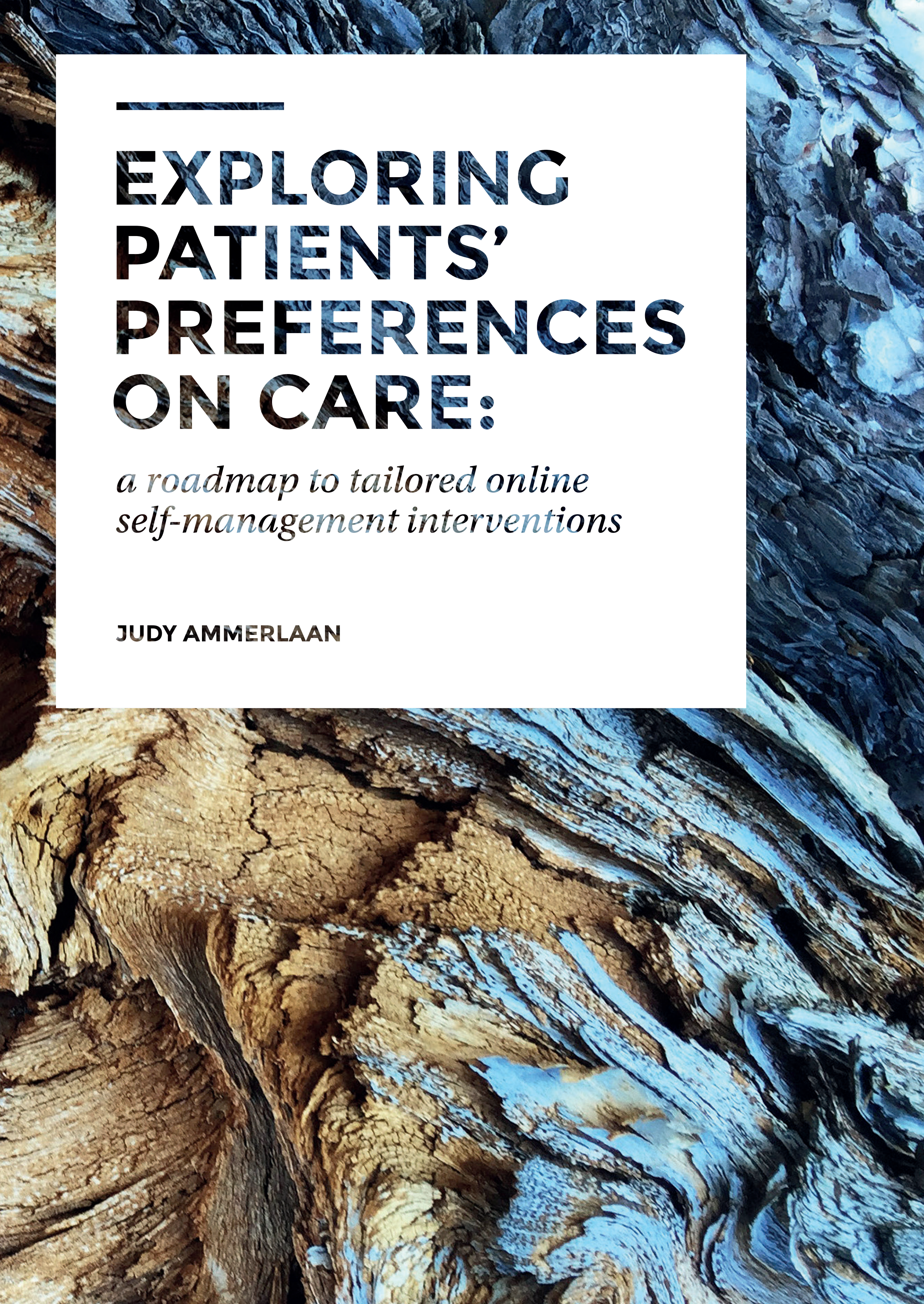

EXPLORING PATIENTS' PREFERENCES ON CARE:

a roadmap to tailored online self-management interventions

JUDY AMMERLAAN

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EXPLORING PATIENTS' PREFERENCES ON CARE:

A roadmap to tailored online self-management interventions

Het verkennen van voorkeuren van patiënten voor zorg:
een roadmap naar online zelfmanagement interventies op maat

(met een samenvatting in het Nederlands)

Proefschrift

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in het openbaar te verdedigen op
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door

Johanna Wilhelmina Ammerlaan
geboren op 18 januari 1966
te De Bilt

Promotoren	Prof. dr. J.W.J. Bijlsma & Prof. dr. A.B.J. Prakken
Copromotoren	Dr. H. van Os-Medendorp & Dr. A.A. Kruize

> *“For me self-management stands for growing up but also to equip myself with strategies and tools so I can survive a storm that I call rheumatism. For me, it is better to bend a little than to break...”*
>
> *- Female, 19 years old, since 9 years Juvenile Idiopathic Arthritis*

CONTENT

> “Self-management for me is looking after yourself, to play an active role in the management of your own disease”
>
> *- Male, 36 years old, since 10 years Rheumatoid Arthritis*

CHAPTER 1

GENERAL INTRODUCTION

Due to research, technology and innovation, people are getting older and therefore the number of people who are suffering from a chronic disease is steadily growing (1, 2). In the Netherlands, 1.4 million people are diagnosed with arthritis and/or osteoarthritis (3). Among them, approximately 3000 children and young adults have a rheumatic disease; the largest group diagnosed with Juvenile Idiopathic Arthritis (JIA) (4). Chronic inflammatory arthritis including Rheumatoid Arthritis and JIA are characterized by inflammation, with a pattern of flares and low physical activity (5, 6). Patients may experience pain, fatigue and physical constraints which contribute to reduced health-quality of life (6-8). Treatment focusses mainly on remission by providing medication including painkillers, non-steroidal anti-inflammatory drugs (NSAIDs), disease modifying anti-rheumatic drugs (DMARDs) or biologicals, often in combination with glucocorticoids (6, 9).

Although in the past decade, many new treatment strategies have been developed, it is still not possible to cure the disease. Consequently, living with a rheumatic disease imposes daily challenges upon those affected and necessitates patients to make daily decisions about the way they manage their illness and life (7, 10, 11). These challenges may be even bigger for young adults, who not only have to develop their own capacity to think critically and to make autonomous informed decisions due to their maturing process, but who gradually also have to become more responsible for their own management of their disease (12-14). Young adults with JIA are usually unprepared for and inexperienced with these tasks (15, 16). Consequently, this may lead to major problems including treatment drop out or passive, resistant behavior towards health professionals and/or parents when they make the transition from child orientated care to adult care (16-18).

Apart from the paediatrician and the rheumatologist, other health care professionals such as rheumatology nurses, physiotherapists and occupational therapists are involved in treatment and support of (young adult) patients (19, 20). Health care professionals are challenged not only to develop personalized treatment and effective treatment (21) but also to develop an organization which accommodates to the diversity of its patient's needs, life stages, questions and preferences (22, 23). In the context of these challenges, self-management is an important concept and is often defined as the individual's ability to deal with symptoms, treatment, physical and psychosocial consequences, and life style changes inherent to a chronic condition (24). However, patients and health care professionals tend to give different meanings to the concept of self-management. Where patients with a rheumatic disease define self-management 'as a way to gain control over their lives' (25), health care professionals relate the concept to adherence to treatment or formalized patient education programs (11). In the last decade several interventions have been developed to support self-management of young adults and adult patients, mostly combining information-based and cognitive-behavioral strategies (26, 27). Some of these interventions are also available online (28-30). However, data on the effectiveness of the self-management interventions is not consistent (22, 26, 27). This might be due to the diversity of interventions, the different theoretical fundaments, the heterogeneity of the studied patient populations and the wide range of assessment tools, used to measure the effects of self-management (19, 22, 26, 27). Until now, it is unclear what the needs of young adult and adult patients for self-management support are, how self-management interventions can be tailored to these needs and what the implications of these interventions mean for daily practice of health care professionals.

Therefore, the aims of this thesis are: 1) to explore the perspectives, needs and preferences of young adults and adult patients with a rheumatic disease regarding self-management, 2) to develop self-management interventions, based on the outcomes of this exploration and 3) to test feasibility and effectiveness of the interventions among the young adults and adult patients.

Outline of the thesis and research questions

In the first part of this thesis, we focus on the youngest patient group: young adults in the age from 16-25 years old, suffering from a juvenile rheumatic disease who making the transition from a paediatric rheumatology department to an adult rheumatology department. In the context of transition, increased involvement of this group in health care and self-management might prevent increasing disease-activity and worsening of symptoms, thus facilitating a successful transition to adult health care (13, 30, 31). But less is shared about the process of changing health care and expectations and needs of young adults concerning healthcare and support of healthcare professionals (16). Also data on the implementation of transitional care in daily rheumatology practice is scarcely reported. We therefore share our experiences on redeveloping transitional health care suitable to the expectations and needs of young adults and their parents in chapter 2.

To improve knowledge and to encourage active involvement of young adults with Juvenile Idiopathic Arthritis (JIA), we developed an informative website with written and video information and an online portal with access to the personal medical record, self-monitoring and eConsult functionalities. According to the new Medical Research Council (MRC) guidance (32) for developing and evaluating complex interventions, the applications were tested on feasibility. Therefore the concepts of ease of use, perceived usefulness and intention to use of the Technology Acceptance Model (TAM) (33) are evaluated in chapter 3: *What is the feasibility of an informative website with written and video information for young adults with a juvenile rheumatic disease and an online portal with access to the personal medical record, self-monitoring and eConsult functionalities, evaluated by young adults with Juvenile Idiopathic Arthritis (JIA)?*

Based on the self-efficacy theory (34), we developed a peer-guided online and a face-to-face version of a self-management intervention for young adults with a rheumatic disease. The aim of this intervention is to support young adults to enhance their skills and knowledge in dealing with the consequences of having a rheumatic disease. We conducted a feasibility study to assess the usefulness, ease of use and user acceptance of the online and face-to-face version among participating young adults and peer-leaders. The results of this study are presented in chapter 4: *What is feasibility of an online and a face-to-face, interactive peer-guided self-management intervention, evaluated by young adults with a juvenile rheumatic disease and peer leaders?*

The online self-management intervention may have an added value in daily health care to support young adults with JIA within the maturing and transition process. According to the MRC guidance (32), before implementing the intervention in daily practice, we investigated its effectiveness among young adults being treated at the transition outpatient clinic of the Rheumatology & Clinical Immunology departments of the University Medical Center Utrecht and the Erasmus Medical Hospital Rotterdam in the Netherlands. A randomized controlled trial with additional qualitative analyses of two interaction components (goals and Chat) was conducted and described in chapter 5: *What is the effectiveness of the web-based self-management intervention, compared with the usual care young adults with JIA received at a transition outpatient clinic?*

The diversity of patient populations, the different theoretical foundations as well as heterogeneity of the self-management interventions, makes it challenging to demonstrate effectiveness with commonly used metrics (22, 35). In response to this issue, the Health Education Impact Questionnaire (heiQ) was developed in Australia (36) and translated into many languages including German (37) and French (38) but not in Dutch. The original heiQ and its translations showed good psychometric properties for evaluating the impact of health education and self-management programs. Consequently, the heiQ might be a valued instrument to evaluate self-management interventions in the Netherlands, including the interventions studied in this thesis. Therefore, we conducted a systematic translation process and studied the psychometric properties among patients with arthritis, atopic dermatitis, food allergy and asthma in chapter 6: *What are the psychometric proportions of the Dutch translation of the Health Education Impact Questionnaire (heiQ) and what is the validity and reliability of the Dutch translation, compared with the English, German and French translations of the heiQ?*

With the expansion of the online self-management intervention for young adults, adults also expressed their need for online self-management support. That brings us to the second part of this thesis with a different age-group. In order to meet the needs for self-management support of adult patients with a rheumatic disease, we investigated preferences for structure and content needs to develop an online self-management intervention. The results of this investigation are detailed in chapter 7: *What are the preferences and needs, regarding the structure and content of a person-centered online self-management support intervention for adult patients with a rheumatic disease?*

A basic assumption about self-management is that when the intervention is customized to the individual needs and situation of the patient, the patient will be more motivated, adhere better and benefit more and for a longer time (7, 22, 25) Based on the results of the comprehensive assessment of preferences and needs among adult patients in chapter 7, a web-based peer-guided and tailored self-management intervention for adults was developed. In chapter 8, *the theoretical considerations that guided the development of the intervention and its content are described. Next to that, we present the results of the usability study we conducted and the study design to measure its effectiveness.*

In the process of redeveloping care and development of self-management interventions, we experienced that small steps, driven by patients' stories and involvement of patients in all phases from development to evaluation, led to meaningful results. But we also experienced that the focus on the patient alone appears not be enough to implement these interventions effectively and sustain patient centered care in daily practice. Also we, being health care professionals, need to be supported. In chapter 9 we share our experiences with this process and we will end this chapter with implications for daily practice.

The thesis ends with a general discussion in chapter 10 with a summary of the main findings, followed by a reflection on three themes: *exploring needs for self-management support, development and feasibility and evaluating self-management interventions.* Next to that some recommendations will be presented.

REFERENCES

1. **Een gezonder Nederland (Internet).** 2014: Ministerie van Volksgezondheid, Welzijn en Sport; 2014. Available from: www.eengezondernederland.nl.

2. **Busse R, Blümel M, Scheller-Kreisen D, Zentner A.** Tackling chronic disease in Europe. Strategies, interventions and challenges. http://www.euro.who.int/__data/assets/pdf_file/0008/96632/E93736.pdf. World Health Organization, on behalf of the European Observatory on Health Systems and Politics. 2010.

3. www.reumafonds.nl.

4. **Szer I, Kimura Y, Malleson P, Southwood T.** Arthritis in children and adolescents (Juvenile Idiopathic Arthritis). New York: Oxford University Press; 2006.

5. **Prakken B, Albani S, Martini A.** Juvenile idiopathic arthritis. Lancet. 2011 Jun 18;377(9783):2138-49.

6. **Calabro A, Caterino AL, Elefante E, Valentini V, Vitale A, Talarico R, et al.** One year in review 2016: novelties in the treatment of rheumatoid arthritis. Clin Exp Rheumatol. 2016 May-Jun;34(3):357-72.

7. **Kristiansen TM, Primdahl J, Antoft R, Horslev-Petersen K.** Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: a focus group study. Musculoskeletal Care. 2012 Mar;10(1):29-38.

8. **Salaffi F, Carotti M, Gasparini S, Intorcia M, Grassi W.** The health-related quality of life in rheumatoid arthritis, ankylosing spondylitis, and psoriatic arthritis: a comparison with a selected sample of healthy people. Health Qual Life Outcomes. 2009 Mar 18;7:25,7525-7-25.

9. **Bijlsma JW, Welsing PM, Woodworth TG, Middelink LM, Petho-Schramm A, Bernasconi C, et al.** Early rheumatoid arthritis treated with tocilizumab, methotrexate, or their combination (U-Act-Early): a multicentre, randomised, double-blind, double-dummy, strategy trial. Lancet. 2016 Jul 23;388(10042):343-55.

10. **Hamnes B, Hauge MI, Kjeken I, Hagen KB.** 'I have come here to learn how to cope with my illness, not to be cured': a qualitative study of patient expectations prior to a one-week self-management programme. Musculoskeletal Care. 2011 Dec;9(4):200-10.

11. **Dwarswaard J, Bakker EJ, van Staa A, Boeije HR.** Self-management support from the perspective of patients with a chronic condition: a thematic synthesis of qualitative studies. Health Expect. 2016 Apr;19(2):194-208.

12. **Suris JC, Michaud PA, Viner R.** The adolescent with a chronic condition. Part I: developmental issues. Arch Dis Child. 2004 Oct;89(10):938-42.

13. **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 Sep;51(3):213-9.

14. **Wells CK, McMorris BJ, Horvath KJ, Garwick AW, Scal PB.** Youth report of healthcare transition counseling and autonomy support from their rheumatologist. Pediatr Rheumatol Online J. 2012 Nov 14;10(1):36,0096-10-36.

15. **Hazel E, Zhang X, Duffy CM, Campillo S.** High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. Pediatr Rheumatol Online J. 2010 Jan 11;8:2.

16. **Lawson EF, Hersh AO, Applebaum MA, Yelin EH, Okumura MJ, von Scheven E.** Self-management skills in adolescents with chronic rheumatic disease: A cross-sectional survey. Pediatr Rheumatol Online J. 2011 Dec 6;9(1):35.

17. **Suris JC, Akre C, Rutishauser C.** How adult specialists deal with the principles of a successful transition. J Adolesc Health. 2009 Dec;45(6):551-5.

18. **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 Nov;37(6):821-32.

19. **Zangi HA, Ndosi M, Adams J, Andersen L, Bode C, Bostrom C, et al.** EULAR recommendations for patient education for people with inflammatory arthritis. Ann Rheum Dis. 2015 Jun;74(6):954-62.

20. **van Eijk-Hustings Y, van Tubergen A, Bostrom C, Braychenko E, Buss B, Felix J, et al.** EULAR recommendations for the role of the nurse in the management of chronic inflammatory arthritis. Ann Rheum Dis. 2012 Jan;71(1):13-9.

21. **Smolen JS, Breedveld FC, Burmester GR, Bykerk V, Dougados M, Emery P, et al.** Treating rheumatoid arthritis to target: 2014 update of the recommendations of an international task force. Ann Rheum Dis. 2016 Jan;75(1):3-15.

22. **Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, et al.** Self-management: one size does not fit all. Patient Educ Couns. 2013 Jul;92(1):134-7.

23. **Mirzaei M, Aspin C, Essue B, Jeon YH, Dugdale P, Usherwood T, et al.** A patient-centred approach to health service delivery: improving health outcomes for people with chronic illness. BMC Health Serv Res. 2013 Jul 3;13:251,6963-13-251.

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 Oct -Nov;48(2):177-87.

25. **Kralik D, Koch T, Price K, Howard N.** Chronic illness self-management: taking action to create order. J Clin Nurs. 2004 Feb;13(2):259-67.

26. **Newman S, Steed L, Mulligan K.** Self-management interventions for chronic illness. Lancet. 2004 Oct 23-29;364(9444):1523-37.

27. **Iversen MD, Hammond A, Betteridge N.** Self-management of rheumatic diseases: state of the art and future perspectives. Ann Rheum Dis. 2010 Jun;69(6):955-63.

28. **Lorig KR, Ritter PL, Laurent DD, Plant K.** The internet-based arthritis self-management program: a one-year randomized trial for patients with arthritis or fibromyalgia. Arthritis Rheum. 2008 Jul 15;59(7):1009-17.

29. **Shigaki CL, Smarr KL, Siva C, Ge B, Musser D, Johnson R.** RAHelp: an online intervention for individuals with rheumatoid arthritis. Arthritis Care Res (Hoboken). 2013 Oct;65(10):1573-81.

30. **Stinson J, Wilson R, Gill N, Yamada J, Holt J.** A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol. 2009 Jun;34(5):495-510.

31. **Stinson JN, Toomey PC, Stevens BJ, Kagan S, Duffy CM, Huber A, et al.** Asking the experts: exploring the self-management needs of adolescents with arthritis. Arthritis Rheum. 2008 Jan 15;59(1):65-72.

32. **Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M.** Developing and evaluating complex interventions: the new Medical Research Council guidance. Int J Nurs Stud. 2013 May;50(5):587-92.

33. **Davis FD.** Perceived usefulness, perceived ease of use and useracceptance of information technology. Management Information Systems Quarterly. 1989;13(3):319-40.

34. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev. 1977 Mar;84(2):191-215.

35. **Nolte S, Elsworth GR, Newman S, Osborne RH.** Measurement issues in the evaluation of chronic disease self-management programs. Qual Life Res. 2013 Sep;22(7):1655-64.

36. **Osborne RH, Elsworth GR, Whitfield K.** The Health Education Impact Questionnaire (heiQ): an outcomes and evaluation measure for patient education and self-management interventions for people with chronic conditions. Patient Educ Couns. 2007 May;66(2):192-201.

37. **Schuler M, Musekamp G, Faller H, Ehlebracht-Konig I, Gutenbrunner C, Kirchhof R, et al.** Assessment of proximal outcomes of self-management programs: translation and psychometric evaluation of a German version of the Health Education Impact Questionnaire (heiQ). Qual Life Res. 2013 Aug;22(6):1391-403.

38. **Epstein J, Osborne RH, Elsworth GR, Beaton DE, Guillemin F.** Cross-cultural adaptation of the Health Education Impact Questionnaire: experimental study showed expert committee, not back-translation, added value. J Clin Epidemiol. 2015 Apr;68(4):360-9.

> “Choosing to do nothing:
> that is self-management
> for me..”
>
> *- Female, 18 years old, since 5 years Juvenile Idiopathic Arthritis*

CHAPTER 2

AN URGE FOR CHANGE: TRANSITIONAL CARE FOR YOUNG ADULTS WITH JUVENILE IDIOPATHIC ARTHRITIS

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INTRODUCTION

> *"I'm not worried about the new doctor; why should I? I'm 17 years old. I can deal with anything that comes my way... My pediatrician told me that I can continue to do my trampoline sport. If the rheumatologist tells me I should quit, I'll never set a foot in that hospital again!"*

This quote of Andrea, suffering from juvenile idiopathic arthritis (JIA), reflects one of the many situations young adults struggle with in living their day-to-day life with a chronic disease. In the Netherlands, 2500 children have JIA (1). Although in the past decade many new treatment options for JIA have been developed, a lasting medication-free disease remission is not always achieved. Many of these children are growing into adulthood with chronic disease (2-4). Obviously, the impact and psychosocial consequences of disease differ depending on age and stage of development of the child, also the appeal on health care providers and their role in assisting chronically ill children and their parents differs. Accumulating evidence reveals that active involvement of the (young) adult with a chronic disease has positive effects on health outcomes (5-7).

Our present health care system does not adequately accommodate an increasing diversity of patient's needs and preferences, especially when patients are in transition from pediatric to adult care because of their age. Although a growing interest in transitional care is noted (2,8-10), data on implementation of transitional care in daily rheumatology practice are scarcely reported. We therefore propose to share our experiences on redeveloping transitional care.

FACING REALITY

In the Netherlands, health policy demands health care for young adults to be transferred from pediatric to adult care at the age of 18. In University Medical Center Utrecht, every year up to 40 young adults with a rheumatic disease make a transition from the pediatrician to the rheumatologist. The departments are housed in different buildings of our hospital, connected by a 400m long subway, to be passed on a scooter. Throughout the years, the adult rheumatologists met significant problems after transition. Young adults confronted the rheumatologists with their experiences of a (too) strong and warm relation with the pediatricians. Parents attributed their pains in 'letting go' of their child and their resistance towards the expected independent behavior of their child.

Rheumatologists encountered high rates of treatment dropout and non-adherence, apparent rebellion towards caregivers and persistent dependency of young adults on their parents. Pediatricians thought that rheumatologists were not caring enough and were insufficiently thinking in terms of family-systems. Rheumatologists felt their colleagues were overprotective and insufficiently focused on self-management of the young adult. Gradually, pediatricians and rheumatologists realized mutual mistrust, lack of communication and coordination which interfered with optimal care during outpatients consultations, but also when young adults had to be admitted to an adult ward.

SHARING RESPONSIBILITY IN TRANSITION

In 2002, a project started, directed to transparency, transferability and positive labeling of the transitional process. The aim was to develop a well prepared, logical, self-management-focused and safe chain of transitional care for the young adult with JIA and his parents with equivalent contributions of both multidisciplinary teams.

First, trained coaches performed an inventory search on the outpatients' clinic of both departments, which resulted in an overview on practical skills of individual caregivers. Also, a conference was organized on the process of transition. All involved co-workers participated: a patient, a parent, pediatricians, rheumatologists, nursing staff, secretary workers, physiotherapists, occupational therapists, orthopedic surgeons, social workers, psychologists and managing staff. The conference was held outside the hospital and was opened with short introductions by patient and parent, followed by a Socratic conversation (11) with both heads of both pediatric and adult departments. Workshops on views of patient, parents and caregivers were followed by forum discussions. At the end of the day, participants expressed a collective responsibility for improving transitional care.

After the conference, a small project group with the key players in transition: a pediatrician, a rheumatologist, an occupational therapist and a clinical nurse specialist was formed. All members felt participation of the young adult to be essential. Consequently, in semi-structured focus group interviews, young adults in the ages of 16-19 years were asked for their expectancies on transitional care and care providers involved. The essential expectations are outlined in Table 1 (12).

Table 1 Expectations of young adults on transitional care and care providers, results from a focus group interview.

- Life style recommendations should fit my daily life.
- Limits to experimenting daily life should be clearly given.
- I don't want to be an exception, just simple.
- Promise open access to caregiver and organization.
- Information should meet my language.
- I want to be prepared and I want to be in charge.

Transition team

The project group members chose a step-by-step change on the job approach. They focused primarily on the appointment of a transition coordinator, i.e. a clinical nurse specialist, active in both departments, who should guarantee coordination and continuity of the processes of transition. Development of self-management skills of the young adult was felt essential. In 2004, our newly formed transition team was installed: a transition coordinator, a pediatrician and a rheumatologist. The team relied on their mutual trust and expertise, their common sense and knowledge on guiding and treating young adults with JIA. Sharing critical reflection, they believed their presence, as a team might be a good fundament for change.

DEVELOPMENT OF A TRANSITION OUTPATIENT CLINIC

Gradually, the transition outpatient clinic attained its present embedment in day-to-day care, where for every young adult in the age of 16-25 years, an individual transition plan is made. In an attempt to keep the young adult in charge, he is encouraged to describe his own goals and actions, and to ask for guidance in achieving these goals, if needed. The process of transition is thought in phases, focused on self-management. Before the age of 16, the young adults meet the transition coordinator. They are encouraged to talk for themselves instead to rely too much on their parents, stimulated to ask questions, to discuss proposed treatments and to appeal on their own abilities to solve problems. In a following phase, while the pediatrician still is in charge, young adults and their parents meet the rheumatologist who eventually will take over care. From that encounter, the rheumatologist is available for contact, if needed. When the young adults are over 18, they move to the other side of the subway, actual care is handed over to the adult outpatient clinic. The transition coordinator remains in charge, and, if needed, the pediatrician remains available for young adults, parents as well as rheumatologist.

NEW TOOLS: ACCESS AND TRANSPARENCY

In the focus group interview, young adults expressed their need for information on consequences of disease and therapy in day-to-day life. None of the Dutch Internet sites were felt to be satisfying because they are directed towards adults over 30 years. Consequently, together with the patients themselves, a website (www.jong-en-reuma.nl) was developed and implemented. The young adults decided the objectives of the website, they told their stories and made video’s about dealing with consequences of their diseases.

Joint use of the same hospital electronic patient file by both departments made it possible to develop an online portal. Patients may send an eConsult to the transition coordinator and are able to monitor their own situation by self-tests and diaries. Also, they may check their latest medicine- and treatment advices from the multidisciplinary team. Young adults judge both tools to be successful in filling gaps in information, in providing them a sense of control and involvement in treatment and care (13).

Encouraged by and in close collaboration with patients who asked for self-management skills, a training program (www.reumauitgedaagd.nl) was developed, with topics as ‘feeling blue’, ‘relations and intimacy’ and ‘having control over your live and arthritis’. The program can be followed online, in a six-week group program with chat sessions, a discussion board and individual exercises. The program can also be followed in a weekend in a holiday resort. Young adults with a rheumatic disease lead both programs. The Dutch Rheumatism Patient League and the University Medical Center specially trained them.

REFLECTIONS AND INTENTIONS

Small steps driven by day-to-day practice can lead to meaningful results. Key persons on the working-floor succeeded in open dialogue, involving colleagues and managers in contemplation on the changing processes. All of the described interventions are developed in close cooperation with our patients and the Dutch Rheumatism Patient League. Evaluations with patients and their parents confirm an actual fitting of the expectations of the young adults and offered interventions. An evaluation study (13) shows that young adults not only are feeling involved but also want to be taught in self- management.

From the start, we strongly felt not only patients but also all care providers should be involved on our journey of change (14). Ever since, seminars were held on a regular base for reflection, feedback and communication. In time, these seminars included presentations and discussions on shared knowledge on care and contents of medicine.

At present, we are working on expanding evidence based knowledge on this subject by carrying out a randomized controlled study to the effectiveness of the online self-management training as an addition to the transition outpatient clinic.

REFERENCES

1. **Szer I, Kimura Y, Malleson P, Southwood T.** Arthritis in children and adolescents. Juvenile idiopathic arthritis. New York: Oxford University Press; 2006.

2. **Hazel EM, Zhang X, Duffy CM, Campillo S.** High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. Pediatric Rheumatology 2010;8:2.

3. **Shaw KL, Southwood TR, McDonagh JE.** User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. Rheumatology 2004;43:770-778.

4. **Wagner-Weiner L.** Pediatric rheumatology for the adult rheumatologist. J Clin Immunol 2008;14:109-119.

5. **Bodenheimer T, Lorig K, Homan H, Grumbach K.** Patient self-management of chronic disease in primary care. JAMA 2002;288:2469-2475.

6. **Marks R, Allegrante JP, Lorig K.** A review of synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (Part11). Health Promotion Practice 2005;6:148-156.

7. **Stinson JN, Toomey PC, Stevens BJ, Kagan S, Duffy CM, Huber A, Malleson P, McGrath PJ, Yeung RSM, Feldman BM.** Asking the experts: exploring the self-management needs of adolescents with arthritis. Arthritis and Rheumatism 2008;59:65-72.

8. **McDonagh, JE.** Young People First, Juvenile idiopathic arthritis second: transitional care in rheumatology. Arthritis and Rheum 2008;59:161-168.

9. **Rosen DS, Blum RW, Britto M, Sawyer SM, Siegel DM.** Transition to adult health care for adolescents and young adults with chronic conditions: position paper of the Society for Adolescent Medicine. J Adolesc Health 2003;33:309-311.

10. **Tucker L, Cabral D.** Transition of the adolescent patient with rheumatic disease: issues to consider. Pediatr Clin North Am 2005;52:641-652.

11. **Schlozman SC.** Using a Socratic Dialogue to tackle thorny issues of psychiatry. Psychiatric Health 2009;84(11):1562-1566.

12. **Gamel CJ, Bijl van der JJ, Ammerlaan JW.** A clinical problem: transition of the young adult with juvenile idiopathic arthritis. Results of a focus group interview. Utrecht: University of Utrecht, Department of Nursing Science; 2005.

13. **Ammerlaan JW, Scholtus LW, Drossaert CHC, Os-Medendorp H, Prakken BJ, Kruize AA, Bijlsma JWJ.** Views and experiences of patients concerning two eHealth applications for young adults with juvenile idiopathic arthritis: views and experiences of patients. Manuscript in preperation.

14. **Berwick DM.** Escape Fires, Lessons for the future of health care. New York: The Commonwealth Fund; 2002.

> *“I like to be independent*
> *but that does not mean*
> *I should not ask for*
> *help. I just struggle*
> *to accept this.”*

- Male, 21 years old, since 9 years Juvenile Idiopathic Arthritis

CHAPTER 3

FEASIBILITY OF A WEBSITE AND A HOSPITAL-BASED ONLINE PORTAL FOR YOUNG ADULTS WITH JUVENILE IDIOPATHIC ARTHRITIS: VIEWS AND EXPERIENCES OF PATIENTS

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ABSTRACT

Background To improve knowledge and to encourage active involvement of young adults with juvenile idiopathic arthritis (JIA), an informative website with written and video information and an online portal with access to the personal medical record, self-monitoring, and e-consult functionalities were developed. Before implementing these applications in daily practice, it is important to gain insight into their feasibility in terms of ease of use, perceived usefulness and intention to use.

Objective The aim of this study was to evaluate and to examine the feasibility of the website and the online portal for young adults with JIA.

Methods A qualitative, feasibility study was conducted among the first users: 13 young adults with JIA. After provided access to the website and online portal, patients were interviewed on perceived usefulness, ease of use, and intention to (re)use the applications.

Results Participants in the study considered the website and online portal as useful and easy-to-use. New medical information and feedback would motivate them to revisit the applications again. On the website, videos showing other young adults, telling how they handle their condition, were found as the most useful. On the portal, access to their medical records was most appreciated: it made the young JIA patients feel in control and it helped them monitor symptoms and disease activity. e-Consults were thought to facilitate communication with physicians.

Conclusion The young adults considered both the website and the online portal as feasible, but they also had valuable suggestions to improve accessibility and use. Based on these findings, a news and event section was added on the website and a direct link was made to a discussion board and social media. To provide and support health information, the website is actively used in daily care. Considering the online portal, the use of self-monitoring tools and e-Consult can be stimulated if there is direct linkage to treatment and feedback from the multidisciplinary team. Feasibility testing, before implementing the website and online portal in daily practice, has proven to be a valuable step. Results led to improvements in terms of integration into standard care and topics for further research.

INTRODUCTION

Living with a chronic rheumatic disease is challenging at any age. However, these challenges may be particularly difficult for young adults with juvenile idiopathic arthritis (JIA), since their chronic condition and treatment affect both physical and socio-emotional development (1,2). One of the main challenges young adults with JIA have to deal with is to take over from their parents the responsibility for their own illness and treatment: they have to become a self-manager (3,4).

In general it is believed that eHealth might contribute to self-management, especially for young adults (5-7). For this study, we followed the definition of eHealth by Eng: "The use of emerging information and communication technology, especially the Internet, to improve or enable health and health care" (8). The use of health information websites and eHealth applications, including online portals in disease management, disease prevention, and health promotion is well-reported (9,10). Unfortunately, many eHealth projects fail to survive the pilot phase and studies that focus on the effectiveness of eHealth applications often do not show any long term effects (9-11). Also, much is developed but not everything is used (10,12,13). For the actual use and acceptance, evaluation and testing of the feasibility before implementing the techniques in daily practice is crucial (11,14-16). Several frameworks have been introduced to increase the uptake and to examine the feasibility of eHealth applications. Among them, the technology acceptance model (TAM) (15,17) stands for a prediction and explanation model of the end-users reaction to a technological innovation. The model states that use or acceptance of a particular innovation can best be predicted by an individual's intention to (re) use the innovation. A comparative model, the Holistic Framework of Gemert (11) suggests that developers of eHealth applications should be aware of interactions between technology, people, and their social-cultural environment. Involvement of end-users in developing eHealth applications is considered to be one of the crucial aspects of acceptance of the tools themselves.

In our specialized transition outpatient department, young adults with JIA and their parents receive multidisciplinary care from a pediatrician and a clinical nurse specialist, in order to support the process of acquiring self-management skills and to guarantee a well-coordinated, continuous process of health care between child and adult (5). To improve knowledge of the disease and to encourage active involvement in this transition process, we developed an informative website and a hospital-based online portal. The website contains information about medical issues and how to deal with consequences of having a rheumatic disease, such as feeling blue, exercise, work, study, relationships and intimacy.

With our secured online portal the young adult has direct access to his medical record; he is enabled to send an e-Consult and may use self-monitoring tools including activity diaries and pain questionnaires. These applications were developed in close cooperation with patients to fit the applications to the specific needs and preferences of this group. Therefore, young adults of the Dutch Youth Network of Rheumatology were asked to perform a central role. In interactive workshops, organized by the multidisciplinary team of the transition outpatient clinic, they determined, together with the professionals, the content and structure of the website and portal.

Both applications may be promising to reach young adults with arthritis and to stimulate their self- management behavior, given their access to and high rates of use of the Internet (7,18,19). Before implementing these applications in daily practice, it is important to gain insight into their feasibility. Therefore, the aim of this study is to evaluate and to examine the feasibility of the website and the online portal for young adults with JIA.

METHODS

Design

A qualitative feasibility study with semistructured, (audiotaped) interviews was conducted among the first users of both eHealth applications in order to explore the views and experiences of the young adult JIA patients with regard to the feasibility outcomes: ease of use, perceived usefulness, and intention to use. These outcomes are part of the technology acceptance model (17). The model states that use or reuse of a particular technical innovation can best be predicted by an individual's intention to (re)use the innovation. This intention is determined by two components: perceived ease of use and perceived usefulness. Perceived ease of use means the degree of ease, associated with the use of the applications. Perceived usefulness can be defined as the degree in which a person believes that using the technical innovation would enhance his or her personal situation.

Participants were asked to use the applications for three months "as needed", without specific instructions, and were interviewed just before or after their subsequent visit to the clinic.

Study Population

Patients diagnosed with JIA, aged between 16 and 25 years old, being able to read and write in Dutch, with access to a home-based computer with Internet were included in the study. Young adults who already participated in the development of the website and/or portal were excluded. Recruitment took place at the transition outpatient department of the University Medical Center Utrecht, the Netherlands. All patients who visited the transition outpatient department within a period of three months were asked by their pediatrician or rheumatologist to participate in the study. An information letter and informed consent was handed out which they could return by post.

A convenience sample was used: all patients who returned the informed consent within two weeks after their visit were included in this study. Because it was a feasibility study with a qualitative design, we aimed to include at least 12 participants. According to the ethics guidelines of our hospital, the nonexperimental and noninvasive nature of this study made ethical approval unnecessary.

Description of the eHealth Applications

The website and online portal are designed to improve knowledge, self-management skills, and involvement in treatment and care. The *website* (in Dutch) (20) is publicly accessible and contains information and tips on five themes: 1) treatment and medication; 2) physical exercise, holidays, alcohol, and drugs; 3) relations, sexuality, and pregnancy; 4) dealing with pain, fatigue, and emotions; and 5) study and work. In addition, videos and written stories from other young adults talking about their lives with JIA are presented. For an impression of home page of the Dutch website, see Figure 1.

Figure 1 Homepage of the Dutch information website

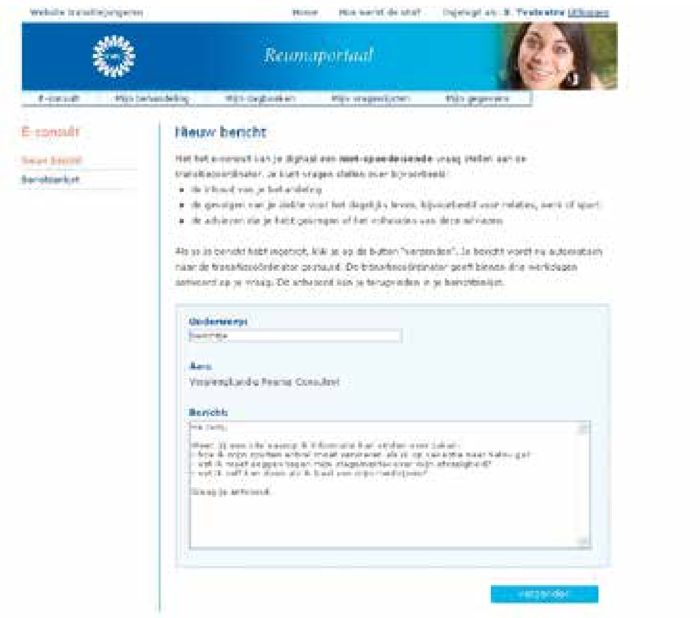

Figure 2 Portal and e-consult tool

The portal (in Dutch) (21) is only accessible to JIA patients of the University Medical Center Utrecht with a personal log-in code. By using the portal, patients may have access to the following tools: 1) e-Consult, through which patients communicate with a clinical nurse specialist; 2) their own medical record, including all written reports of physicians, laboratory results, present medications and appointments with the outpatient clinic; and 3) online self-monitoring, in which patients can fill out self-tests on pain and activities and diaries to monitor their disease. For additional general disease information patients are referred to the website (20). For an impression of the portal and the e-Consult tool, see Figure 2. The content of website and portal are outlined in table 1.

Data Collection

First, participants were asked to complete a brief questionnaire on demographics, illness characteristics, and their general Internet use. Subsequently, a semi structured interview was conducted at the transition outpatient department in a separate, quiet room, by a young independent interviewer (LWS), who was not involved in the care of the young adult.

The first part of the interview was directed at the informative website; in the second part patients' opinions and experiences about the portal were assessed. For both applications, open-ended questions, deriving from TAM were used to get insight into ease of use, perceived usefulness, and intention to (re)use (see Table 2 for an overview of questions). The interviewer encouraged participants to elaborate on all issues, using probes such as: "why?", "please, explain…", "Can you give an example?", "Can you think of any other …" At the end, the participant was asked to grade the applications with a number, where "zero" stands for "not useful" and "ten" stands for "the most useful".

All interviews were audiotaped. Field notes were made immediately after the interview to record the interviewer's impression of the responses to the questions and comfort level of the participants with the interview process. An interview lasted between 30 and 45 minutes.

Table 1 Content of website and portal

WEBSITE, PORTAL	CONTENTS
Website	
Transition	▪ What is transition? ▪ Practical information about the departments, multidisciplinary team
About JIA	▪ What is JIA? ▪ Treatment
Leisure	▪ Exercise and sports ▪ Alcohol and drugs ▪ Holidays/vacation
Love, sex, and kids	▪ Parents and friends ▪ Sex and intimacy ▪ Having kids ▪ Contraception, pregnancy, and heredity
Feeling blue	▪ Having a rheumatic disease ▪ Feeling tired or having pain ▪ Relaxation ▪ Feeling depressed ▪ Adherence and treatment advices
School, work, and money	▪ School ▪ Work ▪ Social insurance
Portal	
Medical record	▪ Appointments ▪ Laboratory results ▪ Written reports of physicians ▪ Actual medication
e-Consult	▪ Sending and receiving e-Consults
Online self-monitoring	▪ Medication diary ▪ Diary on fatigue ▪ Pain score ▪ Activities

Table 2 Interview questions on feasibility of the website and portal

	INTERVIEW QUESTIONS
Use	▪ Have you visited the site/portal? ▪ Did you visit the site/portal with a specific reason? ▪ Which parts did you visit?
Perceived ease of use	▪ Did you experience any difficulties in use of the site/portal?
Perceived usefulness	▪ How useful is the site/ portal for you? ▪ Which parts are most/ least useful? ▪ Can you describe any benefits, or drawbacks? ▪ What are the most and least appealing parts? ▪ Do you miss anything?
Intention to use	▪ Would you visit the site/portal again? ▪ Would you recommend the site/portal to others?
Suggestions	▪ Do you have any suggestions for improvement?

Data Analysis

All interviews were transcribed verbatim. A generic qualitative approach was used for data analysis, including coding, constant comparison, and categorizing. Data collection and analysis were handled as an iterative process (22). The first participant was interviewed with open-ended questions on feasibility and suggestions for improvement of the website and portal. In the following interview, the feasibility was asked again and the suggestions made by the former participant were checked. Then the participant was asked to add his own suggestions. The interviewer checked every interview if new themes emerged and asked further about these themes until saturation was achieved and no new information was obtained. The interviewer (LWS) and a member of the research team (JWA) reviewed and coded all transcripts independently. Relevant fragments were first categorized into the main concepts of TAM and were further categorized into subthemes, using inductive analysis. Results were discussed on several occasions and differences were discussed until consensus was reached.

RESULTS

Participants

Thirty-eight eligible patients received an information letter and informed consent. Patients (n=19) who returned a signed informed consent were included in the study. No information is available from the other 19 patients who did not respond to the invitation to participate in the study. Six patients dropped out after giving their informed consent, with reasons of exacerbation of illness, vacation, and too busy with school. Finally, 13 participants received the URL address of the website and a log-in code for the portal.

The mean age of the sample was 20 years (range: 17-22 years) and consisted of 12 women and 1 man. Most of the participants (n=12) were being treated by a rheumatologist. The mean duration of illness was 8 years (range: 2-20 years). Of the 13 participants, 11 were in high school, and two participants were gainfully employed. They all used the Internet on a daily basis, most participants (n=10) with an average of over two hours a day.

Informative Website

Of the 13 patients, 12 indicated having visited the website several times. One participant visited the website only once. During their visits, all participants had read at least a part of the written information. Most (n=11) had seen one or more videos and read written life stories. The primary reason for (re)visiting the website was curiosity. Other reasons included searching for specific information or for experiences of other patients.

Perceived Ease of Use

Participants did not experience barriers visiting the website. Two participants had issues with loading a video, in retrospect due to their own computer and Internet connection.

Perceived Usefulness

Participants appreciated the website and graded it with a 7.6 (min 6.5, max 9.5) on a scale from 1 to 10. They found the design of the website appealing and the information practical, clear, easy to read, and well-targeted to young adults.

> *"This really is for younger people. I sometimes look at the SLE site but that is mostly for people who are 40, 50 and 60 years of age and their problems. That is not really my cup of tea. This really reveals itself to be more for younger ones. (Female, 21 years)"*

Of all the elements, the videos and life stories were thought to be the best. They enabled recognition and showed new ways to deal with solving problems related to the condition. Patients experienced support and recognition in these stories.

> *"I sometimes think that I am the only one with arthritis. There aren't many with the same problems. Other people don't understand this; they don't see anything on the outside. It is nice to hear people of the same age talking about this problem. That is what I have too! You won't have to say it yourself; somebody else says it for you and I feel the same things." (Female, 21 years)*

> *"It really helped me, especially how to cope with fatigue and pain and how to solve it." (Female, 20 years)*

> *"You can hear and read the experiences from others, things they had to cope with, how they dealt with them. Often you can learn from their experiences, because you recognize them." (Female, 20 years)*

All six themes (in Table 1) on the website were appreciated positively. The participants thought nonmedical themes such as dealing with pain, fatigue, and emotions, physical exercise, holidays, alcohol and drugs, sexuality, study and work, were most appealing.

Some young adults missed detailed information on new developments in medication. Others missed a forum. They thought a link to an existing Dutch discussion board for young adults with JIA and to detailed information about medication on other reliable websites would be valuable supplements.

All participants, except for the youngest, indicated that no or only little new information was added to their knowledge and skills by their visit to the website. Nevertheless, the website was considered useful as a confirmation of what they already knew.

> *"I read that it is important to structure your medication. You have to learn that this is important. Also that before visiting a doctor it's important to make a list with questions. All things you know but important to read again." (Female, 21 years)*

> *"I think I am beyond that age. I was 15 when I got arthritis. I think I would have had more benefit from this between my 15th and 18th. I'm beyond that now. I know what it is; I know how it influences my lifestyle, my alcohol use during the holidays. So I recognize all the subjects but I already have my own opinion about these. It doesn't add anything for me." (Female, 22 years)*

The youngest participant felt she found new and relevant information on the website.

> *"In the hospital, they always talked about JIA. I asked myself: what does that mean? I felt really stupid. I didn't dare to ask. I read on the website that it was Juvenile Arthritis, my disease." (Female, 17 years)*

Finally, participants indicated that the website might be useful for questions of their relatives:

> *"It is very convenient for yourself and your surroundings. My friend sometimes wants to know more and for him it is also a good site to refer to. It is clearly explained what the disease is and how to cope with it." (Female, 21 years)*

In summary, the website was considered useful for three reasons: 1) to find or re-read information (mostly already known), 2) to help in explaining the disease and its consequences to others, but most importantly, 3) to find recognition, and to see that other young people struggle with similar problems.

Intention to Use
Half of the participants intended to revisit the website; the other participants however indicated that they would only revisit the website if new information were added, if news was added or if their personal situation changed. They all recommended the website to other young people with a rheumatic disease.

Online Portal
All participants (n=13) used the portal for a period of two to three months. Within this relatively short time, access to medical records was used most often, whilst fewer participants used the tools for e-Consult (n=4) and four other participants used self-monitoring (n=4). Most of the participants logged in just after a visit to the hospital. "Curiosity about what the doctor or nurse had written" was the main reason to check their medical record. One participant logged in just before her visit. She wanted to prepare herself and read what was discussed during her last consultation.

Perceived Ease of Use
The only problem participants reported (n=5) was the log-in code being too long and complex to be remembered, whereas the code could not be changed. Consequently, they used the portal less often than they wanted.

Perceived Usefulness
Participants graded the portal with a mean grade of 7.8 (min 7.5, max 8.0) on a scale from 1 to 10, and mentioned several advantages (see Table 3).
Four participants used e-Consult, and they considered e-Consult helpful because of easier and better communication with the hospital. It enabled the participants to think more about the questions and to explain it better.

Most participants could not think of any disadvantages of the portal; although one participant suggested other people might think access to medical data is not safe. One participant indicated that too much information about her health had sometimes overwhelmed her.

Table 3 Usefulness of the different parts of the portal, as mentioned by the participants.

MENTIONED EFFECTS, ADVANTAGES	EXAMPLE QUOTE
Access to medical record	
Feeling more in control	*"It's very functional to be able to check your appointment, to check the blood values and what was said during a consult. Being able to control your own treatment." (Female, 22 years)*
Keeping pace with illness activity	*"You have the most recent values and you are able to compare them with the latest values. I once had a liver condition and could see that on the values, it was obvious." (Female, 20 years)*
Reminder tool	*"Recently I wondered what we agree on as treatment and medication and now I was able to look it up. Also when I had an appointment I found out that the appointment was half an hour earlier than I thought." (Female, 20 years)*
Being able to share data with others (parents)	*"My parents never accompany me and now I am able to show them the summary of the consult." (Female, 21 years)*
e-Consult	
Easier communication with the hospital	*"To send a notice is easier than phoning to the hospital. I send a message and it doesn't matter if it's inconvenient. If I call, it might be inconvenient." (Female, 22 years)*
Better communication with doctor/nurse	*"You're able to think things through and to explain it better." (Female, 18 years)*
Self-monitoring	
Facilitates communication with physician	*"If things become worse suddenly, it is handy because you know when it went wrong. The doctor is also able to see when it went wrong. Because sometimes I'm ill and when he asks I don't know when I was ill." (Female, 19 years)*
Provides insight into (the course of) the disease	*"Yes, especially when I thought I'm in pain and very tired, it immediately asks what have you done to prevent it and every time you're not able to answer this question you know that you should have been less active." (Female, 20 years)*

> *"The first time I thought I wish I hadn't read it. That also applies to the website. However it is good to read things about it. It is about you and therefore scary. On the website it is about different people. The portal is very personal." (Female, 17 years)*

Participants mentioned various aspects of usefulness which are summarized in Table 3.
The access to their medical records was considered the best feature of the portal, because it enabled participants to check their appointment, to see all laboratory results, to re-read treatment plans, feeling more in control of their own treatment, but also keeping track of the progression of the illness was expressed. The portal also facilitated sharing these data with their parents.

The opinions on the usefulness of the self-monitoring tools were more diverse. Some participants indicated them as useful to discover why they experience more symptoms at certain times. Others stated that it provides insight into the course of their problems like having pain or being tired.
A few young adults stated that they did not like filling in diaries or monitoring tools and did not want to be too occupied with their condition.

Intention to Use
All participants intended to use the portal again, especially for access to their medical records and using e-Consult. Some participants were certain they would not use the diaries and monitoring tools; other participants would use these in case of exacerbation of their JIA. Some thought they would use the tools only if their physician asked for it, or their physician would use the information during the consultations.

Participants suggested the portal might be even more attractive if elements were added, including a facility for online appointments, access to x-rays, printing forms for blood collection before the consult, and an overview of physicians and clinical nurse specialists and their consulting hours.

DISCUSSION

Principal Findings

In this study, the first users of an informative website and an online portal with opportunities for e-Consult, access to medical records, and tools for self-monitoring were asked to evaluate the feasibility of these applications on ease of use, perceived usefulness and intention to (re)use. Both eHealth applications were found easy-to-use, and the young adults considered them as "clear and understandable" and useful.

Informative Website
On the website, the videos were considered as visually appealing and interactive and as a more pleasant way to learn compared to written information. This appreciation of videos as a source of information was also shown in similar studies in patients with JIA of the same age (6, 14). After seeing other young adults with JIA talking about their lives, some participants felt able to deal/cope with their own condition more adequately. This might indicate effects of modeling and persuasive information, which in fact are methods for enhancing self-efficacy and self-management behavior (23, 24). Most participants indicated they did not encounter information on the website which was new to them, which might be explained because they were relatively experienced patients, as

indicated by mean disease duration. The website might be especially helpful for the relatively inexperienced patients, which would be a valid reason for developing these tools preferably for younger patients and for patients recently diagnosed with JIA. These results are in line with data concluding that patients who feel insecure, concerned or inexperienced are more in need of health related information (25, 26). Otherwise, participants from this study indicated that changes in their personal situation and new information might encourage the patient to visit the website again.

Based on these results, the website is now actively used at the transition outpatient department as the main source to provide and support health information. The young adult is stimulated by all members of the team to use the site for adequate, additional information. Also, a section with news and events is added to the website in order to stimulate re-visiting the website. In these sections, new information on “being young and having a rheumatic disease” is regularly posted. For this purpose, we created links to the website of the Dutch Youth Network on Rheumatology (27) and the Dutch Arthritis Foundation.

Online Portal

Participants indicated that access to their medical record was the most useful tool of the portal, increasing their feelings of being in control and helping them to monitor their symptoms and disease activity. Similar results are also found in a large study on access and usage of Web-based communication among adult patients with a chronic disease (28, 29). In our study, young adults “felt more in control” because they could check their appointments, blood values, and “what was said during a consult”. Feeling more in control as part of perceived usefulness in relation to access to a patient portal was also positively rated and recognized by adults with type 2 diabetes (30), and adults with rheumatoid arthritis (31).

Participants in our study expressed the thought that e-Consult may lead to easier and better communication with physicians. This result is also reported in other studies on use and acceptance of electronic communication among patients with cancer (32), where email or e-Consult were preferred over telephone contact. The finding that most of our participants did not use the e-Consult or self-monitoring tools might be attributed to the short period of time between receiving the log-in code and the interview (two to three months).

Our finding that participants were only moderately enthusiastic about self-monitoring tools is in concordance with qualitative data on the development of a health information technology mode: the uptake of self-monitoring tools and also e-Consult is stimulated if both have a linkage to treatment and to feedback from physicians (25, 33). Consequently, in the implementation phase, the multidisciplinary team will stimulate the young adult to use the tools. Also, the team will address active responses.

Limitations of the Study

Limitations for this study include participation of only a small group of first users of the applications. Although the sample seems to be representative as to age and illness duration (34), given its small size along with the aim of this study, generalization to the whole population of young adults with JIA is limited. Because it was a convenience sample, no effort was made to recruit the same amount of men and women, which resulted in a high percentage of women in this study. Although JIA is more prevalent in women (2), because of the small groups we cannot make a useful comparison. More research on the results within another group of young adults with JIA or another chronic disease is needed. The participants had a limited amount of time to use the applications; so only their first experiences with the website and portal were collected. No information is available on the patients who did not respond to the invitation to participate in this study.

Data collection was performed by rather time consuming semi structured interviews. Within the context of this study, we chose this method to meet the young adults, ask their opinions, and to have the opportunity to encourage them to elaborate on issues of feasibility. In case of a larger sample size, other methods including an online focus group might have been suitable. Focus groups also used to discover perceptions of the participants, often on a limited number of issues, may facilitate the interaction between participants. A recent study on testing feasibility of an eHealth intervention for binge drinking among young people used an online focus group to explore acceptability among 110 adolescents and young adults (34).

For practical reasons, to avoid extra travel time or time lost at school, we chose to plan the interviews before or after a consult. Because this choice might influence our results, the interviews were set up in a separate room, by a young independent interviewer who was not involved in care. Because of their duration of disease, participants were well-accustomed to usual care in our hospital.

Feasibility Testing

With this study we highlighted the importance of conducting feasibility testing prior to implementation of eHealth applications in daily practice. In information science, the involvement of the end-user in the development process of eHealth applications is considered to be a crucial factor for the actual uptake of the applications in daily life (35, 36) . A review of social media in adolescent and young adult health care underpinned our results that targeting health information, based on the needs of the specific group, could stimulate the actual use of an eHealth tool (37).
In this study, we focused on feasibility testing but in the actual development of website and online portal, we collaborated actively with young adults as well. They decided to a large extent the content, layout, and structure of both applications, based on their needs and preferences. The positive outcomes of our study may be attributed to this earlier collaboration. The TAM model, used in this study (17), has already evolved towards a Health Information Technology Acceptance Model (HITAM) (25) ; adding (behavioral) factors such as HIT self-efficacy and health beliefs/concerns.

Based on the results of this study and the high use and acceptance within the group of young adults, we can conclude that the Internet can be a promising tool to provide health information and improve self-management among young adults with rheumatic diseases. This point is also recognized in other studies of young adults with a chronic disease like HIV (38) or Spina Bifida (39) on use and preferences regarding eHealth. Also Stinson's study (14) of young adults with JIA showed similar outcomes. Young adults with JIA believe that *"Web-based interventions are a promising avenue to improve the accessibility and availability of JIA management strategies"* (14). Also mentioned in the HITAM is the "subjective norm", indicating that when a HIT (eg, website or portal) is imbedded in social networks, consumers are more likely to have a positive attitude towards acceptance. Based on these assumptions and the results of our study, we linked the website to a discussion board and social media. However, here some concerns have to be expressed. Several studies show that most young adults primarily use the Internet and social media for contact with peers, for home work or for leisure activities (37, 39-41). Little is known about the actual use of these media in relation to this group and health care. Future research is needed to gain insight into the use and acceptance of these media in relation to health.

Further, website and portals are increasingly used in health care, most in addition to usual care. Further research into the consequences of replacing parts of usual face-to-face care by eHealth interventions, including cost-effectiveness, will be needed.

CONCLUSION

The young adults appreciated both website and online portal as feasible but they also had valuable suggestions to improve accessibility and use. Based on these findings, a news and event section was added on the website and a direct link was made to a discussion board and social media. To provide and support health information, the website is actively used in daily care. As concerns the online portal, the use of self-monitoring tools and e-Consult can be stimulated if there is a direct linkage to treatment and feedback from the multidisciplinary team.

Feasibility testing, before implementing the website and online portal in daily practice, has proven to be valuable. Results led to improvements in terms of integration in usual care and topics for further research.

REFERENCES

1. **Tucker LB, Cabral DA.** Transition of the adolescent patient with rheumatic disease: issues to consider. Rheum Dis Clin North Am 2007 Aug;33(3):661-672. (doi: 10.1016/j.rdc.2007.07.005)

2. **Prakken B, Albani S, Martini A.** Juvenile idiopathic arthritis. Lancet 2011 Jun 18;377(9783):2138-2149. (doi: 10.1016/S0140-6736(11)60244-4)

3. **Shaw KL, Southwood TR, McDonagh JE, British Paediatric Rheumatology Group.** Developing a programme of transitional care for adolescents with juvenile idiopathic arthritis: results of a postal survey. Rheumatology (Oxford) 2004 Feb;43(2):211-219. (doi: 10.1093/rheumatology/keh018)

4. **Wagner-Weiner L.** Pediatric rheumatology for the adult rheumatologist. J Clin Rheumatol 2008 Apr;14(2):109-119. (doi: 10.1097/RHU.0b013e31816b4460)

5. **Ammerlaan JW, Scholtus LW, Bijlsma Hans J W J, Prakken BJ, Kruize AA.** An urge for change: transitional care for young adults with juvenile idiopathic arthritis. Patient Educ Couns 2013 Jul;92(1):127-129. (doi: 10.1016/j.pec.2013.02.006)

6. **Stinson J, Wilson R, Gill N, Yamada J, Holt J.** A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol 2009 Jun;34(5):495-510. (doi: 10.1093/jpepsy/jsn115)

7. **Gray NJ, Klein JD, Noyce PR, Sesselberg TS, Cantrill JA.** Health information-seeking behaviour in adolescence: the place of the internet. Soc Sci Med 2005 Apr;60(7):1467-1478. (doi: 10.1016/j.socscimed.2004.08.010)

8. **Eng TR.** eHealth research and evaluation: challenges and opportunities. J Health Commun 2002 Jul;7(4):267-272. (doi: 10.1080/10810730290001747)

9. **Flynn D, Gregory P, Makki H, Gabbay M.** Expectations and experiences of eHealth in primary care: a qualitative practice-based investigation. Int J Med Inform 2009 Sep;78(9):588-604. (doi: 10.1016/j.ijmedinf.2009.03.008)

10. **Webb TL, Joseph J, Yardley L, Michie S.** Using the internet to promote health behavior change: a systematic review and meta-analysis of the impact of theoretical basis, use of behavior change techniques, and mode of delivery on efficacy. J Med Internet Res 2010;12(1):e4. (doi: 10.2196/jmir.1376)

11. **van Gemert-Pijnen Julia E W C, Nijland N, van LM, Ossebaard HC, Kelders SM, Eysenbach G, et al.** A holistic framework to improve the uptake and impact of eHealth technologies. J Med Internet Res 2011;13(4):e111. (doi: 10.2196/jmir.1672)

12. **Nijland N, van GPJ, Boer H, Steehouder MF, Seydel ER.** Evaluation of internet-based technology for supporting self-care: problems encountered by patients and caregivers when using self-care applications. J Med Internet Res 2008;10(2):e13. (doi: 10.2196/jmir.957)

13. **Nijland N.** Towards a holistic framework for sustainable eHealth technologies. Thesis, University of Twente. 2011. Grounding eHealth URL: http://www.utwente.nl/bms/pgt/bestanden/nijlandproefschrift.pdf

14. **Stinson JN, Toomey PC, Stevens BJ, Kagan S, Duffy CM, Huber A, Yeung Rae S M, et al.** Asking the experts: exploring the self-management needs of adolescents with arthritis. Arthritis Rheum 2008 Jan 15;59(1):65-72. (doi: 10.1002/art.23244)

15. **Holden RJ, Karsh BT.** The technology acceptance model: its past and its future in health care. J Biomed Inform 2010 Feb;43(1):159-172. (doi: 10.1016/j.jbi.2009.07.002)

16. **Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M.** Developing and evaluating complex interventions: the new Medical Research Council guidance. Int J Nurs Stud 2013 May;50(5):587-592. (doi: 10.1016/j.ijnurstu.2012.09.010)

17. **Davis FD.** Perceived Usefulness, Perceived Ease of Use, and User Acceptance of Information Technology. MIS Quarterly 1989 Sep;13(3):319. (doi: 10.2307/249008)

18. **Hargittai E, Hinnant A.** Digital Inequality: Differences in Young Adults' Use of the Internet. Communication Research 2008 Aug 04;35(5):602-621. (doi: 10.1177/0093650208321782)

19. **Stinson J, McGrath P, Hodnett E, Feldman B, Duffy C, Huber A, et al.** Usability testing of an online self-management program for adolescents with juvenile idiopathic arthritis. J Med Internet Res 2010;12(3):e30. (doi: 10.2196/jmir.1349)

20. **jong-en-reuma.** URL: http://www.jong-en-reuma.nl/

21. **umcutrecht (in Dutch).** URL: https://www.umcutrecht.nl/nl/Ziekenhuis/Mijn-UMC-Utrecht

22. **Holloway I, Wheeler S.** Qualitative research in nursing. Oxford: Blackwell Science; 2002.

23. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev 1977 Mar;84(2):191-215.

24. **Barlow JH, Wright CC, Turner AP, Bancroft GV.** A 12-month follow-up study of self-management training for people with chronic disease: are changes maintained over time? Br J Health Psychol 2005 Nov;10(Pt 4):589-599. (doi: 10.1348/135910705X26317)

25. **Kim J, Park HA.** Development of a health information technology acceptance model using consumers' health behavior intention. J Med Internet Res 2012;14(5):e133. (doi: 10.2196/jmir.2143)

26. **Rice RE.** Influences, usage, and outcomes of Internet health information searching: multivariate results from the Pew surveys. Int J Med Inform 2006 Jan;75(1):8-28. (doi: 10.1016/j.ijmedinf.2005.07.032)

27. **Youth-R-Well.** URL: http://www.youth-r-well.com/

28. **Silvestre AL, Sue VM, Allen JY.** If you build it, will they come? The Kaiser Permanente model of online health care. Health Aff (Millwood) 2009;28(2):334-344. (doi: 10.1377/hlthaff.28.2.334)

29. **Hassol A, Walker JM, Kidder D, Rokita K, Young D, Pierdon S, et al.** Patient experiences and attitudes about access to a patient electronic health care record and linked web messaging. J Am Med Inform Assoc 2004;11(6):505-513. (doi: 10.1197/jamia.M1593)

30. **Osborn CY, Mayberry LS, Wallston KA, Johnson KB, Elasy TA.** Understanding patient portal use: implications for medication management. J Med Internet Res 2013;15(7):e133. (doi: 10.2196/jmir.2589)

31. **van der Vaart Rosalie, Drossaert Constance H C, Taal E, Drossaers-Bakker KW, Vonkeman HE, van de Laar Mart A F J.** Impact of patient-accessible electronic medical records in rheumatology: use, satisfaction and effects on empowerment among patients. BMC Musculoskelet Disord 2014;15:102. (doi: 10.1186/1471-2474-15-102)

32. **Gerber DE, Laccetti AL, Chen B, Yan J, Cai J, Gates S, Lee Simon J Craddock.** Predictors and intensity of online access to electronic medical records among patients with cancer. J Oncol Pract 2014 Sep;10(5):e307-e312. (doi: 10.1200/JOP.2013.001347)

33. **Van der Vaart R, Drossaert C, Taal E.** Experiences and preferences of patients with rheumatic diseases regarding an interactive health communication application. In: IEEE Computer Society. 2010 Presented at: eHealth, Telemedicine, and Social Medicine, International Conference on (2010); Feb. 10, 2010 to Feb. 16, 2010; St. Maarten, Netherlands Antilles.

34. **Plaete J, Crombez G, DeSmet A, Deveugele M, Verloigne M, De BI.** What do general practitioners think about an online self-regulation programme for health promotion? Focus group interviews. BMC Fam Pract 2015;16(1):3. (doi: 10.1186/s12875-014-0214-5)

35. **van Gemert-Pijnen Julia E W C, Nijland N, van LM, Ossebaard HC, Kelders SM, Eysenbach G, et al.** A holistic framework to improve the uptake and impact of eHealth technologies. J Med Internet Res 2011;13(4):e111. (doi: 10.2196/jmir.1672)

36. **Nijland N, van Gemert-Pijnen Julia E W C, Kelders SM, Brandenburg BJ, Seydel ER.** Factors influencing the use of a Web-based application for supporting the self-care of patients with type 2 diabetes: a longitudinal study. J Med Internet Res 2011;13(3):e71. (doi: 10.2196/jmir.1603)

37. **Yonker LM, Zan S, Scirica CV, Jethwani K, Kinane TB.** "Friending" teens: systematic review of social media in adolescent and young adult health care. J Med Internet Res 2015;17(1):e4. (doi: 10.2196/jmir.3692)

38. **Flicker S, Goldberg E, Read S, Veinot T, McClelland A, Saulnier P, et al.** HIV-positive youth's perspectives on the Internet and e-health. J Med Internet Res 2004 Sep 29;6(3):e32. (doi: 10.2196/jmir.6.3.e32)

39. **Betz CL, Smith KA, Macias K, Bui K.** Exploration of internet use: profile and preferences of transition-aged adolescents with spina bifida. Pediatr Nurs 2014 Aug;40(4):173-178.

40. **van der Velden Maja, El EK.** "Not all my friends need to know": a qualitative study of teenage patients, privacy, and social media. J Am Med Inform Assoc 2013 Jan 1;20(1):16-24. (doi: 10.1136/amiajnl-2012-000949)

41. **Van Pelt P, Drossaert C, Kruize A.** Use and perceived relevance of health related internet sites and online contact with peers among young people with Juvenile Idiopathic Arthritis. Rheumatology 2015 (in press) 2015. (doi: 10.1093/rheumatology/kev193)

> *“…searching for balance? The continuous search for balance? It tires me out sometimes. Making choices is what self-management is about”*

- Female, 20 years old, since 16 year Juvenile Idiopathic Arthritis

CHAPTER 4

FEASIBILITY OF AN ONLINE AND A FACE-TO-FACE VERSION OF A SELF-MANAGEMENT PROGRAM FOR YOUNG ADULTS WITH A RHEUMATIC DISEASE: EXPERIENCES OF YOUNG ADULTS AND PEER LEADERS

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ABSTRACT

Background Based on the self-efficacy theory, an online and a face-to-face self-management program 'Challenge your Arthritis' for young adults with a rheumatic disease have recently been developed. These two courses are led by young peer leaders. The objective of this study was to test the feasibility of the online and face-to-face self-management programs.

Methods Feasibility was evaluated on items of perceived usefulness, perceived ease of use, user-acceptance and adherence to both programs in young adults and peer leaders. Additional analyses of interactions on the e-Health applications, discussion board and chat, were conducted.

Results Twenty-two young adults with a diagnosed rheumatic disease participated in the study: twelve followed the online program, ten followed the face-to-face program. Both programs appeared to be feasible, especially in dealing with problems in daily life, and the participants indicated the time investment as 'worthwhile'. In using the online program, no technical problems occurred. Participants found the program easy to use, user friendly and liked the 'look and feel' of the program.

Conclusion Both the online and the face-to-face versions of a self-management program 'Challenge your arthritis' were found to be feasible and well appreciated by young adults with a rheumatic disease. Because these programs are likely to be a practical aid to health practices, a randomized controlled study to investigate the effects on patient outcomes is planned.

BACKGROUND

Numerous rheumatic diseases such as arthritis, fibromyalgia and systemic lupus erythematosus affect children (1). These diseases and their treatments put extensive demands on children, young adults and their parents, by precisely scheduling daily medication, regular physical exercises, blood tests and regular visits to a primary care provider or a rheumatologist (2, 3). Most of the young adults with a rheumatic disease experience problems into their adult years (2), as they often have significant disabilities and may receive on-going medical treatment (4). Yet, as all young adults, they have to develop their own identity and independence. As they become more independent, they will gradually have to take over the roles of their parents and become more responsible for their own illness and treatment: to become a self-manager. In the same period of this change, they also make the transition from child care to adult care systems. Often, they are quite unprepared for and inexperienced in assuming all these adult roles especially since they may not had anything like a normal childhood and adolescence (5-7).

To educate and counsel young adults, we first developed an outpatient transition clinic, a digital portal and an information website (8). Users of these e-Health applications and young adults from the outpatient transition clinic expressed their needs for a training program in which they could practice self-management skills. To meet these needs, we developed two versions of a self-management program 'Challenge your Arthritis', based on the Arthritis Self-management Program of the Stanford University (9). The aim of this study was to examine the feasibility of the online and the face-to-face version for young adults and peer leaders with respect to perceived usefulness, perceived ease of use and user acceptance in order to improve and adjust the program is necessary and to implement both versions of the program into daily practice.

METHODS

Study design

Ethical approval was obtained by the ethics committee of University Medical Center Utrecht. The registration number is 10-325.
We developed an online version, using a safe website and a face-to-face version so young patients could choose to participate in the way that best suited their current lifestyle. All these initiatives were taken in close cooperation with the Dutch Rheumatism Patient League (DRPL) and young adults from the outpatient transition clinic of University Medical Center. Both groups participated actively in the development of the program. For the development, the transition outpatient clinic received a grant from the Dutch Arthritis Foundation.

The 'Challenge your Arthritis' program is a multi-component, interactive program of education, self-management strategies and social support. The aim of 'Challenge your Arthritis' is to enhance patients' self-management in coping with their chronic disease. The program is given by peer leaders. To our knowledge, this is the first self-management program for this specific group given by peer leaders in the Netherlands.

We conducted this quantitative feasibility study to evaluate perceived usefulness, perceived ease of use and user acceptance of the online or face-to-face version of 'Challenge your Arthritis' in participating young adults and peer leaders. Additional qualitative analyses of the digital communication within the online version on chat and discussion board were carried out.

Participants

Young adults aged 16-25 years, who had registered for the self-management program 'Challenge your arthritis' on the website www.reumauitgedaagd.nl (in Dutch) in January 2011, were asked to participate. Information about the program was spread by placing leaflets in the waiting room of University Medical Centers in the Netherlands and on the website of the Dutch Rheumatism Patient League. Young adults chose voluntarily either for the online or the face-to-face program. Those, who did not want to participate in the study, were given the opportunity to join the program later on. Inclusion criteria were: 1) each participant had to have a rheumatic diagnosis, made by paediatrician or rheumatologist; 2) access to a home-based computer with Internet; 3) each had to be able to read and write in Dutch; and 4) no previously participation in a self-management.
Informed consent was obtained from the participant and if young adults were younger then 18 years old, also from their parents. The peer leaders, who participated in this study, were young adults in the age of 20-30 years, who had a rheumatic disease themselves.

The self-management program

The online and face-to-face version 'Challenge your arthritis' were based on the self-efficacy theory (10). Self-management may be enhanced by increasing self-efficacy through practicing, observing others (modelling), meeting beliefs of others and by interpretation of physiological and emotional status. All these elements were integrated in 'Challenge your Arthritis'. From the start of the development, young adults played a central role. In interactive workshops, organized by the multidisciplinary team of the transition outpatient clinic, the young adults decided, together with the professionals, the content, style and format of the online and face-to-face training programs.
Next to the program the participants of the program were allowed to use information presented on the website www.jong-en-reuma.nl (in Dutch). This website contains information about medical issues and themes like dealing with the consequences of having a rheumatic disease, feeling blue, exercises, work, study, relationship and intimacy.

The online program consists of password-protected, interactive web-based self-management instruction and three e-Health applications including a chat section, home exercises and a discussion board. Once a week, the group (six participants, two trainers) had a planned group chat with a maximum of 90 minutes. Within the chat, the weekly theme was clarified, goals were set and the participants were allowed to practice, ask questions, play a game or watch a real live story video, based on the weekly theme. The home exercises were discussed and evaluated. After the chat, participants were allowed to work through the program at any time at home and doing the exercises (one hour a week). In addition, a discussion board was used by trainers and participants to offer encouragement and share tips. For six weeks, participants worked chronological through the program using six themes (table 1). On average, the total time investment for the participants was 12 hours in a 6-week-period.

The face-to-face version of the program was organized during a weekend in a holiday resort. In three days, 12 hours in total, participants (8 – 12 young adults) and two peer leaders worked through the same program and themes (table 1) as the online edition. Here also personal goals were set and objectives were clarified.

The 'Challenge your Arthritis' program was administrated by peer leaders, young adults in the age of 20-30 years old, who also had an arthritis disease. The peer leaders were recruited through websites of the DRPL, the arthritis youth network (Youth-R-well.com) and the website of the Dutch Arthritis Foundation. Peer leaders were selected through assessments and interviews conducted by the DRPL. The selection process utilized questions on age, disease duration, motivation, perceptions on self- management and strategies. Finally, the peer leader was trained through a train-the-trainer educational program by the UMC Utrecht and a professional coaching organization (Work21.nl). This program consisted of following the training as participant, knowledge of the different themes and teaching training skills. The peer leaders had a volunteer contract and received a stipend from the Dutch Arthritis Foundation for each program worked.

Two moderators assisted the peer leaders during the online and face-to-face versions of the program, but were not 'visible' to the participants. These moderations were a professional coach of the train-the-trainer program, transition coordinator, or a communication advisor of UMC Utrecht.

Table 1 Themes of the 'Challenge your Arthritis' self-management program

THEME	EXAMPLES OF SUB THEMES
1. Are you a self-manager?	▪ introduce yourself, get in touch with the program and the group ▪ goal setting , action planning
2. Communication	▪ communication strategy ▪ giving and receiving feedback
3. Feeling blue	▪ pain, fatigue, feeling blue ▪ asking and giving help
4. Sport and exercise	▪ being active ▪ maintain your plans ▪ healthy nutrition
5. Relations and intimacy	▪ having a relationship ▪ having sex
6. Having control over your live and arthritis	▪ evaluation of the personal goals ▪ being responsible and make choices

Variables

Demographic variables and internet-related skills

We collected demographic variables including age, sex, diagnosis, disease duration, current treatment and educational level at baseline by means of an online questionnaires as well as data about the use of Internet and self-reported Internet skills by participants of the online program.

Primary outcome measure: Feasibility of the program

Participants

Three months after starting the online version and two weeks after completing the face-to-face version. Feasibility was evaluated using the Technology Acceptance Model (TAM) (11, 12). TAM is a prediction and explanation model of the end-users reaction to Health applications. The model contains three concepts: 1) the perceived ease of use; 2) perceived usefulness; and 3) the determination of the intention (intention to use), which influences the actual use (11). Perceived ease of use means "the degree of ease, associated with the use of the applications". Perceived usefulness can be defined as "the degree in which an individual believes that using the applications will help him to gain or to increase personal performance".

Although the TAM is mostly used to evaluate information technology, we chose the TAM to examine the face-to-face version as well, in order to compare both versions of the program.
The concepts were measured by using online questionnaires for each program (TAM online and TAM live). Both questionnaires contained seven items concerning usefulness and user acceptance. Due to the different form (face-to-face and online), the concept perceived ease of use differs in this questionnaire.

All questions were answered on a categorical scale with room for additional comments. In addition, there was one question on how much each subject appreciated the program on a scale of ‘0’ to ‘10’ (the higher the score, the more appreciated). Concepts and questions of the TAM are described in table 2.

Table 2 Concepts and questions of the Technology Acceptance Model (TAM)

PERCEIVED USEFULNESS

- Carry out treatment
- Dealing with the disease in daily life
- Additional to health care
- Time investment (worth it)

PERCEIVED USER ACCEPTANCE

- Recommend it to others
- Participate again (knowing content and form)

PERCEIVED EASE OF USE

- Experienced problems
- Easy to use
- Easy to understand
- Look and feel of the program

Peer leaders

The peer leaders were asked to fill in an online TAM-trainer questionnaire to give their opinion on perceived usefulness, perceived ease of use and user acceptance of the program in which they participated. In addition, three questions were asked on fulfilling the role of peer leader (preparation and execution) as well as willingness to lead the training again. The peer leaders were also asked to rate their opinion of the program.

Adherence and interaction

Adherence to the program was measured after completing the programs by describing how many people had completed the whole course. Also each participant’s presence during the chats, posting on the discussion board and finishing the exercises of the online program was measured. Content of the interaction on discussion board and chat were available in text and were studied after finishing the online program.

Evaluation of the goals

Participants of both programs were also asked to describe and to evaluate the goals they wanted to achieve within the program. The fulfilling of the goal was evaluated by the participants themselves and written down by the peer leaders. In the face-to-face version of the program, the goals were evaluated verbally during the last session. In the online training goals were evaluated in the last chat session. The young adults were asked to score their satisfaction on achieving their goals on a scale of ‘0’ to ‘10’ (the higher the score, the more satisfied).

Data analyses

All quantitative data were entered into an SPSS 15.0 database and processed using descriptive statistics. The five possible answers on the concepts 'perceived usefulness' were combined in three answers: 'agree', 'agree/disagree' and 'disagree' and analyzed, using frequency scores. An item was considered as sufficient if 2/3 of the participants agreed with the item.

Participation in the e-Health application, chat, discussion board, home exercises, was counted throughout the content management system of the website. Additional comments of the participants were collected, afterwards by the researcher (JA) and were thematically organized and reported. Explorative qualitative analyses of the interaction on the e-Health applications were performed by three researchers (JWA, LS and HVO). They coded all transcripts independently. Relevant fragments were first categorized into concepts of "topics of discussion", "type of questions" and content and subsequently categorized into subthemes, using inductive analysis until consensus was reached. For evaluation of the goals and appreciation of the program, mean, sd and range were computed.

RESULTS

Characteristics of the study population

The online self-management program began in January 2011 with two groups of six participants. Eleven of the participants followed the whole program and participated in the study. One dropped out, due to exacerbation of the illness. One participant was lost to follow up. The face-to-face version started in March 2011. There were no dropouts during the weekend, one participant was lost to follow up. Sixteen participants were female; only 3 males participated in this study. The mean age was 21 years in the face-to-face group and 22 years in the online group. The demographics are described in table 3. All participants of the online program used the Internet on a daily basis. Email, chat and visiting social media were used most often. They all regularly searched for health information on different websites.

Perceived usefulness and user acceptance of online and face-to-face program

Online program

Seven out of ten participants rated the program as useful considering 'dealing with problems in daily life'. Now that they know the program and form, they all would recommend the program to others and participate again. Nine out of ten participants indicated the time investment as 'worthwhile' (table 4).

Face-to-face program

The participants of the face-to-face program valued the program as equally useful when considering 'carry out treatment, daily live and addition to healthcare'. All participants indicated the time investment as 'worthwhile'. Six out of ten would participate again (table 4).

Perceived ease of use

Online program

All participants found the online program 'easy to use' and appreciated the 'look and feel' of the website. There were no great difficulties with logging in or technical bugs and they could easily find what they were looking for.

Table 3 Characteristics of the study population

	FACE TO FACE GROUP (N=9)	ONLINE GROUP (N=10)
Age, mean (range)	20,7 (17-25)	22,3 (17-25)
Female sex (n)	7	9
Education (n)		
Lower[1]	-	1
Middle[2]	6	7
High[3]	3	2
Diseases		
JIA[4]	3	5
FM[5]	2	2
RA[6]	2	2
SpA[7]	1	1
Immune disease (e.g. MCTD[8]**, SLE**[9]**)**	1	-
Duration of the disease, years (SD)	6,4 ± 6,2	5 ± 3,7
Treated by pediatrician or rheumatologist (n)	9	10

[1] Vocational training, [2] Advanced vocational training, [3] College/University training, [4] Juvenile Idiopathic Arthritis, [5] Fibromyalgia, [6] Rheumatoid Arthritis, [7] Spondyloathropathy, [8] Mix Connected Tissue Disease, [9] Systemic Lupus Erythematosus

Table 4 Feasibility results: perceived usefulness online en face-to-face program (participants)

	ONLINE PROGRAM (N=10)			FACE-TO-FACE PROGRAM (N=9)		
Perceived usefulness	agree	neutral	dis-agree	agree	neutral	dis-agree
Carry out treatment, n	1	5	4	7	2	-
Daily live, n	7	3	-	7	2	-
Additional healthcare, n	6	4	-	7	2	-
Time investment, n	9	1	-	9	-	-
Perceived user acceptance						
Recommend the program to others	10			9		
Would participate again	10			6		
Overall score (mean, range)	7.3 (5-10)			7.7 (6-10)		

Face-to-face program

The perceived ease of use of the content of the face-to-face program was judged positively among the participants. Relations & intimacy and 'having control over your life and arthritis' received the highest scores (table 5). Positive statements were shared such as 'very helpful tips and tricks', 'love the mood boards', 'nice video presentations' and 'such a difficult subject like sexuality was also discussed'. Still, there was also some criticism : 'the lack of outdoor activities during the weekend', 'healthy nutrition – as sub theme of sports and exercise – was only generally discussed, 'absence of evaluating the goals' and 'the need for more theoretical interpretation by the peer leaders' were noted.

Table 5 Feasibility results: perceived ease of use of the face-to-face program

PERCEIVED EASE OF USE (N=9)	GOOD	INSUFFICIENT
1. Are you a self-manager?	8	1
2. Communication	8	1
3. Feeling blue	8	1
4. Sport and exercise	8	1
5. Relations and intimacy	9	-
6. Having control over your live and arthritis	9	-

Adherence with the face-to-face and online program

There were no dropouts in the face-to-face weekend program. Twelve participants started the online program; eleven finished it after six weeks. One participant dropped out after three weeks, due to an increase of disease activity. Nearly all online participants logged in, twice a week on the website and participated in the 90 minutes weekly chat. Two participants missed one chat, due to hospitalization and school activities. Ninety-five percent of the home exercises were finished. In total, 191 messages were posted on the discussion board; 17 messages were sent by the trainer, 174 messages were sent by the participants with a mean number of 21 messages per participant.

Interaction on the discussion board and chat in the online program

On the discussion board, the main topic of the posted messages was sharing experiences on the themes of the program (for example: what does arthritis mean to you or how do you handle problems) and discussion of questions about the home exercises. Real life examples were given to clarify their experiences. Tips and tricks about dealing with fatigue and dealing with friends were shared. Support was given in case of concerns about their future or communication problems with the health care provider or the mentor at school or work. Furthermore, individual goals to achieve in the program and future goals were discussed. Feedback and support was given if they had undertaken a step to achieve their goal (for example: talk to the mentor at school).

In the group chat, the homework was discussed and the new theme was explained. Despite the fact that they couldn’t see each other – no webcams were used - the young adults shared a lot of personal experiences (for example about being angry and upset and about intimacy and sexuality). Encouragement, tips, tricks and support were also given in this case.

Goals and achieving goals

The 10 participants of the online training formulated a total of fourteen personal goals; the 9 face-to-face participants formulated a total of fifteen goals. For a summary, see table 6. After six weeks, for the online training, the young adults were asked to evaluate the achievement of their goals with a number between ´0´ and´10´ (the higher the number , the more satisfied). The mean score was 8.4 (range 6 -10). In the face-to-face training, the personal goals were verbally evaluated during the last theme of the weekend. Although goals were achieved, the general comment was that this evaluation deserved more attention and time during the last part of this weekend.

Feasibility as judged by peer leaders

Online program

Concerning the perceived usefulness, three of the four peer leaders of the online training rated the domain ‘dealing with problems in daily live’ as the most useful. All four peer leaders found the program and the online applications ‘easy to use’ and ‘clear in their purpose’. There were no big technical problems or bugs. All peer leaders were willing to give the program again. Two peer leaders were surprised by the level of intimacy and frankness the group reached despite the fact that they could not 'actually see each other live'. Additional comments were made about the preparation ('I didn't plan enough time to prepare myself') and the execution of the program ('is there an online tool available for keeping track of the exercises of the participants?'). The collaboration between the peer leaders was highly appreciated. The program in general received a mean overall score of 7.5 (range 7-8).

Face-to-face program

All peer leaders rated that the face-to-face program 'as useful' on all aspects. Overall scores on content and form for the face-to-face self-management program were good, except for 'healthy nutrition' (a topic of the Sport and Exercise theme). They would recommend the program to others and felt confident 'to fulfil their role as peer leader'. They were also willing to give the training again. Three out of four peer leaders argued that 'there should be more physical activities at the start of the program'. Peer leaders also noticed that more attention should be paid on the theme of feeling blue or depression. The face-to- face program received a mean score of 7.7 (range 6-10).

Table 6 Number and summary personal goals, formulated at the start of the online and face-to-face program

PERSONAL GOALS*	ONLINE	FACE-TO-FACE
Communication about the personal condition with family, friends, school, work	3	2
Handling tiredness, pain and feeling blue	2	3
Carrying responsibility in the personal treatment	1	-
Learn from and modeling with others who have the same problems	1	-
(Maintain) Sports and exercises	2	1
Listing to the signals of the body	1	1
Getting support of others	2	1
Setting boundaries (regarding friends, activities)	1	2
Coping with the frustrations of having a chronic diseases	1	2
Getting to know other youngsters, share experiences and problems	-	1
Anger management	-	1
Feeling an outcast; handling and sharing tips and tricks	-	1

* Participants could formulate > one goal

DISCUSSION

This study assessed the feasibility of an online and a face-to-face version of a recently developed self-management program ‘Challenge your arthritis’. This program was designed to help young adults with rheumatic diseases managing their chronic health condition. Considering the three domains of feasibility, the young adults and peer leaders appreciated the online and face-to-face program both as useful and found the time investment ‘worthwhile’. They all would recommend the program in which they participated to other young adults with a rheumatic disease and all stated that they were likely to participate again, now they know the program’s content and form.

To predict the acceptance of a (e-Health) program, we used the TAM model (12) in order to test not only the technique but also the adherence to and acceptance of the program. Although many Health prediction models on the actual use have been developed over the last years, the TAM is one of the most utilized models to underline the positive relationship between the ease of use, perceived usefulness and the actual use of an intervention. From the start of the process of development, we chose to have an active involvement with the young adults themselves which could have attributed to the positive outcomes of feasibility.

Active involvement of end users is recognized as a critical factor for the actual use of the intervention, by Van Gemert-Pijnen et al (13). They strongly emphasize the importance of a holistic framework for the development of an e-Health application in which a co-creation of technology, patients, caregivers but also an evaluation is included (13, 14). The positive outcomes of the patients and peer leaders on the feasibility of the program may have been influenced by the active involvement of the young adults themselves.

Adherence to both versions of the program was high, the dropout rate was low. This is in contrast with the results of the ‘Eysenbach’ study (15) in which was described that particular self-help applications are known for their high number of dropouts (as being a natural and typical feature). The positive outcomes of this study may be declared by the length of the online program (limited) and ‘the free choice of time to follow the program’ (in their own time, a weekly chat session in the evening) which are known factors to enhance the adherence to an e-Health intervention (16, 17).

Stinson et al. (23) explored that young adults have a strong believe that web based programs are a promising avenue for the accessibility and availability for improving self-management behaviour. We do not know which factors influenced the individual choice for the face-to-face or the online program in our study. Also, we did not examine whether disease duration, or nature (subtype) of the rheumatic disease influenced the decision. Future research is needed to explore these aspects.

Based on our own practice with young adults and the theory of Bandura (10), we know young adults ‘learn the most’ of their peers and modelling is an important factor in stimulating positive self-management. Therefore we educated young adults with rheumatism to give the training. Although there are other self-management programs for adults with a chronic disease in which peer leaders have a leading role (18-20), little is known about the effects of this concept for young adults with a rheumatic disease. In Stinson’s (21) study on the feasibility of an online self-management training for young adults with a rheumatic disease, a health care professional conducted this specific role, but experiences were not described.

Our study showed that young adults were positive about the content and felt comfortable to talk about personal information in the online program. These findings were also recognized in the feasibility study of White (22) among a comparable group of young adults. The anonymity could be an influencing, positive factor of the online program.

CONCLUSIONS

We have shown that both versions of the self-management program ‘Challenge your arthritis’ are feasible and are appreciated by young adults with a rheumatic disease. The involvement of the end user in the development of the program has contributed to the usefulness, ease of use and user acceptance. The support of self-management of chronically ill young patients is an important task of all health care professionals (2, 4, 5, 23). The face-to-face and online program might prove to be a practical aid to adolescent health practice. We believe a randomized controlled study is needed to investigate these effects.

REFERENCES

1. **Prakken B, Albani S, Martini A.** Juvenile idiopathic arthritis. Lancet 2011;377:2138-2149.

2. **Tucker LB, Cabral DA.** Transition of the adolescent patient with rheumatic disease: issues to consider. Rheum Dis Clin North Am 2007;33:661-672.

3. **Adam V, St-Pierre Y, Fautrel B, et al.** What is the impact of adolescent arthritis and rheumatism? Evidence from a national sample of Canadians. J Rheumatol 2005;32:354-361.

4. **Duffy CM.** Health outcomes in pediatric rheumatic diseases. Curr Opin Rheumatol 2004;16:102-108.

5. **Hazel E, Zhang X, Duffy CM, et al.** High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. Pediatr Rheumatol Online J 2010;8:2.

6. **Shaw KL, Southwood TR, McDonagh JE, et al.** Developing a programme of transitional care for adolescents with juvenile idiopathic arthritis: results of a postal survey. Rheumatology (Oxford) 2004;43:211-219.

7. **Lawson EF, Hersh AO, Applebaum MA, et al.** Self-management skills in adolescents with chronic rheumatic disease: A cross-sectional survey. Pediatr Rheumatol Online J 2011;9:35.

8. **Ammerlaan JW, Scholtus LW, Bijlsma HJ, et al.** An urge for change: Transitional care for young adults with juvenile idiopathic arthritis. Patient Educ Couns 2013;92:127-129.

9. **Lorig KR, Sobel DS, Stewart AL, et al.** Evidence suggesting that a chronic disease self-management program can improve health status while reducing hospitalization: a randomized trial. Med Care 1999;37:5-14.

10. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev 1977;84:191-215.

11. **Davis FD.** Perceived usefulness, perceived ease of use and useracceptance of information technology 1989;13:319-340.

12. **Holden RJ, Karsh BT.** The technology acceptance model: its past and its future in health care. J Biomed Inform 2010;43:159-172.

13. **van Gemert-Pijnen JE, Nijland N, van Limburg M, et al.** A holistic framework to improve the uptake and impact of eHealth technologies. J Med Internet Res 2011;13:e111.

14. **Nijland N.** Grounding eHealth. Towards a holistic framework for sustainable eHealth technologies 2011.

15. **Eysenbach G.** The law of attrition. J Med Internet Res 2005;7:e11.

16. **McLean S, Protti D, Sheikh A.** Telehealthcare for long term conditions. BMJ 2011;342:d120.

17. **Shigaki CL, Smarr KL, Yang G, et al.** Social interactions in an online self-management program for rheumatoid arthritis. Chronic Illn 2008;4:239-246.

18. **Tang TS, Funnell MM, Gillard M, et al.** Training peers to provide ongoing diabetes self-management support (DSMS): results from a pilot study. Patient Educ Couns 2011;85:160-168.

19. **Tang TS, Funnell MM, Gillard M, et al.** The development of a pilot training program for peer leaders in diabetes: process and content. Diabetes Educ 2011;37:67-77.

20. **Catalano T, Kendall E, Vandenberg A, et al.** The experiences of leaders of self-management courses in Queensland: exploring Health Professional and Peer Leaders' perceptions of working together. Health Soc Care Community 2009;17:105-115.

21. **Stinson J, McGrath P, Hodnett E, et al.** Usability testing of an online self-management program for adolescents with juvenile idiopathic arthritis. J Med Internet Res 2010;12:e30.

22. **White M, Stinson JN, Lingley-Pottie P, et al.** Exploring therapeutic alliance with an internet-based self-management program with brief telephone support for youth with arthritis: a pilot study. Telemed J E Health 2012;18:271-276.

23. **Stinson J, Wilson R, Gill N, et al.** A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol 2009;34:495-510.

“365 days a year”

- Male, 23 years old, since 8 years Juvenile Idiopathic Arthritis

CHAPTER 5

SHORT TERM EFFECTIVENESS AND EXPERIENCES OF A PEER GUIDED WEB-BASED SELF-MANAGEMENT INTERVENTION FOR YOUNG ADULTS WITH JUVENILE IDIOPATHIC ARTHRITIS

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ABSTRACT

Objective A web-based self-management intervention guided by peer-trainers was developed, aimed to support young adult's self-management in coping with Juvenile Idiopathic Arthritis (JIA). To investigate its effectiveness, a randomized controlled trail (RCT) was conducted. In addition, content of Chat and goals were studied to identify underlying processes.

Methods A RCT with a six months follow up period was conducted among 72 young adults with JIA, randomly assigned to the intervention or to the usual care control group. Analyses according to intention to treat analysis by means of linear mixed models for longitudinal measurements were carried out. Self-efficacy as primary outcome, self-management, disease activity, quality of life, absenteeism of school/work, health care medication use and adherence to the intervention were studied. Interaction on the Chat, goals, personal achievements and the appreciation of the intervention were analyzed using thematic analyses.

Results No significant differences were found on self-efficacy, quality of life and self-management between the participants of the control and the intervention groups. In the intervention group, modelling and sharing experiences were the most recognized. 55 goals were formulated, related to improvement and maintaining balance, setting and recognizing boundaries, communicating and coping with incomprehension. Adherence, appreciation of the own learning experience and personal achievements was rated positively.

Conclusion The web-based intervention did not lead to an improvement of self-efficacy. Additional qualitative analyses showed that the intervention was appreciated and valuable for the participants. More research is needed on how to measure the added value of this intervention compared to the usual care.

SIGNIFICANCE AND INNOVATIONS

- Enhanced self-management of young adults with Juvenile Idiopathic Arthritis might prevent disease exacerbation and facilitate a successful transition towards adult care.

- To increase self-management, a multi-component, interactive web-based peer guided intervention of education, self-management strategies and social support was developed.

- Quantitative results revealed no significant differences between the intervention and control groups in terms of self-efficacy, quality of life and self-management, while additional qualitative analyses of the interaction in the intervention group revealed positive results on adherence and learning experiences.

- Web-based interventions might be a valuable aid to support self-management. More research on the meaning young adults themselves give to the concept of self-management is needed.

INTRODUCTION

eHealth interventions are developed and offered more and more to patients with chronic diseases to improve their self-management (1-3). Since young adults are one of the most active groups of internet users, eHealth interventions like portals and self-management support programs might be promising options also for patients with Juvenile Idiopathic Arthritis (JIA) (1,2,4-12). Next to that, traditional health care services do not always fit with the needs and problems of young adults between 16-25 years with JIA (8-13). JIA is characterized by arthritis of one of more joints, starting before the age of 16 years. In the western world, the reported incidence of JIA varies of 1 to 22 cases per 100.000 children, with a prevalence of 8 to 150 cases per 100.000 (13). The disease and its treatment put extensive demands on children and young adults as well as on their parents, due to precisely scheduled daily medication requirement, regular physical exercise regime and regular visits to the pediatrician or rheumatologist (14,15). Most of these young adults still experience problems into their adult years with on-going medical treatment and significant disability (12-14). Like other young adults, young adults with JIA have to develop their own identity and independence but for them, the path towards adulthood is a lot bumpier (11,15,16). As they become more independent, they will gradually become more responsible for their own illness and the treatment: to be a self-manager. Enhanced self-management may prevent disease exacerbation and facilitate a successful transition to adult care (2,17). Focus group interviews on views and preferences on transitional care among young adults with JIA revealed they would like to be guided online to become a self-manager (9). Self-management can best be described as ‘the individual’s ability to manage the symptoms, treatment, physical and psychological consequences and life style changes, inherent to living with a chronic illness as JIA’ (18). Based on the mentioned interviews, a web-based self-management intervention was developed, in close cooperation with the young adults, the Dutch Youth Network for young people with arthritis (YouthRwell.com) and both multidisciplinary teams of the child and adult rheumatology department (19).

The web-based intervention (which is called “ReumaUitgedaagd!” in Dutch), based on the self-efficacy theory of Bandura (20), is a multi-component, interactive web-based intervention of education, self-management strategies and social support, aimed to enhance the young adult's self-management in coping with JIA. Self-management may be enhanced by increasing self-efficacy (18) and to achieve this aim, the elements of skills mastery, modeling, reinterpreting symptoms and persuasion are embedded in the intervention and discussed during a weekly Chat. In order to display pro-active behaviour 'one needs goals' (21). Therefore goal-setting is a crucial element of the intervention. ReumaUitgedaagd! is led by young peer trainers in the age range of 20-30 years, suffering from arthritis themselves. The first draft of the intervention was evaluated on items of perceived usefulness, perceived ease of use, user acceptance and adherence in 12 young adults and 4 peer trainers and appeared to be feasible, especially in dealing with problems in daily life (19). Although the intervention is thought to be a practical aid in health practices, a randomized controlled trial (RCT) was conducted to investigate its effectiveness on self-efficacy, self-management and quality of life. In addition, thematic analysis (22) within the intervention group was carried out to explore the interaction in the Chat, the goals the participants set and the appreciation of the intervention itself.

PATIENTS AND METHODS

Design

A RCT, registered at the Dutch trial register (www.trialregister.nl, number NTR4679), with a six month follow up period was conducted among young adults with JIA, and treated at the transition outpatient clinics of the University Medical Center Utrecht and the Erasmus MC Rotterdam in the Netherlands. Both university hospitals are seen as the largest national centers for science and care of children with chronic inflammatory diseases in the Netherlands. Participants of the intervention group were given access to the web-based self-management intervention. Among them, qualitative thematic content analyses (22) were used to explore the interaction in the Chat, the content of the goals, personal achievement with regard to their goals and the appreciation of the intervention itself. The control group was a waiting list group which was enabled to follow the web-based intervention after six months.

Patient selection

Young patients, aged between 16 and 25 years were eligible to participate if they were a) diagnosed with JIA b) abled to speak and read Dutch, c) had access to the internet and a mobile phone and d) hadn't participated in a self-management intervention before. Participants were recruited by their pediatrician/rheumatologist at the transition outpatient clinic. Interested participants received information about the study and a telephone contact was set up between the researcher and the participants to check the inclusion criteria and to answer additional questions. Once informed consent had been obtained – for the participants < 18 years, also by their parents - the participants were asked to fill in online baseline measurements, prior to randomization. The medical-ethical review board of the University Medical Center Utrecht in the Netherlands approved the design and the procedures of this study (number: 11-363/K).

Randomization

Because the intervention was group-based with six participants, randomization was carried out each time twelve participants were included. We used a stratified block, randomization for the factor 'gender' to equally divide 'men' and 'women' among both groups, using a computerized intervention with an automated process and no interference from the investigator. After randomization, the participants (and the parents for participants <18 years) received the allocated condition by email and post.

Treatment conditions

Both the control and the intervention groups received usual care, based on medical guidelines (13), consisting of a 3 monthly visit to the transition outpatient clinic where medical problems, questions and treatment plan were discussed and assessment of the disease activity was carried out by the pediatrician/rheumatologist or the transition nurse among the participants of both groups. Also included in usual care, the transition nurse focused on problems and questions in dealing with the consequences of having JIA and coordinated the process of transition. Both control and intervention group were allowed to use information presented on the website jong-en-reuma.nl (in Dutch). This website contains information about medical issues and themes including dealing with the consequences of having a rheumatic disease, feeling depressed, exercises, work and study, relationships and intimacy (6). In addition to usual care, the intervention group started within 1 month, after randomization, with the web-based self-management intervention.

The web-based intervention consists of password-protected, interactive web-based self-management instruction with three components a Chat section, home exercises and a discussion board. Once a week, the group (six participants, two trainers) had a planned group Chat for a maximum of 90 minutes. Within the Chat, the weekly theme was clarified, goals were set and the participants were allowed to practice, ask questions, give and receive feedback, play a game or watch a real live story video-based on the weekly theme. Also, the home exercises were discussed and evaluated. After the Chat, participants were allowed to work through the intervention at any time at home and do the exercises (one hour per week). In addition, a discussion board was used by trainers and participants to offer encouragement and share tips. For six weeks, participants worked chronologically through the intervention using six themes (Table 1). On average, the total time investment for the intervention was 12 hours in a 6-week-period per patient.

Table 1 Themes and content of the web-based self-management intervention ReumaUitgedaagd! (in Dutch)

THEMES	CONTENTS
Are you a self-manager?	▪ introducing yourself, get in touch with the group ▪ what do you (want to) know about your disease? ▪ capacities and talents ▪ goal setting and action planning
Friends, family and communication	▪ communication strategies ▪ communication with school, friends, work, parents, health care providers ▪ giving and receiving feedback ▪ setting boundaries
Feeling blue	▪ receiving therapies (treatment, medication) ▪ pain, fatigue, feeling blue ▪ asking and giving help ▪ relaxation
Sport and exercises	▪ being active ▪ motion and physical activity ▪ maintain your plans
Relations and Intimacy	▪ body images and thoughts ▪ having a relationship ▪ having sex ▪ thinking about kids, pregnancy, heredity
Having control over your life and arthritis	▪ evaluation of your personal goals; how to move on? ▪ being responsible and making choices ▪ celebration and saying goodbye

Outcome measures

In this study quantitative and qualitative outcome measures were collected online with questionnaires, text messages, self-reported by participants. All outcome measures were collected at baseline, 3 and 6 months after randomization. Demographic and disease related variables were collected at baseline, added with data from the medical record. A transcript of the intervention was available for thematic analyses of goals, personal achievements and appreciation of the intervention.

Quantitative outcomes

Primary outcome measurement

Self-efficacy as a determinant of self-management behavior was measured by the online Dutch-Arthritis Self-Efficacy Scale (Dutch-ASES) questionnaire (23). The questionnaire contains eight items. For each item, respondents were asked to indicate on a scale from 1 (very unconfident) to 10 (highly confident) how confident they felt to bring a situation to a good outcome. The mean score of the eight items was calculated where a higher score indicated a higher degree of self-efficacy.

Secondary outcomes measurement

Self-management related outcomes were measured using the Health Education Impact Questionnaire (heiQ) (24) (Dutch translation). This online questionnaire consists of 40 questions, divided into eight independent scales which cover eight self-management domains: 'Positive and active engagement in life', 'Health directed activity', 'Skills and technique acquisition', 'Constructive attitudes and approaches', 'Self-monitoring and insight', 'Health service navigation', 'Social integration and support' and a reversed scale, 'Emotional distress'. Each scale is calculated a mean score (min 1, max 4). A higher mean score indicates a higher degree of self-management in each domain, on each scale.

Quality of life (QoL) was assessed on two dimensions. The first dimension, physical functioning, was measured by the Dutch Consensus Health Assessment Questionnaire Disability Index (HAQ-DI) (25). The second dimension of QoL is a combination of 4 patient reported outcomes: pain, fatigue, general well-being and disease activity. This dimension is assessed with a Numerical Rating Scale (NRS) (26) from '0' to '10' (the higher the score, the more pain, fatigue or disease activity and the worse general well-being). For three days in a row on a fixed time, participants were asked to send their responses via a text message.

Medication use, divided in the categories: Disease Modifying Anti Rheumatic Drug (DMARD), Non Steroid Anti Inflammatory Drug (NSAID), Biologicals and medication reducing pain and health care use (number of consultations to the pediatrician/rheumatologist/transition nurse and day-care center at the hospital), was assessed by analyzing the medical patient record. Absenteeism of school or work was assessed with one question (days of absenteeism of school last month, due to JIA) by a text message on one fixed moment.
At the end of the intervention, participants of the intervention group were asked to indicate with a score from 0 to 10 on a NRS how they rated their own learning experience. Also, in this group, adherence with the web-based intervention was measured by the researcher by counting the amount of total participation in the weekly Chat.

Qualitative outcomes

Interaction in the Chat, goals, personal achievements with regard to their goals and appreciation of the intervention was explored using thematic analyses (22) within the intervention group.

Statistical analysis

Quantitative analyses

The consolidated Standards of Reporting Trails (CONSORT) statement (27) was used to present the results of this study. Quantitative data was entered into a SPSS data base. Based on the theoretical fundament of the intervention, it was hypothesized that participants of the intervention group would have a better result on self-efficacy, compared with the usual care control group participants. Sample size calculation, based on a previous study of Niedermann et al (28), calculated that 72 patients were required to find a difference between both groups of 1.29 (*sd* 1.6) on self-efficacy, with a power of 80% and alpha of .05 and, an estimated loss of 30%.

Demographic variables and absenteeism of school/work, frequency of health use and medication use were presented using descriptive analyses and frequency scores. Adherence of the intervention group in the Chat, and the indication score of their own learning was counted and with a frequency score displayed. Linear mixed models for longitudinal measurements were used to determine effects on self-efficacy, self-management related outcomes and on QoL scores for physical activity, pain, well-being, and fatigue and disease activity. Fixed effects for group, time and group * time were included in the model.

Qualitative analyses

All transcripts of the Chats were inserted into the program Nvivo (QSR International Pty LTD Version 10). To describe the interaction in the Chat, the transcripts were analyzed, using codes like *modelling, mastering, verbal persuasion, goals, sharing experiences, emotional attitude* and *appreciation*, derived from the self-efficacy theory and the elements of the intervention.
Relevant fragments within the transcripts, related to goals, personal achievements and appreciation of the intervention were categorized independently by two members of the research group (JA and NdBN). Results were discussed until consensus was reached.

RESULTS

224 young adults with JIA were found to be eligible to participate in this study and were invited. 152 were excluded where the largest group (55/152) were not able to be reached by phone to give additional information or to check inclusion criteria. 47/152 declined to participate in the study after being informed that the intervention lasted six weeks and participation in all Chat sessions was mandatory. Finally, 72 participants were randomized, equally divided into the intervention and the control group (see Figure 1). 24 participants (67%) of the intervention and 24 (67%) of the control group filled in all questionnaires, after 6 months. There were no significant differences between the intervention and control group on baseline on demographic variables or internet-skills (see table 2). Next to that, there were no differences between the completers and non-completers on demographic and illness-related date on baseline data.

Primary outcome: self-efficacy

No significant differences between the intervention and control group were found on self-efficacy at three and six months (p=0.136) (see table 3 and 4).

Figure 1 Flowchart of the participants

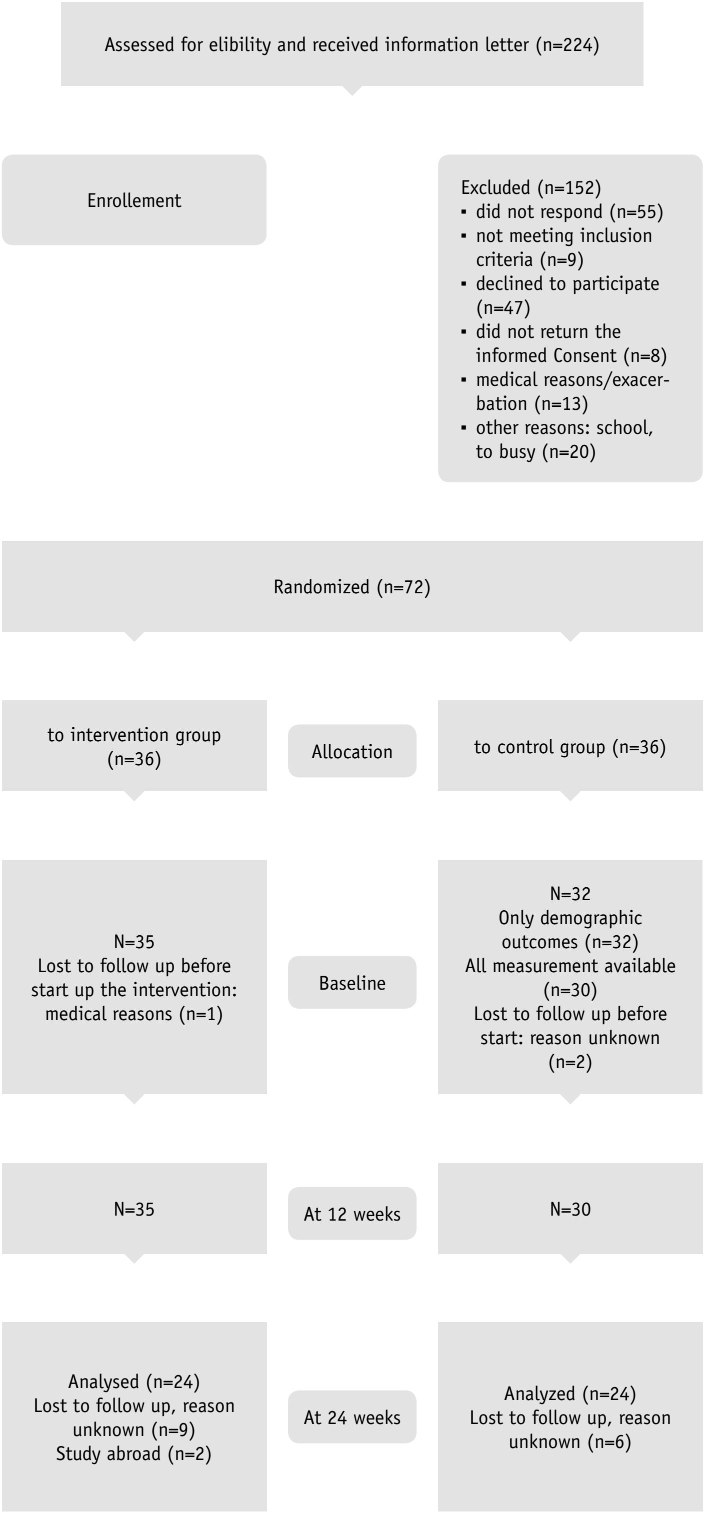

Table 2 Baseline characteristics of the participants

BASELINE CHARACTERISTICS INTERVENTION VERSUS CONTROL

Characteristic	**Total group** (n = 67)	**Intervention** group (n = 35)	**Control** group (n = 32)
Gender – female n(%)	59 (88%)	29 (83%)	30 (94%)
Age, mean year (SD)	19.1 (2.7)	19.2 (2.7)	19.1 (2.9)
Education level completed n(%)			
Primary education[1]	15 (22%)	10 (29%)	5 (16%)
Intermediate vocational education[2]	35 (52%)	17 (49%)	18 (56%)
Bachelor or University	17 (25%)	8 (23%)	9 (28%)
Civil status n(%)			
Living at home with parents	53 (79%)	26 (74%)	27 (84%)
Living independently	14 (21%)	9 (26 %)	5 (16%)
Marital status n(%)			
Single	61 (91%)	32 (91%)	29 (91%)
Living together	3 (5%)	1 (2%)	2 (6%)
Married	3 (5%)	2 (6%)	1 (3%)
Diagnosed disease n(%)			
Oligo-articulair JIA	14 (21%)	8 (23%)	6 (19%)
Poly-articulair JIA	24 (36%)	9 (26%)	15 (47%)
Systemic JIA	8 (12%)	6 (17%)	2 (6%)
Other	21 (31%)	12 (34%)	9 (28%)
Duration of the disease (years, mean, SD)	10.9 (6,4)	10.1 (6,3)	11.8 (6,5)
Frequency of internet use n(%)			
(Almost) daily	62 (93%)	31 (89%)	31 (97%)
Several times a week	3 (5%)	2 (6%)	1 (3%)
Few times a week	1 (2%)	1 (3%)	
Rarely	1 (2%)	1 (3%)	
Purpose use internet ***Search health info n(%)***			
Not	3 (5%)	2 (6%)	1 (3%)
Yes, one single time	30 (45%)	18 (51%)	12 (38%)
Yes, sometimes	28 (28%)	11 (31%)	17 (53 %)
Frequent	6 (9%)	4 (11%)	2 (6%)
Forum visit health/arthritis			
Not	47 (70%)	23 (66%)	24 (75%)
Yes, one single time	12 (18%)	7 (20%)	5 (16%)
Yes, sometimes	6 (9%)	4 (11%)	2 (6%)
Frequent	2 (3%)	1 (3%)	1 (3%)

BASELINE CHARACTERISTICS INTERVENTION VERSUS CONTROL

Characteristic	Total group (n = 67)	Intervention group (n = 35)	Control group (n = 32)
Chat/e-mail with someone with arthritis			
Not	55 (82%)	27 (77%)	28 (88%)
Yes, one single time	5 (7%)	4 (11%)	1 (3%)
Yes, sometimes	2 (3%)	2 (6%)	
Frequent	5 (8%)	2 (6%)	3 (9%)
Visit weblog patient			
Not	55 (82%)	30 (86%)	25 (78%)
Yes, one single time	9 (13%)	2 (6%)	7 (22%)
Yes, sometimes	2 (3%)	2 (6%)	
Frequent	1 (2%)	1 (3%)	
Importance internet in relation			
disease, Scale 1-10 (mean) *(SD)*	3.4 (2.5)	3.1 (2.7)	3.7 (2.1)
peer support, Scale 1-10 (mean) *(SD)*	3.0 (2.5)	3.1 (2.6)	3.0 (2.5)

[1] Lower vocational education, lower general secondary education;

[2] Higher general secondary education, pre-university education; SD: standard deviation

Table 3 Results Type lll Tests of fixed effect – Time * sort group for Dutch-ASES, HAQ-DI, pain, well-being, fatigue, disease-activity and heiQ

	SOURCE	F	SIG.
Dutch Arthritis Self-efficacy Scale	Intercept	1322.47	0
	T	0.51	0.61
	group	0.17	0.68
	T * group	2.07	0.14
Dutch Consensus Health Assessment Questionnaire (HAQ-DI)	Intercept	68.32	0
	T	0.21	0.81
	group	0.01	0.93
	T * group	1.26	0.29
Perceived pain	Intercept	141.02	0
	T	1.34	0.27
	group	0.28	0.60
	T * group	0.25	0.78
Perceived well-being	Intercept	386.33	0
	T	0.88	0.42
	group	0.73	0.40
	T * group	2.50	0.09
Perceived fatigue	Intercept	401.63	0
	T	1.63	0.21
	group	0.43	0.51
	T * group	0.21	0.81
Perceived disease activity	Intercept	146.02	0
	T	1.45	0.24
	group	0.16	0.69
	T * group	2.12	0.13

Table 3 Results Type lll Tests of fixed effect – Time * sort group for Dutch-ASES, HAQ-DI, pain, well-being, fatigue, disease-activity and heiQ

	SOURCE	F	SIG.
Health Education Impact Questionnaire (heiQ)			
Health directed activity	Intercept	2011.89	0
	T	0.33	0.72
	group	1.15	0.29
	T * group	1.66	0.20
Positive and active engagement in life	Intercept	3936.64	0
	T	0.05	0.95
	group	1.61	0.21
	T * group	0.61	0.55
Emotional distress	Intercept	909.73	0
	T	0.21	0.81
	group	0.02	0.90
	T * group	0.43	0.66
Self-monitoring and insight	Intercept	6216.50	0
	T	1.39	0.26
	group	0.83	0.37
	T * group	0.92	0.40
Constructive attitude and approaches	Intercept	3941.58	0
	T	0.17	0.85
	group	0.08	0.78
	T * group	0.91	0.41
Skills and technique acquisition	Intercept	2534.79	0
	T	1.06	0.35
	group	0.70	0.41
	T * group	1.26	0.29
Social integration and support	Intercept	2328.77	0
	T	1.22	0.31
	group	0.50	0.48
	T * group	1.15	0.32
Health service navigation	Intercept	2907.22	0
	T	0.25	0.78
	group	2.29	0.14
	T * group	1.83	0.17

Table 4 Results on Dutch Arthritis Self-efficacy Scale (Dutch-ASES), Pain, Wellbeing, Fatigue, Disease Activity, HAQ-DI, HeiQ, based on linear mixed models

	INTERVENTION GROUP MEAN (95% CI)	CONTROL GROUP MEAN (95% CI)
Dutch-Arthritis Self-efficacy Scale		
T0	6.67 (6.11 - 7.23)	6.99 (6.38 - 7.59)
T1	6.67 (6.41 - 7.54)	6.80 (6.17 - 7.40)
T2	6.51 (5.88 - 7.15)	6.84 (6.19 - 7.50)
Perceived Pain		
T0	3.19 (2.39 - 3.98)	3.06 (2.23 - 3.89)
T1	3.59 (2.69 - 4.50)	3.11 (2.15 - 4.07)
T2	3.65 (2.76 - 4.55)	3.38 (2.44 - 4.32)
Perceived Wellbeing		
T0	4.59 (3.82 - 5.36)	4.24 (3.44 - 5.03)
T1	4.93 (4.25 - 5.62)	4.03 (3.29 - 4.76)
T2	4.12 (3.28 - 4.96)	4.24 (3.36 - 5.12)

	INTERVENTION GROUP MEAN (95% CI)	CONTROL GROUP MEAN (95% CI)
Perceived Fatigue		
T0	5.36 (4.55 - 6.16)	5.05 (4.21 - 5.89)
T1	5.45 (4.66 - 6.23)	4.95 (4.11 - 5.79)
T2	4.94 (4.09 - 5.79)	4.75 (3.86 - 5.64)
Perceived Disease Activity		
T0	3.31 (2.57 - 4.06)	3.01 (2.23 - 3.79)
T1	3.70 (2.81 - 4.58)	3.02 (2.08 - 3.97)
T2	3.42 (2.49 - 4.36)	3.73 (2.75 - 4.71)
Dutch Consensus Health Assessment Questionnaire (HAQ-DI)		
T0	0.66 (0.46 - 0.87)	0.62 (0.40 - 0.85)
T1	0.67 (0.44 - 0.90)	0.59 (0.34 - 0.83)
T2	0.62 (0.37 - 0.87)	0.70 (0.44 - 0.97)
Health Education Impact Questionnaire (heiQ)		
Health directed activity		
T0	2.98 (2.78 - 3.18)	2.94 (2.72 - 3.16)
T1	3.13 (2.92 - 3.33)	2.86 (2.64 - 3.07)
T2	3.08 (2.85 - 3.32)	2.96 (2.72 - 3.21)
Positive and active engagement in life		
T0	3.25 (3.12 - 3.38)	3.18 (3.04 - 3.32)
T1	3.30 (3.13 - 3.47)	3.11 (2.93 - 3.29)
T2	3.28 (3.09 - 3.47)	3.16 (2.96 - 3.36)
Emotional distress		
T0	2.00 (1.84 - 2.16)	1.97 (1.79 - 2.14)
T1	1.93 (1.73 - 2.12)	1.99 (1.78 - 2.20)
T2	1.99 (1.74 - 2.24)	2.01 (1.75 - 2.27)
Selfmonitoring and insight		
T0	2.97 (2.84 - 3.11)	3.01 (2.87 - 3.16)
T1	3.17 (3.01 - 3.32)	3.04 (2.88 - 3.20)
T2	3.11 (2.96 - 3.25)	2.99 (2.84 - 3.13)
Constuctive attitude and approaches		
T0	3.29 (3.14 - 3.43)	3.33 (3.17 - 3.49)
T1	3.34 (3.16 - 3.51)	3.23 (3.05 - 3.41)
T2	3.28 (3.10 - 3.47)	3.26 (3.06 - 3.45)
Skills and technique acquisition		
T0	2.83 (2.65 - 3.00)	2.85 (2.66 - 3.04)
T1	3.00 (2.81 - 3.20)	2.81 (2.61 - 3.01)
T2	3.02 (2.80 - 3.23)	2.90 (2.68 - 3.11)
Social integration and support		
T0	3.10 (2.91 - 3.29)	3.11 (2.90 - 3.31)
T1	3.20 (3.01 - 3.39)	3.10 (2.90 - 3.30)
T2	3.15 (2.93 - 3.37)	2.97 (2.74 - 3.20)
Health service navigation		
T0	3.16 (3.01 - 3.31)	3.12 (2.96 - 3.29)
T1	3.28 (3.09 - 3.46)	3.03 (2.84 - 3.23)
T2	3.31 (3.10 - 3.52)	3.06 (2.84 - 3.28)

T0=baseline, T1=3 months after baseline, T2=6 months after baseline

Secondary outcomes

We found no significant differences between the two groups on the secondary outcomes. Results on health care use showed that participants of both groups have had a consultation with a transition nurse. Considering the consultations with the pediatrician or rheumatologist, the median score of the intervention group was 3 (min 0 – max 28), where the median score of the control group was 3.5 (min 0 – max 28). No differences were found between the control and intervention group on absenteeism of school or work. The use of biologicals was at baseline in the control group higher (31,3%) compared with the intervention group (11,4%) (see table 5).

Table 5 Absenteeism, health care use and medication use

ABSENTEEISM OF SCHOOL/WORK (DAYS, PREVIOUS MONTH)	INTERVENTION GROUP	N	CONTROL GROUP	N
T1: median (min-max)[1]	0.00 (0-10)	30	0.00 (0-10)	27
T2: median (min-max)[2]	0.00 (0-20)	25	0.00 (0-30)	24
Frequency of health use (whole period)				
▪ Consultation Pediatrician/rheumatologist, median (min-max)	3 (0-11)		3.5 (0-28)	
▪ Consultation Transition clinical nurse, median (min-max)	1 (0-7)		0 (0-10)	
▪ Day-care center, median (min-max)	0 (0-11)		0 (0-8)	
Medication use				
Non Steroid Anti-rheumatic Drug (NSAID)				
T0: n (%)	12(34.3%)	35	9(28.1%)	32
T1: n (%)	15(42.9%)	35	10(31.3%)	32
T2: n (%)	12(34.3%)	35	8(25.0%)	32
Disease Modifying Anti Rheumatic Drug (DMARD)				
T0: n (%)	19(54.3%)	35	16(50.0%)	32
T1: n (%)	19(54.3%)	35	15(46.9%)	32
T2: n (%)	16(45.7%)	35	11(34.4%)	32
Biologicals				
T0: n (%)	4(11.4%)	35	10(31.3%)	32
T1: n (%)	7(20%)	35	10(31.3%)	32
T2: n (%)	7(20%)	35	8(25%)	32
Medication to reduce pain				
T0: n (%)	6(17.1%)	35	5(15.6%)	32
T1: n (%)	2(5.7%)	35	4(12.5%)	32
T2: n (%)	2(5.7%)	35	5(15.6%)	32

T0=baseline, T1=3 months after baseline, T2=6 months after baseline

Qualitative outcomes

Within the Chat, modelling and sharing experiences were recognized as the most frequently expressed interaction. In addition, also support and encouragement were given. In total 55 individual goals were formulated by the 32 participants at the start. However, a further 112 personal goals were added during the intervention due to personal reflection or awareness. Most formulated goals were related to 'improve and maintain balance during the day', 'setting and recognizing boundaries', 'improving communication with others' and 'coping with incomprehension'. Next to that, goals on 'increasing knowledge about the disease', 'coping with emotional (fear, uncertainly) and psychical consequences (pain, fatigue)' and 'fitting treatment advices in daily life' were formulated.

A total of 145 personal recognized achievements were categorized into: *'gaining insight and awareness'*, *'making determined informed choices', 'personal influence'* '*understanding'* and *'new goals for the future'.*

The own learning performance was rated with a mean score of 7.1 (min 6, max 8.5; on a scale 0-10). The four participants with a self-appointed grade of 6, did not always relate the learned personal achievement to their originally defined goals.

Some of the participants were surprised by the aim and structure of the intervention, 33 of the 35 participants who started the intervention, stayed adherent and were active during Chat and performing exercises. Two participants stopped due to medical reasons and school activities. All participants appreciated the intervention, especially the input of the trainers. Some remarks were made about the time of the weekly Chats (start time, duration) and the initial pace of the Chat (to slow). Also, not every video led to the outcome that was hoped for.

DISCUSSION

In this RCT on the effectiveness of the web-based self-management intervention ReumaUitgedaagd! among young adults with JIA, no significant differences were found between the intervention group and the control group on self-efficacy, quality of life (QoL) and self-management. On the other hand, participants of the intervention group rated their personal achievements within the intervention positively and adherence and appreciation of the own learning experience was high.

There are some possible explanations for not identifying significant improvements on the patient reported quantitative measurements. Firstly, both control and intervention groups registered relatively high baseline scores on the domains of self-efficacy, self-management and QoL so there was little room for improvement. This could have been influenced by the setting of the hospitals were the participants of this study were recruited. Both university hospitals have a special transition outpatient clinic and a multidisciplinary team and are known as large, tertiary care centers in the Netherlands where already the focus is on guidance towards self-management. Also, both groups were under treatment by a pediatrician, rheumatologist or transition nurse, receiving medical treatment and rated themselves relatively low on disease-activity.

Secondly, although we chose as outcome measure 'self-efficacy', based on the theoretical fundament of the web-based intervention, one might question if this outcome is acceptable to measure the concept of self-management, considering the age and needs of the young adult. To measure self-efficacy, we used the Dutch-ASES, a questionnaire, developed for the adult patient, suffering from a rheumatic disease. As we know from studies on transitional care (9-12) but also from studies on ‘growing up with a chronic disease’ (4,8,11), young adults are experiencing other difficulties in managing arthritis in daily life and have other needs compared with the adult group. So it is possible that not only the concept but also language and aim of the adult self-efficacy scale did not fit with this age group and was therefore not sensitive to change. Clearly, interchangeability of a child and adult questionnaire, measuring the same concept can be problematic. The absence of questionnaires measuring the concept of self-management from the perspective of young adult’s group was also recognized in the study of van Pelt (29). More research is necessary on how to measure the concept of self-management and the meaning of the concept, based on the views of the young adults.

Finally, response shift could have influenced the outcomes of this study. Response shift is defined as ‘a change in the meaning of one’s self evaluation of a target construct as a result of recalibration’ (30) and is recognized as an influencing factor to the outcomes of educational and behavioural interventions. Relatively high scores at baseline could be explained by unawareness of already existing self-management related behaviour, since analyses of Chats revealed gained awareness, increased influence on their own situation and making more informed decisions. It is unknown to what extent the process of awareness had also taken place as a consequence of the usual care both groups received.

The personal goals, set by the participants on the intervention group, reflected the aim of the intervention, and the needs and real-life issues of young adults with JIA which are known from literature on these subjects (4,10,11,16). During the intervention, personal goals were added which can be indicated as a result of awareness of needs. In a study on health care transition in rheumatology (12), awareness is recognized as an important factor towards autonomy. Obtaining and increasing autonomy is, in this study, seen as an important psychosocial developmental task of adolescence. Analyses of the interactions in the Chat revealed that lots of experiences and strategies were shared and support and encouragement was mutually exchanged. Sharing experiences and strategies to solve problems, support and encouragement are recognized as active coping styles and predictors for psychological adjustment in young adults (31). Stimulating the use of these styles may prevent developmental problems in psycho-social functioning.

The aim of the intervention was to enhance the young adult’s self-management in coping with JIA. The qualitative results expose that the intervention ‘delivers what it has to deliver’ and suggest that the intervention fits the needs of this group. These qualitative results and other studies on needs assessments among young adults with JIA (4,8,9) point out the importance of paying attention to the complexity of managing medical needs, together with developmental aspects and the drive towards independence. The web-based intervention is positively appreciated by young adults and can be a valuable aid for both young adults and health care professional to support and improve self-management. But some adjustments have to be made. Future participants should be better informed about the aim and the structure of the intervention. Additionally, support should be provided to identify the personal needs and goals they want to achieve. This can prevent potential dropout and provide an early indication of those patients who will benefit the most. Contact with the peer trainer before administration to the intervention can be helpful in this process and can improve the effectiveness of the intervention.

There are some limitations to our study that should be mentioned. Firstly, we recruited participants from two large tertiary centres in the Netherlands. Some participants expressed that they participated because they had a good relationship with their doctor. This could have affected their results positively and therefore generalizing of the results is limited. Secondly, the qualitative results are only representative of the young adults who participated in the intervention group; we didn't study these outcomes in the control group. And thirdly, although the predetermined calculated number of patients was included, there was some drop-out in the follow up leading to a small sample size at the end of the study. However mixed methods analyses were used in which drop-outs were taken into account (32).

In conclusion, in our study on the effectiveness of the web-based intervention ReumaUitgedaagd! for young adults with JIA, we did not find improvement of self-efficacy, self-management and quality of life. However the intervention was regarded to be a valuable and appreciated aid to influence an active coping style with sharing experiences and enhancing social support, and increasing autonomy and goal-setting behavior. More research is needed on how to measure the added value of this intervention/self-management in this group and what meaning young adults themselves give to the concept of self-management.

REFERENCES

1. **Gray NJ, Klein JD, Noyce PR, Sesselberg TS, Cantrill JA.** Health information-seeking behaviour in adolescence: the place of the internet. Soc Sci Med 2005;60:1467-1478.

2. **Stinson J, Wilson R, Gill N, Yamada J, Holt J.** A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol 2009;34:495-510.

3. **Eland-de Kok P, van Os-Medendorp H, Vergouwe-Meijer A, Bruijnzeel-Koomen C, Ros W.** A systematic review of the effects of e-health on chronically ill patients. J Clin Nurs 2011;20:2997-3010.

4. **Stinson JN, Toomey PC, Stevens BJ, Kagan S, Duffy CM, Huber A, et al.** Asking the experts: exploring the self-management needs of adolescents with arthritis. Arthritis Rheum 2008;59:65-72.

5. **Stinson JN, McGrath PJ, Hodnett ED, Feldman BM, Duffy CM, Huber AM, et al.** An internet-based self-management program with telephone support for adolescents with arthritis: a pilot randomized controlled trial. J Rheumatol 2010;37:1944-1952.

6. **Ammerlaan JJ, Scholtus LW, Drossaert CH, van Os-Medendorp H, Prakken B, Kruize AA, et al.** Feasibility of a Website and a Hospital-Based Online Portal for Young Adults With Juvenile Idiopathic Arthritis: Views and Experiences of Patients. JMIR Res Protoc 2015;4:e102.

7. **van Pelt P, Drossaert C, Kruize A, Huisman J, Dolhain RJEM, Wulffraat N.** Use and perceived relevance of health related internet sites and online contact with peers among young people with Juvenile Idiopathic Arthritis. Rheumatology 2015 (accepted).

8. **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;183:405-409.

9. **Ammerlaan JW, Scholtus LW, Bijlsma HJ, Prakken BJ, Kruize AA.** An urge for change: Transitional care for young adults with juvenile idiopathic arthritis. Patient Educ Couns 2013 Mar 9.

10. **Howland S, Fisher K.** Looking through the patient lens - Improving best practice for young people with juvenile idiopathic arthritis transitioning into adult care. Springerplus 2015;4:111-015-0888-8. eCollection 2015.

11. **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;51:213-219.

12. **Wells CK, McMorris BJ, Horvath KJ, Garwick AW, Scal PB.** Youth report of healthcare transition counseling and autonomy support from their rheumatologist. Pediatr Rheumatol Online J 2012;10:36-0096-10-36.

13. **Prakken B, Albani S, Martini A.** Juvenile idiopathic arthritis. Lancet 2011;377:2138-2149.

14. **Wagner-Weiner L.** Pediatric rheumatology for the adult rheumatologist. J Clin Rheumatol 2008;14:109-119.

15. **Tucker LB, Cabral DA.** Transition of the adolescent patient with rheumatic disease: issues to consider. Rheum Dis Clin North Am 2007;33:661-672.

16. **Shaw KL, Southwood TR, McDonagh JE, British Paediatric Rheumatology Group.** User perspectives of transitional care for adolescents with juvenile idiopathic arthritis. Rheumatology (Oxford) 2004;43(6):770-778.

17. **Lawson EF, Hersh AO, Applebaum MA, Yelin EH, Okumura MJ, von Scheven E.** Self-management skills in adolescents with chronic rheumatic disease: A cross-sectional survey. Pediatr Rheumatol Online J 2011;9(1):35.

18. **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.

19. **Ammerlaan J, van Os-Medendorp H, Scholtus L, de Vos A, Zwier M, Bijlsma H, et al.** Feasibility of an online and a face-to-face version of a self-management program for young adults with a rheumatic disease: experiences of young adults and peer leaders. Pediatr Rheumatol Online J 2014;12:10-0096-12-10.

20. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev 1977;84:191-215.

21. **Lenzen SA, Daniels R, van Bokhoven MA, van der Weijden T, Beurskens A.** Setting goals in chronic care: Shared decision making as self-management support by the family physician. Eur J Gen Pract 2015;21:138-144.

22. **Holloway I, Wheeler S.** Qualitative Research in Nursing. Second edition ed. Oxford: Blackwell Publishing; 2002.

23. **Mueller A, Hartmann M, Mueller K, Eich W.** Validation of the arthritis self-efficacy short-form scale in German fibromyalgia patients. Eur J Pain 2003;7:163-171.

24. **Osborne RH, Elsworth GR, Whitfield K.** The Health Education Impact Questionnaire (heiQ): an outcomes and evaluation measure for patient education and self-management interventions for people with chronic conditions. Patient Educ Couns 2007;66:192-201.

25. **Boers M, Jacobs JW, van Vliet Vlieland TP, van Riel PL.** Consensus Dutch health assessment questionnaire. Ann Rheum Dis 2007;66:132-133.

26. **Englbrecht M, Tarner IH, van der Heijde DM, Manger B, Bombardier C, Muller-Ladner U.** Measuring pain and efficacy of pain treatment in inflammatory arthritis: a systematic literature review. J Rheumatol Suppl 2012;90:3-10.

27. **Moher D, Hopewell S, Schulz KF, Montori V, Gotzsche PC, Devereaux PJ, et al.** CONSORT 2010 explanation and elaboration: updated guidelines for reporting parallel group randomised trials. Int J Surg 2012;10:28-55.

28. **Niedermann K, de Bie RA, Kubli R, Ciurea A, Steurer-Stey C, Villiger PM, et al.** Effectiveness of individual resource-oriented joint protection education in people with rheumatoid arthritis. A randomized controlled trial. Patient Educ Couns 2011;82:42-48.

29. **van Pelt PA, Kruize AA, Goren SS, van der Net J, Uiterwaal C, Kuis W, et al.** Transition of rheumatologic care, from teenager to adult: which health assessment questionnaire can be best used? Clin Exp Rheumatol 2010;28:281-286.

30. **Osborne RH, Hawkins M, Sprangers MA.** Change of perspective: a measurable and desired outcome of chronic disease self-management intervention programs that violates the premise of preintervention/postintervention assessment. Arthritis Rheum 2006;55:458-465.

31. **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.

32. **DeSouza CM, Legedza AT, Sankoh AJ.** An overview of practical approaches for handling missing data in clinical trials. J Biopharm Stat 2009;19:1055-1073.

“I am trying to exercise as much as I can, that is part of self-management. There are some things that you can control yourself. Be positive mentally and exercise as much as possible”

- Female, 44 years old, since 4 years Rheumatoid Arthritis

CHAPTER 6

VALIDATION OF THE DUTCH VERSION OF THE HEALTH EDUCATION IMPACT QUESTIONNAIRE (HEIQ) AND COMPARISON OF THE DUTCH TRANSLATION WITH THE ENGLISH, GERMAN AND FRENCH HEIQ

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ABSTRACT

Background

The Health Education Impact Questionnaire (heiQ) evaluates the effectiveness of health education and self-management programs provided to people dealing with a wide range of conditions. Aim of this study was to translate, culturally adapt and validate the Dutch translation of the heiQ and to compare the results with the English, German and French translations.

Methods

A systematic translation process was undertaken. Psychometric properties were studied among patients with arthritis, atopic dermatitis, food allergy and asthma (n=286). Factorial validity using confirmatory factor analysis, item difficulty (D), item remainder correlation and composite reliability were conducted. Stability was tested using the intra-class correlation coefficient (ICC).

Results

Items were well understood and only minor language adjustments were required. Confirmatory fit indices were >0.95 and item difficulty was D≥0.65 for all items in scales, except for the reversed Emotional distress scale. Composite reliability ranged between 0.67–0.85. Test-retest reliability (n=93). ICC varied between 0.61-0.84. Comparisons with other translations showed comparable fit indices. A lower ICC on Self-monitoring and insight scale was observed.

Conclusions

The Dutch translation of the heiQ was found to be well understood and user friendly by patients with Rheumatoid Arthritis, Atopic Dermatitis, Food allergy and asthma and to have robust psychometric properties for evaluating the impact of health education and self-management programs. Given the wide applications of the heiQ and the comparability of the Dutch results with the English, German and French version, the heiQ is a practical and useful questionnaire to evaluate the impact of self-management support programs in different countries and populations with different diseases.

BACKGROUND

Although several new treatment options for people with chronic conditions like arthritis and atopic dermatitis have been developed in the past decade, patients still experience a large impact of their disease on their daily life (1-5). The chronic nature of the disease imposes daily challenges and patients must make many decisions about the way they manage their lives (6-9). It is not a matter of whether patients self-manage their (chronic) illness, but how they do this (10). An individual's ability to manage the symptoms, treatment, physical and psycho-social consequences and life style changes inherent in living with a chronic condition' is often defined as self-management (11). Over the past decade, several interventions have been developed to improve the self-management of chronically ill patients (12,13). While the initial aim of these interventions was to increase patient's knowledge so they were able to change their behaviour (12,14), the evidence subsequently demonstrated that increased knowledge was not enough. Other theoretical approaches, mostly derived from behavioural sciences, encouraged the movement of self-management interventions towards inclusion of cognitive-behavioural and other approaches (14).

One of the most studied self-management programs, based on Bandura's self-efficacy theory, is the Chronic Disease Self- Management Program (CDSMP) of Stanford University, led by lay people to help people with a chronic disease gain confidence in their ability to control the symptoms and impact of their condition on their lives (12). These programs generated great interest and numerous follow-up studies and government programs ensued (12,15). With the growing opportunities and use of the Internet, online self-management programs for patients with long-term conditions were developed (16, 17). In an uncontrolled longitudinal evaluation of the online CDSMP program, at 12 months small to moderate improvements in health distress, fatigue and pain and self-efficacy were observed. In asthma, it has been shown that online self-management support results in a sustained improvement in disease control and asthma related quality of life (18). This illustrates that self-management interventions can improve quality of life and well-being but their outcomes are varied and include measures of physical, psychological and social well-being (11,14,19). Consequently, the diversity of patient populations, as the different theoretical foundations of the self-management interventions as well as the widely varying educational content of health education programs, make it challenging to demonstrate effectiveness with common metrics (14,19-21).

In response to this issue, Osborne et al (22) developed the Health Education Impact Questionnaire (heiQ) in Australia. The purpose of the heiQ is to provide a direct and realistic measurement of the impact and quality of self-management programs across settings and disease groups and also to provide highly relevant information on the outcomes of self-management programs to health professionals, policymakers and researchers. The development of the heiQ involved a grounded approach rather than a theory-based approach. Techniques used included review of current practice, development of a Program Logic, Concept Mapping, rigorous item development based on the daily language of patients, and extensive item testing and validation in an independent sample. During the development, a wide range of stakeholders were involved, including; patients, health professionals, course leaders, academics and policy makers (22). The original English heiQ (22) contained 42 items across eight independent scales: *Positive and active engagement in life* (five items); *Health directed activity* (four items); *Skill and technique acquisition* (five items); *Constructive attitudes and approaches* (five items); *Self-monitoring and insight* (seven items); *Health service navigation* (five items); *Social integration and support* (five items) and a reversed scale, Emotional distress (formally named Emotional wellbeing), (six items). The Cronbach's alpha of the eight scales varied between α=0.70 (Self-monitoring and insight) and α=0.89 (Emotional distress). Higher values in the heiQ scales indicate better status, except for Emotional distress, where higher values indicate higher distress. Results from additional analysis showed that two items could be removed without compromising the content validity of the questionnaire (23). A recent study on factor structure and measurement invariance of this latest version, confirmed these results (24). Therefore, the revised English heiQ was shortened to 40 items with a 4 point response scale (strongly disagree, disagree, agree and strongly agree) (24).

The heiQ has been translated into many languages including German (23) and French (25). Both versions were translated and culturally adapted using forward and back translation, comprehensibility and content validity testing using interviews (German version) and committee review (French version). The psychometric properties of the German version were studied among 1202 adult patients with a range of chronic diseases from rehabilitation hospitals and the French version was studied among 1030 adult patients with renal failure, diabetes and arthritis. This group was randomly sampled from a health database. Psychometric analysis on reliability, factorial validity and concurrent validity of both the German and French heiQ were consistent with the original English version.

In addition to existing online self-management programs, e.g. in asthma, the UMC Utrecht developed in close cooperation with their patient partners, four online self-management programs that aimed to improve an individual's ability to cope with the symptoms, treatment and consequences of having a chronic disease like atopic dermatitis (AD), food allergy (FA), juvenile idiopathic arthritis (JIA) and arthritis and are developed for different target and age groups like young adults, adults and parents of young patients (26,27). Until now a questionnaire to evaluate the effectiveness of programs across chronic diseases has not been available. The aim for this study was to translate the heiQ into Dutch and then examine its construct validity, stability and reliability. Subsequently, the validity and reliability of the Dutch heiQ was compared with the original English, German and French version heiQ.

METHODS

The study consisted of two steps. First, the original heiQ was translated and culturally adapted into Dutch, following the Deakin University Protocol (28). In the second step, factorial validity, difficulty (D), item remainder correlation, composite reliability and stability (test-retest) were determined. Ultimately, results of the factor structure and reliability of the Dutch translation were compared with results of the original English, revised English, German and French translations of the heiQ.

Translation and cultural adaptation

The original English heiQ (22) was translated by official independent translators using a forward translation followed by a blind back translation. This translation was discussed by three Dutch researchers (JA, HvO, JS) each with fluency in English and experience with the content of questionnaire's constructs, and also the original developer of the questionnaire, native English speaker (RO). Each item was assessed to ensure that the intent was equivalent to the English version and consensus was reached about the translation. The adapted Dutch heiQ was further tested using cognitive interviews in a convenience sample of adult patients with arthritis, atopic dermatitis or food allergy. Following completion of the questionnaire the respondents were asked about their understanding of the questions, acceptability and clarity of wording. All cognitive interview data were discussed to generate a final consensus for the Dutch version.

Psychometric analyses of the Dutch heiQ

Design and procedure

Cross-sectional survey data were used to investigate validity and reliability and two-week follow-up data were obtained on a subset for test-retest reliability. Patients were invited to fill in the questionnaire online after providing informed consent. The Medical Ethics Research Committee (MERC) of the UMC Utrecht and the Leiden UMC declared that this study did not apply to the Medical Research Involving Human Subjects Act and confirmed that official approval was not required. The last step consisted of the comparison of the Dutch heiQ with published reports of original English, revised English, German and French versions.

Population

The study population consisted of adults with atopic dermatitis, food allergy, asthma or rheumatoid arthritis. Inclusion criteria were aged 18 or over; attending the outpatient department of the University Medical Center (UMC) Utrecht the Netherlands, diagnosed with Atopic dermatitis (AD) or Food Allergy (FA) or Rheumatoid Arthritis (RA) or participating in an online self-management program for AD or FA or participating in an internet-based asthma self-management support program by primary care practices in the Leiden region (29) able to read Dutch and have access to Internet.

Sample size estimation

In this study, the heiQ, comprising eight scales (each with 4 to 6 items) was to be subjected to a range of psychometric tests, including confirmatory factor analysis (CFA) to examine how well the hypothesized eight scales fit the data. It has been found that CFA models using relatively small samples of 250-1000 with categorical or ordinal data perform well using the software Mplus (30). We therefore planned to include at least 350 patients.

Psychometric parameters and thresholds

The factorial validity of the Dutch heiQ was investigated by fitting eight single-scale factor models to the data, calculating item-remainder correlations and fitting a full eight-factor model. Fit indices included; Comparative Fit Index (CFI), Tucker Lewis Fit Index (TLI) and Root Mean Square Error of Approximation (RMSEA) and Weighted Root Mean Square Residual (WRMR). Chi-Square Test for Model Fit was also used. We investigated the one-factor model, for each of the eight scales and loadings per item; and the eight-factor model for the total heiQ. We then calculated item Difficulty (D) and Composite Reliability (CR) of scale/individual items. Difficulty was defined as the proportion of positive responses to the item. Stability (test-retest) of the questionnaire was tested using the intra-class correlation co-efficient (ICC). Illness-related variables (duration, severity of complaints/illness, comorbidity) and demographic variables (age, gender, educational level marital status, children) were also collected.

Data analyses

Confirmatory factor analyses were carried out for the one-factor models and the eight-factor model. The mean and variance-adjusted weighted least squares estimator (WLSMV), suitable for the analysis of ordered categorical data, was used for the CFA analyses of the one factor models. The eight-factor model was estimated with robust maximum likelihood (MLR). Difficulty (D) was calculated directly from data on the frequency of item responses while item remainder correlations, composite reliability and their standard errors (SEs) were calculated for each of the eight scales with Mplus code developed by Raykov and colleagues (31).

Following recommendations of Raykov and others et al (32,33) (34), acceptable threshold values for CFI, TLI, RMSEA and WRMR were, respectively, >0.95, >0.95, <0.06 and <1.0. If one factor models were found to have chi-square <0.01 and RMSEA >0.08, correlated residuals were added if suggested by the largest modification index. Acceptable loadings were at least 0.5, according to Nunnally and Burnstein (35). Variation in D gives information on the proportion of respondents who agree or disagree with an item. Therefore a D ranging between 30 and 70% was considered ideal for a scale designed to measure across a relatively broad range of the underlying construct. Item remainder correlations with 95% CI were reported; low item remainder correlation indicated that the question does not fit well in the scale. Composite reliability for full scales without 1 item and for the full scale were computed, including 95% confidence interval; scores of >0.7 were regarded as acceptable (31,36). The ICC was computed to determine test-retest reliability, with ICC >0.7 regarded as acceptable.

Results of the factorial validity and reliability of the Dutch translation were compared with published results of the German (23) and English (22), revised English (24) and French (25) translations. For this comparison, the CFI, RMSEA and Chi-square of model fit, and the composite reliability were collated. Also the performance of eight-factor analyses of the four versions across CFI, TLI and RMSEA (if available) and the intra class correlation per domain of the Dutch and German translation were compared.

RESULTS

Acceptability and comprehensibility of the Dutch translation

Patients with arthritis (n=9), atopic dermatitis (n=4) and food allergy (n=3), 8 men and 8 women with a mean age of 42.5 years (sd 17.3; range 16-73), judged the content and comprehensibility of the Dutch version of the heiQ. Overall, they understood the items as intended and found them acceptable. They had some suggestions like adding specific activity examples for the Dutch population (like biking and swimming) on the Health directed behavior scale which resulted in minor changes.

Psychometric analyses

Participants

A total 286 patients participated in the study; 72% were women, 25% had Atopic Dermatitis, 31% Rheumatoid Arthritis, 23% Food Allergy and 21% asthma. Mean age was 43 years (sd 16; range 18-83 years) (Table 1). Test–retest analyses were carried out with 93 patients, mean age 49 years (sd 17; range 18-83 years). This sample contained 69% woman of whom 12 % had AD, 79% RA and 10% FA.

Table 1 Demographic and clinical characteristics of study populations

	FOOD ALLERGY (FA)	ATOPIC TDERMATITIS (AD)	RHEUMATOID ARTHRITIS (RA)	ASTHMA	TOTAL
n	66	71	88	61	286
Sex: Female n (%)	(77.3)	(64.8)	(69.3)	(83.3)	(72%)
Age: mean, (sd) in years	36.0 (12.5)	34.9 (15.3)	53.6 (15.1)	42.5 (13.7)	42.5 (16.3)
Other demographic data (n)	14	18	88	n.a.	120
Marital status					
Single	50,%	33%	14%		21%
Married	50%	61%	75%		70%
Divorced	0%	0%	9%		7%
Widowed	0%	6%	2%		3%
Level of education[1]					
Low	14%	11%	33%		28%
medium	21%	22%	32%		29%
high	64%	67%	32%		41%
other	0%	0%	3%		3%
Duration of disease mean (sd) in years	15.8 (11.7)	27.3 (14.8)	13.3 (11.8)		15.7 (13.1)
Self-rated health score mean (sd)[3]	7.1 (1.4)	7.3 (1.2)	6.5 (1.6)		6.7 (1.6)
Asthma Control Questionnaire (ACQ) (sd)[4]				1.0 (0.8)	
Food allergy characteristics[2]					
Peanut	50%				
Nuts	73%				
Egg	12%				
Cow's milk	15%				
Other animal products	11%				
Fruits & vegetables	68%				

[1] Low= primary school or lower vocational secondary education, intermediate general secondary education; Middle=intermediate vocational education, higher general secondary education; High= higher vocational education or university education. [2] Individuals may have more than one food allergy; [3] measured on a scale from 1-10 (1 (worst) to 10 (excellent)); [4] ACQ: plausible range 0 - 6, controlled asthma: 0 - 0.75, partly controlled asthma: 0.75 - 1.5, uncontrolled asthma: >1.5.

Table 2 Results of confirmatory factor analyses of the one factor and eight factor model of the Dutch version

					CHI-SQUARE OF MODEL FIT		
	CFI	**RMSEA**	**WRMR**	**TLI**	**VALUE**	**DF**	**P**
One factor models							
Health directed activity	1.00	0.06	0.32	1.00	3.92	2.00	0.14
Positive and active engagement in life	0.97	0.15	0.94	0.95	38.57	5.00	0.00
additional analysis: *heiQ15 WITH heiQ8;*	*1.00*	*0.07*	*0.44*	*1.00*	*10.28*	*4.00*	*0.04*
Emotional distress	0.99	0.10	0.70	0.99	36.21	9.00	0.00
additional analysis: *heiQ12 WITH heiQ4;*	*1.00*	*0.09*	*0.56*	*0.99*	*25.20*	*8.00*	*0.00*
Self-monitoring and insight	0.87	0.16	1.28	0.78	75.91	9.00	0.00
additional analysis: *heiQ17 WITH heiQ3;* *heiQ11 WITH heiQ6;*	*0.99*	*0.06*	*0.49*	*0.98*	*13.01*	*7.00*	*0.07*
Constructive attitudes and approaches	1.00	0.06	0.38	1.00	9.70	5.00	0.08
Skill and technique acquisition	1.00	0.08	0.30	0.99	5.80	2.00	0.05
Social integration and support	0.98	0.15	0.82	0.97	38.35	5.00	0.00
additional analysis *heiQ31 WITH heiQ28;* *heiQ35 WITH heiQ28;*	*1.00*	*0.04*	*0.27*	*1.00*	*4.46*	*3.00*	*0.22*
Health service navigation	0.98	0.18	1.03	0.97	48.82	5.00	0.00
additional analysis: *heiQ33 WITH heiQ32*	*1.00*	*0.09*	*0.47*	*0.99*	*12.97*	*4.00*	*0.01*
Eight-factor model	0.89	0.05	0.064	0.88	1257.77	712	0.00

Acceptable: CFI >0.95; RMSEA < 0.06; WRMR <1.0; TLI >.95

Confirmatory factor analysis

After initial analyses of the one factor models, correlated residuals were added in for five subscales: Positive and active engagement in life, Emotional distress, Self-monitoring and insight, Social integration and support and Health service navigation. This resulted in CFI of all scales being >0.95; RMSEA≤0.06 for four scales: Health directed activity, Self-monitoring and insight, Constructive attitude and approaches, Social integration and support and WRMR <1.0 (Table 2). In table 3 standardized factor loadings with 95% CI of all tested models are shown. Loadings of most scales were >0.5. Two items on the Self-monitoring and insight scale had a lower factor loading in the modified one-factor model, item 3 loaded 0.33 and item 6 loaded 0.48. The RMSEA and the SRMR of the eight factor model indicated good fit. CFI and TLI were, respectively, 0.89 and 0.88.

Item difficulty, item remainder correlation and reliability

Results of item difficulty analysis for Health directed activity, Positive and active engagement in life, Skill and technique acquisition, Self-monitoring and insight, Social integration and support and the reversed scale Emotional distress indicated that the D of items ranged between 0.60 and 0.97 (Table 3). The scales Constructive attitudes and approaches and Health service navigation were found to have D with a smaller range, between 0.87-0.96, indicating that most answers on the items were located in the agree/strongly agree end of the scale.

Item remainder correlation of items (Table 3) of all scales except Self-monitoring and insight were ≥ 0.58, indicating that all of the items hang together well as a scale. Item remainder correlation of items of Self-monitoring and insight varied between 0.35 and 0.76, indicating that there is less cohesion between the items. Composite reliability of the subscale Self-monitoring and insight was 0.67 (95% CI 0.61-0.73). For all other subscales composite reliability was ≥ 0.81. ICC varied between 0.61-0.84 (Table 4).

Table 3 Item Difficulty and Item remainder correlation

	ITEM DIFFICULTY (D)	**95% CONFIDENCE INTERVAL AROUND D**	**ITEM REMAINDER CORRELATION C**	**95% CONFIDENCE INTERVAL AROUND C**	**STANDARDIZED FACTOR LOADINGS ORIGINAL MODEL**	**STANDARDIZED FACTOR LOADINGS MODIFIED MODEL[1]**	**95% CI FOR FACTOR LOADINGS[2]**
Health-directed activity							
heiQ1	0.83	0.78 - 0.87	0.74	0.69 - 0.78	0.83		0.78 - 0.89
heiQ9	0.76	0.71 - 0.81	0.67	0.61 - 0.72	0.78		0.72 – 0.84
heiQ13	0.73	0.68 - 0.78	0.78	0.74 - 0.81	0.90		0.86 – 0.95
heiQ19	0.65	0.59 - 0.70	0.58	0.51 - 0.64	0.67		0.59 – 0.74
Positive and active engagement in life							
heiQ2	0.85	0.80 - 0.88	0.66	0.60 - 0.72	0.78	*0.79*	*0.72 – 0.86*
heiQ5	0.97	0.95 - 0.99	0.65	0.56 - 0.72	0.75	*0.77*	*0.68 – 0.86*
heiQ8	0.83	0.78 - 0.87	0.68	0.61 - 0.73	0.79	*0.70*	*0.62 – 0.78*
heiQ10	0.87	0.82 - 0.90	0.73	0.66 - 0.78	0.84	*0.88*	*0.82 – 0.94*
heiQ15	0.82	0.77 - 0.86	0.60	0.54 - 0.65	0.71	*0.61*	*0.52 – 0.70*
Emotional distress							
heiQ4	0.40	0.34 - 0.46	0.66	0.59 - 0.72	0.73	*0.69*	*0.63 – 0.76*
heiQ7	0.13	0.10 - 0.18	0.66	0.61 - 0.71	0.71	*0.71*	*0.65 – 0.78*
heiQ12	0.25	0.21 - 0.31	0.81	0.78 - 0.85	0.88	*0.86*	*0.82 – 0.90*
heiQ14	0.15	0.12 - 0.20	0.81	0.78 - 0.84	0.90	*0.90*	*0.87 – 0.94*
heiQ18	0.08	0.05 - 0.12	0.83	0.79 - 0.86	0.91	*0.91*	*0.88 - 0.94*
heiQ21	0.12	0.09 - 0.16	0.82	0.79 - 0.85	0.91	*0.91*	*0.88 – 0.94*
Self-monitoring and insight							
heiQ3	0.82	0.77 - 0.86	0.35	0.26 - 0.44	0.52	*0.33*	*0.20 – 0.46*
heiQ6	0.76	0.71 - 0.81	0.43	0.33 - 0.51	0.52	*0.48*	*0.38 – 0.58*
heiQ11	0.94	0.91 - 0.97	0.47	0.39 - 0.55	0.55	*0.51*	*0.38 – 0.64*
heiQ16	0.76	0.71 - 0.81	0.76	0.71 - 0.81	0.67	*0.74*	*0.66 – 0.81*
heiQ17	0.90	0.86 - 0.93	0.75	0.69 - 0.80	0.71	*0.60*	*0.52 - 0.68*
heiQ20	0.87	0.83 - 0.91	0.49	0.39 - 0.57	0.64	*0.70*	*0.62 – 0.78*

	ITEM DIFFICULTY (D)	**95% CONFIDENCE INTERVAL AROUND D**	**ITEM REMAINDER CORRELATION C**	**95% CONFIDENCE INTERVAL AROUND C**	**STANDARDIZED FACTOR LOADINGS ORIGINAL MODEL**	**STANDARDIZED FACTOR LOADINGS MODIFIED MODEL[1]**	**95% CI FOR FACTOR LOADINGS[2]**
Constructive attitude and approaches							
heiQ27	0.96	0.93 - 0.98	0.75	0.68 - 0.80	0.84		0.80 – 0.88
heiQ34	0.92	0.88 - 0.95	0.74	0.70 - 0.78	0.84		0.79 - 0.88
heiQ36	0.91	0.87 - 0.94	0.77	0.74 - 0.80	0.87		0.83 – 0.91
heiQ39	0.92	0.88 - 0.94	0.79	0.75 - 0.83	0.90		0.86 – 0.93
heiQ40	0.90	0.85 - 0.93	0.59	0.54 - 0.64	0.67		0.60 – 0.74
Skill and technique acquisition							
heiQ23	0.69	0.63 - 0.74	0.62	0.57 - 0.67	0.71		0.64 – 0.77
heiQ25	0.84	0.79 - 0.88	0.78	0.73 - 0.83	0.93		0.90 – 0.96
heiQ26	0.86	0.81 - 0.89	0.72	0.67 - 0.77	0.84		0.80 – 0.89
heiQ30	0.80	0.75 - 0.85	0.66	0.62 - 0.70	0.77		0.71 – 0.84
Social integration and support							
heiQ22	0.89	0.85 - 0.92	0.72	0.68 - 0.76	0.80	*0.77*	*0.72 – 0.83*
heiQ28	0.83	0.78 - 0.87	0.69	0.63 - 0.73	0.79	*0.89*	*0.82 – 0.95*
heiQ31	0.71	0.66 - 0.76	0.68	0.62 - 0.74	0.80	*0.83*	*0.78 – 0.88*
heiQ35	0.91	0.87 - 0.94	0.77	0.73 - 0.80	0.86	*0.88*	*0.84 – 0.92*
heiQ37	0.83	0.78 - 0.87	0.72	0.68 - 0.76	0.79	*0.76*	*0.70 – 0.82*
Health service navigation							
heiQ24	0.92	0.88 - 0.95	0.73	0.67 - 0.78	0.86	*0.87*	*0.82 – 0.92*
heiQ29	0.91	0.87 - 0.94	0.69	0.64 - 0.73	0.77	*0.79*	*0.73 – 0.84*
heiQ32	0.96	0.93 - 0.98	0.69	0.63 - 0.74	0.79	*0.70*	*0.62 – 0.77*
heiQ33	0.92	0.88 - 0.94	0.72	0.69 - 0.74	0.85	*0.78*	*0.72 – 0.84*
heiQ38	0.87	0.83 - 0.91	0.76	0.71 - 0.81	0.86	*0.89*	*0.85 – 0.93*

[1] standardized factor loadings of one-factor models (based on the models with correlated residuals as in table 2);
[2] 95% confidence interval of original model (set roman/right up or of the modified model in italic)

Table 4 Composite reliability and intra class correlation per domain

	COMPOSITE RELIABILITY [1]	CONFIDENCE INTERVAL	INTRA CLASS CORRELATION (N=93)	95% CONFIDENCE INTERVAL LOWER BOUND - UPPER BOUND
Health directed activity	0.82	0.79 - 0.86	0.84	0.77 - 0.89
Positive and active engagement in life	0.81	0.78 - 0.85	0.74	0.63 - 0.82
Emotional distress	0.89	0.87 - 0.91	0.86	0.80 - 0.91
Self-monitoring and insight	0.67	0.61 - 0.73	0.61	0.47 - 0.73
Constructive attitudes and approaches	0.86	0.83 - 0.89	0.69	0.57 - 0.78
Skill and technique acquisition	0.82	0.78 - 0.85	0.67	0.55 - 0.77
Social integration and support	0.85	0.83 - 0.88	0.73	0.62 - 0.81
Health service navigation	0.85	0.82 - 0.88	0.65	0.52 - 0.75

[1] Acceptable: CR>0.7

Table 5 Results of confirmatory factor analyses of the Dutch (NL), German (G) and English (ENG) and English Revised (ENG-R) translation of the heiQ

	CFI				RMSEA				CHI-SQUARE OF MODEL FIT P VALUE			
	NL	G	ENG	ENG-R	NL	G	ENG	ENG-R	NL	G	ENG	ENG-R
Health directed activity	1.00	0.99	0.99	1.00	0.06	0.07	0.09	0.08	0.14	0.00	0.00	0.00
additional analysis: *1 with 13;*		*1.00*		*1.00*		*0.03*		*0.02*		*0.13*		*0.10*
Positive and active engagement in life	0.97	0.99	0.99	1.00	0.15	0.04	0.05	0.07	0.00	0.01	0.04	0.00
additional analysis: *15 with 8;*	*1.00*			*1.00*	*0.07*			*0.04*	*0.04*			*0.00*
Emotional distress	0.99	0.98	0.98	0.99	0.10	0.06	0.07	0.10	0.00	<0.00	<0.00	0.00
additional analysis: *12 with 4;*	*1.00*				*0.09*				*0.00*			

	CFI				RMSEA				CHI-SQUARE OF MODEL FIT P VALUE			
	NL	**G**	**ENG**	**ENG-R**	**NL**	**G**	**ENG**	**ENG-R**	**NL**	**G**	**ENG**	**ENG-R**
additional analysis: 4 with 18;		*0.99*				*0.04*				*0.00*		
additional analysis: Without 18;		*1.00*				*0.01*				*0.39*		
Additional analysis: 7 with 4; 21 with 18				*1.00*				*0.07*				*0.00*
Self-monitoring and insight	0.87	0.92	0.99[1]	0.97	0.16	0.09	0.05[1]	0.08	0.00	<0.00	<0.00[1]	0.00
additional analysis: 17 with 3;11 with 6;	*0.99*				*0.06*				*0.07*			
additional analysis: 17 with 3		*0.99*		*0.99*		*0.04*		*0.06*		*0.01*		*0.00*
Constructive attitudes and approaches	1.00	0.99	1.00	1.00	0.06	0.06	0.03	0.06	0.08	<0.00	0.17	0.00
Skill and technique acquisition	1.00	1.00	1.00[2]	1.00	0.08	<0.00	0.00[2]	0.08	0.05	0.59	0.44[2]	0.00
additional analysis: 25 with 23 (or 30 with 26)				*1.00*				*0.03*				*0.04*
Social integration and support	0.98	0.96	0.99	0.99	0.15	0.12	0.05	0.14	0.00	<0.00	0.05	0.00
additional analysis: 31 with 28;35 with 28;	*1.00*				*0.04*				*0.22*			
additional analysis: 28 with 22;		*0.99*		*1.00*		*0.07*		*0.05*		*<0.00*		*0.00*
Health service navigation	0.98	0.96	1.00	0.98	0.18	0.09	0.00	0.13	0.00	<0.00	0.44	0.00
additional analysis: 33 with 32;	*1.00*				*0.09*				*0.01*			
additional analysis: 29 with 32;		*0.99*				*0.06*				*0.00*		
additional analysis: 29 with 32;29 with 24				*1.00*				*0.06*				*0.00*

Acceptable: CFI >0.95; RMSEA <0.06; Chi-square model Fit: p <0.05.

[1] The English heiQ (ENG) Self-monitoring and insight scale contains 7 items (including 'I know when my lifestyle is creating health problems for me).

[2] The English heiQ (ENG) Skill and technique acquisition scale contains 5 items (including 'I have effective skills that help me handle stress')

Comparison of the Dutch heiQ (NL) with the German (G), English (ENG), English revised (ENG-R) and French heiQ translations.

Comparison of confirmatory factor analyses across language versions

Comparison of the one factor validation of the Dutch (NL), German (G) (23), English (ENG) (22) and English revised (ENG-R) (24) translations showed acceptable CFI for all translations (see Table 5). In most translations, the RMSEA was acceptable except for the Dutch translation where only the scale Social integration and support scored 0.04.

The p-value of Chi square Model fit was <0.10; except for Health directed activity (Dutch translation); Constructive attitudes and approaches (English translation), Skills and techniques (German and English translation) and Health service navigation (English translation).

Comparison of composite reliability across language versions

Composite reliability was acceptable for the Dutch, German and English translations except for the domain Self-monitoring and insight of the Dutch translation (see Table 6). The reliability of this domain was also relatively low for the English translation (0.70). The reliability of the French translation ranged from 0.74 to 0.89 (25).

Table 6 Results of the composite reliability of the Dutch, German, English (original) and English (revised) versions of the heiQ

	DUTCH	GERMAN	ENGLISH (ORIGINAL)[3]	ENGLISH (REVISED)
Health directed activity	0.82	0.83	0.80	0.83
Positive and active engagement in life	0.81	0.75	0.86	0.83
Emotional distress	0.89	0.88	0.89	0.86
Self-monitoring and insight[1]	0.67	0.74	0.70[1]	0.74
Constructive attitude and approaches	0.86	0.87	0.81	0.88
Skills and techniques acquisition[2]	0.82	0.77	0.81[2]	0.80
Social integration and support	0.85	0.88	0.86	0.88
Health service navigation	0.85	0.87	0.82	0.85

Acceptable: Composite reliability >0.7

[1] The English heiQ (original) Self-monitoring and insight scale contained 7 items (including 'I know when my lifestyle is creating health problems for me).

[2] The English heiQ (original) Skill and technique acquisition scale contains 5 items (including 'I have effective skills that help me handle stress').

[3] Cronbach's alpha was used in the original English heiQ analysis

Comparison of eight factor models across language versions

Comparison the eight factor model of the Dutch, English, German and French translations showed similar results for CFI and RMSEA (see Table 7).

Table 7 Comparison of the CFI, TLI and RMSEA of the Dutch, German, English and French versions of the heiQ, eight factor model

CFA MODEL	CFI	TLI	RMSEA
Dutch	0.89	0.88	0.05
German	0.92	n.a	0.04
English	0.99	n.a	0.04
French	0.92	0.91	0.04

Acceptable: CFI >0.95; TLI >.95; RMSEA < 0.06

Comparison of Intra Class Correlation (ICC) across language versions

For the comparison of the ICC, only the results of the Dutch and German translation were available. The Dutch and German translations had an ICC <0.7 on the scales Self-monitoring and insight and Health service navigation. The German translation also had ICC <0.7 on Health directed activity, while the Dutch translation had an ICC <0.7 on Constructive attitude and approaches and Skills and techniques acquisition (see Table 8).

Table 8 Comparison of the intra class correlation per domain for the Dutch and German version of the heiQ

	DUTCH N=93	GERMAN N=69
Health directed activity	0.84	0.60
Positive and active engagement in life	0.74	0.72
Emotional distress	0.86	0.77
Self-monitoring and insight	0.61	0.63
Constructive attitude and approaches	0.69	0.77
Skills and techniques acquisition	0.67	0.72
Social integration and support	0.73	0.83
Health service navigation	0.65	0.68

Acceptable: ICC> 0.7

DISCUSSION

In this study, the English heiQ was translated into the Dutch language and psychometric properties were determined. The results show that the Dutch heiQ has good psychometric properties in diverse groups of patients indicating that it is likely to be a robust outcomes measure of health education and self-management programs in The Netherlands. We also showed that psychometric properties of the Dutch translation were comparable with the English, German and French translations, which provides evidence that the heiQ conceptualizes a broad range of self-management skills in a consistent way across chronic conditions, cultures and languages.

Confirmatory Factor Analysis was used to investigate the factorial validity, showing acceptable fit indices for all eight scales of the Dutch translation of the heiQ. The RMSEA and the SRMR of the eight factor model indicated good fit, CFI and TLI were lower. It is known that CFI and TLI do not function well in correctly specified models with larger numbers of variables, while the RMSEA tends to improve in correctly specified models with large numbers of variables (37). The data indicate that the eight factor structure is maintained across settings with different languages, cultures and healthcare systems.

There appears to be an appropriate level and range of difficulty for most scales; Health directed activity, Positive and active engagement in life, Emotional distress, Self-monitoring and insight, Skill and technique acquisition and Social integration and support. However, for Constructive attitudes and approaches and Health service navigation, a more restricted range of difficulties were found which may mean that these scales may not have strong discrimination between persons. Item difficulty is a reflection of the quality of items and the status of the people completing the questionnaire. In our sample, the majority of people answered the items with strongly agree. The respondents were relatively young and the average duration of living with their condition was over 10 years. It is likely that this construct, like many of them, changes greatly over time, and over the course of the disease the respondents in our study came to have a very good understanding of the healthcare system and have a constructive attitude. Further work on the heiQ is needed to explore change over time, including pathways from diagnosis to effective long term self-management.

Reliability of the subscales of the Dutch translation is acceptable. However the domain Self-Monitoring and insight has lower item remainder correlation and composite reliability (0.67) than other scales**.** Self-monitoring and Insight captures *'the individuals' ability to monitor their condition and their physical and or emotional responses that leads to insight and appropriate actions to self-manage'.* The relatively low reliability of this domain accords with the English and German versions. It is possible that this domain consists of two separate concepts: a) self-monitoring (check and action) and b) insight and understanding of the underlying disease processes. We also found relatively low factor loadings on three items in this domain (Table 3), indicating the limited relationship between the items and the latent construct. Future research is needed to identify if this domain could be redeveloped into two separate constructs.

Four domains showed test-retest interclass correlation scores of <0.7 (Self-monitoring and Insight (0.61), Constructive attitude and approaches (0.69), Skills and Technique (0.67) and Health service navigation (0.65)), indicating modest stability, and suggesting that these constructs are less stable over time. People with Rheumatoid Arthritis and Atopic Dermatitis do experience exacerbations and remissions over time (4,5) which influences test-retest estimates. Test-retest was also examined in validation of the German translation, showing comparable results, except for two scales. For the Health directed activity scale the ICC of Dutch translation was 0.84, while the German translation had a substantially lower ICC of 0.60. The opposite was found for Social integration and support, where the ICC for the Dutch translation (0.73) was somewhat lower, compared with the German translation (0.83). Differences in these results could be due to differences in how the items were translated, cultural factors or the study population – regarding the latter, our study mostly comprised people with RA versus orthopedic patients in the German test-retest.

Translation of the original English version of the heiQ was carried out according to international standards, including a back translation and review by a multidisciplinary team, including the developer of the questionnaire. The aim of these standards is to achieve a questionnaire of which results could be compared across different languages. Epstein et al (25) revealed that a multidisciplinary expert team is a critical contributor to psychometric properties. Importantly, in our study, we also used a carefully constructed item intent guidance document, and one of the authors of the heiQ (RO), who has chaired the translation of over 20 language versions of the heiQ.

Comparison of the psychometric properties of the Dutch, German (23), English (22) and French (25) translation of the heiQ showed that most domains have good fit indices. The importance of good fit indices for, e.g., the one-factor models, is that they indicate greater scale unidimensionality (all items measuring the same construct). Unidimensionality is distinct from reliability and is, arguably, at least as important (38). In addition, Brunet et al (39) showed that 5 domains of the French- and English translation measured these domains in the same way across both language groups. The study population of this Dutch study consisted of people with chronic illnesses including Rheumatoid Arthritis, Atopic Dermatitis, Food allergy and asthma, but we did not include other important chronic conditions such as cardiovascular conditions, cancer or diabetes. Given that the heiQ has been widely taken up across different settings and diseases (40,41), and psychometric analyses continue to provide a growing web of evidence that it is has robust properties, the questionnaire is likely to continue to have relevance for different stakeholders and support them in decision-making about the value and impact of self-management and health education programs.

CONCLUSIONS

The Dutch translation of the heiQ was found to be well understood by patients with Rheumatoid Arthritis, Atopic Dermatitis, Food allergy and asthma and to have robust psychometric properties for evaluating the impact of health education and self-management programs. Given the wide applications of the heiQ and the comparability of the Dutch results with the English, German and French version, we conclude that the heiQ is a practical and useful questionnaire to evaluate the impact of self-management support programs in different countries and populations with different diseases.

LIST OF ABBREVIATIONS

ACQ: Asthma Control Questionnaire
AD: Atopic Dermatitis
CDSMP: Chronic Disease Self-management Program
CFA: Confirmatory Factor Analysis
CFI: Comparative Fit Index
CI: Confidence Interval
CR: Composite Reliability
D: Item Difficulty
ENG: English translation
ENG-R: English Revised translation
FA: Food Allergy
G: German translation
heiQ: Health Education Impact Questionnaire
ICC: Intra-class Correlation Coefficient
JIA: Juvenile Idiopathic Arthritis
MERC: Medical Ethics Research Committee
MLR: Maximum Likelihood
NL: Dutch translation
RMSEA: Root Mean Square Error of Approximation
SD: Standard Deviation
SEs: Standard Errors
TLI: Tucker Lewis Fit Index
UMC Utrecht: University Medical Center Utrecht
WLSMV: Weighted Least Squares Estimator
WRMR: Weighted Root Mean Square Residual

REFERENCES

1. **Kiebert G, Sorensen SV, Revicki D, Fagan SC, Doyle JJ, Cohen J, Fivenson D.** Atopic dermatitis is associated with a decrement in health-related quality of life. Int.J.Dermatol. 2002;41:151-158.

2. **Le TM, Flokstra-de Blok BM, van Hoffen E, Lebens AF, Goossens NJ, Dubois AE, Bruijnzeel-Koomen CA, Knulst AC.** Quality of life is more impaired in patients seeking medical care for food allergy. Int.Arch.Allergy Immunol. 2013;162:335-339.

3. **Peniamina RL, Bremer P, Conner TS, Mirosa M.** Understanding the Needs of Food-Allergic Adults. Qual.Health Res. 2014;24:933-945.

4. **Eichenfield LF, Tom WL, Chamlin SL, Feldman SR, Hanifin JM, Simpson EL, Berger TG, Bergman JN, Cohen DE, Cooper KD, Cordoro KM, Davis DM, Krol A, Margolis DJ, Paller AS, Schwarzenberger K, Silverman RA, Williams HC, Elmets CA, Block J, Harrod CG, Begolka WS, Sidbury R.** Guidelines of care for the management of atopic dermatitis: Section 1. Diagnosis and assessment of atopic dermatitis. J.Am.Acad.Dermatol. 2013.

5. **Overman CL, Jurgens MS, Bossema ER, Jacobs JW, Bijlsma JW, Geenen R.** Change of psychological distress and physical disability in patients with rheumatoid arthritis over the last two decades. Arthritis Care.Res.(Hoboken) 2014;66:671-678.

6. **Salaffi F, Carotti M, Gasparini S, Intorcia M, Grassi W.** The health-related quality of life in rheumatoid arthritis, ankylosing spondylitis, and psoriatic arthritis: a comparison with a selected sample of healthy people. Health.Qual.Life.Outcomes 2009;7:25-7525-7-25.

7. **Kristiansen TM, Primdahl J, Antoft R, Horslev-Petersen K.** Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: a focus group study. Musculoskeletal Care. 2012;10:29-38.

8. **Zuberbier T, Orlow SJ, Paller AS, Taieb A, Allen R, Hernanz-Hermosa JM, Ocampo-Candiani J, Cox M, Langeraar J, Simon JC.** Patient perspectives on the management of atopic dermatitis. J.Allergy Clin.Immunol. 2006;118:226-232.

9. **Hamnes B, Hauge MI, Kjeken I, Hagen KB.** 'I have come here to learn how to cope with my illness, not to be cured': a qualitative study of patient expectations prior to a one-week self-management programme. Musculoskeletal Care. 2011;9:200-210.

10. **Bodenheimer T, Lorig K, Holman H, Grumbach K.** Patient self-management of chronic disease in primary care. JAMA 2002;288:2469-2475.

11. **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.

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

13. **Newman S, Steed L, Mulligan K.** Self-management interventions for chronic illness. Lancet 2004;364:1523-1537.

14. **Iversen MD, Hammond A, Betteridge N.** Self-management of rheumatic diseases: state of the art and future perspectives. Ann.Rheum.Dis. 2010;69:955-963.

15. **Osborne RH, Spinks JM, Wicks IP.** Patient education and self-management programs in arthritis. Med.J.Aust. 2004;180:S23-6.

16. **Lorig KR, Ritter PL, Dost A, Plant K, Laurent DD, McNeil I.** The Expert Patients Programme online, a 1-year study of an Internet-based self-management programme for people with long-term conditions. Chronic Illn. 2008;4:247-256.

17. **van der Meer V, Bakker MJ, van den Hout WB, Rabe KF, Sterk PJ, Kievit J, Assendelft WJ, Sont JK, SMASHING (Self-Management in Asthma Supported by Hospitals, ICT, Nurses and General Practitioners) Study Group.** Internet-based self-management plus education compared with usual care in asthma: a randomized trial. Ann.Intern.Med. 2009;151:110-120.

18. **van Gaalen JL, Beerthuizen T, van der Meer V, van Reisen P, Redelijkheid GW, Snoeck-Stroband JB, Sont JK, SMASHING Study Group.** Long-term outcomes of internet-based self-management support in adults with asthma: randomized controlled trial. J.Med.Internet Res. 2013;15:e188.

19. **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.

20. **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.

21. **Kroon FP, van der Burg LR, Buchbinder R, Osborne RH, Johnston RV, Pitt V.** Self-management education programmes for osteoarthritis. Cochrane Database Syst.Rev. 2014;1:CD008963.

22. **Osborne RH, Elsworth GR, Whitfield K.** The Health Education Impact Questionnaire (heiQ): an outcomes and evaluation measure for patient education and self-management interventions for people with chronic conditions. Patient Educ.Couns. 2007;66:192-201.

23. **Schuler M, Musekamp G, Faller H, Ehlebracht-Konig I, Gutenbrunner C, Kirchhof R, Bengel J, Nolte S, Osborne RH, Schwarze M.** Assessment of proximal outcomes of self-management programs: translation and psychometric evaluation of a German version of the Health Education Impact Questionnaire (heiQ). Qual.Life Res. 2013;22:1391-1403.

24. **Elsworth GR, Nolte S, Osborne RH.** Factor structure and measurement invariance of the Health Education Impact Questionnaire: Does the subjectivity of the response perspective threaten the contextual validity of inferences?. SAGE Open Med. 2015;3:2050312115585041.

25. **Epstein J, Osborne RH, Elsworth GR, Beaton DE, Guillemin F.** Cross-cultural adaptation of the Health Education Impact Questionnaire: experimental study showed expert committee, not back-translation, added value. J.Clin.Epidemiol. 2015;68:360-369.

26. **van Os-Medendorp H, van Leent-de Wit I, de Bruin-Weller M, Knulst A.** Usage and users of online self-management programs for adult patients with atopic dermatitis and food allergy: an explorative study. JMIR Res.Protoc. 2015;4:e57.

27. **Ammerlaan J, van Os-Medendorp H, Scholtus L, de Vos A, Zwier M, Bijlsma H, Kruize AA.** Feasibility of an online and a face-to-face version of a self-management program for young adults with a rheumatic disease: experiences of young adults and peer leaders. Pediatr.Rheumatol.Online J. 2014;12:10-0096-12-10.

28. **Hawkins M, Osborne RH.** Questionnaire Translation and Cultural Adaptation Procedure Version 1.0, 2010.

29. **van Gaalen JL, Bakker MJ, van Bodegom-Vos L, Snoeck-Stroband JB, Assendelft WJ, Kaptein AA, van der Meer V, Taube C, Thoonen BP, Sont JK, IMPASSE study group.** Implementation strategies of internet-based asthma self-management support in usual care. Study protocol for the IMPASSE cluster randomized trial. Implement Sci. 2012;7:113-5908-7-113.

30. **Beauducel A, Herzberg PY.** Performance of Maximum Likelihood Versus Means and Variance Adjusted Weighted Least Squares Estimation in CFA. Structural Equation Modeling: A Multidisciplinary Journal 2006;13:186.

31. **Raykov T, Marcoulides, GA.** Introduction to Psychometric Theory. New York: Routledge, 2011.

32. **Hu L, Bentler PM.** Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. Structural Equation Modeling: A Multidisciplinary Journal 1999;6:1.

33. **Browne MW CR.** Alternative ways of assessing model fit. In: Bollen KA LJ ed. Testing Structural Equation Models: Newbury Park, CA: Sage, 1993;136–162.

34. **Schuler M, Musekamp G, Faller H, Ehlebracht-Konig I, Gutenbrunner C, Kirchhof R, Bengel J, Nolte S, Osborne RH, Schwarze M.** Assessment of proximal outcomes of self-management programs: translation and psychometric evaluation of a German version of the Health Education Impact Questionnaire (heiQ). Qual.Life Res. 2013;22:1391-1403.

35. **De Vet HCW, Terwee CB, Mokkink LB, Knol D.** Chapter 4. Field-testing: item reduction and data structure. Measurement in medicine. United Kingdom: Cambridge University Press, 2015;65.

36. **Raykov T.** Scale construction and development using structural equation modeling. In: Hoyle RH ed. Handbook of Structural Equation Modeling. New York: The Guilford Press, 2012;472.

37. **Kenny DA, McCoach DB.** Effect of the Number of Variables on Measures of Fit in Structural Equation Modeling. Struct.Equ.Model. 2003;10:333-351.

38. **Hattie JA.** Methodology review: Assessing unidimensionality of tests and items. Applied Psychological Measurement. 1985;9:139-164.

39. **Brunet J, Lauzier S, Campbell HS, Fillion L, Osborne RH, Maunsell E.** Measurement invariance of English and French Health Education Impact Questionnaire (heiQ) empowerment scales validated for cancer. Qual.Life Res. 2015;24:2375-2384.

40. **Maunsell E, Lauzier S, Brunet J, Pelletier S, Osborne RH, Campbell HS.** Health-related empowerment in cancer: validity of scales from the Health Education Impact Questionnaire. Cancer 2014;120:3228-3236.

41. **Osborne RH, Batterham R, Livingston J.** The evaluation of chronic disease self-management support across settings: the international experience of the health education impact questionnaire quality monitoring system. Nurs.Clin.North Am. 2011;46:255-270.

> *“Everybody applies self-management in their lives, one person more than the other, depending on the situation”*
>
> *- Male, 65 years old, since 12 years Rheumatoid Arthritis*

CHAPTER 7

PREFERENCES AND NEEDS OF PATIENTS WITH A RHEUMATIC DISEASE REGARDING CONTENT AND STRUCTURE OF ONLINE SELF-MANAGEMENT SUPPORT

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ABSTRACT

Objective

Aim of this study was to investigate preferences and needs regarding the structure and content of a person-centered online self-management support intervention for patients with a rheumatic disease.

Methods

A four step procedure, consisting of online focus group interviews, consensus meetings with patient representatives, card sorting task and hierarchical cluster analysis was used to identify the preferences and needs.

Results

Preferences concerning the structure involved 1) suitability to individual needs and questions, 2) fit to the life stage 3) creating the opportunity to share experiences, be in contact with others, 4) have an expert patient as trainer, 5) allow for doing the training at one's own pace and 6) offer a brief intervention. Hierarchical cluster analysis of 55 content needs comprised eleven clusters: 1) treatment knowledge, 2) societal procedures, 3) physical activity, 4) psychological distress, 5) self-efficacy, 6) provider, 7) fluctuations, 8) dealing with rheumatic disease, 9) communication, 10) intimate relationship, and 11) having children.

Conclusion

A comprehensive assessment of preferences and needs in patients with a rheumatic disease is expected to contribute to motivation, adherence to and outcome of self-management-support programs.

Practice Implications

The overview of preferences and needs can be used to build an online self-management intervention.

1. INTRODUCTION

Patients with a rheumatic disease face the challenge of managing their disease and its consequences for daily life. Self-management is the individual's ability to deal with symptoms, treatment, physical and psychosocial consequences, and life style changes inherent to a chronic condition (1). Self-management interventions often combine information-based and cognitive-behavioral strategies (2). In rheumatology, especially self-efficacy theory (3) has guided self-management programs intended to improve the skills necessary to deal with disease-related problems. With the growing opportunities and use of the internet, self-management programs are becoming available online as well (4, 5). There is, however, no consistent (long-term) evidence of the effectiveness of self-management programs for chronic patients in general (2, 6). This might be due to diversity of interventions in terms of format and contents, insufficient theoretical fundament, and heterogeneity of patient populations (2, 6, 7). Moreover, positive mean group outcomes sometimes disguised the substantial proportion of patients who did not comply with or respond to the intervention (2, 6).

A basic assumption in initiating self-management interventions is that the patient will be more motivated, adhere better and benefit more and for a longer time when the intervention is customized to the individual needs and preferences of the patient (6, 8, 9). For instance, while a young parent with a rheumatic disease may want to learn and practice skills needed to raise children, an older person may want help in dealing with frailty due to the disease and old age. A theory, consistent to this assumption, is self-determination theory (10) which emphasizes the importance of keeping goals of behavior change close to the autonomous motivation of people. Behavior change and long-term adherence to changed behavior are expected to be larger when a patient conceives a meaningful rationale for change, values the changed behavior, and aligns it with other central values and lifestyle patterns. Support for this assumption has been offered (11) and emphasizes the need for an individualized and engaged approach to patient guidance, taking their individual needs and circumstances as point of departure.

The contents of self-management interventions that mostly originated form self-efficacy theory comprised elements such as mastering experiences, role modelling, reinterpretation of symptoms, psychological consequences, and provision of information from a persuasive or credible source (1, 12). In the past decade, some studies examined individual and group needs of patients regarding self-management support. Patients with rheumatic diseases wanted health professionals to help them with self-management in general (13) and more specifically, they wanted health professionals to provide information and support to manage pain (14). One study using a scoping review method concluded that patients with RA have informational, emotional, social and practical support needs (15). Another study based upon a study of barriers to self-care proposed that four dimensions should be included in self-management interventions: medical management, communication with healthcare providers, coping with consequences, and lifestyle changes (16). Other needs regarding self-management mentioned in literature were e.g., dealing with limitations (17), working together with health professionals, getting psychosocial support (18), empowerment, and dealing with emotional responses (13,18).

We wanted to extend these previous findings 1) by adopting a truly open approach with individual patients collaborating during the whole research cycle from defining research goals, to choosing the design and offering input, and analyzing and describing the results, 2) by examining specifically needs and preferences of patients with rheumatic diseases, 3) by including not only needs regarding content but also preferences regarding the structure, which was not done before, 4) by offering an overview of needs and preferences that are expressed in a concrete and specific rather than an abstract and global way, and 5) by choosing a method that inventories what kind of self-management support needs an individual patient may have instead of summarizing what a group needs. Therefore, the aim of the present study was to examine the individual needs concerning content and preferences regarding the structure of person-centered online self-management support training in adult patients with a rheumatic disease. To that aim we used a concept mapping technique (19) consisting of online focus groups, card sorting tasks, and hierarchical cluster analysis.

2. METHODS

2.1. Design

A four step procedure was used. First, focus group interviews with adult patients with rheumatic diseases yielded needs and preferences. Second, a project group consisting of patient representatives and professionals from rheumatology, communication sciences, nursing sciences and psychology, analyzed the results of the interviews to differentiate preferences regarding structure and needs regarding content, and summarized the ‘preferences for structure’ during consensus meetings. Third, in a card sorting task, patients with a rheumatic disease grouped the selected content needs by similarity and importance. Fourth, hierarchical cluster analysis was used to classify the needs into clusters.

2.2. Patient selection

Adult (age ≥ 18yrs) patients, diagnosed with a rheumatic disease and able to speak and read Dutch, were recruited for a focus group and card sorting task separately through announcements on the website of the Dutch Arthritis Foundation and Twitter (@reumauitgedaagd). This announcement asked for patients who wanted to help with the development of an online self-management intervention. Patients were asked to share their needs, preferences and thoughts on contents, structure and layout of the new to develop online intervention. Thus, a volunteer convenience sample was used. For focus groups a sample size of at least twelve participants has been suggested to provide a variety of perspectives (20). A sample size between 10 and 20 people has been suggested to be a workable number for a card sorting task (19) and as few as 25-30 participants will likely yield results similar to those of several hundred, provided these participants are representative of actual users and are familiar with the domain being considered (21). All participants received an information letter explaining the aim of the study and a brief self-report questionnaire for demographic characteristics, and all provided written informed consent. The study was conducted according to the principles of the Declaration of Helsinki (22). The medical-ethical review board of the University Medical Center Utrecht decided that the Medical Research Involving Human Subjects Act did not apply to this study.

2.3. Variables, data collection and analyses

2.3.1. Step 1: Identifying content needs and preferences regarding the structure in a focus group

Participants were asked to join a closed, private focus group on Facebook during four weeks. The online focus group was facilitated by two moderators (LS, JA) who guided and stimulated the online, written discussion. They encouraged each participant to elaborate on his or her answer and to react to each other’s comments by using question words like ‘what’ , ‘if’ and ‘how’. The discussion started with an open question. The leading question with respect to content needs was “If you want to work on improvement of your self-management skills, what themes should the self-management training consist of?". Examples of questions to identify preferences for structure were: "would you like to do the training on your own or in an online group?" and "what skills should a trainer have to guide you through the training?". To ensure that the discussion yielded as much as possible needs and preferences, the moderators, summarized regularly and checked if there was anything else the participants wanted to discuss about the subject of self-management.

2.3.2. Step 2: Analyzing the results of the focus group on preferences for structure and content needs

A generic qualitative approach (20) was used for data analysis, including coding, categorizing, and discussion by the project group members. First, the written transcripts of the focus group were copied and sent out to the group. After reading the transcript and assessing the whole scope of the interviews, each member individually coded fragments of the transcript on 'structure' and 'content'. The fragments on structure were categorized and discussed in the project group until consensus was reached. Regarding the fragments on content, corresponding needs were put together, and ambiguous and abstract needs and needs that did not relate to online self-management support were removed. The remaining needs were evaluated by the project group on their suitability and comprehensibility for the card-sort task until consensus was reached.

2.3.3. Step 3: Grouping the needs by similarity and importance in a card sorting task

The selected needs on content (step 2) were numbered randomly and written on separate cards for use in the card sorting task. Each content need fitted the format "I have a need for…". The card sorting task was performed at Utrecht University or, when the patient could not attend this session, at home. Each participant performed, individually, two types of sorting tasks on a table and wrote down the results on a score form. In the first task, participants individually sorted the needs into piles according to similarity of contents. To prevent that too few or too many piles were formed, the following rules applied: all needs had to be placed in a pile; each need could be placed in one pile only; each pile could contain 2-25 needs; and 4-20 piles could be formed. The participants gave the piles a label that could be used by the researchers to interpret the sorting. In a second task, the participants individually sorted the needs based on importance. For this task, the following rules applied: all needs had to be placed in a pile; each need could be placed in one pile only; exactly five piles had to be formed from the least important needs (Pile 1) to most important needs (Pile 5); and needs had to be distributed equally across the five piles.

2.3.4. Step 4: Classifying the needs content into clusters by Hierarchical Cluster Analysis

Hierarchical cluster analysis in SPSS statistical software version 21.0 (SPSS, Chicago, IL) (23) was used to classify needs that were individually sorted by the participants during the card sorting task according to similarity of meaning in a hierarchy of clusters. In cluster analysis, the cells of the input matrix of needs comprised the number of times that two needs were *not* sorted in the same pile. Squared Euclidean distances were computed between each pair of needs and Ward's method was used to derive the hierarchical structure of needs. The main criterion to decide on the number of clusters was that the clusters should reflect distinct components of needs. The final number of clusters was set by all the members of the project group, guided by the dendrogram and the agglomeration schedule produced by the statistical software program showing which statements were being combined at each stage of the hierarchical clustering process.

In the first stage we used a top-down interpretation starting with two clusters, then three and so on until additional clusters did not yield new content. In the second stage, the contents of both a lower and a higher number of clusters were compared to finally decide on the number of clusters, based on consensus of the project group . The scores of the second sorting task were used to calculate for separate needs and clusters a mean importance score and standard deviation.

3. RESULTS

3.1. Participants

Two men and 33 women participated in the online focus group, and 21 women (of which 3 had also participated in the online focus group) and 3 men participated in the card sorting task (table 1). Fourteen participants performed the card sorting at Utrecht University and 10 participants at home.

Table 1 Demographic characteristics of participants in the online focus group and the card sorting task group

	FOCUS GROUP (N=35)	CARD SORTING TASK (N=24)
Female sex, n	33	21
Age, mean (min-max) in years	42 (26-74)	48 (28-62)
Education, n[1]		
Low	1	2
Middle	15	7
High	19	15
Disease, n		
RA (Rheumatoid Arthritis)	13	10
SpA (Spondyloarthritis)	5	2
OA (Osteoarthritis)	3	-
FM (Fibromyalgia)	4	3
PsA (Psoriatic arthritis)	4	3
Sjögren's syndrome	-	2
Systemic Sclerosis	1	-
Palindromic rheumatism	1	1
Comorbid diagnoses[2]	4	3
Mean duration of the disease (min, max) in years	10 (0- 48)	16 (1 - 48)
Under treatment by a rheumatologist, n	29	21
Once participated in a self-management training, n	6	7

[1] Low=primary School or lower vocational secondary education, intermediate general secondary education; Middle=intermediate vocational education, higher general secondary education; High=higher vocational education or university education.

[2] PsA in combination with FM and OA, FM in combination with Tietze's syndrome and hypermobility syndrome, SpA in combination with OA and RA.

3.2. Results focus group (step 1 and 2)

The research team derived 146 topics from the focus group (preferences for structure and content needs).

3.2.1. Preferences for structure

The 43 topics relating to the preferred structure of the program were categorized into six components. These components and some examples of quotes are outlined in textbox 1.

Textbox 1 Components of the online training: preferences (in bold) and example quotes regarding the structure

SUITED TO INDIVIDUAL NEEDS

"… I think it is important to choose something that suits your life and your needs…that varies per person, per phase. One person might only want to learn more about having arthritis while others want to share their life experiences… (female, 26 years old)

"…I want to work on issues that matter most in my life at the moment… children, pregnancy, being a mother in combination with having a rheumatic disease…" (female, 30 years old)

"… I know I made the right decision (not having children)…but I am still sad about it…but we are now living our own life, my husband and me…slowly… but we are not receiving any help…how can I ask for help?…(female, 38 years old)

SUITED TO LIFE STAGE

"…I think my problems differ from older people. I was just looking for people who work, for people with young children and how they manage their lives while having a rheumatic disease. I also wonder how I could combine work and home…. (female, 37 years old)

"…Some themes are not always, at all times, relevant. Especially for the 'younger' generation… also, it depends on the stage where you are with regard to your illness…(male, 52 years old)

SHARE EXPERIENCES AND HAVE CONTACT WITH OTHER PEOPLE WHO HAVE A RHEUMATIC DISEASE

…"I would very much like to hear experiences of others….Maybe I still have something to learn, to adapt better to the situation…(female, 43 years old)

"…Being ill asks for adjustment…having a chronic disease asks for change. Healthcare providers give information and advice. To apply this advice to your own life is something different…by working together (with other patients) and sharing successes, I experience that you can make that change….It can be refreshing… (female, 47 years old)

EXPERT PATIENT AS TRAINER

"…I think having a rheumatic disease is not a special quality, being empathic is….(female, 42 years old)

"…An expert patient who dares to share his experiences…is more a facilitator than a leader. He or she does not pretend that he knows everything…he helps you to work towards your own goals (female, 47 years old)

OWN PACE

"…I want to work at my own pace…behavioral change asks for time and practice…" (female, 42 year old)

LIMITED IN TIME

"…Well, I think it's important to set a time limit (to actually end the training)… to keep me motivated…" (male, 60 years old)

3.2.2. Needs regarding contents

The 103 topics that were identified as needs for the online program were reduced during the consensus meetings to 55. These final needs are shown in the first column of table 2.

3.3. Results card sorting task (step 3)

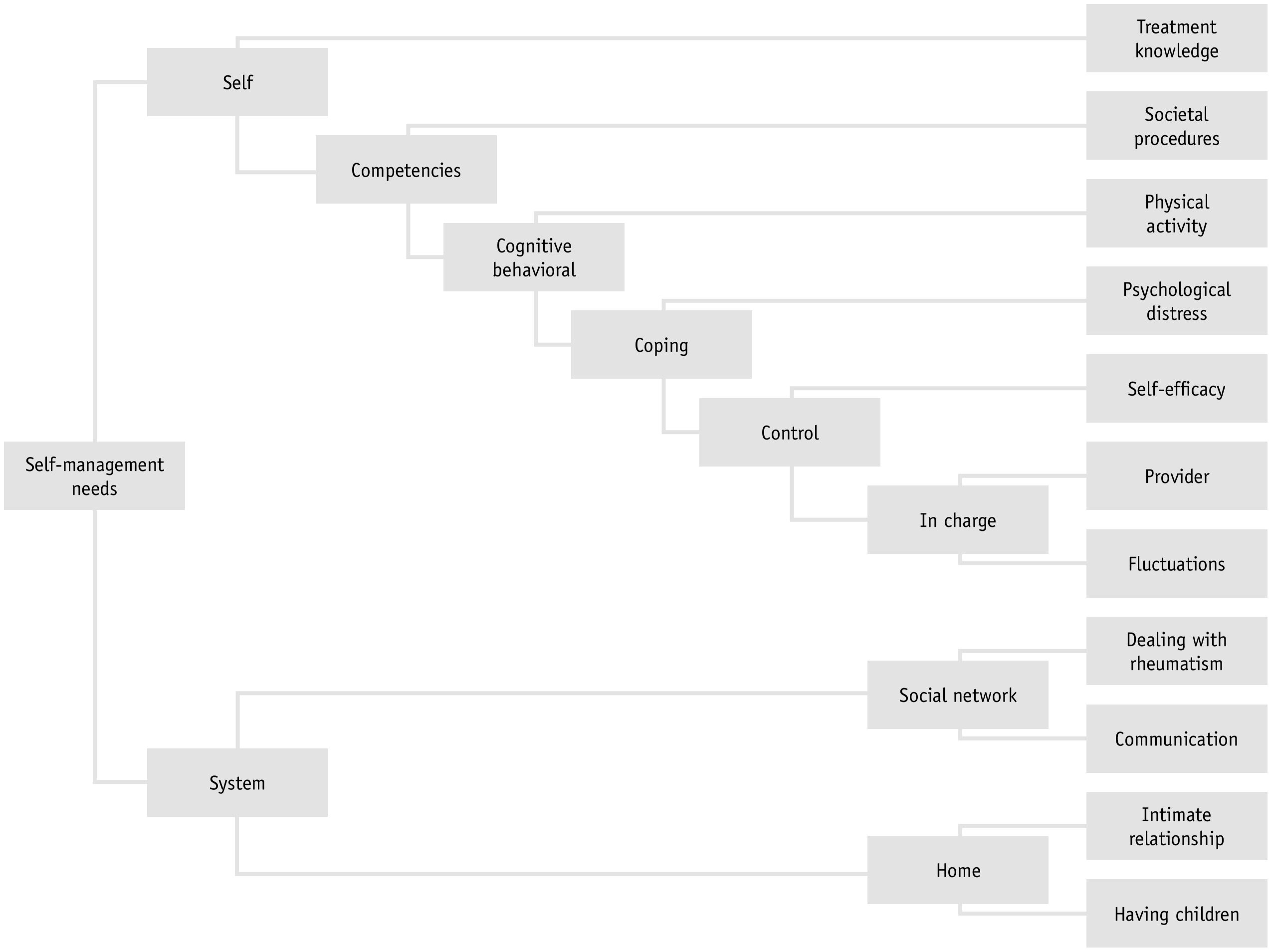
Participants individually sorted the 55 cards with needs regarding content into piles. The mean number of piles was 8 (range 4-12). The number of cards per pile varied from 1 to 22. Individual participants used 56 labels to describe the piles. Frequently chosen labels were: *knowledge about the disease, treatment, medication, psychological problems, sharing experiences, emotions, work, relationships, pregnancy and children, keeping control, self-control, physical consequences, leisure time, sports, exercise* and *standing up for yourself.* These multiple labels were used by the team to interpret the hierarchical cluster analysis and to choose the final labels of the clusters.

3.4. Results hierarchical cluster analysis (step 4)

Based on the dendrogram that was produced by the hierarchical cluster analysis, the number of clusters was set to eleven. A schematic drawing of the dendrogram is shown in figure 1. Table 2 shows which needs are included in the eleven clusters on the right of figure 1. Increasing the number of clusters from eleven to twelve or thirteen divided the clusters 'psychological distress' and 'having children' into new clusters referring to professional help versus learning from peers. As these deviations referred to the process instead of content, we decided to limit the clusters to eleven. Decreasing the eleven clusters to ten combined the clusters 'dealing with rheumatism' and 'communication', which were clearly distinct needs. Thus, the final number of clusters was set to eleven.

Figure 1 shows that in the hierarchical structure of clusters, at the highest level, 'self-management needs' were divided into the categories 'self' and 'system'. 'Self' included both 'knowledge' about the treatment and 'competencies' to self-manage the consequences of the disease. 'System' divided into the broader 'social network' and 'home' including needs relating to the immediate (family) environment.

Figure 1 Schematic drawing of the dendrogram showing the hierarchical structure with needs regarding the content of an online self-management support program



3.4.1. Importance of the clusters

The importance attached to needs is shown in the right column of Table 2. The mean importance rating per cluster varied from 1.9 (*sd* 1.6) for the cluster 'having children' to 4.1 (*sd* 0.5) for the cluster 'self-efficacy'(table 2).

Table 2 The eleven clusters of needs regarding content for an online self-management program with the mean (standard deviation) importance rating of each cluster and statement. All statements started with "I have a need for..."

Cluster Treatment knowledge	**2.8 (0.9)**
16. Knowledge about pain and fatigue caused by the rheumatic disease	3.8 (1.3)
20. Understanding why you have to take the prescribed medication	3.3 (1.4)
41. Understanding how the arthritis medication works	2.9 (1.5)
43. Knowing how best to maintain adherence to the medication	3.0 (1.5)
31. Knowledge of all possible treatments for arthritis	3.0 (1.5)
28. Information about the use of alternative medicine (for example homeopathy and acupuncture) in arthritis	2.2 (1.2)
44. More information in order to judge whether an alternative method is safe and appropriate.	2.0 (1.0)
42. Information about how to get extra help and treatment (for example home care, physiotherapy, psychologist)	2.7 (1.3)
Cluster Societal procedures	**3.0 (0.6)**
52. Knowledge of the laws and regulations that pertain to having a rheumatic disease	2.7 (1.5)
10. Learning how to deal with lack of understanding on the part of social service organizations	3.0 (1.5)
49. Information on accommodations that would make work easier	3.3 (1.2)
50. Information on how to function at work with a rheumatic disease	3.2 (1.2)
37. Hearing how others manage work and chores	3.2 (1.4)
35. Learning how to tell others about your disease at work or school	2.9 (1.2)
27. Finding out how to acquire enough income despite the limitations of arthritis	2.6 (1.4)
Cluster Physical activity	**3.1 (0.9)**
5. Discovering which physical activities or sports are suitable for me	3.5 (1.2)
39. Discovering how much physical activity is healthy with arthritis	3.2 (1.3)
23. Discovering how to keep up physical activities or sports	3.1 (1.1)
36. Information about sports that are possible for people with a rheumatic disease	2.8 (1.3)
Cluster Psychological distress	**3 (0.7)**
33. Learning how to deal with anger, embitterment, irritation and frustration	3.6 (0.9)
54. Learning how to deal with helplessness and sadness	3.5 (1.3)
38. Professional help with psychological or emotional problems caused by the rheumatic disease	2.9 (1.5)
34. Testing to evaluate how serious my psychological problems are	2.2 (1.5)
9. Hearing how others deal with (negative) emotions	2.9 (1.5)
Cluster Self-efficacy	**4.1 (0.5)**
7. Discovering how to remain in control of my life	4.8 (0.5)

30. Discovering how to set boundaries	4.4 (1.1)
18. Learning to set attainable goals	4.2 (1.1)
53. Learning to give direction to your life with arthritis	4.1 (1.3)
19. Learning to say “no” without feeling guilty	4.0 (1.1)
47. Learning how to ask friends, acquaintances and neighbors for help	2.9 (1.0)
Cluster Provider	**3.0 (1.2)**
51. Learning how to best talk to your medical professional	3.2 (1.4)
40. Learning how to express dissatisfaction with the treatment	2.9 (1.3)
Cluster Fluctuations	**3.5 (1.0)**
46. Learning how to control your arthritis or symptoms yourself	3.7 (1.3)
48. Learning how to fit the treatment into daily life	3.3 (1.4)
13. Learning what to do if the arthritis or the symptoms change	3.3 (1.3)
Cluster Dealing with a rheumatic disease	**3.5 (0.8)**
25. Hearing how others keep up their energy all day	3.6 (1.0)
55. Hearing from others how they function with arthritis in daily life	3.6 (1,2)
29. Hearing how others deal with pain and fatigue	3.6 (1.2)
26. Hearing from others how they deal with arthritis in daily life	3.4 (1.3)
24. Hearing from others how they have learned to accept the consequences of a rheumatic disease	3.3 (1.3)
Cluster Communication	**3.0 (0.9)**
2. Learning how to explain to others what it means to have arthritis	3.3 (1.4)
3. Learning how to deal with a lack of understanding on the part of friends, relatives and neighbors.	3.5 (1.2)
4. Learning from others how to maintain a social life while taking account of your arthritis	2.9 (1.3)
17. Hearing from others how they explain what rheumatic disease is	2.4 (1.2)
Cluster Intimate relationships	**2.4 (0.7)**
22. Information about the influence of arthritis on sexuality and intimacy	2.3 (1.1)
32. Information on having sexual relations without pain or discomfort	2.1 (1.1)
11. Getting tips from others on how they deal with sexuality, intimacy and arthritis	2.6 (1.4)
45. Discussing with other patients the influence of arthritis on the partner relationship	3.1 (1.2)
14. Hearing from others what they do with their leisure time and holidays, taking account of their arthritis	2.0 (1.1)
Cluster Having children	**1.9 (1.6)**
1. Expert advice about heredity and rheumatic diseases (for example when hoping to become pregnant)	1.9 (1.3)
15. Information about medication before, during and after a pregnancy	1.8 (1.1)
8. Discussing with others how best to talk to your children about your rheumatic disease	2.1 (1.2)
12. Finding out how to get help with child care	2.0 (1.0)
21. Hearing from others how they involve their partner in child care	2.0 (1.1)
6. Hearing from fellow patients how they decided whether or not to have children	1.8 (1.4)

4. DISCUSSION AND CONCLUSION

4.1. Discussion

This study yielded an extensive overview of the individual preferences regarding structure and content needs of an online self-management program to support patients with a rheumatic disease: six preferences and eleven needs were identified.

Concerning the *structure* of the training, patients emphasized that account should be taken of individual needs and the stages of their disease and life, and that they should be able to follow the program at their own pace and within a limited time period. This is in line with previous studies in which people with arthritis expressed the need for taking individual experiences and an individualized approach as points of departure (6,9,14,15,17) as well as with the ongoing emphasis on customizing training and exercises to the individual (2,7). As a positive example of customized treatment, participants reported 'how arthritis affected their lives' after which the knowledge was used to direct the participants to different parts of a 'Toolkit' of self-management interventions (24). However, although many self-management programs emphasize individual goals of the participant at the start of the program (1, 2, 25) the structure and content are mostly predetermined, protocolled, and based on group preferences (2, 25, 26). Our study emphasized the importance of customizing both structure and content of the program to personal preferences and needs.

The participants in our study also expressed a preference for being guided through the program by a trained patient instead of a professional tutor such as a nurse, psychologist, or social worker, in agreement with the Arthritis Self-Management Program (26) and Dutch Self-management Program for young adults with a rheumatic disease (27). Programs with a patient ''expert" as trainer were observed to be effective (2, 26, 28). This suggests that this wish of patients can be granted, although -to be certain that both are equally efficacious - programs with an expert-patient trainer and a professional trainer should be compared.

Although during recent decades the treatment of rheumatic diseases has improved considerably and psychological distress and physical disability have decreased (18), patients still express the need to have knowledge about their disease, physical activity, managing emotions, and self-regulating their own life, family, work and leisure time. Moreover, having a rheumatic disease affects the patient's family, friends, and coworkers (15, 18,29, 30). Our respondents expressed the need to include relational needs in the self-management support program by working on concrete issues such as asking for help or support from husband or coworkers, communicating with experts, and dealing with pregnancy or intimacy.

Needs within clusters differentiated between getting help from professionals versus peers, in agreement with previous studies (14, 26). Also in a study on self-management support from the perspective of patients with a chronic condition (31), the need for support of different sources (professionals and peers), each with its unique contribution, is recognized. This combination of support from professionals and peers reflects that besides increase of competencies (with help from professionals and peers), social relatedness (with peers) is also an important aspect of behavioral change as is emphasized by self-determination theory (10).

Considering *importance* of the content-needs, the clusters 'self-efficacy', (being in charge of) 'fluctuations' and 'dealing with rheumatic disease' (in the social network) are rated high on average, but the mean importance ratings of most clusters is rather similar which indicates that individuals rate the importance of needs differently. A cluster of needs that is rated less important on average by our sample, can for an individual patient be of primary importance. A strength of the overview of needs in our study is that its comprehensiveness makes it suitable to also quickly screen for needs that are considered important by only some individual patients.

Strengths of this study are that experiences and perspectives of patients were used as input materials and that the project team consisted of patient representatives and professionals that took decisions based on consensus and collaborated during all phases of the study. Moreover, the method that was used allowed a description beyond the subjective interpretation of the researchers, because the patients categorized the needs in meaningful constructs. A limitation of the current study could be that input was especially provided by patients who were interested in contributing to the development of an online self-management intervention. Having a volunteer group, can have resulted in information bias. Moreover, only a few men participated. However, this selection bias that is inherent to a convenience sample might also give a good reflection of the actual participants. In an intervention that was built using the insights of the current study also volunteers are invited to participate (32). Another limitation is that the wording of the needs may have influenced the sorting by patients. Words in the phrasing of needs such as 'knowledge', 'hearing' or 'learning' were sometimes sorted in the same cluster by patients. However, while some patients used these kinds of process variables to sort needs, others used the contents of needs. With respect to sample size, although 10 to 20 people are suggested to be a suitable number for card sorting (19), we noticed that the structure of the hierarchical cluster solution slightly changed by reanalyzing the data while excluding few participants; this was especially true for the higher order structure. Therefore, we focused our results and discussion mainly on the eleven lowest order clusters. Finally, the participation of three participants in both the interviews and the card-sorting is not considered a great problem because these two parts of the study appealed to distinct areas of knowledge of participants.

4.2. Conclusion

A comprehensive assessment of preferences and needs in patients with a rheumatic disease is expected to contribute to motivation, adherence to and outcome of self-management-support programs, which is an area of future research. The current overview of preferences and needs can be used to build a self-management support program, to help in identifying needs at the beginning of the program, and to customize the program to the individual patient.

REFERENCES

1. **Barlow J, Wright C, Sheasby J, et al.** Self-management approaches for people with chronic conditions: a review. Patient Educ Couns 2002;48:177-87.
2. **Iversen MD, Hammond A, Betteridge N.** Self-management of rheumatic diseases: state of the art and future perspectives. Ann Rheum Dis 2010;69:955-63 doi:10.1136/ard.2010.129270.
3. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev 1977;84:191-215.
4. **Lorig KR, Ritter PL, Laurent DD, et al.** The internet-based arthritis self-management program: a one-year randomized trial for patients with arthritis or fibromyalgia. Arthritis Rheum 2008;59:1009-17 doi:10.1002/art.23817.
5. **Shigaki CL, Smarr KL, Siva C, et al.** RAHelp: an online intervention for individuals with rheumatoid arthritis. Arthritis Care Res (Hoboken) 2013;65:1573-81 doi: 10.1002/acr.22042.
6. **Trappenburg J, Jonkman N, Jaarsma T, et al.** Self-management: one size does not fit all. Patient Educ Couns 2013;92:134-7 doi:10.1016/j.pec.2013.02.009.
7. **Zangi HA, Ndosi M, Adams J, et al.** EULAR recommendations for patient education for people with inflammatory arthritis. Ann Rheum Dis 2015;74:954-62 doi:10.1136/annrheumdis-2014-206807.
8. **Kralik D, Koch T, Price K, et al.** Chronic illness self-management: taking action to create order. J Clin Nurs 2004;13:259-67.
9. **Kristiansen TM, Primdahl J, Antoft R, et al.** Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: a focus group study. Musculoskeletal Care 2012;10:29-38 doi:10.1002/msc.224.
10. **Ryan RM, Deci EL.** Self-determination theory and the facilitation of intrinsic motivation, social development, and well-being. Am Psychol 2000;55:68-78.
11. **Knittle K, De Gucht V, Hurkmans E, et al.** Targeting motivation and self-regulation to increase physical activity among patients with rheumatoid arthritis: a randomised controlled trial. Clin Rheumatol 2015;34:231-8 doi:10.1007/s10067-013-2425-x.
12. **Newman S, Steed L, Mulligan K.** Self-management interventions for chronic illness. Lancet 2004; 364:1523-37 doi: 10.1016/S0140-6736(04)17277-2.

13. **van Eijk-Hustings Y, Ammerlaan J, Voorneveld-Nieuwenhuis H, et al.** Patients' needs and expectations with regard to rheumatology nursing care: results of multicentre focus group interviews. Ann Rheum Dis 2013;72:831-5 doi:10.1136/annrheumdis-2012-202810.

14. **Ryan S, Lillie K, Thwaites C, et al.** 'What I want clinicians to know'--experiences of people with arthritis. Br J Nurs 2013;22:808-12 doi:10.12968/bjon.2013.22.14.808.

15. **Zuidema RM, Repping-Wuts H, Evers AW, et al.** What do we know about rheumatoid arthritis patients' support needs for self-management? A scoping review. Int J Nurs Stud 2015 doi:S0020-7489(15)00185-6.

16. **van Houtum L, Rijken M, Heijmans M, et al.** Patient-perceived self-management tasks and support needs of people with chronic illness: generic or disease specific?. Ann Behav Med 2015;49:221-9 doi:10.1007/s12160-014-9649-0.

17. **Hamnes B, Hauge MI, Kjeken I, et al.** 'I have come here to learn how to cope with my illness, not to be cured': a qualitative study of patient expectations prior to a one-week self-management programme. Musculoskeletal Care 2011;9:200-10 doi:10.1002/msc.212.

18. **Dures E, Almeida C, Caesley J, et al.** Patient preferences for psychological support in inflammatory arthritis: a multicentre survey. Ann Rheum Dis 2014 doi: annrheumdis-2014-205636.

19. **Trochim W.** An introduction to concept mapping for planning and evaluation. Evaluation and Program Planning 1989;12:1-16.

20. **Holloway I, Wheeler S.** Qualitative Research in Nursing. Oxford: Blackwell Publishing 2002.

21. **Wood JR, Wood LE.** Card Sorting: Current Practices and Beyond. J Usability Studies 2008;4:1-6.

22. **WMA Declaration of Helsinki - Ethical Principles for Medical Research Involving Human Subjects.** Available at: http://www.wma.net/en/30publications/10policies/b3/

23. **Clatworthy J, Buick D, Hankins M, et al.** The use and reporting of cluster analysis in health psychology: a review. Br J Psychol 2005;10:329-358.

24. **Goeppinger J, Lorig KR, Ritter PL, et al.** Mail-delivered arthritis self-management tool kit: a randomized trial and longitudinal followup. Arthritis Rheum 2009;61:867-75 doi:10.1002/art.24587.

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

26. **Marks R, Allegrante JP, Lorig K.** A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part II). Health Promot Pract 2005;6:148-56 doi:10.1177/1524839904266792.

27. **Ammerlaan J, van Os-Medendorp H, Scholtus L, et al.** Feasibility of an online and a face-to-face version of a self-management program for young adults with a rheumatic disease: experiences of young adults and peer leaders. Pediatr Rheumatol Online J 2014;12:10,0096-12-10 doi:10.1186/1546-0096-12-10.

28. **Chodosh J, Morton SC, Mojica W, et al.** Meta-analysis: chronic disease self-management programs for older adults. Ann Intern Med 2005;143:427-38 doi:143/6/427.

29. **Overman CL, Jurgens MS, Bossema ER, et al.** Change of psychological distress and physical disability in patients with rheumatoid arthritis over the last two decades. Arthritis Care Res (Hoboken) 2014;66:671-8 doi:10.1002/acr.22211.

30. **Wagner EH, Austin BT, Von Korff M.** Organizing care for patients with chronic illness. Milbank Q 1996;74:511-44.

31. **Dwarswaard J, Bakker EJ, van Staa A, et al.** Self-management support from the perspective of patients with a chronic condition: a thematic synthesis of qualitative studies.Health Expect 2016;19:194-208 doi: 10.1111/hex.12346.

32. **Ammerlaan JW, Mulder OK, de Boer-Nijhof NC, et al.** Building a Tailored, Patient-Guided, Web-Based Self-Management Intervention ‘ReumaUitgedaagd!’ for Adults With a Rheumatic Disease: Results of a Usability Study and Design for a Randomized Control Trail. JMIR Res Protoc 2016;5:e113 doi:10.2196/resprot.5735.

“To me, self-management is mainly about setting your own boundaries”

- Female, 41 years old, since 10 years Rheumatoid Arthritis

CHAPTER 8

BUILDING A TAILORED, PATIENT-GUIDED, WEB-BASED SELF-MANAGEMENT INTERVENTION 'REUMAUITGEDAAGD!' FOR ADULTS WITH A RHEUMATIC DISEASE: RESULTS OF A USABILITY STUDY AND DESIGN FOR A RANDOMIZED CONTROLLED TRIAL

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ABSTRACT

Background The chronic nature of rheumatic diseases imposes daily challenges upon those affected and causes patients to make daily decisions about the way they self-manage their illness. Although there is attention to self-management and evidence for the desirability of tailored interventions to support people with a rheumatic disease, interventions based on individual needs and preferences are scarce.

Objective To provide a systematic and comprehensive description of the theoretical considerations for building a Web-based, expert patient-guided, and tailored intervention for adult patients with a rheumatic disease. Also, to present the results of a usability study on the feasibility of this intervention, and its study design in order to measure the effectiveness.

Methods To fit the intervention closely to the autonomy, needs, and preferences of the individual patient, a research team comprising patient representatives, health professionals, Web technicians, and communication experts was formed. The research team followed the new guidance by the Medical Research Council (MRC) for developing and evaluating complex interventions as a guide for the design of the intervention.

Results Considerations from self-determination theory and a comprehensive assessment of preferences and needs in patients with a rheumatic disease guided the development of the Web-based intervention. The usability study showed that the intervention was useful, easy to use, and accepted and appreciated by the target group of patients. The planned randomized controlled trial is designed to be conducted among 120 adults with a rheumatic disease, who are assigned to the self-management intervention or a self-help control group. Both groups will be asked to formulate personal goals they want to achieve concerning their self-management. Progress toward the personal goal is the primary outcome measure of this study. Self-reported Web-based measures will be assessed before randomization at baseline, and 3 and 6 months after randomization. Also, feasibility and adherence to the Web-based self-management intervention as process outcomes will be evaluated.

Conclusion By identifying the individual goals at the beginning of the intervention and customizing the intervention to the individual patient, we aim to improve the usefulness and effectiveness of the Web-based self-management intervention. If proven effective, ReumaUitgedaagd! online will be implemented in the Netherlands.

INTRODUCTION

Background

Having a rheumatic disease often leads to symptoms of pain, fatigue, and physical constraints that are part of a reduced health-related quality of life (1). The chronic nature of this disease imposes daily challenges upon those affected and causes patients to make daily decisions about the way they manage their illness (1-3). The question is not “whether” patients self-manage their (chronic) illness, but “how” they do this (4). The ‘(individual’s) ability to manage the symptoms, treatment, physical, and psychosocial consequences and life style changes inherent in living with a chronic condition’ has been defined as self-management (5). Interventions to improve self-management commonly combine information-based and cognitive-behavioral strategies (6). In the last decade, several interventions have been developed to improve self-management. For people with a rheumatic disease, the Arthritis Self-Management intervention (ASMP) of Stanford University is the most recognized and studied self-management intervention (7). The ASMP intervention, based on the self-efficacy theory of Bandura (8) is led by expert patients and is designed to help people with arthritis gain confidence in their ability to control their symptoms and the impact of their condition on their lives (7,9,10). For patients with rheumatoid arthritis and osteoarthritis, participating in an ASMP led to improved health behavior (cognitive symptom management, communication with physicians, dietary habit, exercises, and relaxation) and a decrease of depression. However, decreases in fatigue and anxiety were found not to be significant (6,7).

With the growing opportunities and use of the Internet, a Web-based self-management version of the ASMP intervention for patients with long-term conditions was developed in 2007 (11). Evaluation of the effectiveness of this intervention after 12 months showed significant improvements on health status measures like distress, pain, and self-efficacy. In 2011, based on the ASMP intervention and the self-efficacy theory of Bandura and in cooperation with the Dutch Arthritis Foundation and young adults from the transition outpatient clinic of University Medical Center Utrecht, we developed a Web-based self-management intervention for young adults up to the age of 25 years (12). The aim of that intervention was to enhance young adults’ self-management in coping with their rheumatic disease.

With the expansion of the Web-based intervention in the Netherlands, older adults with rheumatic diseases also expressed their need for a Web-based self-management intervention. In order to meet this need, the Dutch Arthritis Foundation gave us a grant to develop a Web-based intervention for adults from the age of 25 years and older. The goal of this research protocol was to describe 1) the theoretical considerations that guided the development of this Web-based intervention for adult patients with rheumatic diseases, 2) the contents of the intervention, 3) the results of a pilot study to study the usability of the intervention, and 4) the study design in order to examine the effectiveness of the intervention.

Toward a Patient-Guided Intervention

As we inferred from our experiences with the development and research pertaining to the Web-based intervention for the young adult group, collaboration with the end-users in all phases of development of a Web-based self-management intervention is crucial and influences the actual use, adherence, and effectiveness (13). Based on this notion and to fit the intervention closely to the autonomy, needs, and preferences of the patients, we formed a research team consisting of patient representatives of different ages, health professionals, Web technicians, and communication experts. The research team followed the new guidance for developing and evaluating complex interventions by the Medical Research Council (MRC) (14) as a guide for the design of the intervention. The guidance included the following phases: *development*, *feasibility and piloting*, and *evaluation* and *implementation*. During the development phase, the aims were to determine a theoretical foundation and to develop the structure and content of the first draft. To achieve these aims, we first screened the scientific literature for theoretical considerations and effectiveness of (Web-based) self-management interventions for people with a chronic or rheumatic disease. The search for qualitative and quantitative articles was conducted in Medline, the Cumulative Index to Nursing and Allied Health Literature, Web of science, PsycINFO, and Pubmed. We searched for studies published in English, which used the words: "self-management," "chronic disease," "rheumatic disease," "adults," "theoretical foundations/considerations," and "effectiveness" in different combinations. No publication year limit was used. Secondly, a focus group and concept mapping study was performed to assess preferences and needs of adult patients with a rheumatic disease regarding the structure and content of the future Web-based self-management intervention.

Theoretical Considerations

Although there is growing attention for interventions that are customized to individual patients with chronic diseases, the structure and contents are generally still protocol-based on group preferences (6,15). And, disappointingly, to date there is no consistent (long-term) evidence of the efficacy of self-management interventions for patients with a chronic disease in general (6,16). This might be due to various reasons, including diversity of interventions, insufficient theoretical foundation, and the heterogeneity of the patient populations (16). Moreover, positive mean group outcomes may disguise that a substantial proportion of patients did not comply with or respond to the intervention (6,16). A basic assumption about self-management is that when the intervention is customized to the individual needs and situation of the patient, the patient will be more motivated, adhere better, and benefit more and for a longer time (6,15,16). Thereby, a change in behavior and long-term adherence to changed behavior is expected to be greater when a patient experiences a meaningful rationale for change, values the change in behavior positively, and aligns it with other central values and lifestyle patterns (2,3,17). Consistent with these assumptions is Self-determination Theory of Ryan and Deci (17), which emphasizes the importance of keeping goals of behavior change (like improvement of self-management) close to the autonomous motivation of people. In this theory, three basic needs determine motivation: *autonomy*, *competence*, and *social relatedness* (17). Among these three, autonomy is considered as the most central need: if a behavior is autonomous, it is voluntary, originating from one's own values and self-determination. Competence refers to the necessity to experience that one is really able to achieve something, and is related to the construct of self-efficacy (10). The third basic need, social relatedness, is the extent to which one finds support in one's environment, including support from a trainer or professional. High levels of autonomy, competence, and social relatedness enhance self-regulation.

Needs Assessment

An important part of the development phase consisted of a needs assessment, conducted by the combination of a focus group and concept mapping design (J. W. Ammerlaan, et al., unpublished data, 2016). Online focus group interviews among adult patients with rheumatic diseases in the Netherlands, a card sorting task, and hierarchical cluster analysis yielded an extensive overview of the individual preferences regarding structure and content. Patients preferred an intervention tailored to their needs, stage of life, and goals. Also, an expert patient as a trainer, the opportunity to be in contact and to share with others, and the ability to follow the intervention at one's own pace were preferred. With respect to needs for content of the intervention, hierarchical cluster analysis yielded 11 clusters involving increasing individual knowledge of treatment and consequences for daily life, skills including managing emotions, managing, the fluctuations of disease, and dealing with health professionals and social authorities. Self-regulating their own lives, including requesting support from their spouse, family, or coworkers, setting boundaries and the ability to communicate adequately, and dealing with pregnancy or intimacy issues and taking care of kids. Based on the data from this needs assessment and the theoretical considerations, the first draft of the Web-based self-management intervention (in Dutch: ReumaUitgedaagd! online) was developed.

METHODS

Design of the Web-Based Intervention

ReumaUitgedaagd! Online is a Web-based, password protected, tailored, self- management intervention for adults with a rheumatic disease, aimed at enhancing patients' self-management skills. The participants perform the intervention individually, are coached by a trainer, and have online contact with other participants on a discussion board. The role of the trainer is to support participants during the Web-based intervention in becoming a good self-manager and achieving their personal goals. The trainers are adults who also have a rheumatic disease. They are recruited through the website of the Dutch Arthritis Foundation and selected through assessments and interviews conducted by a professional coaching organization (Work21), in close cooperation with the Dutch Arthritis Foundation and the University Medical Center Utrecht. The selection process used questions about motivation, perceptions of self-management, the self-determination theory and strategies derived from the theoretical foundation, and goals of the Web-based intervention to identify those trainers who could adhere to the basic tenets of the intervention. Finally, the expert trainer was trained through a 3-day train-the-trainer (TTT) educational intervention. The TTT intervention consisted of following the intervention as a participant, knowledge of different themes, and teaching Web-based training skills. The trainers are given a volunteer contract and receive a stipend from the Dutch Arthritis Foundation.

The basic needs of autonomy, competence, and social relatedness, derived from Self-Determination theory, are embedded in the intervention and combined with elements of skills training and modelling, based on the Self-efficacy theory (8). Autonomy is taken into account by customizing the intervention to three individual needs and goals, which the participants choose at the beginning of the intervention. The participants choose thematic modules based on these individual needs and goals. For example: a woman who wants to learn more about coping with the consequences of her disease at work, may choose the 'Work' module, while a man who wants to increase his physical fitness may choose the 'Exercises' module. Competence is increased by making action plans, reflecting on one's own behavior by performing exercises, or sharing the output of exercises on the discussion board to receive feedback or support from other participants and the expert trainer. Social relatedness is achieved through the support of the expert trainer via individual chats and the message box, and also by sharing experiences and giving feedback and support with other participants.

Content of the Web-Based Intervention

The Web-based self-management intervention consists of four components: nine thematic modules (willing, knowing, skills, feeling, living together, influence, exercise, work, and moving on), a chat application, a discussion board, and a message box.

Figure 1 Screen of the homepage, showing the nine modules of the online ReumaUitgedaagd! (in Dutch)

Modules

Each module involves a specific theme. Both informative text about the theme and exercises are included. The information and the exercises are supported by short videos in which people with a rheumatic disease or a member of the multidisciplinary team tell about their experiences with arthritis. The content of the modules is described in Table 1. The participant performs the intervention individually and has 2 months to complete it. The total time investment for the participants is between 4 and 9 hours (approximately 30-60 minutes per module). The first three modules and the last module ('willing', 'knowing', 'skills', and 'moving on') are mandatory for all participants. The participant can choose other modules depending on his or hers personal goals. The nine modules are displayed in Figure 1.

Chat Sessions

The intervention includes three chat sessions between the participant with the trainer (after finishing module 1, after finishing module 3, and after finishing module 9). During the chat sessions, the trainer discusses the progress of the intervention and answers questions from participants. The duration of a chat session is approximately 15 to 30 minutes. The participant also has the ability to individually contact the trainer via a message box.

Discussion Board

The purpose of the discussion board is to exchange experiences between participants and trainers. In some exercises the participants put their output on the discussion board to start a discussion. For instance, they report how they tend to deal with being dispirited and whether or not they feel the strategy is proving to be successful.

'Look and Feel' of the intervention

Based on the preferences of the research team, the design of the Web-based intervention was made attractive by using 'colorful, real-life pictures of people of different ages' to support information and exercises. Secondly, pictures of people, performing activities based on the content of the module, were used as pictograms to navigate. Thirdly, the videos to support the informative text of the modules were directed and produced by a professional company. Finally, a voice over was added to assist visually impaired participants.

Table 1 Content of the Nine Modules of the Web-Based Self-Management Intervention and Exercises

MODULE	CONTENTS	EXERCISES
1. Willing (formulating personal goals)	▪ Self-management ▪ Priorities in life (getting to know yourself) ▪ Setting and achieving personal goals	▪ Awareness of self-management ▪ Evaluating self-management ▪ Life values (priorities in life) ▪ Formulating personal goals for the training
2. Knowing (disease-specific information and treatment)	▪ A rheumatic disease: what does that mean? ▪ Treatment possibilities ▪ Getting control over one's disease and treatment	▪ Knowledge Quiz: what do you (already) know of your disease? ▪ Gaining insight into treatment and treatment goals ▪ Working together with your physician and health professional ▪ Pain and fatigue diary
Additional	▪ Medication ▪ A consultation in the hospital: how do you prepare yourself?	▪ Practice and evaluation of consultation in the hospital
3. Skills (self-management skills)	▪ Being in charge: making choices ▪ Problem solving ▪ Communication ▪ To give and receive feedback ▪ Setting boundaries ▪ Coping: dealing with consequences	▪ Evaluating your own behavior: making choices ▪ Circle of influence and engagement ▪ Feedback in your daily life ▪ Saying no ▪ Recognizing your own coping scale
Additional		▪ Self-assertiveness test
4. Feeling (body, mind, and emotions)	▪ Having a rheumatic disease; what's next? ▪ Consequences of having a rheumatic disease on your body, your mind, and socially ▪ Pain, fatigue, and negative emotions ▪ Your own influence	▪ Loss of health; what does that mean to you? ▪ Feeling blue ▪ Evaluation of a situation to get insight into the influence of one's thoughts, behavior, and feelings ▪ Evaluation of the pain and fatigue diaries
Additional	▪ Processing phases in the loss of health ▪ Tips for handling pain ▪ Tips for handling fatigue ▪ To rack one's brain: what can one do about it?	▪ Dealing with the loss of health ▪ Relaxation exercises ▪ To puzzle over: what can you do?
5. Living together (family and spouses)	▪ Communicating with family and friends ▪ Kids and stuff	▪ Relationships ▪ Intimacy ▪ Sexuality ▪ Asking for help from your representatives or friends

Table 1 Content of the Nine Modules of the Web-Based Self-Management Intervention and Exercises

MODULE	CONTENTS	EXERCISES
Additional	▪ Getting pregnant and having kids ▪ Taking care of kids ▪ Communicating with one's children	▪ Communicating with your partner
6. Influence (interaction with your environment)	▪ How to influence one's environment? ▪ Dealing with lack of understanding (invalidation) ▪ Asking for help	▪ Explain your disease and consequences ▪ Asking for help: sharing experiences
7. Exercise (sport, exert, and relaxation)	▪ Exercise and having a rheumatic disease ▪ Motion and physical activity ▪ Pain and overload ▪ Exertion and relaxation	▪ Your exercises ▪ Exercise diary ▪ Action plan ▪ Relaxation
Additional	▪ Exercise and different rheumatic diseases	
8. Work (daily activities)	▪ Suitable work ▪ Dealing with invalidation at work ▪ Dealing with fatigue and stress at work ▪ Rights and obligations ▪ Going to school or university ▪ To apply for a job	▪ What's a suitable job for you? ▪ Who knows that you have a rheumatic disease at work? ▪ Dealing with obstacles
Additional	▪ Preparing for an interview with your colleagues or boss ▪ Being sick and getting back to work/school ▪ Work adaptions	▪ Preparing for an interview with your colleagues or boss
9. Moving on (evaluation and looking forward)	▪ Your personal goals ▪ Action plan for the future ▪ Evaluation	▪ Self-management: reflection of your own knowledge and skills ▪ Action plan for the future ▪ Evaluating your own goals
Additional	▪ An example of an action plan	

Usability Testing

Design

The first draft of the Web-based self-management intervention was tested in a quantitative pilot study, using the three concepts of the Technology Acceptance Model (TAM) (19): perceived usefulness, perceived ease of use, and intention to use. According to TAM, the usability of a particular technical innovation can best be predicted by an individual's intention to use or re-use the innovation. This intention is determined by two components: (1) perceived ease of use, which can be defined as "the degree of ease, associated with the use of the applications," and (2) perceived usefulness, which can be defined as "the degree to which an individual believes that using applications will help him to attain gains or to increase personal performance." (19).

The participants of the pilot study were given 3-weeks' access to the Web-based self-management intervention to examine and apply the contents of the intervention. After 3 weeks, the participants completed a Web-based questionnaire on usability (based on the TAM).

Population

Adult patients with access to a computer with Internet, sufficient Internet skills, diagnosed with a rheumatic disease, and being able to read and write in Dutch were included. Participants were recruited through websites, Facebook, and Twitter accounts of the Dutch Arthritis Foundation (20) and ReumaUitgedaagd! (21). All patients gave informed consent via the Internet.

Variables and Outcome Measures

Demographic variables like age and type of rheumatic disease and self-reported Internet-skills (measured on a 5-point Likert scale from very bad to very good) were collected to describe the group. Usability as primary outcome measure was operationalized using the three concepts of the TAM with 11 questions on a 5-point Likert scale (from totally disagree to totally agree) with the possibility to give additional comments. One question on 'overall satisfaction' was added, using a numeric rating system (NRS) from 0 (not satisfied) to 10 (most satisfied) (see Textbox 1).

Textbox 1 Questions to Measure the Three Concepts of the Technology Acceptance Model on Usability (All questions start with: "Now that you have seen the Web-based intervention…")

PERCEIVED USEFULNESS

- Did you perceive the content of the intervention to be useful?
- Did you perceive the content of the intervention as understandable?
- Did you perceive the exercises in the intervention to be useful?
- Did you perceive the content of the exercises as understandable?
- Did you perceive the intervention to be useful as a supplement to usual health care?
- Did you perceive the intervention to be useful in dealing with the consequences of having a rheumatic disease in daily life?

PERCEIVED EASE OF USE

- Did you perceive the Web-based self-management intervention to be easy to navigate?
- Could you easily find what you were looking for?

INTENTION TO USE

- Would you participate again, knowing now the content and structure?
- Would you recommend the Web-based self-management intervention to others (knowing now the content and structure)?

OVERALL SATISFACTION

- How do you rate your overall satisfaction with the intervention?
- How do you rate the look and feel of the intervention?

Results

Twenty-three respondents (22 women, mean age of 47 years) were given access to the Web-based intervention to test the usability. Most of them were diagnosed with inflammatory arthritis (16/23, 70%). Other diagnoses were osteoarthritis and fibromyalgia. Ninety-one percent (21/23) of participants rated their Internet-skills as 'very good'. Two participants rated their skills as average.

Ninety-one percent (21/23) of participants indicated the content and exercises as easy to understand and useful (ie, agree/totally agree on the Likert-scale). The majority of the participants (21/23, 91%) indicated the intervention to be useful in dealing with the consequences of having a rheumatic disease in daily life. The navigation on the site itself was rated somewhat lower with 70% (16/ 23) of participants being critical about the menu with thematic modules on the homepage and finding their way on the website. The look and feel of the intervention was recognized by 78% (18/23) of participants as pleasant.

In terms of intention to use: 78% (18/23) would participate in the Web-based intervention themselves and 91% (21/23) would recommend it to others. The mean satisfaction score of the Web-based intervention was rated 7.9 (range 4-10) on a scale of 0 (not satisfied) to 10 (most satisfied).

Conclusion

Considering the three concepts, we concluded that the Web-based intervention was to be recognized as being useful and easy to use. Participants stated that they were likely to participate; now they were familiar with the content and structure. To improve the navigation and menu of the intervention, numbers were added to each module in order to indicate the sequence of the modules**.**

Study Design in Order to Measure the Effectiveness of the Newly Developed Web-Based Intervention

Design

To evaluate the Web-based self-management intervention, we have planned a randomized controlled trial with an intervention and a self-help control group and a 6-month follow-up period among adults in the Netherlands having a rheumatic disease. The control group will be placed on a waiting list and will cross-over to the intervention after 6 months. Participants in the intervention group will be given access to the Web-based self-management intervention ReumaUitgedaagd! Both groups will receive usual care, based on the medical standard guidelines of the Dutch Association of Rheumatology (22), which also includes attention for self-management by the use of informational and educational materials that are normally used by patients to promote self-management. These materials are offered on the website of the Dutch Arthritis Foundation. Measuring the effectiveness means that we investigate whether there is an additional effect of the Web-based intervention in the intervention group on top of the care that is usually offered. The medical-ethical review board of the University Medical Center Utrecht in the Netherlands has approved the design and the procedures of this study.

Participants

Because we already have a Web-based self-management intervention for young adults (from 16-25 years), adults ≥26 years, having a rheumatic disease, diagnosed at least 2 years before inclusion by a rheumatologist or a General Practitioner, are eligible for this study. In addition to having an Internet connection, patients need to have proficiency in the Dutch language and not having previously participated in a self-management intervention. Having a psychiatric disorder or being under (recent) treatment by a psychologist or psychiatrist are criteria for exclusion from this study. The participants will be recruited via the Internet through websites, Facebook, and Twitter accounts of the Dutch Arthritis Foundation (20) and ReumaUitgedaagd! (21). After having signed consent forms, they receive information about the study and an information paper on goal setting with instruction and some examples of goals derived from the study on needs (J.W. Ammerlaan, et al, unpublished data, 2016). After 1 week, a telephone call will be set up between the researcher and the participant to check the inclusion criteria and to answer any questions about the study. The participants will also be asked to think about three individual goals concerning their self-management they want to achieve. One week later, a second phone call will take place between the researcher and the participant to set the final three individual goals and to inform the participant about the randomization procedure. To warrant objectivity and standardization as much as possible, standardized scripts will be used for the two contacts. The telephone calls are conducted by an independent interviewer (OM) who is not involved in the care of patients with a rheumatic disease.

Randomization

Randomization will take place after informed consent and completion of the goal-setting procedure, using a computerized application of the University Medical Center Utrecht. This is an automated process with no interference from the investigators. We will use a stratified block randomization to decrease the likelihood of imbalance between three conditions (arthritis, osteoarthritis, and soft-tissue rheumatism). After randomization, the participants will be informed by the researcher if they are assigned to the intervention or control group. The participants of the intervention group will then start with the Web-based self-management intervention and be asked to work through the intervention within 2 months.

Outcome Measurements

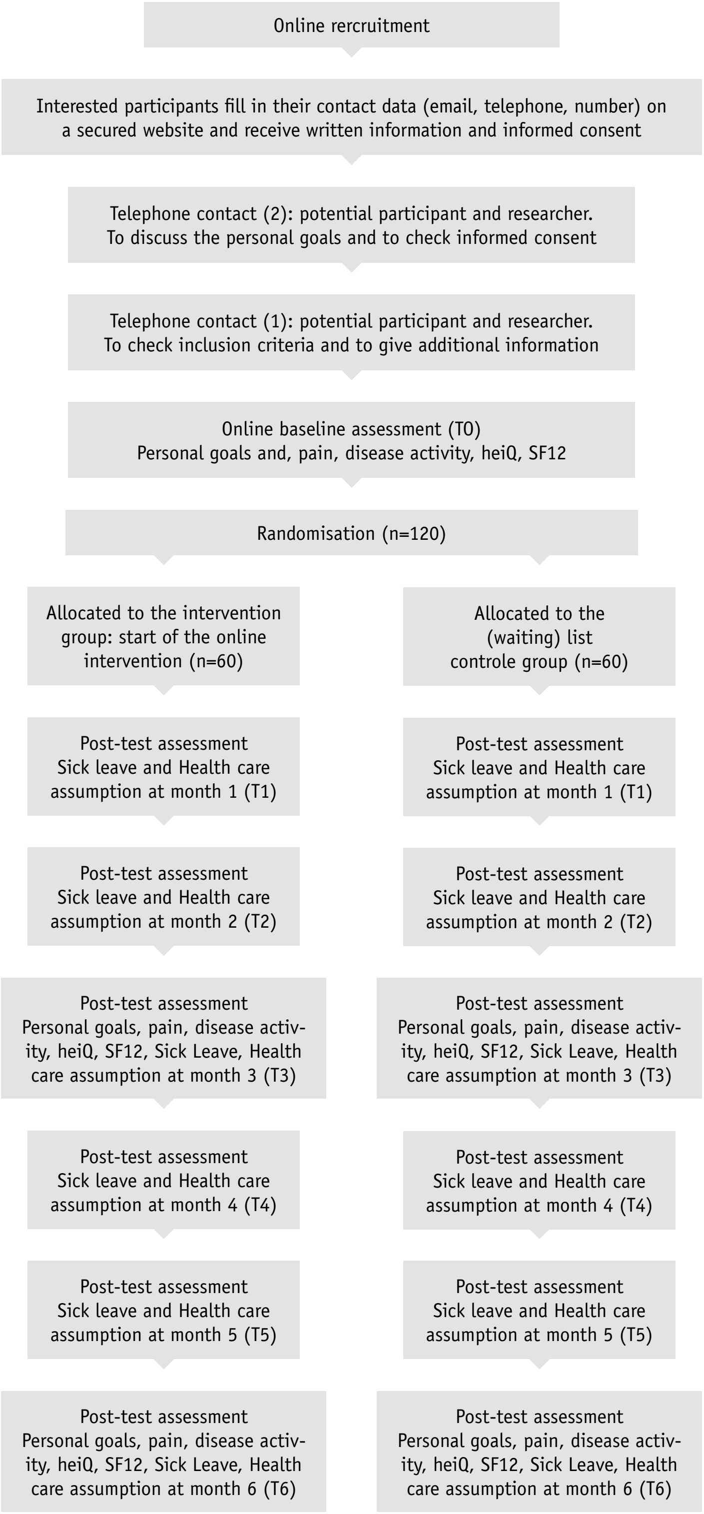
In this study individual outcome measures, generic outcome measures, and process outcomes measures will be collected, most of them via the Internet with questionnaires, self-reported by the participants. Demographic variables including age, sex, disease duration, diagnosis, marital status, current treatment, education level, work, and comorbidity at baseline will be collected in order to characterize the group of participants. The timeframe for collecting the outcome measures is displayed in Figure 2.

Individual Outcomes: Goal Accomplishment

A crucial and novel aspect of this study is that the intervention is customized to the needs of the participant. This is one of the reasons why the change –progress toward– the main personal goal is chosen as the primary individual outcome measure. Studies show that on individual outcome measures the effect is larger than on generic outcome measures (23-25), which is not unexpected because many of the patients are already on a functional level of generic outcome measures; even more so, given that patients with psychopathology are excluded. The three personal goals that the patient wants to accomplish are in a telephone interview with the researcher checked according to the following criteria: 1) the individual goals are aligned to the content and overall aim of the self-management intervention, 2) the goals focus on 'knowledge' and 'skills', and 3) the participant feels that each goal is achievable. In addition, the participant is asked to prioritize these goals (the first goal being the most important goal). Progress on the highest priority personal goal is the primary outcome of the study. Evaluations of the second and third personal goals are also conducted as (secondary) individual outcome measures.

The personal goals are measured with a Web-based NRS. The participant is asked to indicate with a score from 0 to 10 on the NRS to what extent he or she achieved this goal. The content of the primary goal can differ per person but the rate of change can be compared between subjects because they are measured on the same scale.

Figure 2 Time frame and flow-chard study design



Generic Outcomes

The following generic outcome measures are assessed (all self-reports): pain, disease activity, self-management skills, quality of life, and sick leave.

Self-reported pain and disease activity will be scored by the participant on a Web-based NRS from 0 to 10. The higher the score, the more pain or disease activity. Self-management will be measured with the Dutch translation of the Web-based Health Education Impact Questionnaire (heiQ) (26), which consists of 40 questions with scores ranging from 1 (not at all true) to 4 (exactly true) and are organized into a set of eight scales: health-directed behavior, positive and active engagement in life, emotional well-being, self-monitoring and insight, constructive attitudes and approaches, skills and technique acquisition, social integration and support, and health navigation. In a recent study of patients with a chronic disease, the heiQ scales showed good internal consistency, with Cronbach's alpha ranging from 0.70 to 0.89 on the eight independent scales, and high construct validity (27). Quality of life will be assessed with the Web-based Medical Outcomes Study 12-item Short Form Health Survey (SF-12) (28), which includes eight questions on functional status, three questions on general well-being, and one question on general health. The psychometric properties of the SF-12 are good (28). Sick leave is measured with three questions regarding 1) working in a paid job (yes/no/how many days a week), 2) sick leave during the past month, and 3) reasons for sick leave. Two measures of health care assumption are recorded as follows: self-reported visits to general practice, medical specialist, or physiotherapist, and whether or by whom support is offered to achieve the personal goals.

Process Outcomes

Feasibility is measured as a process outcome in the effectiveness study to evaluate the intervention in real-life in a larger group. Feasibility will be measured within the intervention group using the three concepts of the TAM (19,29) (see Textbox 1).

Use and adherence of the Web-based self-management intervention are digitally measured by Google Analytics within the intervention group. This was done by counting: 1) the number of starting and finishing participants within the time period, 2) the number of started and finished exercises, 3) the number of logins, 4) the number of messages that were put on the discussion board, 5) the number of contact moments with the expert trainers, and 6) the number of messages on the message box.

Power Calculation

To be able to compare our results with previous evaluations of self-management interventions, power calculation was based on the generic outcome parameters. In previous research, the generic measures of self-efficacy (which is close to our measurement of self-management skills) and functioning (which is part of our quality of life measurement), small to moderate effect-sizes (d) were found varying from 0.21 to 0.42 (11,30,31). An effect-size d of 0.30 is similar to an effect-size f of 0.15 in repeated measures analysis of variance. In the current study, to be able to find a small to moderate difference (f=0.15) between the experimental and control groups using repeated measures analysis of variance, the total sample size needs to be N=90 (2 groups of n=45): G*Power3: f=0.15, 1-ß=.80, α=.05 two-tailed, r=.50, two groups, two repeated measures (baseline vs one post-therapy measurement) (32). Taking a dropout rate of 25% into account, we decided to recruit 120 participants. The expectation of a small to moderate effect-size on these generic outcome measures may be explained because patients already have reasonable scores on self-management and quality of life at the start of the study. And there is little reason to expect that the intervention will affect other generic measures such as disease activity and sick leave.

As the crucial aspect of the current study is that the intervention is customized to the needs of the individual participant, the change on the main individual goal is chosen as primary outcome measure, and the change on the other two individual outcome measures (evaluation of the second and third personal goal) is considered important as well. Our sample size is large enough to examine differences in this primary outcome measure. Based on previous studies with individual outcome measures (23-25), we expect a moderate effect-size (d=0.5) for the control group and a large effect-size (d = 1.2) for the intervention group resulting in a moderate to large (d=0.7, f=0.35) effect-size when comparing the intervention group and the control group using the three individual primary and secondary outcome measures. The calculated sample size is therefore considered to be safely chosen to test the main individual goal and both secondary individual outcome measures.

Statistical Analysis

Demographic and disease-specific outcomes will be descriptively presented per group, where possible, with means and standard deviations. The Consolidated Standards of Reporting Trials statement (33) will be used to report the results of this study. Quantitative data will be entered into a SPSS database. Effect analyses will be done according to intention to treat analysis by means of linear mixed-models for longitudinal measurements with random intercept. Fixed effects for group, time, and group × time will be included in the model. Sick leave and health care use will be counted and differences between both groups will be analyzed using parametric tests or nonparametric tests, depending on the distribution of the data. Process outcomes, feasibility, use, and adherence will be analyzed with descriptive statistics.

RESULTS

Patient inclusion and data collection will be completed in February 2017.

DISCUSSION

Implications of the Intervention

A comprehensive assessment of the preferences and needs of patients with a rheumatic disease was used to build ReumaUitgedaagd! Online, guided by Self-Determination theory (17). The usability study showed that the intervention was considered useful, easy to use, and accepted and appreciated by the target group of patients. These results predict that the intervention will be used to improve the use and effectiveness of this intervention, individual goals based on personal needs are identified at the beginning of the intervention and the intervention is customized to the individual patient. Because the intervention is personalized and guided by needs and preferences of patients, a low drop-out rate is expected.

According to the MRC framework (14), which was used by the research team to develop and evaluate the Web-based self-management intervention, this intervention can be defined as complex, taking into consideration the components, the required behaviors, and level of difficulty for both participants and trainer. The intervention is also flexible and customized to the individual participant. Although the MRC framework does not recommend active involvement of the users in the development or evaluation of the intervention, the knowledge and experiences of patient representatives were embedded in all phases of the framework. The aim of involving the users was to the use, acceptance, adherence, and effectiveness of the intervention (13,19). Although we conducted a usability study in an earlier phase of the MRC model, we will measure additional process outcomes like feasibility, use, and adherence to gain knowledge of the working elements of the newly developed Web-based intervention.

Conclusion

Strong features of this Web-based intervention are that it is guided by needs and preferences of patients, that the precise contents of the interventions are customized to the individual patient, and that also the outcome measures fit the self-management goals that are really important to the individual patient. This makes the intervention an example of personalized, patient-centered care. If proven effective, ReumaUitgedaagd! online will be implemented in the Netherlands.

REFERENCES

1. **Salaffi F, Carotti M, Gasparini S, Intorcia M, Grassi W.** The health-related quality of life in rheumatoid arthritis, ankylosingspondylitis, and psoriatic arthritis: a comparison with a selected sample of healthy people. Health Qual Life Outcomes 2009;7:2. (doi: 10.1186/1477-7525-7-25)

2. **Kristiansen TM, Primdahl J, Antoft R, Hørslev-Petersen K.** Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: a focus group study. Musculoskeletal Care 2012;10:29-38. (doi: 10.1002/msc.224)

3. **Hamnes B, Hauge MI, Kjeken I, Hagen KB.** 'I have come here to learn how to cope with my illness, not to be cured': a qualitative study of patient expectations prior to a one-week self-management programme. Musculoskeletal Care 2011;9:200-210. (doi: 10.1002/msc.212)

4. **Bodenheimer T, Lorig K, Holman H, Grumbach K.** Patient self-management of chronic disease in primary care. JAMA 2002;288:2469-2475.

5. **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.

6. **Iversen MD, Hammond A, Betteridge N.** Self-management of rheumatic diseases: state of the art and future perspectives. Ann Rheum Dis 2010;69:955-963. (doi: 10.1136/ard.2010.129270)

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

8. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev 1977;84:191-215.

9. **Marks R, Allegrante JP, Lorig K.** A review and synthesis of research evidence for self-efficacy-enhancing interventions for reducing chronic disability: implications for health education practice (part II). Health Promot Pract 2005;6:148-156. (doi: 10.1177/1524839904266792)

10. **Holloway A, Watson HE.** Role of self-efficacy and behaviour change. Int J Nurs Pract 2002;8:106-115.

11. **Lorig KR, Ritter PL, Laurent DD, Plant K.** The internet-based arthritis self-management program: a one-year randomized trial for patients with arthritis or fibromyalgia. Arthritis Rheum 2008;59:1009-1017. (doi: 10.1002/art.23817)

12. **Ammerlaan J, van OH, Scholtus L, de VA, Zwier M, Bijlsma H, et al.** Feasibility of an online and a face-to-face version of a self-management program for young adults with a rheumatic disease: experiences of young adults and peer leaders. Pediatr Rheumatol Online J 2014;12:10. (doi: 10.1186/1546-0096-12-10)

13. **van Gemert-Pijnen JE, Nijland N, van LM, Ossebaard HC, Kelders SM, Eysenbach G, et al.** A holistic framework to improve the uptake and impact of eHealth technologies. J Med Internet Res 2011;13(4):e111. (doi: 10.2196/jmir.1672)

14. **Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M.** Developing and evaluating complex interventions: the new Medical Research Council guidance. Int J Nurs Stud 2013;50:587-592. (doi: 10.1016/j.ijnurstu.2012.09.010)

15. **Zangi HA, Ndosi M, Adams J, Andersen L, Bode C, Boström C, European League Against Rheumatism (EULAR).** EULAR recommendations for patient education for people with inflammatory arthritis. Ann Rheum Dis 2015;74:954-962. (doi: 10.1136/annrheumdis-2014-206807)

16. **Trappenburg J, Jonkman N, Jaarsma T, van Os-Medendorp H, Kort H, de Wit N, et al.** Self-management: one size does not fit all. Patient Educ Couns 2013;92:134-137. (doi: 10.1016/j.pec.2013.02.009)

17. **Ryan RM, Deci EL.** Self-determination theory and the facilitation of intrinsic motivation, social development, and well-being. Am Psychol 2000;55:68-78.

18. **Davis FD.** Perceived usefulness, perceived ease of use and user acceptance of information technology. Management Information Systems Quarterly 1989;13:319-340.

19. **Dutch Arthritis Foundation. 2016.** URL: http://www.reumafonds.nl/

20. **Reuma Uitgedaagd!. 2016.** URL: https://www.reuma-uitgedaagd.nl/

21. **Dutch Association of Rheumatology.** Richtlijnen, standpunten en zorgpaden. URL: http://www.nvr.nl/nvr-richtlijnen-en-standpunten

22. **Khan F, Pallant JF, Turner-Stokes L.** Use of goal attainment scaling in inpatient rehabilitation for persons with multiple sclerosis. Arch Phys Med Rehabil 2008;89:652-659. (doi: 10.1016/j.apmr.2007.09.049)

23. **Rushton PW, Miller WC.** Goal attainment scaling in the rehabilitation of patients with lower-extremity amputations: a pilot study. Arch Phys Med Rehabil 2002;83:771-775.

24. **Gordon JE, Powell C, Rockwood K.** Goal attainment scaling as a measure of clinically important change in nursing-home patients. Age Ageing 1999;28:275-281.

25. **Osborne RH, Elsworth GR, Whitfield K.** The Health Education Impact Questionnaire (heiQ): an outcomes and evaluation measure for patient education and self-management interventions for people with chronic conditions. Patient Educ Couns 2007;66:192-201. (doi: 10.1016/j.pec.2006.12.002)

26. **Kroon FP, van der Burg LR, Buchbinder R, Osborne RH, Johnston RV, Pitt V.** Self-management education programmes for osteoarthritis. Cochrane Database Syst Rev 2014;1:CD008963. (doi: 10.1002/14651858.CD008963.pub2)

27. **Gandhi SK, Salmon JW, Zhao SZ, Lambert BL, Gore PR, Conrad K.** Psychometric evaluation of the 12-item short-form health survey (SF-12) in osteoarthritis and rheumatoid arthritis clinical trials. Clin Ther 2001;23:1080-1098.

28. **Holden RJ, Karsh B.** The technology acceptance model: its past and its future in health care. J Biomed Inform 2010;43:159-172. (doi: 10.1016/j.jbi.2009.07.002)

29. **Lorig KR, Ritter PL, Dost A, Plant K, Laurent DD, McNeil I.** The Expert Patients Programme online, a 1-year study of an Internet-based self-management programme for people with long-term conditions. Chronic Illn 2008;4:247-256. (doi: 10.1177/1742395308098886)

30. **Ory MG, Smith ML, Ahn S, Jiang L, Lorig K, Whitelaw N.** National study of chronic disease self-management: age comparison of outcome findings. Health Educ Behav 2014;41(Suppl):34S-42S. (doi: 10.1177/1090198114543008)

31. **Faul F, Erdfelder E, Lang A, Buchner A.** G*Power 3: a flexible statistical power analysis program for the social, behavioral, and biomedical sciences. Behav Res Methods 2007;39:175-191.

32. **Moher D, Hopewell S, Schulz KF, Montori V, Gøtzsche PC, Devereaux PJ, CONSORT.** CONSORT 2010 explanation and elaboration: updated guidelines for reporting parallel group randomised trials. Int J Surg 2012;10:28-55. (doi: 10.1016/j.ijsu.2011.10.001)

"'What's most important' drives the head, heart, and hands. When we know what's most important to a patient we can make decisions more easily by aligning priorities. So be sure to ask this crucial question of your patients and of yourself"

- @Twitter/KB

CHAPTER 9

THE MOST IMPORTANT NEEDS AND PREFERENCES OF PATIENTS FOR SUPPORT FROM HEALTH CARE PROFESSIONALS: A REFLECTIVE PRACTICE ON TRANSITIONAL CARE FOR YOUNG ADULTS WITH JUVENILE IDIOPATHIC ARTHRITIS

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Introduction

Due to research, technology and innovation, we are living longer and therefore the number of people suffering from a chronic disease is increasing (1, 2). Having a chronic disease, such as arthritis, imposes daily challenges upon those affected and necessitates patients to make daily decisions about the way they manage their illness (3-5). The question is not *whether* patients manage their (chronic) illness, but *how* they do this. The (individual) ability to manage sometimes debilitating symptoms, changing treatment strategies, day to day physical and psychosocial consequences, the flexibility to perform life style changes inherent in living with a chronic disease, is often defined as self-management (6). Living with a rheumatic disease is challenging at any age. However, these challenges may be particularly difficult for young adults with juvenile idiopathic arthritis (JIA), since their chronic condition and treatment affect both physical and socio-emotional development (7, 8). Although in the past decade many new treatment options for JIA have been developed, to date it is not possible to cure this disease (9). And so, many of these children are growing into adulthood with JIA. One of the main challenges young adults with JIA have to deal with is to increase their own capability to think critically and to make autonomous, informed decisions: they have to become a self-manager (10, 11). To enhance young adult's self-management, we as child- and adult treatment teams, we paediatricians, rheumatologists and rheumatology nurses, have to adjust care (12). At the paediatric Rheumatology department and the adult Rheumatology & Clinical Immunology department, we developed the organisation of our care using patient-stories in dialogue with young adults to 'put them in the centre of our care' and worked with them to develop innovations to fit closely to their needs and preferences. As we inferred from our personal experiences, collaboration with the young adults in all phases of development was crucial and influenced the actual use, adherence and effectiveness of these innovations. Presently, we are expanding these experiences towards developing care interventions for the adult group also. The aim of this article is to share our experiences, the lessons we have learned, and to share our reflections on the question: are we there yet?

Box 1 Anne – personal notes

The pediatrician introduced the next patient to the outpatient clinic as a vulnerable young-girl, with no friends and severe systemic juvenile arthritis. The door opened and there was Anne, a young girl of 17 years, sitting in a wheelchair. Her mother pushed the wheelchair into the consultation room. Her father came along too. He walked with the coats and the bags of Anne and her mother and in a second, the consultation room was filled with the pediatrician, a family, a wheelchair and me. The pediatrician started the consultation and asked: "How are you today, Anne?" Anne replied, quietly, as she looked at the ground, "good", while father and mother looked worried. I looked at Anne's nose and lips where impressive piercings were placed. I also noticed a bracelet on her wrist with the name of a popular festival on it, a sign that Anne had been there. Mother took the floor and expressed her concerns. She had tried not interfering with the medication regime and to leave it to Anne, as had been agreed during the previous consultation. But it hadn't worked. Anne had not taken any pills and mother felt helpless. Father also took the floor: he was worried about Anne. She did not go to school and seemed to have no interest in her future. Discussions about this subject caused a lot of stress and had led to father and daughter not being on speaking terms for the last two weeks. Anne gave her father the silent treatment. The pediatrician nodded understandably and tried to make eye contact with Anne. He cleared his throat and stressed the importance of taking the medication and of focusing on the future. Anne nodded too and murmured that she understood what he was saying. The parents still looked worried. Anne looked at the ground.

May I ask how you think we should organize your treatment?

Our present health care system does not adequately accommodate an increasing diversity of patient's needs and preferences, especially those of young adults with a rheumatic disease who are in transition from child to adulthood (7, 12-14). Throughout the years, we have experienced many problems after the young adult made the transfer from child to adult hospital. Problems like high rates of treatment dropout and passive, resistant behaviour towards caregivers and/or parents were recognizable (12, 13). 'What young adults want and what their priorities are' and 'what health care professionals want and what their priorities are' may differ considerably. In order to bridge the gap between these two worlds, we initiated conversations with the young adults and asked them to share their experiences on growing up with a rheumatic disease and to reflect on their needs and preferences for support from health care professionals. Two questions were leading in these conversations: 'What is most important for you?' and 'How would you like us to organize your treatment'? The answers were honest, simple and down to earth. *"listen more and speak less", "I don't want to be an exception"*, "*your recommendations should fit into my daily life*", "*information should meet my language*" and "*I want to be prepared and I want to be in charge*" were needs they shared. The young adults also shared some solutions like the use of Internet to contact and provide information and the need for a professional who's main focus is 'self-management and dealing with the consequences of rheumatism on daily life'.

Box 2 Anne – personal notes

Due to a technical problem with the computer, the pediatrician took Anne's parents to another room to discuss the X-rays of Anne's hands. Anne did not want to go. I asked her why she was not interested in her X-rays. From her point of view, she already knew the results: "ugly hands and painful hands". So Anne and I stayed behind. It was suddenly strangely silent and Anne sighed. I smiled at her, she smiled back. "You're a nurse, right?" she asked. I nodded. Some tears dripped from her eyes and she squeezed her hands forcefully.
"Can I ask you something?" she said. I gave her an encouraging nod and moved my chair closer to her wheelchair. "A few weeks ago, I had sex at a party and did not use condoms.
I think I am pregnant".

I nodded and swallowed. It became somewhat hotter in the consultation room.

Formulating answers

Asking questions and young adults sharing their needs and preferences provided the fundament for some innovations we developed with and for the young adults. We developed an outpatient transition clinic where young adults and their parents/caregivers were able to meet their future rheumatologist at an early age and where a transition nurse guided and supported them during the transfer from child to adult care and self-management (12). To improve the young adults' knowledge and to meet the needs for information fit to their language and preferences, an informative website www.jong-en-reuma.nl with written and video information, was installed. To encourage active involvement and to stimulate the feelings of control, a web-based portal with access to the personal medical record, self-monitoring and eConsult functionalities was developed (15). Young adults who used these eHealth applications expressed their needs for a training program, where they could practice self-management skills. To meet these needs, we developed two versions of a self-management training program: a face-to-face and a web-based version (16). The two programs www.reuma-uitgedaagd.nl are led by young peers themselves. And, although the treatment of rheumatic diseases for adults with a rheumatic disease has improved considerably in the past years, these patients also expressed their need for an individualized self-management intervention focused on their life-stages and problems (17). Based on an extensive needs assessment, we also developed peer guided web-based intervention, specially targeted at the age of 26 years and older (17).

Keep asking the question

The interventions we developed to support our patients in self-management, proved to be feasible and were appreciated. We experienced that especially small steps, driven by dialogue and by patient stories, in day-to-day practice, gave meaningful results for our patients. But are we there yet? We don't think so. Our focus on the patient alone, appears not be enough to change daily practice of professionals. Also we ourselves, being professionals, need to be supported towards patient-centered care. We still have a way to go.

Reflecting on the case of Anne as described in boxes 1 and 2, we nowadays start our consultation asking 'what's the most important thing you want to discuss with me?' Asking this question might invite our patients, like Anne, to adopt a more active role, to share their thoughts and worries and might increase self-management and adherence to medical treatment. Considering the needs of our patients, we are constantly aware that 'we are guests in their lives' (18). Most of the work Anne and our other patients perform, managing their life with a chronic disease, is done at home: alone or with their parents or spouses. Gradually we experienced that the concept of patient-centered care implies a change towards a less dominant role for us. Integrating patients' expertise and respecting their decisions are skills that do not naturally fit in our traditional role as health care professionals (19-21). We should talk less and listen more, through being present (22). Patient centeredness demands professional attention and genuine commitment (22, 23). Its success depends on how health care professionals are acquainted with the concept and to which extent we are equipped with the skills the concept requires. Our communication and listening skills need constant attention and training, and therefore they have to be imbedded in professional education as well as in daily supervision on the work floor (23-25). By integrating the patients' expertise and supervision in the professional education of our students and ourselves, we can improve both skills. Also important is the support of colleagues and management next to evaluation with our patients to reflect on our performance, especially when we doubt or feel insecure with redesigning our role (20, 25). But increasing knowledge, training skills, integrating patients' expertise and support isn't enough; there is more to it.

The success of patient-centered care depends on shared values of the concept (18, 19, 25, 26). In a study on the challenges of fostering the adoption of the empowerment paradigm (25), including patient-centered care, the results revealed that, even after intensive education and training, health care professionals still were not able to implement or sustain the paradigm in daily practice. The authors discussed that the meaning the health professional gives to the underlying philosophy of patient-centered care – the personal believes, goals and priorities – determines his or her actions (24-26). An understanding of how health professionals personally think about patient-centered care is essential and needs to be discussed in daily care. Thus, being a health care professional, we can only implement patient-centered care when we are intrinsically motivated to do so. Like a patient can only be a self-manager and adhere to treatment when they are intrinsically motivated. Consequently, it determines the questions asked to our patient, the actions taken and decisions made. It determines the way of being present with attention and commitment in daily health care. It affects the *hands, head* and *heart*. To achieve patient centeredness, we have to work side-by-side with our patients. This implies that daily reflection on shared answers to the question ‘what’s the most important’ is mandatory. So be sure to ask this question to your patients as well as to yourself.

REFERENCES

1. **(A healthier Netherland).** Available at: www.eengezondernederland.nl.

2. **Tackling chronic disease in Europe.** Strategies, intervention and challenges. Available at: http://www.euro.who.int/en/health-topics/Life-stages/healthy-ageing/publications/2010/tackling-chronic-disease-in-europe-strategies,-interventions-and-challenges.

3. **Salaffi F, Carotti M, Gasparini S, et al.** The health-related quality of life in rheumatoid arthritis, ankylosing spondylitis, and psoriatic arthritis: a comparison with a selected sample of healthy people. Health Qual Life Outcomes 2009;7:25,7525-7-25 doi:10.1186/1477-7525-7-25.

4. **Hamnes B, Hauge MI, Kjeken I, et al.** 'I have come here to learn how to cope with my illness, not to be cured': a qualitative study of patient expectations prior to a one-week self-management programme. Musculoskeletal Care 2011;9:200-10 doi:10.1002/msc.212; 10.1002/msc.212.

5. **Kristiansen TM, Primdahl J, Antoft R, et al.** Everyday life with rheumatoid arthritis and implications for patient education and clinical practice: a focus group study. Musculoskeletal Care 2012;10:29-38 doi:10.1002/msc.224.

6. **Barlow J, Wright C, Sheasby J, et al.** Self-management approaches for people with chronic conditions: a review. Patient Educ Couns 2002;48:177-87.

7. **Bloom SR, Kuhlthau K, Van Cleave J, et al.** Health care transition for youth with special health care needs. J Adolesc Health 2012;51:213-9 doi:10.1016/j.jadohealth.2012.01.007.

8. **Suris JC, Michaud PA, Viner R.** The adolescent with a chronic condition. Part I: developmental issues. Arch Dis Child 2004;89:938-42 doi:10.1136/adc.2003.045369.

9. **Prakken B, Albani S, Martini A.** Juvenile idiopathic arthritis. Lancet 2011;377:2138-49 doi:10.1016/S0140-6736(11)60244-4.

10. **Wagner-Weiner L.** Pediatric rheumatology for the adult rheumatologist. J Clin Rheumatol 2008;14:109-19 doi:10.1097/RHU.0b013e31816b4460.

11. **Shaw KL, Southwood TR, McDonagh JE, et al.** Developing a programme of transitional care for adolescents with juvenile idiopathic arthritis: results of a postal survey. Rheumatology (Oxford) 2004;43:211-9 doi:10.1093/rheumatology/keh018.

12. **Ammerlaan JW, Scholtus LW, Bijlsma HJ, et al.** An urge for change: Transitional care for young adults with juvenile idiopathic arthritis. Patient Educ Couns 2013 doi:10.1016/j.pec.2013.02.006.

13. **Hazel E, Zhang X, Duffy CM, et al.** High rates of unsuccessful transfer to adult care among young adults with juvenile idiopathic arthritis. Pediatr Rheumatol Online J 2010;8:2 doi:10.1186/1546-0096-8-2.

14. **Stinson JN, Toomey PC, Stevens BJ, et al.** Asking the experts: exploring the self-management needs of adolescents with arthritis. Arthritis Rheum 2008;59:65-72 doi:10.1002/art.23244.

15. **Ammerlaan JJ, Scholtus LW, Drossaert CH, et al.** Feasibility of a Website and a Hospital-Based Online Portal for Young Adults With Juvenile Idiopathic Arthritis: Views and Experiences of Patients. JMIR Res Protoc 2015;4:e102 doi:10.2196/resprot.3952.

16. **Ammerlaan J, van Os-Medendorp H, Scholtus L, et al.** Feasibility of an online and a face-to-face version of a self-management program for young adults with a rheumatic disease: experiences of young adults and peer leaders. Pediatr Rheumatol Online J 2014;12:10,0096-12-10 doi:10.1186/1546-0096-12-10.

17. **Ammerlaan JW, Mulder OK, de Boer-Nijhof NC, et al.** Building a Tailored, Patient-Guided, Web-Based Self-Management Intervention 'ReumaUitgedaagd!' for Adults With a Rheumatic Disease: Results of a Usability Study and Design for a Randomized Control Trail. JMIR Res Protoc 2016;5:e113 doi:10.2196/resprot.5735.

18. **Berwick DM.** What 'patient-centered' should mean: confessions of an extremist. Health Aff (Millwood) 2009;28:w555-65 doi:10.1377/hlthaff.28.4.w555.

19. **Dwarswaard J, van de Bovenkamp H.** Self-management support: A qualitative study of ethical dilemmas experienced by nurses. Patient Educ Couns 2015;98:1131-6 doi:10.1016/j.pec.2015.05.017.

20. **Holm AL, Severinsson E.** Reflections on the ethical dilemmas involved in promoting self-management. Nurs Ethics 2014;21:402-13 doi:10.1177/0969733013500806.

21. **Mudge S, Kayes N, McPherson K.** Who is in control? Clinicians' view on their role in self-management approaches: a qualitative metasynthesis. BMJ Open 2015;5:e007413,2014-007413 doi:10.1136/bmjopen-2014-007413.

22. **Klaver K, Baart A.** How can attending physicians be more attentive? On being attentive versus producing attentiveness. Med Health Care Philos 2016 doi:10.1007/s11019-015-9669-y.

23. **Dwamena F, Holmes-Rovner M, Gaulden CM, et al.** Interventions for providers to promote a patient-centred approach in clinical consultations. Cochrane Database Syst Rev 2012;12:CD003267 doi:10.1002/14651858.CD003267.pub2.

24. **Anderson RM, Funnell MM.** Patient empowerment: myths and misconceptions. Patient Educ Couns 2010;79:277-82 doi:10.1016/j.pec.2009.07.025.

25. **Anderson RM, Funnell MM.** Patient empowerment: reflections on the challenge of fostering the adoption of a new paradigm. Patient Educ Couns 2005;57:153-7 doi:S0738-3991(04)00165-X (pii).

26. **Been-Dahmen JM, Dwarswaard J, Hazes JM, et al.** Nurses' views on patient self-management: a qualitative study. J Adv Nurs 2015;71:2834-45 doi:10.1111/jan.12767.

CHAPTER 10

GENERAL DISCUSSION

This final chapter of this thesis starts with a summary of the findings, described in the chapters of this thesis, followed by a reflection on three themes: exploring needs for self-management support, development and feasibility and evaluating self-management interventions. These themes form the important elements of our roadmap toward tailored online interventions. The chapter ends with some recommendations.

Summarizing the findings

Redeveloping health care (chapter 2 and 9)

Reflections on redeveloping health care to accommodate the diversity of young adults' needs and preferences for care and support lead to the awareness that an essential part of this process is about integrating the patients individual situation, their experiences, and their expectations of daily health care. To implement patient centered care, we realize, as shown in chapter 9, that it is not only our patient that needs to be supported towards self-management. Also, we as health care professionals need to gain and regain knowledge and skills continuously. Even of more importance, we should be aware of our own values towards concepts of patient centered care (1-3). A daily reflection on shared answers of patients and health care professionals to the question 'what is most important to you?' may be helpful to processes of redeveloping health care.

Feasibility of a website and hospital based portal for young adults (chapter 3)

An informative website and a hospital based online portal were developed in close cooperation with the young adult group themselves aimed to support the young adult. We tested the feasibility of these eHealth applications using the three concepts of the Technology Acceptance Model (TAM)(4): ease of use, usefulness and intention to use. The young adults evaluated both applications as useful in dealing with problems in daily life and easy to use. They reported that especially new medical information and feedback of health care professionals during consultations do motivate them to revisit the applications. Based on these results, the informative website and patient portal have been integrated in usual care.

Feasibility of face-to-face and online self-management intervention for young adults (chapter 4)

To support self-management of the young adults with a juvenile rheumatic disease, we developed a peer guided self-management intervention which can be followed online for 6 weeks, or during a weekend, based on the self-efficacy theory (5). Also in this chapter, we studied the feasibility of both self-management interventions among young adults and peer leaders before implementing in daily care, using concepts of the TAM (4). Both patient groups showed to appreciate the face-to-face and online self-management support interventions as easy to use and useful considering dealing with problems in daily life. The peer leaders of the online patient group were surprised by the levels of intimacy and frankness of communication that were reached despite the fact participants and peer leaders of the online interventions could not see each other. Apparently, a safe atmosphere where participants could share and discuss their situation and problems was created.

Effectiveness of the online self-management intervention for young adults (chapter 5)

We conducted a randomized controlled trial with additional qualitative analyses of two interaction components (goals and Chat). The online self-management intervention did not lead to an improvement of self-efficacy, quality of life and self-management of young adults with Juvenile Idiopathic Arthritis (JIA). Qualitative data revealed that patients report their goals are achieved. Analysis of the chat showed modelling and sharing experiences as the most used interactions. Adherence to the intervention was high, appreciation of the own learning experiences and personal achievements in the online training was rated positively. More research is needed in order to measure possible added values of this intervention compared to usual care.

Translation and validation of a self-management questionnaire (chapter 6)

The Health Education Questionnaire (heiQ) (6) was chosen to capture the patients perspective in the evaluation of self-management. Therefore, we first conducted a systematic translation process of the original English heiQ (6). After that, the psychometric properties of the Dutch translation were studied to test its value to evaluate self-management interventions among patients with a chronic disease. Data show that the Dutch heiQ has good psychometric properties in patient groups with rheumatoid arthritis, atopic dermatitis, food allergy and asthma, indicating that it is a robust outcome measure of health education and self-management programs also in The Netherlands. The psychometric properties of the Dutch translation were comparable with the English (6), German (7) and French versions (8), which provide evidence that the heiQ conceptualizes a broad range of self-management skills in a consistent way across chronic conditions, cultures and languages.

Exploring the needs and preferences for self-management support among adults (chapter 7)

We studied individual preferences on structure and needs for content of an online self-management intervention to support adult patients with a rheumatic disease. Considering structure, participants prefer a tailored intervention with personal needs and circumstances taken into account. They indicate they want to follow the intervention at their own pace in their own time. They also prefer to be guided by a trained patient, rather than a health professional. In addition to gaining knowledge about disease and treatment, participants identify as important content of the intervention the clusters about increasing competences in self-efficacy, dealing with societal procedures and working together with health care providers, handling disease fluctuations, psychological distress and maintaining and increasing physical activity. Also clusters on relational needs towards family, friends, children and co-workers were expressed to be valuable.

Developing an online self-management intervention for adults (chapter 8)

Based on the results of the comprehensive assessment of preferences and needs of adult patients as shown in chapter 7, a web-based peer-guided and tailored self-management intervention for adults was developed and tested on feasibility among adults with a rheumatic disease, using the concepts of the TAM (4). The participants evaluated the intervention positively on usefulness and ease of use. Participants stated that they were likely to participate in daily practice since they are now familiar with content and structure. In this chapter we also present the study design of the RCT which will be conducted in due time. Consistent with the Self-Determination Theory (9) – the theoretical fundament of the online intervention - progress towards the personal goal will be the primary outcome measure of the intervention.

Exploring needs for self-management support

Reflecting on supportive care for young adults (chapter 2), their needs appear to be strongly related to their life stage and thereby their maturing processes. The young adults in our study express that they are living in the moment, and want to experience and to experiment as their peers do. They certainly do not want to label or present themselves as being sick. These outcomes are confirmed by other studies on experiences of young adults with a chronic disease (10, 11). The young adults, who participated in the presented studies, are all in the age of 16-25 years. This life-stage is recognized as a crucial, turbulent and unique period of growth and development between child and adulthood, where numerous biological, psychological and social changes take place (10, 12, 13). These changes may influence their ability to reflect on behavior and its consequences, for instance non-compliancy to treatment. This might also explain why young adult are mainly living in the moment, with less focus on the future and on long term consequences of actual behavior.

Also adult patients expressed that supportive care should integrate their individual life experiences, questions and problems, as is concluded in chapter 7. But in contrast to young adults, their focus is more related to being sick and dealing with its consequences, in their social life as in their personal relationships. They indicated informational, emotional, social and practical support as needed categories, which is in line with the needs for support of other patient groups with a chronic disease (14, 15). Despite a considerably improvement of medical treatment in the last 10 years, especially in rheumatology, and the already implemented multidisciplinary support (16-18), patients in our study still express the need to be supported by peers. They want to increase the knowledge, skills and competences about their disease, on physical activity, managing emotions, and self-regulating their own life, on family life, work and leisure time. Participants in our study expressed clearly that having a rheumatic disease affects the relationship with their family, friends, and coworkers, so this should also be a subject for support.

In summary, exploring the needs of self-management support among young and adult patients with a rheumatic disease, the development of self-management interventions tailored to the individual needs, circumstances, life stages and problems of the individual patient, is essential and inevitable. Here also the question 'what's most important to you within the context of care' which we proposed in chapter 9, might help to define mutual goals of supportive care.

Development and feasibility

Self-management is complex and requires interventions that have different qualities including flexibility, modes of delivery (face-to-face, online), usage of knowledge, elements of motivation and behavior and diversity in themes of content. Due to these requirements, self-management support interventions are complex interventions and consequently a systematic and person-based approach to develop is essential (19-21). In this thesis (chapter 3, 4 and 8), the new Medical Research Council (MRC) guidance (21) is followed. This guidance includes the following phases: Development, Feasibility and Piloting and Evaluation and Implementation (see figure 1).

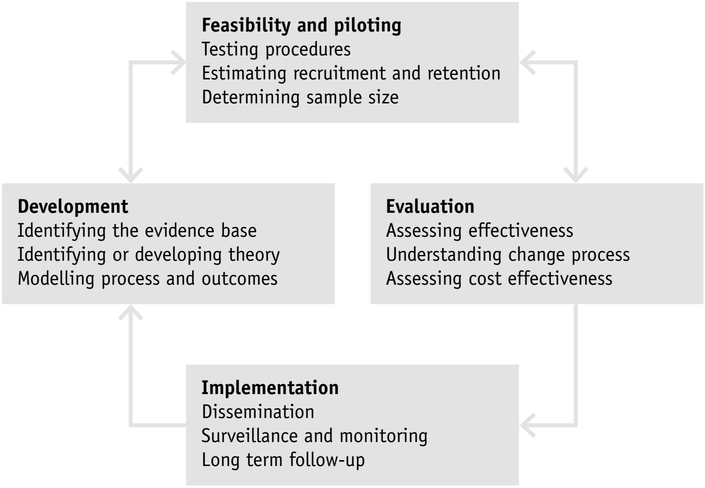

Figure 1 The phases of the new Medical Research Council guidance (21)

Based on earlier experiences, we chose to work intensively together with patient partners and patient representatives of the Dutch Arthritis Foundation in the development and research of the self-management interventions. This to stay close to their needs and preferences and to bridge the gap between real life and the world of care. Several levels of patient participating in research have been described, varying from participating as a study subject to contributing to set the research agenda (22, 23). We chose to work with patients during the whole cycle: from defining research goals, to choosing the design and offering input and analyzing and describing the results. In our department, it was a new experience to work so closely with our patients. In this challenging journey, some aspects might be worthwhile to mention. First of all, the contribution of patients was appreciated as extremely valuable. They shared their knowledge and their experiences. They gave feedback and asked questions on why and for whom a certain matter might be that important. It made us more aware of the existence of a gap between professionals' and patients' perspectives on supporting management. The results of this collaboration on content, layout, language and style made our interventions more applicable, and may indeed have influenced the positive evaluation of the usefulness of the applications, as described in chapter 3, 4 and 8. Our decision to involve patients is supported by other studies, mostly derived from information science and development of eHealth applications, where the involvement of end-users in developing eHealth applications is described to be one of the crucial aspects of acceptance of the tools themselves (24, 25). We experienced that a clear plan with a description of the specific goals, tasks and responsibilities helped us to keep focused. During the project, we initiated regular meetings, firstly to evaluate the progress of the project, but mostly to reflect on working together as patients and professionals, respecting each other's role, capacity and input. In this process, we followed the recommendations of the European League Against Rheumatism (EULAR) for the involvement of patient representatives in scientific projects (26).

Feasibility

To gain insight on use and acceptance, we tested the website and portal (chapter 3) and the self-management interventions (chapter 4 and 8) on feasibility, before implementing these tools in daily life of our patients. We used the Technology Acceptance (TAM) (4) (see figure 2, see next page), a prediction and explanation model of the end-users reaction to an innovation. Although originally TAM was not developed for health care, it was used and validated in its generic form in several studies on eHealth interventions (27, 28). TAM posits two factors: perceived usefulness (U) and perceived ease of use (E) to determine the intention to use (BI) which influences the actual use. In our feasibility studies, we focused mainly on perceived usefulness and ease of use. Reflection on this decision learned that focusing on these two factors alone may be subject to some limitations. Also age, social influence and motivation may influence as well usefulness as ease of use. So, with gained insight, we now recommend the Unified Theory of Acceptance and Use of Technology (UTAUT) (29) to use in feasibility testing. In this latest version of the TAM, factors including age, social influence, voluntariness of use and effort-expectancy are added.

Evaluating self-management interventions

Based on our aim to increase knowledge and self-management skills of the young adults, we chose as theoretical fundament the Self-efficacy theory (5). But, as shown in chapter 5, focusing on knowledge and skills alone appeared not to be enough. Also individual needs and circumstances of the young adult should be taken into account. It has been reported that also motivation is an important drive for behavioral change (30, 31) and the self-determination theory (9) emphasizes the importance of keeping goals of behavior change close to the autonomous motivation and circumstances of people. Therefore, an expansion of the theoretical fundament of the online intervention for young adults with elements of the self-determination theory (9), as we did for the adult version, maybe helpful to improve acceptability and effectiveness of the intervention.

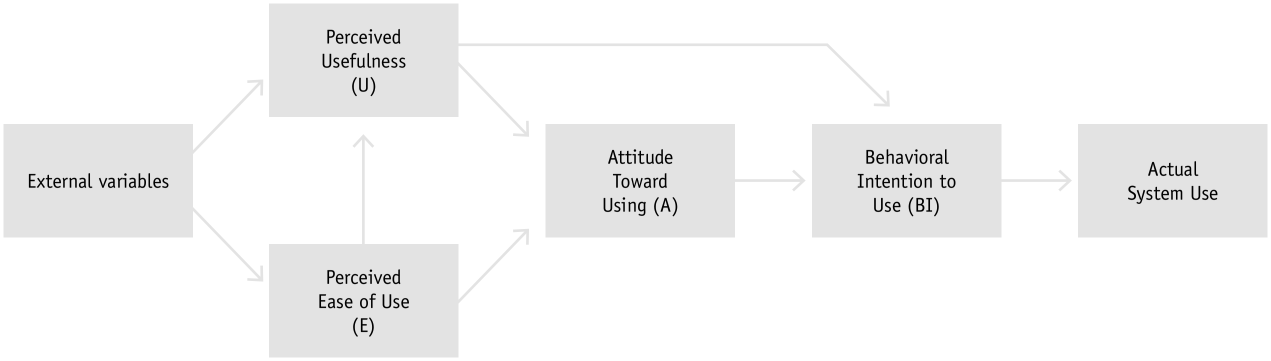

Figure 2 The Technology Acceptance Model (TAM) (4)

Patient reported outcomes (PROs)

In our studies, patient reported outcomes (PROs) as personal goals (chapter 8) and self-efficacy items (chapter 5) were chosen to capture the patients' perspectives on the effectiveness of the self-management interventions. Other PROs as functionality, quality of life and absenteeism were added in order to compare our outcomes with other studies on self-management interventions (32-34). In rheumatology, PROs for pain, functionality and patient global wellbeing to evaluate interventions, treatment strategies or trials are commonly used (35-38). Studies focusing on self-management and support show that PROs are more sensitive to change compared to clinical outcome measurements (37, 39). But the PROs, and also the questionnaires used to measure self-management may vary in the different studies, which makes it difficult to generalize the results. The Health Education Impact Questionnaire (6) may be of additional value, since we demonstrated in chapter 6 its good psychometric properties to measure a broad range of self-management skills across chronic conditions, cultures and languages.

Recommendations

The recruitment of young adults from the outpatient transitional departments of university hospitals limits the generalizability of our findings. Firstly, usual care in these university hospitals includes the availability of a multidisciplinary team, of which a transition nurse supports self-management. This might explain the relatively high base line scores on self-efficacy, quality of life and self-management of the participants in the effectiveness study of chapter 5. Secondly, university hospitals mostly treat patients, referred by regional hospitals partly because of the complexity of their disease or treatment including biologicals. Further research in a broader patient group might address the question who will benefit the most of the self-management intervention and whether it is necessary to determine specific inclusion criteria.

Adults with interest in online interaction and survey participated in our studies on preferences and needs (chapter 7), and usability (chapter 8), while patients who might be less experienced or less interested did not. Our recommendation would therefore be to study experiences and self-management in a broad range of participants, including those without particular interest in online communication.

We choose to guide the self-management programs for young adults by peers because Bandura's self-efficacy theory (5) addresses modelling to be an important factor to increase self-management. Others stress that learning and identification with peers is a way to educate and to increase coping styles among this age group (11, 40, 41). Also the participants of the adult self-management intervention expressed to prefer to be guided by peer trainers (chapter 7). Several studies suggest that well trained peers teach as well as, and probably better than, health professionals (42, 43). Our analyses of the interactions in the online intervention for young adults (chapter 5) showed that the contribution of the peer trainers varied in different groups. We did not examine the influence of the peer trainer on the outcome of the online intervention. Focus group discussions with young adults might be an appropriate way to gain additional information. As it is expected, also the peer trainers are undergoing a learning experience while performing their role in the intervention. We therefore recommend exploring the perspectives of the peer trainers themselves and their development with regard to self-management.

Developing eHealth applications and interventions in health care should always be started with determining possible additional values for patients, health care professionals and others. We should avoid that eHealth becomes a goal in itself (44). We feel that equal attention has to be given to the last phases of the project: the implementation phase, 'what happens with the eHealth intervention when the grant funding is over?' and the dissimilation phase 'how do we adjust the eHealth interventions to the results of ongoing studies and the technology of today?'.

Therefore a constructive plan is essential, where the roles and responsibilities of each stakeholder - patient, professional, management, technique and financer - is described (24, 45). As the aim would be to create a continuous, ongoing model of (financial) support for post development and sustainability of the eHealth applications, collaborating with partners and stakeholders, including Health Insurances companies, is important. Within this context, in our presented research projects, we closely worked together not only with patients, but as inspiring, with web developers and technicians (www.hollandswild.nl), coaches (www.work21.nl) and the Dutch Arthritis Foundation. The Dutch Arthritis Foundation also financed some of our research projects and expands the availability of the developed self-management interventions to all patients with a rheumatic disease in the Netherlands.

REFERENCES

1. **Anderson RM, Funnell MM.** Patient empowerment: reflections on the challenge of fostering the adoption of a new paradigm. Patient Educ Couns. 2005 May;57(2):153-7.

2. **Berwick DM.** What 'patient-centered' should mean: confessions of an extremist. Health Aff (Millwood). 2009 Jul-Aug;28(4):w555-65.

3. **Holmstrom I, Roing M.** The relation between patient-centeredness and patient empowerment: a discussion on concepts. Patient Educ Couns. 2010 May;79(2):167-72.

4. **Davis FD.** Perceived usefulness, perceived ease of use and useracceptance of information technology. Management Information Systems Quarterly. 1989;13(3):319-40.

5. **Bandura A.** Self-efficacy: toward a unifying theory of behavioral change. Psychol Rev. 1977 Mar;84(2):191-215.

6. **Osborne RH, Elsworth GR, Whitfield K.** The Health Education Impact Questionnaire (heiQ): an outcomes and evaluation measure for patient education and self-management interventions for people with chronic conditions. Patient Educ Couns. 2007 May;66(2):192-201.

7. **Schuler M, Musekamp G, Faller H, Ehlebracht-Konig I, Gutenbrunner C, Kirchhof R, et al.** Assessment of proximal outcomes of self-management programs: translation and psychometric evaluation of a German version of the Health Education Impact Questionnaire (heiQ). Qual Life Res. 2013 Aug;22(6):1391-403.

8. **Epstein J, Osborne RH, Elsworth GR, Beaton DE, Guillemin F.** Cross-cultural adaptation of the Health Education Impact Questionnaire: experimental study showed expert committee, not back-translation, added value. J Clin Epidemiol. 2015 Apr;68(4):360-9.

9. **Ryan RM, Deci EL.** Self-determination theory and the facilitation of intrinsic motivation, social development, and well-being. Am Psychol. 2000 Jan;55(1):68-78.

10. **Suris JC, Michaud PA, Viner R.** The adolescent with a chronic condition. Part I: developmental issues. Arch Dis Child. 2004 Oct;89(10):938-42.

11. **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 Sep;51(3):213-9.

12. **Blum RW, Bastos FI, Kabiru CW, Le LC.** Adolescent health in the 21st century. Lancet. 2012 Apr 28;379(9826):1567-8.

13. **Eleftheriou D, Isenberg DA, Wedderburn LR, Ioannou Y.** The coming of age of adolescent rheumatology. Nat Rev Rheumatol. 2014 Mar;10(3):187-93.

14. **van Houtum L, Rijken M, Heijmans M, Groenewegen P.** Patient-perceived self-management tasks and support needs of people with chronic illness: generic or disease specific? Ann Behav Med. 2015 Apr;49(2):221-9.

15. **Dwarswaard J, Bakker EJ, van Staa A, Boeije HR.** Self-management support from the perspective of patients with a chronic condition: a thematic synthesis of qualitative studies. Health Expect. 2016 Apr;19(2):194-208.

16. **Zuidema RM, Repping-Wuts H, Evers AW, Van Gaal BG, Van Achterberg T.** What do we know about rheumatoid arthritis patients' support needs for self-management? A scoping review. Int J Nurs Stud. 2015 Jun 9.

17. **Zangi HA, Ndosi M, Adams J, Andersen L, Bode C, Bostrom C, et al.** EULAR recommendations for patient education for people with inflammatory arthritis. Ann Rheum Dis. 2015 Jun;74(6):954-62.

18. **van Eijk-Hustings Y, Ammerlaan J, Voorneveld-Nieuwenhuis H, Maat B, Veldhuizen C, Repping-Wuts H.** Patients' needs and expectations with regard to rheumatology nursing care: results of multicentre focus group interviews. Ann Rheum Dis. 2013 Jun;72(6):831-5.

19. **Kreuter MW, Skinner CS.** Tailoring: what's in a name? Health Educ Res. 2000 Feb;15(1):1-4.

20. **Yardley L, Morrison L, Bradbury K, Muller I.** The person-based approach to intervention development: application to digital health-related behavior change interventions. J Med Internet Res. 2015 Jan 30;17(1):e30.

21. **Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M.** Developing and evaluating complex interventions: the new Medical Research Council guidance. Int J Nurs Stud. 2013 May;50(5):587-92.

22. **Sacristan JA, Aguaron A, Avendano-Sola C, Garrido P, Carrion J, Gutierrez A, et al.** Patient involvement in clinical research: why, when, and how. Patient Prefer Adherence. 2016 Apr 27;10:631-40.

23. **de Wit MP, Kvien TK, Gossec L.** Patient participation as an integral part of patient-reported outcomes development ensures the representation of the patient voice: a case study from the field of rheumatology. RMD Open. 2015 Aug 5;1(1):e000129,2015-000129. eCollection 2015.

24. **van Gemert-Pijnen JE, Nijland N, van Limburg M, Ossebaard HC, Kelders SM, Eysenbach G, et al.** A holistic framework to improve the uptake and impact of eHealth technologies. J Med Internet Res. 2011 Dec 5;13(4):e111.

25. **Nijland N.** Grounding eHealth. Towards a holistic framework for sustainable eHealth technologies. . 2011.

26. **de Wit MP, Berlo SE, Aanerud GJ, Aletaha D, Bijlsma JW, Croucher L, et al.** European League Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects. Ann Rheum Dis. 2011 May;70(5):722-6.

27. **Yarbrough AK, Smith TB.** Technology acceptance among physicians: a new take on TAM. Med Care Res Rev. 2007 Dec;64(6):650-72.

28. **Holden RJ, Karsh BT.** The technology acceptance model: its past and its future in health care. J Biomed Inform. 2010 Feb;43(1):159-72.

29. **Venkatesh V, Morris MG, Davis GB, Davis FD.** User acceptance of Information Technology: Toward a Unified view. MIS Quaterly. 2003;27:425-78.

30. **Knittle K, De Gucht V, Maes S.** Lifestyle- and behaviour-change interventions in musculoskeletal conditions. Best Pract Res Clin Rheumatol. 2012 Jun;26(3):293-304.

31. **Knittle K, De Gucht V, Hurkmans E, Peeters A, Ronday K, Maes S, et al.** Targeting motivation and self-regulation to increase physical activity among patients with rheumatoid arthritis: a randomised controlled trial. Clin Rheumatol. 2015 Feb;34(2):231-8.

32. **Lorig KR, Ritter PL, Laurent DD, Plant K.** The internet-based arthritis self-management program: a one-year randomized trial for patients with arthritis or fibromyalgia. Arthritis Rheum. 2008 Jul 15;59(7):1009-17.

33. **Shigaki CL, Smarr KL, Siva C, Ge B, Musser D, Johnson R.** RAHelp: an online intervention for individuals with rheumatoid arthritis. Arthritis Care Res (Hoboken). 2013 Oct;65(10):1573-81.

34. **Iversen MD, Hammond A, Betteridge N.** Self-management of rheumatic diseases: state of the art and future perspectives. Ann Rheum Dis. 2010 Jun;69(6):955-63.

35. **Luca NJ, Feldman BM.** Health outcomes of pediatric rheumatic diseases. Best Pract Res Clin Rheumatol. 2014 Apr;28(2):331-50.

36. **Hersh AO, Salimian PK, Weitzman ER.** Using Patient-Reported Outcome Measures to Capture the Patient's Voice in Research and Care of Juvenile Idiopathic Arthritis. Rheum Dis Clin North Am. 2016 May;42(2):333-46.

37. **Nolte S, Elsworth GR, Newman S, Osborne RH.** Measurement issues in the evaluation of chronic disease self-management programs. Qual Life Res. 2013 Sep;22(7):1655-64.

38. **Gossec L, Dougados M, Dixon W.** Patient-reported outcomes as end points in clinical trials in rheumatoid arthritis. RMD Open. 2015 Apr 2;1(1):e000019,2014-000019. eCollection 2015.

39. **Gordon JE, Powell C, Rockwood K.** Goal attainment scaling as a measure of clinically important change in nursing-home patients. Age Ageing. 1999 May;28(3):275-81.

40. **Stinson J, Wilson R, Gill N, Yamada J, Holt J.** A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol. 2009 Jun;34(5):495-510.

41. **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 May;54(9):1453-61.

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

43. **Baksi AK, Al-Mrayat M, Hogan D, Whittingstall E, Wilson P, Wex J.** Peer advisers compared with specialist health professionals in delivering a training programme on self-management to people with diabetes: a randomized controlled trial. Diabet Med. 2008 Sep;25(9):1076-82.

44. **Pluut B.** Wat is patiënt gerichte e-Health? TVZ. 2016;4:19-21.

45. **van Limburg M, van Gemert-Pijnen JE, Nijland N, Ossebaard HC, Hendrix RM, Seydel ER.** Why business modeling is crucial in the development of eHealth technologies. J Med Internet Res. 2011 Dec 28;13(4):e124.

Quotes of young adults with Juvenile Idiopathic Arthritis (JIA) and adults
with Rheumatoid Arthritis (RA) on living with a rheumatic disease and self-management

> *“365 days a year”*
>
> *- Male, 23 years old, since 8 years JIA*

> *“Self-management for me is looking after yourself, to play an active role in management of your own disease”*
>
> *- Male, 36 years old, since 10 years RA*

> *“Self-management; what does it mean? I think it’s politically driven, not patient driven. To control costs, not to support or empower people who are sick”*
>
> *- Female, 61 years old, since 10 years RA*

> *“To me self-management is mainly about setting your own boundaries”*
>
> *- Female, 41 years old, since 10 years RA*

> *“...searching for balance? The continuous search for balance? It tires me out sometimes. Making choices is what self-management is about”*
>
> *- Female, 20 years old, since 16 year JIA*

> *“Self-management asks for an active role, to acquire information from other people, put things into your own hands and be part of decision making”*
>
> *- Male, 65 years old, since 12 years RA*

“Self-management to me is having an active role in what you are able and not able to do. I am using knowledge to manage my disease”

- Female, 37 years old, since 14 years RA

“You do not need to have an illness to use self-management”

- Female, 64 years old, since 4 years RA

“Choosing to do nothing: that is self-management for me..”

- Female, 18 years old, since 5 years JIA

“I like to be independent but that does not mean I should not ask for help. I just struggle to accept this. This is self-management for me”

- Male, 21 years old, since 9 years JIA

“Everybody applies self-management in their lives, one person more than the other, depending on the situation”

- Male, 65 years old, since 12 years RA

“I am trying to exercise as much as I can, that is part of self-management. There are some things that you can control yourself. Be positive mentally and exercise as much as possible”

- Female, 44 years old, since 4 years RA

SUMMARY

The aims of this thesis are to explore the perspectives, needs and preferences of young adult and adult patients with a rheumatic disease regarding self-management in order to develop self-management interventions, based on the outcomes of this exploration and to test feasibility and effectiveness of the interventions among these patients.

In the first part of this thesis, we focus on the youngest patient group*:* young adults in the age from 16-25 years old, suffering from a juvenile rheumatic disease who are making the transition from a paediatric rheumatology department to an adult rheumatology department. In the context of this transition, an increased involvement of this group in health care and self-management might prevent increasing disease-activity and worsening of symptoms, thus facilitating a successful transition to adult health care. Intriguingly, 'what young adults want and what their priorities are' and 'what health care professionals want and what their priorities are' may differ considerably. As we describe in *chapter 2,* to start with an attempt to understand the young adults' world, their problems and preferences, might bridge the gap between the needs of the young adult and the thoughts of the health care professional on self-management support. In this way, asking questions and young adults sharing their needs and preferences, provided a fundament for some innovations we developed with and for the young adults. We developed a transition outpatient clinic where young adults and their parents/caregivers meet their future rheumatologist at an early age and where a transition nurse guides and supports them during the transfer from child into adult care.

Also described in *chapter 2,* the young adults suggested to use the Internet for knowledge improvement, active involvement and support. Based on their suggestions, in close cooperation with the young adult group themselves, we developed an informative website (www.jongenreuma.nl) with written and video information and an online portal with access to the personal medical record, self-monitoring and eConsult functionalities, which we describe in *chapter 3*. According to the new Medical Research Council (MRC) guidance for developing and evaluating complex interventions, the applications were firstly tested on feasibility. Ease of use, usefulness and intention to use, were evaluated according to the Technology Acceptance Model (TAM). The results revealed that the young adults appreciated both applications as useful in dealing with problems in daily life and as easy to use. They reported that especially new medical information and feedback of health care professionals during consultations do motivate them to revisit the applications. Based on these results, the informative website and online portal have been integrated in usual care at the outpatient transition clinic. Next to that, the young adults expressed their needs for a training program in which they could actually practice self-management skills.

To answer the need of young adults with a juvenile idiopathic arthritis for self-management support, and to enhance their skills and knowledge in dealing with the consequences of having a rheumatic disease, we developed a peer guided self-management intervention, which can be followed online for 6 weeks, or face-to-face during a weekend. The theoretical fundament of the self-management intervention is the self-efficacy theory as described in *chapter* 4. The results of a feasibility study among young adults and peer leaders, using concepts of the TAM are presented. Both patient groups expressed to appreciate the face-to-face and online self-management support interventions as easy to use and as useful to dealing with problems in daily life. The peer leaders of the online patient group were surprised by the levels of intimacy and frankness of communication which were reached despite the fact participants and peer leaders of the online interventions could not see each other. Apparently, a safe atmosphere was created where participants could share and discuss their situation and problems.

Before implementing the intervention in daily practice, we investigated its effectiveness among young adults being treated at the transition outpatient clinic of the Rheumatology & Clinical Immunology departments of the University Medical Center Utrecht and the Erasmus Medical Hospital Rotterdam in the Netherlands. A randomized controlled trial with additional qualitative analyses of two interaction components of the intervention (goals and Chat) was conducted among 72 young adults with Juvenile Idiopathic Arthritis (JIA), as described in *chapter 5.* No differences on self-efficacy, quality of life and self-management items were shown between the intervention group who followed the online self-management intervention and the control group who received usual care. However, qualitative analysis within the intervention group did reveal indeed that participants achieved their personal defined goals. Adherence to the intervention was high; appreciation of the own learning experiences and personal achievements in the online training was rated positively. Analysis of the Chat showed *modelling* and *sharing experiences* as the most used interactions. Although encouraging, more research is needed to measure possible added values of this intervention compared to usual care.

In general, the diversity of patient populations, the different theoretical foundations as well as heterogeneity of the self-management interventions used makes it challenging to demonstrate any effectiveness with the most commonly used metrics. We therefore translated the Health Education Impact Questionnaire (heiQ), which was developed in Australia, to capture the patients' perspective in the evaluation of self-management, into Dutch. Results of its psychometric features, presented in *chapter 6,* may illustrate that the Dutch heiQ has good psychometric properties in patient groups with rheumatoid arthritis, atopic dermatitis, food allergy and asthma. The psychometric properties of the Dutch translation were comparable with the English, German and French versions, which provide evidence that the heiQ conceptualizes a broad range of self-management skills in a consistent way across chronic conditions, cultures and languages.

With the expansion of the online intervention for young adults, adults also expressed their need for online self-management support. That brings us to the second part of this thesis with a different age group. In order to meet the needs for support of adult patients with a rheumatic disease, we investigated preferences for structure and content needs to develop an online self-management intervention. The results are detailed in *chapter 7*. In terms of structure, participants prefer a tailored intervention which also takes into account their personal needs and circumstances. They prefer to be enabled to follow the intervention at their own pace in their own time. They also prefer to be guided by a trained patient, rather than a health professional. In addition to gaining knowledge about disease and treatment, participants identify as important content of the intervention the clusters about increasing competences in self-efficacy, dealing with societal procedures and working together with health care providers, handling disease fluctuations, psychological distress and maintaining and increasing physical activity. Also clusters on relational needs towards family, friends, children and co-workers were expressed to be valuable.

A basic assumption about self-management is that when the intervention is customized to the individual needs and situation of the patient, the patient will be more motivated, adhere better and benefit more and for a longer time. Based on the results of the comprehensive assessment of preferences and needs among adult patients as described in in *chapter 7*, a web-based peer-guided and tailored self-management intervention for adults was developed and tested on feasibility among adults with a rheumatic disease, using the concepts of the TAM, as shown in *chapter 8.* The participants evaluated the intervention positively on usefulness and ease of use. Participants stated that they were likely to participate also in daily practice since they are now familiar with content and structure. In this chapter we also present the study design of the RCT which will be conducted in due time. Consistent with the self-determination theory – the theoretical fundament of the online intervention - progress towards the personal goal will be the primary outcome measure of the intervention.

In *chapter 9*, we reflect on the process of redeveloping care and the development of self-management interventions in this thesis. We experienced that exploring the needs of self-management support among young and adult patients with a rheumatic disease, the development of self-management interventions tailored to the individual needs, circumstances, life stages and problems of the individual patient, is essential and inevitable. But we also recognized that a focus on the patient alone is not enough to implement these interventions effectively and sustain patient centered care in daily practice. Being health care professionals, we also need to be supported and to gain and regain knowledge and skills continuously. Even of more importance, we should be aware of our own values towards concepts of patient centered care. A daily reflection on shared answers of patients and health care professionals to the question 'what is most important to you?' might be helpful to processes of redeveloping health care.

In addition to summarizing the findings, in *chapter 10* (the general discussion), we also reflect on three themes: *exploring needs for self-management support, development and feasibility* and *evaluating self-management interventions*. We allowed ourselves to end this chapter with some recommendations.

Reflection on the theme *exploring needs for self-management support* taught us that the needs of young adults with a rheumatic disease appeared to be strongly related to their life stage and thereby their maturing processes. This might also explain why young adult are mainly living in the moment, with less focus on future and long-term consequences of actual behavior.
Adult patients with a rheumatic disease expressed that supportive care should integrate their individual life experiences, questions and problems. In contrast to young adults, their focus is more related to being sick and dealing with its consequences, in their social life as in their personal relationships.

Reflection on the theme *development and feasibility* made us realize that a systematic, person based approach is conditional for the development of self-management interventions, where also patient involvement and collaboration are important factors to bridge the gap between real life and the medical world. The results of this collaboration on content, layout, language and style made our interventions more applicable, and may indeed have influenced the positive evaluation of the usefulness of the applications.
Considering feasibility testing, we used the Technology Acceptance (TAM) where we focused mainly on perceived usefulness and ease of use. Reflection on these decisions raised questions whether focusing on these two factors alone might be subject to some limitations. Also age, social influence and motivation may influence as well usefulness as ease of use. So, with gained insight, when using feasibility testing, we now recommend the Unified Theory of Acceptance and Use of Technology (UTAUT). In this latest version of the TAM, factors age, social influence, voluntariness of use and effort-expectancy are included.

Considering *evaluating self-management interventions,* as shown in *chapter 5*, focusing on knowledge and skills alone appears not to be enough. Also individual needs and circumstances of the young adult patient should be taken into account. Therefore, an extension of the theoretical fundament of the online intervention for young adults with elements of the self-determination theory, as we did for the adult version, may be helpful to improve acceptability and effectiveness.

We end the discussion with recommendations to a) study experiences and self-management in a broad range of participants, including those without particular interest in online communication; b) exploring the perspectives of the peer trainers themselves and their development with regard to self-management and c) create a continuous, on-going model of (financial) support for post development and sustainability of the eHealth applications, collaborating with partners and stakeholders.

SAMENVATTING

In dit proefschrift zijn de behoeften en voorkeuren van jong volwassen en volwassen patienten met reuma met betrekking tot zelfmanagement support verkend. Op basis van deze verkenning zijn, in nauwe samenwerking met beide groepen, interventies ontwikkeld. De zelfmanagementinterventies zijn onderzocht op haalbaarheid door zowel jongeren als volwassenen en op effectiviteit onder jongeren met Juveniele Idiopathische Artritis (JIA).

In het eerste deel van het proefschrift focussen we op de jongste patiëntgroep: jongvolwassenen in de leeftijd van 16-25 jaar, gediagnosticeerd met een reumatische aandoening die de transitie maken van de kindergeneeskunde naar de zorg voor volwassenen. In deze periode kan het actief betrekken van de jongere bij zijn/haar behandeling en zelfmanagement een positieve invloed hebben op een succesvolle transitie en verergering van de ziekteactiviteit voorkomen. Echter, wat jongeren willen, verwachten en wat hun prioriteiten zijn in deze periode van hun leven en wat zorgverleners willen en verwachten in deze fase van behandeling kan behoorlijk verschillen.

In *hoofdstuk 2* verkennen we de voorkeuren voor zorg en zelfmanagementondersteuning onder jongeren met een reumatische aandoening. Dit hebben we gedaan door de dialoog aan te gaan met jongeren en op basis van de uitkomsten van deze gesprekken zijn interventies ontwikkeld. Zo is er een transitiepolikliniek ontworpen en geïmplementeerd in de dagelijkse zorg op de poliklinieken van de kinderreumatologie en de volwassenreumatologie, waar de jongere en zijn/haar ouders begeleid worden door een transitieteam, bestaande uit een kinderarts, reumatoloog en een transitiecoördinator. Ook adviseerden jongeren digitale hulpmiddelen te gebruiken voor kennisvergroting, betrokkenheid en ondersteuning. Als antwoord daarop hebben we, in nauwe samenwerking met de doelgroep, een informatieve website gemaakt (www.jongenreuma.nl) waarop informatie, real-life video's, tips en ervaringsverhalen worden gedeeld. Daarnaast is er een online portaal ontwikkeld waarmee de jongere via een beveiligde verbinding zijn/haar persoonlijk medisch dossier kan inzien. Ook kan de jongere, door het zenden van een e-consult, digitaal contact hebben met zijn/haar behandelteam, de conditie- en symptomen monitoren, zijn/haar medicatie overzicht en laboratoriumuitslagen inzien.

Conform de richtlijn van de Medical Research Guidance (MRC) voor het ontwikkelen en evalueren van complexe interventies, zijn beide online hulpmiddelen onderzocht op haalbaarheid voordat deze in de praktijk zijn geïmplementeerd. Hiervoor zijn de concepten van het Technology Acceptance Model (TAM); nut, gebruikersgemak en intentie tot gebruik, geëvalueerd in *hoofdstuk 3.* Uit het haalbaarheidsonderzoek blijkt dat jongeren de website en het portaal nuttig vinden voor het omgaan met problemen in het dagelijks leven en het gebruikersgemak positief beoordelen. Vooral de video's waarin andere jongeren met reuma ervaringen delen, worden positief gewaardeerd, naast het digitaal toegang hebben tot het eigen medisch dossier. Volgens de jongeren zal het gebruik van de website en het portaal toenemen als er regelmatig nieuwe informatie op de website geplaatst wordt en feedback door de zorgverlener gegeven wordt op de inhoud en het gebruik van het portaal. Op basis van deze resultaten, zijn de informatieve website en het portaal aangepast en geïntegreerd op de transitiepolikliniek. Verder gaven jongeren aan dat zij behoefte hadden aan een digitaal hulpmiddel waarmee zij zelfmanagementvaardigheden kunnen leren en vergroten.

Om aan deze behoefte voldoen, hebben we, op basis van de Self-efficacy theory van Bandura, samen met jongeren een zelfmanagementinterventie ontwikkeld. Deze interventie, ReumaUitgedaagd! genaamd, wordt begeleid door jongeren die zelf een vorm van reuma hebben en kan zowel in een weekend als online gevolgd worden. Het doel van ReumaUitgedaagd! is het vergroten van kennis en vaardigheden in het omgaan met de gevolgen van het hebben van een reumatische aandoening. In *hoofdstuk 4* worden de resultaten van de haalbaarheidsstudie gepresenteerd. Deze is gehouden onder jongeren en jongeren trainers van de weekend en de online versie van ReumaUitgedaagd! waarvoor de concepten van het Technology Acceptance Model (TAM) zijn geëvalueerd. Zowel de jongeren als de jongeren trainers vonden ReumaUitgedaagd! nuttig voor het omgaan met problemen in het dagelijks leven en een aanvulling op de gebruikelijke zorg. Ook zouden ze ReumaUitgedaagd! aanraden aan andere jongeren met reuma. De jongeren trainers van de online versie waren verrast door het niveau van intimiteit en openhartigheid onder de deelnemers. Deze lijkt voldoende veiligheid te bieden waardoor deelnemers, ondanks dat ze elkaar niet kennen en zien, hun situatie en problemen durven te bespreken met elkaar.

In *hoofdstuk 5* zijn de resultaten van een gerandomiseerde, gecontroleerde studie met 3 meetmomenten (op baseline, 3 maanden en 6 maanden na randomisatie) naar de effectiviteit van de online versie van ReumaUitgedaagd! op self-efficacy, kwaliteit van leven en zelfmanagement beschreven. Hiervoor werden 72 jongvolwassenen met Juveniele Idiopathische Artritis (JIA) geincludeerd en random toegewezen aan een interventie- of een controlegroep. Alle deelnemers aan dit onderzoek ontvingen de gebruikelijke zorg op een transitiepolikliniek; de interventiegroep volgde aanvullend ReumaUitgedaagd!. Naast een kwantitatieve analyse is er een kwalitatieve analyse uitgevoerd naar twee onderdelen van de interventie (doelen en Chat) onder de deelnemers van de interventiegroep. Er werden geen significante verschillen op self-efficacy, kwaliteit van leven en zelfmanagement aangetoond tussen de deelnemers van de interventie- en de controlegroep. Door de deelnemers van de interventiegroep werden in totaal 55 doelen geformuleerd voornamelijk gericht op het behouden en vergroten van balans, het herkennen en stellen van grenzen, communicatie, omgaan met de gevolgen van reuma en onbegrip. Modelling en delen van ervaringen werden als meest voorkomende interactie binnen de Chat geïdentificeerd. De adherence met ReumaUitgedaagd! was groot en tevredenheid met de eigen leerervaringen en de interventie werden als positief en waardevol gewaardeerd. Meer onderzoek is nodig om de aanvullende waarde van ReumaUitgedaagd! aan de gebruikelijke zorg aan te tonen.

In *hoofdstuk 6* wordt de Australische Health Education Questionnaire (heiQ) vertaald in het Nederlands en onderzocht op validiteit, betrouwbaarheid en stabiliteit. Het doel van de heiQ is om zelfmanagementvaardigheden van en educatie voor patiënten met een chronische aandoening te evalueren. De vragenlijst is vertaald en beoordeeld op begrijpelijkheid en bruikbaarheid onder volwassen patiënten met artritis, eczeem en voedselallergie. Daarna zijn de psychometrische kenmerken van de Nederlandse vertaling bestudeerd onder patiënten met artritis, eczeem, voedselallergie en astma (n=286). De resultaten laten zien dat de vragenlijst als begrijpelijk, acceptabel en gebruikersvriendelijk wordt gewaardeerd. De psychometrische kenmerken zijn stabiel en robuust en vergelijkbaar met de Engelse, Duitse en Franse vertalingen. De Nederlandse heiQ lijkt hiermee een geschikte vragenlijst om een breed scala van zelfmanagementvaardigheden op een consistente manier te evalueren.

Met de verspreiding van de online zelfmanagementinterventie onder jongvolwassenen met reuma bleek dat ook volwassenen behoefte hadden aan een online zelfmanagementinterventie. Dit brengt ons bij het tweede deel van dit proefschrift dat focust op een andere leeftijdsgroep: de volwassen patiënt met een reumatische aandoening.

Om inzicht te krijgen in de voorkeuren en behoeften aan zelfmanagementondersteuning bij volwassenen met reuma, hebben we een behoefteonderzoek naar de structuur en de inhoud van een online zelfmanagementinterventie verricht in *hoofdstuk 7*. Tijdens dit proces hebben we intensief samengewerkt met patiënt partners en zijn er vier stappen uitgevoerd om tot het

uiteindelijke resultaat te komen: een online focus groep interview, consensus bijeenkomsten, een kaart sorteeropdracht en hiërarchische clusteranalyse.
Uit de resultaten van deze stappen blijken deelnemers behoefte te hebben aan een zelfmanagementinterventie op maat, passend bij hun persoonlijke omstandigheden, vragen en problemen. Zij willen de interventie individueel doorlopen, in eigen tempo, maar tijdens de interventie wel digitaal contact hebben met andere mensen die de interventie ook volgen. Daarnaast verkiezen zij een expert patiënt als trainer boven een hulpverlener. In de hiërarchische clusteranalyse werden 55 behoeften geïdentificeerd ten behoeve van de inhoud van de interventie. Deze behoeften konden, op basis van een dendrogram, worden samengebracht tot 11 inhoudsclusters waarbij deelnemers, naast het cluster 'kennis over ziekte en behandeling', de clusters 'zelfeffectiviteit', 'omgaan met reuma en je sociale omgeving (bijvoorbeeld hulp vragen aan je partner of collega's)', 'samenwerken met zorgverleners', 'omgaan met grilligheid van de ziekte', 'managen van emoties' en 'het behouden en vergroten van fysieke activiteit' als belangrijk waarderen. De overview van behoeften en voorkeuren voor structuur en inhoud kan gebruikt worden om de online zelfmanagementinterventie te ontwikkelen. Daarnaast kan deze overview de deelnemer helpen om zijn doelen (wat wil ik leren/verbeteren) te formuleren en de interventie daarop aan te passen.

Op basis van de resultaten van het behoeftenonderzoek in *hoofdstuk 7*, hebben we, in samenwerking met patiënt partners , een online zelfmanagementinterventie ontwikkeld (ReumaUitgedaagd! online genaamd) voor volwassenen met reuma, waarbij de deelnemer op basis van zijn eigen doelen de interventie doorloopt. De deelnemer wordt hierbij begeleid door een trainer die zelf reuma heeft en is opgeleid door Work21 (www.work21.nl), in samenwerking met het Reumafonds en het UMC Utrecht. Voor digitaal contact met andere deelnemers, kan de deelnemer gebruik maken van een forum. In *hoofdstuk 8* is de haalbaarheid van de ReumaUitgedaagd! online geëvalueerd onder volwassenen met reuma (n=23) met behulp van de concepten van het Technology Acceptance Model (TAM). De deelnemers waardeerden de interventie positief op nut en gebruikersgemak en 21 van de 23 deelnemers zou de interventie ook aanraden aan andere mensen met reuma. Verder wordt in *hoofdstuk 8* het design van een gerandomiseerde, gecontroleerde studie naar de effectiviteit van ReumaUitgedaagd! online voor volwassenen beschreven. Consistent met het theoretisch fundament van deze interventie – de Self-Determination theorie van Ryan & Deci – is verbetering van de persoonlijke doelen van de deelnemer de primaire uitkomst maat naast pijn, ziekte activiteit, zelfmanagement, kwaliteit van leven en verzuim.

In *hoofdstuk 9* wordt gereflecteerd op het proces van ontwikkeling van zorg en zelfmanagementinterventies voor jongeren en volwassenen met reuma. Het verkennen van behoeften van zelfmanagementondersteuning en het ontwikkelen van interventies die toegesneden zijn op deze behoeften, in nauwe samenwerking met de doelgroepen zelf, is noodzakelijk en onvermijdelijk. Maar het is niet alleen de patiënt die ondersteuning kan gebruiken; om zorg op maat te leveren en de patiënt centraal te stellen heeft ook de zorgverlener kennis en vaardigheden nodig. Maar wellicht nog belangrijker; het succes hangt voor een groot deel af van de waarde, de overtuiging en de intrinsieke motivatie van de zorgverlener. Om de patiënt centraal te stellen en zorg op maat te leveren, is samenwerking en communicatie met de patiënt van essentieel belang. Een frequente reflectie op het antwoord van zowel de patiënt als de zorgverlener op de vraag 'wat is het meest belangrijke voor jou?' kan wellicht helpen om verandering en afstemming van zorg en behoeften te bewerkstelligen.

In *hoofdstuk 10,* de algemene discussie, zijn de bevindingen van alle hoofdstukken samengevat. In dit hoofdstuk wordt ook gereflecteerd op drie thema's: 1) het verkennen van behoeften voor zelfmanagementondersteuning, 2) ontwikkeling en haalbaarheid en 3) het evalueren van zelfmanagementinterventies. Hoofdstuk 10 eindigt met een aantal aanbevelingen.

Reflectie op het thema “verkennen van behoeften voor zelfmanagementondersteuning” heeft ons geleerd dat de behoeften van jongvolwassenen met een reumatische ziekte sterk samenhangen met het natuurlijke ontwikkelingsproces naar volwassenheid. Jongeren zijn meer gericht op het ‘hier en nu’, willen vooral niet verschillen van andere leeftijdgenoten en gelabeld zijn als ziek. Zij zijn minder bezig met de effecten van reuma op latere leeftijd en overzien niet altijd de consequenties van hun gedrag op lange termijn. In tegenstelling tot jongeren richten de behoeften van volwassenen zich meer op omgaan met de ziekte in het dagelijks leven en de consequenties daarvan, zowel op hun sociale omgeving als hun persoonlijke relaties.

Reflectie op het thema “ontwikkeling en haalbaarheid” heeft ons bewust gemaakt dat een systematische aanpak, met oog voor de diversiteit van personen, essentieel is. Het betrekken van en samenwerken met patiënten, niet alleen bij de ontwikkeling van de interventies maar ook bij alle fasen van het onderzoek, zijn belangrijke factoren geweest om de kloof tussen patiënt (en de wereld thuis) en behandelaar (en de wereld in het ziekenhuis) te dichten en daarmee haalbare en acceptabele interventies te ontwikkelen. Voor het evalueren van de haalbaarheid van de interventies, hebben we in dit proefschrift gebruik gemaakt het Technology Acceptance Model (TAM) waarbij wij ons vooral hebben gefocust op de twee concepten nut en bruikbaarheid. Achteraf realiseren we dat enkel focussen op deze concepten wel een beperking met zich mee brengt: ook leeftijd, sociale omgeving en motivatie beïnvloedt de haalbaarheid van een interventie. Op basis van deze reflectie raden we nu de Unified Theory of Acceptance and Use of Tehcnology (UTAUT) aan. In deze nieuwe versie van de TAM zijn deze factoren opgenomen.

Reflectie het thema “evaluatie van zelfmanagementinterventies” leert ons in *hoofdstuk 5* dat enkel focussen op kennis en vaardigheden om zelfmanagement te vergroten niet voldoende is. De individuele behoeften en omstandigheden van de jongere zijn eveneens van belang, naast de factor motivatie. Op basis hiervan concluderen we dat een uitbreiding van het theoretische fundament van de zelfmanagementinterventie voor jongeren noodzakelijk, zoals we al bij de volwassen online interventie hebben toegepast. Deze uitbreiding kan nuttig zijn om de effectiviteit van de interventie voor de jongeren te vergroten.

Het proefschrift eindigt met een drietal aanbevelingen. Deze betreffen: a) zelfmanagement verder exploreren onder een grotere, diverse groep, waaronder mensen die geen bijzondere belangstelling hebben voor online communicatie, b) een studie naar de perspectieven van de (jongeren) trainers op zelfmanagement en hun ontwikkeling ten behoeve van het trainerschap en c) een plan, samen met patiënten, stakeholders en partners, ontwikkelen waarbinnen post-ontwikkeling van de interventies, duurzaamheid en de (financiële) ondersteuning thema’s zijn.

DANKWOORD

“Surround yourself with the dreamers and the doers, the believers and thinkers, but most of all, surround yourself with those who see the greatness within you, even when you don’t see it yourself”

Dit proefschrift was niet tot stand gekomen zonder *the dreamers, the doers, the believers* en *the thinkers*. Zowel in mijn privé- als in mijn werksituatie ben ik omringd geweest door mensen die in mijn geloofden en mij hebben geholpen. Een dankwoord aan hen is zeker op z'n plaats.

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Surround yourself with those who see the greatness within you, even when you don't see it yourself.
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CURRICULUM VITAE

Judy Ammerlaan was born on the 18th of January 1966 in de Bilt, the Netherlands. In 1984 she finished secondary school at the St. Gregorius College in Utrecht and obtained a bachelor's degree in nursing (HBO-V) at the Hogeschool Utrecht in 1988. During and after her study, she worked as a nurse at the Willem Arntsz Hoeve (a psychiatric clinic) in Den Dolder, the internal diseases and AIDS unit of the Amsterdam Medical Center in Amsterdam and the rheumatology department of the University Medical Center Utrecht. In 1991 she obtained the certificate of nursing manager from the Hogeschool Midden Nederland te Leusden. She worked as a team manager of the rheumatology and internal diseases clinic at the University Medical Center Utrecht for six years and then obtained a job as clinical nurse specialist at the outpatient rheumatology clinic. In 2002 she enrolled in the Health Sciences program at the Utrecht University, majoring in Nursing Sciences. For her master thesis, she investigated the perspectives of patients and health care professionals on patient centered care at the rheumatology outpatient clinic under the supervision of Dr. Jaap van der Bijl and Dr. Claudia Gamel. She obtained her master's degree (cum laude) from the Utrecht University in 2006. During the last year of her study, she started to work as a transition rheumatology nurse for young adults with a rheumatic disease and as nurse researcher at the University Medical Center Utrecht. Based on the needs and preferences of this group, she developed and implemented, together with the two multidisciplinary teams of the childrens and adult rheumatology departments, the Dutch Rheumatism Patient League and young adults themselves, a transition outpatient clinic and three (eHealth) interventions, all described in this thesis. The transition outpatient clinic aim to to create a safe chain of care between the child and adult hospital for the young adult, his/her parents-caregivers and the multidisciplairy team. The (eHealth) interventions aim to empower the young adults and to support them in their self-managagement.

Based on the needs and preferences for self-management support of adult patients with rheumatism, she developed and implemented together with patient partners, web technicians, health professionals, communication experts and the Dutch Arthritis Foundation a peer-guided online self-management version of ReumaUitgedaagd! for this group as well.

Judy teaches at the nursing rheumatology education programme of the St. Antonius Academy in Nieuwegein and is a member of the research group of the Dutch Rheumatology Nurses Association (V&VN Reumatologie). Judy lives with Bob Wielinga in Leerdam and they have 2 kids: a son Jesse (1994) and a daughter Kristy (1998).