L, Stringer E, Hazel B, Hochman J, Reiss J, Kaufman M: A systematic review of transition readiness and transfer satisfaction measures for adolescents with chronic illness. *Int J Adolesc Med Health* 2014, 26(2):159-174.

17. Busse FP, Hiermann P, Galler A, Stumvoll M, Wiessner T, Kiess W, Kapellen TM: Evaluation of patients' opinion and metabolic control after transfer of young adults with type 1 diabetes from a pediatric diabetes clinic to adult care. *Horm Res* 2007, 67(3):132-138.
18. Latzman RD, Majumdar S, Bigelow C, Elkin TD, Smith MG, Megason GC, Iyer R: Transitioning to adult care among adolescents with sickle cell disease: A transitioning clinic based on patient and caregiver concerns and needs. *Int J Child Adolesc Health* 2010, 3:537-545.
19. Garvey KC, Wolpert HA, Laffel LM, Rhodes ET, Wolfsdorf JL, Finkelstein JA: Health care transition in young adults with type 1 diabetes: barriers to timely establishment of adult diabetes care. *Endocr Pract* 2013, 19(6):946-952.
20. Terwee CB, Mokkink LB, Knol DL, Ostelo RW, Bouter LM, de Vet HC: Rating the methodological quality in systematic reviews of studies on measurement properties: a scoring system for the COSMIN checklist. *Qual Life Res* 2012, 21(4):651-657.
21. Shaw KL, Southwood TR, McDonagh JE, British Society of Paediatric and Adolescent Rheumatology: Development and preliminary validation of the 'Mind the Gap' scale to assess satisfaction with transitional health care among adolescents with juvenile idiopathic arthritis. *Child Care Health Dev* 2007, 33(4):380-388.
22. Colver AF, Merrick H, Deverill M, Le Couteur A, Parr J, Pearce MS, Rapley T, Vale L, Watson R, McConachie H, Transition Collaborative Group: Study protocol: Longitudinal study of the transition of young people with complex health needs from child to adult health services. *BMC Public Health* 2013, 13:675.
23. 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.
24. van Staa AL, 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.
25. Jenkinson C, Coulter A, Bruster S, Richards N, Chandola T: Patients' experiences and satisfaction with health care: results of a questionnaire study of specific aspects of care. *Qual Saf Health Care* 2002, 11(4):335-339.
26. Delnoij DM, ten Asbroek G, Arah OA, de Koning JS, Stam P, Poll A, Vriens B, Schmidt P, Klazinga NS: Made in the USA: the import of American Consumer Assessment of Health Plan Surveys (CAHPS) into the Dutch social insurance system. *Eur J Public Health* 2006, 16(6):652-659.
27. Simeoni MC, Schmidt S, Muehlan H, Debensason D, Bullinger M, DISABKIDS Group: 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.
28. 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.
29. Steinkamp G, Ullrich G, Muller C, Fabel H, von der Hardt H: Transition of adult patients with cystic fibrosis from paediatric to adult care--the patients' perspective before and after start-up of an adult clinic. *Eur J Med Res* 2001, 6(2):85-92.
30. Rutishauser C, Sawyer SM, Ambresin AE: Transition of young people with chronic conditions: a cross-sectional study of patient perceptions before and after transfer from pediatric to adult health care. *Eur J Pediatr* 2014, 173(8):1067-1074.
31. Britto MT, DeVellis RF, Hornung RW, DeFries GH, Atherton HD, Slap GB: Health care preferences and priorities of adolescents with chronic illnesses. *Pediatrics* 2004, 114(5):1272-1280.
32. Peter NG, Forke CM, Ginsburg KR, Schwarz DF: Transition from pediatric to adult care: internists' perspectives. *Pediatrics* 2009, 123(2):417-423.

33. Suris JC, Akre C, Rutishauser C: How adult specialists deal with the principles of a successful transition. *J Adolesc Health* 2009, 45(6):551-555.
34. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, Reiss J, Wood D: 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.
35. Zhang LF, Ho JS, Kennedy SE: A systematic review of the psychometric properties of transition readiness assessment tools in adolescents with chronic disease. *BMC Pediatr* 2014, 14:4.
36. Gleeson H, McCartney S, Lidstone V: 'Everybody's business': transition and the role of adult physicians. *Clin Med* 2012, 12(6):561-566.
37. Kennedy A, Sawyer S: Transition from pediatric to adult services: are we getting it right? *Curr Opin Pediatr* 2008, 20(4):403-409.
38. Downing J, Gleeson HK, Clayton PE, Davis JR, Wales JK, Callery P: Transition in endocrinology: the challenge of maintaining continuity. *Clin Endocrinol (Oxf)* 2013, 78(1):29-35.
39. Godbout A, Tejedor I, Malivoir S, Polak M, Touraine P: Transition from pediatric to adult healthcare: assessment of specific needs of patients with chronic endocrine conditions. *Horm Res Paediatr* 2012, 78(4):247-255.
40. Dabadie A, Troadec F, Heresbach D, Siproudhis L, Pagenault M, Bretagne JF: Transition of patients with inflammatory bowel disease from pediatric to adult care. *Gastroenterol Clin Biol* 2008, 32(5 Pt 1):451-459.
41. Got Transition? Center for Health Care Transition Improvement. The Six Core Elements of Health Care Transition. Available at <http://www.gottransition.org/6-core-elements> [accessed 11 April 2014]

Appendix C6.1 Items of the two subscales of the On Your Own Feet Transfer Experiences Scale (OYOF-TES): means, scores, factor loadings, and correlations with overall transfer satisfaction (VAS) (n=315)

	Item mean (SD) ^a	Agree (%) ^b	Factor loading	Correlation with transfer satisfaction ^c
Subscale A – Alignment between pediatric and adult care (mean = 37.47 (8.64); Cronbach's alpha = .91)				.73
I can manage well on my own during hospital consultations, also without my parents	4.00 (0.96)	78.7	.35	.25
I have confidence in my adult health care providers	3.98 (0.92)	80.9	.78	.56
I am happy with the care I receive in the adult care setting	3.79 (0.95)	73.6	.82	.60
My new care provider was well informed about me and my condition	3.67 (1.10)	67.0	.84	.62
I was taken care of very well in the adult care setting	3.64 (1.04)	65.4	.72	.64
Treatment recommendations in the adult care setting are similar to those I used to receive in pediatric care	3.36 (1.17)	56.2	.85	.56
There was good collaboration between pediatric and adult care	3.35 (1.09)	51.4	.78	.64
The way of working and dealing with patients in adult care are similar to what I was used to in pediatric care	3.20 (1.17)	47.9	.85	.56
When I first met my adult care provider, I knew exactly what was expected of me and what I could expect from him / her	3.05 (1.14)	38.1	.55	.51
I don't really experience many differences between pediatric and adult care	3.03 (1.22)	41.3	.75	.51
Before the transfer I had already met my new health care providers	2.40 (1.25)	23.5	.45	.33
Subscale B – Preparation for transfer (mean = 24.36 (5.98); Cronbach's alpha = .89)				.62
The transfer to adult care was announced timely and did not come as a surprise	3.91 (1.05)	77.2	.80	.46
My parents were ready to transfer to adult care	3.80 (0.99)	69.5	.77	.42
I was ready to transfer to adult care	3.70 (1.12)	63.8	.85	.46
The timing of the transfer was just about right	3.46 (1.03)	51.8	.80	.53
I was well prepared for the transfer to adult care	3.30 (1.09)	49.2	.67	.58
I received enough information about the transfer to adult care	3.17 (1.12)	43.8	.59	.52
I had a say in the timing of the transfer	3.02 (1.25)	39.7	.76	.40

^a 4-point Likert scale (1=no, definitely not; 2=no, probably not; 3=yes, probably; 4=yes certainly).

^b Only those young adults who scored 'agree' and 'strongly agree'.

^c Pearson's correlation coefficient *r* (all *P*<.001)

7

How is self-management related to health-related quality of life in young adults with chronic conditions?



Jane N.T. Sattoe, Jos M. Latour, Susan Jedeloo, Roland Bal, AnneLoes van Staa

Submitted

ABSTRACT

Purpose

Self-management is assumed to contribute to Health-Related Quality of Life (HRQoL), but it is not explored in the most commonly used models or definitions of HRQoL. Conflicting findings are reported about the contribution of self-management interventions to HRQoL. The aim of this study was to explore the relationship between self-management and HRQoL in young adults with chronic conditions.

Methods

Adolescents with various chronic conditions participating in a survey in 2006 (T₀) were re-invited for a follow-up study (T₁) in 2012. The young adults (18-25 years of age) reported on background variables, self-management related variables, and HRQoL. The development of HRQoL over time was studied with a paired-sample T-test, while Structural Equation Modeling was used to explore the associations between HRQoL and self-management at T₁ with a multivariate multiple regression analysis.

Results

Young adults with chronic conditions reported a decrease in general HRQoL after their transition to adulthood. Young adults' self-management contributed to their HRQoL (R^2 ranged between .22 and .42). Medical management and emotion management were associated with all domains of HRQoL. Role management was associated only with independence and the social domains of HRQoL.

Conclusions

The findings emphasize the need for 'holistic' support of young people with chronic conditions in their uptake of self-management, in both pediatric and adult care. The inter-relatedness of the self-management domains and the shared associations with HRQoL independence and social domains further justify the notion that self-management support should pay attention to young people's psychosocial needs.

INTRODUCTION

Adolescents growing up with a chronic condition are expected to gradually develop the knowledge, attitudes and skills required for successful self-management. They have to balance common developmental tasks with the medical challenges presented by the chronic condition [1, 2]. Studies show that chronically ill adolescents and young adults are at risk for poorer psychosocial development [2, 3] and less autonomy in social participation [4] than their healthy peers, and have fewer opportunities in adulthood [5]. Also, suboptimal transition from a pediatric to adult health care setting can be associated with adverse outcomes like no-show in medical follow-up or non-adherence to medical treatment [6]. Successful transition to adulthood and engagement in self-management is crucial for a satisfying adult life and fulfillment of social roles [1, 7, 8]. Self-management support is therefore considered an integral part of care for these young people [9-11].

Self-management is described as “the individual’s ability to manage the symptoms, treatment, physical and psychosocial consequences and life style changes inherent in living with a chronic condition”, and the ultimate goal of self-management is defined as maintaining “a satisfactory quality of life” [12]. This is not surprising, since health-related quality of life (HRQoL) incorporates physical, psychological and social functioning [13] – domains that are relevant to self-management as well [14]. As such, HRQoL often serves as a patient-reported outcome measure in the evaluation of self-management interventions [15].

Yet, self-management is neither mentioned nor explored in the most commonly used HRQoL models [16] or HRQoL definitions in the pediatric population [13]. Several reviews on the effectiveness of self-management interventions in (young) people with chronic conditions reported conflicting findings about the contribution of these interventions to HRQoL, suggesting that there is no good insight into the relationship between HRQoL and self-management [17-21]. Gaining insight into this relationship is important, because it may help to adequately support self-management in this group of patients. Therefore, this study aims to explore the associations between self-management and HRQoL in young adults (YA) with chronic conditions.

METHODS

Study design and participants

A longitudinal survey study was conducted in which participants of a web-based survey in 2006 (T₀) [7, 22] were re-invited for a similar survey six years later (T₁). The web-based follow-up questionnaire was basically the same as the previous questionnaire, except that questions on social participation were added. It was pilot tested in face-to-face

(n=7) and telephone (n=3) interviews with young university students with a chronic condition, who were not included in the final sample.

In 2006, participants were adolescents aged 12 to 18 years who had been under treatment at the Erasmus MC – Sophia's Children's Hospital Rotterdam for more than three years. They presented with a broad range of chronic somatic conditions without intellectual disabilities. In 2012, current contact addresses and death notices were retrieved from the hospital's electronic patient records. Eligible YA were sent a letter providing relevant information and a unique password to log in on a secured website. Included was a postcard on which they could state they did not want to participate. Those who did not respond within one month were sent a reminder by mail. After another month, non-responders were reminded through a phone call. Respondents were entered in a lottery to win one of twenty-five cookbooks, two smart phones, or an iPad provided by local suppliers. The Erasmus MC medical ethical review board approved the study (MEC 2012-022) and all data was processed anonymously. Participants provided digital consent for their participation in the study.

Description of survey

The survey addressed three domains: background characteristics, HRQoL, and self-management behaviors and attitudes.

Background characteristics: Age and gender were recorded in both surveys (To and T1). Data of the To questionnaire served to assess physical limitations in mobility (0=no; 1=yes), and ethnicity (1=Dutch surname, 2=non-Dutch surname). At T1, the present level of education was assessed (1=low [pre-vocational education or secondary vocational education; level 1-3]; 2=medium [pre-vocational education or secondary vocational education; level 4]; 3=high [pre-university / senior general secondary education or higher educational institutions]).

Health-Related Quality of Life was assessed using the self-report version of the DISABKIDS questionnaire, originally designed for children and adolescents. The To questionnaire used the short form (index) of the DISABKIDS (DCGM-10) [23], and scores had been transformed to a scale of 0-100. At T1, five domains of HRQoL were measured with the DCGM-37 on a 5-point Likert scale (from 1='often' to 5='never'): Independence (6 items, Cronbach's $\alpha=.85$); Physical (6 items, Cronbach's $\alpha=.84$); Emotion (7 items, Cronbach's $\alpha=.90$); Social exclusion (6 items, Cronbach's $\alpha=.81$); and Social inclusion (6 items, Cronbach's $\alpha=.82$) [23]. While social exclusion refers to feelings of being left out, social inclusion refers to understanding of others and experiencing positive social relationships. A general score was computed by combining all items (Cronbach's $\alpha=.95$). To compare To and T1 scores,

the short form index was also computed with T1 data (as indicated in the DISABKIDS guidelines). Higher scores reflect better HRQoL.

Self-management was operationalized through various self-reported measures addressing the three tasks of self-management: medical management (re. treatment and symptoms), role management (re. social participation), and emotion or identity management (re. emotional consequences of being ill) [14]. This operationalization was based on findings of a review on content of self-management interventions for young people with chronic conditions (Sattoe et al., 2015).

Medical management-related variables:

- 'No show', i.e. the number of missed consultations in the past three years (retrieved from the hospital's electronic patient records at T1 measurement);
- Self-reported adherence to medication regimen measured with the 5-item Medication Adherence Report Scale (MARS-5) at T1 [24, 25]. Items are scored on a 5-point Likert scale (from 1='always true' to 5='never true'; $\alpha=.74$), with higher scores reflecting better adherence;

Role management-related variable:

- Autonomy in social participation, classified using the Rotterdam Transition Profile (RTP) [26]. Participation in five life areas was addressed: 1) employment and education; 2) housing; 3) intimate relationships; 4) transportation; and 5) leisure. Full autonomy in social participation on these domains was recorded as 1, while 0 referred to dependency on adults (e.g. parents), display of typical child behavior, or experimenting with adult behavior or orienting to it [4].

Emotion management-related variable:

- Self-reported self-efficacy measured by the On Your Own Feet Self-Efficacy Scale (OYOF-SES) using a 4-point Likert scale for every item (from 1='yes certainly' to 4='no, definitely not') [7]. The scale consists of three domains: self-efficacy in coping with the condition (4 items, Cronbach's $\alpha=.82$), self-efficacy in knowing about the condition (6 items, Cronbach's $\alpha=.78$), and self-efficacy considering competencies during consultations (6 items, Cronbach's $\alpha=.85$). The original OYOF-SES consists of 17 items, but one item ("I know what will happen to me when I transfer to adult care") was deleted, because it did not apply to those already transferred at T1. A higher total score (16 items, Cronbach's $\alpha=.87$) reflects higher self-efficacy.

Data analysis

Backward logistic analysis served to detect selective response; determinants of study non-response are expressed in odds ratios (ORs) with 95% confidence intervals (95% CI), and Nagelkerke R^2 indicates the proportion of explained variance. Model fit was tested

with the Hosmer and Lemeshow test. Descriptive statistics were used to describe the study sample.

SPSS 21 was used for descriptive statistics. Differences between T₀ and T₁ scores on HRQoL and self-efficacy were tested with paired samples t-tests. Bivariate correlations (Pearson's *r* or Spearman's *ρ*) between the outcome and other variables were computed. Structural Equation Modeling (SEM) using Mplus 7 was then applied to explore the associations of background variables and medical, role, and emotion management variables with the different domains of HRQoL in multivariate multiple regression analysis. This method allows studying the associations between multiple independent and multiple dependent variables, which implies we could explore associations of the background and self-management variables with the interrelated HRQoL domains. The conceptual model is presented in Figure 1. Only variables significantly correlated ($p < .05$) with the outcome variables were included in the final regression model. We used the following indicators of model fit: 1) relative χ^2 (χ^2/df), 2) comparative fit index (CFI), 3) standardized root mean square residual (SRMR), and 4) root mean square error of approximation (RMSEA). They indicate good model fit if relative χ^2 is below 2 [27], CFI is above .95 [28], SRMR is below .08 [29], and RMSEA is below .05 [30].

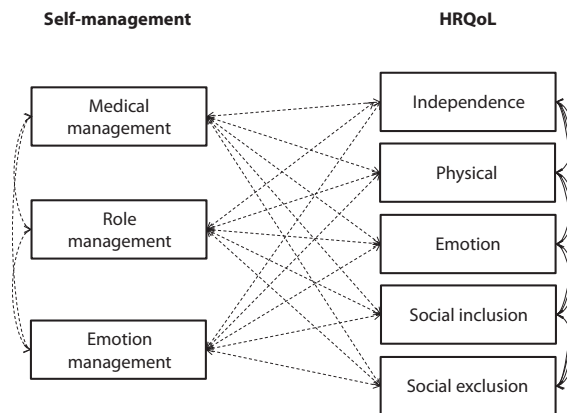


Figure 1 Conceptual model

RESULTS

Response and study sample

Of the 1,039 participants at T₀, 13 (1.3%) had died and 25 (2.4%) could not be traced. Consequently 1,001 were invited 88 (8.8%) of whom returned a postcard stating they declined from participation. Eventually, 518 (51.8%) YA completed the T₁ survey online. Non-response (39.4%) was associated with male gender (OR, .57; 95% CI, .43–.74), and

non-Dutch ethnicity (OR, .49; 95% CI, .33–.74) ($\chi^2=29.0$, $df=2$; $P<.001$; $R^2=.04$; Hosmer and Lemeshow test $P=.77$). Thirty YA (5.8%) were excluded as they did not provide information on HRQoL at both times. The final study sample for analysis included 478 YA (Table 1).

Table 1 Description of the study sample

	n	Study sample at T0	Study sample at T1
Background characteristics			
Age [18-25] [^] , mean in years (\pm SD)	478	-	20.61 (1.90)
Gender, n=male (%)	478	-	180 (37.7)
Ethnicity, n=non-Dutch surname (%)	478	-	51 (10.7)
Educational level, n=high (%)	471	-	212 (45.0)
Physical limitation(s), n=yes (%)	478	-	137 (28.7)
Health-Related Quality of Life [0-100] [^] , mean (\pm SD)			
General HRQoL (short index)	478	80.50 (15.41)*	78.26 (18.22)*
Independence domain	478	-	83.99 (15.87)
Physical domain	478	-	71.16 (22.46)
Emotion domain	478	-	76.31 (21.01)
Social inclusion domain	478	-	76.46 (19.45)
Social exclusion domain	478	-	83.36 (18.06)
Medical management , mean (\pm SD)			
No-show at consultations in the past three years	471	-	.30 (.88)
Adherence [5-25] [^]	212	-	21.83 (2.92)
Role management , n=full autonomy (%)			
Employment and education	478	-	130 (27.2)
Housing	478	-	90 (18.8)
Intimate relationships	478	-	192 (40.2)
Transportation	478	-	441 (92.3)
Leisure	478	-	262 (54.8)
Emotion management , mean (\pm SD)			
Self-efficacy [10-64] [^]	353	52.81 (6.87)*	55.83 (6.56)*

[^] theoretical range

* Paired samples T-test: $p<.01$

HRQoL development over time

Mean general HRQoL at To was 80.50 ± 15.41 . At T1 this was significantly lower $T_1: 78.26\pm18.22$ ($p<.01$). The use of an index of HRQoL at To limited further exploration of the domains in the longitudinal analysis. At T1, on average YA scored highest on the

independence domain of HRQoL (83.99 ± 15.87), followed by the social exclusion domain (83.36 ± 18.06). The mean score was lowest on the physical domain (71.16 ± 22.46) (Table 1).

The final multivariate multiple regression model

Bivariate correlation analysis (Table 2) determined which of the T1 variables should be included in the final model (Figure 2), because including all independent variables for each dependent variable would have resulted in a saturated model. Significant correlations were those with a p -value $< .05$.

The variable 'self-reported adherence' was excluded from the final multivariate model because only 212 of the 478 YA had a medical regimen to adhere to, and filled out the MARS-5. Otherwise, over one-third of the cases would have to be disregarded.

For the independence, physical, emotion, social exclusion and social exclusion domains of HRQoL the background and self-management variables explained 30%, 22%, 24%, 28% and 42% of the total variance, respectively, see Table 3. The covariance between independent variables was not significant, indicating the absence of multicollinearity. The correlations between the domains of HRQoL is presented in Table 4. The model fit indices indicated good model fit: $\chi^2(11) = 13.45$, $p = .120$; CFI = .997; SRMR = .019; RMSEA = .031.

Associations with HRQoL

All self-management domains were associated with scores on the independence domain. The emotion management variable, i.e. self-efficacy, had the strongest association ($\beta = .39$; $p < .001$), followed by autonomy in social participation on the leisure domain ($\beta = .13$; $p < .01$). Men reported higher HRQoL on the independence domain than did women ($\beta = .12$; $p < .01$), while YA with physical limitations reported lower HRQoL on this domain ($\beta = -.15$; $p < .01$) (Table 3).

All but the role management-related variables were associated with HRQoL score on the physical domain. The disease-related background variable and medical management-related variable had the strongest associations: YA with physical limitations or more missed consultations reported lower HRQoL on the physical domain ($\beta = -.30$; $p < .001$ and $\beta = -.15$; $p < .01$ respectively). Self-efficacy had the strongest association with the emotion domain of HRQoL ($\beta = .32$; $p < .001$), but this domain was also associated with missed consultations ($\beta = -.15$; $p < .01$), gender ($\beta = .19$; $p < .001$), and the presence of physical limitations ($\beta = -.18$; $p < .001$) (Table 3).

Furthermore, scores on the social exclusion domain were also most strongly associated with self-efficacy ($\beta = .38$; $p < .001$), followed by the presence of physical limitations ($\beta = -.21$; $p < .001$), and missed consultations ($\beta = -.10$; $p < .05$). The role management-related variables were associated with the social inclusion domain, but not with the social exclusion domain. Social participation on the leisure domain had a relatively

Table 2 Bivariate correlations of variables with the HRQoL domains (T1): Pearson's or Spearman's correlation coefficient r/p

	n	Independence	p	Physical	p	Emotion	p	Social exclusion	p	Social inclusion	p
Background											
Age	478	-.05	.276	-.07	.129	-.13	.004	-.11	.021	-.03	.560
Gender (male)	478	.17	.000	.21	.000	.23	.000	.10	.026	.16	.001
Ethnicity (non-Dutch surname)	478	-.06	.162	-.04	.342	-.11	.017	-.08	.069	-.06	.200
Educational level (high)	471	.08	.071	.07	.107	.06	.206	.12	.008	.10	.037
Presence of physical limitations	478	-.23	.000	-.34	.000	-.22	.000	-.26	.000	-.20	.000
Medical management											
Missed consultations	471	-.12	.009	-.19	.000	-.15	.001	-.11	.020	-.12	.012
Adherence	212	.16	.019	.11	.101	.12	.072	.17	.014	.11	.105
Role management											
Social participation: vocational	478	-.03	.538	.04	.436	-.06	.172	-.02	.655	.02	.706
Social participation: living	478	.01	.832	-.01	.817	-.07	.113	-.03	.583	.00	.992
Social participation: relations	478	.04	.345	-.01	.842	-.06	.224	.09	.051	.06	.225
Social participation: mobility	478	.19	.000	.18	.000	.12	.008	.20	.000	.21	.000
Social participation: leisure	478	.29	.000	.19	.000	.19	.000	.17	.000	.37	.000
Emotion management											
Self-efficacy T1	369	.46	.000	.20	.000	.36	.000	.43	.000	.55	.000
HRQoL											
Short index T0	478	.42	.000	.36	.000	.43	.000	.42	.000	.36	.000

high beta ($\beta = .24$; $p < .001$), next to self-efficacy ($\beta = .46$; $p < .001$). Having more missed consultations was also associated with lower HRQoL on the social inclusion domain ($\beta = -.14$; $p < .01$). Gender was weakly associated with both social domains of HRQoL ($\beta = .10$; $p < .05$ and $\beta = .08$ $p < .05$) (Table 3).

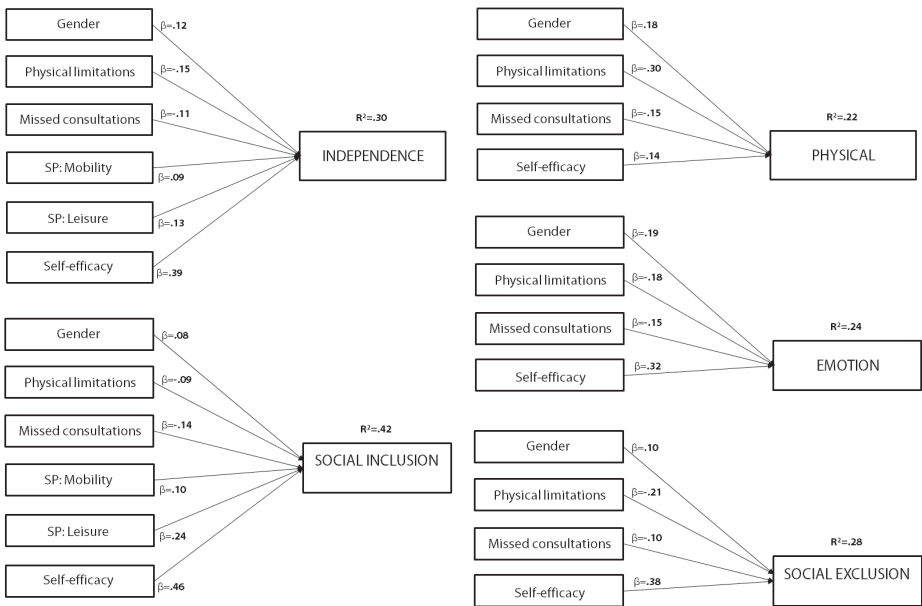


Figure 2 Final model

Table 3 Multivariate multiple regression model of associations with domains of HRQoL (n=359)^a

	Standardized estimates (β)	Standard errors	p-value
INDEPENDENCE ($R^2 = .30$)			
Gender (male)	.12	.04	.007
Presence of physical limitations (yes)	-.15	.05	.001
Missed consultations	-.11	.04	.011
Social participation: mobility	.09	.05	.045
Social participation: leisure	.13	.05	.005
Self-efficacy	.39	.04	.000
PHYSICAL ($R^2 = .22$)			
Gender (male)	.18	.05	.000
Presence of physical limitations (yes)	-.30	.05	.000
Missed consultations	-.15	.05	.002
Social participation: mobility	.07	.05	.140
Social participation: leisure	.07	.05	.161
Self-efficacy	.14	.05	.005
EMOTION ($R^2 = .24$)			
Age	-.04	.04	.254
Gender (male)	.19	.05	.000
Ethnicity (non-Dutch surname)	-.04	.03	.183
Presence of physical limitations (yes)	-.18	.05	.000
Missed consultations	-.15	.05	.002
Social participation: mobility	.01	.05	.783
Social participation: leisure	.05	.05	.289
Self-efficacy	.32	.05	.000
SOCIAL EXCLUSION ($R^2 = .28$)			
Age	-.05	.04	.178
Gender (male)	.10	.05	.024
Educational level	.08	.04	.034
Presence of physical limitations (yes)	-.21	.05	.000
Missed consultations	-.10	.05	.021
Social participation: mobility	.09	.05	.063
Social participation: leisure	.01	.05	.858
Self-efficacy	.38	.04	.000
SOCIAL INCLUSION ($R^2 = .42$)			
Gender (male)	.08	.04	.030
Educational level	.01	.03	.708
Presence of physical limitations (yes)	-.09	.04	.032
Missed consultations	-.14	.04	.001
Social participation: mobility	.10	.04	.016
Social participation: leisure	.24	.04	.000
Self-efficacy	.46	.04	.000

^aNote: $\chi^2 (11) = 13.45$, $p = .120$; CFI = .997; SRMR = .019; RMSEA = .031

Table 4 Correlations between domains of HRQoL ($p < .001$)

	Social inclusion	Social exclusion	Emotion	Physical
Independence	.52	.55	.63	.57
Physical	.47	.44	.60	
Emotion	.36	.59		
Social exclusion	.39			

DISCUSSION

YA perceived HRQoL on average had decreased after the transition from adolescence to young adulthood. Although there is evidence in some populations that older chronically ill adolescents report lower HRQoL than do younger adolescents and children [31-33], there is little understanding of how HRQoL evolves with transition into adulthood. Taylor and colleagues stressed the influence of developmental state on HRQoL. They found that chronically ill adolescents' urge to strive for normality increases as they develop [13]. Having to deal with barriers encountered in their efforts to be on par with healthy peers might perhaps explain the lower HRQoL at young adult age. Note that most YA with chronic conditions are at risk to lag behind in social participation and reaching developmental milestones compared to healthy populations [2, 4, 34]. This may also explain why having a physical limitation was negatively associated with all domains of HRQoL, for it is highly likely that someone with a physical limitation faces more barriers in daily life than someone without physical limitations.

Male gender was positively associated with all domains of HRQoL. While gender differences in HRQoL are not always present during childhood, females in older chronically ill populations often report lower general HRQoL [33, 35-37]. In our sample, associations were weak in the social domains of HRQoL. A recent study among young adults with beneficiary benefits (recipients under the Dutch Income Provision Act for Disabled Young People) in the Netherlands showed that female gender was negatively associated with the physical domain of HRQoL, but not with the mental domain [37]. These gender differences may suggest that women may need more support in living with a chronic condition, although greater need of support in part may be explained by socioeconomic status [38, 39].

Furthermore, higher self-efficacy in young adulthood was associated with better HRQoL on all domains. Self-efficacy has been reported as a predictor for positive health outcomes or self-care in, for example, diabetes [40-42] and asthma [43]. Higher self-efficacy is also associated with better school performance of young people with cystic fibrosis (CF) [44], better emotional outcomes in YA with CF or pediatric cancer survivors [45], better adaption to the chronic condition and less condition-related distress in diabetes [46, 47], better quality of life in adolescents with juvenile idiopathic arthritis

and diabetes [48] and more transfer readiness in young people with a chronic condition in general [22]. Our finding that self-efficacy is associated with all domains of HRQoL and that it was the most important predictor for all but the physical domain supports the use of interventions that aim to enhance self-efficacy of young people with a chronic condition.

Missing consultations in current care was negatively related to HRQoL on all domains. This suggests that special attention for young people in transition to adulthood should be continued after transfer to adult care. Missed consultations may be an important indicator of a lack of continuity of care for YA, but also for problems in psychosocial functioning. Continuous monitoring of medical but also the psychosocial needs of young people is essential. Especially since social participation in the mobility and leisure domains seemed important for HRQoL on the social inclusion and independence domains. Interventions such as the KLIK PROfile [49, 50], which allows for systematic monitoring of patient-reported outcomes, including HRQoL, might be useful to organize the much-needed attention for young adults in adult healthcare settings.

Despite the similarity between the domains of HRQoL and those of self-management, we found no one-to-one association between them. The medical management domain, for instance, was associated with all HRQoL domains and not only the physical one (as was to be expected). Similarly, the emotion management domain did not only correlate with the emotion domain of HRQoL, but also with all the other domains. These findings emphasize strong interrelatedness of self-management and HRQoL, and consequently the need for holistic self-management support. Future research should further clarify which self-management processes are linked to HRQoL, and should explore possible intermediating variables.

Other considerations

Our study was performed in the largest university hospital in the Netherlands, which comprises all major pediatric subspecialties. It included a large sample of young persons that was heterogeneous in terms of congenital and acquired conditions, and in age. The wide range of chronic conditions made it impossible, however, to explore the impact of nature of the disease and that of disease severity. Disease- or age-specific studies could yield more specified results. On the other hand, studying chronic conditions in general is not considered a flaw, because the challenges and adaptive tasks young people have are similar across conditions [3].

We operationalized medical management by measuring missed consultations, role management by measuring autonomy in social participation, and emotion management by measuring self-efficacy. Since self-management is a broad and multidimensional construct, in this way we probably did not capture all relevant elements, as is confirmed by our model's explained variance. We could for instance not include therapeutic adher-

ence. Further studies are needed to investigate which elements of self-management interact with or contribute to HRQoL.

Lastly, the DISABKIDS (DCGM-37) questionnaire was included in both surveys. This instrument was developed for use in children and adolescents, and has not been validated for the young adult population. Still, the Cronbach's alpha values for both the subscales and the total score in our sample were high, supporting its use. On the other hand, the fact that we measured the short index only at T0 limited the over-time analysis. We could for instance not elaborate on the changes in specific domains. More studies are needed to research HRQoL of young people with chronic conditions across their life span.

CONCLUSIONS

Chronically ill YA reported a decrease in general HRQoL after their transition to adulthood. Their self-management was related to their HRQoL. While medical management and emotion management were associated with all domains of HRQoL, role management was particularly associated with the independence and social domains of HRQoL. These findings emphasize the need for 'holistic' self-management support taking into account the developmental tasks of these young people, both in pediatric and adult care. Such self-management support might very well aim to build self-efficacy. The interrelatedness of the self-management domains and the shared associations with the independence and social domains further support the notion that self-management support should include attention for young people's psychosocial needs.

ACKNOWLEDGMENTS

We thank Heleen van der Stege and Sander Hilberink for their contribution to the design, execution, and reporting of the study. Kimberly Onderdijk and Esmee Kuppen are thanked for their help in pretesting the questionnaire, while Laila Oumansour, Wendy de Boer, Marjolijn Bal, and Mariëlle Peeters are thanked for their assistance in during data collection. We are grateful to Mathilde Strating (from Erasmus University Rotterdam) for her statistical advice considering our analysis. Finally, we thank Prof. Bert van der Heijden, then head of the Department of Pediatrics of Erasmus Medical Center, for providing us with the original idea to perform this study.

REFERENCES

1. Turkel S, Pao M: Late consequences of chronic pediatric illness. *Psychiatr Clin North Am* 2007, 30(4): 819-835.
2. 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.
3. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.
4. Sattoe JNT, Hilberink SR, van Staa AL, Bal R: Lagging behind or not? Four distinctive social participation patterns among young adults with chronic conditions. *J Adolesc Health* 2014, 54(4):397-403.
5. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, Reiss J, Wood D: 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.
6. Lugasi T, Achille M, Stevenson M: Patients' perspective on factors that facilitate transition from child-centered to adult-centered health care: a theory integrated metasummary of quantitative and qualitative studies. *J Adolesc Health* 2011, 48(5):429-440.
7. Van Staa AL: On Your Own Feet: Adolescents with chronic conditions and their preferences and competencies for care (Doctoral dissertation). Rotterdam: Rotterdam University; 2012.
8. 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.
9. Harvey PW, Petkov JN, Misan G, Fuller J, Battersby MW, Cayetano TN, Warren K, Holmes P: Self-management support and training for patients with chronic and complex conditions improves health-related behaviour and health outcomes. *Aust Health Rev* 2008, 32(2):330-338.
10. Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, Hoes A, Schuurmans M: Self-management: one size does not fit all. *Patient Educ Couns* 2013, 92(1):134-137.
11. Wagner EH, Austin BT, Davis C, Hindmarsh M, Schaefer J, Bonomi A: Improving chronic illness care: translating evidence into action. *Health Aff (Millwood)* 2001, 20(6):64-78.
12. 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.
13. 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.
14. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26(1):1-7.
15. Santana MJ, Feeny D: Framework to assess the effects of using patient-reported outcome measures in chronic care management. *Qual Life Res* 2014, 23(5):1505-1513.
16. Bakas T, McLennon SM, Carpenter JS, Buelow JM, Otte JL, Hanna KM, Ellett ML, Hadler KA, Welch JL: Systematic review of health-related quality of life models. *Health Qual Life Outcomes* 2012, 10: 134.
17. Lindsay S, Kingsnorth S, McDougall C, Keating H: A systematic review of self-management interventions for children and youth with physical disabilities. *Disabil Rehabil* 2014, 36(4):276-88.
18. Novak M, Costantini L, Schneider S, Beanlands H: Approaches to self-management in chronic illness. *Semin Dial* 2013, 26(2):188-194.
19. Jonsdottir H: Self-management programmes for people living with chronic obstructive pulmonary disease: a call for a reconceptualisation. *J Clin Nurs* 2013, 22(5-6):621-637.

20. Newman S, Steed L, Mulligan K: Self-management interventions for chronic illness. *Lancet* 2004, 364(9444):1523-1537.
21. Kirk S, Beatty S, Callery P, Gellatly J, Milnes L, Prymachuk S: The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child Care Health Dev* 2013, 39(3):305-324.
22. van Staa AL, 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.
23. Ravens-Sieberer U, Schmidt S, Gosch A, Erhart M, Petersen C, Bullinger M: Measuring subjective health in children and adolescents: results of the European KIDSCREEN/DISABKIDS Project. *Psychosoc Med* 2007, 4:Doco8.
24. Horne R, Hankins M: The Medication Adherence Report Scale (MARS): a new measurement tool for eliciting patients' reports of non-adherence. Unpublished Working Paper; 2007.
25. Mahler C, Hermann K, Horne R, Ludt S, Haefeli WE, Szecsenyi J, Jank S: Assessing reported adherence to pharmacological treatment recommendations. Translation and evaluation of the Medication Adherence Report Scale (MARS) in Germany. *J Eval Clin Pract* 2010, 16(3):574-579.
26. Donkervoort M, Wiegerink DJ, van Meeteren J, Stam HJ, Roebroek ME: Transition to adulthood: validation of the Rotterdam Transition Profile for young adults with cerebral palsy and normal intelligence. *Dev Med Child Neurol* 2009, 51(1):53-62.
27. Bollen KA: Structural equations with latent variables. New York: Wiley; 1989.
28. Bentler PM: Comparative fit indexes in structural models. *Psychol Bull* 1990, 107(2):238-246.
29. Hu L, Bentler PM: Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. *Struc Equ Modeling* 1999, 6(1):1-55.
30. MacCallum RC, Browne MW, Sugawara HM: Power analysis and determination of sample size for covariance structure modeling. *Psychological Methods* 1996, 1(2):130-149.
31. van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, Wensink-Boonstra AE, Stam HJ, Roebroek 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.
32. Raty LK, Wilde Larsson BM, Soderfeldt BA: Health-related quality of life in youth: a comparison between adolescents and young adults with uncomplicated epilepsy and healthy controls. *J Adolesc Health* 2003, 33(4):252-258.
33. Michel G, Bisegger C, Fuhr DC, Abel T: Age and gender differences in health-related quality of life of children and adolescents in Europe: a multilevel analysis. *Qual Life Res* 2009, 18(9): 1147-1157.
34. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis M: 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.
35. Bisegger C, Cloetta B, von Rueden U, Abel T, Ravens-Sieberer U: Health-related quality of life: gender differences in childhood and adolescence. *Soz Praventivmed* 2005, 50(5):281-291.
36. Jorngarden A, Wettergen L, von Essen L: Measuring health-related quality of life in adolescents and young adults: Swedish normative data for the SF-36 and the HADS, and the influence of age, gender, and method of administration. *Health Qual Life Outcomes* 2006, 4:91.
37. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis M: Health-related quality of life, anxiety and depression in young adults with disability benefits due to childhood-onset somatic conditions. *Child Adolesc Psychiatry Ment Health* 2013, 7(1):12.
38. Cherepanov D, Palta M, Fryback DG, Robert SA: Gender differences in health-related quality-of-life are partly explained by sociodemographic and socioeconomic variation between adult men

- and women in the US: evidence from four US nationally representative data sets. *Qual Life Res* 2010, 19(8):1115-1124.
39. Cherepanov D, Palta M, Fryback DG, Robert SA, Hays RD, Kaplan RM: Gender differences in multiple underlying dimensions of health-related quality of life are associated with sociodemographic and socioeconomic status. *Med Care* 2011, 49(11):1021-1030.
 40. Austin S, Guay F, Senecal C, Fernet C, Nouwen A: Longitudinal testing of a dietary self-care motivational model in adolescents with diabetes. *J Psychosom Res* 2013, 75(2):153-159.
 41. Berg CA, King PS, Butler JM, Pham P, Palmer D, Wiebe DJ: Parental involvement and adolescents' diabetes management: the mediating role of self-efficacy and externalizing and internalizing behaviors. *J Pediatr Psychol* 2011, 36(3):329-339.
 42. Chih AH, Jan CF, Shu SG, Lue BH: Self-efficacy affects blood sugar control among adolescents with type I diabetes mellitus. *J Formos Med Assoc* 2010, 109(7):503-510.
 43. Gandhi PK, Kenzik KM, Thompson LA, DeWalt DA, Revicki DA, Shenkman EA, Huang IC: Exploring factors influencing asthma control and asthma-specific health-related quality of life among children. *Respir Res* 2013, 14:26.
 44. Grieve AJ, Tluczek A, Racine-Gilles CN, Laxova A, Albers CA, Farrell PM: Associations between academic achievement and psychosocial variables in adolescents with cystic fibrosis. *J Sch Health* 2011, 81(11):713-720.
 45. Schwartz LA, Drotar D: Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *J Pediatr Psychol* 2009, 34(9):954-965.
 46. Whittemore R, Jaser S, Guo J, Grey M: A conceptual model of childhood adaptation to type 1 diabetes. *Nurs Outlook* 2010, 58(5):242-251.
 47. Law GU, Walsh J, Queral V, Nouwen A: Adolescent and parent diabetes distress in type 1 diabetes: the role of self-efficacy, perceived consequences, family responsibility and adolescent-parent discrepancies. *J Psychosom Res* 2013, 74(4):334-339.
 48. Cramm JM, Strating MM, Nieboer AP: The Importance of General Self-Efficacy for the Quality of Life of Adolescents with Diabetes or Juvenile Rheumatoid Arthritis Over Time: A Longitudinal Study among Adolescents and Parents. *Front Pediatr* 2013, 1:40.
 49. Haverman L, Engelen V, van Rossum MA, Heymans HS, Grootenhuys MA: Monitoring health-related quality of life in paediatric practice: development of an innovative web-based application. *BMC Pediatr* 2011, 11:3.
 50. Haverman L, van Rossum MA, van Veenendaal M, van den Berg JM, Dolman KM, Swart J, Kuijpers TW, Grootenhuys MA: Effectiveness of a web-based application to monitor health-related quality of life. *Pediatrics* 2013, 131(2):e533-543.

PART III

Self-management support for young
people with chronic conditions:
Intervention effectiveness



8

Exploring effectiveness and effective components of self-management interventions for young people with chronic conditions: A systematic review



Marjolijn I. Bal, Jane N.T. Sattoe, Pepijn D.D.M. Roelofs, Roland Bal,
AnneLoes van Staa, Harald S. Miedema

Submitted

ABSTRACT

Importance

There is a lack of clarity regarding effectiveness and effective intervention components of self-management support for young people with a variety of chronic conditions.

Objective

To systematically explore the effectiveness and effective intervention components of self-management support for young people with a variety of chronic conditions.

Evidence review

Databases searched were Embase, Medline, PsycINFO, Web-of-Science, CINAHL, and Cochrane; and additionally relevant reviews' reference lists were scrutinized. Study selection was based on the following criteria: Original articles in English published between 2003 and February 2014; focusing on youth aged 7-25 years with somatic chronic conditions/physical disabilities; describing self-management interventions; with clear outcome measures; using a RCT design. Methodological quality was independently assessed using the methodology checklists of the Scottish Intercollegiate Guidelines Network. Quality of evidence was rated according the Grades of Recommendation, Assessment, Development and Evaluation Working Group approach. Data were extracted on study design; outcome measures and results; and intervention components.

Findings

31 RCTs were included, showing a trend in favor of interventions targeting disease knowledge, adherence and dealing with a chronic condition; a trend in favor of usual care regarding improvement of symptoms and school attendance; no clear pattern regarding psychological outcomes and quality of life. Interventions focused on medical management, provided individually in a clinical setting by a mono-disciplinary team showed a trend in improving adherence. Interventions in home setting and those delivered individually showed a trend in improving dealing with the chronic condition.

Conclusions and Relevance

Adherence could be improved through interventions focused on medical management, provided individually in a clinical setting by a multidisciplinary team. Interventions focused on dealing with the chronic condition in daily life might be provided individually and through telemedicine programs facilitating peer-support. More solid evidence should come from more standardized effectiveness research on self-management interventions.

INTRODUCTION

Advancements in medical care result in increasing numbers of young people with chronic conditions making the transition into adulthood [1, 2]. This transition, which implies becoming an independent adult fulfilling different roles in society, requires complicated life changes [3,4]. For these young people the transition may be more complicated, however, as they often lag behind in social and emotional development compared to healthy peers [5, 6]. It seems crucial, therefore, to help them develop self-management skills [7, 9].

Barlow and colleagues (2002) define self-management as “the individual’s ability to manage the symptoms and the consequences of living with a chronic condition, including treatment, physical, social, and lifestyle changes” [10-12]. As such, self-management and self-management support encompass three elements: medical management (considering the treatment), role management (considering participation in society), and emotion management (considering emotional consequences of being ill) [13]. This broad definition of self-management is widely used these days, as it fits with the World Health Organization’s definition of health in terms of physical, mental and social well-being [14].

A range of pediatric interventions are available for health care providers to support self-management in chronic disease [7-9, 15, 16]. These have been reviewed in some studies [17, 18], but solid evidence on effectiveness of self-management interventions (SMI) for young people with chronic conditions in general, i.e. irrespective of diagnosis, is still lacking [17-23]. Specifically, Kirk et al. (2013) restricted themselves to asthma, cystic fibrosis and diabetes [18], while Lindsay et al. (2014) restricted themselves to physical disabilities, excluding people with somatic chronic conditions [17]. Moreover, although Kirk and colleagues reviewed effective components of SMI for young people with asthma, cystic fibrosis and diabetes [18], insights into effective intervention components of self-management support for young people with a variety of chronic conditions are still lacking.

Still, it is important to look at self-management support in a way that goes beyond particular chronic conditions [24]. Despite disease-specific differences, there are many commonalities between young people with chronic condition, because they all face comparable challenges and similar adaptive tasks while growing up [19]. A non-categorical approach to self-management support may benefit both research and health service delivery [25]. First, because this approach could facilitate development and evaluation of interventions. Second, because it could allow pediatric specialist teams to learn from each other, for instance by using (partly) the same SMI [25].

In a parallel paper including SMI evaluation studies, we provided an overview of the content of self-management support for youth with chronic conditions in general [26].

However, we did not include results about effectiveness. Therefore, in the current paper, we reviewed published randomized controlled trials of SMI elaborating on a) evidence regarding effectiveness of SMI focused on youth with chronic conditions; and b) effective intervention components of these SMI, by employing a non-categorical approach to chronic conditions. Such insights may provide understanding of what elements of self-management support might be effective across conditions, and hence may be valuable to further improve self-management support in pediatric care.

METHODS

Study design

We applied a systematic review methodology which Grant and Booth defined as “systematically searching for, appraising and synthesizing research evidence, often adhering to guidelines for conducting a review” [27]. Characteristics of a systematic review methodology are: exhaustive and comprehensive searching, quality assessment, narrative synthesis with tabular accompaniment, results of uncertainty around findings, recommendations for practice and future research [27]. The PRISMA statement guided the review process [28].

Search strategy

The search strategy employed variations and Boolean connections (AND, OR) of the following terms: self-management, children and adolescents, chronic illness, and intervention. Relevant variations of search terms were derived from database thesauruses and relevant review articles. An information specialist helped define the final search strategy, employing a combination of free-text and thesaurus terms. The strategy used in Embase is presented in Box 1. Besides Embase, five other databases were searched: Medline,

Box 1 Search strategy in Embase

```
((('self care'/de OR 'self medication'/de OR 'self help'/de OR 'drug self administration'/de OR (((self OR shared) NEAR/3 (manag* OR care* OR medicat* OR efficac* OR help*))) :ab,ti) OR (((('coping behavior'/exp OR 'health education'/de OR 'patient education'/de OR emotion/de OR emotionality/de) AND ('intervention study'/de OR psychotherapy/exp OR 'program development'/de)) OR (psychotherap* OR ((coping OR cope OR cognitiv* OR behavior* OR emotion* OR education* OR psycholog* NEAR/6 (therap* OR interven* OR program*))) :ab,ti)) AND ('chronic disease'/de OR 'genetic and familial disorders'/exp OR 'congenital disorder'/exp OR 'disabled person'/de OR 'handicapped child'/de OR disability/exp OR (((chronic* OR longterm OR 'long term' OR 'end stage' OR endstage* OR degenerat* OR persisten* OR genetic* OR familial* OR congenit*) NEAR/3 (ill* OR disease* OR condition* OR disorder*)) OR (physic* NEAR/3 (handicap* OR disab* OR challeng*))) :de,ab,ti) AND (child/exp OR adolescent/exp OR adolescence/exp OR 'child health care'/de OR 'child care'/de OR 'child hospitalization'/de OR 'handicapped child'/de OR (young OR youth OR child* OR adolescen* OR teenage* OR teen OR teens OR juvenile*) :ab,ti) AND ('comparative effectiveness'/de OR 'clinical effectiveness'/de OR evaluation/de OR 'self evaluation'/de OR (effectiv* OR evaluat*) :ab,ti))
```

PsycINFO, Web-of-Science, CINAHL, and Cochrane. Two researchers (JS, MB) completed the database searches by scrutinizing relevant reviews' references for additional relevant publications.

Inclusion criteria

- *Study design*: studies using a randomized controlled study design.
- *Study types*: only original research articles in English language published from 2003 - February 2014, because the focus in literature on self-management rapidly increased since 2003 [29].
- *Interventions*: studies focusing on the evaluation of SMI and describing the SMI or referring to previous description(s) of the intervention.
- *Outcome measures*: studies considering clearly defined outcome measures.
- *Participants*: studies focusing on young people aged 7–25 years with somatic chronic conditions or physical disability. The age of seven years is considered a developmentally appropriate age to start the development of independence [30]. Since young adults are still developing their full potential, the age range was extended from 18 to 25 years [31, 32]. Studies had to meet all inclusion criteria to be included for further analysis.

Selection, quality assessment, and data extraction

Retrieved records identified in databases and relevant reviews' reference lists were imported into Endnote X7.2®. Two reviewers (JS, MB) independently categorized studies into: 'include', 'exclude' or 'not clear' based on title and abstract. Thereafter, the reviewers discussed the studies for which inclusion/exclusion was unclear and aimed to reach consensus on the decision. If doubt remained, a third reviewer was consulted (PR). Then, the two reviewers independently decided on the inclusion of articles based on the full text. Three reviewers (JS, MB, PR) independently assessed methodological quality of all included studies using the methodology checklists of the Scottish Intercollegiate Guidelines Network (SIGN) for randomized controlled trials [33]. Methodological quality was defined as high when at least eight out of ten criteria were met; as moderate when from five till eight were met; and as low when fewer than five criteria were met [33]. Any discrepancies in assessment of methodological quality were resolved by discussion. Two reviewers (JS, MB) extracted data on study design; study sample; self-management domain considered during the intervention; interventions' theoretical base, formats, elements, settings, interventionists; outcome measures and study results [34]. Data were recorded in an electronic extraction form.

Analysis

The following seven categories of outcome measures were inductively derived from the data: 1) quality of life; 2) disease knowledge; 3) symptoms; 4) adherence; 5) dealing with the chronic condition (including self-efficacy, coping and problem-solving); 6) psychological outcomes (including depression, stress, anxiety); and 7) school attendance. Two reviewers (JS, MB) independently clustered the specific study outcome measures into these categories.

For each category of outcome measures the quality of evidence was rated as low, moderate and high as suggested in the Grades of Recommendation, Assessment, Development and Evaluation Working Group (GRADE) approach [35]. Basically, evidence of randomized controlled trials (RCT's) is rated as high rating but may be modified downwards on the basis of five different criteria: (1) risk of bias: assessment of methodological quality; (2) inconsistency: heterogeneity or variability in results across studies; (3) indirectness of evidence: indirect comparison of interventions; (4) imprecision of results: wide confidence intervals around the estimate of the effect; and (5) likelihood of publication bias: underestimating or overestimating of the effect due to the selective publication of study [36-40]. Problems on a criterion was defined as serious when 60 - 80 percent of the studies showed limitations, and very serious when > 80 percent of the studies showed limitations on the criterion. Quality of evidence was single downgraded for serious study limitations and double downgraded for very serious study limitations.

Analyses to explore the effectiveness and effective intervention components of self-management support were performed for each outcome measure separately. The random effects model analysis was applied in which standardized mean differences per study were calculated [41]. In this way, we could compare effect sizes of statistically heterogeneous studies and provide an overview of effects on an outcome measure measured with different measurement instruments. Pooled estimates were not calculated, because interventions were clinically diverse and studies statistically diverse. In addition, to evaluate the effective intervention components, we compared the effect sizes of included studies that differed on particular intervention components. For instance, effect sizes of individual intervention programs were compared with effect sizes of group intervention programs. Likewise, interventions' focus on self-management domain, elements, interventionists, and settings were compared. We speak of 'a trend in favor of', because we could not perform a meta-analysis (due to the heterogeneity in measurement instruments) and thus cannot state whether an intervention or its components are indeed effective or not.

A pattern of effects was described as a trend in favor of the intervention or usual care, if more than 67% of the studies on a particular outcome measure pointed that way. Otherwise, the pattern was described as showing no clear effects. We set this 67% cut-off point by ourselves. A pattern was only described if there were more than three

studies with a particular intervention component. Individual studies that showed a significant effect were weighted twice and individual studies that showed a trend (but not a significant effect) were weighted once. Review Manager version 5.1 software was used for the analyses.

RESULTS

Inclusion process

The selection process and reasons for exclusion are presented in Figure 1. The search strategy identified 6373 publications. The two reviewers judged 490 articles as potentially eligible for inclusion. After having read the full-texts of these 490 articles, they eventually included 31 studies.

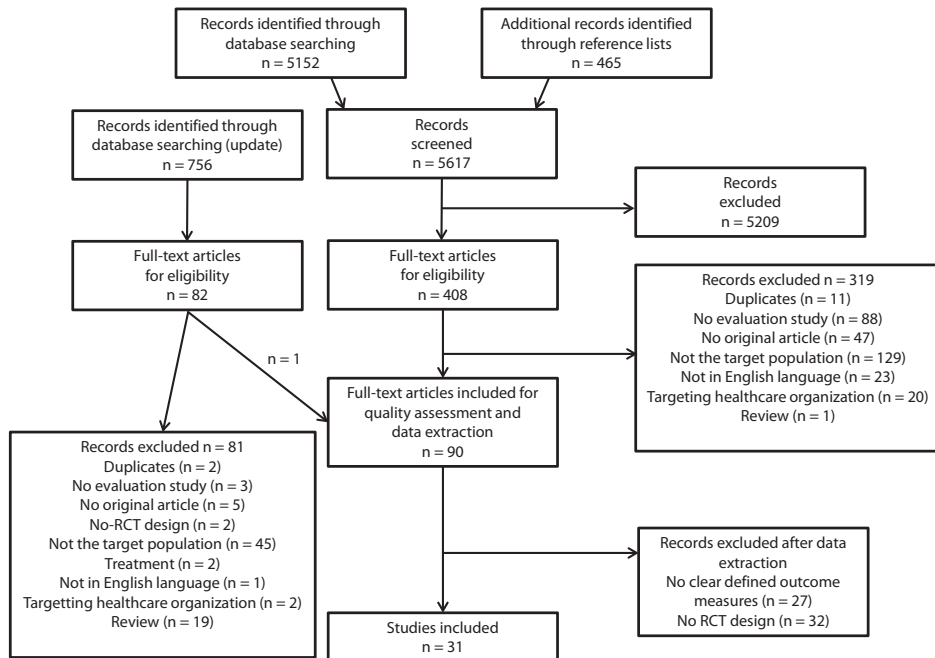


Figure 1 Selection process

Study and intervention components

Study and intervention components are summarized in Table 1. Detailed description of intervention components per outcome measure is presented in Appendix C8.1.

Most of the other studies referred to learning theories like Bandura's (cognitive) social learning theory (n=3), cognitive behavioral theory (n=2) or a combination of these (n=1).

Table 1 Study characteristics and intervention components

Background			Participants		Study characteristics			Components of self-management interventions				
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventions	Setting
Barakat 2010	USA	Sickle cell disease	N = 37 I = 17 C = 20	Low	Another specific intervention	Symptoms Dealing with the chronic condition in daily life School-attendance Disease knowledge	NA	MM	Individual	Education Cognitive restructuring Self-monitoring Relaxation training	Psychologist	Home
Beebe 2010	USA	Asthma	N = 22	Low	Waitlist	Quality of Life Psychological outcomes	NA	MM+EM	Group	Art-therapy	NA	School
Betz 2010	USA	Spina Bifida	N = 65 I = 31 C = 34	Low	Usual care	Psychological outcomes	NA	MM+RM	Group	Goal-setting skills	NA	Clinic
Butz 2005	USA	Asthma	N = 200 I = 112 C = 98	Moderate	Usual care	Quality of Life Dealing with the chronic condition in daily life Disease knowledge	NA	MM	Group	Education	Asthma educator	School
Chiang 2009	Taiwan	Asthma	N = 48 I = 22 C = 26	Moderate	Another specific intervention	Quality of Life Symptoms Psychological outcomes	NA	MM	Individual	Education Self-monitoring Relaxation training	Nurse (undergraduate degree)	Home Clinic
Christian 2006	USA	Cystic Fibrosis	N = 116 I = 58 C = 58	High	Usual care	Quality of Life Symptoms	NA	RM	Individual Group	Education Problem solving skills Peer-support	NA	Home Clinic

Table 1 Study characteristics and intervention components (continued)

Background		Participants		Study characteristics			Components of self-management interventions					
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventions	Setting
Connelly 2006	USA	Migraine	N = 31 I = 14 C = 17	High	Waitlist	Symptoms	NA	MM+EM	Individual	Education Relaxation training Cognitive restructuring	NA	Home
Davis et al. 2004	USA	Cystic Fibrosis	N = 47 I = 25 C = 22	High	Waitlist	Dealing with the chronic condition in daily life Disease Knowledge	NA	MM+RM+ EM	Individual	Education	NA	Home
Downs 2006	Australia	Cystic Fibrosis	N = 43 I = 18 C = 25	Moderate	Not clear	Adherence Disease knowledge	CSL	MM	Individual	Education Problem-solving skills Decision-making skills	Healthcare professional	Clinic
Franklin 2006	UK	Diabetes	N = 90 I1 = 32 I2 = 31 C = 27	Moderate	Another specific intervention	Symptoms Dealing with the chronic condition in daily life Adherence Disease knowledge	CSL	MM	Individual	Telemedicine Goal-setting skills	Diabetes healthcare team	Clinic
Huss 2003	USA	Asthma	N = 101 I = 56 C = 45	Moderate	Usual care	Disease knowledge	NA	MM	Individual	Education	NA	Home
Jan 2007	Taiwan	Asthma	N = 164 I = 88 C = 76	Moderate	Another specific intervention	Symptoms	NA	MM	Individual	Education Self-monitoring	NA	Clinic (out-patient)

Table 1 Study characteristics and intervention components (continued)

Background		Participants		Study characteristics		Components of self-management interventions						
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventions	Setting
Jones 2010	USA	Cancer	N = 65 I = 35 C = 30	Moderate	Another specific intervention	Quality of Life Dealing with the chronic condition in daily life Disease knowledge	NA	MM+RM	Individual	Education	Healthcare professional	Home
Joseph 2007	USA	Asthma	N = 314 I = 162 C = 152	Moderate	Another specific intervention	Quality of Life Symptoms School-attendance	Trans-theoretical/Health belief model	MM	Individual	Education	NA	School
Kashikar-Zuck 2012	USA	Juvenile Fibromyalgia	N = 114 I = 57 C = 57	High	Another specific intervention	Symptoms Psychological outcomes	CBT	MM	Individual	Cognitive restructuring Education Relaxation training Problem-solving skills	Psychologist	Clinic
Kato 2008	USA	Cancer	N = 371 I = 195 C = 176	High	Another specific intervention	Quality of Life Dealing with the chronic condition in daily life Psychological outcomes Adherence Disease knowledge	CSL	MM	Individual	Education	NA	NA
Koontz 2004	USA	Sickle cell disease	N = 24 I = 10 C = 14	Low	Another specific intervention	School-attendance Disease knowledge	NA	MM+RM	Group	Education Peer-support	Teachers	School

Table 1 Study characteristics and intervention components (continued)

Background		Participants		Study characteristics			Components of self-management interventions					
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventions	Setting
Krishna 2003	USA	Asthma	N = 228 I = 107 C = 121	Moderate	Another specific intervention	Symptoms School-attendance Disease knowledge	NA	MM	Individual	Education Decision-making skills Social skills	Multi-disciplinary team	Clinic
Kumar 2004	USA	Diabetes	N = 40 I = 19 C = 21	Moderate	Another specific intervention	Symptoms	NA	MM	Individual	Self-monitoring	NA	Online
Laffel 2003	USA	Diabetes	N = 100 I = 50 C = 50	Moderate	Usual care	Quality of Life Symptoms	NA	MM	Individual	Family therapy Education Goal-setting skills	Research assistant	Clinic
Newcombe, 2012	Australia	Chronic respiratory condition	N = 39 I = 19 C = 20	High	Waitlist	Psychological outcomes Dealing with the chronic condition in daily life	NA	MM	Individual Group (online)	Education Self-monitoring Peer-support	NA	Online
Ng, 2008	China	Asthma	N = 37 I = 20 C = 17	Moderate	Waitlist	Quality of Life Symptoms	NA	MM+RM+EM	Group	Education	NA	Clinic
Palermo 2009	USA	Chronic pain	N = 48 I = 26 C = 22	High	Waitlist	Symptoms Psychological outcomes	CBT, CSL	MM	Individual	Family therapy Education Cognitive restructuring Relaxation training	Psychologist	Online

Table 1 Study characteristics and intervention components (continued)

Background		Participants		Study characteristics			Components of self-management interventions					
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-man-agement ^b	Formats	Elements	Interven-tionits	Setting
Pulgaron 2010	USA	Asthma	N = 41 I = 20 C = 21	Moderate	Usual care	Disease knowledge Dealing with the chronic condition in daily life	NA	MM	Group	Education Problem solving skills Peer-support	Psychologist (under-graduate degree)	Camp
Rhee 2011	USA	Asthma	N = 91 I = 46 C = 45	Moderate	Another specific inter-vention	Quality of Life Symptoms	NA	MM+RM	Group	Discussion Education Tele-medicine Peer support	NA	Camp
Shames 2004	USA	Asthma	N = 119 I = 59 C = 60	Moderate	Usual care	Symptoms Disease knowledge	NA	MM	Individual	Education Tele-medicine	Case manager Clinician Nurse	Clinic Home
Staab 2006	Germany	Atopic derma-titis	N = 823 I = 446 C = 377	Moderate	Another specific inter-vention	Symptoms	NA	MM+EM	Group	Education Peer-support Relaxation training	Dermatologist Pediatrician Psychologist Dieticians	Clinic
Stinson 2010	Canada	Juvenile Idio-pathic Arthritis	N = 46 I = 22 C = 24	High	Another specific inter-vention	Quality of Life Symptoms Psychological outcomes Adherence Disease knowledge Dealing with the chronic condition in daily life	NA	MM+RM+EM	Individual	Education Telemedicine	Psychologist (under-graduate degree)	Online
Stulemeijer 2005	Nether-lands	Chronic Fatigue Syn-drome	N = 71 I = 36 C = 35	High	Waiting list	Symptoms School attendance	CBT	MM+RM+ EM	Individual	Education Cognitive restructuring	Psychologist	Clinic

Table 1 Study characteristics and intervention components (continued)

Background		Participants		Study characteristics			Components of self-management interventions					
Studies	Country	Diagnosis	N	Methodological Quality	Control	Outcome measure	Theoretical base (Yes) ^a	Domain of self-management ^b	Formats	Elements	Interventionists	Setting
Velsor-Friedrich 2005	USA	Asthma	N = 52 I = 28 C = 24	Low	Usual care	Symptoms	Orem's Self-Care Deficit Theory of Nursing	MM+ RM	Individual Group	Education Peer-support	Nurse practitioner	School
Wysocki 2007	USA	Diabetes	N = 104 I = 28 C1 = 31 C2 = 26	Moderate	Another specific intervention Usual care	Symptoms Adherence	NA	MM+ RM	Individual	Problem solving skills Family therapy Communication skills Education Cognitive restructuring	Psychologist	Clinic

^a NA= Not available; CSL = Cognitive social learning theory; CBT = cognitive behavioral theory^b MM = Medical Management; EM = Emotion Management; RM = Role Management

Many interventions were solely aimed at medical management (54.8%), which refers to health and healthcare related tasks and to tasks or topics associated with or related to a specific diagnosis. One intervention considered only role management (5.9%), referring to topics related to social participation, such as communicating and assertiveness. None of the interventions considered emotion management separately. The other interventions addressed multiple domains (39.3%). Interventions were either applied at individual level (61.3%), at group level (25.8%) or both (9.7%). Most interventions included education (96.8%), cognitive restructuring (22.3%), relaxation training (22.3%), peer support (19.4%) or self-monitoring (19.4%). In general, neither the domain of self-management considered during interventions nor formats and elements of interventions were specific for a certain theoretical base. Intervention settings were inpatient or outpatient clinics (35.5%), home (13.5%), school (13.5%), online (23.5%), home and clinic (17.6%) or camping sites (11.8%). Settings were not exclusive for formats and elements of interventions.

Effectiveness of self-management interventions

Symptoms

Twenty studies focused on the severity of symptoms as an outcome measure. The quality of evidence was low (Table 2). In general, SMI showed a trend in favor of usual care. Interventions focused on medical management, medical and role management, or medical management, emotion management and role management showed a trend in favor of usual care. The same pattern held for interventions delivered by a psychologist, or interventions provided in a clinic, online, or home and clinic. Also mono-disciplinary interventions showed a trend in favor of usual care, but this was also the case for multi-disciplinary interventions (Figure 2A-2E).

Disease knowledge

Thirteen studies focused on disease knowledge as an outcome measure. The quality of evidence was low (Table 2). In general, SMI showed a trend in favor of the intervention.

Interventions focused on medical management and interventions delivered mono-disciplinary, showed a trend in favor of the intervention condition. No clear patterns were revealed for other intervention components (Figure 3A-3B).

Adherence

Five studies focused on adherence as an outcome measure. The quality of evidence was moderate (Table 2). Overall, SMI showed a positive trend on adherence. Interventions focused on medical management showed trend in favor of intervention condition. The same pattern held for interventions provided individually, mono-disciplinary or in a clinic (Figure 4A-4D).

Table 2 Evidence profile

Outcome (No. of studies)	Quality assessment					Number of patients		Quality of the evidence
	Limitation in design	Inconsistency	Indirectness	Imprecision	Publication bias	C ^a	I ^b	
Symptoms (20)	No serious limitation	No serious inconsistency	Very serious indirectness	Serious imprecision	Unlikely	868	902	Low
Disease knowledge (13)	No serious limitation	Serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	516	554	Low
Adherence (5)	No serious limitation	No serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	242	263	Moderate
Dealing with chronic condition in daily life (9)	No serious limitation	No serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	360	391	Moderate
Psycho-logical outcomes (8)	No serious limitation	No serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	324	344	Moderate
School attendance (5)	Serious limitation	Serious inconsistency	Very serious indirectness	No serious imprecision	Unlikely	305	285	Very low
Quality of life (11)	No serious limitation	Serious inconsistency	No serious indirectness	No serious imprecision	Unlikely	576	625	Moderate

^a number of patients that participated in the control condition.^b number of patients that participated in the intervention condition

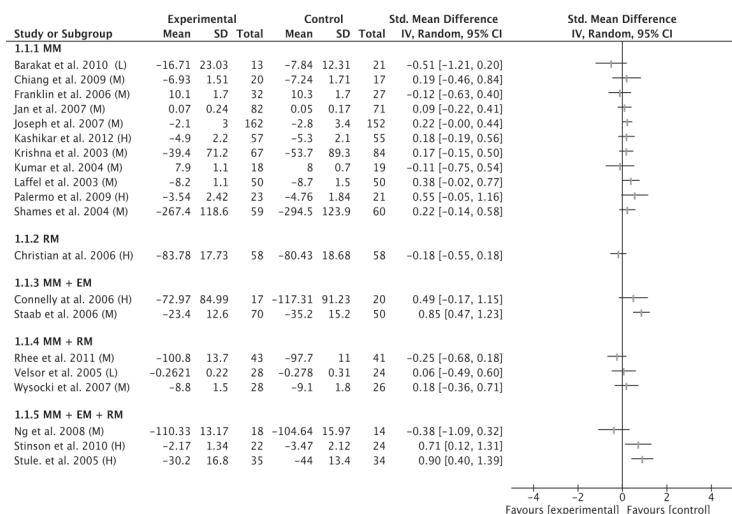


Figure 2A Effects of domain of self-management on symptoms

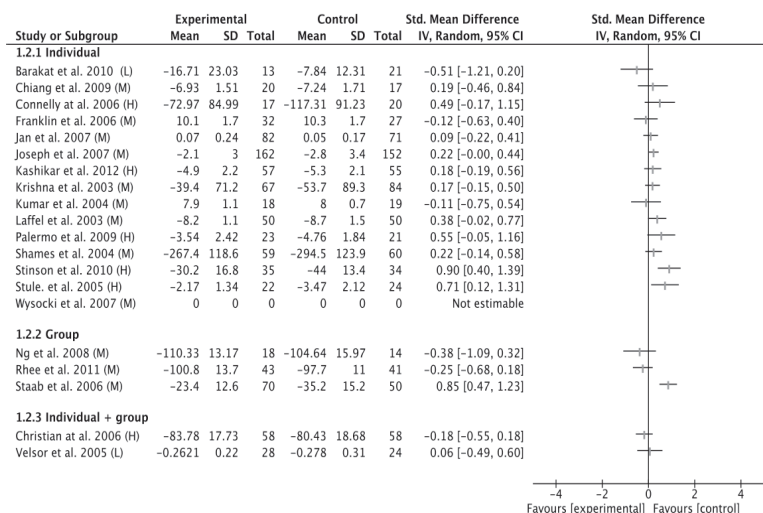


Figure 2B Effects of intervention format on symptoms

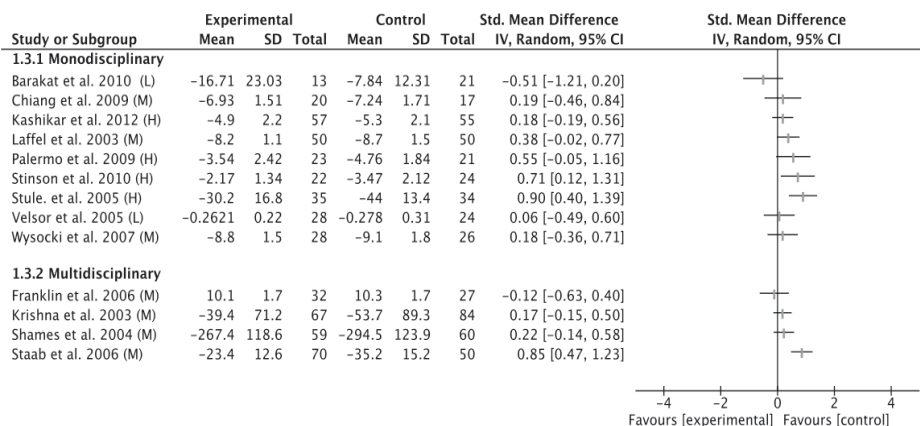


Figure 2C Effects of involved disciplines on symptoms

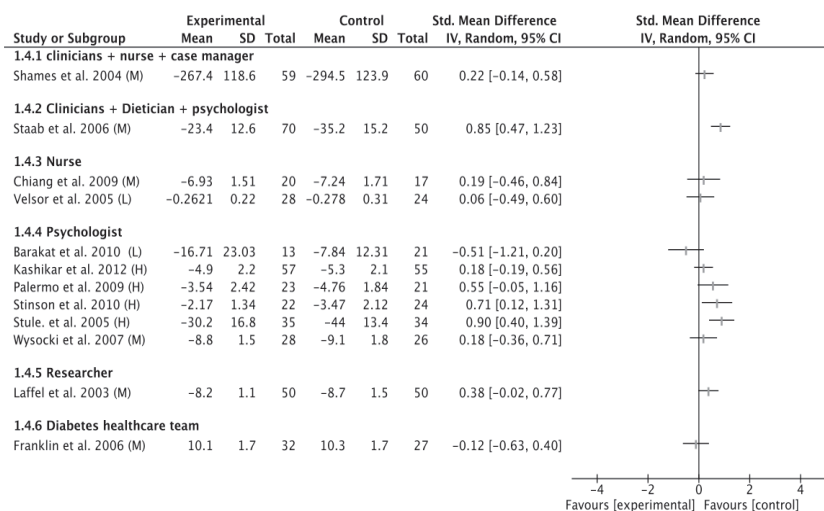


Figure 2D Effects of interventionists on symptoms

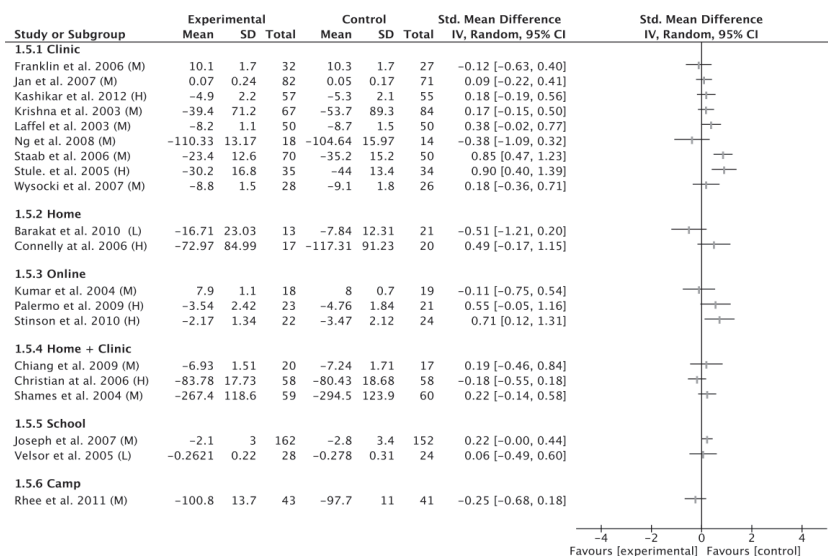


Figure 2E Effects of setting on symptoms

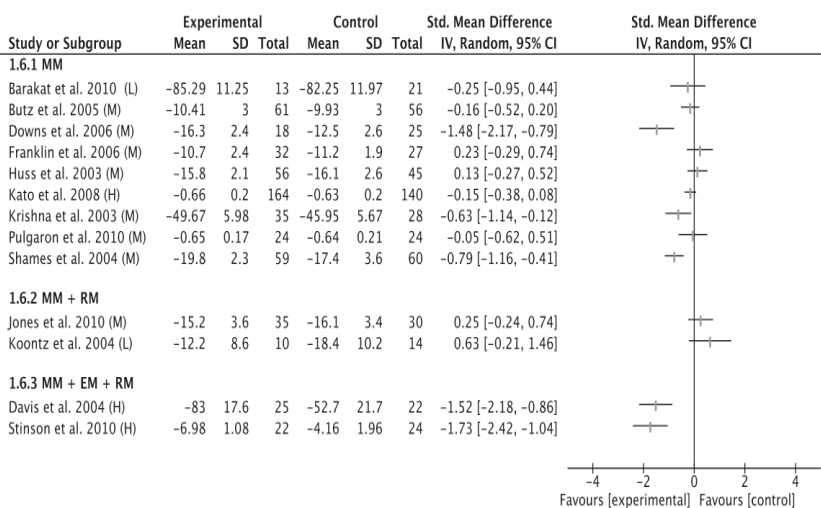
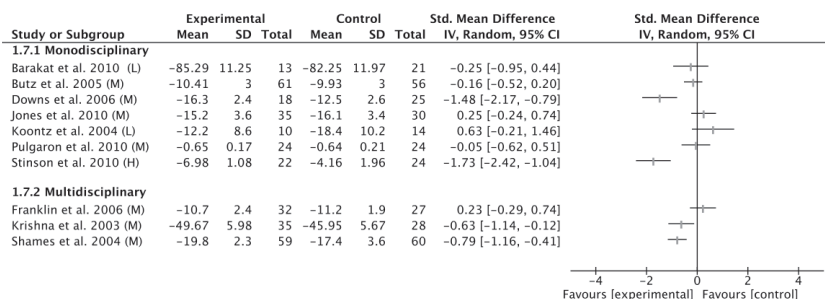
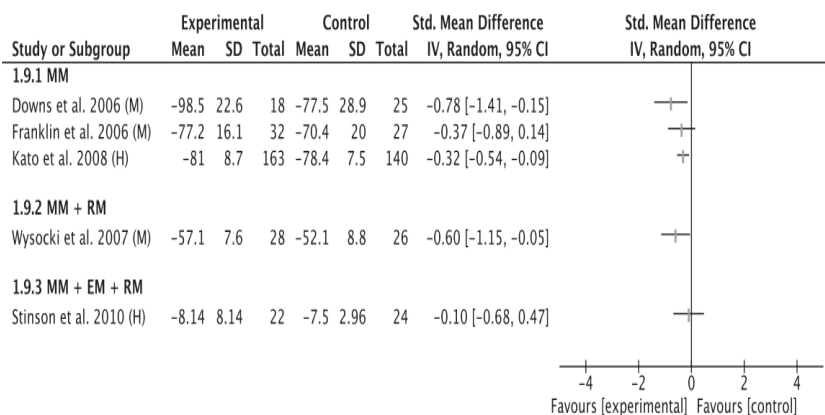
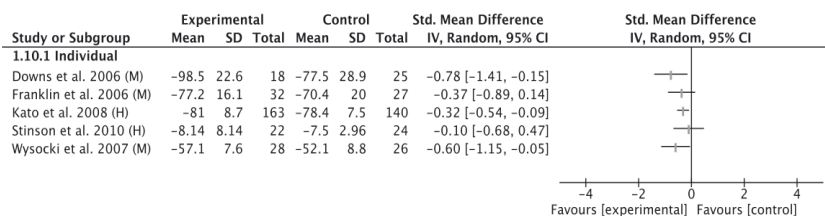


Figure 3A Effects of domain of self-management on disease knowledge

**Figure 3B** Effects of involved disciplines on disease knowledge**Figure 4A** Effects of domain of self-management on adherence**Figure 4B** Effects of intervention format on adherence

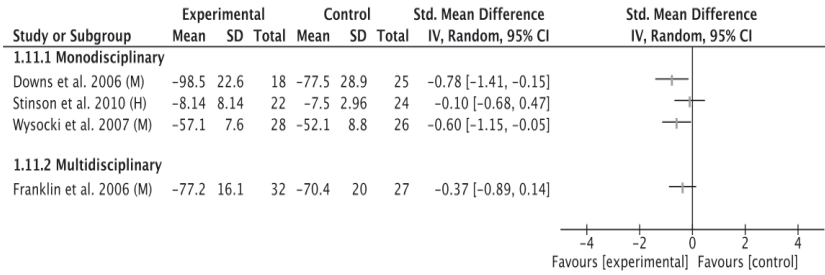


Figure 4C Effects of involved disciplines on adherence

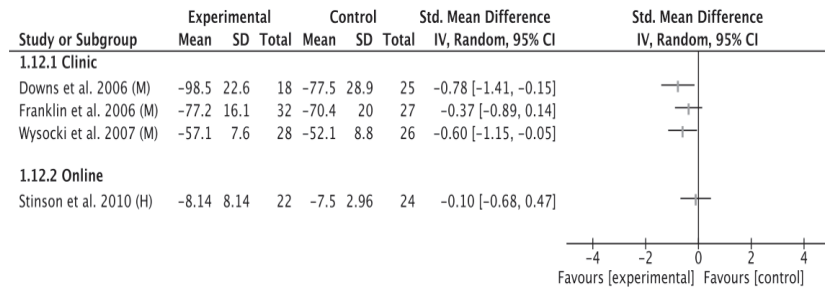


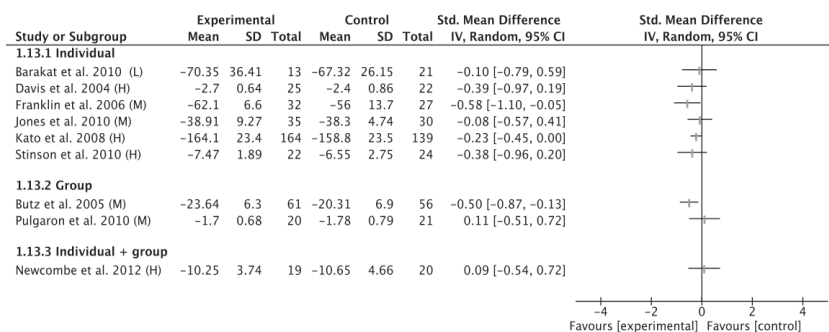
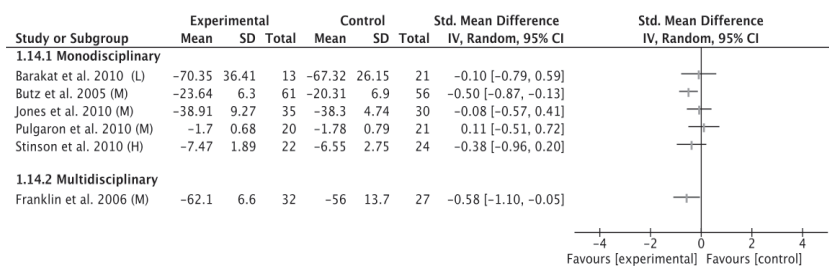
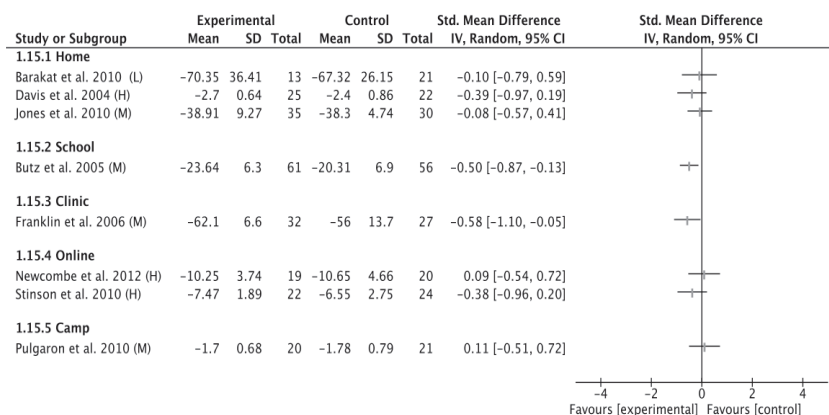
Figure 4D Effects of setting on adherence

Dealing with chronic condition in daily life

Nine studies focused on dealing with the chronic condition in daily life as an outcome measure. The quality of evidence was moderate (Table 2). In general, SMI showed a positive trend on dealing with the chronic condition in daily life. Mono-disciplinary interventions showed a trend in favor of the intervention condition. The same pattern held for interventions provided at home and for interventions provided individually (Figure 5A-5C).

Psychological outcomes

Eight studies focused on psychological outcomes. The quality of evidence was moderate (Table 2). In general, no clear effects of SMI were found on this outcome measure. Mono-disciplinary interventions and interventions provided online showed a trend in favor of usual care. No clear pattern was revealed for other intervention components (Figure 6A-6B).

**Figure 5A** Effects of format on dealing with a chronic condition in daily life**Figure 5B** Effects of involved disciplines on dealing with a chronic condition in daily life**Figure 5C** Effects of setting on dealing with a chronic condition in daily life

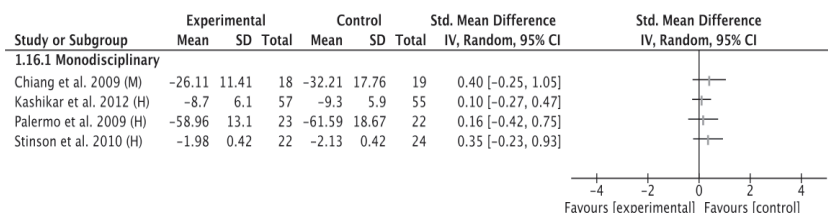


Figure 6A Effects of involved disciplines on psychological outcomes

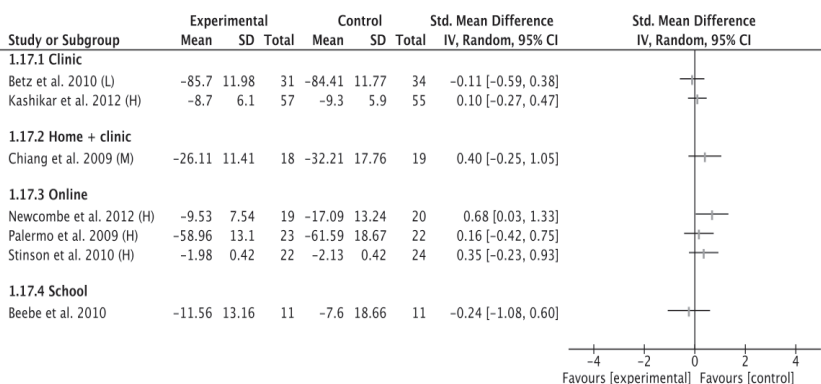


Figure 6B Effects of setting on psychological outcomes

School attendance

Five studies focused on school attendance as an outcome measure. The quality of evidence was very low (Table 2). In general, SMI showed a trend in favor of usual care on school attendance. No clear pattern was revealed for any intervention components.

Quality of life

Eleven studies focused on quality of life as an outcome measure. The quality of evidence was moderate (Table 2). Overall, no clear effects of SMI were found on quality of life. No clear pattern was revealed for any intervention components.

DISCUSSION

We reviewed randomized controlled trials to explore the effectiveness and effective intervention components of *pediatric* SMI. In contrast to existing literature, we looked at pediatric SMI in general, i.e. irrespective of type of condition. Although no strong

conclusions can be drawn, our findings suggests that pediatric SMI might be effective at influencing disease knowledge, adherence, and dealing with the chronic condition, but not symptom severity and school attendance. Conflicting evidence was found for effectiveness of SMI on psychological outcomes and quality of life. Furthermore some evidence was found for effective intervention components, but this differs per outcome measure.

Effectiveness of self-management interventions

Possible evidence for effectiveness of SMI on disease knowledge and dealing with the chronic condition was also reported in previous reviews on SMI for adult patients [10, 74, 75]. Others showed possible evidence for effectiveness of SMI on disease knowledge of youth with spina bifida, arthritis, asthma, or diabetes [17, 18, 76] and for effectiveness of pediatric SMI on adherence [77-79]. Moreover, earlier studies on youth showed that both knowledge about the condition and medication adherence are important to maintain health [80].

Although Kirk and colleagues [18] underlined the need to incorporate patient-centered outcomes such as quality of life and psychosocial well-being into SMI evaluation studies, conflicting evidence for effectiveness of SMI on these outcome measures was found and inconsistent results have been reported by others [10, 74]. Nolte et al. [81] pointed out that patient reported outcomes measures (e.g. quality of life and psychosocial well-being) are highly variable, since these require the highest degree of personal appraisal. Specifically, people may define depression or quality of life differently before SMI compared to after intervention, influencing the reliability of measurements. These biases could have influenced the results of included studies and, thus, our results. However, patient reported outcomes do provide insights into patients' lived experiences. Therefore, future studies designed to explore response shifts are recommended. For instance, qualitative methods may shed light on how a patient's context interacts with their lived experiences. This is in line with the recently posed notion that patient's context is important to consider when researching self-management [82].

Effective components of self-management interventions

In addition to previous research on effectiveness of SMI [17, 18, 21], this review revealed some suggestions about components of SMI to be effective. Specifically, SMI aimed to improve disease knowledge, could target medical management and could be provided mono-disciplinary. This is not unexpected since medical management is primary focused on topics associated with or related to a specific diagnosis. A specialist, too, should provide education, mono-disciplinary.

In addition, our review suggests that SMI aimed to improve adherence might be effective when focused on medical management and provided individually in a clinic

by a mono-disciplinary team. This is not surprising, since adherence is part of medical management and individual attention may enhance insights into personal barriers to adherence. In addition, adherence is a specialism of medical professionals.

Furthermore, our review suggests that SMI aimed to support young people to deal with the chronic condition in daily life might be mono-disciplinary programs, provided individually and at home. These findings are somewhat unexpected since peer-support was found to be an appropriate element to enhance self-efficacy [83, 84]. However, taking a closer look at elements used in these SMI, we conclude that these programs facilitate peer-support using telemedicine element. This indicates that online peer-support could also be an appropriate element to improve self-efficacy, problem solving skills and pro-active coping behavior, as was earlier mentioned in literature [18].

Our review suggests, too, that a mono-disciplinary team and an online setting might not be effective intervention components of self-management support focused on psychological outcomes, i.e. stress, anxiety and depression. Since a low self-esteem is either a correlate or risk factor for depressive symptoms [85, 86], it might be necessary to incorporate element that promote people's sense of empowerment, such as peer-support i.e. group-sessions, into SMI interventions if aiming to improve psychological outcomes [87, 88]. However, this review could not provide evidence for this hypothesis as only few interventions focused on psychological outcomes were provided in group sessions. It seems that the development of SMI is often not based on theories. Therefore, it is recommended to further standardize the development of SMI, i.e. explicit use of theory in designing interventions.

Furthermore, some indications were found that SMI focused on symptom reduction should not target medical management; medical management and role management; or medical management, role management and emotion management. In addition we found that a psychologist should not provide these interventions. This is not surprising, since symptoms reduction may also be the specialism of clinicians or nurses. Also, we found that interventions should not be provided in a clinic, online, or at home and in a clinic. An explanation for our findings could be that symptoms arise in interacting with the social context. In line with this reasoning, SMI could be more effective when only provided in people's daily life. For example, Reeves et al. [89] indicated the importance of social community to fulfill self-management tasks. However, Krieger et al. [90] reviewed pediatric asthma interventions and found home and clinical setting to be effective for symptom reduction. It seems that symptom reduction is too disease-specific and therefore could not be targeted with SMI focused on variety of chronic conditions. However, again, intervention elements are too diverse and the sample sizes were too small to provide evidence for this hypothesis. Future research should further investigate this hypothesis.

Strengths, limitations and other considerations

SMI for young people across the wide age range of 7-25 years were included, while content or themes obviously are not applicable to the whole age range, e.g. employment pertains to older adolescents only. It would be advisable, therefore, to distinguish between age groups. However, an additional analysis -not reported in this paper- showed that interventions' theoretical base, formats and elements did not differ for different age groups.

This study looked at many types of SMI across a wide range of chronic conditions. The breadth of this study is a potential strength. The findings enable researchers and healthcare professionals to look at general self-management support, which goes beyond particular chronic conditions. In addition, subgroup analyses -not presented in this paper- showed no differences in effects between interventions focused on a distinctive diagnosis.

Last, it was not possible to perform a meta-analysis on the effects of SMI, since the outcome measures and intervention programs were heterogeneous. Different conceptualizations of self-management might be one explanation for the variety in outcome measures used in SMI evaluation studies, which points at a more fundamental challenge to provide precise definitions [91]. Therefore, results about effectiveness should be interpreted with some caution. Others, too, recommended to further standardize the development and evaluation of SMI [16, 17, 74]. Intervention mapping methodology [84] is a recommended stepwise approach for theory and evidence based development and implementation of interventions that helps to fit intervention goals with program development and program evaluation. In addition, an evidence based theoretically derived intervention would make for a more effective SMI, using components which have been found to be effective in changing that specific outcome [92].

CONCLUSIONS

Although, no strong conclusions can be drawn considering SMI effectiveness, our review suggests that pediatric SMI might be effective at influencing disease knowledge, adherence and dealing with the chronic condition, across a wide range of conditions. There are indications that SMI aimed to improve *adherence* should be focused on *medical management*, and should be provided *individually in a clinical setting by a mono-disciplinary team*. Furthermore, an *individual format and home setting* combined with *online peer-support* might be effective intervention components for SMI focused on *dealing with the chronic condition* in daily life. These combinations of expected outcomes, focus and intervention elements seemed effective irrespective of diagnosis, and may therefore act as good starting points for further research into and improvement of self-management support

of young people with chronic conditions. Results underlined the need to systematically develop and evaluate SMI, since it may provide more evidence for effectiveness and effective intervention components.

ACKNOWLEDGEMENTS

The authors thank the information specialist of the Erasmus MC Medical library, drs. W. Bramer, for his assistance in defining the search strategy. The study was part of the Self-management and Participation Innovation Lab, supported by SIA-RAAK, the Foundation Innovation Alliance with funding from the Dutch ministry of Education, Culture and Science (OCW) (PRO-02-025).

REFERENCES

1. White PH: Access to health care: health insurance considerations for young adults with special health care needs/disabilities. *Pediatrics* 2002, 110(6):1328-1335.
2. World Health Organization. World report on disability. Available at <http://whqlibdoc.who.int/publications/2011> [accessed 21 January 2015].
3. Verhoof E, Maurice-Stam H, Heymans H, Grootenhuis M: Growing into disability benefits? Psycho-social course of life of young adults with a chronic somatic disease or disability. *Acta Paediatr* 2012, 101(1):19-26.
4. Ratanachadawan Y: A response to Koch T, Jenkin P, Kralik D. (2004). Chronic illness self-management: locating the 'self'. *J Adv Nurs* 2005, 52(5):571-572.
5. Donkervoort M, Roebroek M, Wiegerink D, van der Heijden-Maessen H, Stam H, Transition Research Group South West N: Determinants of functioning of adolescents and young adults with cerebral palsy. *Disab Rehab* 2007, 29(6):453-463.
6. 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.
7. Harvey PW, Petkov JN, Misan G, Fuller J, Battersby MW, Cayetano TN, Warren K, Holmes P: Self-management support and training for patients with chronic and complex conditions improves health-related behaviour and health outcomes. *Aust Health Rev* 2008, 32:330-338.
8. Wagner EH: Chronic disease management: what will it take to improve care for chronic illness? *Eff Clin Pract* 1998, 1(1):2-4.
9. Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, Hoes A, Schuurmans M: Self-management: One size does not fit all. *Patient Educ Couns* 2013, 92:134-137.
10. 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.
11. Coleman MT, Newton KS: Supporting self-management in patients with chronic illness. *Am Fam Physician* 2005, 72(8):1503-1510.
12. Bourbeau J: Disease-specific self-management programs in patients with advanced chronic obstructive pulmonary disease: a comprehensive and critical evaluation. *Dis Manag Health Outc* 2003, 11(5):311-319.
13. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26(1):1-7.
14. World Health Organization. Constitution of the World Health Organization. Available at http://www.who.int/governance/eb/who_constitution_en.pdf [accessed 21 January 2015].
15. Wagner EH, Austin BT, Davis C, Hindmarsh M, Schaefer J, Bonomi A: Improving chronic illness care: translating evidence into action. *Health Aff* 2001, 20(6):64-78.
16. Nolte E, McKee M: Caring for people with chronic conditions: an introduction. In: Nolte E, McKee M, eds. *Caring for people with chronic conditions. A health system perspective*. Berkshire: Open University Press; 2008.
17. Lindsay S, Kingsnorth S, McDougall C, Keating H: A systematic review of self-management interventions for children and youth with physical disabilities. *Disab Rehab* 2014, 36(4):276-288.
18. Kirk S, Beatty S, Callery P, Gellatly J, Milnes L, Pryjmachuk S: The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child Care Health Dev* 2012, 39(3):305-324.
19. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.

20. Wittgenstein L. *Philosophical Investigations*. Oxford: Blackwell; 1958.
21. Newman S, Steed L, Mulligan K. *Chronic physical illness: Self-management and behavioural interventions*. New York: Open University Press; 2009.
22. Jonsdottir H: Self-management programmes for people living with chronic obstructive pulmonary disease: a call for a reconceptualisation. *J Clin Nurs* 2013, 22(5-6):621-637.
23. Schulman-Green D, Jaser S, Martin F, Alonzo A, Grey M, McCorkle R, Redeker NS, Reynolds N, Whittemore R: Processes of self-management in chronic illness. *J Nurs Scholarsh* 2012, 44(2):136-144.
24. Perrin JM, MacLean WE, Gortmaker SL, Asher KN: Improving the psychological status of children with asthma: a randomized controlled trial. *J Dev Behav Pediatr* 1992, 13(4):241-247.
25. 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.
26. Sattoe JNT, Bal MI, Roelofs PDDM, Bal R, Miedema HS, van Staa AL: Self-management interventions for young people with chronic conditions: A Systematic Overview. *Patient Educ Couns* 2015, doi: 10.1016/j.pec.2015.03.004. [Epub ahead of print].
27. Grant MJ, Booth A: A typology of reviews: an analysis of 14 review types and associated methodologies. *Health Info Libr J* 2009, 26(2):91-108.
28. Moher D, Liberati A, Tetzlaff J, Altman DG: *Preferred Reporting for Systematic Reviews and Meta-Analyses: The PRISMA Statement*. PLoS Med; 2009.
29. Lu Y, Li Z, Arthur D: Mapping publication status and exploring hotspots in a research field: chronic disease self-management. *J Adv Nurs* 2014, 70(8):1837-1844.
30. Gall C, Kingsnorth S, Healy H. Growing up ready: a shared management approach. *Phys Occup Ther Pediatr* 2006, 26(4):47-62.
31. Irwin CW: Young Adults Are Worse Off Than Adolescents. *J Adol Health* 2010, 46(5):405-406.
32. Beresford B, Stuttard L: Young adults as users of adult healthcare: experiences of young adults with complex or life-limiting conditions. *Clin Med* 2014, 14(4):404-408.
33. Scottish Intercollegiate Guidelines Network. Critical appraisal: notes and checklists. Available at <http://www.sign.ac.uk/methodology/checklists.html#> [accessed 17 June 2013].
34. Davidson KW, Goldstein M, Kaplan RM, Kaufmann PG, Knatterud GL, Orleans CT, Spring B, Trudeau KJ, Whitlock EP: Evidence-Based Behavioral Medicine: What Is It and How Do We Achieve It? *Ann Behav Med* 2003, 26(3):161-171.
35. Higgins JPT, Green ST. *Cochrane Handbook for Systematic Reviews of Interventions version 5.1.0*. The cochrane Collaboration; 2011.
36. Guyatt G, Oxman AD, Akl EA, Kunz R, Vist G, Brozek J, Norris S, Falck-Ytter Y, Glasziou P, DeBeer H, Jaeschke R, Rind D, Meerpohl J, Dahm P, Schünemann HJ: GRADE guidelines: 1. Introduction- GRADE evidence profiles and summary of findings tables. *J Clin Epidemiol*. 2011;64(4):383-394.
37. Guyatt GH, Oxman AD, Kunz R, Woodcock J, Brozek J, Helfand M, Alonso-Coello P, Falck-Ytter Y, Jaeschke R, Vist G, Akl EA, Post PN, Norris S, Meerpohl J, Shukla VK, Nasser M, Schünemann HJ; GRADE Working Group: GRADE guidelines: 8. Rating the quality of evidence-indirectness. *J Clin Epidemiol* 2011, 64(12):1303-1310.
38. Guyatt GH, Oxman AD, Kunz R, Woodcock J, Brozek J, Helfand M, Alonso-Coello P, Glasziou P, Jaeschke R, Akl EA, Norris S, Vist G, Dahm P, Shukla VK, Higgins J, Falck-Ytter Y, Schünemann HJ; GRADE Working Group: GRADE guidelines: 7. Rating the quality of evidence-inconsistency. *J Clin Epidemiol* 2011, 64(12):1294-1302.
39. Guyatt GH, Oxman AD, Kunz R, Brozek J, Alonso-Coello P, Rind D, Devereaux PJ, Montori VM, Freyschuss B, Vist G, Jaeschke R, Williams JW Jr, Murad MH, Sinclair D, Falck-Ytter Y, Meerpohl J,

- Whittington C, Thorlund K, Andrews J, Schünemann HJ: GRADE guidelines 6. Rating the quality of evidence-imprecision. *J Clin Epidemiol* 2011, 64(12):1283-1293.
40. Brozek JL, Akl AE, Alonso-Coello P, Lang D, Jaeschke R, Williams JW, Phillips B, Lelgemann M, Lethaby A, Bousquet J, Guyatt GH, Schünemann HJ; GRADE Working Group: Grading quality of evidence and strength of recommendations in clinical practice guidelines. Part 1 of 3. An overview of the GRADE approach and grading quality of evidence about interventions. *Allergy* 2009, 64(5): 669-677.
 41. DerSimonian R, Laird N: Meta-analysis in clinical trials. *Control Clin Trial* 1986, 7:177-188.
 42. Barakat LP, Schwartz LA, Salamon KS, Radcliffe J: A family-based randomized controlled trial of pain intervention for adolescents with sickle cell disease. *J Pediatr Hematol Oncol* 2010, 32(7): 540-547.
 43. Beebe A, Gelfand EW, Bender B: A randomized trial to test the effectiveness of art therapy for children with asthma. *J Allergy Clin Immunol* 2010, 126:263-U14.
 44. Betz CL, Smith K, Macias K: Testing the transition preparation training program: A randomized controlled trial. *Int J Child Adolesc health* 2011, 3:595-607.
 45. Butz A, Pham L, Lewis L, Lewis C, Hill K, Walker J, Winkelstein M: Rural children with asthma: Impact of a parent and child asthma education program. *J Asthma* 2005, 42:813-821.
 46. Chiang LC, Ma WF, Huang JL, Tseng LF, Hsueh KC: Effect of relaxation-breathing training on anxiety and asthma signs/symptoms of children with moderate-to-severe asthma: A randomized controlled trial. *Int J Nurs Stud* 2009, 46:1061-1070.
 47. Christian BJ, D'Auria JP: Building life skills for children with cystic fibrosis: Effectiveness of an intervention. *Nurs Res* 2006, 55:300-307.
 48. Connelly M, Rapoff MA, Thompson N, Connelly W: Headstrong: A pilot study of a CD-ROM intervention for recurrent pediatric headache. *J Pediatr Psychol* 2006, 31:737-747.
 49. Davis MA, Quittner AL, Stack CM, Yang MCK: Controlled evaluation of the STARBRIGHT CD-ROM program for children and adolescents with cystic fibrosis. *J Pediatr Psychol* 2004, 29:259-267.
 50. Downs JA, Roberts CM, Blackmore AM, Le Souef PN, Jenkins SC: Benefits of an education programme on the self-management of aerosol and airway clearance treatments for children with cystic fibrosis. *Chronic Respir Dis* 2006, 3(1):19-27.
 51. Franklin VL, Waller A, Pagliari C, Greene SA: A randomized controlled trial of Sweet Talk, a text-messaging system to support young people with diabetes. *Diabet Med* 2006, 23(12):1332-1338.
 52. Huss K, Winkelstein M, Nanda J, Naumann PL, Sloand ED, Huss RW: Computer game for inner-city children does not improve asthma outcomes. *J Pediatr Health Care* 2003, 17(2):72-78.
 53. Jan RL, Wang JY, Huang MC, Tseng SM, Su HJ, Liu LF: An internet-based interactive telemonitoring system for improving childhood asthma outcomes in Taiwan. *Telemed J E Health* 2007, 13(3): 257-268.
 54. Jones JK, Kamani SA, Bush PJ, Hennessy KA, Marfatia A, Shad AT: Development and evaluation of an educational interactive CD-ROM for teens with cancer. *Pediatr Blood Cancer* 2010, 55(3):512-519.
 55. Joseph CL, Peterson E, Havstad S, et al. A web-based, tailored asthma management program for urban African-American high school students. *Am J Respir Crit Care Med* 2007, 175(9):888-895.
 56. Kashikar-Zuck S, Ting TV, Arnold LM, et al. Cognitive behavioral therapy for the treatment of juvenile fibromyalgia: A multisite, single-blind, randomized, controlled clinical trial. *Arthritis Rheum* 2012, 64(1):297-305.
 57. Kato PM, Cole SW, Bradlyn AS, Pollock BH: A video game improves behavioral outcomes in adolescents and young adults with cancer: a randomized trial. *Pediatrics* 2008, 122(2):305-317.

58. Koontz K, Short AD, Kalinyak K, Noll RB: A randomized, controlled pilot trial of a school intervention for children with sickle cell anemia. *J Pediatr Psychol* 2004, 29:7-17.
59. Krishna S, Francisco BD, Balas EA, Konig P, Graff GR, Madsen RW: Internet-enabled interactive multimedia asthma education program: a randomized trial. *Pediatr* 2003, 111:503-510.
60. Kumar VS, Wentzell KJ, Mikkelsen T, Pentland A, Laffel LM: The DAILY (Daily Automated Intensive Log for Youth) trial: a wireless, portable system to improve adherence and glycemic control in youth with diabetes. *Diabetes Technol Ther* 2004, 6:445-453.
61. Laffel LM, Vangsness L, Connell A, Goebel-Fabbri A, Butler D, Anderson BJ: Impact of ambulatory, family-focused teamwork intervention on glycemic control in youth with type 1 diabetes. *J Pediatr* 2003, 142(4):409-416.
62. Newcombe PA, Dunn TL, Casey LM, Sheffield JK, Petsky H, Anderson-James S, Chang AB: Breathe Easier Online: Evaluation of a randomized controlled pilot trial of an Internet-based intervention to improve well-being in children and adolescents with a chronic respiratory condition. *J Med Internet Res* 2012, 14:115-126.
63. Ng SM, Li AM, Lou VW, Tso IF, Wan PY, Chan DF: Incorporating family therapy into asthma group intervention: a randomized waitlist-controlled trial. *Fam process* 2008, 47:115-130.
64. Palermo TM, Wilson AC, Peters M, Lewandowski A, Somhegyi H: Randomized controlled trial of an Internet-delivered family cognitive-behavioral therapy intervention for children and adolescents with chronic pain. *Pain* 2009, 146(1-2):205-213.
65. Pulgaron ER, Salamon KS, Patterson CA, Barakat LP: A problem-solving intervention for children with persistent asthma: A pilot of a randomized trial at a pediatric summer camp. *J Asthma* 2010, 47:1031-1039.
66. Rami B, Popow C, Horn W, Waldhoer T, Schober E: Telemedical support to improve glycemic control in adolescents with type 1 diabetes mellitus. *Eur J Pediatr* 2006, 165(10):701-705.
67. Rhee H, Belyea MJ, Hunt JF, Brasch J: Effects of a peer-led asthma self-management program for adolescents. *Arch Pediatr Adolesc Med* 2011, 165(6):513-519.
68. Shames RS, Sharek P, Mayer M, Robinson TN, Hoyte EG, Gonzalez-Hensley F, Bergman DA, Umetsu DT: Effectiveness of a multicomponent self-management program in at-risk, school-aged children with asthma. *Ann Allergy Asthma Immunol* 2004, 92:611-618.
69. Staab D, Diepgen TL, Fartasch M, Kupfer J, Lob-Corzilius T, Ring J, Scheewe S, Scheidt R, Schmid-Ott G, Schnopp C, Szczepanski R, Werfel T, Wittenmeier M, Wahn U, Gieler U: Age related, structured educational programmes for the management of atopic dermatitis in children and adolescents: multicentre, randomised controlled trial. *BMJ* 2006, 332:933-8.
70. Stinson JN, McGrath PJ, Hodnett ED, Feldman BM, Duffy CM, Huber AM, Tucker LB, Hetherington CR, Tse SM, Spiegel LR, Campillo S, Gill NK, White ME: An internet-based self-management program with telephone support for adolescents with arthritis: a pilot randomized controlled trial. *J Rheumatol* 2010, 37:1944-1952.
71. Stulemeijer M, De Jong LWAM, Fiselier TJW, Hoogveld SWB, Bleijenberg G: Cognitive behaviour therapy for adolescents with chronic fatigue syndrome: Randomised controlled trial. *Br Med J* 2005, 330(7481):14-17.
72. Velsor-Friedrich B, Pigott T, Srof B: A practitioner-based asthma intervention program with African American inner-city school children. *J Pediatr Health Care* 2005, 19(3):163-171.
73. Wysocki T, Harris MA, Buckloh LM, Mertlich D, Lochrie AS, Maura N, White NH: Randomized trial of behavioral family systems therapy for diabetes: maintenance of effects on diabetes outcomes in adolescents. *Diabetes Care* 2007, 30:555-560.

74. Nolte S, Osborne RH: A systematic review of outcomes of chronic disease self-management interventions. *Qual Life Res* 2013, 22(7):1805-1816.
75. Franek J: Self-Management Support Interventions for Persons With Chronic Disease: An Evidence-Based Analysis. *Ont Health Technol Assess Ser* 2013, 13(9):1-60.
76. DeShazo J, Harris L, Pratt W: Effective intervention or child's play? A review of video games for diabetes education. *Diabetes Technol Ther* 2010, 12(10):815-822.
77. Dean AJ, Walters J, Hall A: A systematic review of interventions to enhance medication adherence in children and adolescents with chronic illness. *Archives of disease in childhood* 2010, 95(9):717-723.
78. Graves MM, Roberts MC, Rapoff M, Boyer A: The efficacy of adherence interventions for chronically ill children: a meta-analytic review. *J Pediatr Psychol* 2010, 35(4):368-382.
79. Kahana S, Drotar D, Frazier T: Meta-analysis of psychological interventions to promote adherence to treatment in pediatric chronic health conditions. *J Pediatr Psychol* 2008, 33(6):590-611.
80. Kingsnorth S, Healy H, Macarthur C: Preparing for adulthood: a systematic review of life skill programs for youth with physical disabilities. *J Adol Health* 2007, 41(4):323-332.
81. Nolte S, Elsworth GR, Newman S, Osborne RH: Measurement issues in the evaluation of chronic disease self-management programs. *Qual Life Res* 2013, 22(7):1655-1664.
82. Thirsk ML, Clark AM: What is the 'self' in chronic disease self-management? *Int J Nurs Stud* 2014, 51(5):691-693.
83. Bandura A: *Social Learning Theory*. New York: General Learning Press; 1977.
84. Bartholomew LK, Parcel GS, Kok G, Gottlieb NH, Fernandez ME: *Planning Health Promotion Programs: An Intervention Mapping Approach*. 3rd edition. San-Francisco: Jossey-Bass; 2011.
85. Roberts JE, Kassel JD, Gotlib IH: Level and stability of self-esteem as predictors of depressive symptoms. *Person Indiv Diff* 1995, 19(2):217-224.
86. Roberts JE, Gamble SA: Current mood-state and past depression as predictors of self-esteem and dysfunctional attitudes among adolescents. *Person Indiv Diff* 2001, 30:1023-1037.
87. Dishman RK, Hales DP, Pfeiffer KA, Felton GA, Saunders R, Ward DS, Dowda M, Pate RR: Physical self-concept and self-esteem mediate cross-sectional relations of physical activity and sport participation with depression symptoms among adolescent girls. *Health Psychol* 2006, 25(3):396-407.
88. Rogers ES, Chamberlin J, Ellison ML, Crean T: A consumer-constructed scale to measure empowerment among users of mental health services. *Psychiatr Serv* 1997, 48(8):1042-1047.
89. Reeves D, Blickem C, Vassilev I, Brooks H, Kennedy A, Richardson G, Rogers A: The contribution of social networks to the health and self-management of patients with long-term conditions: a longitudinal study. *PloS one* 2014, 9(6):1-12.
90. Krieger J, Takaro TK, Song L, Beaudet N, Edwards K: A Randomized Controlled Trial of Asthma Self-management Support Comparing Clinic-Based Nurses and In-Home Community Health Workers. The Seattle-King County Healthy Homes II Project. *Arch Pediatr Adolesc Med* 2009, 163(2):141-149.
91. Rijken M, Jones M, Heijmans M, Dixon A: Supporting self-management. In: Nolte E, McKee M, eds. *Caring for people with chronic conditions. A health system perspective*. Berkshire: Open University Press; 2008.
92. Michie S, Prestwich A: Are interventions theory-based? Developory coding scheme. *Health Psychol* 2010, 29(1):1-8.

Appendix C8.1 Components of self-management interventions

Domain of self-management				Format				Interventionists										Setting														
Outcome measure (quality of evidence)	Number of studies	MM	EM	RM	MM + RM	MM + EM	MM+EM+RM	Individual	Group	Combined	Multidisciplinary	Not mentioned	Clinicians	Nurse (practitioner)	Asthma educator	Dietician	Psychologist	Peers	Researcher	Teacher	Healthcare professional	Clinician + Nurse + case manager	Clinician + dietician+ pediatrician + psychologist	Diabetes healthcare team	Not mentioned	Online	Home	Clinic	Home & clinic	School	Camp	Not mentioned
		20																														
Symptoms (low)																																
Number of HQ & MQ studies with significant effect	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
Number of HQ & MQ studies with positive trend	5	2	0	1	1	0	1	2	2	1	1	4	0	0	0	0	0	0	0	0	0	0	0	0	5	1	0	2	1	0	1	0
Number of HQ & MQ studies with lack of effect	13	8	0	0	1	2	2	12	1	0	3	3	0	1	0	0	5	0	1	0	0	1	1	1	3	2	1	7	2	1	0	0
Number of LQ studies with positive trend	1	1	0	0	0	0	0	1	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0
Number of LQ studies with lack of effect	1	0	0	0	1	0	0	0	0	1	0	0	0	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	0	0
Disease knowledge (low)																																
Number of HQ & MQ studies with significant effect	5	3	0	0	0	2	5	0	0	2	1	0	0	0	0	0	1	0	0	0	1	1	0	1	1	1	1	2	1	0	0	0

Appendix C8.1 Components of self-management interventions (continued)

Domain of self-management			Format			Interventionists										Setting																		
Outcome measure (quality of evidence)	Number of studies	9	MM	EM	RM	MM + RM	MM + EM	MM+EM+RM	Individual	Group	Combined	Multidisciplinary	Not mentioned	Clinicians	Nurse (practitioner)	Asthma educator	Dietician	Psychologist	Peers	Researcher	Teacher	Healthcare professional	Clinician + Nurse + case manager	Clinician + dietician+ pediatrician + psychologist	Diabetes healthcare team	Not mentioned	Online	Home	Clinic	Home & clinic	School	Camp	Not mentioned	
			2	2	0	0	0	0	0	1	1	0	0	1	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0	0	1	0	1	0
Dealing with the chronic condition in daily life (moderate)																																		
Number of HQ & MQ studies with significant effect			4	1	0	0	1	0	2	4	0	0	0	2	0	0	0	0	1	0	0	0	1	0	0	0	2	1	2	0	0	0	0	1
Number of HQ & MQ studies with positive trend			2	2	0	0	0	0	0	1	1	0	1	0	0	0	0	0	1	0	0	0	0	0	0	0	1	1	0	0	0	0	1	0
Number of HQ & MQ studies with lack of effect																																		
Number of LQ studies with positive trend			1	1	0	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0
Psychological outcomes (moderate)			8																															
Number of HQ & MQ studies with significant effect			1	1	0	0	0	0	1	0	0	0	1	0	0	0	0	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	1	

Appendix C8.1 Components of self-management interventions (continued)

Domain of self-management				Format			Interventionists										Setting				
</																					

Appendix C8.1 Components of self-management interventions (continued)

Domain of self-management				Format			Interventionists										Setting																
Outcome measure (quality of evidence)	Number of studies	MM	EM	RM	MM + RM	MM + EM	MM+EM+RM	Individual	Group	Combined	Multidisciplinary	Not mentioned	Clinicians	Nurse (practitioner)	Asthma educator	Dietician	Psychologist	Peers	Researcher	Teacher	Healthcare professional	Clinician + Nurse + case manager	Clinician + dietician+ pediatrician + psychologist	Diabetes healthcare team	Not mentioned	Online	Home	Clinic	Home & clinic	School	Camp	Not mentioned	
		2	2	0	0	0	0	0	2	0	0	1	1	0	0	0	0	0	0	0	0	0	0	0	2	0	0	0	1	0	1	0	0
Number of HQ & MQ studies with lack of effect																																	
Number of LQ studies with positive trend	1	1	0	0	0	0	0	1	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0
Number of LQ studies with lack of effect	1	0	0	0	1	0	0	0	0	1	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	0	0	0	0	0	1	0	0
Quality of life (moderate)	11																																
Number of HQ & MQ studies with significant effect	3	1	0	0	1	0	1	0	3	0	0	2	0	0	1	0	0	0	0	0	0	0	0	0	2	0	0	0	1	0	1	1	0
Number of HQ & MQ studies with positive trend	3	3	0	0	0	0	0	3	0	0	0	2	0	1	0	0	0	0	0	0	0	0	0	2	0	0	0	0	1	1	0	1	0
Number of HQ & MQ studies with lack of effect	4	1	0	1	1	0	1	3	0	1	0	1	0	0	0	0	1	0	1	0	1	0	0	1	1	1	1	0	1	1	0	0	0
Number of LQ studies with positive trend	1	0	0	0	1	0	0	1	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	1	0	0	0	0	0	1	0	0	0

9

Effective peer-to-peer support for young people with end-stage renal disease: A mixed methods evaluation of Camp COOL



Jane N.T. Sattoe, Susan Jedeloo, AnneLoes van Staa

BMC Nephrology. 2013; 14:279

ABSTRACT

Background

The Camp COOL programme aims to help young Dutch people with end-stage renal disease (ESRD) develop self-management skills. Fellow patients already treated in adult care (hereafter referred to as 'buddies') organise the day-to-day program, run the camp, counsel the attendees, and also participate in the activities. The attendees are young people who still have to transfer to adult care. This study aimed to explore the effects of this specific form of peer-to-peer support on self-management of young people (16-25 years) with ESRD who participated in Camp COOL (CC) (hereafter referred to as 'participants').

Methods

A mixed methods research design was employed. Semi-structured interviews (n=19) with initiators/staff, participants, and healthcare professionals were conducted. These were combined with retrospective and pre-post surveys among participants (n=62), and observations during two camp weeks.

Results

Self-reported effects of participants were: increased self-confidence, more disease-related knowledge, feeling capable of being more responsible and open towards others, and daring to stand up for yourself. According to participants, being a buddy or having one positively affected them. Self-efficacy of attendees and independence of buddies increased, while attendees' sense of social inclusion decreased (measured as domains of health-related quality of life). The buddy role was a pro-active combination of being supervisor, advisor, and leader.

Conclusions

Camp COOL allowed young people to support each other in adjusting to everyday life with ESRD. Participating in the camp positively influenced self-management in this group. Peer-to-peer support through buddies was much appreciated. Support from young adults was not only beneficial for adolescent attendees, but also for young adult buddies. Paediatric nephrologists are encouraged to refer patients to CC and to facilitate such initiatives. Together with nephrologists in adult care, they could take on a role in selecting buddies.

BACKGROUND

Young people with end-stage renal disease (ESRD) often achieve fewer developmental milestones and lag behind in development compared to both healthy peers and peers with other chronic conditions [1]. In general, the transition into adulthood is especially challenging for adolescents with chronic conditions, because they have to balance the usual developmental tasks with the medical challenges presented by the chronic condition [2]. Also, negative family exchanges like overprotection may hamper autonomy and self-advocacy development [3, 4]. Young people with ESRD are known to be a vulnerable and unique group [5]. They are at risk for cognitive impairments, low educational attainment, and psychosocial and psychiatric problems [6-12]. Psychosocial development is closely linked to health-related quality of life and social participation [13]. Young adults who reached fewer developmental milestones in childhood and adolescence therefore experienced greater impact of their condition on their daily lives [13], while sound psychosocial development in early life was associated with successful social participation (e.g. [14]).

Since adolescence involves a shift from parental influences to peer relationships [15], and peers can provide psychosocial support [16, 17] and influence treatment-related behaviors [18], creating opportunities for young people with chronic conditions to support each other is gaining popularity [19]. One popular method is the organization of recreation camps. There is some evidence that participation in recreation camps has psychosocial benefits for children with chronic conditions. Various studies reported increased health-related quality of life [20-24], improved self-esteem, self-confidence, self-image or self-efficacy and sense of mastery [22, 25-28], positive attitudes towards illness [29, 30], increased disease-specific knowledge [26, 31-33], and fostered independence, responsibility or self-management skills [26, 33, 34]. Yet, most studies have samples with an age range of 10 - 16 years on average [35], and further exploration of the benefits of participating in recreation camps for an older age group is needed [36, 37]. Furthermore, relatively little is known about these camps' working mechanisms [36, 37], and there is a lack of qualitative or mixed-methods studies into participant experiences and the effects of recreational camping for young people with chronic conditions [35].

In the Netherlands, young people with ESRD can attend a yearly, nationwide one-week camp (Camp COOL) since 2007. Funded by the Dutch Kidney Foundation and private sponsors, the camp is free of charge for the participants. Pediatric healthcare professionals throughout the country refer patients to the camp. A unique feature is that fellow patients already treated in adult care (hereafter called 'buddies') organize the day-to-day program, run the camp and counsel the attendees, next to actively participating in the activities. Attendees are young people who still have to transfer to adult care. Only one other study reports on a more active role of adolescents with rheumatic disorders in

organizing and designing a camping program, but this more active role was not evaluated [28]. Our study aimed to explore the effects of this specific peer-to-peer support on self-management of all young people (16-25 years) with ESRD who participated in Camp COOL (CC) (hereafter called ‘participants’).

METHODS

Study design & Ethics

Epstein and colleagues [20] advocated the use of Mixed Methods Research (MMR) [38] to evaluate the effects of therapeutic camping for chronically ill, because the use of complementary quantitative and qualitative designs could lead to more enriched findings [20]. We used this method not only for this reason, but also because MMR was expected to contribute to the comprehensiveness and validity of the study [31, 39]. The guidelines for Good Reporting of A Mixed Methods Study (GRAMMS) were followed [40], see Table 1. Quantitative measures such as questionnaires were combined with semi-structured interviews and participant observations during the camp weeks. Furthermore, different perspectives were explored by including healthcare professionals, buddies, attendees, and the initiators/staff of CC in the study sample. The qualitative component of our study adheres to the qualitative research review guidelines (RATS) [41].

Table 1 Guidelines for Reporting of A Mixed Methods Study (GRAMMS)*

Criteria description	
1.	Describe the design in terms of the purpose, priority and sequence of methods
2.	Describe the justification for using a mixed methods approach to the research question
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

*From: O’Cathain et al. 2008 [37]

More specifically, included in the study sample were: 1) all young people with ESRD that had once participated in CC during 2007-2010 (n=52) or were visiting the camp in 2011 and/or 2012 (n=38); 2) all pediatric nephrology professionals in the country that referred to CC (n=5); and 3) the initiators/staff of CC (n=4). The staff consisted of adults that stayed at the camp to assist the buddies in case they encountered problems they could not solve themselves. They kept themselves at the background and let the buddies run the camp.

The study was conducted in two consecutive phases, presented in Table 2. Participants were assured of confidentiality and data were processed anonymously. They received written information about the study and participants aged 12 years or older gave informed consent. Parents also provided informed consent for minors (<18 years). There were separate parts on the consent form for each of the study components (i.e. questionnaires, interviews and observations). The Medical Ethics Committee of the Erasmus MC University Medical Center approved all study procedures.

Table 2 Mixed Methods research Camp COOL

<i>Study sample:</i>	Young people that participated in Camp COOL	Initiators or staff of Camp COOL	Nephrology professionals that refer patients to Camp COOL
<i>Study phases:</i>			
Phase 1: Gaining insight into Camp COOL	<ul style="list-style-type: none"> ▪ February 2011 Semi-structured interviews (n=2) ▪ March-June 2011 Retrospective questionnaire (n=24, response: 46%) 	<ul style="list-style-type: none"> ▪ January 2011 Semi-structured interviews (n=2) 	<ul style="list-style-type: none"> ▪ January 2011 Semi-structured interviews (n=3)
Phase 2: Evaluation of Camp COOL in 2011 and 2012	<ul style="list-style-type: none"> ▪ September 2011, and October 2012 Participant observations during camp ▪ September 2011, and October 2012 Pre-post questionnaires (n=36, response: 95%) ▪ December 2011/2012, and January 2012/2013 Semi-structured interviews after camp (n=10) 	<ul style="list-style-type: none"> ▪ December 2012 Semi-structured interviews (n=2) 	

Phase 1: Gaining insight into Camp COOL

The aims of phase 1 were:

- 1) To gain insight into the underlying principles of CC as an intervention for young people with ESRD, and the context in which it takes place. These insights were also used to develop our study materials for the evaluation of CC.
- 2) To pre-test our questionnaire and to gather preliminary information about the effects CC may have on participants.

Semi-structured interviews

First, semi-structured interviews were held with the original initiators of CC (n=2), with nephrology professionals referring patients to CC (n=3), and with a buddy (n=1) and an attendee (n=1) who had participated in the previous camps (2007-2010). All original

initiators and healthcare professionals were invited to participate, and were approached through e-mail. Initiators recruited former participants in this phase of the study.

For all interviews, topic guides were used. Professionals reflected on what they knew about CC, their rationale for referring patients to CC, the criteria used for selecting patients for CC, and their expectations considering the camp's impact on both buddies and attendees. The initiators explained their aims for organizing CC, the concepts and ideas integrated in the program, and what they considered to be the camp's impact on buddies and attendees. Former participants reflected on their experiences during CC and on the benefits.

Questionnaire

Information from the semi-structured interviews with the initiators and healthcare professionals served as a basis for the retrospective questionnaire. A pilot version was tested in the interviews with the buddy and the attendee. Subsequently, all former participants (n=52) were contacted by the initiators who sent out information letters and questionnaires by mail. Participants received three reminders: by mail (four weeks after initial invitation), by e-mail (two weeks after first reminder), and by phone (two weeks after the second reminder). Respondents were entered in a lottery to win one out of four vouchers worth €25. The questionnaire contained questions on participants' background, self-management and participation and Camp COOL. The measured socio-demographic and disease-related characteristics [42], and the instruments used to measure general and disease-related self-efficacy [43-44], Health-related Quality of Life [45], and social participation [46], including their psychometrics are presented in Table 3. The questions specifically developed for this study and considering the influence of Camp COOL on the participants are presented in Appendix C9.1.

Phase 2: Evaluation of Camp COOL in 2011 and 2012

The aims of phase 2 were:

- 1) To gain insight into the effect of peer-to-peer support as working mechanism of CC.
- 2) To study the effects participating in CC has on self-management of young people with ESRD.

Prior to the camp, participants of the camp in 2011 and 2012 received a letter informing them about the research and asking for their consent, and in case of minors for parental consent as well. They filled out an informed consent form, agreeing to all research methods.

Table 3 Content and psychometrics of the measurement instruments (questionnaire)

	Measured characteristics or constructs	Measurement instrument	Answer categories or scales	α^1
Socio-demographics	Age			
	Gender		Male / Female	
	Educational level		Low / High	
Disease-related characteristics	Age at diagnosis		0 years / 1-5 years / 6-12 years / 13-16 years	
	Treatment type		Pre-dialysis / Haemodialysis / Peritoneal dialysis / Kidney transplantation / Other	
	Limitations in mobility	Medical outcomes Study (MOS) 6-Items Short Form Health Survey [42]	3-point scale: 1=severely limited / 2=somewhat limited / 3=not limited at all	.78
Self-management and social participation	General self-efficacy	10-item General Self-Efficacy Scale [43]	4-point Likert scale: 1=not right / 2=hardly right / 3=somewhat right / 4=totally right	.71
	Disease-related self-efficacy	16-item ² On Your Own Feet Self-Efficacy Scale (OYOF-SES) [44]	4-point Likert scale: 1=yes certainly / 2=yes probably / 3=no probably not / 4=no, definitely not	.90
	Health-Related Quality of Life	37-item European DISABKIDS condition generic questionnaire (DCGM-37) [45] with six domains: independence (I), social inclusion (SI), social exclusion (SE), emotion (E), physical (P), medication (M); and a general score (range: 0-100)	5-point Likert scale: 1=often / 2=quite often / 3=sometimes / 4=almost never / 5=never	I: .86 SI: .70 SE: .85 E: .81 P: .46 M: .79
	Social participation	Rotterdam Transition Profile (RTP) [46] with seven life areas: school/work, finances, (independent) living, (intimate) relationships, leisure, and mobility	Four transition (to adulthood) phases (0-3) ³	na ⁴
Camp COOL	Influence of living with the condition	10 items Effects of CC Scale See Appendix C8.1.	5-point Likert scale: 1=completely disagree / 2=disagree / 3=somewhat agree / 4=agree / 5=completely agree	.92
	Value of peer-to-peer (i.e. buddy-to-attende) support	Value of peer-to-peer support (2 items for buddies and 2 items for attendees) See Appendix C8.1.	5-point Likert scale: 1=completely disagree / 2=disagree / 3=somewhat agree / 4=agree / 5=completely agree	
	Overall liking score for CC		10-point Visual Analogue Scale: 1=lowest possible liking / 10=highest possible liking	

¹ α = Cronbach's Alpha²This instrument originally consists of 17 items assigned to knowledge, coping and skills for hospital consultations. However, one item about expecting to be ready for the transfer to adult care was deleted, because it did not apply to our full sample.³Young persons in phases 0 and 1 are still fully dependent on adults, e.g. parents, or display typical child behaviour. Young persons in phase 2 experiment with adult behaviour or orient to it. Phase 3 refers to full autonomy in participation. Because we were interested in successful transition to adulthood, the phases were dichotomised as follows: 0 = phases 0-2, 1 = phase 3.⁴Construct validity was established in a previous study [45].

Observations

Participant observations were conducted to gain insight into the establishment of peer-to-peer support during CC. Participants received information before the camp and provided consent. Two researchers (JS & SJ) and four trained nursing and physical therapy students observed participants during CC 2011 and CC 2012 and were introduced during the first activity of CC. They took field notes and filled out structured forms about participants' attitudes and behavior, and topics discussed. Special attention was paid to buddy-attendee interaction. Other broad themes on the forms were: general description of the event (e.g. description of the setting and format), topics addressed during the event, interaction between participants, and other notable happenings. Observers wrote down their findings per theme in narratives. Some activities required the group to be split into smaller groups. Therefore, to be able to observe the same activity in different groups, three to four observers were present at CC 2011 and CC 2012. At least one of the researchers teamed up with the trained students during observations, and the observers were present at every activity or event.

Semi-structured interviews

Two staff persons were interviewed at the campsite in 2011 and 2012. They talked about the daily program of CC and about the perceived impact of CC on buddies and attendees. They were selected because they were the only staff persons not interviewed during phase 1. All participants had been requested to indicate their willingness to participate in semi-structured interviews performed 4-12 weeks after the camp. Ten participants who attended CC 2011 or CC 2012 (31.3%) were willing to participate and were subsequently interviewed. They reflected on their experiences, the different elements of the program, the buddy-to-attendee support, and the benefits of participating in CC.

Pre-post questionnaires

All participants of the camps organized in 2011 or 2012 ($n=38$), filled out pre-post questionnaires containing questions similar to the ones in the retrospective questionnaire. In the pre-questionnaire, administered at camp start (T_0), the questions considering the camp experiences had been rephrased to reflect expectations. The post-questionnaire, administered at camp closure (T_1), asked after outcomes of these expectations.

Data analyses

Interviews were all digitally recorded and transcribed ad verbatim. The interview transcripts and the observation forms were imported into separate files in the qualitative software package Atlas.ti 6.2. (www.atlasti.com). Thematic analysis was applied on both data sets, and data from different parties (buddies versus attendees, and participants versus initiators/staff) were constantly compared. In Atlas.ti, initial codes (themes)

were formulated on the basis of the interview guides and the observation form. These were complemented with newly formed codes. Broad themes were derived from the interview guide, while subthemes were empirically derived from the data. Themes for instance considered 'going to CC, 'at the camp', 'peer-support' and 'CC and transition to adulthood/adult care'. Subthemes were for example 'reasons to participate', 'value of participating', 'program elements', 'buddies', and 'becoming independent'.

SPSS 20.0 (SPSS Inc., Chicago, IL) was used for all the statistical analyses. Means, standard deviations and proportions were used for descriptive analyses. Non-parametric tests were used for pre-post analyses. Finally, effect sizes were estimated for significant differences (Cohen's *d*).

Validation & Integration

Method triangulation and peer-review enhanced validation for the qualitative findings. Two researchers (JS & SJ) discussed all preliminary analyses of the observations and interviews; the final analyses were presented to and discussed with the supervisor (AvS) and the members of the advisory board. Validation for the quantitative findings was enhanced through pre-testing the questionnaire with one buddy and one attendee. None of the respondents had difficulties in answering the questions, but they had some useful suggestions considering the formulation of questions. Filling out the questionnaire took approximately 20 minutes.

Findings from the MMR were integrated in different ways. First, the qualitative findings from Phase 1 were summarized and used to develop the questionnaires. Also, statistical comparison of first phase quantitative results with the second phase quantitative results led to integration. Final integration was achieved through comparing the qualitative and quantitative findings of both phases, and drafting this manuscript.

RESULTS

First, we present the final study samples. Then, the origins and goals of CC are presented to enhance understanding of CC as intervention for young people with ESRD. This section is based on the results from the interviews with initiators and healthcare professionals. Next, the results from the observations, interviews with all three parties, and questionnaires are presented. The findings are integrated in the last paragraph.

Study samples

In the two phases, 19 respondents were interviewed: 4 initiators/staff, 3 healthcare professionals, 6 buddies, and 6 attendees (Table 4). Buddies were on average 21 years old (range: 18-25 years), while for attendees this was 17 years (range: 16-18 years).

Table 4 Characteristics of interviewed respondents

Respondent code	Type of Respondent	Gender	Attendance at Camp COOL
A	Initiator (Parent)	Female	yes
B	Initiator (Paediatric nephrologist)	Male	yes
C	Paediatric nephrologist	Female	no
D	Social worker	Female	no
E	Social worker	Male	yes
F	Buddy	Female	4 x buddy
G	Attendee	Female	2 x attendant
H	Buddy in 2011	Female	1 x buddy, 1 x attendant
I	Buddy in 2011	Female	2 x attendant
J	Buddy in 2011	Male	2 x buddy
K	Attendee in 2011	Male	first time
L	Attendee in 2011	Male	first time
M	Attendee in 2011	Female	first time
N	Buddy in 2012	Female	3 x buddy, 2 x attendant
O	Buddy in 2012	Female	1 x attendant
P	Attendee in 2012	Female	first time
Q	Attendee in 2012	Male	first time
R	Staff	Male	yes
S	Staff	Female	yes

In Phase 1, 24 out of 52 former participants (46%) filled out the retrospective questionnaire. Most of them were girls, and had received kidney transplantation. Mean age of the respondents was 20.8 (± 3.2) years, and half of them had been attendees only; while the other half had been both attendees and buddies. Background and self-management characteristics are summarized in Table 5.

In Phase 2, 38 participants of CC in 2011 and/or 2012 were asked to fill out pre and post questionnaires. Four attended both camps and filled out the questionnaires twice. Only the data from 2011 were used for the analysis, because this was their first experience with CC. Two respondents did not fill out the post questionnaire, because they had left to undergo treatment. Consequently, the pre-post sample consisted of 32 (84%) young persons with ESRD. Most of them were boys, and had had kidney transplantation. Mean age was 19.1 (± 2.4) years. Background and self-management characteristics are summarized in Table 5. Participants were observed during CC 2011 and 2012; in total on 8 out of 10 days. The program elements observed are presented in Table 6.

Table 5 Characteristics of questionnaire respondents: n (%) or mean (\pm SD)

	R* (n=24)	T0* (n=32)	T1* (n=32)	p; Cohen's d**
Background				
Age (at time of questionnaire) [15-29]^	20.8 (\pm 3.2)	19.1 (\pm 2.4)		
Gender (male)	8 (33.3)	17 (53.1)		
Educational level (high)	8 (50.0) ¹	11 (39.3) ⁴		
Age at diagnosis				
0 years	11 (45.8)	15 (46.9)		
1-5 years	3 (12.5)	8 (25.0)		
6-12 years	5 (20.8)	5 (15.6)		
13-16 years	5 (20.8)	4 (12.5)		
Treatment type				
Pre-dialysis	-	2 (6.3)		
Haemodialysis	4 (16.7)	6 (18.8)		
Peritoneal dialysis	-	-		
Kidney transplant	20 (82.3)	18 (56.3)		
Other	-	6 (18.8)		
Limitations in mobility [6-18]^	7.9 (\pm 2.0) ²	7.6 (\pm 2.0) ⁵		
Self-management				
General self-efficacy [10-40]^	27.7 (\pm 3.0)	30.7 (\pm 4.5)	32.1 (\pm 4.7)	<.05; .31
Disease-related self-efficacy				
Coping domain [4-16]^	14.3 (\pm 1.9)	13.8 (\pm 2.3) ⁷	13.7 (\pm 2.0) ²	ns
Knowledge domain [6-24]^	22.0 (\pm 2.1)	21.7 (\pm 2.6) ⁹	21.5 (\pm 2.5) ¹⁰	ns
Skills for hospital consultations [6-24]^	21.3 (\pm 3.5)	20.8 (\pm 3.2) ¹¹		
HRQoL [0-100]^				
General HRQoL	73.9 (\pm 11.4)	72.4 (\pm 17.0) ²	72.1 (\pm 14.2)	ns
Independence domain	82.9 (\pm 14.0)	78.1 (\pm 13.2)	83.9 (\pm 15.0)	<.01; .44
Emotion domain	63.2 (\pm 13.5)	71.1 (\pm 23.3) ⁵	71.3 (\pm 18.4) ²	ns
Social inclusion domain	75.7 (\pm 14.1)	74.1 (\pm 18.9) ⁶	70.5 (\pm 15.8)	<.05; -.19
Social exclusion domain	77.4 (\pm 18.8)	77.1 (\pm 17.6) ⁵	75.2 (\pm 18.2) ²	ns
Physical domain	68.2 (\pm 15.9)	60.6 (\pm 19.4)	60.1 (\pm 16.4) ²	ns
Medication domain	77.9 (\pm 16.4)	71.0 (\pm 20.4) ²	72.2 (\pm 21.7) ²	ns
Autonomy in social participation (yes independent)				
Finances	14 (58.3)	3 (15.0) ⁵		
Employment	7 (29.2)	3 (15.0) ⁶		
Living	6 (25.0)	3 (15.0) ⁶		
Relationships	15 (65.2) ²	16 (80.0) ⁶		
Sexuality	11 (50.0) ³	9 (52.9) ¹⁰		
Transportation	22 (100) ³	14 (70.0) ⁶		
Leisure	17 (70.8)	13 (68.4) ⁵		

*R=retrospective; T0=pre-camp; T1=post-camp

^Theoretical range

**Wilcoxon Signed Ranks Test (paired) for differences between T0 and T1 measurements, and Cohen's d for effect sizes.

Missing values: ¹n=8, ²n=1, ³n=2, ⁴n=4, ⁵n=13, ⁶n=12, ⁷n=3, ⁸n=1, ⁹n=22, ¹⁰n=21, ¹¹n=9

Camp COOL: the intervention

The rationale behind Camp COOL

One of the initiators had heard about a ‘transition camp’ in the UK [28] and felt this approach might be helpful for young people with ESRD in the Netherlands as well. He discussed his idea with parents and fellow professionals, and together they explored the specific needs of young people with ESRD. Realizing that acquiring autonomy and independence was especially hard for these young people, they widened the scope of the camp (particularly preparing for transition from pediatric to adult care and self-care) to a self-management camp (aimed at independent living with ESRD, i.e. the transition to adult care and adulthood). *“Self-management is the main theme of Camp COOL. It [...] requires self-confidence, self-efficacy, and self-consciousness”* (A). Next to this, knowledge of the disease and various skills are important for self-management.

Acquiring self-management skills was facilitated by buddy-to-attendee support. This implied that buddies – fellow patients already gone through the transition to adulthood and adult care – lead the day-to-day program, run the camp and counsel the attendees who have not moved on to adult care yet. Initiator A explained: *“They manage the week. We are present, but are invisible. We are only available if there is really something they need to know. But even then, we always let them come up with their own solutions first and ask them what they think is needed to solve a problem.”* The concept of buddy-to-attendee support presupposes that buddies will share their lived experiences, allowing for transfer of experiential knowledge. Also, it is hoped that buddies become role models. Buddies are not formally selected or trained, but receive some coaching during the two days before start of the actual camp. Also, buddies have a ‘buddy meeting’ every day to discuss anything that requires attention. Initiators select former attendees and ask them to become buddies, but apply no explicit selection criteria.

Furthermore, the program elements support building general competencies, e.g. a ‘how to present yourself’ workshop. There are no activities focused on the disease; attendees will not be lectured about side effects, for example. Although buddies lead the day-to-day program, in 2011 the initiators/staff had pre-selected the program elements. However, in 2012, the buddies had more to say about the program by selecting specific elements, presented in Table 6. This was done as a first step to evolve the buddy role, because it was noticed in the past years that buddies benefited from this role. In both years, a hospital social worker and an initiator were present.

The referring role of healthcare professionals

C (nephrologist) defined her referring role as being a *“counsellor”* who *“recruits young people”* with ESRD. Furthermore she mentioned that professionals may be asked to

take over the “background” role of the initiators during the camp, “only interfering when needed”.

All professionals agreed that age was the major selection criterion; 16 years or older in general. A social worker added that she also considers impact of the condition on the person’s daily life: *“Especially those who daily take medication and are on a diet. Or those who do not know how to deal with the condition at school, and those who have yet to learn to become independent”* (D).

Table 6 Program elements Camp COOL 2011 and 2012

CC-2011	CC-2012
<ul style="list-style-type: none"> ▪ Workshop ‘Present yourself’ ▪ Movie making workshop & self-made movie about Rating Camp COOL ▪ Dancing (Zumba) workshop ▪ Sports ▪ Cooking teams ▪ Free time 	<ul style="list-style-type: none"> ▪ Theater performance by professional artists on transition to adulthood (in general) ▪ Art workshop, creating a self-portrait ▪ Acting workshop & self-made talk show about transition, independence, and living on your own ▪ Drumming workshop ▪ Free time

Observations during the camp

Notably the first-timers needed to get acquainted with the new people they met and with the camp’s routine. Buddies helped breaking the ice. They started conversations with attendees, encouraged attendees to talk with one another, and told a lot about themselves to create an open atmosphere. There was an observable difference between first-timers and attendees who had joined previous camps. The latter were less hesitant to interact with others, and less often relied on their buddies. Buddies proactively engaged the new attendees in conversations. As the first day progressed, the ice had melted, and there was a warm and relaxed atmosphere.

Participants talked a lot with each other during activities and free time, a great deal about medical and social aspects of ESRD. Side effects of medication were discussed, in particular Prednisone. Insomnia, feeling hungry, and a “fat head” were often mentioned as annoying side effects. Participants during CC 2011 even came up with a story about a “Prednisone park” when they presented a show as one of the activities. Still, participants joked a lot about side effects. Other medical topics were transplantation, diets, treatment frequency, and treatment options. Social topics addressed were school, work, sports, risky behaviours like smoking, drinking or doing drugs, but also dealing with ESRD in social life. A major issue was the influence of ESRD and its treatment on school carrier, i.e. either or not being enrolled in special education and whether they felt pushed by their environment to do so. Another hot topic was ‘how to become independent from parents’. Participants during CC 2012 presented this in their evening show.

During certain activities the buddy role was more prominent, for instance during the 'Present yourself' workshop and the acting workshop. Buddies encouraged the attendees to actively participate in workshops. During the moviemaking workshop, one of the buddies urged attendees to come up with ideas: *"Hello, listen, I'm talking all the time here. You guys could come up with something as well!"* During free time, the buddy role varied from telling their attendees it was their turn to do the dishes to reminding them of their diets.

The buddy role was less prominent in the art workshop and preparations for the evening show. Here, the buddies seemed to adopt a more passive role and let the attendees figure things out on their own. In the preparations for the evening show, they only offered ideas on how the selected themes should be presented. Consequently, the show was largely the work of the attendees.

Interviews: The value of Camp COOL

All interviewed parties acknowledged that young people with ESRD needed to be supported in their development of self-management. Professionals mostly emphasized that young people with ESRD in adult care tended to show lack of independence, and initiators held the opinion they should actively develop autonomy and readiness for adult care and adult life. A former buddy (F) reasoned that adult care requires certain skills that are not necessarily trained for in pediatric care: *"You have to be attentive yourself. In pediatric care they arranged everything for you [...]. You must ensure that they won't just let you be. This happens. Other buddies had the same experience."*

Buddies and attendees had different reasons to participate in CC. While buddies thought of CC as a place to meet the others again and to enjoy themselves, attendees in general had to be encouraged by their parents to join. *"At first, I wasn't really up for it. My father signed me up. But I did not regret going to Camp COOL"* (M).

The most valued aspect of CC was peer support. Participants did not only appreciate the informative or instructional character of the peer support, but also found that meeting others *"who have been through the same"* helped them to *"put"* themselves and their ESRD *"into perspective"*. J (buddy) explained: *"Realizing that you are not the only one, or even that your own condition is not as bad as that of others. For instance, I saw that I was not the only one that got tired easily during sports."* Social comparison seems to be an inherent part of peer activities, as mentioned by K (attendee): *"Well, having heard stories of others, I feel lucky that things aren't going that bad for me. Some said they have been on dialysis for years or are still waiting for kidney transplantation. Yes, I think I am lucky that I do not have to wait anymore."* Young people emphasized that contacts with others in their social network differed from contacts with peers with ESRD: *"Other ESRD patients will understand your condition better than your own family or friends"* (L). N gave specific

examples: *"The freedom to take your medication without anyone asking you why you have to do this. And, that you do not have to hide a shunt from the outside world."*

Participants particularly appreciated the informative character of peer support. The sharing of experiences gave them new information on dealing with healthcare professionals, treatment options, and possible side effects. M (attendee) said: *"I didn't even know that I had side effects. [...] I sat down and said I was hungry again. And they said 'Prednisone.' I asked: 'Prednisone?!' And they said, yes, [being hungry] is one of the side effects of Prednisone. I went like, side effects?!"* Young people also learned more about generic issues of living with ESRD. P (buddy) mentioned living independently as an example: *"I learned something about being independent, because we talked about living on your own and how to arrange for that to happen."* Other issues mentioned were school, work, and dealing with friends.

Finally, buddies and attendees ascertained that the program elements had helped them to develop more *"self-confidence"* and *"perseverance"*, and had made it easier for them to *"be more daring"* and *"open towards others"*. The healthcare professionals, however, were less certain about the exact effects of CC. *"I cannot imagine it having no effects at all. Still, I can't specifically point out what the effects are"* (E). Their reluctance was related to the question whether or not any positive effects were directly attributable to the camp.

Interviews: Buddy-to-attendee support

The buddy is an important part of CC, and was much appreciated. The attendees mostly viewed the buddy as a companion who helped them through the first day and who guided the activities. *"I think it is important to have a buddy when you first get there. That he or she helps you to get used to the new environment. I had a very experienced buddy, who told me a lot"* (L, attendee). They appreciated that they could learn from their buddies, because: *"A buddy is more experienced [in living with ESRD]. So, it's a good thing that he is here. [...] A doctor can tell you all of it, but doesn't experience things. A buddy does"* (Q, attendee).

The initiators noticed that buddy-to-attendee support did not only benefit attendees, but that buddies themselves grew wiser from managing the camp too. *"The responsibility for the camp and the attendees makes them grow"* (B). Buddies in general indeed described having *"responsibility"* as the most important aspect of their role as and found this role to be threefold: 1) looking after others, 2) giving advice to others, and 3) running the program. The supervising role relates to monitoring medical regimen adherence, but also seeing to it that the attendee feels well and enjoys the activities. *"Especially the medication, she tried to hold off taking them. So, I tried to convince her it's crucial to take it on time"* (N, buddy). Buddy O said this about her attendee: *"You almost had to feed her. I really had to take care she ate enough; I sort of had to force her to do so."*

The advisor role revolves around listening to the attendees' stories and being able to advise them if asked to. Questions often concerned living with ESRD but could be medically oriented as well. Buddy O, for example, was asked about types of dialysis: *"I did both types of dialysis and therefore could tell them about the differences and consequences of choosing one method over the other"* (O). Finally, smooth running of the program is the responsibility of the buddies in their leader role: *"We as buddies take care of the daily camping program, we lead the camp"* (J).

All buddies mentioned that being a buddy was fruitful for them: they learned a lot and it increased their self-confidence. However, some felt insecure at times. Buddy N said: *"I found that difficult, because I could understand her feelings [of being misunderstood by family and friends], and of course I can advise her, but it made me feel like a psychologist and that is not my task"*. This goes to show that the buddy role is a challenging one. Buddy O had come to realize this: *"I do not get angry easily, but sometimes that's what is needed. So, if someone is extremely annoying, I would not know how to deal with it"*. Fortunately, the buddies would work together if needed and discuss problems during the buddy meeting.

Quantitative results: self-management of young people with ESRD and pre-post effects of Camp COOL

On average, all participants scored relatively high on self-efficacy measures and on health-related quality of life (Table 5). As for social participation, most of the respondents still lived with their parents (respectively 75% and 85% in the retrospective and 2011-2012 groups), and were involved in a romantic relationship (65.2% and 80.0%). Also, half of them or more were independent in the areas of sexuality (50.0% and 52.9%), transportation (100% and 70%), leisure (70.8%), and 68.4%). The young adults in the retrospective group were more frequently financially self-supporting (58.3%) than the participants in 2011-2012 (15.0%) (Table 5).

The 2011-2012 group reported significantly higher general self-efficacy after CC (Cohen's $d=.31$; $p<.05$). Disease-related self-efficacy did not differ between the T1 and T0 assessments. The mean score on the independence domain after CC was significantly higher ($d=.44$; $p<.01$), but the mean score on the social inclusion domain was significantly lower ($d=-.19$; $p<.05$) (Table 5). Discriminating between buddies and attendees, only attendees reported a significantly higher score on general self-efficacy ($d=.37$; $p<.05$) after CC. Also, only attendees perceived significantly lower HRQoL on the social inclusion domain after CC ($d=-.33$; $p<.05$). Buddies reported significantly higher HRQoL on the independence domain afterwards ($d=1.1$; $p<.05$) (Table 7).

A reasonably large proportion of respondents, i.e. half or more, found that participating in CC had positively influenced their daily lives on several areas, e.g. attitude toward illness, independence, self-confidence, ability to socially interact with others, knowl-

Table 7 Buddy-attendee comparison: n (%) or mean (\pm SD)

	BUDDIES (n=18)		p; Cohen's d**	ATTENDEES (n=14)		p; Cohen's d**
	T0*	T1*		T0*	T1*	
Background						
Age	20.7 (±2.0)			17.1 (±1.1)		
Gender (male)	10 (55.6)			7 (50.0)		
Educational level (high)	7 (50.0) ⁵			4 (28.6)		
Age at diagnosis						
0 years	9 (50.0)			6 (42.9)		
1-5 years	5 (27.8)			3 (21.4)		
6-12 years	1 (5.6)			4 (28.6)		
13-16 years	3 (16.7)			1 (7.1)		
Treatment type						
Pre-dialysis	1 (5.6)			1 (7.1)		
Haemodialysis	5 (27.8)			1 (7.1)		
Kidney transplant	11 (61.1)			7 (50.0)		
Other	1 (5.6)			5 (35.7)		
Limitations in mobility [6-18]^	7.0 (±2.0) ¹			7.8 (±2.0) ²		
Self-management						
General self-efficacy [10-40]^	31.2 (±4.1) ²	32.1 (±4.2) ²	ns	30.2 (±5.1) ²	32.1 (±5.6) ³	<.05; .37
Disease-related self-efficacy						
Coping domain						
[4-20]^	14.4 (±1.8) ³	13.8 (±1.8)	ns	13.1 (±2.8) ²	13.6 (±2.4) ²	ns
Knowledge domain						
[7-35]^	26.2 (±2.9) ⁴	25.8 (±3.1) ¹	ns	23.6 (±3.3) ⁴	24.2 (±2.8) ⁴	ns
HRQoL [0-100]^						
General HRQoL	73.3 (±13.2) ¹	74.0 (±11.6) ²	ns	72.0 (±18.9) ²	69.8 (±16.9)	ns
Independence domain	77.9 (±7.4)	86.1 (±10.9)	<.05; 1.1	78.5 (±18.9) ²	81.0 (±19.0)	ns
Emotion domain	66.7 (±20.7) ¹	73.1 (±15.1) ²	ns	73.2 (±24.9) ²	69.1 (±22.2)	ns
Social inclusion domain	72.2 (±12.0) ¹	72.5 (±11.8)	ns	74.9 (±21.5)	67.9 (±20.0)	<.05; .33
Social exclusion domain	83.8 (±15.8) ¹	79.4 (±15.7) ²	ns	74.0 (±18.2) ²	70.1 (±20.4)	ns
Physical domain	59.9 (±13.7)	58.7 (±11.7)	ns	61.6 (±25.5)	62.2 (±21.8) ²	ns
Medication domain	75.3 (±17.5)	75.0 (±20.3) ²	ns	65.2 (±23.3) ²	68.8 (±23.6)	ns
Overall score for CC [1-10]^	9.2 (±.73)			8.4 (±.68)		

*T0=pre-camp; T1=post-camp

** Wilcoxon Signed Ranks Test (paired) for differences between T0 en T1 measurements, and Cohen's d for effect sizes.

Missing values: ¹n=12, ²n=1, ³n=2, ⁴n=13, ⁵n=4[^]Theoretical range

edge of the condition, and insight into what the transition to adult care involves. The least influence was perceived on healthier living (respectively 16.7% and 37.5% in the retrospective and 2011-2012 groups) (Table 8). The majority of the attendees appreciated having a buddy (91% and 85.7%), but the 'personal' buddy was not always the one they learned the most from. More than half of the buddies in the 2011-2012 group (57.2%)

Table 8 Rating Camp COOL: frequency (%) of respondents agreeing or totally agreeing with the statements; mean (\pm SD) for overall score

	R* (n=24) outcomes	T0* (n=32) expectations	T1* (n=32) outcomes
I expect (T0) / found (R and T1) CC to positively influence my:			
<i>Dealing with physical limitations</i>	9 (37.5)**	21 (65.6)	21 (65.6)**
<i>Attitude toward illness</i>	11 (45.8)***	19 (59.4)	24 (75.0)***
<i>Healthier living</i>	4 (16.7)	8 (25.0)	12 (37.5)
<i>Knowledge of the condition</i>	9 (37.5)**	20 (62.5)	18 (56.3)**
<i>Independence</i>	7 (29.2)	21 (65.6)	16 (50.0)
<i>Self-confidence</i>	11 (45.8)	16 (50.0)	16 (50.0)
<i>Ability to socially interact</i>	10 (41.7)	12 (37.6)	16 (50.0)
<i>Insight into what the transition to adult care holds</i>	10 (43.5) ¹	19 (61.3) ¹	18 (51.3)
<i>Being prepared for transition to adult care</i>	7 (30.4) ¹	15 (62.5) ⁵	12 (52.2) ¹
<i>Assertiveness</i>	8 (33.3) ¹	11 (35.5) ¹	14 (43.8)
The value of buddy-to-attendee support (yes):			
<i>As an attendant, I appreciated having a buddy</i>	10 (91.0) ²		12 (85.7) ⁶
<i>As an attendant, I learned the most from my buddy</i>	5 (45.5) ²		8 (57.2) ⁶
<i>As a buddy, I learned more during CC than I did as attendant</i>	2 (28.6) ³		8 (57.1) ⁴
<i>As a buddy, I would recommend being a buddy to others</i>	8 (80.0) ⁴		15 (93.8) ⁷
Overall score for CC [1-10][^]	8.0 (\pm1.2)		8.9 (\pm.82)¹

*R=retrospective; T0=pre camp; T1=post camp

[^]Theoretical range

**p<.05; Wilcoxon Signed Ranks Test (independent) for differences between R and T1 (at mean level)

***p<.01; Wilcoxon Signed Ranks Test (independent) for differences between R and T1 (at mean level)

Missing values: ¹n=1, ²n=13 (attendees only), ³n=17 (buddies only), ⁴n=14 (buddies only), ⁵n=8, ⁶n=18 (attendees only), ⁷n=16 (buddies only)

thought they learned more from being a buddy than from being an attendee, but in the retrospective group fewer buddies agreed with this statement (28.6%). The majority in both groups would recommend being a buddy to others. The mean (\pm SD) overall CC appreciation score assigned by participants in the retrospective group was 8.0 (\pm 1.2) on a scale from 1 to 10, versus 8.9 (\pm .82) by participants in the 2011-2012 group. Respondents in the 2011-2012 group were also more positive about the perceived effects of CC on dealing with physical limitations, attitude toward illness, and knowledge of the condition than those in the retrospective group (Table 8). There were no significant differences between expectations and outcomes in the 2011-2012 group.

Integration of findings

The 2007-2010 and 2011-2012 groups were very similar when considering HRQoL and social participation. The first group was more financially self-supporting, but then, their

mean age was higher at time of the questionnaire. All parties acknowledged that young people need support in their development of self-management. This was also implicitly observed during the camp: becoming independent was a hot topic, and was processed in activities by the campers.

The perceived effects of CC mentioned in the interviews were increased self-confidence, more knowledge of ESRD, feeling capable of being more responsible and open towards others, and daring to stand up for yourself. In the quantitative evaluation of CC half or more of the participants reported the same effects. Furthermore, the pre-post analyses showed that general self-efficacy of attendees, and independence as domain of HRQoL of buddies had increased after attending CC, whereas social inclusion as domain of HRQoL of attendees had decreased. Peer support was the most valued aspect of CC, both mentioned in the interviews and found in the questionnaires. It was perceived as informative, but even more importantly as a great opportunity to meet fellow patients. This was also observed during CC.

Appreciation of buddy-to-attendee support was demonstrated in both the interviews and questionnaires. Buddies were expected to transfer knowledge and to be an example for attendees. Indeed, during the interviews attendees mentioned that they learned a lot from buddies, and observations showed the same. Buddies shared experiences and knowledge, looked after their attendees, and led the camp. The buddy role was given shape as a pro-active combination of supervisor, advisor, and leader.

DISCUSSION

Self-management support, effects of CC, and the buddy role

It would seem evident that young people with ESRD need support in developing self-management skills. When it comes to social participation, for instance, young people in our samples most resemble those we labelled as “outgoing laggards” in another study, with little autonomy in the areas of finances, employment, and living, while at the same time enjoying romantic relationships and socialization with peers [47]. Becoming independent in the areas of living, employment and finances was much discussed during CC, showing that young people with ESRD seem to be lagging behind in these areas. This finding is in line with the results of other studies [6, 7], and calls for more specific support for work-participation. The different attitude towards self-management found for the majority of the older participants, despite similar HRQoL and social participation, indicates that age is an important determinant of self-management.

The positive effects we encountered – e.g. increased self-efficacy, self-confidence, and knowledge of ESRD – were also reported previously as benefits of therapeutic camping for young people with a variety of chronic conditions [20, 25-27, 31, 33, 48, 49],

and benefits of peer support [16]. It seems that Camp COOL creates an environment that allows for “mastery experiences” and “learning by examples” [50]. Greater self-efficacy can positively affect different levels of functioning in young people with ESRD. This is especially valuable for those who still have to transfer to adult care and adulthood, and provides support to pediatric nephrologists for referring young people to CC or initiating such camps.

However, we also found diminished sense of social inclusion (as part of HRQoL) of attendees after CC. This may be due to the fact that a subculture is created during the camp in which the attendees perceive themselves as being different from others. This was identified in previous studies as a possible disadvantage of peer support [19], and requires attention. Olsson and colleagues [19] argued that this “over-identification” might be counteracted by addressing it in the group. This may be an important recommendation for future camps.

Participating as a buddy during CC had a positive effect on the independence domain of HRQoL, implying that being a buddy fosters confidence in future living without impairments caused by ESRD. Positive effects of a challenging buddy role have been reported previously for renal peer support volunteers [51], and peer leaders in an asthma self-management camp [52]. Also, the buddies’ combined roles of supervisor, advisor and leader for seems to match with the three types of assistance identified with peer support based on experiential knowledge (i.e. emotional, appraisal and informational assistance) [53, 54]. Still, this combined role might be too challenging for untrained buddies. Although buddies receive some coaching and have buddy meetings, for the buddy role to be effective a buddy should possess the skills and knowledge required to act as a role model [55]. Selection and training of peer supporters is important. Therefore, a recommendation for CC in the future is to more carefully select buddies and to specifically train or coach them to be models. This could counteract any negative effects of peer support [16, 19]. Pediatric nephrologists could involve their counterparts from adult care in selecting potential buddies.

Strengths and limitations

This study is one among the first to evaluate therapeutic camping for young people with ESRD and one of the few considering effects of therapeutic camping in chronically ill young people in MMR. To our knowledge, it is the first that more specifically looks at the benefits of buddy-to-attendee support during therapeutic camping. Furthermore, the use of MMR added to the comprehensiveness of this study, and led to a broader insight into CC. Mixed methods research also partially overcomes the disadvantage of a convenience sample and of the small sample size inherent to this specific disease group, because it allows for exploration of findings from different angles and at different levels. Although randomized controlled trials are seen as the golden standard of research

evidence, conducting this type of research was not considered feasible. One reason for this was the low prevalence of childhood ESRD and the (presumed) difficulty in getting a powered sample. We also considered the ethical challenge associated with randomizing young people with ESRD to a potentially beneficial intervention [35].

A limitation of our study is the lack of an appropriate comparison group. In 2012, 518 young people with chronic conditions responded to a questionnaire about self-management that contained the same measures used in this study [47]. Unfortunately, a few respondents had ESRD, so that we could not create a comparison group.

Also, a printing error in the pre-post questionnaires in 2011 led to missing data in the self-efficacy questionnaires, thereby weakening the results of the quantitative evaluation. Furthermore, the measurements in the 2011-2012 group were timed just before and after CC, not allowing for exploration of any long-term effects. However, some long-term effects were explored by comparing this group with the retrospective sample. Although they mentioned similar effects of CC in the interviews, the quantitative results showed that the latter group, which participated longer ago, was slightly less positive about the effects. Future studies should include more measurement moments after the camp to explore the long-term effects. Finally, allowing buddies to determine the final camping program led to different activities during the two camps and a more manifested role for buddies in CC 2012, which may have influenced our findings. However, since results from both years were compared and yielded the same findings, we expect this influence to be small.

CONCLUSIONS

Participating in CC seems to have a positive influence on self-management of young people with ESRD aged 16-25 years. Peer-to-peer support in the form of buddy-to-attendee support is very much appreciated and support from young adults is not only beneficial for adolescent attendees, but also for the young adult buddies. It is therefore recommended to keep or start organizing CC for these young people. Pediatric nephrologists are encouraged to refer patients to CC and to facilitate such initiatives. Together with nephrologists in adult care, they could take on a role in selecting buddies. Also, since young people with other chronic conditions may also benefit from CC, it is advised to explore the possibilities to organize the camp for other groups as well. When organizing future camps, more attention should be given to the selection and training of buddies, and to the imminent effect of over-identification in order to counteract any negative effects. Future evaluation studies could benefit from a MMR approach, the inclusion of a control group and more measurement moments.

ACKNOWLEDGEMENTS

This study was funded by the Dutch Kidney Foundation. The authors thank the young people, healthcare professionals, and initiators/staff that participated in this study. Furthermore, former students of Bachelor in Nursing and Bachelor in Physical therapy, Charlotte de Haan, Karin van den Berge, Nathalie Breedveld, Marlies Verweij, Robert de Ruiter, and Karin Volkers (Rotterdam University for Applied Sciences) are thanked for their assistance in data collection. Next, we are grateful to our advisory board: Jane de la Fosse (COOL Foundation), Harry Weezeman (Dutch Kidney Patient League), and Eefje Verhoof (Amsterdam Medical Center). Jane de la Fosse (Camp COOL Foundation) and Kees Kuin (Amsterdam Medical Center) helped executing the study in a fantastic way. Jaap Groothoff (Amsterdam Medical Center) is thanked for his useful input about Camp COOL and for his comments on the quantitative design. Finally, we thank Roland Bal (Erasmus University Rotterdam) for his valuable comments on an earlier draft of this paper.

REFERENCES

1. 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.
2. Nicholas DB, Picone G, Selkirk EK: The lived experiences of children and adolescents with end-stage renal disease. *Qual Health Res* 2011, 21(2):162-173.
3. Pomaki G, Delongis A, Anagnostopoulou T, Heininger J: Can't live with you, can't live without you: negative family exchanges and adaptation in end-stage renal disease patients. *J Health Psychol* 2011, 16(3):520-529.
4. Jansen DL, Heijmans M, Rijken M, Kaptein AA: The development of and first experiences with a behavioural self-regulation intervention for end-stage renal disease patients and their partners. *J Health Psychol* 2011, 16(2):274-283.
5. Kiberd JA, Acott P, Kiberd BA: Kidney transplant survival in pediatric and young adults. *BMC Nephrol* 2011, 12:54.
6. Icard PF, Hower SJ, Kuchenreuther AR, Hooper SR, Gipson DS: The transition from childhood to adulthood with ESRD: educational and social challenges. *Clin Nephrol* 2008, 69(1):1-7.
7. 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.
8. 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.
9. Darbyshire P, Oster C, Henning P: Children's and young people's experiences of chronic renal disease: a review of the literature, methodological commentary and an alternative proposal. *J Clin Nurs* 2006, 15(6):751-760.
10. Tjaden L, Tong A, Henning P, Groothoff J, Craig JC: Children's experiences of dialysis: a systematic review of qualitative studies. *Arch Dis Child* 2012, 97(5):395-402.
11. Tong A, Henning P, Wong G, McTaggart S, Mackie F, Carroll RP, Craig JC: Experiences and perspectives of adolescents and young adults with advanced CKD. *Am J Kidney Dis* 2013, 61(3):375-384.
12. Jansen DL, Grootendorst DC, Rijken M, Heijmans M, Kaptein AA, Boeschoten EW, Dekker FW, PREPARE-2 Study Group: Pre-dialysis patients' perceived autonomy, self-esteem and labor participation: associations with illness perceptions and treatment perceptions. A cross-sectional study. *BMC Nephrol* 2010, 11:35.
13. 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.
14. Maurice-Stam H, Verhoof EJ, Caron HN, Grootenhuis MA: Are survivors of childhood cancer with an unfavourable psychosocial developmental trajectory more likely to apply for disability benefits? *Psycho-oncol* 2011, 22(3):708-714.
15. Pendley JS, Kasmen LJ, Miller DL, Donze J, Swenson C, Reeves G: Peer and family support in children and adolescents with type 1 diabetes. *J Pediatr Psychol* 2002, 27(5):429-438.
16. Hughes J, Wood E, Smith G: Exploring kidney patients' experiences of receiving individual peer support. *Health Expect* 2009, 12(4):396-406.
17. Waldrup AM, Malcolm KT, Jensen-Campbell LA: With a Little Help from Your Friends: The Importance of High-quality Friendships on Early Adolescent Adjustment. *Soc Dev* 2008, 17(4):832-852.

18. Wells F, Ritchie D, McPherson AC: 'It is life threatening but I don't mind'. A qualitative study using photo elicitation interviews to explore adolescents' experiences of renal replacement therapies. *Child Care Health Dev* 2013, 39(4):602-612.
19. Olsson CA, Boyce MF, Toubourou JW, Sawyer SM: The Role of Peer Support in Facilitating Psychosocial Adjustment to Chronic Illness in Adolescence. *Clin Child Psychol Psychiatry* 2005, 10(1): 78-87.
20. Epstein I, Stinson J, Stevens B: The effects of camp on health-related quality of life in children with chronic illnesses: a review of the literature. *J Pediatr Oncol Nurs* 2005, 22(2):89-103.
21. Bekesi A, Torok S, Kokonyei G, Bokretas I, Szentes A, Telepoczki G, European KG: Health-related quality of life changes of children and adolescents with chronic disease after participation in therapeutic recreation camping program. *Health Qual Life Outcomes* 2011, 9:43.
22. Kiernan G, Gormley M, MacLachlan M: Outcomes associated with participation in a therapeutic recreation camping programme for children from 15 European countries: Data from the 'Barretstown Studies'. *Soc Sci Med* 2004, 59(5):903-913.
23. Shepanski MA, Hurd LB, Culton K, Markowitz JE, Mamula P, Baldassano RN: Health-related quality of life improves in children and adolescents with inflammatory bowel disease after attending a camp sponsored by the Crohn's and Colitis Foundation of America. *Inflamm Bowel Dis* 2005, 11(2): 164-170.
24. Moons P, Barrea C, Suys B, Ovaert C, Boshoff D, Eyskens B, Vandenrijn C, Sluysmans T: Improved perceived health status persists three months after a special sports camp for children with congenital heart disease. *Eur J Pediatr* 2006, 165(11):767-772.
25. Warady BA: Therapeutic camping for children with end-stage renal disease. *Pediatr nephrol* 1994, 8(3):387-390.
26. Cushner-Weinstein S, Berl M, Salpekar JA, Johnson JL, Pearl PL, Conry JA, Kolodgie M, Scully A, Gaillard WD, Weinstein SL: The benefits of a camp designed for children with epilepsy: evaluating adaptive behaviors over 3 years. *Epilepsy Behav* 2007, 10(1):170-178.
27. Torok S, Kokonyei G, Karolyi L, Ittzes A, Tomcsanyi T: Outcome effectiveness of therapeutic recreation camping program for adolescents living with cancer and diabetes. *J Adolesc Health* 2006, 39(3):445-447.
28. Hackett J, Johnson B, Shaw KL, McDonagh JE: Friends United: an Evaluation of an Innovative Residential Self-Management Programme in Adolescent Rheumatology. *Br J Occup Ther* 2005, 68(12):567-573.
29. Briery BG, Rabian B: Psychosocial changes associated with participation in a pediatric summer camp. *J Pediatr Psychol* 1999, 24(2):183-190.
30. Sawin KJ, Lannon SL, Austin JK: Camp experiences and attitudes toward epilepsy: a pilot study. *J Neurosci Nurs* 2001, 33(1):57-64.
31. Bluebond-Langner M, Perkel D, Goertzel T: Pediatric cancer patients' peer relationships: The impact of an oncology camping experience. *J Psychosoc Oncol* 1991, 9(2):67-80.
32. Cheung R, Young Cureton V, Canham DL: Quality of life in adolescents with type 1 diabetes who participate in diabetes camp. *J Sch Nurs* 2006, 22(1):53-58.
33. Gillard A, Witt PA, Watts CE: Outcomes and processes at a camp for youth with HIV/AIDS. *Qual Health Res* 2011, 21(11):1508-1526.
34. O'Mahar K, Holmbeck GN, Jandasek B, Zukerman J: A camp-based intervention targeting independence among individuals with spina bifida. *J Pediatr Psychol* 2010, 35(8):848-856.
35. Moola FJ, Faulkner GEJ, White L, Kirsh JA: The psychological and social impact of camp for children with chronic illnesses: a systematic review update. *Child Care Health Dev* 2014, 40(5):615-631.

36. Walker DA, Pearman D: Therapeutic recreation camps: an effective intervention for children and young people with chronic illness? *Arch Dis Child* 2009, 94(5):401-406.
37. Hunter HL, Rosnov DL, Koontz D, Roberts MC: Camping Programs for Children with Chronic Illness as a Modality for Recreation, Treatment, and Evaluation: An Example of a Mission-Based Program Evaluation of a Diabetes Camp. *J Clin Psychol Med S* 2006, 13(1):64-77.
38. Creswell JW: Research design: Qualitative, quantitative and mixed methods. London: SAGE; 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. O'Cathain A, Murphy E, Nicholl J: The quality of mixed methods studies in health services research. *J Health Serv Res Polic* 2008, 13(2):92-98.
41. Clark JP: How to peer review a qualitative manuscript. In *Peer Review in Health Sciences*. Second edition. Edited by Godlee F, Jefferson T. London: BMJ Books; 2003:219-235
42. Ware JE: Memorandum: Users of the Medical Outcomes Study (MOS) 20-Items Short-Form Health Survey (SF-20) Santa Monica, Calif: The Rand Co; 1989.
43. Schwarzer R, Jerusalem M: Generalized Self-Efficacy scale. In: *Measures in health psychology: A user's portfolio Causal and control beliefs*. Edited by Weinman J, Wright S, Johnston M. Windsor, UK: NFER-NELSON; 1995: 35-37.
44. Van Staa AL: On Your Own Feet: Adolescents with chronic conditions and their preferences and competencies for care (Doctoral dissertation). Rotterdam: Rotterdam University; 2012.
45. Schmidt S, Peterson C, Mühlen H: The DISABKIDS Questionnaires-Handbook incl. CD-Rom. Lengerich: Pabst Science Publishers; 2006.
46. Donkervoort M, Wiegerink DJ, van Meeteren J, Stam HJ, Roebroek ME, Transition Research Group South West N: Transition to adulthood: validation of the Rotterdam Transition Profile for young adults with cerebral palsy and normal intelligence. *Dev Med Child Neurol* 2009, 51(1):53-62.
47. Sattoe JNT, Hilberink SR, van Staa A, Bal R: Lagging behind or not? Four distinctive social participation patterns among young adults with chronic conditions *J Adol Health* 2014, 54(4):397-403.
48. Plante WA, Lobato D, Engel R: Review of group interventions for pediatric chronic conditions. *J Pediatr Psychol* 2001, 26(7):435-453.
49. Woods K, Mayes S, Bartley E, Fedele D, Ryan J: An Evaluation of Psychosocial Outcomes for Children and Adolescents Attending a Summer Camp for Youth with Chronic Illness. *Child Health Care* 2013, 42(1):86-98.
50. Bandura A: Self-efficacy: the exercise of control. New York: W.H. Freeman and Company; 1997.
51. Brunier G, Graydon J, Rothman B, Sherman C, Liadsky R: The psychological well-being of renal peer support volunteers. *J Adv Nurs* 2002, 38(1):40-49.
52. Rhee H, McQuillan BE, Belyea MJ: Evaluation of a Peer-Led Asthma Self-Management Program and Benefits of the Program for Adolescent Peer Leaders. *Respir Care* 2012, 57(12):2082-2089.
53. Dennis CL: Peer support within a health care context: a concept analysis. *Int J Nurs Stud* 2003, 40(3):321-332.
54. Embuldeniya G, Veinot P, Bell E, Bell M, Nyhof-Young J, Sale JE, Britten N: The experience and impact of chronic disease peer support interventions: A qualitative synthesis. *Patient Educ Couns* 2013, 92(1):3-12.
55. Bartholomew LK, Parcel GS, Kok G, Gottlieb NH, Fernandez ME: Planning Health Promotion Programs: An Intervention Mapping Approach, 3rd edn. San-Francisco: Jossey-Bass; 2011.

Appendix C9.1 Questions considering: Perceived influence on living with the condition (of Camp COOL)

Indicate which of the boxes below best describes your position.

	Completely Disagree	Disagree	Somewhat Agree	Agree	Completely Agree
After participating in Camp COOL, I am more capable in dealing with the physical limitations my condition and my treatment entail.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I feel more positive about living with my condition and my treatment.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I will start living a healthier life (for example: <i>Not drinking when going out, taking my medication on time, and paying more attention to my fluid restriction</i>).	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I've become more knowledgeable about my condition and my treatment.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I've become more independent (for example: <i>I'll be able go to the doctor for consultation by myself</i>)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I've gained more confidence (for example: <i>I'll be more proactive in asking my doctor questions</i>)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I'll be better at connecting with people socially.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I'm more informed about what it means to make the transfer to a hospital or department for adults.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I'll be better suited to making the transfer to a hospital or department for adults (<i>if you have already made this transfer, leave the boxes blank</i>).	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
After participating in Camp COOL, I feel I'm more courageous (for example: <i>I'll be able to tell my boss I want to work more / less more easily</i>)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Questions considering: value of the buddy-attendant concept*For attendants:*

Indicate which of the boxes below best describes your position.

	Completely Disagree	Disagree	Somewhat Agree	Agree	Completely Agree
At Camp COOL 2012, I learnt the most from the buddies.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I liked the fact that there were buddies present at Camp COOL, who also suffer from a kidney condition.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

For buddies:

Indicate which of the boxes below best describes your position.

	Completely Disagree	Disagree	Somewhat Agree	Agree	Completely Agree
At Camp COOL, I learnt more as a buddy than as a participant <i>(if you've never been a participant at Camp COOL, leave the boxes blank)</i>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
I would definitely recommend being a buddy at Camp COOL to other kidney patients.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

10

CONCLUSION

Main findings, general discussion &
future directions



OUTLINE

This thesis explored self-management of young people growing up with chronic conditions. The first part addressed the concepts of self-management and self-management support, the second part looked at the development of self-management and influencing factors in a cohort of young people with various chronic conditions, while the third part explored the effectiveness of self-management interventions for young people with chronic conditions. This concluding chapter opens with an overview of the main findings in the light of current literature, followed by methodological considerations, and a general discussion. It closes with lessons and future directions for practice and research, and lastly presents key messages.

MAIN FINDINGS

Self-management and self-management support: Shifting between dimensions and roles

Self-management is a multifaceted and contested concept. In the introduction chapter, a holistic view on self-management was introduced, in which young people with chronic conditions have three tasks: medical management, role management, and emotion (or identity) management [1]. Such broad scope allows for the consideration of developmental trajectories from childhood into adolescence and young adulthood, and therefore was deemed appropriate for use in health care for young people growing up with chronic conditions. This holistic view was presented to experts in the field of self-management in a Delphi study (*chapter 1*) and to health care professionals in pediatric nephrology (*chapter 2*). This led to a great deal of discussion about the concept of self-management.

Remarkably, it were not the domains of self-management themselves that were subject of discussion. All researchers and policy advisors in the Delphi study found Lorig & Holman's model useful for the conceptualization of self-management, and the interviewed health care professionals generally agreed with the domains addressed in the *Skills for Growing Up in pediatric Nephrology* tool. Also, the review of self-management interventions (*chapter 4*) showed that some interventions addressed multiple domains of self-management, instead of medical management alone. Participants in the first two studies seemed to disagree, however, on the supposed focus of self-management in the context of healthcare. While the researchers and policy advisors in the Delphi study seemed to focus on role and emotion management (*chapter 2*), medical professionals (*chapter 3*) and current approaches to self-management support (*chapter 4*) were more often focused on medical management. Those with a narrower focus on medical

management were usually more concerned with good clinical outcomes and a leading role for health care professionals, whereas those with a broader focus generally linked self-management to quality of life and shared decision-making.

Similar differences in perspectives are found in the international self-management literature. Kendall and colleagues, for instance, identified three approaches to self-management [2]. Two of these, named the policy discourse and the professional discourse, respectively, narrowly emphasize medical management. The third, the patient discourse, is of a broader scope by focusing not merely on one domain of self-management [2]. While these discourses of self-management can be linked one-to-one to specific groups – in this case policy makers, health care professionals and patients – other studies report different perspectives on self-management within groups, like in our Delphi study (*chapter 2*). A Q-study, for instance, distinguished four different preference profiles among adolescents with chronic conditions [3], and another study found that nurses' views on self-management support differed and could be distinguished into four distinct perspectives [4]. Interestingly, although the profiles in these studies represent different and sometimes conflicting views, the authors suggest that one could fit more than one profile, i.e. apart from the dominant profile. Nurses should even need to switch between profiles to provide tailored self-management support [4]. Another study, too, found that people with chronic conditions switched between up to four different self-management patterns [5].

This reasoning is in line with the notion of 'shifting perspectives' that conceptualizes living with a chronic condition as a continuous shifting between the illness-on-the-foreground and wellness-on-the-foreground perspectives [6]. The course of illness, for example, can be of influence on self-management support needs [7, 8], in that priorities and goals may change along the course. This implicates that self-management is a dynamic process rather than a fixed reality, and which requires flexibility in the operationalization of self-management support and in the role fulfillment of involved parties. In *chapter 2*, self-management support is therefore presented as a twofold task for professionals: first gaining understanding of the person's needs in dealing with a medical condition and its treatment, and of lived experiences and societal roles, and next empowering the person to engage in self-management and facilitating the development of self-management skills. Note that self-management goes beyond the individual to a wider social unit. After all, everyone is part of a social and material environment, and others will help the patient to self-manage [9] or influence how he or she will self-manage [10]. Enabling people to self-manage also involves role shifts, because sometimes people prefer to hear from health care professionals what they should do and at other times they feel capable enough to co-decide or even to decide for themselves (which is nicely illustrated by the preference profiles of adolescents [3]). Self-management support thus requires shifting between dimensions of self-management and between roles of involved parties, as

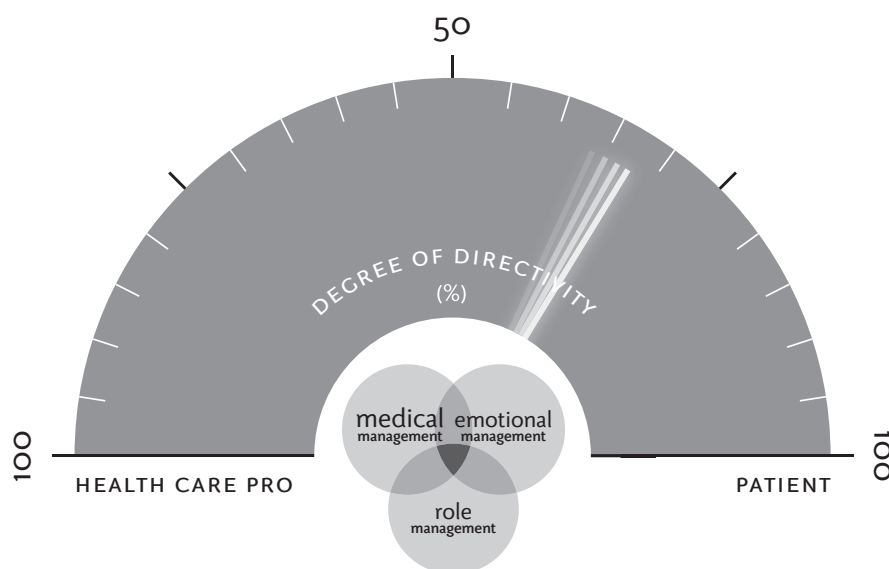


Figure 1 Self-management support: Shifting between dimensions and roles

presented in Figure 1. What this means in the special case of young people growing up with chronic conditions, will be addressed further below under the heading general discussion. The next two sections paragraphs further elaborate on young people's self-management support needs, and the factors associated with it.

Self-management support needs of young people with chronic conditions: Transitioning to adulthood and adult care

Young people growing up with chronic conditions in general go through two transitions while dealing with the task of gradually taking up self-management: the developmental transition to adulthood and the factual transition to adult care [11]. While the transition to adulthood is aimed at becoming an autonomous adult in society in general (in life areas such as education, employment or living independently), the transition to adult care implies taking up adult roles in the context of health care. An extra complication could be that additional health and illness transitions may also take place during puberty.

Most of the young adults in the cohort study presented in part 2 of this thesis did not yet experience a full transition to adulthood – thereby lagging behind their healthy age-peers in autonomy in social participation in one or more life areas (*chapter 5*). Similar findings were reported in other studies, too [12-15]. With respect to the transition to adult care, many of the young adults rated the actual process as unsatisfactory (*chapter 6*). Previous studies, in which this process was also seen as a complicated one, concluded

that developmentally appropriate transitional care is required to support young people during this transition [11, 16-19]. Overall, HRQoL of the studied cohort had decreased after transition (*chapter 7*). These findings underscore the notion that young people indeed need support in their uptake of self-management tasks and need to prepare for the transition to adult care. Moreover, the variety in social participation patterns and transfer experiences emphasizes that support needs differ between young people, indicating that tailored and personalized health care is a must.

Transitions of young people with chronic conditions: Associated factors

To date, outcomes of young people growing up with chronic conditions have been examined in a few studies [20, 21], but to our knowledge none followed a cohort of adolescents over time into young adulthood. By doing precisely that, the cohort study reported in the second part of this thesis (*chapters 5, 6, 7*) provides unique insights into factors associated with transitions of these adolescents. It indicates what aspects should be taken into account when deciding on how to support these young people.

Our results suggest that there are gender differences in the way the transitions are experienced. Male gender was more often associated with the ‘slow developers’ social participation pattern, but also with better transfer experiences, higher satisfaction with the transfer to adult care, and higher HRQoL. The transition to adulthood could also be less successful for those young people with physical limitations, those who followed special education, and those who received disability benefits. Thus, while for males and those suffering from physical disabilities it may be appropriate to focus self-management support more on the transition to adulthood, females may more specifically need emotional support, and support to deal with the transfer to adult care.

If we look at the associations between adolescents’ reports and young adults’ experiences with the transitions, only the attitude towards transition (to adult care) during adolescence was related to the actual transfer experiences reported in young adulthood. A more positive attitude related to better transfer experiences. Others have already suggested that young people’s transfer experiences can be enhanced by paying attention to transition readiness in adolescent care [22-24]. Our finding confirms this and shows that a more positive attitude towards transition during adolescence, which is an indicator of better readiness [24], indeed leads to more positive ratings of the transfer to adult care. Therefore, the use of interventions or support in adolescent care aimed to enhance transition awareness and readiness should be encouraged.

Furthermore, adolescents who reported higher independence during consultations achieved more autonomy in social participation in young adulthood. This may suggest that fulfilling adult roles in the broader context of society may also be beneficial for the fulfillment of an adult role in the health care context. In turn, learning to become a partner in health care could provide the opportunity to practice more general life skills.

So, while a large part of self-management activities takes place outside the health care context, health care providers may positively influence self-management by activating or empowering young people during consultations. It may be useful, for example, to see the adolescent alone, without parents, during consultations, or part thereof [25-28], or to design an individual transition plan that engages young people, parents and health care professionals in shared goal-setting [29, 30].

Our study confirmed that support should not be confined to pediatric or adolescent care. Higher perceived patient-centeredness of health care providers in adult care was correlated with both better transfer experiences and higher satisfaction with the transfer. Others also found that provider characteristics are important, and that it is essential for young people to be acknowledged as a partner in health care [31-33]. Still, today's debate around transition focuses on better preparation of adolescents, whereas less attention is paid to making adult services more responsive to young adults' needs [34-36].

Furthermore, the relationship between independence during consultations and autonomy in social participation persisted in young adulthood. Similarly, higher self-efficacy in young adulthood was also associated with more autonomy in social participation. Although no conclusions could be drawn about the direction of this relation, previous studies established self-efficacy as an important determinant of self-management. Adolescents' self-efficacy is reported to be a predictor for positive health outcomes or self-care in, for example, diabetes [37-39] and asthma [40]. Higher self-efficacy is also associated with better school performance of young people with cystic fibrosis [41], better emotional outcomes in young adults with cystic fibrosis or pediatric cancer survivors [42], better adaption to the chronic condition and less condition-related distress in diabetes [43, 44], better quality of life in adolescents with juvenile idiopathic arthritis and diabetes [45] and more transfer readiness [24] in young people with chronic conditions in general. Since being more self-efficacious can positively affect several important life domains, self-management support for young people should be aimed at enhancing self-efficacy, in both pediatric and adult care.

The implications of these insights into self-management support needs and associated factors will be further dealt with in the general discussion. The next sections elaborate on current approaches to self-management support for these young people and the (study of) effectiveness of self-management interventions.

Self-management support: A multidisciplinary task in health care for young people with chronic conditions

Chapter 4 showed that most current self-management interventions for young people with chronic conditions only address the medical domain, while few cover all domains of self-management. Apparently, there is a strong focus on medical management, at least as represented in the literature. Widening the range of self-management support

requires health care providers to move beyond the medical domain, and to look at self-management as the outcome of interactions between medical, role and emotion management.

In *chapter 3*, health care professionals' experiences with the Skills for Growing Up in pediatric Nephrology tool were explored. This tool includes all three domains of self-management, and some of the health care professionals indeed raised the question whether or not their responsibility as a health care professional should extend beyond medical management. To deal with this issue, most teams selected the nurse as the interventionist. The nurse had the task to explore patients' independence, goals for change, and need for support across the different domains, and to report her findings during multidisciplinary team meetings. In this way she was able to select other support sources for the patient. When, for instance, a young person had questions about school and work, she could refer him or her to the social worker. As such, the nurse fulfills an important part of self-management support. This is in line with ideas about the role of nurses in care for people of all ages with chronic conditions [46].

Still, it should also be noted that most self-management behavior takes part outside the hospital. A recent thematic synthesis of qualitative studies found that there are several sources for self-management support that serve different purposes [47]. This implies that support should also be available outside the hospital. Interviews with adolescents with cystic fibrosis for instance showed that their family played an important role in providing treatment-related support, while friends formed an important source for relational and emotional support [48]. Another study among young adults with cancer found that peer support is particularly important to achieve 'normality' [49]. *Chapter 8* also suggests that peer support could be effective in dealing with the chronic condition in daily life. This is confirmed in *chapter 9*, where we demonstrated that a peer intervention such as Camp COOL indeed allowed young people to support each other. Support from young adult peers was not only beneficial for adolescent participants, but also for the young adults who had co-organized the camp themselves.

Thus, next to health care professionals, others are able to provide support. Needs assessment therefore also should include the identification of other support sources. Depending on the patient's needs and social environment, health care professionals' support can be more or less intensive. In case of the Skills for Growing Up in pediatric Nephrology tool (*chapter 3*), for example, creating awareness and acting as an 'eye-opener' for parents could in some instances be enough to support families in achieving more autonomy in young people.

Current approaches to self-management support in health care for young people with chronic conditions

The modes and formats, elements, settings and interventionists of the self-management interventions reviewed in *chapter 4* were comparable to those found in a review of self-management interventions for young persons with physical disabilities [50] and in another review of self-management approaches for people with chronic conditions in general [51]. Self-management interventions varied greatly in their modes and formats, elements, settings and interventionists, and none of these characteristics was restricted to the content of interventions. However, the descriptions of modes, formats, elements, settings and interventionists were often limited. Perhaps this might be explained by the type of study design. Most of these studies had a quantitative design including a pre-post measurement, leaving little room for intervention descriptions and the exploration of characteristics of interventions. Others also recognized this and advocated that broader descriptions of the context of interventions be provided in randomized controlled trials [52]. Also, the need for other study designs has been stressed [51].

There seems to be a lack of qualitative evaluation studies for self-management interventions. Since qualitative studies could provide deeper insight into the context of interventions, future studies are recommended to add a qualitative component to the evaluation of interventions, i.e. to employ a mixed-methods evaluation approach. This would also benefit research into specific working mechanisms and contextual factors of self-management interventions, which is needed to answer the question of what works for whom. The next section further reviews the findings related to evaluation of self-management interventions in health care for young people with chronic conditions.

Self-management interventions: Evaluation and effectiveness

Chapter 4 and *chapter 8* raised questions regarding the selection of outcomes to define self-management interventions' impacts. Outcome measures or themes used to evaluate the effectiveness of self-management interventions varied greatly between the reviewed studies and even within the reviewed self-management interventions for specific diagnosis groups and content. It seems that in current evaluation studies the focus on what is aimed at is often lost, leading to the use of many different outcomes that are unrelated to the content of interventions. This hampers conclusions about their effectiveness in meta-analyses. In *chapter 8*, this was one of the explanations for not being able to draw any hard conclusions on effectiveness and effective intervention elements. Other authors also mentioned the uncritical use of outcomes in self-management intervention evaluations and advocate the establishment of a core set of measurement instruments for the evaluation of self-management interventions [50, 53, 54]. Our conceptual measurement framework presented in *chapter 4* is a first attempt towards this. Also, the lack of clarity surrounding the conceptualization of self-management and consequently the

methodological limitations inherent to the designs of current evaluation studies add to the complexity of researching the effectiveness self-management interventions.

METHODOLOGICAL CONSIDERATIONS

The studies described in this thesis considered self-management of young people growing up with chronic conditions. In our exploration of the concept of self-management (*chapter 1*), we did not explicitly study the unique views young people and their parents may have. However, our research builds on results of the On Your Own Feet research program that extensively studied the preferences and views of young people with chronic conditions, and also included parent views [11]. Moreover, we did include young people with chronic kidney disease in the Delphi expert panel to develop the Skills for Growing Up in pediatric Nephrology tool (*chapter 3*). As such, we did consider their views on important aspects of growing up with a chronic condition. Also, young people were interviewed about their experiences during Camp COOL (*chapter 9*).

Our cohort study included a large sample of young persons with a wide range of chronic conditions (*chapters 5, 6, 7*). The sample was heterogeneous in terms of congenital and acquired conditions, and in age. It originates from a large 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. Furthermore, although the response rate at follow-up measurement in 2012 was reasonable for a population of young people (52%), the response rate at base-line measurement was quite low (36%) [11]. Non-response analysis revealed that non-responders at both measurements more frequently had a non-Dutch surname and were males. This indicates a risk for selection bias, and implies that caution is warranted when generalizing our results to a broader context, as these may not be representative for all young people growing up with chronic conditions.

Gaining insight into effectiveness of self-management interventions was hindered by the heterogeneity in outcome measures found in the literature review (*chapter 4*). We could not draw solid conclusions about effectiveness or effective elements of self-management interventions (*chapter 8*). Also, we did not study the effectiveness of the Skills for Growing Up in pediatric Nephrology tool or participants' views on its value (*chapter 3*). In the evaluation of Camp COOL (*chapter 9*), we could say something about effects, but it should be kept in mind that we were not able to include a control group, and we did not directly study long-term effects of this intervention. Therefore, we provided valuable insights for further research into self-management intervention effectiveness rather than conclusive evidence on this topic.

GENERAL DISCUSSION

At the crossroads of medical, role and emotion management, shifting child-parent roles, and other sources of self-management support

Conceptualizing self-management as a dynamic process that requires flexibility in shifting between different content and roles when supporting it, implicates that self-management support takes place on a continuum of strategies that combine diverse content with different roles of all parties involved. Current perspectives on self-management seem to presume certain combinations of these shifts. When looking at the classification of self-management perspectives, the ‘professional discourse’ [2] or narrow view on self-management combines medical management with an expert role for health care professionals. The broader view identified in *part I* links a focus on role and emotion management to the concept of partnership with a power balance between patients and health care professionals (i.e. shared decision-making), while the ‘patient discourse’ perspective [2] does not distinguish between domains and advocates an expert role for patients. However, as patients’ needs determine the nature of self-management support, the question is not whether one perspective is to be preferred over the other, but rather ‘what works for whom and at what stage of the developmental trajectory?’ Although this question applies to the individual level, there are two important points to be made relating to the whole population of young people growing up with a chronic condition.

The first has to do with the dimensions of self-management. Young people growing up with chronic conditions go through the normal developmental stages of adolescence and young adulthood, and have the additional challenge of taking up medical management [11]. The broad view that takes medical, role and emotion management tasks as domains of self-management [1], acknowledges these developmental tasks. This is important, because a chronic illness and its treatment can have manifold effects on different areas of daily life and development, while developmental changes during adolescence reciprocally affect chronic illness and its treatment [55-57]. A recent study, for instance, showed that wanting to be normal and feeling controlled by your condition were perceived barriers for adolescents to adhere to their medical treatment [58]. Young people thus have to balance the usual developmental tasks with the adaptive tasks presented by their chronic condition. In this light, it is essential to note that even though self-management is operationalized in medical, role and emotion domains, these domains are interrelated. Consequently, the core self-management task for young people is to learn how to coordinate the tasks and priorities related to each domain. Although this “articulation work” [59] is not a specific challenge for young people, but for all living with chronic illness, the fact that they are developing – and thus are in the process of acquiring new life skills – makes it more challenging. Think of John – the sixteen-year-old boy with chronic kidney disease in the story in the introduction chapter – who wanted

to go out with his friends (and thus fulfill his task of being part of a social group), but who also had to get his treatment done (and thus fulfill his medical management task). At first, he got angry, because he obviously preferred the first over the second. Later, he seemed to have found a way to deal with both: he underwent his treatment right away, playing a computer game with a friend, and planned to go out later that week. Professionals providing self-management support for young people with chronic conditions should take this extra assignment of 'coordination of tasks' into account by exploring patients' needs at the crossroads of medical, role and emotion management.

This brings us to the second point. While, or even before, actually engaging in "articulation work" [59], young people have to gain insight into the tasks related to medical, role and emotion management and have to learn how to perform such tasks. They also have to define their own preferences and priorities in life. The second point thus relates to the patient role of the young person. As mentioned before, self-management is not only linked to the individual, but is formed in and influenced by the individual's social context. This means that significant others will take part in self-management, notably the parents or guardians. Parents often fulfill part of the young person's 'patient role' by performing medical management tasks for their children [60] or assisting them in these tasks. A systematic review of the experiences of parents of children with chronic kidney disease found that these parents performed additional tasks such as forming partnerships with health care providers, seeking information and organizing transportation and finances [61]. In the case of John, for instance, his father helped him to set up the dialysis machine. However, while growing up, these young people are expected to gradually take over these tasks in order to perform their full 'patient role' and accompanying responsibilities, while parents should support this acquisition of autonomy in their children and cede control [62-64]. This role change is not always self-evident [65]. Moreover, young people and their parents may have different perspectives, for instance on quality of life [66] or health care, but also on the possibilities for and desirability of full social participation such as living independently, leisure activities and (intimate) relationships [67]. Self-management support for these young people should therefore also focus on the gradual shifts in tasks, and include support for both young people and their parents. In diabetes care, there has been some evidence that including parents in psychosocial interventions for young people enhanced effectiveness of the interventions [68]. Also, different studies showed that parents of young people need support during the process of 'letting go' [64, 65, 67, 69].

Peers play an increasingly important role in young peoples' daily lives. John, for instance, had a peer, Ron, who happened to get his treatment at the same time and invited him to an online game. As such, Ron relieved John's sense of isolation. Olsson and colleagues mention nine psychosocial mechanisms of peer support among young people with chronic illness: "learning new coping techniques, learning how to influence social environments, enlarging perspectives on what is normal, examining alternative

perspectives, understanding the causes of personal stressors, confirmation of positive changes in attitudes, reduced sense of isolation, enhanced social identity through group approval, and extending help to others” [70]. Through these mechanisms, peer support may help to deal with a chronic condition in daily life and to orient to adult roles. *Chapter 9* for instance showed that peer support is indeed effective in enhancing young people’s feeling of being independent. Orienting towards adult roles in other contexts may also influence the way young people participate in health care. This idea fits with the Positive Youth Development (PYD) perspective that promotes the development of generic attributes like confidence and social connection, and suggests that having such attributes will lead to positive outcomes like improved wellbeing and better self-care [71]. As such, peer support may be an important ‘out of the health care’ source of self-management support for young people growing up with chronic conditions.

While supporting self-management or the development of self-management of young people with chronic conditions, health care professionals thus should pay attention to medical, role and emotion management, shifting child-parent roles, other sources of self-management support (social context), and articulation work (i.e., the interaction between the domains and the coordination of tasks in the social network); as presented in Figure 2. The next section discusses what this implies for current health care practices and health care professionals.



Figure 2 Self-management support for young people

Self-management support in health care practice for young people with chronic conditions

Young people's support needs are not limited to the health care context, stressing the importance of holistic support for young people growing up with chronic conditions. Widening the range of self-management support in health care implies a huge task for professionals, which not everyone is able to take up individually. Self-management support in the health care context is rather a multidisciplinary task for teams of health care professionals. One professional may then be appointed as the prime provider of self-management support. In case of the *Skills for Growing Up in pediatric Nephrology* (chapter 3), this was the nurse. Others have also proposed the nurse as the person to take up the task of self-management support [72-74], but it is also acknowledged that she needs to include other providers in self-management support [4, 75].

Yet, when care is not provided in multidisciplinary teams, it becomes more complicated to provide holistic self-management support. A recent study showed that in 44% of the outpatient clinics in a children's hospital in the Netherlands no regular multidisciplinary team meetings were held. Also, nurses were not always involved in care provision in outpatient clinics [76]. Perhaps in such cases, professionals could point towards other sources of self-management support, such as Camp COOL (chapter 9) or other community programs. Furthermore, in 56% of these clinics no team meetings were planned to discuss transfers and transitional care with the adult care staff [76]. Still, the fact that little is currently known about the optimal timing of transitions and needed assistance [77] calls for holistic attention all the way through pediatric, adolescent and adult care. Most attention for self-management of young people with chronic conditions is now centered in pediatric and adolescent care, and the need to engage adult care providers in transitional care has only been highlighted recently [11, 34]. Since young adults with chronic conditions form a unique group in health care, adult care providers may benefit from more training in providing self-management support and the use of interventions in this group of patients. In our review (chapter 4) we found that only 6% of the self-management interventions were aimed at young people over the age of 18 years.

Still, this does not imply that health care providers in current pediatric and adolescent care are already fully equipped and ready to provide holistic self-management support to young people. Although they are more familiar with the developmental needs of young people growing up with chronic conditions and initiatives have been taken to widen their repertoire of care, there is a lack of clear intervention or program descriptions, and little is known about the development, focus and effectiveness, and the experiences of young people with such programs [11, 78-80]. Thus, while there should be paid more attention to the specific needs of young people growing up with chronic conditions in adult care, it is also essential to gain more insight into good practices of self-management support of this group of patients during their transitions. The next

section discusses the implications of our findings for self-management interventions for young people with chronic conditions.

Approaches to self-management support for young people with chronic conditions

From the holistic view, the ultimate goal of self-management is maintaining “a satisfactory quality of life” [51]. As a consequence, and as shown in *chapter 4*, HRQoL is increasingly used as a patient-reported outcome measure in the evaluation of self-management interventions for young people. Although self-management is assumed to contribute to HRQoL, it is neither mentioned nor explored in the most commonly used HRQoL models or definitions [81]. To date, several studies have explored the relationship between elements of self-management and domains of HRQoL. For instance, between self-efficacy or social participation and HRQoL [82-84]. In *chapter 7*, relationships between the domains of self-management and those of HRQoL were studied, and the suggestion that self-management domains are interrelated was confirmed. Moreover, finding shared associations of the self-management domains with the domains of HRQoL again emphasized that attention is warranted for young people’s psychosocial needs.

Despite the finding that HRQoL and self-management domains are associated with each other, one could question the claim of ‘a satisfactory HRQoL’ as the ultimate goal or outcome of self-management. *Chapter 7* showed a general decrease in HRQoL of young people with chronic conditions after their transition to adulthood and adult care. Additionally, *chapter 6* showed that an increase in HRQoL from adolescence to young adulthood was associated with better transfer experiences and more satisfaction with the transfer. However, at the same time, *chapter 5* revealed that young adults with a higher level of autonomy in social participation did not necessarily have higher HRQoL, but did report higher self-efficacy and independence at both measurements (and vice versa). The inability of the quantitative measurement of HRQoL to fully apprehend the experience of living with a chronic conditions has been highlighted already [11]. To really understand what it means to grow up with a chronic condition and what the personal expectations about life transitions are, it is essential to gain insight into young people’s lived experiences. The measurement of HRQoL may assist in this, for instance by regularly using HRQoL reports to facilitate the discussion of psychosocial aspects during medical consultations [85]. This has proven to be effective in enhancing communication about psychosocial issues in care for children with juvenile idiopathic arthritis [86]. The monitoring of HRQoL as such may help health care professionals to gain insight into the person’s self-management needs (i.e., dealing with a medical condition and its treatment, lived experiences, and societal roles).

As for the subsequent task to enable people to self-manage, our findings suggest that self-efficacy is an important determinant to aim at. This is in line with social learn-

ing theory that argues that people learn from others, and in general aim at enhancing self-efficacy [87]. The review of self-management interventions for young people with chronic conditions (*chapter 4*) showed that most of the interventions for which a theoretical base was mentioned were based on social learning theory. In this theory, self-management support facilitates environments that allow ‘learning from others’ and gaining ‘mastery experiences’. As mentioned earlier, these environments can be both in and outside the health care setting. Furthermore, it is important to empower independent behavior of young people during consultations, and to pay attention to transition readiness. However, no hard conclusions could be drawn considering the question which approaches to self-management support are best for whom. Therefore, more research on the content of self-management interventions and their effectiveness is needed. The next sections discuss the implications of our findings for future practice and research of self-management interventions.

A non-categorical approach to self-management and self-management support

Over thirty years ago, Stein and Jessop already pleaded for a non-categorical approach to chronic illness in pediatric care [88]. The core of this approach is the notion that young people growing up with different chronic conditions face similar adaptive challenges regardless of type of condition. In this respect, young people within a specific diagnostic group may differ as much as those in different diagnostic groups [11, 88]. More recently, Modi and colleagues emphasized the usefulness of a generic approach and the need to consider adaptive tasks in self-management of young people with chronic conditions [89]. Remarkably, only 7% of the self-management interventions found in the systematic literature review (*chapters 4 and 8*) were developed for or applied to all kinds of chronic conditions.

A non-categorical approach to self-management support may be beneficial for both research and practice. It could overcome the problem of achieving effectiveness and cost-effectiveness of disease-specific self-management interventions because specific pediatric or young adult diagnostic groups are often small [11, 90]. It is for instance important to consider the question of disease-specific versus general measurement instruments when evaluating self-management interventions. Apart from asthma and diabetes, the numbers of studies per chronic condition were small in our review study. If these studies use disease-specific instruments to evaluate effectiveness of the intervention, it remains hard to research the effectiveness of self-management support for young people within their study population. Moreover, a non-categorical approach allows professionals of different disciplines to learn from each other and would prevent the wheel from being invented everywhere. This advantage was evident in our study

with the *Skills for Growing Up* tool that was developed in rehabilitation care before being adapted for use in nephrology (*chapter 3*).

However, this is not to say that disease-specific characteristics do not matter at all. Interventions should include disease-specific content. The content of the *Skills for Growing Up* tool for instance had to be adapted to enable medical management in young people with a chronic kidney disease. The systematic overview of self-management interventions for young people with chronic conditions (*chapter 4*) also showed that much of the medical content of interventions consists of disease-specific aspects like understanding medication, treatment regimen, and side effects, or using specific treatment devices or techniques (e.g. peak flow meter for asthma). Since the tasks associated with these aspects are also part of self-management, they are indeed important to consider. Also, they could alter the articulation tasks of young people as these tasks have to be aligned with other aspects of their lives. Therefore, rather than employing a solely generic focus, a non-categorical approach to self-management support of young people growing up with chronic conditions should be complemented by disease-specific considerations. Interventions or combinations of interventions should address more general and disease-specific self-management tasks. Research studies are recommended to use disease-specific measurement instruments if their intervention intervenes at disease-specific medical management, but to use general measurement instruments if the intervention intervenes at general medical management or other domains of self-management.

A measurement framework to research self-management interventions' effectiveness

In *chapter 4* we constructed a framework for measuring self-management by categorizing sets of currently used outcomes per domain of self-management. The outcomes of the reviewed studies matched those found in comparable reviews of self-management interventions [50, 54, 91, 92], which strengthens the validity of the framework. It may be used as a selection tool to define outcome measures based on the content of the intervention(s). However, it is a global classification and decisions have to be made about what measurement outcomes and measurement instruments would be appropriate. The heterogeneity in the measurement instruments used in the reviewed studies did not allow for the formulation of a core set of instruments. More studies into valid measurement instruments for self-management interventions for young people with chronic conditions are needed to further sharpen the selection tool. Besides, more systematic approaches to intervention evaluation are needed. Such an evaluation approach could globally include the four steps presented in Figure 3.

Intervention mapping is an example of the application of such stepwise approach. Intervention mapping is usually concerned with the development of interventions, but

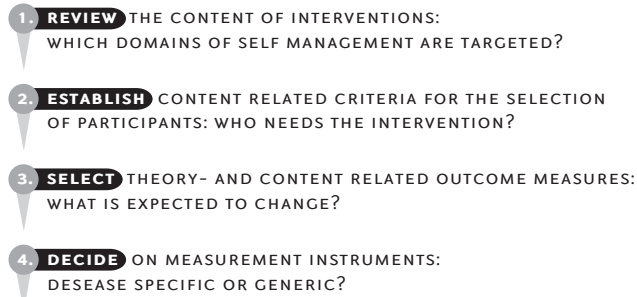


Figure 3 Global steps in the evaluation of self-management interventions

may also be used for their evaluation [93]. An advantage of an intervention mapping approach is that it allows for explicit consideration of the theoretical bases of interventions [94]. *Chapter 4* showed that most of current intervention studies do not clarify interventions' underlying theoretical principles. Yet it is important to consider these, because theories inform the assumptions about what is expected to change or what may be working elements of interventions.

Nevertheless, a measurement framework as presented in *chapter 4* may never be able to seize the full breadth of self-management support outcomes. This is the case, first, because outcomes of self-management interventions could be unexpected. In Camp COOL for instance (*chapter 9*), participants gained more knowledge about the disease, despite the fact that the content, i.e. the day-to-day camping program, purposely did not include disease-specific elements. Interventions can thus have multiple and heterogeneous effects which cannot all be foreseen. Another reason is that the core self-management task of young people to learn how to coordinate the tasks and priorities related to each domain of self-management, i.e. their 'articulation work', is not captured by simply combining domain-related outcomes. These issues may be circumvented perhaps by employing a mixed-methods design with a quantitative part and a qualitative part. In such a design, the elements and outcomes of self-management support, e.g. the roles of involved parties and outcomes specifically related to medical, role and emotion management can then be studied quantitatively while the qualitative part addresses the shifting between roles of involved parties and dimensions of self-management, as well as the influence of the intervention on these aspects. As such, it may provide opportunities to explore possible unexpected outcomes and experiences, e.g. young persons' articulation tasks, role balancing between young people and their parents, and other sources of support.

LESSONS AND FUTURE DIRECTIONS

Implications for clinical practice

Multidisciplinary team work

Nurses have a leading role in exploring patients' self-management support needs, but holistic support for young people growing up with chronic conditions should ideally be provided in multidisciplinary teams. It could also be beneficial to identify other sources of support outside the health care context. Peers are an important source of social support.

Non-categorical approach to self-management

A non-categorical approach to self-management support could allow professionals from different disciplines to learn from each other. For example in the form of thematic group meetings in which knowledge and experiences are exchanged.

Building bridges between pediatric and adult care

Self-management support for young people growing up and living with chronic conditions should be specific and should be provided all the way from pediatric into adult care. It is essential to build bridges between pediatric and adult care, for instance in the form of joint transition clinics.

Gaining insight into support needs

Young people's needs should be explored at the crossroads of medical, role and emotion management; therefore attention should be paid to medical as well as psychosocial needs. HRQoL monitoring can give insight into psychosocial functioning and needs, and enable conversations about this.

A shift in responsibility

Professionals should encourage the gradual shift in responsibility from parents to young persons. Young people should be activated or empowered during medical visits, for instance by asking the parents not to be present for part of the consultation or by formulating individual transition plans together with the young person. It is important to consider the role of parents since they may also need support.

The nature of self-management interventions

Self-management interventions should be tailored to individual needs. There is little insight into good practices, but self-efficacy seems to be an important determinant of

self-management. Therefore, the use of interventions that create a learning environment and aim to enhance self-efficacy is recommended.

Future directions for research

Timing of transitions and support needs

Little is known about the timing of transitions and support needs of young people growing up with chronic conditions. More longitudinal research is needed to understand how self-management of people with chronic conditions develops over the life span.

Non-categorical approach to intervention evaluation

In small specific pediatric or young adult diagnostic groups, a non-categorical approach to intervention evaluation could overcome the problem of achieving effectiveness and cost-effectiveness of disease-specific self-management interventions.

Systematic approach to intervention evaluation

More systematic approaches with respect to the evaluation of self-management interventions are needed. Intervention mapping is an example of a stepwise approach that could be applied to evaluation as well as intervention design.

Mixed methods research of interventions

When researching effectiveness of self-management interventions, it is important to pay attention to the contexts in which interventions are provided, as well as participants' social context and articulation tasks. Detailed information on interventions and young people's experiences with interventions is valuable, too. Therefore, a mixed methods research approach combining qualitative and quantitative research is recommended for the evaluation of self-management interventions.

KEY MESSAGES

- Self-management is a dynamic process whose support requires flexibility in adapting to different content and roles. As a consequence, self-management support takes place on a continuum of strategies that combine diverse content with different roles of all parties involved.
- Self-management support for young people with chronic conditions should go beyond medical management. It includes attention for medical, role and emotion management, shifting child-parent roles, and young people's articulation work.

- Rather than employing a solely generic focus, a non-categorical approach to self-management support of young people growing up with chronic conditions should be complemented by disease-specific considerations.
- If possible, a nurse may be appointed as the prime provider of self-management support, but other providers also should be included in this task. Also, it is important to consider other sources of self-management support – outside the health care context.
- Monitoring of HRQoL may help health care professionals to understand what self-management tasks could be problematic.
- As for the subsequent task to enable people to self-manage, self-efficacy is an important determinant to aim at. Self-management support thus should facilitate environments that allow 'learning from others' and gaining 'mastery experiences'. Also, it is important to empower independent behavior of young people during consultations, and to pay attention to transition readiness.
- More attention should be given to the specific needs of young people with chronic conditions in adult care, and there is a need for studies to research transition 'beyond pediatrics'.
- It is also essential to gain more insight into good practices of self-management support for young people with chronic conditions during their transitions, in both pediatric and adult care.
- Studies evaluating self-management interventions should employ a mixed-methods design, and give more detailed descriptions of interventions and contexts.

REFERENCES

1. Lorig KR, Holman H: Self-management education: history, definition, outcomes, and mechanisms. *Ann Behav Med* 2003, 26(1):1-7.
2. Kendall E, Ehrlich C, Sunderland N, Muenchberger H, Rushton C: Self-managing versus self-management: reinvigorating the socio-political dimensions of self-management. *Chronic illn* 2011, 7(1):87-98.
3. 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.
4. van Hooft SM, Dwarswaard J, Jedeloo S, Bal R, van Staa A: Four perspectives on self-management support by nurses for people with chronic conditions: A Q-methodological study. *Int J Nurs Stud* 2015, 52(1):157-166.
5. Audulv A: The over time development of chronic illness self-management patterns: a longitudinal qualitative study. *BMC public health* 2013, 13:452.
6. Paterson BL: The shifting perspectives model of chronic illness. *J Nurs Scholarsh* 2001, 33(1):21-26.
7. van Houtum L, Rijken M, Heijmans M, Groenewegen P: Self-management support needs of patients with chronic illness: do needs for support differ according to the course of illness? *Patient Educ Couns* 2013, 93(3):626-632.
8. Rijken M, Heijmans M, Jansen D, Rademakers J: Developments in patient activation of people with chronic illness and the impact of changes in self-reported health: results of a nationwide longitudinal study in The Netherlands. *Patient Educ Couns* 2014, 97(3):383-390.
9. Richard AA, Shea K: Delineation of self-care and associated concepts. *J Nurs Scholarsh* 2011, 43(3):255-264.
10. Thirsk LM, Clark AM: What is the 'self' in chronic disease self-management? *Int J Nurs Stud* 2014, 51(5):691-693.
11. van Staa AL: On Your Own Feet. Preferences and competencies for care of adolescents with chronic conditions. Rotterdam: Hogeschool Rotterdam; 2012.
12. 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.
13. Roebroek 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.
14. Maslow GR, Haydon A, McRee AL, Ford CA, Halpern CT: Growing up with a chronic illness: social success, educational/vocational distress. *J Adolesc Health* 2011, 49(2):206-212.
15. Maslow GR, Haydon AA, Ford CA, Halpern CT: Young adult outcomes of children growing up with chronic illness: an analysis of the National Longitudinal Study of Adolescent Health. *Arch Pediatr Adolesc Med* 2011, 165(3):256-261.
16. 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.
17. Monaghan M, Hilliard M, Sweenie R, Riekert K: Transition readiness in adolescents and emerging adults with diabetes: the role of patient-provider communication. *Curr Diab Rep* 2013, 13(6):900-908.

18. Price CS, Corbett S, Lewis-Barned N, Morgan J, Oliver LE, Dovey-Pearce G: Implementing a transition pathway in diabetes: a qualitative study of the experiences and suggestions of young people with diabetes. *Child Care Health Dev* 2011, 37(6):852-860.
19. Cook K, Siden H, Jack S, Thabane L, Browne G: Up against the System: A Case Study of Young Adult Perspectives Transitioning from Pediatric Palliative Care. *Nurs Res Pract* 2013, 2013:286751.
20. Opic P, Roos-Hesselink JW, Cuypers JA, Witsenburg M, van den Bosch A, van Domburg RT, Bogers AJ, Utens EM: Psychosocial functioning of adults with congenital heart disease: outcomes of a 30-43 year longitudinal follow-up. *Clin Res Cardiol* 2014.
21. Opic P, Utens EM, Ruys TP, van Domburg RT, Witsenburg M, Bogers AJ, Roos-Hesselink JW: Long-term psychosocial outcome of adults with tetralogy of Fallot and transposition of the great arteries: a historical comparison. *Cardiol Young* 2014, 24(4):593-604.
22. Sawicki GS, Lukens-Bull K, Yin X, Demars N, Huang IC, Livingood W, Reiss J, Wood D: 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.
23. Stinson J, Kohut SA, Spiegel L, White M, Gill N, Colbourne G, Sigurdson S, Duffy KW, Tucker L, Stringer E *et al*: A systematic review of transition readiness and transfer satisfaction measures for adolescents with chronic illness. *Int J Adolesc Med Health* 2014, 26(2):159-174.
24. 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.
25. van Staa A: 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.
26. Hargreaves DS: Revised You're Welcome criteria and future developments in adolescent health-care. *J Clin Res Pediatr Endocrinol* 2011, 3(2):43-50.
27. Committee on Adolescence American Academy of Pediatrics: Achieving quality health services for adolescents. *Pediatrics* 2008, 121(6):1263-1270.
28. Wilkinson D, Robinson AJ: You're Welcome quality criteria: making health services young-people-friendly. *Sex Transm Infect* 2009, 85(3):233-234.
29. Betz CL, Smith K, Macias K: Testing the transition preparation training program: A randomized controlled trial. *Int J Child Adolesc Health* 2010, 3(4):595-607.
30. 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.
31. Fegran L, Hall EO, Uhrenfeldt L, Aagaard H, Ludvigsen MS: Adolescents' and young adults' transition experiences when transferring from paediatric to adult care: a qualitative metasynthesis. *Int J Nurs Stud* 2014, 51(1):123-135.
32. Betz CL, Lobo ML, Nehring WM, Bui K: Voices not heard: a systematic review of adolescents' and emerging adults' perspectives of health care transition. *Nurs Outlook* 2013, 61(5):311-336.
33. Lugasi T, Achille M, Stevenson M: Patients' perspective on factors that facilitate transition from child-centered to adult-centered health care: a theory integrated metasummary of quantitative and qualitative studies. *J Adolesc Health* 2011, 48(5):429-440.
34. Gleeson H, McCartney S, Lidstone V: 'Everybody's business': transition and the role of adult physicians. *Clin Med (London)* 2012, 12(6):561-566.
35. Hilderson D, Eyckmans L, Van der Elst K, Westhovens R, Wouters C, Moons P: Transfer from paediatric rheumatology to the adult rheumatology setting: experiences and expectations of young adults with juvenile idiopathic arthritis. *Clin Rheumatol* 2013, 32(5):575-583.

36. Neinstein LS, Irwin CE, Jr.: Young adults remain worse off than adolescents. *J Adolesc Health* 2013, 53(5):559-561.
37. Austin S, Guay F, Senecal C, Fernet C, Nouwen A: Longitudinal testing of a dietary self-care motivational model in adolescents with diabetes. *J Psychosom Res* 2013, 75(2):153-159.
38. Berg CA, King PS, Butler JM, Pham P, Palmer D, Wiebe DJ: Parental involvement and adolescents' diabetes management: the mediating role of self-efficacy and externalizing and internalizing behaviors. *J Pediatr Psychol* 2011, 36(3):329-339.
39. Chih AH, Jan CF, Shu SG, Lue BH: Self-efficacy affects blood sugar control among adolescents with type I diabetes mellitus. *J Formos Med Assoc* 2010, 109(7):503-510.
40. Gandhi PK, Kenzik KM, Thompson LA, DeWalt DA, Revicki DA, Shenkman EA, Huang IC: Exploring factors influencing asthma control and asthma-specific health-related quality of life among children. *Respir Res* 2013, 14:26.
41. Grieve AJ, Tluczek A, Racine-Gilles CN, Laxova A, Albers CA, Farrell PM: Associations between academic achievement and psychosocial variables in adolescents with cystic fibrosis. *J Sch Health* 2011, 81(11):713-720.
42. Schwartz LA, Drotar D: Health-related hindrance of personal goal pursuit and well-being of young adults with cystic fibrosis, pediatric cancer survivors, and peers without a history of chronic illness. *J Pediatr Psychol* 2009, 34(9):954-965.
43. Whittemore R, Jaser S, Guo J, Grey M: A conceptual model of childhood adaptation to type 1 diabetes. *Nurs Outlook* 2010, 58(5):242-251.
44. Law GU, Walsh J, Queralto V, Nouwen A: Adolescent and parent diabetes distress in type 1 diabetes: the role of self-efficacy, perceived consequences, family responsibility and adolescent-parent discrepancies. *J Psychosom Res* 2013, 74(4):334-339.
45. Cramm JM, Strating MM, Nieboer AP: The Importance of General Self-Efficacy for the Quality of Life of Adolescents with Diabetes or Juvenile Rheumatoid Arthritis Over Time: A Longitudinal Study among Adolescents and Parents. *Front Pediatr* 2013, 1:40.
46. Alleyne G, Hancock C, Hughes P: Chronic and non-communicable diseases: a critical challenge for nurses globally. *Int Nurs Rev* 2011, 58(3):328-331.
47. Dwarswaard J, Bakker EJM, Van Staa AL, Boeije HR: Self-management support from the perspective of patients with a chronic condition: a thematic synthesis of qualitative studies. *Health Expect* 2015 (Accepted for publication).
48. Barker DH, Driscoll KA, Modi AC, Light MJ, Quittner AL: Supporting cystic fibrosis disease management during adolescence: the role of family and friends. *Child Care Health Dev* 2012, 38(4):497-504.
49. David CL, Williamson K, Tilsley DW: A small scale, qualitative focus group to investigate the psychosocial support needs of teenage young adult cancer patients undergoing radiotherapy in Wales. *Eur J Oncol Nurs* 2012, 16(4):375-379.
50. Lindsay S, Kingsnorth S, McDougall C, Keating H: A systematic review of self-management interventions for children and youth with physical disabilities. *Disabil Rehabil* 2014, 36:276-288.
51. 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.
52. Langstrup H, Ross Winthereik B: The Making of Self-Monitoring Asthma Patients: Mending a Split Reality with Comparative Ethnography. *Comp Sociol* 2008, 7(3):362-386.
53. Nolte S, Elsworth GR, Newman S, Osborne RH: Measurement issues in the evaluation of chronic disease self-management programs. *Qual Life Res* 2013, 22(7):1655-1664.
54. Nolte S, Osborne RH: A systematic review of outcomes of chronic disease self-management interventions. *Qual Life Res* 2013, 22(7):1805-1816.

55. Suris JC, Michaud PA, Viner R: The adolescent with a chronic condition. Part I: developmental issues. *Archives of disease in childhood* 2004, 89(10):938-942.
56. Yeo M, Sawyer S: Chronic illness and disability. *BMJ* 2005, 330(7493):721-723.
57. Sawyer SM, Drew S, Yeo MS, Britto MT: Adolescents with a chronic condition: challenges living, challenges treating. *Lancet* 2007, 369(9571):1481-1489.
58. Hanghoj S, Boisen KA: Self-reported barriers to medication adherence among chronically ill adolescents: a systematic review. *J Adolesc Health* 2014, 54(2):121-138.
59. Corbin JM, Strauss AL: Unending work and care: managing chronic illness at home: Jossey-Bass Publishers; 1988.
60. Smith J, Cheater F, Bekker H: Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature. *Health Expect* 2013, Jan 14 [Epub ahead of print].
61. Tong A, Lowe A, Sainsbury P, Craig JC: Experiences of parents who have children with chronic kidney disease: a systematic review of qualitative studies. *Pediatrics* 2008, 121(2):349-360.
62. Gall C, Kingsnorth S, Healy H: Growing up ready: a shared management approach. *Phys Occup Ther Pediatr* 2006, 26(4):47-62.
63. Kirk S: Transitions in the lives of young people with complex healthcare needs. *Child Care Health Dev* 2008, 34(5):567-575.
64. Akre C, Suris JC: From controlling to letting go: what are the psychosocial needs of parents of adolescents with a chronic illness? *Health Educ Res* 2014, 29(5):764-772.
65. Williams B, Mukhopadhyay S, Dowell J, Coyle J: From child to adult: an exploration of shifting family roles and responsibilities in managing physiotherapy for cystic fibrosis. *Soc Sci Med* 2007, 65(10):2135-2146.
66. Sattoe JNT, van Staa A, Moll HA: The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. *Health Qual Life Outcomes* 2012, 10:10.
67. Peeters MA, Hilberink SR, van Staa A: The road to independence: lived experiences of youth with chronic conditions and their parents compared. *J Pediatr Rehabil Med* 2014, 7(1):33-42.
68. Murphy HR, Wadham C, Rayman G, Skinner TC: Approaches to integrating paediatric diabetes care and structured education: experiences from the Families, Adolescents, and Children's Team-work Study (FACTS). *Diabetic Med* 2007, 24(11):1261-1268.
69. Meah A, Callery P, Milnes L, Rogers S: Thinking 'taller': sharing responsibility in the everyday lives of children with asthma. *J Clin Nurs* 2010, 19(13-14):1952-1959.
70. Olsson CA, Boyce MF, Toumbourou JW, Sawyer SM: The Role of Peer Support in Facilitating Psychosocial Adjustment to Chronic Illness in Adolescence. *Clin Child Psychol Psychiatry* 2005, 10(1):78-87.
71. Maslow GR, Chung RJ: Systematic review of positive youth development programs for adolescents with chronic illness. *Pediatrics* 2013, 131(5):e1605-1618.
72. Meleis AI, Sawyer LM, Im EO, Hilfinger Messias DK, Schumacher K: Experiencing transitions: an emerging middle-range theory. *ANS Adv Nurs Sci* 2000, 23(1):12-28.
73. Goossens E, Hilderson D, Moons P: Coaching through transition: a challenge for critical care nurses. *Aust Crit Care* 2011, 25(1):1-2.
74. Betz CL: Health care transition for adolescents with special healthcare needs: where is nursing? *Nurs Outlook* 2013, 61(5):258-265.
75. Betz CL: Health care transitions: a peek into the future. *J Pediatr Nurs* 2012, 27(1):1-2.
76. Van Hooft SM, Van der Knaap L, Latour JM, Jedeloo S, Sattoe JNT, Van Staa AL: Wat zijn de huidige werkwijzen rondom de ondersteuning van zelfmanagement in de zorg voor jongeren met een

- chronische aandoening binnen het Erasmus MC-Sophia. In. Rotterdam: Hogeschool Rotterdam, Kenniscentrum Zorginnovatie; 2012.
77. Sawyer SM, Ambresin AE: Successful transitions: beyond disease control to better life chances. *J Adolesc Health* 2014, 54(4):365-366.
78. 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.
79. Bloom SR, Kuhlthau K, Van Cleave J, Knapp AA, Newacheck P, Perrin JM: Health care transition for youth with special health care needs. *J Adolesc Health* 2012, 51(3):213-219.
80. Davis AM, Brown RF, Taylor JL, Epstein RA, McPheeters ML: Transition care for children with special health care needs. *Pediatrics* 2014, 134(5):900-908.
81. 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.
82. Bent N, Jones A, Molloy I, Chamberlain MA, Tennant A: Factors determining participation in young adults with a physical disability: a pilot study. *Clin Rehabil* 2001, 15(5):552-561.
83. van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, Wensink-Boonstra AE, Stam HJ, Roebroek 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.
84. Cramm JM, Strating MM, Roebroek ME, Nieboer AP: The Importance of General Self-Efficacy for the Quality of Life of Adolescents with Chronic Conditions. *Soc Indic Res* 2013, 113(1):551-561.
85. 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.
86. Haverman L, van Rossum MA, van Veenendaal M, van den Berg JM, Dolman KM, Swart J, Kuijpers TW, Grootenhuis MA: Effectiveness of a web-based application to monitor health-related quality of life. *Pediatrics* 2013, 131(2):e533-543.
87. Bandura A: Social Learning Theory. New York: General Learning Press; 1977.
88. 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.
89. Modi AC, Pai AL, Hommel KA, Hood KK, Cortina S, Hilliard ME, Guilfoyle SM, Gray WN, Drotar D: Pediatric self-management: a framework for research, practice, and policy. *Pediatrics* 2012, 129(2):e473-485.
90. Newman S, Mulligan K, Steed L: What is meant by self-management and how can its efficacy be established? *Rheumatology (Oxford)* 2001, 40(1):1-4.
91. Du S, Yuan C: Evaluation of patient self-management outcomes in health care: a systematic review. *Int Nurs Rev* 2010, 57(2):159-167.
92. Kirk S, Beatty S, Callery P, Gellatly J, Milnes L, Prymachuk S: The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child Care Health Dev* 2012, 39(3):305-324.
93. Bartholomew LK, Parcel GS, Kok G, Gottlieb NH, Fernandez ME: Planning Health Promotion Programs: An Intervention Mapping Approach, vol. 3rd edition. San Francisco: Jossey-Bass; 2011.
94. Michie S, Prestwich A: Are interventions theory-based? Development of a theory coding scheme. *Health Psychol* 2010, 29(1):1-8.

Summary



Young people form an increasingly important group in current healthcare. As a result of improved survival, more children grow up with a chronic disease, and more of them reach adulthood than ever before. Becoming an adult often proves extra challenging for those who grow up with chronic conditions, because the adaptive tasks related to living with a chronic condition can clash with normal developmental milestones. Finding a good balance and integrating these tasks in daily life is also referred to as 'self-management'. Taking up self-management is no easy task; appropriate support can be of good use to young people growing up with a chronic condition. For this reason, there is an increasingly stronger emphasis on the importance of self-management in chronic care.

Still, a definition in which self-management is regarded as more than just managing a chronic condition and the medical treatment thereof has only recently been recognized in the literature. A broader definition that accounts for normal developmental tasks and psychosocial functioning seems particularly relevant for young people. Such a definition establishes three domains related to self-management: medical management (concerning the condition and treatment thereof), role management (concerning social participation and roles) and emotion management (concerning emotional well-being). Not much is known about how self-management is embedded in the current care practice for young people. There is also little insight into factors that affect their self-management, and into effective interventions for targeted self-management support. These were reasons to start the research described in this thesis.

The studies described in this thesis were conducted in the framework of the research program 'Self-management & Participation Innovation Lab' (SPIL). Started in 2011, this four year research program aims to improve self-management and self-management support for young people with chronic conditions. SPIL is a continuation of the 'On Your Own Feet' research program. Some of the studies were conducted in the Erasmus MC – Sophia Children's Hospital, others in different academic hospitals in the Netherlands. More information concerning the research programs and projects can be found at: www.opeigenbenen.nu.

The thesis consists of three parts. The first part explores the concept of self-management and self-management support for young people with chronic conditions (*chapters 2, 3 and 4*). The second part addresses young people's transitions to adulthood and to adult care, and explores the development of self-management and its influencing factors (*chapters 5, 6 and 7*). The last part describes the methods by which self-management interventions are evaluated as well as the current knowledge about the effects of current interventions (*chapters 8 and 9*).

Chapter 2 describes a Delphi study into researchers' and policy advisors' visions of self-management in general. Like in the literature, there was ambiguity concerning the

concept of self-management within the group of experts. However, in this case there was a tendency to relate self-management more to person-centered concepts and less to medical management. Although healthcare providers should be discouraged to limit self-management to medical management, it is important that researchers and policy advisors understand that medical treatment is an essential part of daily life with a chronic disease. Self-management support begins with acquiring insight into individual needs arising from a person's adaptive tasks (not only in the medical domain, but also in the role and emotional domains). These needs determine the focus of the self-management support. To stimulate the development of evidence-based interventions and to inform (future) policy, researchers and policy advisors are advised to be more explicit about the needs that self-management support targets, and thus the goals to be achieved.

Chapter 3 studies the first experiences of healthcare providers with the development and implementation of a self-management intervention – the *Skills for Growing Up tool* – for young people with chronic kidney disease. The *Skills for Growing Up tool* was developed together with healthcare providers, adolescents, their parents and a number of experts. The tool is meant to stimulate a 'normal' development of autonomy and independence. All parties reached consensus about the life domains addressed by the tool. These domains contained items about emotion management, social participation (role management) and medical management of chronic kidney diseases. The healthcare providers used and valued the tool, but indicated that digitalizing and making it available online was important to make it feasible. They also needed to get used to the new method of working, which required a more passive role, while young people and their parents had the responsibility of (partly) deciding the course of the conversation and coming up with action plans. Healthcare providers in pediatric nephrology are challenged to look beyond medical management, and to keep an eye on the developmental tasks of young people while they are guided to adulthood. Insight into the experiences and needs of young people is essential for good self-management support.

Chapter 4 reports on the findings of a systematic literature review of self-management interventions for children and young people (7-25 years). The goal of the review was to gain insight into the characteristics, content, underlying theories and expected outcomes of these interventions. Most interventions are offered on a group level and are oriented towards education and/or enhancing competencies. A quarter of the interventions were meant for both young people and their parents. There was a diverse range of settings in which interventions were applied, but they were mostly implemented in outpatient clinics. Many different professionals were involved in implementation. The content of a number of interventions was related to all three domains of self-management, but the majority was oriented towards medical management only. Medical, role

and emotion management were not related to specific intervention characteristics (like type of healthcare provider, setting, format), but the content was adjusted to the age of the group to which the intervention was offered. A large portion of the studies did not report the theoretical backgrounds of the interventions. The most frequently named theories were the social learning theory and cognitive behavioral theory. Expected outcomes were very diverse; most common were clinical and healthcare outcomes, quality of life and knowledge of the condition and/or treatment.

From the literature review it can be concluded that the current self-management interventions address the self-management skills young people with chronic conditions need to develop. On the other hand, there is a strong focus on medical management. It is important that healthcare professionals are also aware of the importance of role and emotional management. Furthermore, an experience-based approach, oriented towards learning from others and ‘mastering through experience’, would be fitting for young people, considering the developmental tasks they have. Future evaluation studies are recommended to give a detailed description of interventions and their theoretical backgrounds and to relate expected outcomes to the content and characteristics of interventions. The use of more generic outcome measures could aid comparison between different studies. The content-related evaluation framework and overview of the content, characteristics and outcomes of interventions, as presented in *chapter 4*, could help with this. Furthermore, this overview could give professionals insight into the broad range of self-management support and help them determine the range and focus of support they offer. Mixed-methods research is recommended to gain additional insight into the (social) context and working elements of self-management interventions.

Chapter 5 examines the different patterns of social participation of a cohort of young adults with chronic conditions, and the differences between those with differing patterns. In general, young adults with a chronic condition lag behind in social participation in comparison to their healthy peers. Four patterns were found: ‘*typical developers*’, ‘*financially secure laggards*’, ‘*slow developers*’ and ‘*outgoing laggards*’, each with their own background characteristics. Differences were found in gender and level of education, but also in terms of whether or not they attended special education, received state benefits for young people with a disability or whether they had a physical disability. Remarkably, more social participation was not always related to a higher quality of life (or vice versa), but it did seem to be related to more self-efficacy and independence. The variety of patterns shows that reaching independence does not run synchronously across all life domains, and emphasizes that the combination of these life domains constitutes reality for young people with chronic conditions. Healthcare providers should be aware of this and can use different interventions to obtain insight into the experiences of these young people and their need for self-management support. Our results emphasize that ‘*one*

size fits all does not apply here, and assert the importance of tailored self-management support.

Chapter 6 evaluates experiences and degree of satisfaction with the transition to adult care in a cohort of young adults with chronic conditions. After a six-year follow-up, around 60% had transitioned to adult care and 14% were still in pediatric care, while the place of treatment of a quarter of the group was unknown. There was no general tendency in terms of satisfaction concerning the transition: About a third was very satisfied with the transition, but a fifth of the group was not. Furthermore, half of the young adults felt properly prepared at the time of the transition, and 24% had had met their new healthcare provider in advance. In general, men were more positive about their experiences and reported a higher level of satisfaction than women. Patient-centeredness of the healthcare provider in adult care was the most important determinant for the transfer experience. Higher self-efficacy in young adults was related to a more positive transfer experience, but not to higher satisfaction. Satisfaction was higher for those who transferred to adult care within the same hospital.

Our results show that while young people need to be better prepared for and involved in the transfer, the first priority should be to build a bridge between child and adult care. The responsibility for a safe and smooth transition goes beyond pediatric care; transition care should therefore not be limited to pediatric care. Gaining trust and investing in new personal relations is the way forward for all parties involved: transition is about responding and bonding.

Chapter 7 studies the relationship between self-management and health-related quality of life in a cohort of young adults with chronic conditions. In general, they experienced a decrease in health-related quality of life if compared to their score six years earlier. Health-related quality of life and self-management were related. While medical and emotional management were related to all domains of health-related quality of life, role management was mostly related to the social domain and the domain of independency. Our results emphasize that young people with chronic conditions need holistic support in acquiring self-management skills. Specific self-management support that accounts for the developmental tasks of young people should also be offered in both pediatric and adult care. Thus, the self-management domains are interrelated, and share associations with the social and independency domains of health-related quality of life. This further emphasizes the importance of including psychosocial aspects in self-management support.

Chapter 8 describes a systematic literature review of current self-management interventions for young people with chronic conditions. The primary goal of this study was

to explore the effects of such interventions from a non-categorical approach to self-management. Another goal was to gain insight into working elements of interventions. Although no strong conclusions can be drawn on intervention effectiveness, results suggest that pediatric self-management interventions might be effective at influencing disease knowledge, adherence and dealing with the chronic condition across a wide range of conditions. There are indications that self-management interventions aimed to stimulate adherence should be focused on medical management, and should be provided individually in a clinical setting by a multidisciplinary team. Furthermore, an individual mode and home setting combined with online peer-support might be effective elements of self-management interventions focused on dealing with the chronic condition in daily life. Also, a mono-disciplinary medical management intervention might stimulate disease knowledge. These combinations of expected outcomes, focus and intervention elements seemed effective irrespective of diagnosis, and may therefore act as good starting points for further research into and improvement of self-management support of young people with chronic conditions. The results underline the need for systematic development and evaluation of self-management interventions. The use of a core set of outcomes could help with this, because this enables comparison between studies and can deliver more concrete evidence about the effectiveness of interventions.

Chapter 9 describes a mixed-methods study into the effects of a recreational camp – *Camp COOL* – as a self-management intervention for young people with a chronic kidney disease (16-25 years). Participating in Camp COOL seems to have a positive influence on the self-management of young people. Peer-to-peer support in the form of a buddy-participant concept was greatly appreciated. Support from young adults is not only beneficial for adolescent attendees, but also for the young adult buddies. It is therefore recommended to keep or start organizing these camps. Pediatric nephrologists are encouraged to refer patients to Camp COOL and to facilitate such initiatives. Together with nephrologists in adult care, they could take on a role in selecting buddies. Also, since young people with other chronic conditions may also benefit from attending, it is advised to explore the possibilities to organize the camp for other groups as well. More attention should be given, however, to the selection and training of buddies, and to the imminent effect of over-identification in order to counteract any negative effects. Future evaluation studies could benefit from a mixed-methods approach, the inclusion of a control group and more measurement moments.

Conclusion

Self-management support for young people growing up with chronic conditions goes beyond medical management. There should be attention for medical, role and

emotional management, for the shifting of parent-child roles and for the coordination of different tasks (articulation work). The dynamic of self-management requires that self-management support is adapted to shifting needs and roles. As a consequence, self-management support takes place on a continuum of strategies that combine different content and roles of involved parties. Instead of a disease-specific approach to self-management, a generic approach supplemented with disease-specific elements is recommended.

Nurses have a main role in self-management support of young people, but other healthcare providers definitely also need to be involved. It is also important to look at other forms of self-management support outside the context of healthcare. Monitoring quality of life can give healthcare providers insight into possible problems young people may experience in daily life. Self-management support should be oriented towards facilitating an environment in which young people can learn from others and 'master through experience'. It is also important to stimulate independent behavior during consultations and to pay attention to transition readiness.

Furthermore, the specific needs of young people with chronic conditions in adult care require attention. There is a need for more transition studies that go beyond pediatric care. Also, it is important to gain additional insight into good practices of self-management support, both in pediatric and adult care. A better understanding of the effects of and working elements in self-management interventions could be gained from evaluation studies that adopt a mixed-methods approach, give detailed intervention descriptions and present information about the context in which interventions take place.

SAMENVATTING

Jongeren vormen een steeds belangrijkere doelgroep in de gezondheidszorg. Door toegenomen overlevingskansen zijn er steeds meer kinderen met een chronische aandoening en steeds vaker bereiken ze de volwassen leeftijd. Volwassen worden vormt een extra uitdaging voor deze jongeren. De aanpassingstaken die een chronische aandoening met zich meebrengt, kunnen namelijk botsen met de normale ontwikkelingsmijlpalen. Een goede balans vinden tussen deze taken en mijlpalen, waarbij men zodanig omgaat met de aandoening dat deze optimaal wordt geïntegreerd in het dagelijks leven, wordt ook wel 'zelfmanagement' genoemd. Jongeren die opgroeien met een chronische aandoening kunnen hier ondersteuning bij gebruiken. In de chronische zorg komt daarom steeds meer nadruk te liggen op zelfmanagementondersteuning.

In de afgelopen jaren is de aandacht voor zelfmanagement toegenomen. Toch is er in de literatuur nog weinig instemming met een definitie waarin zelfmanagement wordt beschouwd als meer dan alleen het managen van een chronische conditie en de medische behandeling daarvan. Zeker voor jongeren lijkt een bredere definitie van belang, waarbij rekening wordt gehouden met normale ontwikkelingstaken en psychosociaal functioneren. Zo'n definitie gaat ervan uit dat zelfmanagement zich uitstrekt over drie domeinen: medisch management (betrekking hebbend op de aandoening en behandeling hiervan), rolmanagement (betrekking hebbend op sociale participatie en rollen) en emotiemanagement (betrekking hebbend op emotioneel welzijn).

Er is nog weinig bekend over hoe zelfmanagement is ingebed in de huidige zorgpraktijk voor de jongeren die opgroeien met een chronische aandoening. Ook is er weinig inzicht in factoren die van invloed zijn op zelfmanagement van jongeren en is er nog niet veel onderzoek gedaan naar de effectiviteit van interventies voor zelfmanagementondersteuning aan deze groep. Dit alles vormde de aanleiding voor dit promotieonderzoek.

Het onderzoek dat in dit proefschrift is beschreven, is onderdeel van het onderzoeksprogramma 'Self-management & Participation Innovation Lab' (SPIL). Dit vierjarige onderzoeksprogramma is in 2011 gestart met als doel zelfmanagement en zelfmanagementondersteuning aan jongeren met chronische aandoeningen te bevorderen. SPIL is een vervolg op het onderzoeksprogramma 'Op Eigen Benen'. De studies voor dit proefschrift zijn deels in het Erasmus MC – Sophia en deels in verschillende academische ziekenhuizen in Nederland uitgevoerd. Meer informatie over de onderzoeksprogramma's en -projecten is te vinden op: www.opeigenbenen.nu.

Het proefschrift bestaat uit drie delen. Het eerste deel verkent de concepten zelfmanagement en zelfmanagementondersteuning voor jongeren met chronische aandoeningen (hoofdstuk 2, 3 en 4). Het tweede deel gaat nader in op de transities van jongeren naar zowel de volwassenheid als de zorg voor volwassenen. Vervolgens onderzoekt het

de ontwikkeling van zelfmanagement en beïnvloedende factoren (hoofdstuk 5, 6 en 7). Het laatste deel gaat over de manier waarop zelfmanagementinterventies worden geëvalueerd en gaat na wat er bekend is over de effectiviteit van de huidige interventies bij kinderen en jongeren (hoofdstuk 8 en 9).

Hoofdstuk 2 beschrijft een Delphi-studie naar de visies van onderzoekers en beleidsadviseurs op zelfmanagement in het algemeen. Net als in de literatuur, is er binnen de groep van experts onduidelijkheid over het concept zelfmanagement, maar in dit geval was er een tendens om zelfmanagement meer te relateren aan persoon-gebonden concepten en minder aan medisch management. Terwijl zorgverleners moeten worden ontmoedigd om zelfmanagement te beperken tot medisch management, is het voor onderzoekers en beleidsadviseurs belangrijk te onderkennen dat ook de medische behandeling een essentieel onderdeel is van het dagelijkse leven met een chronische aandoening. Zelfmanagementondersteuning begint met het verkrijgen van inzicht in de individuele behoeften van de jongeren, zoals die voortkomen uit hun adaptieve opgaven (in zowel het medische als het sociale en emotionele domein). Deze behoeften bepalen de focus van de zelfmanagementondersteuning. Bij de ontwikkeling van *evidence based* interventies en het informeren van (toekomstig) beleid, is het advies aan onderzoekers en beleidsadviseurs om explicieter te zijn over de behoeften waarop zelfmanagementondersteuning inspeelt en dus over de beoogde doelen.

Hoofdstuk 3 onderzoekt de eerste ervaringen van zorgverleners met het ontwikkelen en implementeren van een zelfmanagementinterventie – de *Nier Groei-wijzer* – voor jongeren met een chronische nieraandoening. De *Nier Groei-wijzer* is samen met zorgverleners, jongeren, hun ouders en een aantal experts ontwikkeld. Het instrument is bedoeld om een ‘normale’ ontwikkeling van autonomie en zelfstandigheid te stimuleren. Alle partijen bereikten consensus over de levensdomeinen die in het instrument aan bod komen. Ze bevatten items over emotiemanagement, sociale participatie (rolmanagement) en medisch management voor jongeren met chronische nieraandoeningen. De zorgverleners gebruikten en waardeerden het instrument, maar gaven aan dat het voor de haalbaarheid van het instrument belangrijk is dit te digitaliseren en online beschikbaar te maken. Ook moesten zij wennen aan de nieuwe methode van werken waarin zij een minder actieve rol hadden, terwijl de jongeren en hun ouders meer verantwoordelijkheid moesten nemen bij het bepalen van de gespreksagenda en de actieplannen. De uitdaging voor zorgverleners in de kinder nefrologie is verder te kijken dan medisch management en de bredere ontwikkeling van jongeren in ogenschouw te nemen. Zo kunnen deze jongeren optimaal begeleid worden naar volwassenheid. Inzicht in hun ervaringen en behoeften is essentieel voor goede zelfmanagementondersteuning.

Hoofdstuk 4 gaat in op een systematische literatuurreview van zelfmanagementinterventies voor kinderen en jongeren (7-25 jaar). Het doel van de review was inzicht te krijgen in de kenmerken, de inhoud, de onderliggende theorieën en de verwachte uitkomsten van bestaande zelfmanagementinterventies. De meeste interventies werden aangeboden op groepsniveau en richtten zich op educatie en/of vaardigheidstraining. Een kwart van de interventies was bedoeld voor zowel jongeren als hun ouders. Er was een breed scala aan settingen waarin interventies werden ingezet, maar meestal vonden ze plaats op de polikliniek. Er waren veel verschillende professionals betrokken bij de uitvoering ervan. Een aantal interventies was gericht op alle drie de domeinen van zelfmanagement, maar het merendeel alleen op medisch management. Het medische, rol- en emotiemanagement was niet specifiek voor bepaalde interventiekenmerken (zoals type zorgverlener, setting of format), maar werd wel aangepast aan de leeftijd van de groep die de interventie aangeboden kreeg. Bij een groot deel van de interventies werd niet gerapporteerd over hun theoretische onderbouwing. De meest genoemde theorieën waren de *social learning*-theorie en de cognitieve gedragstheorie. Verwachte uitkomsten waren erg divers; meestal ging het om klinische en gezondheidsuitkomsten, kwaliteit van leven en kennis over de aandoening en/of behandeling.

Uit de literatuurreview kan worden geconcludeerd dat de huidige zelfmanagementinterventies goed aansluiten bij de zelfmanagementtaken van jongeren met chronische aandoeningen en de zelfmanagementvaardigheden die zij nodig hebben. Wel is er een sterke focus op medisch management. Het is belangrijk dat zorgprofessionals zich bewust zijn van het belang van rol- en emotiemanagement. Verder zou een ervaringsaanpak, gericht op het leren van anderen en het 'beheersen door te ervaren', passend zijn voor jongeren, gezien de ontwikkelingstaken waar zij voor staan. Er wordt geadviseerd om in toekomstige evaluatiestudies de interventies en hun theoretische achtergrond gedetailleerd te beschrijven en de verwachte uitkomsten te relateren aan de inhoud en kenmerken van de interventies. Ook kan het gebruik van meer generieke uitkomstmaten in de evaluatie van zelfmanagementinterventies onderlinge vergelijkingen tussen studies ten goede komen. Het inhoudelijke evaluatiekader en het overzicht van de inhoud, kenmerken en uitkomsten van interventies, zoals gepresenteerd in hoofdstuk 4, zou hierbij kunnen helpen. Verder zou dit overzicht professionals inzicht kunnen verschaffen in de diversiteit van zelfmanagementondersteuning en hen kunnen helpen bij het vaststellen van de breedte en focus van de ondersteuning die zij bieden. *Mixed-methods* onderzoek wordt aanbevolen om meer inzicht te krijgen in de (sociale) context en de werkende elementen van zelfmanagementinterventies.

Hoofdstuk 5 kijkt naar verschillende patronen van sociale participatie van een cohort van jongvolwassenen met chronische aandoeningen, en naar verschillen tussen degenen met verschillende participatiepatronen. Over het algemeen lopen jongvolwassenen

met een chronische aandoening achter in sociale participatie vergeleken met gezonde leeftijdsgenoten. Er werden vier patronen gevonden: *'typical developers'*, *'financially secure ladders'*, *'slow developers'*, en *'outgoing ladders'*. Jongvolwassenen met verschillende patronen verschilden op achtergrondkenmerken zoals geslacht en opleidingsniveau, maar ook in het al dan niet gevolgd hebben van speciaal onderwijs, de aanwezigheid van een Wajong-uitkering en een fysieke beperking. Opvallend was dat meer sociale participatie niet altijd gerelateerd was aan een hogere kwaliteit van leven (of andersom), maar wel gerelateerd leek te zijn aan meer zelfeffectiviteit en ervaren zelfstandigheid. De variëteit aan patronen laat zien dat het bereiken van zelfstandigheid niet synchroon loopt in alle levensdomeinen, en geeft aan dat de combinatie van deze levensdomeinen de realiteit vormt voor jongvolwassenen met chronische aandoeningen. Zorgverleners zouden zich hier meer bewust van moeten zijn en kunnen verschillende interventies gebruiken om inzicht te krijgen in de ervaringen en de behoeften van jongeren op het gebied van zelfmanagementondersteuning. Onze resultaten bevestigen dat er geen benadering is die voor iedereen bruikbaar is. Ze onderstrepen het belang van maatwerk in de zelfmanagementondersteuning voor jongeren.

Hoofdstuk 6 evalueert de ervaringen en tevredenheid met de overstap naar de volwassenenzorg in een cohort van jongvolwassenen met chronische aandoeningen. Na een follow-up van zes jaar was ongeveer 60% overgestapt naar de zorg voor volwassenen en werd 14% nog in de kinderzorg behandeld, terwijl van een kwart onbekend was of en waar zij onder behandeling waren. Er was geen algemene tendens in tevredenheid met de overstap te vinden: ongeveer een derde van de jongvolwassenen was erg tevreden, maar een vijfde gaf een onvoldoende voor het totale overgangsproces. Verder voelde de helft van de jongvolwassenen zich goed voorbereid en had 24% vooraf kennis gemaakt met zijn of haar nieuwe zorgverlener. Mannen waren over het algemeen positiever over hun ervaringen en rapporteerden hogere tevredenheid dan vrouwen. Patiëntgerichtheid van de zorgverlener in de volwassenenzorg was de belangrijkste determinant voor positieve ervaringen. Hogere zelfeffectiviteit in jongvolwassenheid was gerelateerd aan een positievere ervaring van de overstap, maar niet aan meer tevredenheid. De tevredenheid was wel hoger onder degenen die binnen hetzelfde ziekenhuis overstapten naar de zorg voor volwassenen.

Onze resultaten laten zien dat, terwijl adolescenten beter voorbereid moeten worden op en meer betrokken moeten worden bij de transitie, de eerste prioriteit is om bruggen te bouwen tussen de kinder- en volwassenenzorg zelf. De verantwoordelijkheid voor een veilige en soepele overgang gaat verder dan de kinderzorg; transitiezorg moet daarom niet tot de kinderzorg beperkt blijven. Het verkrijgen van vertrouwen en investeren in nieuwe persoonlijke relaties is de weg voorwaarts voor alle betrokken partijen.

Hoofdstuk 7 onderzoekt de relatie tussen zelfmanagement en kwaliteit van leven in een cohort van jongvolwassenen met chronische aandoeningen. Over het algemeen ervoeren de jongvolwassenen een afname in kwaliteit van leven ten opzichte van de score zes jaar daarvoor. Kwaliteit van leven en zelfmanagement waren met elkaar gecorreleerd. Terwijl medisch en emotiemanagement aan alle domeinen van de kwaliteit van leven gerelateerd waren, was rolmanagement vooral gerelateerd aan de sociale- en zelfstandigheidsdomeinen van kwaliteit van leven. Onze resultaten tonen aan dat jongeren met chronische aandoeningen holistische ondersteuning nodig hebben bij het oppakken van zelfmanagement. Ook zou specifieke zelfmanagementondersteuning, gericht op de ontwikkelingstaken van jongeren, zowel in de kinder- als de volwassenenzorg aangeboden moeten worden. De onderlinge samenhang tussen de drie zelfmanagementdomeinen en de relatie die zij alle drie hebben met de sociale- en zelfstandigheidsdomeinen van kwaliteit van leven, onderstrepen dat aandacht voor de psychosociale aspecten belangrijk is in zelfmanagementondersteuning.

Hoofdstuk 8 beschrijft een systematische literatuurreview van bestaande zelfmanagementinterventies voor jongeren met chronische aandoeningen. Het doel van deze review was te onderzoeken wat er gezegd kan worden over de effecten van dergelijke interventies vanuit een generieke benadering van zelfmanagement. Een ander doel was inzicht te krijgen in de werkzame elementen van interventies.

Hoewel er geen harde conclusies kunnen worden getrokken over de effectiviteit van huidige interventies, suggereren de uitkomsten dat ze in staat zijn om kennis over de aandoening, therapietrouw en omgang met de aandoening positief te beïnvloeden bij een breed scala aan verschillende chronische aandoeningen. Er zijn aanwijzingen dat interventies die door een multidisciplinair team in een poliklinische setting aan de individuele jongere worden aangeboden, gericht op medisch management, therapietrouw kunnen bevorderen. Interventies gericht op het omgaan met de aandoening in het dagelijkse leven zouden daarentegen individueel in de thuissituatie aangeboden moeten worden, en gecombineerd moeten worden met online peer-support. Een monodisciplinaire interventie gericht op louter medisch management zou jongeren kunnen helpen meer kennis over de aandoening te krijgen. Deze combinaties van verwachte uitkomsten, focus en werkzame elementen van de interventies bleken effectief ongeacht de aandoening en kunnen daardoor als startpunt dienen voor verder onderzoek. De resultaten onderstrepen de noodzaak om zelfmanagementinterventies systematisch te ontwikkelen en evalueren. Het gebruik van een kern-set van uitkomsten kan hierbij behulpzaam zijn, omdat dit vergelijking tussen studies mogelijk maakt en sterker bewijs kan leveren voor de effectiviteit van interventies.

Hoofdstuk 9 beschrijft een *mixed-methods* studie naar de effecten van een recreatie-kamp – *Camp COOL* – als zelfmanagementinterventie voor jongeren met een chronische nieraandoening (16-25 jaar). Participeren in *Camp COOL* lijkt een positieve invloed te hebben op het zelfmanagement van jongeren. De *peer-to-peer*-support waarbij een buddy en een deelnemer aan elkaar worden gekoppeld, wordt door alle deelnemers erg gewaardeerd. Ondersteuning door jongvolwassen lotgenoten levert niet alleen een meerwaarde op voor de deelnemende adolescenten, maar ook voor deze jongvolwassenen zelf. Daarom wordt aanbevolen om door te gaan met het organiseren van *Camp COOL* en de mogelijkheden te verkennen om het kamp ook voor andere doelgroepen te organiseren. We bevelen zorgverleners aan om jongeren naar dergelijke initiatieven door te verwijzen. Daarnaast kunnen zorgverleners uit zowel de kinder- als volwassenenzorg een rol spelen bij het selecteren van buddy's. Om eventuele negatieve effecten tegen te gaan, zou bij toekomstige kampen meer aandacht gegeven kunnen worden aan de selectie en training van buddy's en aan het mogelijke effect van over-identificatie binnen een groep. Toekomstige evaluatiestudies zouden kunnen profiteren van een *mixed-methods* aanpak, het gebruik van een controlegroep en meerdere meetmomenten.

Conclusie

Zelfmanagementondersteuning voor jongeren die opgroeien met chronische aandoeningen gaat verder dan medisch management. Er moet meer aandacht komen voor medisch, rol- en emotiemanagement, voor rolverschuivingen tussen ouder en kind en voor de coördinatie van alle taken en verantwoordelijkheden (*articulation work*). De dynamiek van zelfmanagement vereist dat zelfmanagementondersteuning aangepast wordt aan de verschuivende behoeften en rollen. Zelfmanagementondersteuning vindt dus plaats binnen een continuüm van strategieën die verschillende inhoud en rollen van betrokken partijen combineren. In plaats van een ziektespecifieke benadering van zelfmanagement is een generieke benadering van zelfmanagement voor jongeren met chronische aandoeningen aan te bevelen, aangevuld met ziektespecifieke elementen.

Verpleegkundigen kunnen een hoofdrol vervullen bij zelfmanagementondersteuning van jongeren, maar ook andere zorgverleners moeten er zeker bij betrokken worden. Ook is het belangrijk om te kijken naar vormen van zelfmanagementondersteuning buiten de context van de gezondheidszorg. Regelmatige monitoring van kwaliteit van leven zou zorgverleners inzicht kunnen geven in problemen die jongeren bij hun zelfmanagement ervaren. Verder is zelfeffectiviteit een belangrijke determinant van zelfmanagement. Zelfmanagementondersteuning moet daarom een omgeving creëren waarin jongeren kunnen leren van anderen en kunnen 'beheersen door te ervaren'. Ook is het belangrijk om zelfstandig gedrag in de spreekkamer te stimuleren en van jongs af aan aandacht te besteden aan de ontwikkeling van '*transition readiness*'.

Er moet meer aandacht komen voor de specifieke behoeften van jongvolwassenen met chronische aandoeningen in de volwassenenzorg. Er moeten dan ook meer studies worden uitgevoerd die verder gaan dan de kindergeneeskunde. Daarnaast is het belangrijk om meer inzicht te krijgen in *good practices* van zelfmanagementondersteuning voor jongeren met chronische aandoeningen, in zowel de kindergeneeskunde als de volwassenenzorg. Voor studies die zelfmanagementinterventies evalueren wordt een *mixed-methods* aanpak aanbevolen. Ook gedetailleerde interventiebeschrijvingen en informatie over de context waarbinnen interventies plaatsvinden, zijn noodzakelijk om goed inzicht te krijgen in de effecten en werkzame elementen van zelfmanagementinterventies.

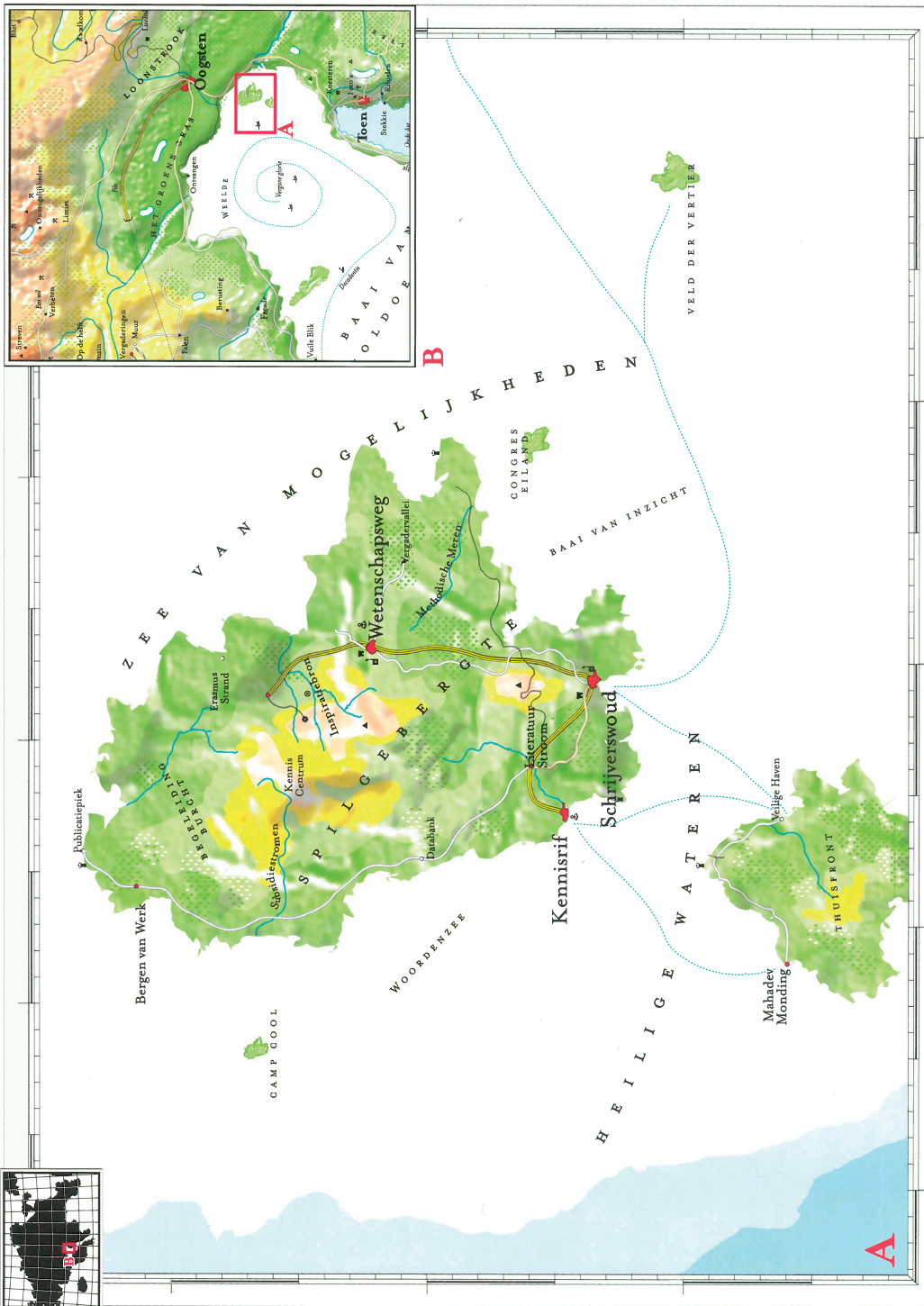
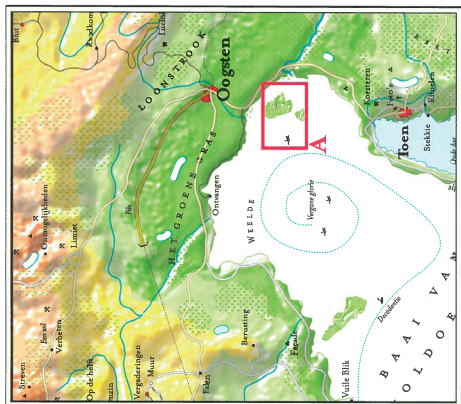
Appendices



DE BELEVINGSWERELD



TAAL VAN WEELDE (BERGEN V. WERK)



DANKWOORD

Alleen zijn we één druppel; samen vormen we een oceaan. Een *zee van mogelijkheden*.

Mijn *Reis der Promotie* was avontuurlijk, uitdagend en leerzaam. Van de *Publicatiepiek* tot het *Veld der Vertier*, een 'proeftuin' waar ik gedurende mijn transitie naar wetenschapper mijn 'mastery experiences' mocht opdoen. Er zijn velen die gezamenlijk mijn *zee van mogelijkheden* vorm gaven, die mij de ruimte gaven, maar die ook ondersteunden, (bege)leidden, onderwezen, een voorbeeld waren, een luisterend oor boden of zorgden voor de broodnodige afleiding.

AnneLoes, we ontmoetten elkaar op het *Erasmusstrand* en ik mocht met je meereizen naar het *Kennis Centrum*. Als co-promotor zat je vaak in de *Begeleiding Burcht*, maar we hebben ook *Bergen van Werk* verzet op vele andere mooie en minder mooie (lees: Bobigny) plekken. Ik weet dat je het ongemakkelijk vindt als je de hemel in geprezen wordt – je hebt het me zelfs 'verboden'. Toch mag gezegd worden dat je met jouw veelzijdigheid, betrokkenheid en kritische blik een parel van een begeleider bent. Je daagt me precies op de goede momenten in de goede dingen uit, om net weer boven mezelf uit te stijgen. Ik heb veel van je geleerd. Dank voor het vertrouwen, de kansen, het enthousiasme en de inspiratie. Ik hoop dat we nog wel een tijdje samen mogen door reizen.

Roland, ook jou kwam ik op het *Erasmusstrand* tegen en ook jij zat, als promotor, vaak in de *Begeleiding Burcht*. Toch was je daarbuiten ook makkelijk bereikbaar en kwam je vaak genoeg naar het *Kennis Centrum*. Onze gesprekken voerden we vaak langs de *Baai van Inzicht*, maar we hebben ook samen in het *Schrijverswoud* gezeten. Jouw inbreng zorgde altijd weer voor een frisse blik en nieuwe gedachten. Dank voor de inzichten, inspiratie en het vertrouwen. Bedankt dat je samen met AnneLoes mijn wegwijzer wilde zijn. Ik hoop dat ik je nog vaak mag tegenkomen op volgende reizen.

Vanaf het SPILgebergte waren er meerderen die regelmatig met mij mee liepen. De *Wetenschapsweg* op, via de *Vergadervallei*, langs de *Databank*, de *Literatuur Stroom*, het *Kennisrif* tot aan het *Schrijverswoud* en terug. Het SPIL Sophia onderzoeksteam stond mij altijd bij met raad en daad. AnneLoes, Jos, Susan, Linda en Sander, en later ook Mariëlle en Erwin naast de betrokken stagiaires en studenten, dank voor alle inspanningen en het meedenken. Maar ook het SPIL Revalidatie onderzoeksteam en de SPIL stuurgroep hebben meegedacht. Bedankt voor jullie input tijdens onze bijeenkomsten. De Health Care Governance sectie (Erasmus Universiteit) en Zelfmanagement en Participatie onderzoeksgroep (Hogeschool Rotterdam) dank ik voor het bieden van een podium - met

vragenronde - voor mijn praatjes zo nu en dan. Veel dank ben ik ook verschuldigd aan iedereen die heeft meegewerkt aan (delen van) dit onderzoek, in het bijzonder Jane de la Fosse (*Camp COOL*), de kindernefrologische teams in de academische ziekenhuizen en alle jongeren die als respondent hebben meegedaan. Ik hoop dat de resultaten uit dit proefschrift de brug slaan naar *Oogsten* (in de praktijk). Het Nationaal Regieorgaan Praktijkgericht Onderzoek, de Nierstichting en de Hogeschool Rotterdam waren belangrijke *Subsidiestromen*.

In het centrum van het *Kennis Centrum* kon ik altijd weer terecht voor de ondersteuning rond de SPIL projecten. Hanneke, bedankt voor het geduld, de inspanningen en de creatieve oplossingen. Elly, Nadia, nu ook Brigitte, jullie ook bedankt. Ilona, dank voor de mooie posters voor mijn reisjes naar het *Congreseiland*.

Ko Hagoort, veel dank voor het temmen van de golven van mijn Engelse *Woordenzee*. Ik bewonder je 'inkortingsvermogen'! Hans Schoots, bedankt voor het editen van mijn Nederlandse samenvatting van dit proefschrift. Maarten Thissen en Diederik Corvers, de illustraties in mijn discussie hoofdstuk en het omslag van dit proefschrift zijn prachtig geworden. Veel dank hiervoor.

Alle collega's bij het Kenniscentrum Zorginnovatie dank ik voor de fijne werksfeer en gezelligheid. Dat maakt het allemaal nog leuker.

Bijzondere dank gaat uit naar Heleen, Hanny, Sander, Janet, Mariëlle en Marjolijn, naast alle anderen. Voor de kopjes koffie en thee, gesprekken over alles en werk, gezelligheid en wijsheid. Heleen, ondanks dat we geen gezamenlijke projecten hadden, hebben we veel beleefd op het *Congreseiland*. Wie weet komt er nog een gezamenlijk project aan. Hanny, dank voor dit creatieve idee. Sander, weet je nog dat we naar het AMC gingen? Janet, fijn dat je er weer bent; we hebben je gemist. Mariëlle, ik ben blij dat jij me nog even gezelschap houdt op het *SPILgebergte*. Zullen we weer wat leuks plannen?

Marjolijn, lieve paranimf, jij verdient een bijzonder plekje. Je bent zoveel meer dan mijn mede-SPIL-promovendus. Je bent een vriendin en luisterend oor. Daarom ben ik blij dat je naast me wilt staan straks. En daarna gaan we vrolijk door. Van de etentjes op het *Veld der Vertier* (met de andere kuikens) tot de *Publicatiepiek*. Ideeën zijn er genoeg, maar eerst nog een reisje naar het *Congreseiland*. Dank je wel voor alles.

Dan kom ik eindelijk bij het *Thuisfront* waar het allemaal begon. Lieve mama, er zijn geen woorden genoeg om jou te bedanken en geen woorden kunnen beschrijven wat jij voor mij betekent. Ik bewonder je en leer elke dag weer meer van je. Met jou heb

ik veel avonturen beleefd op onze binnen- en buitenlandse, jaarlijkse en wekelijkse yatra's. Over de Heilige Wateren en bij de Mahadev Monding. Ik zal ze nooit vergeten en ik hoop dat we nog veel samen mogen meemaken. Lieve papa, ook van jou heb ik veel geleerd. We delen veel; muziek, films, hobby's. Wanneer gaan we weer knutselen met hout? Amar, mijn grote *Bhaiyya*, je bent één van de meest wijze mensen die ik ken en ik kan altijd op je rekenen. Niet gek dan, dat ik vaak jou bel als er iets is, leuk of minder leuk. Grote broer, laten we nog veel samen doen, samen met Cindy en kleine Chayen. Misschien weer samen op vakantie, binnenkort? Anoop, mijn kleine *Bhaiyya*, je bent één van de sterkste mensen die ik ken – ook al heb je dat zelf niet altijd door. Ook op jou kan ik altijd rekenen en je toont vaak interesse in waar ik allemaal mee bezig ben. Samantha, dat geldt ook voor jou natuurlijk. Dank jullie wel. Gaan jullie mee op reis?

"Family is a training ground where you rise beyond likes and dislikes" (Sadhguru). Jullie hebben mij gevormd en ik sta daar straks mede dankzij jullie. Dank jullie wel voor alle steun, warmte en liefde; voor de *Veilige Haven*. Natuurlijk zijn er nog anderen op het *Thuisfront*. Lieve familie en vrienden, allemaal bedankt voor de vele uitstapjes naar het *Veld der Vertier*. Angela, Saraja, ik koester mijn vriendschap met jullie. Pallvi, dank je wel dat je mijn 'sister from another mother' bent.

Een bijzonder plekje voor mijn andere paranimf, Savitri. *Phoewa*, je bent één van de liefste. Toen ik mijn VWO diploma behaalde, kreeg ik van jou een mooie pen cadeau – met een mededeling: "Nooit stoppen met schrijven". Dat sloeg natuurlijk op mijn voorliefde voor lezen en schrijven en zie hier, ik heb het ten harte genomen. Dank je wel voor je steun, enthousiasme en warmte. Ik ben blij dat je naast me wilt staan straks.

और आप भोलेनथ, आप तो मेरे आराध्य हो, इष्ट हो |
सब कुछ आप को अर्पित है |

ॐ नमः शिवाय

PHD PORTFOLIO

Name PhD student: Jane Sattoe
 Department: Erasmus University Rotterdam, Institute of Health Policy and Management
 PhD period: 2011 – 2015
 Promotor: Prof. dr. Roland Bal
 Supervisor: Dr. AnneLoes van Staa

PhD training	Year
<i>Research skills</i>	
- Academic writing, Rotterdam University	2011
- Focus group discussions, Evers Research & Training	2011
- Atlas.ti for qualitative data analysis, Evers Research & Training	2011 & 2013
- Structure Equation Modelling, Utrecht University (Summer Course)	2012
- Multilevel statistical analyses, Rotterdam University	2014
<i>Research groups</i>	
- Research meetings Self-management & Participation Innovation Lab	2011 – now
- Meetings 'Promovendiclub Zelfmanagement'	2012 – now
<i>Presentations at international conferences</i>	
- 'Camp COOL: Buddies in charge, peer-support among adolescents with ESRD'; International Association for Adolescent Health, 17 th European Annual Meeting, Antalya Turkey	2012
- 'The Dutch Skills for Growing Up checklists in paediatric Nephrology (dSGU-N), a self-management support tool'; International Association for Adolescent Health, 17 th European Annual Meeting, Antalya Turkey	2012
- 'Evaluating self-management interventions for young people with chronic conditions: A review'; 18 th European Congress of the International Association for Adolescent Health, Paris, France	2014
- 'What determines the impact of medication regimen in young adults with chronic conditions?'; 18 th European Congress of the International Association for Adolescent Health, Paris, France	2014
- 'Discussing non-medical issues during consultations with chronically ill adolescents: Experiences with Individual Transition Plans'; European Association of Communication in Healthcare, 12 th International Conference on Communication in Healthcare, Amsterdam, the Netherlands	2014
<i>Other</i>	
- Peer reviewer for international academic journals (<i>Health and Quality of Life Outcomes, Health Expectations, Social Psychiatry and Psychiatric Epidemiology, Health Care: Current Reviews, Journal of Adolescent Health</i>)	2012 – now
Teaching qualifications and activities	
<i>Training</i>	
- Didactics, Rotterdam University	2012 – 2013
<i>Lecturing</i>	
- Bachelor Human Technology (The Hague University): Quantitative research	2012 – 2013
- Bachelor Instituut voor Gezondheidszorg (Rotterdam University): diverse bijdragen aan minoronderwijs en projectbegeleiding (minor Kind & Jeugd en minor GGZ)	2012 – now
- Bachelor Nursing (Rotterdam University): Schrijven van een essay	2013 – now
<i>Supervising and appraising theses</i>	
- Master Advanced Nursing Practice (Rotterdam University)	2012 – 2013
- Bachelor Nursing (Rotterdam University)	2012 – now
<i>Other</i>	
- Supervising interns and trainees	2012 – now

LIST OF PUBLICATIONS (NOT INCLUDED IN THIS THESIS)

Sattoe JNT, van Staa AL, Moll HA: The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. *Health and Quality of Life Outcomes* 2012, 10:10.

Van Staa AL, **Sattoe JNT**: Op Eigen Benen Vooruit! Betere transitiezorg voor jongeren met nierziekten. *Dialyse & Nefrologie Magazine* 2013, 31(3):10-17.

Van Staa AL, **Sattoe JNT**: Betere Transitiezorg bij chronische nierziekten. *Nederlands Tijdschrift voor Nefrologie* 2014, 4(2):5-11.

Saan MC, Boeijs H, **Sattoe JNT**, Bal MI, Missler MA, van Wesel F: Accounting for the role of stakeholders in systematic reviews: demonstration of a Tool for Recording and Accounting for Stakeholder Involvement (TRASI). *Health Information and Libraries Journal* 2015, doi: 10.1111/hir.12099. [Epub ahead of print].

CURRICULUM VITAE

Jane Sattoe was born in Rotterdam on the 5th of April 1988. At age of 9 she moved to Surinam for four years. There she followed her first year of secondary education at Arthur Alex Hogendoorn Atheneum (2000-2001). When she returned, Jane continued secondary education at Melanchthon College (2001-2006). She studied health sciences and got her Bachelor's degree in 2009. In 2010, she graduated in health economics, policy and law at the Erasmus University Rotterdam. After six months working as a junior researcher, in 2011, she started her PhD project at Research Centre Innovations in Care, Rotterdam University. Jane's research focuses on self-management and transitions of young people with congenital or childhood-onset conditions. She is currently involved in the evaluation of transition clinics.

