Self-management in transition for patients with a rheumatic disease

Margot Walter

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Self-management in Transition for Patients with a Rheumatic Disease

Zelfmanagement in transitie voor patiënten
met een reumatische aandoening

Proefschrift

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CHAPTER 1

General Introduction

INTRODUCTION

Rheumatic disease

Rheumatic disease (RD) is a chronic condition which predominantly affects the joints and which can have a pervasive impact (1-3). A distinction is made between inflammatory RD (e.g. rheumatoid arthritis, arthritis psoriatic, juvenile arthritis, SLE) and non-inflammatory RD (osteoarthritis, fibromyalgia). The predominant symptoms are swollen joints, pain and stiffness, and physical disability may be the result (4).

In the past decades the treatment of inflammatory RD has much improved due to treat-to-target and tight-control strategies, as well as biological treatment (5, 6). Inflammatory RD should be treated in the most effective way to prevent radiographic damage and to increase physical capacity (7,8). The purpose of intensive treatment is to reach remission. Many studies have shown good results concerning remission and structural radiographic damage of the joints with the use of treat-to-target and tight-control strategies (5,6,9).

Still, patients with a RD may be burdened by, for example physical and psychosocial consequences of living with the condition (10,11). Besides pain, stiffness and disabilities, RD can have a pervasive impact on a patient's psychology and social environment; like limited work participation (12), family activities or social activities (13), sports and simply enjoying life as it is. This could lead to a significantly reduced quality of life (10).

Receiving the diagnosis of an RD or living with a chronic rheumatic disease is a life-altering event (14). How someone copes with it depends for instance on one's individual traits, the disease activity or the extent of social support (15, page 10).

Juvenile idiopathic arthritis

RDs do not solely affect elderly; children may suffer from a RD as well. The condition then is referred to as juvenile idiopathic arthritis (JIA), a clinically heterogeneous group of arthritis conditions of an unknown cause, with onset before the age of 16 (16). The treatment options are in many ways similar to those used in adult RD. The treatment goals are suppression of inflammation, pain management and the prevention of joint destruction (4, page 410). The impact and burden of having a JIA can be immense. The young people (YP) have to deal not only with the physical consequences such as pain, stiffness, and disability, but also with the effect on the psychosocial wellbeing. YP with a JIA are faced with an extra task: giving the disease a place in their self-image, which can make them extra vulnerable. They are always pressured to find a balance between growing up with a disease and the "normal life" (17) and becoming an independent person can be delayed (4, page 415). It is important to support the YP in this process of changes.

Transition

Health professionals should realize that patients are in a process of transition, e.g. growing up, from a healthy person to a patient with a chronic illness or in a transition from having a disease flare-up. Transition is defined as "the change from one situation, form, or state to another, which might temporally disrupt normal life and ask for adjustments" (18). During these transitions, patients may have a substantially greater need for care and support from health professionals. They have to adjust their lives to cope with the new challenges. A successfully completed transition is one in which the patient has found a personal answer to a situation and has rediscovered a new life balance – and thus is able to self-manage again.

Meleis, a prominent nurse sociologist, developed a nursing transition theory which considers the nature of transition; types, patterns, properties of transition, the conditions; facilitating or barriers for successful transition, the pattern of response (process indicators and outcome indicators) and the related nursing interventions (18). These interventions are intended to promote healthy transitions. Self-management is an important concept within the transition. Figure 1 depicts the relations between the different aspects of the transition process.

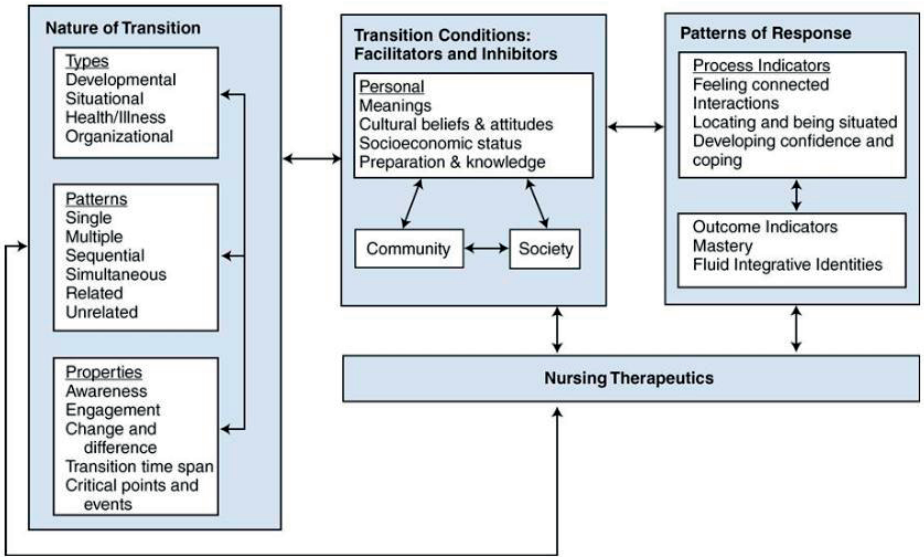

Figure 1 Transition model, middle-range transitions theory [18]

Transition is a far-reaching process, where patients have a tendency to be more vulnerable for risks that could cause health issues. Typical problems that occur if transition of care for young people is not well managed are a high risk for lack of self-management skills and independence, missed appointments or even dropout of care, non-compliance

with medication and ultimately poor disease outcome (19-21). It is important to recognize in which phase of transition patients are and which support of self-management is needed.

Self-management

The purpose of self-management is to get hold on one's disease in a way that allows to reach the best possible quality of life. Self-management is seen as "empowering the individual's ability to deal with symptoms, treatment, physical and social consequences and lifestyle changes inherent in living with a chronic disease" (22). For patients with an RD, self-management is the effort to find a balance between the demands of the disease and the activities of daily living (23). Although they are expected to take an active role (24), they generally are not expected to self-manage their disease entirely independently; they will need support from health care professionals (25).

Self-management support can be defined as the provision of interventions to increase patients' skills and confidence in managing their chronic condition (26). The components of self-management support include education / information, symptom management by self-monitoring, management of psychological consequences, social support, communication and coping (22). A range of self-management interventions have been studied, however there is limited empirical evidence, based on lived experiences (27,28) of the support that patients with an RD need. Most of the studied self-management interventions provide education, sometimes supplemented with cognitive behavioural therapy and goal-setting. The authors of a recent review on self-management support concluded that the most effective intervention was the one that was focusing on the intrinsic motivation and self-efficacy, while providing education only was the least effective (29). They further concluded that information provision should be tailored to a person's individual needs.

Health professionals have a shared responsibility in facilitating patients' self-management. It should not be forgotten, though, that patients wish to be seen as the daily life experts (30). This wish fits well with the concept of self-monitoring. One of the activities of self-monitoring is self-measurement of symptoms and vital signs obtained through measurements, recordings or observations. Self-monitoring is an essential part of self-management and can help to achieve more control over one's life (31). Over the past decade, new technology has been developed that can support self-management and self-monitoring, including EHealth or smartphone apps. These apps perfectly fit the idea that patients prefer a more active role in self-management (32). Both, self-monitoring and e-Health, are investigated in this thesis. But, when designing effective interventions for self-management support there has to be a good understanding of patients' (unmet) needs.

AIMS AND OUTLINES

The main rationale for this thesis is to gain a better understanding of how self-management support interventions can improve the care for adult and young patients with a RD. We posed the following questions:

1. What do patients with an RD prefer with regard to self-management support and what are the unmet needs that might impede self-management (support)?
2. What self-management support interventions can improve the care for adult and young patients with an RD?

To answer these questions the following studies were conducted.

In the study reported in chapter 2, we explored the unmet needs and the self-management of adolescents (and their parents) in the phase of transition and developed a clinical transition pathway. The study reported in chapter 3 made use of focus-groups and interviews to explore what support in self-management patients with various RD preferred to receive. Fatigue is common problem for many patients and a constraining factor for self-management. In the study reported in chapter 4, we therefore investigated aspects of fatigue more in depth. As patients and health professionals have different opinions on disease activity, we tried to unravel this discrepancy through focus-groups. (chapter 5).

In the study reported in chapter 6 we investigated whether it is possible to develop a tool for self-monitoring the disease activity by patient-reported outcomes. We developed a smartphone app and performed a pre- and post-study (chapter 7) to find out whether use of this app would increase patients' self-management behaviour. The clinical transition pathway to support the transition process and self-management for adolescent patients was evaluated in a quantitative study reported in chapter 8.

The main findings of these studies are discussed in chapter 9.

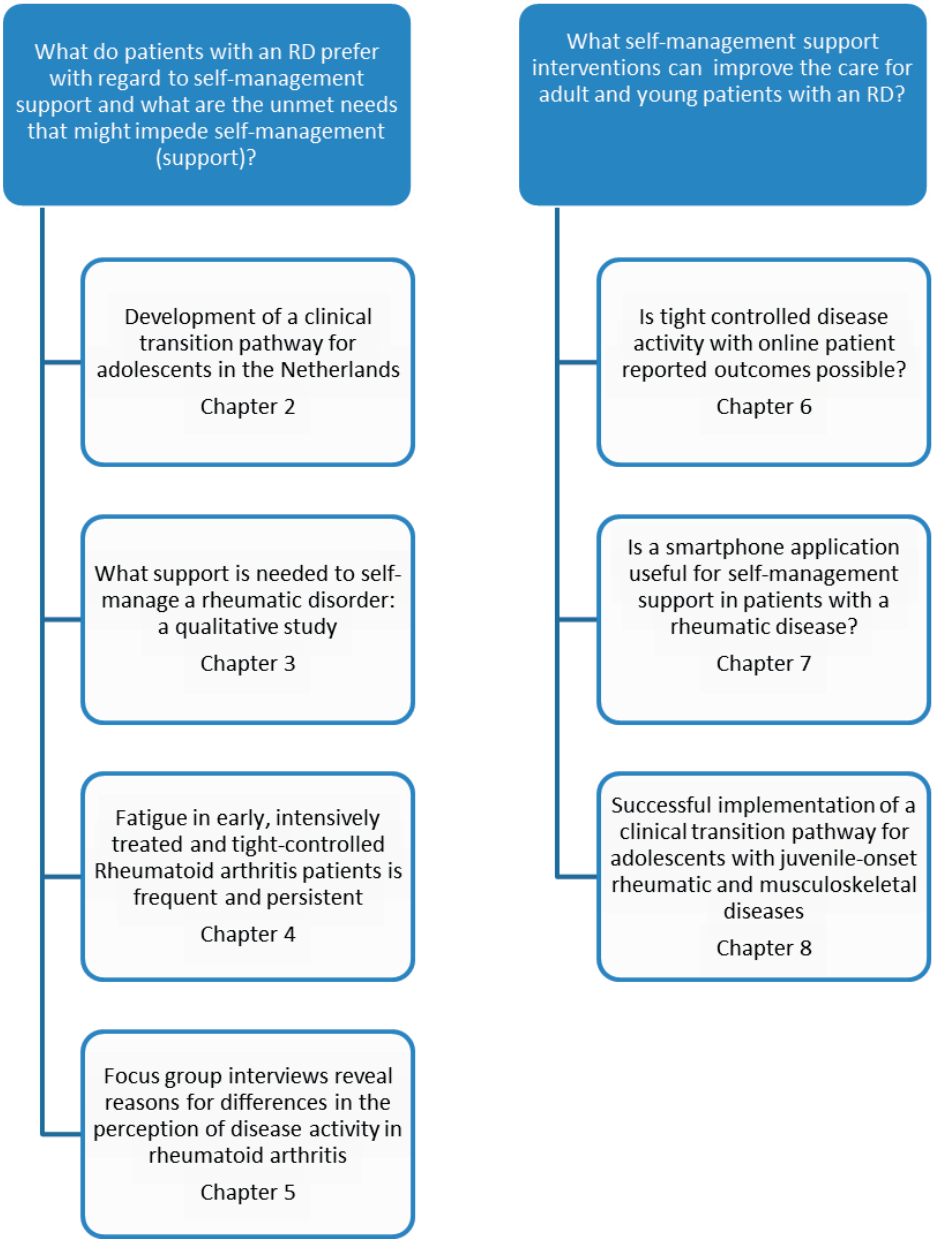

Figure 2 Overview of the outline

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CHAPTER 2

Development of a clinical transition pathway for adolescents in the Netherlands

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ABSTRACT

Aims

To explore how young people with juvenile-onset rheumatic and musculoskeletal diseases (jRMDs) and their parent(s) experience care during preparation for the upcoming transfer to adult services, and to develop a clinical transition pathway.

Methods

A survey was conducted with 32 young people aged between 14 and 20 years with jRMDs, and their parents (*n*=33), treated at the department of paediatric rheumatology in a tertiary care children's hospital in the Netherlands.

Results

More than 30% of young people would have liked to discuss topics such as educational and vocational choices in a clinic, but did not get the opportunity. Preparation for transition was poor as was training in self-management skills. One third of parents had feelings of anxiety about the upcoming transfer. Results from the survey and evidence-based principles of transitional care were used to develop the clinical transition pathway. The pathway focuses on starting transition early, developing self-management skills, joint consultations and supporting parents in giving young people control of their clinical care.

Conclusion

Current care does not meet the needs of young people in the process of transition to adult rheumatology services. The clinical transition pathway developed as a result of the study is a tool that may improve this process.

INTRODUCTION

Transition is the planned transfer of care for young people with a chronic disease from children's to adult services [1-3]. It involves a period of preparation, the transition process, during which young people are trained in the skills necessary for a successful transfer [2,3]. If transition is not well managed, however, it will have an adverse effect on the quality, continuity and efficiency of care [4].

Several transition programmes and guidelines have been developed over the past decade to support the process [2,5-8]. These transition programmes result in better quality of life, increased knowledge and satisfaction for adolescents, and better pre-vocational readiness [5,9,10]. However, implementation of transitional care is not widespread in routine clinical practice for young people with chronic diseases, including juvenile-onset rheumatic and musculoskeletal diseases (jRMDs) [2,6,11]. Despite the availability of transition programmes for these young people, half of them do not make a successful transfer and they are often inadequately supported during the transition process [2,8,12,13].

A clinical transition pathway is a tool to make implementation of transition programmes into routine clinical practice easier. Reviews on transition and transition guidelines have reported several important points that need to be incorporated into care. Overarching principles are planned, developmentally appropriate and holistic care. A transition coordinator is essential, as is good collaboration between children's and adult's services [2,8].

During the development process for a specific clinical pathway, it is important to investigate the status of care and identify potential problems and bottlenecks experienced by patients [14]. Surveys that explore patients' preferences and opinions can be used for this purpose [15].

AIMS

The aims of this study were to:

- Explore how young people with jRMDs and their parent(s) experience care during preparation for the upcoming transfer to adult services.
- Develop a clinical transition pathway.

METHODS

All young people with jRMDs aged 14-20 years, and their parents, treated at the department of paediatric rheumatology in a tertiary care children's hospital in the Netherlands were invited to participate. Separate surveys were developed for patients and their parents based on evidence from the literature and discussions with a paediatric rheumatologist (SK), a rheumatologist from the adult care team (RD) and two specialist rheumatology nurses from the paediatric (AD) and adult care team (MW). The survey included four open and 35 multiple-choice questions (MCQs) for patients and five open and 13 MCQs for parents. The surveys included questions about demographics, topics that should have been discussed in clinic but were not, experiences of preparation for transfer and achievement of self-management skills. At the time the study was conducted, the department of paediatric rheumatology had no specific policy or protocol about the process of transition.

The development of the clinical transition pathway was based on the principles of Vanhaecht and Sermeus [14,16]. This method consists of 11 steps in four phases: the plan-do-check-act cycle [17]. Results from the patient and parent surveys were incorporated into the clinical transition pathway.

Data analysis

Descriptive statistics were used to analyse the results from the MCQs in the surveys. Answers to open questions were labelled and coded; these codes were grouped into concepts. Two researchers (SK, MW) independently analysed the open questions. Differences were discussed until consensus was reached.

Ethical considerations

All patients and parents gave informed consent and the ethical review board of the hospital approved the study.

RESULTS

A total of 48 surveys were sent to patients and a total of 48 were sent to parents. The overall response rate was 67% (*n*=32) patients and 69% (*n*=33) parents. Patients' mean age was 17 years (range 14-20) with a mean disease duration of 6.2 years (range 0.5-16). Most (72%) had juvenile idiopathic arthritis (Table 1).

Table 1 Demographics of participating with young people with juvenile rheumatic diseases

Respondents	***n*=32**
Age (mean, range)	17 (14-20)
Female *n* (%)	22 (69)
Diagnosis *n* (%):	
• Juvenile idiopathic arthritis	23 (72)
• Childhood-onset systemic lupus erythematosus	6 (19)
• Granulomatosis with polyangiitis	1 (3)
• Eosinophilic fasciitis	1 (3)
• Henoch-Schönlein purpura	1 (3)
Age since diagnosis (mean, range)	6.2 (0.5-16)

Clinic discussions

When asking the young people if they missed topics that were never discussed during consultations, 35% (*n*=10) reported they missed discussion of at least one of the following topics:

- Educational and vocational choices.
- Alcohol/drugs.
- Sexuality.
- Leisure-time activities.

Of parents, 21% (*n*=7) said they missed topics and would have liked to discuss at least one of the same topics during consultations. Two parents also wanted to receive information about fertility and medication.

Experiences of preparation for transfer

Six questions investigated preparation for transition. The answers are summarised below. Almost half of patients never thought about the transfer to adult care and 50% were not informed about the upcoming transfer. The other half heard about transition during the study through a verbal remark that they would soon be transferred to the adult team. No further information was given according to patients. Only three received more information about the differences between children's and adult clinics. Of patients 16% (n=5) experienced anxiety about the transition. Half said they would like to meet the adult care team before the transfer.

About one third of patients did not expect a difference between the approaches of the children's and adults' teams. Those who did expect a difference mentioned a change in approach with less personal attention and older patients in the waiting room, while others did not know what the difference would be.

Of parents, 70% (n=23) had thought about the transfer to the adult team, but did not find it necessary to discuss this subject at an earlier stage. However, when specifically asked about the upcoming transfer, almost one third (n=10) of parents had feelings of anxiety and insecurity, and 10% (n=4) had mixed feelings. Most parents expected to experience a difference; they mentioned a less personal approach and being treated more maturely.

Patients' self-management skills

Eleven questions referred to self-management skills (Table 2). The first question on self-management was preparation for the consultation. A total of 59% of the patients prepared questions before the consultation and only half asked questions during the consultation. In total, 29 (91%) patients were familiar with their diagnosis and 28 (88%) indicated they had enough knowledge about their disease to take care of themselves independently. This was in line with the results from the parents' survey; most parents thought their child had enough knowledge about the disease and medication to take care of themselves. Most (n=30, 94%) patients knew what medication they used, but only 18 (56%) knew its side effects. Only 12 (38%) patients thought about taking the medication themselves without the help of their parents. Additionally, only eight (25%) patients ordered prescriptions from the pharmacy and five (16%) made the clinic appointment themselves.

The patients' degree of independence is an important self-management skill. Almost all 88% (n=28) patients attended the outpatient clinic with their parents. Reasons for attending with their parents included fear of forgetting what physicians had told them, finding it difficult to ask questions independently, fear of forgetting questions that needed to be asked, requiring transport to the hospital and parents who insisted on attending. Of parents, 38% (n=13) thought their child could not visit the clinic independently. Half of the parents recognised the importance of their child visiting the hospital independently, but this was not yet current practice. The parents who recognised the importance of independent visits preferred to make this transition in a gradual way.

Table 2 Young people's survey questions referring to self-management tasks

Questions concerning Self-management skills	Yes (N=32)	%
Prepared questions before the consultation	19	59%
Asked questions during the consultation	16	50%
Made appointment independently		
Patients alone	5	16%
Parents and patients	9	28%
Parents alone	18	56%
Familiar with diagnosis	29	91%
Had enough knowledge about the disease to take care of themselves independently	28	88%
Knew what medication to use	30	94%
Knew what the medication was used for	27	84%
Knew the medication side effects	18	56%
Thought about taking the medication		
Patients alone	12	38%
Patients and parents	15	47%
Parents alone	4	13%
Ordered the medication at the pharmacy		
Patients alone	8	25%
Patients and parents	3	9%
Parents alone	20	63%
Young person injected the medication themselves	9(n=15)	60%

DEVELOPING THE CLINICAL TRANSITION PATHWAY

All results from the patient and parent survey were used to develop a clinical transition pathway, based on the principles of Vanhaecht and Sermeus [14,16].

In brief, the principles for developing the clinical pathway were: starting to investigate the problem, forming a project team, performing a literature search and benchmarking, implementing the clinical pathway, testing the clinical pathway and making adjustments. The last phase is the evaluation.

Evidence-based principles of transitional care were incorporated into the design of the clinical transition pathway [2,18,19]. These principles are also described in the NICE [7] guideline on transition [8]. The principles are:

- The availability of a transition coordinator and age-appropriate care.
- An early start: 12-14 years.
- The use of an individual transition plan (ITP).
- Joint consultations with professionals from children's and adults' services.

- Support for parents in allowing their children to take control of managing their disease [3,9,10,18,20,21].
- Holistic person-centred care, which addresses not only medical care but also psychosocial and vocational needs [22,23].

The ITP developed by McDonagh et al [24,25] was used, with permission, to develop the clinical transition pathway. The ITP was divided into three age categories (12-14, 14-16 and 16-18+ years), each with age-appropriate questions covering ten domains (Supplementary Table 1).

The ITP includes checklists at the end of each age category, which reflect which domain goals are achieved and which domains need specific attention in the following period (supplementary, table 1). An ITP to support and guide parents in the transition process was also developed (Supplementary Table 1) [24,25]. The Childhood Health Assessment Questionnaire (CHAQ) was incorporated in the transition pathway. The CHAQ measures functional status and is validated for different juvenile rheumatic conditions [26,27].

The availability of a transition coordinator is vital for the success of the clinical transition pathway. The specialist rheumatology nurse from the paediatric rheumatology team is the designated transition coordinator and coordinates the clinical transition pathway from the start to the transfer to adult care. This nurse provides substantive patient support and ensures the smooth running of the process. After transfer to adult care, a member of the adult care team takes over as transition coordinator. Both transition coordinators are in close contact with each other.

Figure 1 depicts the clinical transition pathway. All patients aged 12 years and older who visit the outpatient clinic of the department of paediatric rheumatology receive care as outlined in Figure 1. When patients reach the age of 16 and 17, joint consultations are planned with the paediatric rheumatologist, the rheumatologist, the specialist paediatric rheumatology nurse and the nurse from the adult team. The purpose of the joint consultation is to allow patients and their parents to become acquainted with the adult team and ask questions. The adult team also gets the chance to become acquainted with the young person and be informed about the treatment plan. Knowledge of the treatment plan in childhood and understanding why certain choices were made is important for the rheumatologist to ensure alignment of treatment after transfer. Actual transfer to the adult clinic occurs around the age of 16-18 years, where timing of the transfer depends on the patient's achieved skills and personal wishes.

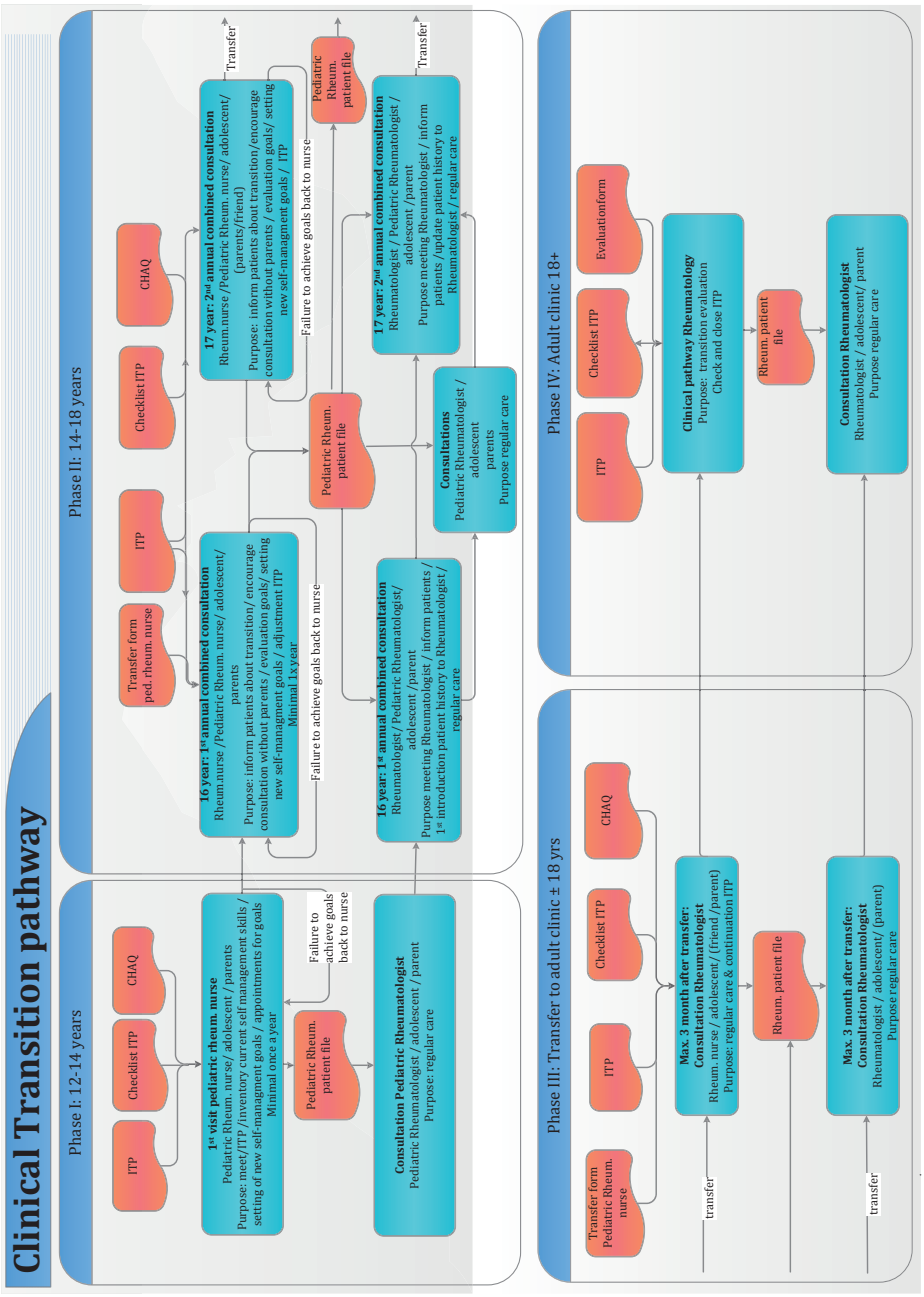

Figure 1 Clinical transition pathway

DISCUSSION

This study explored how patients with jRMDs and their parent(s) experience care during the preparation for the upcoming transfer to adult services, and used the results to develop a clinical transition pathway.

Patients and their parents thought current care at the paediatric rheumatology department did not prepare them well enough for the upcoming transfer to adult care. This included lack of discussion of relevant topics in clinic, neglect in self-management skills training and insufficient knowledge about the differences in care between the children's and adult departments. These themes influenced the development of the clinical transition pathway.

The lack of discussion of topics such as education, vocational needs, sexuality and use of alcohol has been reported in other studies [28,29]. Discussing these topics and providing information is important and may support the development of self-management skills in decision-making [21,30,31]. Adolescents often do not address these topics themselves during consultations [25], possibly because they are accompanied by their parents. In the study, almost none of the patients visited healthcare professionals independently. Confidence to see health professionals without parents is an important skill for successful transition and supports the development of autonomy [9,32,33]. Adolescents who visit the doctor without their parents tend to participate more actively in the consultations [34]. Hence, preparing patients from a young age to attend consultations independently is important.

Other self-management skills were also underdeveloped, for example, making appointments, seeking advice for intercurrent complaints and renewing prescriptions. By implementing an ITP early in the transition phase that addresses all these aspects, goals can be formulated, and plans made to stimulate and amplify self-management skills. This will teach young people skills necessary for a successful transition and help them to cope with their disease later in life [9,33]. A transition plan is also recommended [7,8]. Many studies support the transition process starting early, ideally at the beginning of adolescence (11-12 years) and at the latest by the age of 14 [2,23,35,36,37].

When asked about the differences between children's and adult care, patients and their parents had little understanding. For example, on the one hand you have adult care that is disease oriented and, on the other hand, you have children's care which is socially oriented. Another difference is the emphasis on treatment and compliance with little or no attention to development, school/study or social function in adult care [38]. In addition, they were not always aware of the upcoming transfer to adult care. It is important for patients and parents to understand the differences between the two departments and be prepared for the culture gap [9]. This may diminish the feelings of anxiety and uncertainty that parents and patients reported when asked about the

upcoming transfer to adult care. Anxiety is an obstruction in the transition process [38]. These feelings may be decreased further if patients and their parents can meet their new healthcare provider before the actual transfer [25,30,39]. A minimum of two annual combined consultations at ages 16 and 17 have been incorporated into the clinical transition pathway (Figure 1). Here, young people and their parents are offered a joint consultation with the children's and adult care team together in the same room.

Parents need to be guided during the transition process, because it is important for them to feel confident about their changing role. Parents can be overprotective, which may have a negative effect on their child's developing independence [40]. Therefore, an ITP to support parents in giving young people control of their clinical care was incorporated in the clinical transition pathway.

Studies have shown the importance of transition coordinators in guiding young people and ensuring all their transition needs are met [7,8,41,42]. This role is best fulfilled by nurse specialists as they are trained to address young people's physical, psychosocial and educational needs [25,37] and they can improve transition outcomes [43].

The clinical transition pathway has been incorporated into the routine care of the rheumatology department and is now under evaluation. Its effect will be reported in a future article.

Limitations

Study limitations include the small sample size, of patients receiving treatment in a tertiary care hospital in one year. Although the sample was small, the response rate was relatively high (32 out of 48) for a survey [44]. It is possible that those who did not complete surveys would have responded differently; however, the results were comparable with other studies.

Conclusion

This study explored the experience of care in the transition phase. We found that the clinical care does not meet the needs of young people with jRMDs during the transition process. These unmet needs existed because of a lack of discussion of relevant topics, neglected self-management skills training and insufficient knowledge about the differences in care between the children's and adult departments. Parents were also not guided in the transition process of their child which led to feelings of anxiety for the upcoming transfer to adult care. The clinical transition pathway, developed in the light of the survey results, is a tool that can be used to improve this transition process by providing holistic care appropriate to young people's developmental stage and support their parents in this process.

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Supplementary Table 1 Individual transition plans for YP and parents

Individual Transition plan: young people 12-14 years

Name:	**Start date:**
	Dates when Plan reviewed:

Transition skills	**Yes, I can do this on my own and don't feel I need any extra advice**	**I would like some extra advice/ help with this**	**Action/ date**
1. I can describe my condition			
2. I ask my own questions in clinic			
2. I feel ready to start preparing to go alone for part of the clinic visit in the future			
2. I am able to manage my fatigue (tiredness)			
2. I usually sleep well			
2. I am able to manage my pain			
2. I can look after myself at home in terms of dressing and bathing / showering etc			
3. I know my medication regime – names, doses, how often etc			
3. I understand the risks of not taking my medication			
4. I understand the meaning of 'transition'			
5. I understand the importance of exercise/activity for both my general health and my condition			
6. I understand being overweight can be extra troubling for both my general health and my condition			
6. I am aware that my condition can influence my puberty development			
7. I am comfortable with the way I look to others			
8. I understand the risks of alcohol, drugs and smoking for my health in combination with my medication			
9. I see my friends outside school hours			
9. I have friends or I know someone that I can talk to when I feel sad or fed-up			
9. I know how to deal with unwelcome comments or bullying			
9. I have hobbies, I am a member at a (sport) club			
10. I am managing at school e.g. getting to and around school, deal with my schoolwork, gym at school, chore, friends etc			
10. I know what I want to do when I leave school			
Please list anything else you would like help/advice with:			

Individual Transition Plan: young people 14-16 years

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
1. I understand the medical terms/words and procedures relevant to my condition			
2. I feel confident to go by myself for part of the clinic visit or the complete visit			
2. I understand my rights and responsibilities regarding disease information, privacy and decision-making and consent.			
2. I am able to manage my own pain			
2. I am able to manage my own fatigue (tiredness)			
2. I usually sleep well			
2. I am responsible for my own medication at home			
2. I am responsible for a particular household chore(s) at home			
2. I can look after myself at home in terms of dressing and bathing/showering etc			
3. I understand what the effect of each of my medications is and what their side effects might be			
3. I understand the risks of not taking my medication on a regular basis			
4. I know what each member of the rheumatology team can do for me			
4. I understand the differences between pediatric and adult health care			
5. I exercise regularly / have an active lifestyle			
6. I understand being overweight can be extra troubling for both my general health and my condition			
7. I am comfortable with the way I look to others			
8. I understand the risk of drugs, alcohol and smoking for my health in combination with my medication			
9. I know how to deal with unwelcome comments / bullying			
9. I have friends or I know someone that I can talk to when I feel sad or fed-up			
9. I have hobbies, I am a member at a (sport) club			
10. I am managing at school e.g. getting to and around school, deal with my schoolwork, gym at school, chore, friends etc			
10. I know what I want to do when I leave school			
10. I have (some) work experience			
10. I am aware of the potential impact of my condition for my education and/or work opportunities			
11. I know my disease can affect my sex life (if applicable):			

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
11. I understand the implications of my condition and medication on pregnancy/parenting (if applicable)			
12. I know how to access reliable information about my disease (sexual health, work, relations, organisations for young people with a chronic disease)			
Please list anything else you would like help or advice with:			

Individual Transition Plan: young people 16-18 years +

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
1. I am confident that I have enough knowledge regarding my disease and it's medication			
2. I am able to manage my own pain			
2. I am able to manage my own fatigue (tiredness)			
2. I usually sleep well			
2. I feel confident to go by myself to the clinic			
2. I take care of my medication			
2. I order and collect my (renewed) prescriptions and book my clinic appointments			
2. I call the hospital myself if I have a question about my disease and / or medication			
2. I am responsible for a particular household chore(s) at home			
2. I can look after myself at home in terms of dressing and bathing/showering etc			
3. I know how to plan ahead for being away from home, for example for (overseas) trips and how to store my medication, where to get information regarding vaccinations			
3. I understand the risks of not taking my medication on a regular basis			
5. I exercise regularly / have an active life style			
7. I am comfortable with the way I look to others			
8. I understand the risk of drugs, alcohol and smoking for my health in combination with my medication			
9. I know how to deal with unwelcome comments/bullying			
9. I have friends or I know someone that I can talk to when I feel sad or fed-up			
9. I have hobbies, I am a member at a (sport)club			
10. I have a career plan (please specify)			
10. I have (voluntary) work experience			
10. I am aware of the potential impact (if any) of my condition on my future career plans			
10. I know what to tell a potential employer about my disease			
11. I know my disease can affect my sex life (if applicable)			
11. I understand the implications of my condition and medication on pregnancy/ parenting (if applicable)			
12. I know that there are possibilities in terms of financial support for young adults with chronic illness (if applicable)			

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
12. I understand my eligibility for receiving specific student grants developed for YP with a chronic disease			
13. I know how it works at the adult care and what I can expect of the adult rheumatology care team			
13. I can or am learning to drive a car			
13. I have plans for a study, job and living on my own (please specify)			
Please list anything else you would like help or advice with:			

Source McDonagh JE, Southwood TR, Shaw KL. Growing up and moving on in rheumatology: development and preliminary evaluation of a transitional care programme for a multicentre cohort of adolescents with juvenile idiopathic arthritis. J Child Health Care 2006; 10(1):22-42.
McDonagh JE, Hackett J, McGee M, Southwood T, Shaw KL. The evidence base for transition is bigger than you might think. Arch Dis Child Educ Pract Ed. 2015 Dec;100(6):321-2

Individual transition plan: parents

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
I think that my son / daughter has sufficient knowledge with regard to the disease and medication			
I am able to help my son / daughter to deal with pain			
I know how to support son / daughter to deal with fatigue			
I encourage my son / daughter to be responsible for household chores			
I know how I can support my son / daughter to be responsible for the disease and the use of medication (medication intake, prescriptions including renewals, clinic visits)			
I encourage my son / daughter to be independent at home (general daily necessities, preparing meals, etc.)			
I feel confident when my son / daughter, goes to part or the whole consultation in clinic without me			
I understand the right my son / daughter has to information, privacy and confidentiality			
I can support my son / daughter in when and how contact should be made with the rheumatology and how to obtain prescription renewals.			
I know how to advise my son / daughter when planning a weekend away or holiday, including how to deal with medication (storage, quantity) and if necessary where to get advice for vaccinations			
I know the members and their role in the adult rheumatology team			
I understand the difference between the pediatric and the adult outpatient clinics			
I know the future plans regarding the rheumatology care for my son / daughter after transfer (which hospital, which rheumatologist)			
I understand the intent of the transition process, namely working towards independence of my son / daughter			
I encourage my son / daughter to have an active lifestyle			
I understand the importance of a healthy lifestyle and preventing overweight for my son / daughter			
I know how to help my son / daughter when he / she does not feel comfortable with his / her looks			
I understand the risks of alcohol, drugs and smoking in combination with medication for the health of my son / daughter			

Transition skills	Yes, I can do this on my own and don't feel I need any extra advice	I would like some extra advice/ help with this	Action/ date
I recognize the importance of having friends especially during puberty and think my son / daughter is competent to make friends and I know how to support this process			
I know how to support my son /daughter in dealing with unwelcome comments / bullying			
I know that my son / daughter has someone to talk to when he / she feels sad			
I know websites or patients associations for parents with a child with the same disease			
I know where I can get advice / help if there are problems at the school of my son / daughter			
I understand the importance of work experience for the career of my son / daughter			
I am aware of the possible impact that my child's disease may have on education and/or work abilities			
I know what my son / daughter should tell about the disease to a potential employer			
I understand that the disease / medication can have consequences for pregnancy / parenting of my son / daughter.			
I know where I can get reliable information about sexuality for young people and their parents			
I know that there are possibilities in terms of financial support for young adults with chronic illness			
I know agencies that support parents with a child with a rheumatic disease			
I know what vocation my son / daughter would like to pursue after high school			
I understand the (mental, emotional, physical) changes of puberty and that this may affect the experiences of the disease of my son / daughter			
I've talked with my child about how the disease can be when he / she is an adult			
Please list anything else you would like help or advice with:			

Checklist domains YP	**12-14**		**14-16**		**16-18+**	
1= finished 2= needs attention	**1**	**2**	**1**	**2**	**1**	**2**
1. Information / knowledge disease						
2. Self-management						
3. Information / knowledge medication adherence						
4. Information transition						
5. Sport / Exercise/ relaxation						
6. Health and lifestyle						
7. Self-image						
8. Alcohol, drugs, smoking						
9. Social participation						
10. Vocational, work						
11. Sexuality						
12. Social service						
13. Future prospects						

The numbers on the ITP are referring to the numbers on the checklist.

CHAPTER 3

What support is needed to self-manage a rheumatic disorder: a qualitative study

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ABSTRACT

Background

Today, patients are expected to take an active role in the form of self-management. Given the burden of a rheumatic disorder, the patients cannot be expected to self-manage on their own. In order to develop self-management interventions that fit patients' needs and preferences, it is essential to examine patients' perspective on how support can be optimized. This study aimed to identify support needs of outpatients with rheumatic disorders and preferences for who should provide self-management support.

Methods

A qualitative study was conducted using focus groups and individual interviews with outpatients with rheumatic disorders treated in a Dutch university hospital. Interview data was analysed with Directed Content Analysis and coded with predetermined codes derived from our model about support needs of chronically ill patients. This model distinguished three types of support: instrumental, psychosocial and relational support.

Results

Fourteen patients participated in two focus group interviews and six were interviewed individually. Most patients preferred an active role in self-management. Nonetheless, they notably needed support in developing skills for self-managing their rheumatic disorder in daily life. The extent of support needs was influenced by disease stage, presence of symptoms and changes in one's situation. A trusted relationship and partnership were conditional for receiving any kind of professional support. Patients wanted to be seen as experienced experts of living with a rheumatic disorder. Acquiring specific disease-related knowledge, learning how to deal with symptoms and fluctuations, talking about emotional aspects, and discussing daily life issues and disease-related information were identified as important elements of self-management support. It was considered crucial that support be tailored to individual needs and expertise. Professionals and relatives were preferred as support givers. Few patients desired support from fellow patients.

Conclusion

Self-management was primarily seen as patient's own task. Above all, patients wanted to be seen as the experienced experts. Professionals' self-management support should be focused on coaching patients in developing problem-solving skills, for which practical tools and training are needed.

INTRODUCTION

Having a rheumatic disorder requires ongoing psychosocial adjustment and behavioral change to deal with fluctuations, pain, restricted mobility and fatigue in daily life [1,2]. It may also affect one's mood, self-esteem, role, relationships, and control perceptions [3]. Today, patients are expected to take an active role [4, 5] in the form of self-management, defined as "managing one or more chronic conditions (e.g. symptoms, treatment, physical and psychosocial consequences, and lifestyle changes) and integrate them in day-to-day life with the aim of achieving optimal quality of life"[6: p.547,7: p.178]. Given the burden of a rheumatic disorder, however, the patients cannot be expected to self-manage on their own; they will need support not only from health care professionals [8] but also from relatives and fellow-patients [9].

Many self-management support (SMS) interventions aimed at patients with a rheumatic disorder are available, including educational programs [9], cognitive behavioral therapy [10, 11] and goal setting interventions [12]. At outpatient clinics, SMS is mostly provided by nurses [13]. There is limited empirical evidence, based on lived experiences [14, 15], on what kind of support outpatients with rheumatic disorders desire. A recent scoping review showed that people with rheumatoid arthritis desire informational, emotional, social and practical support [16]. Another recent qualitative review presented a model of various chronic patients' support needs distinguishing three types of support: instrumental, psychosocial and relational support [17]. Moreover, professional SMS is often medically oriented, with a tendency to overlook social and psychosocial problems [13, 18]. It must be noted, that patients' support needs are unique and may change over time [17]. While their needs can be disease-specific, recent research had indicated that challenges in self-management are partly generic. Most types of Chronic Conditions had a small effect on patients' self-management challenges [19]. Both disease-related and individual factors, e.g. flare up of symptoms, cultural backgrounds, gender, and changes in patient's personal situation, seem to influence one's self-management support needs [17].

Professionals could facilitate patients' self-management by seeing healthcare as a shared responsibility. Patients want to be seen as the daily life experts [20]. Good understanding of patients' needs could help professionals in designing effective interventions.

We used the model of 'SMS needs' [17] to identify what kind of support outpatients with rheumatic disorders need and who they would like to receive support from. This study is part of an intervention mapping process [21] that is expected to lead to the development of a nurse-led self-management intervention that fits patients' needs and preferences for support.

MATERIALS AND METHODS

Design

A cross-sectional qualitative study was applied involving a variety of outpatients with rheumatic disorders and using the directed content analysis.

Sample and participants

A full sampling strategy was used, inviting Dutch-speaking patients treated at the outpatient clinic of the Rheumatology department of the Erasmus MC, University Medical Center Rotterdam. During seven weeks, three rheumatologists and one nurse practitioner (MW) distributed a flyer with information about the focus groups to eligible patients.

Eligible patients were those diagnosed with rheumatoid arthritis (RA), psoriatic arthritis or ankylosing spondylitis and a minimum age of eighteen years. These patients were recruited because they represent the most common disease of our outpatient clinic. Patients who have been diagnosed recently were excluded. Sixty-three patients were actually invited. Using principles of purposeful sampling [22] in order to create a sample with maximum variation in terms of age, employment, disease type and years of diagnosis, 63 patients were finally invited for group or individual interviews.

Data collection

Between March 2014 and February 2015, in-depth information was gathered through focus group interviews and face-to-face interviews. Both methods were used because not all patients could attend the focus group sessions. Focus group interviews were considered an appropriate data collection method because participants can be encouraged to discuss and react to others' remarks. This type of intensive interaction enables a broad exploration of experiences and attitudes, which can enrich data [22, 23, 24]. Additionally, individual interviews were held to allow for maximum variation sampling. Individual interviews helped us to gain a deeper understanding of the topics discussed during focus group interviews because participants could explain their view more elaborately. In the analysis, results of both interview types were pooled to develop a comprehensive understanding of patients' needs and to validate conclusions [22].

The primary researcher, a nurse with basic training in qualitative research methods (JB), conducted the focus group interviews assisted by an independent moderator, a psychologist and psychotherapist who was very experienced with group interaction. This moderator stimulated patients to share their ideas and opinions, but was not involved in data analysis.

These interviews lasted about two hours and were held in a private location outside the hospital. Face-to-face interviews were conducted by JD, an experienced qualitative researcher. These lasted about one hour and were conducted in a private space in

the hospital. Leading interview questions are shown in Table 1. Prior to the interview, patients did not receive any information about what kind of support could be provided by whom. This was done in order to encourage them to freely describe their needs for support and preferences for any team member who should provide this support. All interviews were audio-recorded and transcribed verbatim.

Table 1 Leading interview questions

- **What can you tell me about your life with a rheumatic disorder?**
- **What kind of support do you receive in dealing with your rheumatic disorder?**
- **What kind of support would you need and/or prefer in dealing with your rheumatic disorder?**
- **Who would you preferably like to provide this self-management support?**

Ethical considerations

All invited patients received a flyer and all included participants provided informed consent. Participants were assured of confidentiality and data were processed anonymously by the first researcher. The researchers (JB, JD, AvS, and EI) had no access to patient records, while MW and JH –who were involved in the medical care of some patients- were neither involved in data collection nor had access to non-anonymous data. The study protocol was approved by the Medical Ethical Committee of the Erasmus MC (MEC-2013-350).

Data analysis

Patients' support needs were explored through the Directed Content Analysis (DCA) approach, which is appropriate when prior research exists about a phenomenon [25]. Of the two DCA coding strategies, we opted for the one that starts with applying predetermined codes from an existing theoretical framework, in this case the model of 'SMS needs' further detailed below [17]. First, the first and second author (JB and MW) read the interview transcripts to gain an overall impression of the contents. Subsequently, they applied predetermined codes based on the different components of the model of SMS needs: (need for support) knowledge – information and instruction, internalizing knowledge, instrumental, adjusting daily life, recognition of emotional aspects, building self-confidence and empowerment, partnership and sympathy. Subthemes of these codes were (support from) professionals, relatives and fellow patients. Factors contributing to the uniqueness of this support were also coded. JB and MW discussed and refined these codes during the coding process. Data considered interesting but which could not be coded with this initial coding scheme were analyzed later "to determine if they represent a new category or a subcategory of an existing code" [25].

Data saturation was achieved after having analyzed two focus group interviews and four individual interviews when the data became repetitive [22].

Theoretical framework: model of SMS needs

To analyze the data, we used the model of SMS needs (figure 1), constructed by Dwarswaard and colleagues [17]. This generic model, developed in a qualitative review of 37 articles, distinguishes three types of support to be provided by professionals, relatives (family and friends) and fellow patients to chronically ill patients: relational, instrumental, and psychosocial [17]. This model of SMS needs will be explained more clearly in the Results section and in Table 3.

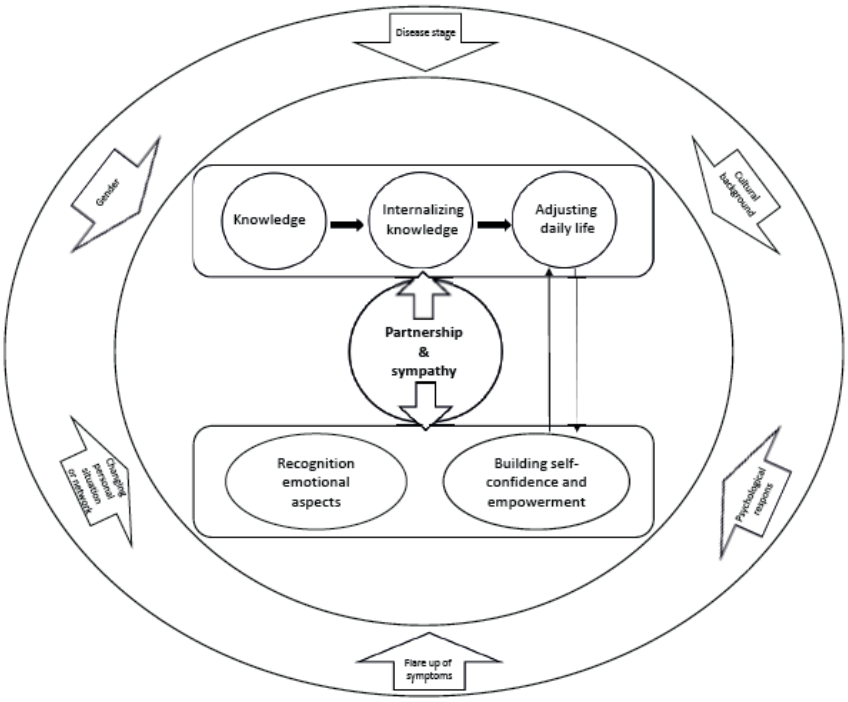

Figure 1 Model of SMS needs [17]

Strategies to establish rigor

Both researcher and method triangulation [22] were used to enhance the validity of the data. All data was collected and analyzed in a team-based fashion. Agreement in coding was reached by consensus between the two coders. To increase the dependability of the research, the design, methods, (preliminary and final) analyses and results were all discussed within the research team. Readers can conclude on the degree of transferability from the provided details of the participants and settings. The description of the methods also contributes to the conformability of this study.

RESULTS

Forty-three (68%) patients declined to participate, mostly due to logistical difficulties with planning. Eventually, fourteen patients participated in two focus groups interviews (FGI) and six were interviewed individually (II). Sample characteristics are shown in Table 2.

Table 2 Sample characteristics

	Face-to-face interviews N (%)	Focus group interviews N (%)	Total N (%)
Gender			
Female	3 (50.0%)	11 (78.6%)	14 (70.0%)
Male	3 (50.0%)	3 (21.4%)	6 (30.0%)
Age			
34 – 44 years	2 (33.3%)	0 (0.0%)	2 (10.0%))
45 – 54 years	0 (0.0%)	3 (21.4%)	3 (15.0%)
55 – 64 years	3 (50.0%)	7 (50.0%)	10 (50.0%)
> 65 years	1 (16.7%)	4 (28.5%)	5 (25.0%)
Marital state			
Cohabiting / married	4 (67.3%)	8 (57.2%)	12 (60.0%)
Widow	0 (0.0%)	1 (7.1%)	1 (5.0%)
Single	2 (33.3%)	5 (35.7%)	7 (35.0%)
Diagnosis			
Rheumatoid arthritis	6 (100%)	10 (71.4%)	16 (80.0%)
Psoriatic arthritis	0 (0.0%)	2 (14.3%)	2 (10.0%)
Ankylosing spondylitis	0 (0.0%)	2 (14.3%)	2 (10.0%)
Years of diagnosis			
< 5 years	0 (0.0%)	1 (7.1%)	1 (5.0%)
5-10 years	2 (33.3%)	7 (50.0%)	9 (45.0%)
> 10 years	4 (66.7%)	6 (42.9%)	10 (50.0%)
Employment			
Yes	4 (66.7%)	2 (14.2%)	5 (25.0%)
No	2 (33.3%)	12 (85.8%)	15 (75.0%)

Views on self-management

Self-management was primarily seen as one's own task: "I want to do it [managing a rheumatic disorder] myself" (II-R1). Most patients preferred an active role, thinking that others could not manage the rheumatic disorder for them: "Finally, I'm in charge. I want to experience things myself. Other persons cannot explain everything" (II-R5). Ultimately, they themselves have to deal with the disorder: "in the end no one can really help" (FG1-R1). Patients wished to "determine [themselves] what works or does not work […]" (II-R4). Problems are solved by trial and error: "Initially, you ask too much of your

own body…. But at some point you'll recognize your limits. To get there, you must be familiar with your own body" (FG2-R2). Still, actively adapting to the rheumatic disorder can be difficult: e.g. "Sometimes, I go beyond my physical limits. But eventually, you'll hit a brick wall" (FG1-R7).

Support needs

Even though self-management was primarily seen as the patient's responsibility, support from professionals (doctors and nurses), relatives and fellow patients could be accepted. Support might strengthen their empowerment: "I often have inflammations in my wrist. The pain is terrible. Apart from taking pills, I did not know other solutions. A nurse helped me by sharing the experiences of other patients... At some point I learned to live with it. However, I would like to be guided in managing these challenges in daily life" (II-R2). Preferences are described below following the 'SMS needs' model [17]. Table 3 provides an explanation.

Table 3 *Model of SMS needs [17]*

Themes	Subthemes	Quotations to explain the model*
Relational support refers to supporting aspects of interactions with other persons. This involves two subthemes: partnership, and sympathy.	Partnership	*"It is not possible to hold professionals responsible for everything. It ought to be co-operation. Every patient should consider what is good for him of her" (II-R5)*
	Sympathy	Patients highly appreciate when their symptoms and side effects are taken seriously: *"Action was taken immediately. In a few days I felt better. I was really accepted" (FG2-R3)*
Instrumental support is related to the medical management of a chronic condition, This involves three subthemes: Knowledge – information and instruction, internalizing knowledge, and adjusting daily life.	Knowledge – information and instruction	*"For example, I want information about what can happen if I do not wish to be operated on my hand" (FG2-R4)*
	Internalizing knowledge	Having the opportunity to discuss disease-related information: *"I calm down when a nurse tells me how to interpret side effects I've noticed" (II-R4)*
	Adjusting daily life	*"I liked to get advice on how to deal with a rheumatic disorder in daily life. To hear that on the one day you're capable of house cleaning and the next day you're not" (FG1-R2)*
Psychosocial support pertains to the resources needed to manage the emotional and psychosocial aspects in living with a chronic condition. This involves two subthemes: recognition of emotional aspects of the chronic condition, and building self-confidence and empowerment.	Recognition of emotional aspects of the chronic condition	*"Just ventilating [my emotions or feelings] is enough" (II-R3)*
	Building self-confidence and empowerment	*"For me, it was a psychological transition to inject myself. First, the nurse showed me how to administer this medication. Then she instructed me stepwise. Afterwards I felt confident enough do it myself" (II-R3)*

* Quotations were derived from the focus group (FGI) and individual interviews (II)

1. Relational support

Partnership and sympathy

Having a trusting relationship with professionals, relatives and fellow patients was seen as conditional for receiving SMS. Only then, one may comfortably talk about problems at home or work, express one's own opinion and feel one can rely on the capabilities of the other person. If such relationship is lacking, one may be less open to support: "I did not want any kind of support from her [a specific professional]… She was not unfriendly, but I did not trust her" (II-R1). This applies also to relatives and fellow patients: "First, they [relatives] need to show some genuine interest in me" (FG1-R1). Sympathy can affect this level of trust. A sympathetic person was defined as a good listener, someone who is empathic, shows interest and understands.

Continuity of care was important for those who preferred support from professionals. Continuous rotation was seen as counterproductive for building a relationship of trust as becomes clear from a discussion in one of the focus groups: "At first, I had different doctors. This was very annoying" (FG2-R7). "Yes, that is really annoying" (FG2-R2). "Every time I had to repeat my story. There was a story in the computer, e.g. about blood levels. However, this was not my personal story" (FG2-R7). Confidence in professionals "needs to emerge over time" (II-R1).

Apart from trust, also partnership with professionals was seen as an important component of SMS: "It is not possible to hold professionals responsible for everything. It should be a matter of co-operation. Every patient should consider what is good for him of her" (II-R5). Patients wished to be involved in decision-making and preferred to "think together about treatment options" (II-R4). Even though professionals were seen as the medical experts, patients wanted professionals to "respect the choices" (FG1-R8) they make. Above all, they wanted to be seen as experienced experts of living with a rheumatic disorder.

2. Instrumental support

Knowledge – information and instruction

Patients said they needed specific disease-related knowledge (e.g. about diagnosis, symptoms, treatment options, assistive devices, and the necessity of physical exercise). Not everyone needed the same amount and type of information at the same time. Once they had received the diagnosis, patients just wanted information about their rheumatic disorder or how to recognize early symptoms. They did not wish to hear about all possible complications: because, "I am not ready for it" (FG2-R6). They were not open to this kind of information until after a certain degree of acceptance has been reached. Some time after diagnosis, patients wanted to receive information related to their personal situation (e.g. about new devices, medication, or symptoms related to complications.

Patients' information needs are also influenced by the disease activity and the symptoms experienced.

In this study, patients preferred a stepwise knowledge provision tailored to personal needs. Failure to provide tailored education carries the risk of patients being "overwhelmed by all information" (FG2-R6). Most patients prefer advice about reliable literature: "Nowadays, you can find information anywhere. Professionals could help by offering information about reliable sources" (II-R5).

In terms of knowledge provision, not much was expected from relatives. However, patients found it important that professionals provide tailored information about the rheumatic disorder to relatives, as lack of knowledge could lead to less optimal support.

Internalizing knowledge

Having the opportunity to discuss disease-related information with professionals, relatives, and fellow patients was seen as a way to internalize knowledge. "I calm down when a nurse tells me how to interpret side effects I've noticed" (II-R4) and "It helps me to talk with […], someone [a fellow patient] who knows what it means to have a rheumatic disorder" (II-R2). However, not everyone liked this kind of support from fellow patients: "I don't need this [support from fellow patients], because they will constantly talk about their ailment. It gets worse and worse" (FG1-R6).

Adjusting to daily life

Since "nothing is as difficult as changing your lifestyle" (II-R2), almost all patients needed support in integrating their rheumatic disorder in daily life. The extent of support need was influenced by the disease stage, the presence of symptoms and changes in one's situation. Right after diagnosis, more and specific support is needed: "In the beginning I needed a lot of support. I felt I had my back to the wall. You do not know where it will go" (FG1-R7).

Patients highly appreciated professionals who just "listen and ask how you are doing at home and work" (II-R4). Besides, professionals could give practical advice about dealing with the disorder: "peeling potatoes is very hard for me, professionals can advise me on appropriate assistive devices" (II-R4). Disease fluctuations can be hard to handle. Patients wanted to know how to deal with these.

Some patients needed relatives to monitor their limits: "Sometimes it is helpful when someone else tells you it is enough" (II-R2). However, others said: "I just want to do this all by myself" (II-R1). Patients were less ready to accept this kind of monitoring from their children than from their partners. While relatives may provide practical support such as cleaning and cooking, for some patients "it is difficult to accept help" (II-R5).

Most patients said they did not need support from fellow patients. Some acknowledged that "it is good to know that they [fellow patients] understand how you feel"

(FG1-R4). Several patients also felt supported by experiential stories in the patient association's magazine. One patient was active in a social media group because, "you can ask fellow patients how they are dealing with certain symptoms…. these people face similar problems. A professional does not have this experience" (II-R4).

3. Psychosocial support

Recognition of emotional aspects of a rheumatic disorder

Accepting that a rheumatic disorder is a lifelong disease was a deep emotional process for many: "for me, it felt like an execution" (FG2-R7); and "I was really panicking after diagnosis" (FG1-R4). Mostly it was already helpful when professionals proactively asked and listened: "just ventilating [my emotions or feelings] is enough" (II-R3). For some of the patients this was insufficient, however, because they had long-term problems: "the pain and sadness remain" (FG1-R2). These patients needed to "receive guidance" (FG2-R7) from a specialist e.g. psychologist or social worker, to accept a life with a rheumatic disorder.

Generally, it was easier for patients to discuss emotional aspects when professionals proactively asked about these. Not all patients had the courage to discuss these kinds of problems, sometimes because they "do not want to be perceived as a bore" (FG1-R1). Patients preferred to discuss emotional issues with a nurse, because nurses tended to be "able to create a moment to listen" (FG1-R6).

Most patients just wanted a listening ear from relatives, but some pointed out that relatives did not always recognize their emotional issues. Not all relatives were able to "imagine what it is to be a chronic patient with daily pain" (II-R4). As a result, not all patients received the support they needed. Compared to children and friends, partners seemed more capable in recognizing such emotional issues.

Fellow patients could be of help when they have the same experiences: "I want to talk with someone who is experiencing the same" (II-R4). However, patients were not interested in meeting fellow patients in a group session organized by the hospital. Some patients preferred to meet them informally.

Building self-confidence and empowerment

Although described implicitly, encouragement and reassurance supported the building self-confidence and empowerment: "For me, it was a psychological transition to inject myself. First, the nurse showed me how to administer this medication. Then she instructed me stepwise. Afterwards I felt confident enough do it myself" (II-R3). Positive reinforcement seems to help patients to solve problems or change behavior. For example, when a physician told a patient "that she would be able to exercise" (FG1-R7) and that it should help her, she felt confident to exercise more often so that her body became more flexible. It could also be helpful to see other patients exercising. On the

other hand, some thought it would be confrontational to see the consequences of rheumatism in others.

DISCUSSION

In this qualitative study we explored the support needs of people living with rheumatic disorders. The analysis learned that they saw self-management primarily as a task for themselves but nevertheless appreciated support to help them achieve this. Most of the interviewed outpatients preferred support from professionals and relatives; only few appreciated psychosocial support from fellow patients.

Although the concept of self-management assumes an active role for patients in managing and integrating a chronic condition(s) in daily life [6, 7], it was striking to find that this concept seems to fit so well to outpatients with a rheumatic disorder. However, even when patients appear to be autonomous self-managers their need for support should not be underestimated. It is not reasonable to expect patients to manage a rheumatic disorder on their own [26, 27]. All patients need encouragement [28] to develop enough self-confidence to manage a disorder. Bandura found self-efficacy to be an accurate predictor of patients' fulfillment in managing a disorder [29, 30]. The core element of professionals' support should therefore be coaching patients to develop problem-solving skills. It should be remembered, however, that not everyone believes in their capacity of self-managing. Patient with less confidence need more encouragement and recommendations from others e.g. professionals and relatives [31].

We found that learning how to deal with symptoms and fluctuations, talking about emotional aspects, and discussing daily life issues e.g. work and household were important aspects. All important aspects of the broad definition about self-management that we used in this study [6, 7]. Chronically ill patients are challenged to deal with the medical, emotional and social issues of their disorder in daily life [8]. These aspects also came to the fore in two reviews [16, 17]. One of these reviews shows that patient-related factors influence support needs [17]. In this qualitative study we did not found any difference for gender, age and work status. Moreover, the time since diagnosis and course of the rheumatic disorder affected support needs. Patients who experienced more problems or disease activity had more need for support, in line with previous research [32]. It would be worthwhile to study how SMS could be tailored to individual needs and expertise [33].

The interviewees in this study saw partnership and a trusted relationship as conditional for receiving SMS. Continuity of care and professionals taking the problems seriously could help build a trusted relationship. A good professional-patient relationship is therefore the cornerstone of care, especially in view of achieving behavioral change [34,

35]. Partnership is generally recognized as an important part of SMS [8, 36, 37]. However, it can be difficult for professionals to achieve collaborative partnership [38-40] as they may be inclined to play the expert role [18]. Patients in this study appreciated support from nurses and doctors alike. Usually, nurses took more time to discuss emotional and social aspects.

Partnership and a relation of trust were not only conditional for support from professionals, but also from relatives and fellow patients. Relatives were especially prized for their emotional and practical support. Fellow patients can help by sharing their lived experiences. However, not all patients appreciate this kind of support, unless this can help in managing a chronic condition well [4]. Modeling, observing others in performing new behavior patterns successfully, can serve as a guide for translating behavioral conceptions to appreciate actions [26].

Operationalizing SMS may not be easy for professionals [13]. They tend to resort to traditional (standardized) patient education [18], instead of providing the recommended tailored patient education [41]. Moreover, interventions that solely provide education have been found least successful [42, 43]. Interventions focusing on patients' intrinsic processes seem to be most successful [44]. Focusing on more internal perceived locus of control is important for persistence and performance of new behavior [45]. Still, professionals lack skills to facilitate psychosocial challenges in self-management [20]. Additional training could help professionals to incorporate coaching into their repertoire of SMS interventions.

The model of 'SMS needs' (figure 1)[17] we employed was helpful in that we benefitted from previous descriptions and could create a deeper understanding of the support needs of people with a rheumatic disorder. On the other hand, the DCA approach carries the risk of fitting data to the predetermined coding scheme. Relevant data can be missed when applying this highly structured method. To minimize this risk, we also applied inductive coding if data could not be categorized. This enabled us to unravel the importance of the 'self' in self-management for patients with rheumatic disorders. Lastly, collecting data from not only focus group interviews but also face-to-face interviews was very useful. Individually interviewed participants in elaborated more on their experiences, which helped to create a comprehensive understanding of patients' needs. However, findings from the two interview types did not differ essentially.

A possible limitation of this study is that mostly elderly, retired patients with RA participated in the focus groups. It was difficult to recruit younger persons for the focus groups. Still, given that the prevalence of RA is much higher than the prevalence of psoriatic arthritis and ankylosing spondylitis, that RA occurs at older age, and that most of the RA patients were women, the composition of our sample seems to correspond to the normal distribution in the general population [46]. However, to minimize the risk

of selection bias, we purposefully searched for younger or employed patients for the individual interviews.

In this study, we decided to exclude patients who have been diagnosed recently and to ask patients in retrospect what their supports needs were at the time. Thus we did not provide insight in support needs of recently diagnosed patients. Furthermore, all data was collected in one hospital in the Netherlands and the findings may therefore not be representative for patients in others countries. Hence, we recommend to study whether of outpatients in other countries may perhaps have other SMS needs.

CONCLUSION

Self-management was primarily seen as one's own task, but patients still appreciated support to help achieve this. Above all, they wanted to be seen as experienced experts of living with a rheumatic disorder. Preferred support givers were professionals and relatives. Professionals' self-management support should be focused on coaching patients in developing problem-solving skills for managing the medical, emotional and social challenges experienced in dealing with a rheumatic disorder in daily life. Practical tools and training are needed to operationalize coaching as a part of professional self-management support in working routines.

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CHAPTER 4

Fatigue in early, intensively treated and tight-controlled rheumatoid arthritis patients is frequent and persistent: a prospective study

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ABSTRACT

Background

Fatigue has a large impact on quality of life and is still unmanageable for many patients. Study aims were describe (1) the prevalence and pattern of fatigue over time in patients with early rheumatoid arthritis under a treat-to-target strategy and (2) identify predictive factors for worsening and recovering of fatigue over time.

Methods

Data from the tREACH study were used, comparing different treatment strategies with fatigue as secondary objective. Patient outcomes on fatigue, quality of life, depression, and coping were obtained every 6 months and clinically assessed every 3 months. Prediction of fatigue at 12 months was investigated with an ROC curve. Analysis was stratified into non-fatigue and fatigue at baseline. Logistic regression was used for the evolution of fatigue in relation with the covariates over time.

Results

Almost half of all patients (n=246) had high fatigue levels at baseline, decreasing slightly over time. At 12 months, 43% of patients were fatigued; while 23% of the initially fatigued patients showed lower levels of fatigue, the fatigue level had increased in 15% of the initially non-fatigued patients.The strongest predictor of fatigue was the previous fatigue levels (AUC 0.89). Higher score on the depression-scale and coping with limitations was associated with developing fatigue over time in the initially non-fatigued group.

Conclusion

Despite a strict treat-to-target strategy, fatigue remained an overall problem during the first year of treatment, and was mainly predicted by its baseline status. In subgroups, a small additional effect of depression was seen. Monitoring fatigue and depression may be important in managing fatigue.

BACKGROUND

Studies have shown good results concerning remission and structural radiographic damage of the joints by tight control and treat-to-target management [1,2]. Despite these effective strategies and reaching remission of disease activity, patients with rheumatoid arthritis (RA) still may experience a burden of the diseases like pain and fatigue [3]. From 40% to 80% of the RA patients are fatigued, which may affects their lives [4-6]. The impact of fatigue permeates through every aspect of their lives, limiting work participation [7], family activities or social activities [8], sports and simply enjoying life as it is.

Thus, fatigue is an important aspect for many patients with a high impact on patients by influencing the choices they make in their social life. Moreover, fatigue is associated with a reduced health-related quality of life and depression [8, 9] and is the most limiting factor for the ability to work [10]. Because of this large impact, it is important to study fatigue. So far, little is known about fatigue during the disease course in early RA. According to patients, reducing fatigue is an important treatment target, but is not often addressed during consultations [11,12].

The evolvement of fatigue over time in patients with early RA had been addressed in a few previous studies. An 8-year study from The Netherlands suggested little change of fatigue levels over time at group level, while individual levels fluctuated over time [13]. In contrast, a study in early RA revealed an improvement in fatigue for 40% of the patients, while fatigue levels increased in another 24% [14]. These were both cohort studies in which treatment was left to the discretion of the physicians.

Therefore, the objective of the present study was (1) to describe the prevalence and pattern of the fatigue over time in patients with early RA under a treat-to-target strategy and (2) to identify predictive factors for worsening and recovering of fatigue over time.

METHODS

Study Participants

Data from the tREACH study (treatment in the Rotterdam Early Arthritis Cohort, 2007-2013), comparing different treatment strategies with fatigue as secondary objective, and patients fulfilling the ACR-EULAR 2010 criteria for RA, were used for this analysis [15]. This multi-centered trial compared different initial treatment strategies in early RA patients. Inclusion criteria for the tREACH study were: age ≥ 18 years, arthritis in one or more joint(s) and symptom duration < 1 year. Patients were recruited from the outpatient clinics of all participating centres between July 2007 and April 2011. Initial treatment arms were: I methotrexate, sulfasalazine, and hydroxychloroquine (HCQ) +glucocorticosteroids (GCs) intramuscularly; II methotrexate, sulfasalazine (SASP), and HCQ + oral GC

tapering scheme; III MTX + oral GC. Treatment was escalated to biologicals if DAS44 > 2.4 [16-18]. Details can be found in Claessen et al [19]. The medical ethics committee at each participating center approved the study protocol and all patients gave written informed consent before inclusion (METC 2006-252, trial protocol number 2006-005771-18).

Data collection

Patients' demographic and clinical characteristics as well as the frequency of erosions were recorded at baseline. Disease activity measures and adjustments to treatments were applied every 3 months. Fatigue, coping strategies for pain and physical limitations, health-related quality of life, and symptoms of anxiety and depression were assessed every 6 months.

Clinical evaluation of disease activity

The disease activity was assessed by the disease activity score (DAS28) score, which is a composite score assessing swollen joints, tender joints, and the erythrocyte sedimentation rate (ESR), and includes a visual analog scale (VAS) global (range 0-10). Higher score indicates a higher disease activity [20]. To investigate the relation with fatigue and painful joints, we used the separated tender joint count (TJC 44).

Patient-reported outcome measures

Fatigue

Fatigue level was measured by VAS and the Fatigue Assessment Scale (FAS). The VAS (100 mm) fatigue involves the severity of the fatigue over the past week with the anchors: no fatigue (0 mm) and extremely fatigued (100 mm). The scale is sensitive to change, valid, and reliable, but no cut-off point has been determined [21, 22]. The FAS is a ten-item fatigue scale with a good internal consistency, reliability, and validity [23,24]. Five questions reflect physical fatigue and five questions reflect mental fatigue. The instruction is directed at how a person usually feels. Each item is scored on a five-point rating scale ranging from 1 'never' to 5 'always'. The total scores thus ranges from 10 to 50 and are interpreted as follows: 10-21 no fatigue; ≥ 22-34 substantial fatigue; and ≥ 35-50 extreme fatigue [25, 26].

Disease-related

The Rheumatoid Arthritis Disease Activity Index (RADAI) measures self-reported disease activity [27]. It contains 5 items: global disease activity during the last month, today's disease activity in terms of swollen and tender joints, and today's severity of arthritis pain and stiffness and self-assessed tender joints. It is measured on a scale ranging from 0 to 10, where higher scores indicate more disease activity [28].

General health

The health-related quality of life (HRQOL) was scored with the SF-36 (range 0-100). A higher score indicates a better HRQOL. It assesses eight health concepts: physical functioning, bodily pain, role limitations due to physical health problems, role limitations due to personal or emotional problems, emotional well-being, social functioning, energy/fatigue, and general health perceptions which are summarized in a physical component summary (PCS) and mental component summary (MCS) score [29].

Psychosocial

Coping was measured via the Coping with Rheumatoid Stressors (CORS) scale. The subscales dealing with pain, decreasing activities (range 8-32) and limitations (range10-40) were included in tREACH study. A higher sum score indicates more frequent use of the coping strategy. Both subscales have good internal consistency and high test-retest reliability [30, 31].

Depression and anxiety were measured by the Hospital Anxiety and Depression Scale (HADS). Two subscales with each seven items are calculated with higher scores indicating more severe symptoms of anxiety or depression [32]. Categorical scores are available. Scores between 0 and 7 represent 'no case'; 8 to 10 'possible case'; and 11 to 21 'probable case of anxiety or depression' [32, 33].

Statistical analysis

Simple descriptive techniques were used to describe the prevalence of fatigue and its associations with other covariates at baseline. Mean and SD or percentages were described, as appropriate. As longitudinal fatigue evolvement was diverse, we stratified the analysis into two clinically relevant patient samples: those with no fatigue (FAS values 10-21) and those with fatigue (FAS values 22-50) at baseline [25, 26]. The baseline differences between fatigued and non-fatigued patients among continuous variables were tested with the unpaired t-test or Mann-Whitney U test as appropriate. Categorical variables were tested using Pearson's Chi-square test.

Prediction of fatigue at 12 months was investigated with an ROC curve with fatigue as a continuous variable. To investigate variables that are important for change of fatigue over time, logistic regression analyses predicting fatigue status at 12 months by baseline covariates were performed for each stratum. First, univariable analyses were performed. Thereafter, starting with full models, backward elimination was performed until all remaining variables reached a significance level of $p<0.10$. Age and gender were forced into the models regardless of their levels of significance. Missing values were imputed by multiple imputation with chained equations using m=100 imputation datasets. P-values <0.05 were considered statistically significant.

RESULTS

Baseline fatigue data were available for 246 of 270 individuals participating in the tREACH trial. The mean age was 53 years (SD 14.3 years), the DAS score was 4.8 and 68% were females (see table 1). At baseline, rheumatoid factor and anti-CCP antibodies were present in 73% and 77% of patients, respectively. Erosions were present in 18% of patients. The 24 patients that missed their baseline fatigue level worked less often (p=0.05) but did not differ with respect to DAS score (p=0.7), the presence of erosions (p=0.76) or the treatment arm that they had been randomized to (I vs. II p=0.79; I vs. III p=0.90; II vs. III p=0.88) (data not shown).

Table 1 Baseline characteristics, total, high fatigued patients and fatigue and non-fatigued patients

N=246	All patients (n=246)	Fatigued patients (n=113; FAS (≥22)	No fatigued patients (n=133; FAS <22)	p
Age, in years $\bar{T}$	53.3 (14.3)	51.3 (14.1)	55.0 (14.3)	0.04
Sex, female, (%)	68%	75%	62%	0.03**
Working status (%)	55%	52%	60%	0.21**
Native, Dutch (%)	83%	81%	85%	0.35**
Symptom duration(days)	161.5 (88.8)	166.0 (91.0)	158.14 (87.0)	0.48
RF-positive, %	73%	76%	69%	0.01**
ACPA-positive, %	77%	76%	80%	0.12**
DAS28 (range 0-10)*	4.8 (4.0-5.7)	4.9 (4.3-6.0)	4.7 (3.7-5.4)	0.004‡
• **Tender joints (range 0-44)***	10 (5-15)	11 (6-18)	8 (4-13)	<0.001‡
• **Swollen joints (range0-44)***	8 (4-12)	9 (4-13)	7 (4-11)	0.12‡
• **ESR ***	24 (14-42)	23 (13-44)	24 (15-39)	0.71‡
• **VAS global (range 0-100)***	53 (34-69)	60 (49-73)	49 (28-63)	<0.001‡
VAS fatigue (range 0-100)*	53 (31-73)	70 (55-80)	36 (36-54)	<0.001‡
FAS (range 10-50) *	21 (17-27)	27 (25-31)	17 (15-19)	<0.001‡
RADAI (range 0-10) *	4.1 (2.8-5.5)	4.7 (3.3-6.0)	3.6 (2.3-4.8)	<0.001‡
Coping pain (range 8–32)*	15 (11-19)	17 (14-21)	13 (10-16)	<0.001‡
Coping limitations (range 8–40)*	23 (17-29)	25 (20-30)	21 (16-27)	<0.001‡
HADS anxiety (range 0–21)*	5 (3-8)	7 (5-10)	4 (2-6)	<0.001‡
HADS depression (range 0–21)*	4 (2-7)	5 (4-8)	2 (1-4)	<0.001‡
Possible case depression (HADS-D≥ 8, n / (%))	49 (19.9%)	37 (32.7%)	12 (9%)	<0.001
SF-36 PCS (range 0-100) *	39.9 (35.7-44.9)	37.9 (33.4-41.8)	42.0 (37.4-45.9)	<0.001‡
SF-36 MCS (range 0-100) *	45.4 (41.0-50.5)	43.4 (38.7-48.2)	47.5 (43.7-51.9)	<0.001‡
SF-36 vitality (range 0-100)*	55.0 (40-70)	40.0 (30-50)	70.0 (55-80)	<0.001‡

Abb. RF: rheumatoid factor; ACPA: anti-citrullinated protein antibodies ; ESR: erythrocyte sedimentation rate; VAS: visual analog scale; FAS: fatigue assessment scale; RADAI: Rheumatoid arthritis disease activity index; HADS Hospital Anxiety and Depression Scale; SF-36: Short Form 36; PCS: physical component summary; MCS: mental component summary, $\bar{T}$mean (SD) / *median (IQR) **Pearson's Chi-square ‡Mann-Whitney U test

Prevalence and pattern of fatigue

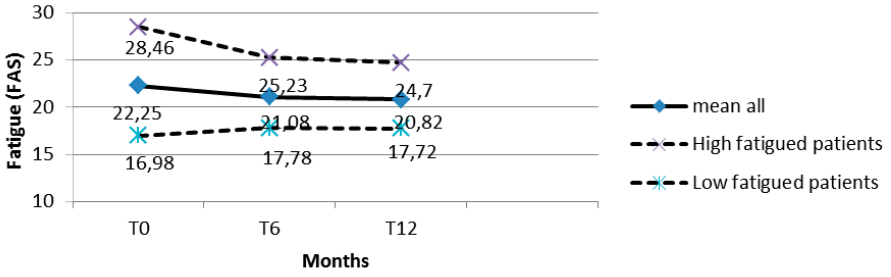
At baseline, the mean VAS fatigue score was 53 (SD 26); and the mean FAS score was 21 (SD 7) and 45% of the patients were categorized as fatigued (FAS > 21) .

Table 1 summarizes the baseline results for all patients and broken down for the 113 fatigued and 133 non-fatigued patients. Fatigue was most commonly present in younger females. The two fatigue groups differed in disease-related characteristics and patient-reported outcomes (table 1). Of note, 32% of the fatigued patients reached the cut point of 8 in the HADS that bears clinical relevant levels for depression, compared to 9% of the non-fatigued patients.

Over time, the FAS fatigue score on average decreased slightly, by 1.4 points in all patients and by 3.8 points in the fatigued patients, while it increased by 0.8 points in the non-fatigued patients (figure 1). At 12 months, 43% of all patients were still fatigued. Individual patient profiles showed varying patterns.

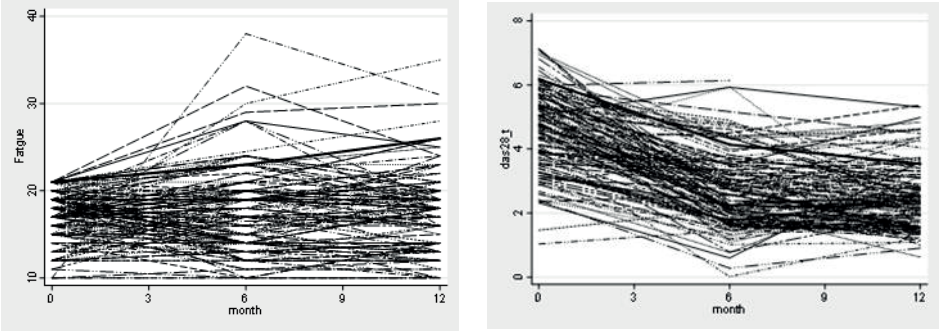
Of all fatigued patients (n=113) at baseline, the fatigue level decreased to below the level of no fatigue in only 23%, while 15% of the non-fatigued patients (n=133) became fatigued.

Figure 1a Evolution over time of fatigue



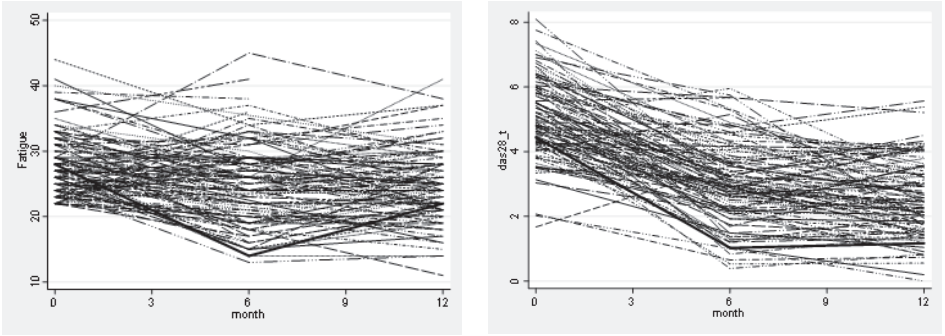
Fatigue measured with the FAS (range 10-50)

Figure 1b Individual profiles patients with no fatigue – panel A – FAS; panel B- DAS28



Panel A Fatigue measured with the FAS (range 10-50) Panel B DAS28 (0-10)

Figure 1c Individual profiles patients with high fatigue, – panel A – FAS; panel B- DAS28



Panel A Fatigue measured with the FAS (range 10-50) Panel B DAS28 (0-10)

Predicting fatigue

The strongest predictor of fatigue was the previous fatigue levels. In an area under curve (AUC) model, baseline fatigue predicted fatigue over time with an AUC of 0.89 (figure 2). Adding other variables did not improve the model.

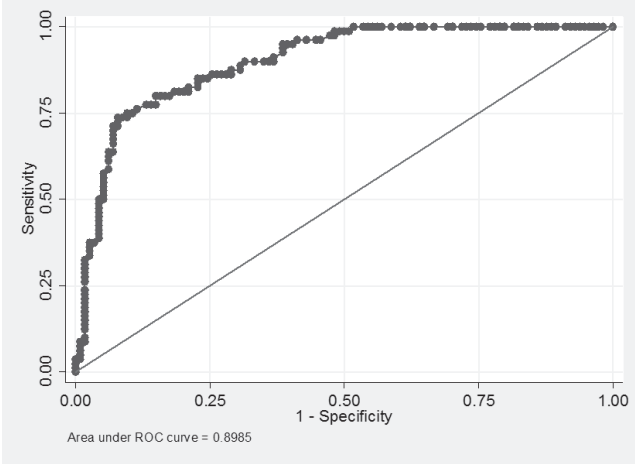


Figure 2 ROC model with only baseline fatigue

Factors associated with the strata of fatigue at 12 months

The univariable analysis of patients stratified by baseline fatigue status showed a significantly higher VAS global score in the no fatigue group, as well as a lower score on the Mental Component summary of the SF-36, higher scores on the HADS depression and anxiety, more painful joints and higher levels of DAS. In the multivariable analysis, only a higher HADS depression score and higher scores on coping with limitations were associated with developing fatigue over time (table 2).

In the fatigued patients, no factors apart from the severity of fatigue itself explained the recovery of fatigue in the univariable and multivariable analyses (table 2).

Table 2 Univariable and multivariable analyses for developing fatigue after 12 month for low fatigued and fatigued patients

	Univariable Odds ratio (CI95%)		Multivariable Odds ratio (CI95%)	
	Fatigue (<21)	**FAS (≥22)**	**Fatigue (<21)**	**FAS (≥22)**
Sex, female	2.60 (0.83-8.09)	1.28 (0.48-3.40)	3.01 (0.84-10.73)	1.83 (0.65-5.01)
Age, per year	0.98 (0.95-1.02)	1.00 (0.97-1.04)	0.97 (0.93-1.01)	1.00 (0.97-1.04)
Education	1.01 (-0.03-2.07)	-0.03 (-1.08-1.01)		
Working status (Y/N)	1.89 (0.67-5.30)	1.28 (0.53-3.06)		
Nationality Natively/Dutch	1.33 (0.29-6.01)	2.98 (0.89-9.95)	7.45 (0.74-74.83)	3.43 (0.99-11.82)*
DAS28	1.96 (1.07-3.59) *	1.45 (0.93-2.25)		
ESR	1.007 (0.98-1.02)	1.00 (0.98-1.02)		
Tender joints (0-44)	1.12 (1.01-1.23)**	1.03 (0.96-1.10)		
Swollen joints (0-44)	1.03 (0.95-1.12)	1.02 (0.96-1.08)		
VAS global (0-100)	1.02 (1.00-1.05)**	1.01 (0.99-1.04)		
Radai (0-10)	1.19 (0.88-1.61)	0.97 (0.75-1.25)		
Hads depression (0-21)	1.20 (1.03-1.40)**	1.04 (0.91-1.19)	1.33 (1.08-1.62)**	
Hads anxiety (0-21)	1.15 (1.01-1.32)*	0.98 (0.86-1.12)		
Coping limitations (8-40)	1.06 (0.99-1.14)	0.98 (0.92-1.04)	1.09 (1.00-1.18)*	
Coping pain (8-32)	1.08 (0.98-1.19)	0.96 (0.87-1.05)		
Physical health (SF36, 0-100)	0.94 (0.86-1.02)	1.00 (0.94-1.07)		
Mental health (SF 36, 0-100)	0.87 (0.80-0.96)**	0.98 (0.92-1.05)		

Level of significance *p=0.05/**p=0.01/***p=0.001 Cut point for FAS≤21 non-fatigue>22 fatigued
Multivariable analysis corrected for sex and age
Abb. ESR: erythrocyte sedimentation rate; VAS: visual analog scale; RADAI: Rheumatoid arthritis disease activity index; HADS Hospital Anxiety and Depression Scale; SF-36: Short Form 36; PCS: physical component summary; MCS: mental component summary

DISCUSSION

In this study, almost half of the early RA patients were fatigued over the first year after diagnosis, although they had been treated by an early, intensive, and tight-controlled strategy. Of those who had no fatigue at baseline, 15% became fatigued, while most of those who were fatigued at baseline (77%) remained fatigued despite lesser disease activity. The minor change in fatigue levels was also reflected in the AUC analysis of all patients, which showed that baseline level of fatigue was the strongest factor in predicting follow-up levels of fatigue. This factor was so strong that adding other variables did not improve the model. In a stratified analysis among the non-fatigued patients at

baseline, higher levels on the HADS were associated with higher levels of fatigue later on.

The literature on the course of fatigue in early RA has been conflicting. A cohort study in early RA patients reported recovery of fatigue over time [14]. Recovery over time is more often observed in studies evaluating biological treatment with longstanding RA patients [34, 35]. However, another cohort study showed persistent fatigue over time with almost no change since diagnosis [13]. Moreover, a recent meta-analysis found that treatment with biologicals only led to a small, but statistically significant, improvement in levels of fatigue [36]. Since our study was performed in an early RA population treated with induction by conventional DMARDs, it is interesting to see a similar pattern, with fatigue decreasing by only 6% over 1 year of follow-up.

At baseline, both inflammatory disease characteristics and patient-reported characteristics were more pronounced in the fatigued than in the non-fatigued patients. It is not clear; therefore, whether fatigue in the present study was related to the disease or to other more personal characteristics. Some direct and indirect observations suggest a less prominent relationship with the disease itself. Over time, disease activity decreased, while fatigue remained present in most patients. In the initially non-fatigued patients, the presence of symptoms related to depression and anxiety, more painful joints, lower scores on the Mental Component summary of the SF-36, and higher levels of DAS, were predictive in the univariate analysis for the development of fatigue. This may suggest that other pre-existing factors contribute to the presence of fatigue, of which depression / anxiety is the most powerful relation. Moreover, the relation of depression in the fatigued patients was also pronounced at baseline. Depression seems to interplay with fatigue in our early RA study population. Depressive symptoms are a common feature of both established and early RA [37] and are associated with fatigue [38]. The direction of the association, thus is depression induced by fatigue or fatigue induced by depression, is under debate. According to Druce et al., both directions are possible [39]. A dynamic conceptual model of RA fatigue showed the bi-directional relation for depression and fatigue [40]. Irrespective of the direction of this relation, the high levels of symptoms related to depression warrant monitoring over time and further examination by a psychologist if symptoms persist.

Several aspects of this study need further discussion. There are many ways to analyse fatigue over time. We used simple logistic regression and the AUC, but also considered longitudinal models taking into account individual patient profiles. These models did not lead to different results and insights than described in the analysis presented here.

In 2003, it had been decided to use the FAS and the VAS fatigue, at a time when not many specific RA fatigue instruments were available. The FAS has a good internal consistency reliability and validity [24, 25]. The lack of a standardized VAS cut-off point for high and low fatigue prevented a clear interpretation of the VAS fatigue scores. We were

able to analyse a substantial number of covariates influencing fatigue, but data, on for example, sleep quality or the presence of symptoms of fibromyalgia were not available. Strong points of this study include its longitudinal design and the use of data of the protocolled medication and tight-controlled treatment. As this study was not an RCT, we could study the longitudinal evolvement of fatigue and the development of fatigue among those patients with initial low fatigue level at baseline and recovery of fatigue among those with initial high level of fatigue. Medication had no effect on the decrease of fatigue in both groups (data not shown).

Given that many patients in this study in early RA showed fatigue, it seems advisable to quantify fatigue and depression in daily care. Our results and those of others suggest that fatigue does not resolve by itself. To facilitate (more) self-management behaviour, it is important to inform patients about the course of fatigue. Nurses are ideally suited to address this topic during consultations.

Screening on fatigue and depressive symptoms at baseline and follow-up will make a patient feel that his or her fatigue is acknowledged, and may also improve patient satisfaction and treatment outcomes.

Conclusion: Despite a strict treat-to-target strategy in early RA patients , fatigue is and remains a problem for many patients. Initial fatigue level is the main predictor for fatigue at follow-up. Higher levels of depression were associated with developing fatigue in initially non-fatigued patients. Discussing and monitoring fatigue and depression at baseline and follow-up might be important to acknowledge its presence and improve patients' self-management of fatigue. Referral to a psychologist may be indicated.

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CHAPTER 5

Focus group interviews reveal reasons for differences in the perception of disease activity in rheumatoid arthritis

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ABSTRACT

Objective

Doctors frequently see patients who have difficulties coping with their disease and rate their disease activity high, despite the fact that according to the doctors the disease activity is low. This study explored the patients' perspectives on this discordance that may help to understand why for some patients usual care seems to be insufficient.

Methods

In our qualitative study we conducted focus group interviews where questions were used as a guideline. Transcripts were analyzed using inductive thematic analysis.

Findings

29 patients participated in four focus groups. Participants could not put their finger exactly on why doctors estimated that their disease activity was low while they experienced high levels of disease activity. During the in-depth focus interviews seven themes emerged that appeared related to high experienced disease activity: 1) perceived stress, 2) balancing activities and rest, 3) medication intake, 4) social stress, 5) relationship with professionals, 6) comorbidity, and 7) physical fitness.

Conclusion

When patients were asked why their view of their disease activity was different from that of their physician, seven themes emerged. The way participants coped with these themes seemed to be the predominant concept. Specific interventions that focus on one or more of the reported themes and on coping may improve not only the quality of life of these patients but also the satisfaction with the patient-doctor relationship for both parties.

Key messages

- Seven themes are identified for discordance of disease activity between patients and physicians namely perceived stress, balancing activities and rest, medication intake, social stress, relationship with professionals, comorbidity, and physical fitness.
- The predominant concept that emerged was the way in which patients ineffectively coped with these themes
- Specific interventions focused on these themes and on coping may improve not only the quality of life of these patients but also the satisfaction with the patient-doctor relationship and the quality of care.

INTRODUCTION

In rheumatoid arthritis (RA), patients and physicians do not always rate disease activity equally [1-4]. Despite the fact that treatment is regarded effective on commonly used disease activity measures (e.g. Disease activity score, Clinical disease activity index) about one third of patients with low disease activity report high levels of pain, functional disability and fatigue [1-7]. This difference is undesirable, as it may affect the patient's satisfaction, adherence to treatment [8,9] and outcome [8]. Differences between patients and physicians regarding the perception of disease activity are not well understood and may relate to various factors.

In the context of shared decision making and patient-centered care it is important to know the patients' thoughts about the high disease activity that they perceive and this possible discordance. Data from cohort studies suggest that in the perception of the patient the most relevant disease activity parameters are pain and fatigue [6], while for the physician the most important parameter is the number of swollen joints [4,10]. Moreover, previous studies suggest a role for factors influencing discordance such as education, health literacy and the concurrent presence of depression [2,4,11]. Furthermore, qualitative studies showed that pain, mobility, fatigue, physical capacity and wellbeing are seen as an important outcome for patients [12,13].

A better understanding of factors – according to the individual patient – that influence the high self-reported disease activity may help to understand why for some patients usual care seems to be insufficient. Therefore, the aim of this study was to explore the patient's perspective on the patient-physician discordance with regard to disease activity in rheumatoid arthritis.

METHODS

To explore patients' perspectives in breadth and depth on the discordance of the disease activity between patients and physicians, a qualitative study was performed by using focus group interviews. Focus groups allowed an interactive discussion on the topic of discordance. This method enabled researchers to explore the experiences, concerns, collective understanding and opinions of participants by discussing specific topics related to discordance of disease activity and generate data [14].

Initial cohort

Patients from the RAPPORT study (Rheumatoid Arthritis Patients rePort Onset ReacTivation), an observational cohort [15], were invited to participate in this study by letter. In brief, RA patients were eligible for this study if they were aged 18 years or older, were

able to read and write Dutch. Further study details can be found in Walter [15]. Of the initial 159 RAPPORTstudy patients, 82 patients (52%) were willing to participate. No significant differences between responders and non-responders were found with regard to demographic characteristics, previous disease activity and previous patient-reported outcome (PRO) scores [15]. The disease activity was measured with the disease activity score (DAS28). This score ranges from 0 to 10 containing swollen joints, tender joints, visual analog scale (VAS) global and erythrocyte sedimentation rate (ESR), where a higher score indicates a higher disease activity.

Patients were asked to complete web-based questionnaires (Health Assessment Questionnaire/HAQ, Rheumatoid Arthritis Disease Activity Index/RADAI and Visual analog scale/VAS fatigue) three times, at three-week intervals, and were clinically assessed by their consultant rheumatology once. Based on the online PROs and disease activity as rated by the physicians, 29 patients were identified as being discordant. According to regulations in the Netherlands (WMO), approval from the ethical review board was not needed. All patients gave written informed consent before inclusion in the focus groups.

Patient selection for the focus group interviews

Patients who had a discrepancy between patient-reported outcomes (PROs) and physician-assessed disease activity were invited to take part in focus groups. Patients were regarded as discordant if they had low disease activity according to the physician and a high PRO score (HAQ>1 [16], RADAI>2.2 [17] and VAS fatigue >50) for two or three consecutive time points. The interview schedule was devised after discussions between a clinical psychologist, nurse practitioner and an epidemiologist as well as a literature search (table 1).

The duration of the sessions was 1-1.5 hours. They were led by a male psychologist. The moderator introduced himself as being interested in this topic and emphasized the confidentiality of the interviews. All interviews were audio taped, transcribed verbatim and anonymized.

Table 1 Interview questions

General question
'What makes you say that your disease activity is high, while the rheumatologist indicates that the disease activity is low?'
Additional questions
- What do you do when your disease activity is high?
- Was your intervention effective?
- Do you believe there are influencing factors that can reduce your disease activity?

Data analysis

Data analysis was based on grounded theory [18]. We adopted not a constructivist approach to grounded theory [19], but adopted a more thematic analysis of the data. After each interview, emergent themes were identified. The principle of data saturation was used. Interviews were held until themes and categories in the data become repetitive and redundant, such that no new information could be gleaned [20,21]. At this point saturation has been reached and depth and breadth of the information was achieved [21]. No new concepts emerged after 4 focus groups (data saturation). Emergent themes from the first interviews were incorporated into the next interviews. When themes were not discussed spontaneously, groups were asked explicitly about themes from previous interviews. After all interviews were held, the transcripts were read and re-read by the principal investigator to gain an overall understanding of the interviews. Then the transcripts were examined, and open coding was applied to individual quotes. Codes were then grouped into concepts and then into major themes. All transcripts were analyzed independently by two of the four investigators (MW, AvtS, AP, JL). To guarantee uniformity, MW was involved in the analysis of all group transcripts. Differences in opinion were discussed until consensus was reached. The final transcript yielded no new codes, indicating data saturation. Analyses were completed using Atlas.ti software.

FINDINGS

Of the 29 patient who were identified as discordant, all subsequently attended a focus group. The demographic characteristics of participants are summarized in table 2.

Patients confirmed that they experienced high levels of self-reported disease activity despite the low disease activity reported by the professionals. If asked directly what could explain the difference between their own experience of high disease activity and the low disease activity according to the doctor, participants could not come up with a clear explanation. However, a clearer picture emerged during discussions of issues brought up by the participants themselves. Seven themes were identified during the inductive analysis, in line with our thematic analysis. In the following section, we elaborate on these themes. The quotes are identified by a participant number (e.g. P1) and a group number (e.g. FG1).

Table 2 Demographic data for focus group participants

	Group 1 (n=7)	Group 2 (n=8)	Group 3 (n=8)	Group 4 (n=6)	Overall 1-4 (n=29)
Female (no.)	5	5	7	6	23
Duration RA, yrs mean (IQR)	8	15.4	16.7	9	12.3 (4-10)
Erosive disease (no.)	1	4	4	4	13
Medication					
DMARD use (no.)	7	7	7	5	26
Biological use (no.)	2	6	3	3	14
Age, yrs (mean,SD)	57.5	56.1	58.7	55.2	56.8 (8.9)
VAS fatigue 0-100 (mean,SD)					67 (12.8)
HAQ, 0-3 (mean,SD)					1.1 (0.6)
RADAI, 0-10 (mean,SD)					2.7 (1.8)

Theme 1 Perceived stress

The participants in our study indicated that higher levels of cognitive stress were associated with pain and functional disability. For example, if they appraised a situation as taxing and stressful and they considered themselves incapable of dealing with it, this increased the likelihood of experiencing symptoms.

Stressful situations at work or in private situations were discussed as an inducer of increased pain and fatigue:

> *Stress is also a big wrongdoer. And I have experienced a lot of stress for several years; and you do notice this, that it has a negative effect on everything, particularly the pain. (P2,FG4)*

Besides the direct impact of stress, patients struggled how to cope with the high self-reported disease activity when they were feeling stressed.

> *Do you not also think that stress, unexpected events can have an influence on this? In my case they do. Like, for instance, the physical examination for my work is a very bad time for me, I feel that in my joints and everything. Because it's on your mind a lot more. What will be the consequences, how will my employer react to this, how will I cope at home, how will I cope financially, so…. (P5,FG2)*

Although they were well aware of the positive impact of low stress levels, it was hard to implement stress-reducing techniques. Patients reported feeling much better

at times when they were able to cope with the pressure or during periods with low external stressors.

Another factor affecting stress was highlighted by specific remarks about the context of health care. If patients felt that their symptoms were not understood by health-care professionals (including doctors) and therefore referred to other health professionals this was experienced as stressful for some patients.

> *Being sent from pillar to post was very stressful for me (P8, FG2)*

Theme 2 Balancing activities and rest

Patients mentioned that activities and rest need to be properly balanced in order to cope with the disease.

> *You have to find the right balance, and no-one can tell you what the right balance is between physical strain and relaxation (P4, FG2)*

To manage activities they used strategies such as planning, adapting and avoidance of regular activities. In order to cope with the pain and fatigue that they expected to experience when taking part in specific activities, patients often made sure that they rested or relaxed in advance. Taking sufficient rest and restricting oneself in activities was hard for many patients. It was notable that patients regarded the need to make adjustments in daily life as a loss. They saw adjustments as an obligation rather than as an investment required to engage in valued activities. The need to rest, which was viewed as loss of usable time rather than energy renewal.

Another way of coping with high levels of disease activity was to ignore the symptoms and carry on. Some patients accepted that this would result in "off days" .

> *And a day like today, I couldn't plan this on time, so for me this is actually already too much. I just know: tomorrow I'll be ill. (P3, FG2)*

A number of patients also simply accepted ongoing high levels of disease activity as they did not wish to – or could not – reduce the level of their activities.

> *To exceed your limits, because either you can't do it any other way or you don't want to do it any other way. And that causes more problems the days after. (P4,FG1)*

The patients also talked about how prior experiences with activities influenced how they made choices regarding current activities. Some patients avoid activities because of these bad experiences:

> *It is an obstacle that you don't do things just because you know they will cause you pain later. (P6,FG1)*

This not only changed their behavior but also made them feel that they were not able to do what "normal" people are capable of.

Theme 3 Medication intake

Most patients felt that medication had a negative influence on their general well-being that was not picked up by the physician's measure of disease activity. They considered the medication side effects to be a potential cause of fatigue.

> *Get tired because of all those medicines. (P1,FG1)*

Because of these side effects some patients considered weighing the side effects against the severity of their disease in their decision whether or not to continue their medication.

> *At some point I really felt like a walking chemical factory.*
>
> *Can you imagine: every week you inject MTX, you take proton pump inhibitors, Naproxen, Arava, an anti-malaria drug, and I was injected with boosts of prednisone. If you still manage to feel OK after that… terrible. (P2,FG4)*

Other patients mentioned that it was hard to disentangle the symptoms caused by the disease from those caused by medication use.

Theme 4 Social stress

Social stress is defined as stress arising from a lack of social support or inappropriate social support.

Patients talked about the lack of social support and affiliation with family and friends. They felt that these people had limited understanding of their illness. Patients discussed feeling down related to the social response as a result of the invisibility of symptoms.

> *I can't shake hands, I usually explain why. The common reaction of people is: It can't be that bad, I don't see anything (P2,FG3)*

For some patients a lack of understanding about the chronicity of the disease created negative thoughts, they always had to explain their condition resulting in more experienced stress.

> *I am being misunderstood by people around me; they keep asking: Have you still not recovered yet? I noticed that being misunderstood also causes me a lot of stress. (P4,FG2)*

Some of them mentioned the same effect of feeling down if doctors ignored their pain and fatigue. Although most encounters only had a brief effect on patients, their recurrence made this a relevant topic for them.

Repeatedly having to ask for help was also difficult for some patients. It created a feeling of dependence on family or friends for everyday life. On the other hand, some patients found it difficult to deal with meddling by their family.

Theme 5 Relationship with professionals

The relationship with professionals and the professionals attitude was not regarded as having a direct negative effect on the self-reported disease activity, but it was suggested that good guidance, personal attention, listening, and taking time during consultations all alleviate self-reported disease activity.

> *If you leave the consultation room feeling badly, this will really affect you a lot. (P7,FG1)*

Patients discussed the importance of being heard, and being listened to, were signs that professionals took them seriously.

> *Well, actually, it is not enough just to treat the illness; you have to treat the patient. And this patient is more than just that illness. (P2,FG2)*

Theme 6 Comorbidity

Patients believed that comorbidity plays a part in the high disease burden, but it was difficult to interpret. Patients could not explain whether high self-reported disease activity was the result of RA or result of comorbidity. For example, if patients experienced pain in their joints they could not distinguish between osteoarthritis and arthritis .

> *And osteoarthritis is also very painful. And that also confuses me, when they ask: are your joints painful? Well, yes, they always hurt. (P6,FG 4)*

A few patients discussed the causes of fatigue. Fibromyalgia or other diseases were mentioned as inducers of fatigue, but so was their RA. Female patients going through the menopause also talked about fatigue, disturbed sleeping and pain in the joint. But they could not say whether it was due to RA or menopause.

> *And menopause, that I have to open the windows, and then you're still exhausted because it's early in the morning and you're still waking up. (P5, FG1)*

Theme 7 Physical fitness

This theme was perceived as having a negative impact on well-being, both psychologically and physically. Patients mentioned that a lack of exercise led to more disease symptoms.

> *So when I sit down I get more overall complaints from my body; you become drowsy, you become tired. But when you move, your back doesn't hurt as much, your hips don't hurt as much, and so on (P5, FG 3)*

> *Becoming unfit, that might worsen the illness. Well, worsen, maybe you just experience a symptom sooner (P6,FG1)*

But the opposite was also reported: more pain and fatigue after exercise or other intense physical activities were mentioned as a reason for staying away from exercise. Although patients were aware of the benefits of exercise, their concerns about the pain and fatigue after exercising resulted in them avoiding all activities related to exercise.

> *If you exercise more, you get more energy. Well, it turned out to be so exhausting that I had to stop doing other things I liked. And it certainly didn't give me more energy, so…it doesn't help. (P4,FG2).*

DISCUSSION

Main findings

We explored the discordance between the patients self-reported high level of disease activity, while at the same time their doctors believed that their disease activity was low. Summarizing the data of the focus groups we found seven themes that according to patients were relevant to high disease activity: perceived stress, balancing activities and rest, medication intake, social stress, relationship with professionals, comorbidity and physical fitness.

Comparison with existing literature

Considering the seven themes that emerged in our study it seemed that the way in which patients coped with these themes may have played an important role in managing the impact of the disease on their daily life. Coping is the process by which people try to manage (e.g. reduce, minimize, master, or tolerate) the internal and external demands that are appraised as taxing or exceeding the resources of the person [22,23]. Maladaptive coping may lead to loss of confidence, increased likelihood of perceiving a situation as stressful and loss of control [23], all of which hinder a person's ability to adapt to living with a chronic illness [24,25]. Results from RA studies suggest that problem-focused coping strategies are helpful in placing the disease into perspective, thereby allowing patients to better manage the disease burden [26]. Furthermore, studies have shown that active coping strategies appeared to be useful in RA patients and that these strategies improve psychological well-being [27].

Appraisal of the situation and employment of coping strategies differ considerably between patients. Some patients successfully adjusted to their disease while others did not, which is known from the literature [23,28]. This was for example seen in physical activity, patients often tried to follow advice to do more exercise. However, some experienced more complaints hereafter. An explanation for this finding might be that initially RA comes with high levels of pain related to movement and exercise. These high levels of pain could lead to avoidance behaviour, passive coping, based on fear of more pain [29] leading to a decrease in physical condition.

Also, the need for medication was viewed by some patients as a necessary evil with side effects implicating lower levels of health. From previous studies we know that negative beliefs about medication can influence medication uptake, adherence and the degree to which side effects are experienced [30,31]. Patients with a more positive outlook on medication were more confident about the ability of DMARD treatment to control their RA [32]. It is therefore possible that negative thinking about medication, has an impact on the experienced symptom reduction of known effective drugs (i.e. the nocebo effect).

At the same time, passive coping with pain and neglecting social support is known to have a negative impact on long-term disability and pain [33]. In this study, patients mentioned the importance of the support of professionals. This was in line with other studies where it was found that psychosocial factors influenced patient outcomes, but not disease activity [34,35].

In the present study, we formulated the main question from the perspective of the professionals as we regarded low disease activity measured by DAS28 as treatment effectiveness. Some work has been done in this field by asking what patients perceived as disease remission. This resulted in themes such as absence or reduction of symptoms, ability to do valued activities and the ability to cope with the disease [36]. The patients

and the doctors perspective on low disease activity thus appear to be based on different factors.

Strengths and weaknesses

There are a number of limitations to this study. First, the results applied to patients with a median disease of 5 years. It is possible that patients with recently diagnosed RA would have identified different themes. Second, patients who were willing to participate were mainly Caucasian. It is possible that other ethnic groups would have discussed other themes. Third, we think that coping style may be seen as an important factor in the difference between physicians and patients assessment of the disease activity. Although coping style is often seen as a stable personality trait, coping style in chronic patients may be influenced by prolonged experience of stress that comes with rheumatoid symptoms. Therefore, coping style may have changed resulting in less optimal coping strategies. It is possible that our patients used less often problem-focused strategies, which hinder these patients to manage the burden in a more effective way [26]. This relationship could be bidirectional as prolonged experience of stress influences the presence of rheumatoid symptoms. This is not unlikely, because prolonged worry and anticipatory stress can influence somatic immunologic functioning [37].

Fourth, the focus groups consisted mostly of female patients (80%). This could be the result of the fact that women are more likely to be discordant with the physicians [38]. However, male patients might have added different items which makes that the results should be applied to female RA patients. Furthermore, depression was not measured in this study. Previous studies suggest a role for depression in patients with high levels of symptoms and estimated low disease activity. It is known that depression influences coping style, and thus depression could have influenced our results [33]. Although co-morbidity was a theme of the discordance in disease activity, this study was not designed to specifically analyse this issue.

Implications for clinical practice

Our results suggest that there are options for improving the quality of care for RA patients, which involve changing maladaptive coping styles by teaching them in problem solving techniques, transform negative thoughts (i.e. cognitive training), physical training and the need for rest to renew energy. Doctors frequently see patients who had difficulties coping with their disease and who are discontented, despite the fact that, according to the same doctors, their disease activity is low. This discontentment may even lead to inappropriate drug treatment as patients may request to intervene in their symptoms, possibly initiating expensive drug instead of more appropriate non-pharmacologic management interventions. Specific interventions focused on one or more of the themes reported here may help to increase contentment, thereby improving not

only the quality of life but also the satisfaction with the patient-doctor relationship for both parties.

When we asked patients why their view of their disease activity was different from that of their physician, seven themes emerged. The way in which patients coped with these themes in demanding situations may be the overarching theme.

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CHAPTER 6

Is tight controlled disease activity with online patient reported outcomes possible?

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ABSTRACT

Objective

To evaluate the performance of patient reported outcomes (PROs) as primary indices for identification and prediction of a DAS28>3.2 among patients with RA.

Methods

RA arthritis patients completed monthly online PROs (HAQ, RADAI, VAS fatigue) and were clinically assessed every 3-months by the DAS28. Simple descriptive statistics, logistic regression and Bayesian joint modelling approach were used to analyse the data. The Bayesian joint model combines the scores and changes in scores of three PROs to predict a DAS28>3.2 at the subsequent time point.

Results

159 RA patients participated in this study. Stratified summaries of the PROs by DAS28 categories at baseline provided incremental values of the PROs for more active disease. However, on individual level the DAS28 and the PROs fluctuated over time. The prediction of subsequent DAS score by a single instrument at single time points resulted in moderate sensitivity and specificity. Using the intercept and slope of the combined PROs of the first 3 measurements to predict the DAS28 state at 3 months resulted in a sensitivity of 0.81 and a specificity of 0.92. After 10-fold cross validation the model had a sensitivity of 0.61 and specificity of 0.75 to identify patients with a DAS28 >3.2.

Conclusion

PROs showed fluctuating levels of disease activity over time while on group level disease activity stayed the same. Using the evolution of RADAI, HAQ and VAS fatigue over time to predict future DAS28>3.2 resulted after the internal cross validation of the model in moderate performance (sensitivity 0.61, specificity 0.75).

INTRODUCTION

Tight control of disease activity is commonly monitored by clinical disease activity measures such as the Disease Activity Score(DAS) [1] or SDAI [2]. These measures, administered by physicians and nurses, combine tender joint count (TJC), swollen joint count (SJC), ESR and a patient reported measure of experienced global disease activity (VAS global). The SDAI also includes the evaluator global assessment of disease activity. As the DAS/SDAI are administered clinically and are therefore only measured during consultations, they may miss relevant fluctuation in disease activity over time, especially in patients who attend the clinic only once or twice a year.

An alternative way to assess disease activity could be the use of patient reported outcomes (PROs).These measures can be provided to patients in a web based manner enabling distant-monitoring of disease activity by the rheumatologists. There are many disease-related self-reported instruments such as the HAQ [3], RADAI [4], SF-36 [5], and the AIMS2 [6], which may have the ability to function as self-monitoring instruments at home. There is no single "gold standard" measure [7, 8] that can serve for identification and prediction of a high disease activity state (DAS28>3.2) in patients with RA, that is indicative for treatment escalation [9]. There is also no core set of patient reported outcomes generally used in studies that seems to fit the purpose of monitoring disease activity by patients [10, 11]. Self-assessment of the DAS components demonstrated moderate to low correlations with a trained assessor especially for the swollen joint count [12, 13]. Besides the unavailability of ESR limits the use of the unmodified DAS28 score by patients themselves. While there are many studies of self-reported measures, no studies are available that investigated combined self-reported measures as an instrument that uses PRO data to predict subsequent DAS28>3.2.

In this study we aim i) to describe the change of functional status (HAQ), self-reported disease activity (RADAI), VAS fatigue and the DAS 28 ii) to identify patients with DAS28 >3.2 by the proposed cut off points for RADAI and the HAQ iii) to evaluate the performance of the HAQ, VAS fatigue and RADAI as primary indices for identification and prediction of high disease activity (DAS28>3.2).

METHODS

Participants

We recruited patients with clinical RA from one outpatient rheumatology clinic in Rotterdam. Participants were identified from the hospital record database and invited by their rheumatologists. Data was collected on demographics, DAS28 and patient reported outcomes. Patients were eligible for inclusion if they were aged 18 years or older, were

able to read and write in Dutch and had access to a computer with internet and email facilities. As some patients wanted to participate but had no computer we accepted that they could participate by completing the questionnaires in paper version. We excluded patients with severe psychiatric illness or personality disorders. All patients signed an informed consent before study enrolment. The study was approved by the independent medical ethic committee of the Erasmus MC.

Procedures

The duration of this study was one year. The patients were clinically evaluated using the DAS28 every three months in standard care by his/her rheumatologist or nurse practitioner. These patients were also asked to complete a web-based patient reported questionnaires each month over a 1 year period. The web-based questionnaires were easy to complete, no specific instruction was given. They were reminded for their next questionnaire by email. Non responders received two reminders by email and one phone call.

Primary outcome measures

The DAS28 was used as primary outcome and as reference standard for a moderate to high disease activity (DAS28>3.2). The DAS28 is a score ranging from 0 to 10, where a higher score indicates a higher disease activity. Treatment escalation was indicated if DAS28 exceeded 3.2 points according to Dutch guidelines for treatment of RA. We used the clinical cut off point of 3.2 for the identification of a moderate to high disease activity [9, 14]. Data from the DAS28 were collected at baseline and at 3, 6, 9 and 12 months.

Patient reported Outcomes

A choice was made for instruments that reflects different aspects of disease activity and that were available in Dutch. Over a one year period we measured the following PROs monthly: HAQ [3], RADAI [4], VAS global [15] and the VAS-fatigue [16]. The HAQ (Health Assessment Questionnaire) measures functional status using 20 questions phrased in eight subscales [3]. The score ranges from 0 to 3 (3 worst health) with a minimal clinical important difference of 0.20 [17, 18]. The HAQ score suggested for remission '≤ 0.5', representing patients with almost no difficulties in daily activity. A HAQ score between 0.5 and 1.0 could be regarded as low activity, while a score above 1 would indicate moderate to high disease activity with major problems in preforming daily activities [19]. The HAQ is commonly used in clinical trials. It is an effective and sensitive tool for measuring the functional status and it is correlated with the DAS score [20]. The HAQ is also a predictor of severe long-term outcomes, so it measures both disease course as well as outcome [21]. The RADAI (Rheumatoid Arthritis Disease Activity Index) measures self-reported disease activity [4]. It uses a scale ranging from 0 to 10, where higher scores indicate

more disease activity. The cut points for the RADAI are: < 2.2 for low disease activity, ≥ 2.2 and ≤ 4.9 for moderate disease activity and > 4.9. for high disease activity [8]. The RADAI measures similar domains as the DAS, but laboratory values are not required. It has shown to be reliable and sensitive for change compared to the DAS28 although low concordance for absolute values between RADAI and DAS28 were observed [22]. The VAS (visual analogues scale) fatigue and VAS global are both single-item scales and measure one domain each. The VAS fatigue asks about the severity of the fatigue over the past week with the anchors: no fatigue (0 mm) and extremely fatigued (100mm) The scale is sensitive to change[16, 23], valid [16, 23] and reliable [16, 23]. Although there are minimal important differences for the VAS fatigue for improvement and worsening available, there is no guidance on the choice of cut off points [24], while in general 10 mm change of the VAS seem to be a clinically detectable difference for patients [25]. The VAS global was also considered but as it is one component of the DAS28, it was not evaluated for predictive capacity of the DAS28 states. Each of the PROs was completed at 1-month intervals.

Covariates

In addition to the PROs, we also measured the effect of coping, self-efficacy and illness perception on both the primary outcomes and the PROs. We selected these questionnaires as the underlying constructs may influence the observed relationship. Coping was measured via the CORS scale which is based on the frequency of individual coping efforts. It is divided into five scales of which we used dealing with pain (decreasing activities), limitations and optimism [26]. The self-efficacy was measured with the Dutch version of the arthritis self-efficacy scale, which constitutes of two subscales related to self-efficacy to deal with pain and to deal with other symptoms (depression, fatigue, frustration) [27]. Illness perception was assessed by an 11 item list including aspects like perception, causes, experience of symptoms, consequences, timeline and controllability of the disease [28].

Statistical analysis

Sample size

No formal sample size calculation was preformed due to the exploratory nature of this study. We estimated to find around 20% patients with a change into high disease activity annually [29], leaving us with at least 30 cases of a flare of the disease activity among 150 patients.

Analysis

Simple descriptive statistics and diagrams were used for the pattern of change over time of the HAQ, RADAI, VAS fatigue, VAS global, medication and the state of the DAS28. The cross sectional relationship between the DAS28 and the PROs was evaluated by

Spearman's rank correlation. Sensitivity and specificity of the proposed cut points of the HAQ and RADAI to identify a high disease activity state were compared to the clinical DAS28>3.2. Difference in PROs between patients with missing values on the DAS28 and patients without missing values were tested with the Mann-Whitney test.

To evaluate the predictive capacity of the PROs for individual patients we used a Bayesian joint modeling approach. This method is described in detail in MohdDin et al (submitted). With this model we try to predict a moderate disease activity state (DAS28>3.2) at subsequent time point by the evolvement of the HAQ, RADAI and VAS fatigue over time. In summary, the following steps were taken: first the skewed distributions of the PROs were transformed to values between 0 and 1 which, after a logit transformation, results in a normal distribution [30]. Second, the evolution of each PRO during a three month period was summarized into a random intercept and random slope by fitting linear mixed effects models. These two parts of each PRO reflect the part that is stable for the individual patient (random intercept) and the part that changes over time (random slope). Third, the intercepts and slopes of each PRO were used to estimate the DAS28 at the subsequent time point corrected for age, sex, self-efficacy, coping with pain, and two questions of the illness perception. The latter two steps were done at once in the Bayesian joint model, but for simplicity described here as two steps. This model resulted in predicted DAS28 values. These were classified into DAS28 ≤ 3.2 and DAS28 >3.2. The predicted responses were then compared to the observed values of DAS28 and fitted by an ROC for discrimination and plotted for calibration [31]. The model described here was developed using the measurements recorded at baseline, months one and two to predict the DAS28 at month three. This selection of data was made because after three months the clinical evaluation could alter the course of disease by the adjustment of medication. This was likely to impact the scores on both the PROs as well on the subsequent DAS score. The model developed on the first three months data was internally validated by 10-fold cross validation.

Simple descriptive statistics were performed in STATA (version 11). The transformation of the PROs was performed in SAS. The Bayesian joint model was developed in WinBUGS and R [32].

RESULTS

Patients

159 out of 174 invited patients with RA consented to the study; 76% were female; the average age was 54 yrs (sd13.26); with a median disease duration of 4.5 years (min-max:1-38 yrs). 37% of the patients had radiographic damage. Over time, medication did not change for 33% of the patients, 16% had a decrease in their medication by either

type or dose, 39% had an increase and 10 % had a temporally increase via a glucocorticoid injection (triamcinolonacetonide intramuscular or intra-articulair). Further details can be found in table 1.

The majority of patients (90%) participated by internet questionnaires while 10% completed paper versions. Complete DAS28 data was available for 97 out of 159 patients (61%), 19% missed one DAS28 evaluation and 20% missed 2 time points or more. There was no significant difference between patients in DAS28 and PROs with one or more missing values of the DAS28 using the Mann- Whitney test. For the PROs complete data for all 13 time points were available for 64 out of 159 patients (47%), 29% missed 1-3 self-reported time points and 24% missed 4 or more time points.

Table 1 Patients' Characteristics at baseline (n=159)

Characteristics	Estimates
Demografic details	
Age, years, median (IQR)	55.7 (45.6-62.8)
Female, (%)	76.1
Ethnicity, (%)	
Native, (Dutch)	80.3
Duration of disease, median, yrs (IQR)	4.5 (2.0-7.0)
Working status, (%) (n=154)	
Paid work	41.5
Education (%) (n=154)	
Higher education	23.1
Clinical measures	
Rheumatoid Factor positive,(%)	60.8
Anti-CCP positive,(%)	52.8
Radiographic damage,(%)	37,0
DMARD Medication (%)	
No medication	6.3
DMARD single	23.9
DMARD combination	49.7
Biological	20.1
DAS 28, median (IQR)	2.66 (2.01-3.44)
Patient reported measures	
HAQ score (0-3), median (IQR)	0.62 (0.13-1.00)
RADAI (0-100), median (IQR)	2.00 (0.84-2.99)
VAS-fatigue (0-100), median (IQR)	50 (29-70)
VAS global (0-100), median (IQR)	40 (21-58)

Values are the mean, IQR unless otherwise indicated. DMARD= disease modifying anti rheumatic drugs; RF= rheumatoid factor; DAS 28= Disease activity score in 28 joints; HAQ= health assessment questionnaire; RADAI=rheumatoid arthritis disease activity index;VAS= visual analog scale.

Clinical disease activity and evolvement over time

Median disease activity measured by the DAS28 was 2.66 (IQR 2.01-3.44) with 0.88 (sd 1.82) swollen joints and 2.1 (sd 4.34) tender joints at baseline. 47% patients were in DAS28 remission (DAS28<2.6), 19% patients had low disease activity and 34% patient had moderate to high disease activity (DAS28>3.2). Over time there was little evolution of the DAS28 on group level while on individual level patients showed changes in their DAS28 score (figure 1). Table 2 shows the percentage of patients changing. Between 6% and 14% of the patients had a change to a high disease activity (DAS28>3.2) at any of the time points. Continuous low disease activity was seen in 48% (n=76) of the patients.

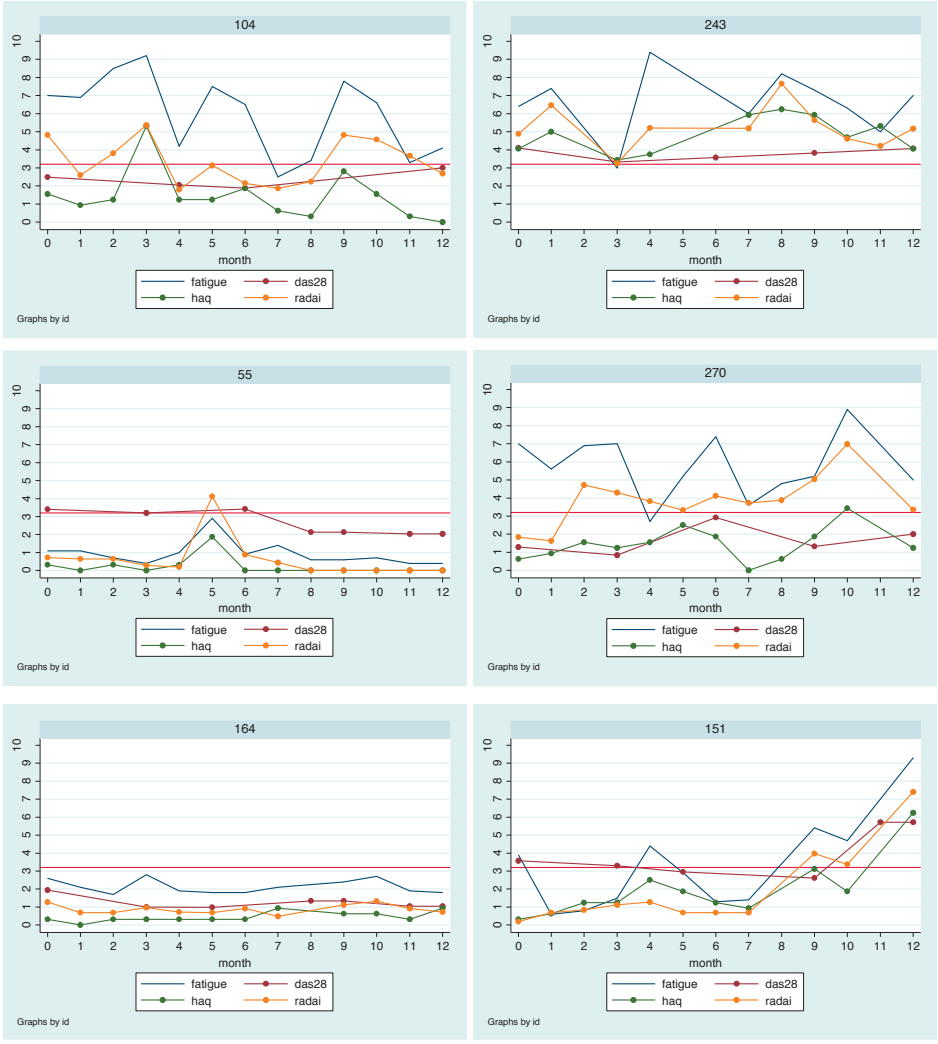

Figure 1 Individual patterns from six patients of DAS28 and PROs

Table 2 DAS28 changes over time presented as number of patients with a change (%)

	Change ≤0.6	**>0.6 & ≤ 1.2**	**> 1.2**	**DAS28≤3.2**	**DAS28>3.2**
T0=T3 (n=146*)	81 (56.3)	43 (30.0%)	20 (13.9%)	16 (11.1%)	15 (10.4%)
T3-T6 (n=130)	77 (59.2%)	30 (23.1%)	23 (17.7%)	18 (13.9%)	8 (6.2%)
T6-T9 (n=112)	61 (54.6%)	30 (26.8%)	21 (18.8%)	9 (8.0%)	16 (14.3%)
T9-T12 (n=102)	58 (56.9%)	25 (24.5%)	19 (18.6%)	10 (9.8%)	8 (7.8%)

*Data presented for patients who had data for both time points

Self-reported measures

Self-reported physical functioning on the HAQ resulted in a median of 0.62 (IQR 0.125-1.00) at baseline. Self-reported disease activity (RADAI) was low with a median of 2.0 (median; IQR 0.84-2.99). General health (VAS global) was scored 40 (median, IQR 21-58) just below half of the scale while fatigue had a median value of 50 (IQR 29-70). The median values of the different PROs did not evolve much over time, while individuals showed substantial variation as reflected in figure 1 and table 3. Figure 1 shows the variation in self-reported measures rescaled to 0-10 scores for six patients (whom all reflected different patterns) and table 3 provides the minimal clinical important worsening for three subsequent monthly measures.

Table 3 Number of patients with a worsening on the PROs using the minimally clinically important difference*

	HAQ (MCID: 0.2)	**RADAI** (MCID: 1.0)	**VAS Fatigue** (MCID: 10mm)	**VAS global** (MCID: 10 mm)
T0-T1 (n=140)	33 (23.6%)	66 (46.1%)	47 (34.1%) (n=138)	35 (25.0%)
T1-T2 (n=133)	25 (18.8%)	22 (16.3%)	30 (23.1%) (n=130)	30 (22.9%)
T2-T3 (n=135)	18 (11.1%)	18 (13.5%)	37 (25.2%) (n=147)	25 (18.6%)
T0-T3 (n=142)	27 (19%)	43 (30.3%)	36 (24.8%) (n=145)	40 (28.1%)

Association between PROs and DAS-28 scores

We used various ways to estimate the relationship between PROs and DAS28. First, we looked at group level at the correlation between DAS28 and RADAI, HAQ, VAS fatigue. The Spearman rank correlation coefficient varied between 0.29 and 0.51 with the lowest correlation for the VAS fatigue. Second, we categorized the DAS28 in disease activity states and summarized the PROs per disease state. This provided incremental values of the PROs for more active disease (table 4).

Third, we tested the discriminatory properties of the PROs using their proposed cut points to identify patients with moderate to high levels of disease activity (DAS28>3.2). For the RADAI (cut point <2.2) sensitivity at time point two for the DAS28 at time point three was 0.63 (CI 0.48-0.77) and the specificity 0.71 (CI 0.59-0.79). For the HAQ this was 0.43 (0.29-0.59) and 0.90 (0.81-0.96) for the sensitivity and specificity, respectively.

Table 4 Patient reported outcomes stratified for disease activity states according to the Das28

DAS28	HAQ (median; IQR)	RADAI (median; IQR)	VAS Fatigue (median; IQR)
<2.6 (n=72)	0.19 (0.00-0.50)	1.09 (0.60-1.82)	43 (20-66)
2.6 ≤3.2 (n=29)	0.50 (0.25-1.12)	1.47 (0.92-2.20)	53 (21-79)
>3.2&<5.1 (n=39)	0.75 (0.38-1.38)	2.73 (1.46-3.61)	59 (40-70)
>5.1 (n=9)	1.25(1.00-2.13)	4.53 (4.05-4.81)	80 (75-82)
Overall (n=159)	0.50 (0.13-1.00)	1.63 (0.84-2.99)	56 (29-70)

Change of PROs to predict DAS28

Our aim of this study was to identify patients with a DAS28>3.2 by the changes of the PROs over time. We therefore modelled the DAS28 at month three using the combined scores and evolvement over time of the HAQ, the RADAI and the VAS fatigue. This was done using a joint modelling technique of Bayesian approach. The RADAI and HAQ of the random intercept had a "Bayesianly significant" positive relationship with the DAS28 at month 3, while male had a significantly negative relationship (table 5).

Table 5 * Bounded outcome score responses for HAQ, RADAI, and VAS Fatigue

Parameter		DAS_T3 vs 3 PROs month 0,1,2		
		Mean	SE	95% CI
Intercept	$\beta 0$	3.04	0.53	(2.01, 4.06)
HAQ				
Intercept	β_1	0.19	0.09	(0.02, 0.36)
Slope	$\beta 2$	-0.06	0.96	(-1.99, 1.85)
RADAI				
Intercept	$\beta 3$	0.28	0.12	(0.05, 0.53)
Slope	$\beta 4$	1.08	1.02	(-0.04, 4.12)
Vas fatigue				
Intercept	β_5	-0.01	0.11	(-0.23, 0.20)
Slope	β_6	-1.95	2.24	(-7.33, 1.33)
Age	β_7	0.01	0.01	(-0.01, 0.02)
Sex (Male)	β_8	-0.78	0.25	(-1.26, -0.29)
Self-efficacy	β_9	-0.004	0.01	(-0.02, 0.02)
Coping pain	β_{10}	-0.10	0.05	(-0.20, -0.0002)
Coping optimism	β_{11}	-0.02	0.05	(-0.11, 0.07)
Control Illness	$\beta 1_2$	-0.02	0.04	(-0.09, 0.05)
Experience symptoms	$\beta 1_3$	0.07	0.04	(-0.01, 0.16)
Sensitivity			0.61* (0.81)	
Specificity			0.75* (0.92)	

*Results of the 10 fold cross validated model (initial values of the development model)
Estimate = posterior mean of estimated regression coefficient, CI= confidence interval

In addition, gender and coping with pain were significantly related (table 5). Our initial development model would correctly identify 81% of the patients with a DAS28>3.2 and 92% of the patients with a DAS≤3.2. However after 10-fold cross validation, a technique that corrects for over optimism in the development model, 61% of the DAS28>3.2 patients were correctly identified and 75% of the patients with a DAS28 ≤3.2. The positive likelihood ratio was 2.7 and the negative likelihood ratio 0.51.

DISCUSSION

Patient reported outcome measures (PROs) are valuable tools in the clinic to guide treatment in addition of disease activity measures performed by the physician. In this study we aimed to assess the predictive capacity of PROs in relation to the subsequent scores on the DAS28. Moderate sensitivity and specificity were seen for the performance of one single PRO as primary index. Combining the monthly measurements of three PROs using an advanced Bayesian statistical model, taken into account the score and the evolvement of the PROs over time, the sensitivity to identify patients with a DAS28 >3.2 at month three was 0.61 and the specificity was 0.75. From discussions with rheumatologists we know that there is a strong need to reduce the pressure on their schedule. One way they would like to do that is by observing the patients with PROs in those with low levels of clinical disease activity. The patients could complete the PROs at home and with stable levels of the PROs they only need to come in once a year for clinical assessment. With the current instruments this approach is not feasible because a substantial portion of patients with high levels of clinical disease activity would be missed.

To the authors' knowledge, this is the first study that investigates distant monitoring of disease activity using web-based PROs to predict a high disease activity state. Although there are studies that investigated the relation between the PROs and the disease activity, none of them evaluated the predictive value of the PROs. One study used the MCID on the PROs to predict treatment response on a subsequent time point but did not report any risk parameter that quantified the contributions of PROs in the prediction of low disease activity [33].

An important finding of this study was that individual patients show fluctuating patterns of the patient reported outcome instruments and the DAS28 (figure 1). While on group level the measurement did not evolve much over time. This individual fluctuation on PROs was also demonstrated by Blanchais using the RAPID4/3 (patient reported measurement: physical function, pain, global estimate and self-reported joint counts) weekly [34]. One would expect that an increase of the PRO values over time, that indicates patient reported disease worsening, would result in a high disease activity state as measured by the DAS28 at subsequent time point. This was however not the

case. The PROs slopes (evolvements over time), which were captured in the Bayesian model, did not significant contribute to the prediction of the high disease activity. This may indicate several things. First, it could be that the time frame of the study should be weekly rather than monthly to be able to measure closer to the moment of clinical disease flare. Second, three time points that reflect an individual patient trajectory may be too little. The possible change of medication after each clinical visit limited us to use more of three consecutive monthly measures to predict the outcome of the subsequent clinical DAS28. Maybe the use of more measurements in this model would improve prediction of individual outcomes. Third, the fluctuating patterns may be influenced by other factors unrelated to disease activity (e.g comorbidity). This may be especially true for fatigue which seems not only be driven by disease activity.

As we suspected several personality aspects to influence the relationship between DAS and PROs, we measured coping, self-efficacy and illness perception at baseline. However, in the analysis most of the measured personality aspects did not contributed to prediction of the model. Only gender and coping with pain were significant.

In this study we needed to make choices that may raise discussion. First our choice of using the DAS28 to measure high disease activity. In daily clinical practice the DAS28>3.2 is commonly used for treatment intensification and when the study was designed probably also a valid option in our study design. Regarding the recent discussions within the OMERACT working group on flare this maybe a conservative way to identify patients needed treatment intensification. In their view, shared by us, a flare represents a change in multiple variables that requires treatment change [35]. These variables are patient global assessment, pain, swollen joints, tender joints, function, physician global assessment, and fatigue [36]. Second, there are several PROs available (RAPID3, PASS, RADAI-5 and the RAID) that could be used for the evaluation of disease activity, but there is no consensus what would be best to use. Hence, we chose PROs that were familiar to us, which were validated (in Dutch) and reflecting different domains of disease activity: HAQ,VAS global, VAS fatigue and RADAI. Reanalysing the data with the RADAI-5 [37, 38] and a modified version of the RAPID3 [39] did not alter the observation of moderate performance of the PROs. In an additional cross sectional analysis (time 3 to time 3) the Spearman rank correlation coefficient between the Das28 and the modified RAPID3 was 0.54. The correlation between the DAS28 and RADAI-5 0.49. In the longitudinal analysis the predictive value (time 2 PRO predicting time 3 DAS28) resulted in a sensitivity of the modified RAPID 3 of 0.29 and 0.24 for the RADAI-5. The specificity was 0.84 for the RAPID3 and 0.86 for the RADAI-5.

Limitations of our study include the choice of the timeframe for clinical evaluation and the limited number of patients that had a relevant change of the DAS28 score. Ideally, we would have liked to evaluate the DAS28 each month as we did with the PROs. However, that meant that patients had to come to the clinic each month which was regarded not

feasible for them. We therefore decided to go with a timeframe of each 3 months. The change in the DAS28 >1.2 points we regarded relevant occurred in 20 patients in the first 3 months. Which is good from a clinical view point as most patients were under control, but from a prediction view point this change may be too little to have sufficient power to assess the impact of the PROs. One way to solve this is to study larger samples as established patient well-under control are expecting to change little in levels of their disease activity. In addition, we had to deal with missing values on both the clinical DAS28 as well as the PROs. The missing values on the PROs are likely related to the high frequency of measurements (13 time points). The monthly questionnaires were for some of the patients a too high burden. To reflect the observable patterns in the data we decided not to impute data if patients had one time point of PRO data missing. The problem of missing data on the outcome (DAS28) was solved by the Bayesian model.

A strength of our study was using a web based environment allowing patients to assess their disease activity at home. Collecting data in a web based environment has been tested and validated before on patients with a rheumatic disease [40-43], and worked well in this study. Computerized versions offers advantage over the paper version (less time-consuming) without compromising on the data validity.

Conclusion

PROs are very valuable and may give additional information about the patients. Patients reported outcomes showed fluctuating levels of disease activity over time while on group level disease activity stayed the same. Using the score and evolvement of RADAI, HAQ and VAS fatigue over time to predict future DAS28 moderate to high disease activity resulted in sensitivity of 0.81 and a specificity of 0.92 in the development set. However, the internal cross validation of the model resulted in moderate performance (sensitivity 0.61, specificity 0.75). Further research is needed to investigate the possibilities of using PROs as predictors for clinical disease activity.

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CHAPTER 7

Is a smartphone application useful for self-management support in patients with a rheumatic disease?

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ABSTRACT

Aims

This study 1) investigates whether the use of an app improved patients' self-management 2) investigates which factors are associated with the use of the RD-app, 3) explored patients' experiences with this RD-app.

Background

Self-management is an important aspect in the care for patients with a rheumatic disease (RD). To support patients' self-management, we developed and implemented a smartphone-application (RD-app).

Design

A prospective before-after study was performed among patients with a RD.

Methods

The primary outcome was patients' self-management measured with the Partners in Health-scale (PIH). Survey questions addressed whether the RD-app had contributed to get more hold on the disease and how. A paired t-test was used to evaluate changes in the PIH-scale score after three months. Logistic regression analyses served to investigate variables that are important for using the RD-app.

Results

Of the 1511 eligible patients, 397 completed both the baseline and the follow-up surveys. Participants with positive expectations of the RD-app for getting hold on the disease were more likely to use the RD-app. 114 participants used the RD-app, of which forty-two percent of the app-users perceived that use of the RD-App had contributed to get more hold on the disease. This percentage was higher for those who used the RD-app more frequently (p=0.04). The PIH-scale score in the app-users group had not changed after 3 months. Receiving tips, information on exercises and gaining insight in self-reported disease activity contributed to get more hold on the disease.

Conclusion

Almost one third used the RD-app. If they used the RD-app, almost half experienced more hold on the disease. Positive expectations are an important factor for the use of the app.

Relevance to clinical practice

The RD-app can be useful for additional self-management support in a clinical pactice.

INTRODUCTION

Patients with rheumatic disease (RD) can face several physical (e.g. pain, stiffness, disability and fatigue) and psychosocial problems that might influence their activities in daily life [1]. Sometimes emotional, psychosocial adjustments and behavioral changes are needed [2, 3]. This might require a great effort; patients daily have to make decisions to self-manage the disease and are expected to take an active role in this process. Self-management is considered highly in chronic care and it needs to be integrated in a patient's life [4, 5, 6].

Self-management is not clearly defined; a commonly used definition is: 'the individual's ability to manage the symptoms, treatment, physical and psychosocial consequences and life style changes inherent in living with a chronic condition' [7]. Effective self-management approaches might result in physical and psychosocial benefits and improve quality of life [7, 8]. Self-management in RD has primarily been seen as patient's own task, although receiving support to optimize self-management behavior is appreciated [9, 10, 11].

Self-management support can be defined as the provision of interventions to increase patients' skills and confidence in managing their chronic condition [12]. Patients are activated and facilitated to play an active role concerning living with a chronic disease. Besides the support given by nurses in daily care, an additional way to assist self-management support is by using a health-related application (App).

Background

Apps are becoming a part of the nursing practice and it has been suggested that 'nurses must be able to recommend and integrate apps into their clinical practice' [13]. Health-related apps might contribute to assisting self-management by providing information, advice, support, encouragement and tools for monitoring the disease activity and might overcome several barriers like time and distance [14, 15, 16]. Furthermore, apps can provide efficient and individual tailored information at the time suitable for patients [17, 18] and a way for patients to become more involved with their self-management of the disease [19].

There are many commercial health-apps available, however, most of the apps have a paucity of high-quality and the content is not evidence based [19, 20]. Only a few studies have been done on health-apps for patients suffering from a RD [21, 22]. These studies showed that apps can be useful to assess gait pattern in rheumatoid arthritis patients [22] and to predict the disease activity by combining subjective measurements of joint symptoms, degree of disability and objective gait balance measurements [21, 22]. Patients with rheumatoid arthritis stated they would certainly use an app for self-management support [23].

In 2015, the Rheumatology department in the Erasmus University Medical Center, Rotterdam, the Netherlands, developed and implemented an app for patients with a RD with the purpose of assisting patients to get more hold on the disease.

The aim of this study was 1) to investigate whether the implementation of this newly developed RD-app can help improve self-management of patients with a RD and 2) to investigate which factors are associated with use of the RD-app, 3) to explore patients' experiences with this RD-app.

METHODS

Design and data collection

A before-after study was designed to measure the effects of use of the RD-app on patients' self-management behavior after implementation of the RD-app.

We recruited patients with RD from the outpatient rheumatology clinic of the Erasmus University Medical Center in Rotterdam between August 2015-July 2016. Patients were eligible for inclusion if they had access to a computer with internet and email facilities. All eligible patients whose email address was noted in the electronic patient record received an invitation by email to participate in this study (n=1511). After they confirmed to be willing to participate, they received a questionnaire before (baseline) the launch of the RD-app. All patients who participated at baseline received a second questionnaire three months after the launch of the RD-app. Reminders to return the questionnaire were sent after one and two weeks at baseline and after the second questionnaire by email. At baseline, demographic data, working status, diagnosis and time since diagnosis were obtained.

No extra instructions were given on how to use the app, as we thought it was self-explanatory. Furthermore, no instructions were provided to the (minimum) number of times participants had to use the app nor which categories they had to use specifically.

The study protocol was approved by the Medical Ethical Committee of the University Medical Center Rotterdam (MEC-2015-317). All participants gave written informed consent before completing the baseline questionnaire.

Development of the RD-app

The RD-app was developed by a team of specialist nurses, rheumatologists, a professional software developer and patients. Different self-management components were incorporated, such as education with disease-specific information, self-monitoring and medication management, lifestyle e.g. exercise, as suggested by studies on self-management interventions [7]. The development of the app started by creating a functional design by the team mentioned above. To make sure the design fulfilled the needs

and expectations of patients with RD, a total of 61 patients completed a questionnaire asking for their opinions. This design process resulted in a definitive design consisting of the following six functionalities:

1. Patients can monitor the disease activity using specific self-reported outcomes (e.g. Rheumatoid Arthritis Disease Activity Index, Bath Ankylosing Spondylitis Disease Activity Index, Health Assessment Questionnaire). Thus, this category can be used for self-monitoring the disease.
2. Overall wellbeing and activity level are each rated on a visual analog scale (VAS). The ratings over the last weeks or months can be visualized. This category is trying to gain more insight information on the course of the overall wellbeing in combination with perceived physical activity level.
3. Different physical exercises, explained with video's, are provided to stimulate activity. Promoting healthy moving by evidence based exercises might be helpful for patients to change their physical activity.
4. The app also provides reminders for medication intake and appointments. With this category the adherence of medication as well as appointments are addressed.
5. A game was developed to increase patients' knowledge about the process of inflammation. Education and understanding the disease is a part of self-management.
6. Tips and health information form the last category. To tailor this app, information categories (tips and health information) can be switched on or off. This tailored evidence-based information is divided into 10 categories: exercises for joints, fatigue, more hold on the disease, medication, pain, sexuality, tips using hands, holidays, working tips and pregnancy. To perform in daily life and incorporate the disease many tips are provided monthly.

Feedback of the patients was used to develop the app and therefore this is an user driven approached app. Thereafter, a prototype of the app was used for the test rounds, to get feedback on the functionality and missing topics, in two rounds, by 30 and 40 patients, respectively. The first test round resulted in modifications in functionality, minor additions and textual adjustments. After the second test round no more adjustments were made. This app does not include a feedback function, due to privacy legislation.

The RD-app was incorporated in the usual nursing care: patients received information and explanation about this app during nursing consultations, were given written information and received a newsletter from the department of rheumatology to inform them about the app. Furthermore, the app was launched on several different (social) media platforms. The RD-App (Dutch: Reuma app) is freely available from both the iOS and the Android store, thus not only for patients treated in this hospital.

Outcome measures

The primary outcome of this study was the self-management as measured by a generic validated 12-item self-rated scale, the Partners in Health scale (PIH scale), with total score ranging from 12 to 96 points. Higher scores indicate better self-management behavior [24, 25, 26]. The PIH scale is a reliable and valid instrument for measuring self-management of chronic conditions, including arthritis. Additionally, participants were asked if the app helped them to get more hold on their disease with one dichotomized question, yes or no.

The secondary outcomes were health-related quality of life (HRQOL), self-efficacy, pain, fatigue, and experiences with the app. HRQOL was scored with the validated SF-36 (range score 0-100). A higher score indicates a better HRQOL. It assesses eight health concepts: physical functioning, bodily pain, role limitations due to physical health problems, role limitations due to personal or emotional problems, emotional well-being, social functioning, energy/fatigue, and general health perceptions, which are summarized in a physical component summary and mental component summary score [27]. Self-efficacy was measured with the validated Dutch version of the arthritis self-efficacy scale, which consists of two subscales related to self-efficacy to deal with pain and to deal with other symptoms (depression, fatigue, frustration) [28]. Self-efficacy is 'the belief of patients in their own capability to perform activities to produce a desired outcome' [29]. Pain and fatigue were each measured with a visual analogue scale (VAS) ranging from 0-10, where higher scores are regarded as more fatigue or pain. The VAS scale is valid to detect changes in pain and fatigue in RD patients [30, 31].

Experiences with the app

To measure experiences of the participants who used the RD-app (app-users) an open-ended question was asked: How did the app help you to gain more hold on your disease? Participants who did not use the app (non-users) were asked to state the reason why they did not use the app.

Statistical analysis

Quantitative data

We used descriptive statistics to describe the study sample. Because not all participants used the RD-App, we divided the participants in two groups: app-users and non-users. A paired T-test was used to investigate whether the use of the RD-app had contributed to an increase in the PIH-scale score and other patient reported outcomes (PROs).

Logistic regression analyses served to investigate variables that are important for using the RD-app. First, univariate analyses were performed. All significant variables were put in the full models; thereafter backward elimination was performed. Results were

considered statistically significant when the p-values were less than 0.05. STATA version 13.0 was used to analyze data.

Qualitative data

Answers to the open questions about the experience of the app were labeled and grouped. Two researchers (JB, MW) independently analyzed answers to the open-ended question using open coding. Differences were discussed until consensus was reached.

RESULTS

At baseline, 679 of the 1511 eligible patients, participated (response rate 43%). Of those 679 participants, 397 also filled in the second questionnaire. The latter were older ($p<0.01$), more often unemployed ($p=0.04$) and had higher expectations the app would help them to get more hold on their disease ($p=0.01$), than those who responded only at baseline. All other baseline characteristics and PRO did not show differences.

Participants who completed questionnaires (n=397) at both periods were most frequently diagnosed with RA, 65% was female, the mean age was 52.0 (SD 15.6) years (table 1). Almost all participants had a smartphone (89%). At baseline, 70% thought they would use the RD-app for self-management support and 63% expected it would give them more hold on the disease (table 1).

Of the 397 participants, 19% (n=75) had missing data regarding the question "did you use the app?". Of those participants who missed this question 72% (n=54) did not possess a smartphone. Furthermore, there was a difference between the participants who did or did not answer this question, participants with the missing data worked less often ($p=<0.001$) and were less highly educated ($p=0.006$). Further analysis did not show differences on demographics and patients reported outcomes (PROs).

Table 1 Demographics baseline of participants who filled in both questionnaires, app-users and non-users

	Total n=397
Age (mean, SD)	52.0 (15.6)
Gender (Female) (n,%)	258 (65)
Education (College or higher, n,%)	284 (71)
Employed (yes) n(%)	180 (45)
Diagnosis (n,%)	
• Rheumatoid arthritis	163 (41.2)
• Spondyloarthritis	23 (5.8)
• Systemic Lupus Erythematosus	15 (3.8)
• Juvenile arthritis	9 (2.2)
• Fibromyalgia	24 (6.1)
• Osteo-arthritis	37 (9.3)
• Gout	20 (5.0)
• Arthritis psoriatic	54 (13.6)
• Other (e.g. PMR, arthritis eci, sarcoidose)	51 (12.9)
Time since diagnosis (n,%)	
• <6 months	7 (1.8)
• 6 months-1 year	20 (5.0)
• 1-2 years	46 (11.6)
• 2-5 years	69 (17.4)
• >5 years	254 (64.1)
Nationality (Netherlands) (n,%)	383 (96.4)
Smartphone present (yes, n,%)	344 (89.3)
Intend to use the app (yes, n,%)	242 (70.4)
Expect the app will increase the self-management skills (yes,n, %)	242 (62.7)

App users

After three months, almost one third (n=114) of the participants used the app. App-users had a mean age of 51.3 (SD 16.4). Eighty-eight percent of the app-users (n=98) who had responded positively to the baseline question "do you think you will use the app", actually used it after three months. App-users who used the app five times or more were younger (44.4 vs. 52.1; p=0.03) and were higher educated (p=0.001). Eighty-two percent of the app users would recommend this app to others. Almost all who used the app five times or more (96%) recommended this app to others. To investigate who did use the RD-app univariate analysis showed a role for gender, positive expectations of the RD-app, help needed to get more hold on the disease and the VAS global as associated factors with actually using the RD-app (table 2). In the multivariate analysis only expectations of the RD-app remained significantly associated with actually using the RD-app (p=<0.001)

(table 2). Thus, if participants had positive expectations of the RD-app for getting more hold on the disease, they were more likely to use the RD-app compared to those who did not believe the RD-app would help them to get more hold on their disease.

Table 2 Prediction model for using the RD-app

Used the RD-app	Uni variate			Multi variate		
	Coef.	95% CI	P value	Coef.	95% CI	P value
Age	-0.0008	-0.004-0.002	0.62			
Gender	0.11	0.008-0.22	0.03	0.09	0.01-0.20	0.1
Time since diagnosis	0.01	-0.03-0.06	0.60			
Employed	0.02	-0.08-0.12	0.42			
Education	-0.04	-0.16-0.07	0.42			
Self-efficacy	0.03	-0.29-0.09	0.31			
Expectations of RD-app	0.21	0.11-0.32	<0.001	0.20	0.10-0.31	<0.001
VAS global	-0.02	-0.05-0.0002	0.05			
SF-36 PCS	-0.003	-0.008-0.0005	0.08			
SF-36 MCS	-0.002	-0.008-0.002	0.25			
Partners in Health scale	-0.0005	-0.005-0.004	0.82			
No help needed	-0.11	-0.22- -0.007	0.03			

Abbrev: VAS visual analog scale; SF-36 short form 36; PCS Physical Component Summary, MCS Mental Component Summary

Evolution of self-management and PRO over time

One hundred eleven app-users completed the PIH-scale at baseline and after three months. After three months, the mean score had not significantly changed (p=0.8) (table 3). Forty-two percent of the app-users agreed the app had contributed to get more hold on the disease. This percentage was significantly higher for app-users who used the app five times or more compared to those who used it less frequently (59% vs. 37%; p=0.04).

The secondary outcomes self-efficacy, VAS pain and VAS fatigue, and SF-36 also did not change over time (table 3).

Table 3 Patients reported outcome at baseline and after 3 months

Outcome variables	App Users (n=114)		
	Baseline	**Post test**	**P-value**
Partners in Health scale (12-96)	79.47 (11.75)	79.20 (11.55)	0.8
VAS fatigue (0-10)	6.07 (2.39)	6.03 (2.22)	0.8
VAS pain (0-10)	4.96 (2.25)	4.97 (2.18)	0.9
SF-36 PCS (0-100)	36.44 (11.39)	36.73(11.12)	0.7
SF-36 MCS (0-100)	48.44 (10.64)	48.37 (10.72)	0.9
Self-efficacy (1-5)	2.55 (0.83)	2.48 (0.77)	0.1
More grip on Rheumatic disease due to app (yes, %)		42% (n=48)	
Recommended app to others		82% (94)	

Abbrev: PIH – partners in Health scale, VAS visual analog scale, SF-36 short form 36, PCS Physical Component Summary,MCS Mental Component Summary

Experiences with the app

The answers to the question: How has the app helped you getting more hold on the disease, could be divided into three categories: 1) receiving tips, 2) information on exercises and 3) gaining insight into the self-reported disease activity.

App-users stated that practical tips were helpful, for example to learn more about the disease; how to gain more control in daily life, and how to deal with fatigue or devices in daily life. Some app-users mentioned that they could use the tips whenever necessary. App-users mentioned that video instruction on physical exercises stimulated them to do more exercises. Lastly, app-users appreciated that the evolution of their self-reported disease activity could be visualized.

Reasons for not using the app

Non-users (n=208) mentioned different reasons for not using the RD-app. The most stated reasons were 'no interest at all' or 'low disease activity. Other reasons were; 'not any added value due to e.g. online information sources', 'not wishing to be confronted with the disease', and 'no time for downloading the app'. Although, some of them stated they want to download the app later on. Some non-users struggled with technical issues as they did not know how to download or did not have enough remaining space on their phone.

DISCUSSION

This study aimed to evaluate if the RD-App had a positive effect on patients' self-management behavior. It appeared that use of the app helped 42% to get more hold on the

disease by the provided tips, exercises and the insight in self-reported disease activity. This percentage was even higher for those used the app more frequently. However, a beneficial effect on self-management behavior three months after the launch of the RD-App could not be shown.

Self-management is a complex concept and the assessment of interventions on self-management is complex as well [32]. A review on evaluation of self-management support by using apps in chronic illnesses showed conflicting results: only three out of the nine studies demonstrated an improvement in symptom management through self-management when the intervention comprised an app only, thus without a feedback tool integrated in the app [16]. Another review showed that apps were rarely successful in improving self-efficacy or quality of life [14]. In the present study, the absence of improvement in the PIH scale might be explained by app-users being convinced they had sufficient self-management skills, since most of them had an established RD. On the other hand, it cannot be excluded that the PIH scale fails to pick up any changes important for a patient's experience to get more hold on the disease. Interestingly, almost half of the app-users, even those with an established RD, responded positively to the question: "Did the RD-app help you to get more hold on the disease". Thus, the perception of more hold on the disease increased by using this RD-app. Therefore, this RD-app must not be dismissed as it might be useful for self-management support from the perspective of the users, which is something that is not picked up in the used questionnaires. Besides, app-users appreciated the received tips, information on exercises and gaining insight into the self-reported disease activity. This is in line with a study on users' perceptions of apps, which found that personalized and tailored information was a motivator for using apps [33].

The second aim of the study was to investigate which factors were associated with the use of the RD-app. In the multivariate analysis, only having positive expectations beforehand was found to be associated with actually using the RD-app. This is in line with other studies on smartphone apps, they found that positive expectations and a positive attitude were of great importance for the utilization of apps [18, 34]. Nurses may play a role here by recommending, integrating and communicating positively about the app. However, assessing the quality of existing apps can be difficult [19]. Nurses can take the lead in integrating technological possibilities, like health-related apps, and using them to improve care targeted at supporting self-management.

In this study we found a remarkable discrepancy in the willingness to use the app and the actual use. At baseline, 70% of all participants indicated they would use the app. This compares well with a study in patients with epilepsy, in which 65% were willing to use an app for self-management support [35]. This percentage was higher in a study of willingness of using apps for patients with RA (85%), even if they had to pay for it [23]. In the present study, however, only one third of the participants did actually use

the app after three months. A similar gap was seen in a study in patients with diabetes, which identified a lack of time and insufficient technological skills as influential factors [36]. These factors were also mentioned by the non-users in our study. But, non-users also stated, in the qualitative analysis, that they did not need an app because they had already received enough information. This finding was confirmed in the univariate analysis, an association for not using the RD-app was found if participant did not need help at all. Similarly, in a study investigating the needs of patients toward self-management and eHealth for self-management, patients had already received most information from health professionals [34].

Some limitations of this study need to be addressed. First, we had missing data with regard to the question "did you use the app?". This might have affected the outcomes. However, of those who missed this question almost three quarters did not possess a smartphone. After adding these participants to the non-users group, no different outcomes were found for the prediction of the use of the RD-app. The other participants who missed this question had similar results on the of the PIH scale and expectations compared to the other participants. Second, we additionally used a dichotomous question for measuring the feeling of getting more hold on the disease. Self-management is a difficult construct and moreover it is difficult what outcomes must be used [37, 38]. Patient reported outcomes should provide key information of the perspective of the patient of getting more grip on the RD. The use of a one single question provided an overall image of the feeling of more grip on the RD, while an extended questionnaire may be useful to address more specific issues. Therefore, we feel that the used single item question is informative to measure how patients perceived hold on their disease. Finally, the effects of the app were measured three months after it was launched. This period may have been too short to detect changes in complex outcomes like self-management. App-users who used the app more often than average found they were getting more hold on the disease. Thus, more long-term evaluation, after 6 and 12 months for example, might show changes in outcomes.

Relevance to clinical practice

Although patients feel self-management has been seen as patient's own task, patients appreciate support on self-management. The RD-app can be useful for additional self-management support. The results are important as they help understand how new technology can improve patients' self-management in daily care.

Conclusion

Although the RD-app is used in only one third of the patients and despite the fact that self-management questionnaires did not show any difference over time, the RD app must not be dismissed. As it seemed to be beneficial for participants in their need to get

more hold on the disease by receiving tips, information on exercises and gaining insight in self-reported disease activity. Positive expectations are an important factor for the use of the app, which might increase by better communication about the app.

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CHAPTER 8

Successful implementation of a clinical transition pathway for adolescents with juvenile-onset rheumatic and musculoskeletal diseases

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ABSTRACT

Background

In 2008 a clinical transition pathway for young people with juvenile-onset rheumatic and musculoskeletal diseases (jRMD) aiming at improving transitional care was instituted. Historical data on dropout rate in our clinic was 35%, one year before the implementation of the transition pathway. This study aims to I) evaluate the effectiveness of the clinical transition pathway, II) evaluate the experiences and satisfaction of YP with the transitional process and evaluate their perceived self-management skills.

Methods

Young people with any jRMD transferred from the pediatric to the adult rheumatology department in our academic center were eligible to enroll in this quantitative cross-sectional observational study between 2009-2015. Notably in 2012, we created a dedicated adolescent JIA -clinic, located at the adult rheumatology department. Electronic patient records from all young people that were transferred between 2009-2015 were reviewed for drop-out of care. Young people were asked to rate a VAS for 'satisfaction with transition' and to complete the "on your own feet transfer experience scale" (OYOF-TES)-questionnaire regarding their experiences and satisfaction with transition. Self-management skills were measured with the "on your own feet self-efficacy scale" (OYOF-SES)-questionnaire.

Results

154 young people were transferred to the adult department, of which 76 were transferred to the dedicated adolescent JIA-clinic. The mean age at transfer was 17.8 years for YP transferred to the adult clinic and 15.2 years for transfer to the adolescent clinic. Drop-out of care rate one year after transfer was 5.1% in the adult clinic and 1.3% in the adolescent JIA-clinic. Response rate of the returned questionnaires was 61% for the adolescent JIA clinic and 36% for the adult clinic. There was no difference between responders and non-responders in demographics and disease type besides age (non-responders were significantly younger). Young people transferred to the adult and adolescent JIA-clinic both had high scores on the satisfaction scale (7.7 and 7.5 on the VAS-scale and 72.0 and 74.5 on the OYOF-TES). Self-efficacy scores were high for both groups, with OYOF-SES 59.7 for those transferred to the adult clinic and 58.2 for those transferred to the adolescent JIA-clinic.

Conclusion

The implementation of the clinical transition pathway has led to a substantial improvement of patient care during the transitional process leading to low drop-out of care

rate and high scores on satisfaction with transition. High scores on the self-reported self-efficacy scale suggests confidence of young people to have achieved sufficient skills to successfully manage their disease.

BACKGROUND

It is generally accepted that young people (YP) with juvenile-onset rheumatic and musculoskeletal diseases (jRMD) need to be prepared, well in advance, to achieve a successful transfer to adult rheumatology care [1-5]. Well-coordinated and effective management of the transitional process during adolescence is necessary to enable YP to acquire sufficient skills to independently manage their disease. Previously, we found that YP with jRMD who attended the pediatric rheumatology service before a dedicated transition program was established, were dissatisfied with the preparation for transfer to adult care [6]. The central points of YP in this study were that i) training self-management skills was neglected, ii) specific topics like education, vocation as well as the upcoming transfer to adult care were not discussed in clinic and iii) they didn't feel confident to attend the consultation without the support of their parents [6]. These findings are in line with results from other studies where ad hoc, unprepared transfer to adult care carried a high risk for lack of self-management skills and independence of YP, missed appointments and drop-out of care, non-compliance with medication and ultimately poor disease outcome [7-9].

Transition of care is a complex intervention, only few studies are available regarding the evaluation of the transitional process and there is a lack of clarity regarding which outcomes are relevant to determine the success of the transitional process. Drop-out of care rates are used as outcome measure for successful transition, being between 25-52% even after implementation of a transition program [10-13]. Other outcomes used are the satisfaction of the YP with the transitional process and development of self-management skills [5, 14, 15]. Several studies also used physical, psychosocial and rheumatic specific health status as outcome measure for the success of implementing a transition program [11, 16-19].

However, there is no gold standard for successful transition [1, 5, 15, 18, 20]. Furthermore, it is also not clear as to whose perception the success should be measured - success as perceived by the young person, parent and/or professional - and at which time point success of transition should be measured. These questions continue to be debated and are an area for further research.

Recently, EULAR endorsed recommendations for transition were published that include the definition of specific outcomes that can be used in the future to measure successful transition to adult care [5]. Outcomes can be categorized in the area of health service outcomes (e.g attending medical appointments/ drop-out, patients first visit no later than 3-6 months after transfer), individual outcomes (e.g self-management, adherence, quality of life, satisfaction with transition), social outcomes (having a social network) and alignment; assuring a good coordination between pediatric and adult health care professionals [1, 15].

The aims of the present study were I) to evaluate the effectiveness of a clinical transition pathway implemented in our institution since 2008 II) to evaluate the experiences and satisfaction of YP with jRMD with the transitional process III) to evaluate the perceived self-management skills of YP with jRMD.

METHODS

Participants and study design

A quantitative cross-sectional observational study in a real life setting was conducted. All YP with any jRMD who were transferred from the pediatric to the adult rheumatology clinic between 2009-2015 in our academic hospital were eligible to enroll in this study. Notably, all patients preferred to continue care in our center after transfer, none transferred to another medical center due to 1) our active policy to advocate continuing care in the same center, 2) the expertise with adolescent care in our center and 3) the limited travel distances in our country. Patients with the subtype oligo-articular JIA with medication-free disease remission for ≥ one year before transfer, were not included as it is our policy to routinely discharge these patients from clinical care. All YP and their parents (if age <18 yrs) gave written informed consent before inclusion.

Clinical transition pathway

The clinical transition pathway was described previously [6]. The most important points of this clinical transition pathway are an early start (12-14 years) with focus on development of self-management skills and independency using an individual transition plan (ITP) for each patient [6]. The ITP developed by McDonagh et al (2006, 2015) was used [21, 22]. The ITP is an important tool for developing self-management skills and self-efficacy with attention for the entire transitional process (suppl. Table 1 Ch.3).

During the study inclusion period (2009-2015), we created a dedicated adolescent JIA-clinic in 2012 at the adult rheumatology department. This was primarily developed for YP with JIA in the age of 12-23 years to optimally support the specific needs of adolescents and the transitional process (suppl figure 1: clinical transition pathway, adjusted from [6]). In addition, patients with other (rare) diseases that were only seen by the rheumatologist were also transferred to the dedicated JIA-adolescent clinic.

The adolescent JIA-clinic health care team consists of a mixture of paediatric and adult-care health professionals including (pediatric) rheumatologists, pediatric physiotherapists, specialized rheumatology nurses and nurse practitioners with certified training in pediatrics. The actual transfer to the adolescent clinic is between 12 and 14 years. The time of transfer is decided in cooperation with the patient and physician.

Clinical care for YP with systemic autoimmune diseases after 2012 continued to be delivered at the pediatric rheumatology department in the children's hospital, due to the necessity of involvement of other pediatric subspecialties (e.g.pediatric neurology, hematology, nephrology). Actual transfer to the adult clinic occurs around the age of 17-18 years, where timing of the transfer depends on the patient's achieved skills, personal wishes in discussion with health professionals (clinical transition pathway as published previously [6]).

Data collection

To evaluate effectiveness, electronic patient records (n=154) were reviewed on frequency of drop-out of care and the use of ITPs. YP were asked at study inclusion to complete a questionnaire that inventoried their experiences and satisfaction with the transitional process. They received the questionnaire once, a reminder by letter was sent after two weeks. The questionnaire included the following topics: demographic parameters, age, ethnicity, education, vocation, self-efficacy, self-management and satisfaction with transition.

Outcome measurements

Primary outcomes

The primary outcomes in this study were drop-out of care at one, two and three years after transfer and satisfaction with the transitional process. Drop-out was defined as YP who did not attend any follow up appointment at the dedicated adolescent JIA / adult clinic one year after the last appointment in the pediatric clinic.

Satisfaction with transition was measured by the "on your own feet transfer experience scale" (OYOF-TES) and a visual analogue scale assessing satisfaction with transition [10]. The OYOF-TES is a validated eighteen item patient reported outcome that measures experiences in transition, rated on five-point Likert scales (1 = strongly disagree to 5 = strongly agree) [10]. Seven items reflect the preparation for transition and eleven items reflect the alignment and collaboration between the pediatric and adult care [10]. A higher score on the OYOF-TES (score range 18-90) expresses higher experienced satisfaction of the YP with the transitional process, a specific cut-off point is not defined [10].

The VAS is a one dimensional question asking: How satisfied were you with the overall process of transfer to the adolescent JIA / adult clinic on a Likert scale ranging from 1-10 (1=completely unsatisfied to 10=completely satisfied) [10].

Secondary outcomes

Secondary outcomes were independent visits of YP with the nurse and physicians defined as a visit without their parents being present for the whole visit, the number of ITP's present at transfer, self-management skills and the self-efficacy of the YP. To measure the

efficacy of the implemented clinical transition pathway, the number of adolescents with an ITP were collected from the electronic patient records. Self-efficacy is measured with the on your own feet self-efficacy scale (OYOF-SES) [23]. This self-reported questionnaire consists of 17 items with a Likert scale from 1 to 4 (ranging from 1=no, certainly not- 4=yes, certainly). Four items reflect self-efficacy coping with the condition, six items reflect self-efficacy in disease-knowledge and six items reflect self-efficacy in skills for independent behavior. A higher score on the OYOF-SES (score range 10-64) expresses the perception of YP to have more self-reliance to manage the disease by themselves [23]. We asked YP an additional eight questions reflecting self-management skills. These questions were copied from a previous study [6]. Furthermore, we asked YP three additional questions regarding whether their disease had affected their educational achievements and whether the disease had affected their career choice.

Statistical analysis

We used descriptive statistics to describe the study sample by using STATA14. For continuous variables, the mean and standard deviation (SD) are shown, for categorical data percentages were presented. To compare the group of questionnaire responders and non-responders and the differences between YP transferred to the adolescents JIA clinic or adult clinic unpaired t-test or Wilcoxon's rank sum test, if data were not normally distributed, were used. Categorical variables were tested using Pear-son's chi-square test. P-value <0.05 was considered statistically significant. Data from YP with JIA who were transferred to the dedicated adolescent JIA clinic and those transferred to the adult clinic, were separately analyzed.

RESULTS

All electronic patient records of YP with jRMD who were transferred from the pediatric to the adult rheumatology department between 2009-2015 were reviewed (n=154). In total 78 YP were transferred to the adult clinic and 76 to the dedicated adolescent JIA-clinic. Demographics and disease activity of all YP who were transferred are shown in table 1. As expected the mean age at study inclusion of the YP transferred to the adult clinic was higher when compared with the YP transferred to the dedicated adolescent JIA-clinic. The majority of all study patients had oligo-articular or poly-articular JIA .

Table 1. Demographics of YP transferred from pediatric rheumatology to the adult rheumatology clinic and the adolescent JIA-clinic between 2009-2015.

	YP transferred to the adult clinic (n=78)	YP transferred to the adolescent JIA-clinic (n=76)
Transfer period	2009-2015	2012-2015
Gender Female (%)	71%	56%
Mean age (SD) at time of study inclusion	22.2 (2.4)	18.6 (2.3)
Mean age (SD) at transfer	17.8 (1.5)	15.2 (2.1)
Diagnosis (n / %)		
• JIA – oligoarticular	21 / 27	26 / 37
• JIA – polyarticular	15 / 19	21 /30
• JIA – PSA	3 / 4	3 /4
• JIA – ERA	8 / 10	7 /10
• SLE	15 / 19	1 /2
• Other (e.g. GPA, FMF)	16 / 21	12 /17
Disease activity before transfer (median, IQR)		
Number of tender joints	0 (0-0)	0 (0-0)
Number of swollen joints	0 (0-0)	0 (0-0)
ESR	8 (3-17)	8 (3-15)
Disease activity after transfer (median, IQR)		
Number of tender joints	0.(0-0)	0 (0-1)
Number of swollen joints	0.(0-0)	0 (0-1)
ESR	9 (4-19)	8 (3-15)

Abbreviations: JIA: Juvenile Idiopathic Arthritis, PsA: Psoriatic Arthritis, ERA: Enthesitis Related Arthritis, SLE: Systemic Lupus Erythematosus, GPA Granulomatosis with Polyangiitis, FMF: Familial Mediterranean Fever

Results electronic patient records

The overall frequency of 'drop-out of care' one year after transfer to the adult clinic was low, with only 5.1% of YP that did not attend any clinical appointment within the first year after transfer. The drop-out of care frequency remained stable in the next two years after transfer to the adult rheumatology team (Table 2). This percentage of drop-out of care was 1.3 % for YP who were transferred to the dedicated adolescent-JIA clinic one year after transfer. The drop-out of care frequency of YP transferred to the adolescent JIA-clinic decreased to zero after three years (table 2).

Table 2. Drop-out rate of YP after transfer to adult rheumatology clinic and adolescent JIA clinic

Data electronic patient records	YP transferred to the adult clinic N=78	YP transferred to the adolescent JIA clinic N=76	p-value
Drop-out rate after 1 year (% / N)	5.1% (4 of 78)	1.3% (1 of 76)	0.18
Drop-out rate after 2 years (% / N)	6.7% (5 of 74) #	2.7% (2 of 75)	0.51
Drop-out rate after 3 years (% / N)	5.7% (4 of 69) #	0% (0 of 73)	0.10
Total drop-out rate (1-3 years after transfer)	**16.6% (13 of 78)**	**3.9% (3 of 76)**	
Presence of ITP at transfer (%)	**55%**	**94%**	<0.01 *

Abbreviations: YP: Young People, JIA: Juvenile Idiopathic Arthritis *: P-value <0.05 was considered statically significant #: drop-out rate f*or year 2 and year 3: the number of remaining patients in care were taken as the total number of patients in that year of follow up.*

Almost all YP (94%) were transferred to the dedicated adolescent JIA-clinic with an ITP. This was in contrast to 55% ITPs in the group of YP who had been transferred to the adult clinic. As this is a real life study: the frequency of not only discussing the ITP but also filing the results in the ITP became more and more incorporated in clinical care during the inclusion period of the study. This is reflected by the higher number of ITPs in the adolescent JIA clinic, as this study group primarily includes patients in a later time period of inclusion (2012-2015).

Results Questionnaires

In total 46/76 YP transferred to the adolescent JIA-clinic and 28/78 YP transferred to the adult clinic returned questionnaires, leading to a response rate of resp. 61 and 36%. There were no differences between questionnaire-responders and non-responders regarding demographics or disease-type besides age (non-responders were younger 19.7 vs 21.4, p=<0.001) and drop out from care. All of the responders did not drop out from care (p=0.02). The mean age of the responders at the time of study inclusion was 18.6 years (SD 2.3) for the dedicated adolescent JIA-clinic and 22.2 (SD 2.4) for the YP transferred to the adult clinic.

The satisfaction with the transitional process for both scales (OYOF-TES and VAS transition) of YP who either transferred to the adult clinic or to the dedicated adolescent JIA-clinic was high (Table 3). The questions regarding the alignment and collaboration between pediatric and adult care [10], resulted in high scores for both groups; about 80% thought their new care provider was well informed about the condition of the YP and their treatment. Furthermore, 78.6% of the YP transferred to the adult clinic and 80% transferred to the adolescent JIA-clinic were convinced that there was a good collaboration between pediatric and adult care.

Table 3 Questionnaire results of YP transferred to the adult or adolescent JIA-clinic

Questionnaires	Responders transferred to the adult clinic N=28	Responders transferred to the adolescent JIA clinic N=46	p-value
VAS satisfaction with transition ± (mean, SD)	7.7 (0.8)	7.5 (1.9)	0.79
OYOF-TES±± (mean, SD)	72.0 (14.7)	74.5 (12.1)	0.44
Treatment recommendations in the adult care setting are similar to those I used to receive in pediatric care (agreed,%)	85.7	79.5	0.51
There was good collaboration between pediatric and adult care (agreed,%)	78.6	80	0.88
OYOF-SES ±±± (mean, SD)	59.7 (2.9)	58.2 (5.0)	0.42
Independent visits physician (yes)	69%	47%	0.06
Independent visits nurse (yes)	75%	44%	0.01*
Important topics discussed (yes)	97%	96%	0.83
I order my medication at the pharmacy by myself (yes)	83%	52%	0.008*
Thinking about taking medication by myself (yes)	90%	80%	0.27
Forgetting medication (yes)	52%	80%	0.01*
Making appointments independently (yes)	86%	42%	<0.01*
Transfer discussed on time (yes)	75%	79%	0.78
Education negatively influenced by the disease?			
• No	• 30%	• 29%	0.76
• Repeating a class	• 23%	• 19%	
• Lower level	• 12%	• 12%	
• More absenteeism	• 19%	• 29%	
• Other	• 15%	• 12%	
Taking the disease into account at choice for vocation (yes)	59%	42%	0.13
Restriction in career options by the disease (yes)	48%	25%	0.96

*: p-value * <0.05 was considered statically significant. ±: VAS satisfaction with transition: score range 1-10, higher score reflects higher satisfaction ±±: score-range of OYOF-TES 18-90, higher score reflects higher satisfaction of YP with transition ±±±: score-range of OYOF-SES 10-64, higher score reflects higher self-efficacy of YP. Abbreviations: YP: Young People, JIA: Juvenile Idiopathic Arthritis. VAS: visual analogue scale, OYOF-TES: on your own feet transfer experience scale, OYOF-SES: on your own feet self-efficacy scale

Self-management, measured with the OYOF-SES, was high in both groups (Table 3). More positive responses to the eight self-reported questions measuring self-management skills (e.g. independently visiting the nurse or physicians, ordering medication and making appointments independently) were seen in the group transferred to the adult clinic (Table 3). Furthermore, almost all YP in both transfer groups agreed that all

important topics were discussed during consultations. The transfer was discussed on time according to the YP in more than three quarters for both groups.

The answers to the three questions regarding education and vocation showed that the majority of all YP thought that their education was negatively influenced by the disease (Table 3). This was seen in YP transferred to the adult clinic as well as in YP transferred to the adolescent JIA-clinic. In addition, around half of all YP had taken the disease into account when choosing their vocation. Almost half of the YP transferred to the adult clinic and a quarter of YP transferred to the adolescent clinic stated that they were restricted in career options by their disease.

DISCUSSION

The implementation of the clinical transition pathway for YP with jRMD was successful: a low drop-out of care rate was seen and YP were satisfied with the transitional process in both groups. High levels on the self-efficacy scale were reported, suggesting confidence of all YP in this study to have achieved sufficient self-management skills and underlining the efficacy of the clinical transition pathway.

As a primary outcome of a successful transitional process, we evaluated the effectiveness of the clinical transition pathway by using the drop-out of care rate. Despite implementing transition programs, preventing drop-out of care is challenging. Previous studies showed drop-out frequencies between 25-52% after implementation of an transition program [10-13, 22]. In historical data from the electronic patient records in our hospital, we saw a drop-out rate of 35% after transfer to the adult clinical in 2007, one year before the implementation of our clinical transition pathway (unpublished data). At that time, there was no transition policy and preparation for transfer to adult care was not structurally given. Although these drop-out rates cannot be compared one-to-one, it gives an impression of the drop-out rates before implementation of the clinical transition pathway. The EULAR guidelines defined drop-out of care as a quality indicator to measure good transition [5]. We showed low frequencies of drop-out of care (5.1% and 1.3%) in the first year after transfer to the adult and adolescent JIA-clinic. This indicates that the implemented clinical transition pathway can improve attendance and adherence to follow up clinic visits, and prevent drop-out of care. This low percentage might also be influenced by our active policy to continue care in the same academic hospital, the expertise with adolescent care and the limited travel distances in our country. Notably, the low drop-out of care rate of YP transferred to the adolescent JIA-clinic seems logical, as the mean age at transfer was lower (15.2 years) and we assume that parents are still making the clinic appointments and are more involved in the care process. However, three years later (mean age 18.0) years, the drop-out of care rate was even

lower and decreased to zero. Additionally, when considering YP that transferred to the adolescent JIA-clinic at the age of 16 years and older up to 21 years, we still found a low drop-out of care rate of 6%. Literature showed that the implementation of a dedicated adolescent clinic has shown to improve clinical outcome [23]. Our data of the adolescent JIA-clinic underline this conclusion.

Another quality indicator according to the EULAR guidelines on the individual outcome of the transitional process, is satisfaction of the YP with the transitional process [5]. In our study the overall satisfaction with the transitional process was high for both groups in our study. Another study with YP with jRMD resulted in almost similar scores of the VAS satisfaction after instituting a transition program [11]. One study of van Staa (2014) who investigated satisfaction with transition programs of YP with a broad range of chronic somatic conditions showed lower percentages on both scales (respectively 61.8 and 6.6) [10].

Good alignment and collaboration between the pediatric and adult departments was seen as an important factor for higher satisfaction of YP with the transitional process [10].The presence of good coordination between pediatric and adult health care professionals is also recommended as an outcome for successful transition [5, 15]. According to the EULAR guidelines this can be realized by direct communication before and after transfer by combining consultations [5]. We embedded this in the clinical transition pathway. Moreover, we evaluated the alignment between the pediatric and adult clinic from the perspective of the YP with the OYOF-TES. Almost all YP in both groups were convinced there was a good collaboration between both departments.

Self-management is an important individual outcome for successful transition. The high score on the self-reported self-efficacy scale assumes more responsibility in the self-management skills of the YP for both groups. Another important measured question regarding self-management skills, is independent visits, meaning visits of YP with care professionals without their parents [7, 14, 15, 24]. According to a study of Hilderson, many YP are not given the opportunity for independent visits with the physician [25]. Previous studies showed percentages between 46-59% of independent visits for YP between 15-23 years [26-28]. In our study, almost half of the YP transferred to the adolescent JIA-clinic had independent visits. More independent visits were reported in the group YP transferred to the adult clinic, which seemed logical because of the older age compared to those transferred to the adolescent clinic. This was also observed in a study with YP with JIA, where frequency of independent visits increased with older age [17]. Notably, our historical data obtained before a clinical transition pathway was implemented, showed that 88% of YP between 14-20 years did not visit the physicians independently [6]. This suggests improved autonomy during clinic visits of the YP and may be contributed by implementing the use of an ITP in the clinical transition pathway. Many items in the ITP are designed to help YP develop self-management skills and goals

are set during the consultations. Indeed, the use of an ITP in the transitional process is strongly recommended by the EULAR guidelines for transitional care [5, 29, 30].

An important finding was that many YP experienced limitations in their education and vocation because of the disease. This was also found in other studies, where the time needed to finish studies tended to be longer and absenteeism was higher [31, 32]. These limitations might influence their future career as higher rates of unemployment were found in JIA patients [32-37]. These findings support the importance of vocational guidance and discussing vocation by health professionals [38] and should be a prominent subject in any transition plan.

This study has limitations. Paper-questionnaires were used with a mean response rate of 50%. Using electronic questionnaires might have led to higher response rates and therefore could have had an influence on the results. In addition, the number of YP discontent with the transitional process might be higher in the non-responders, leading to bias. This however seems not likely as the drop-out rate in the non-responders group was low. Additionally, the success of this study is attributed in part to the fact that none of the patients transferred to another institution besides the Erasmus University MC, this influences the external validity of the study. However the transition process should start at an early age and prepare young people to be ready for the actual transfer to adult rheumatology at age 17-18. The transition pathway is a suitable instrument to instruct the adult rheumatologist elsewhere regarding the follow up of care for this vulnerable patient group. The transition coordinator can play a role in this process and support the adult rheumatologist elsewhere. The mean age of YP transferred to the JIA clinic was 15 years and as such these YP are still in the midst of the transition process, this has influenced the results of the study.

Conclusion

In conclusion, the implementation of the clinical transition pathway for YP with jRMD has led to a substantial improvement of patient care during the transitional process leading to low drop-out rate, improved self-reported autonomy during the clinic visit, improved perceived self-management skills for both groups and high satisfaction of YP with the transitional process. This is important as these findings underlines the efficacy of this clinical transition pathway on relevant outcomes according to the most recent guidelines.

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Supplemental figure 1

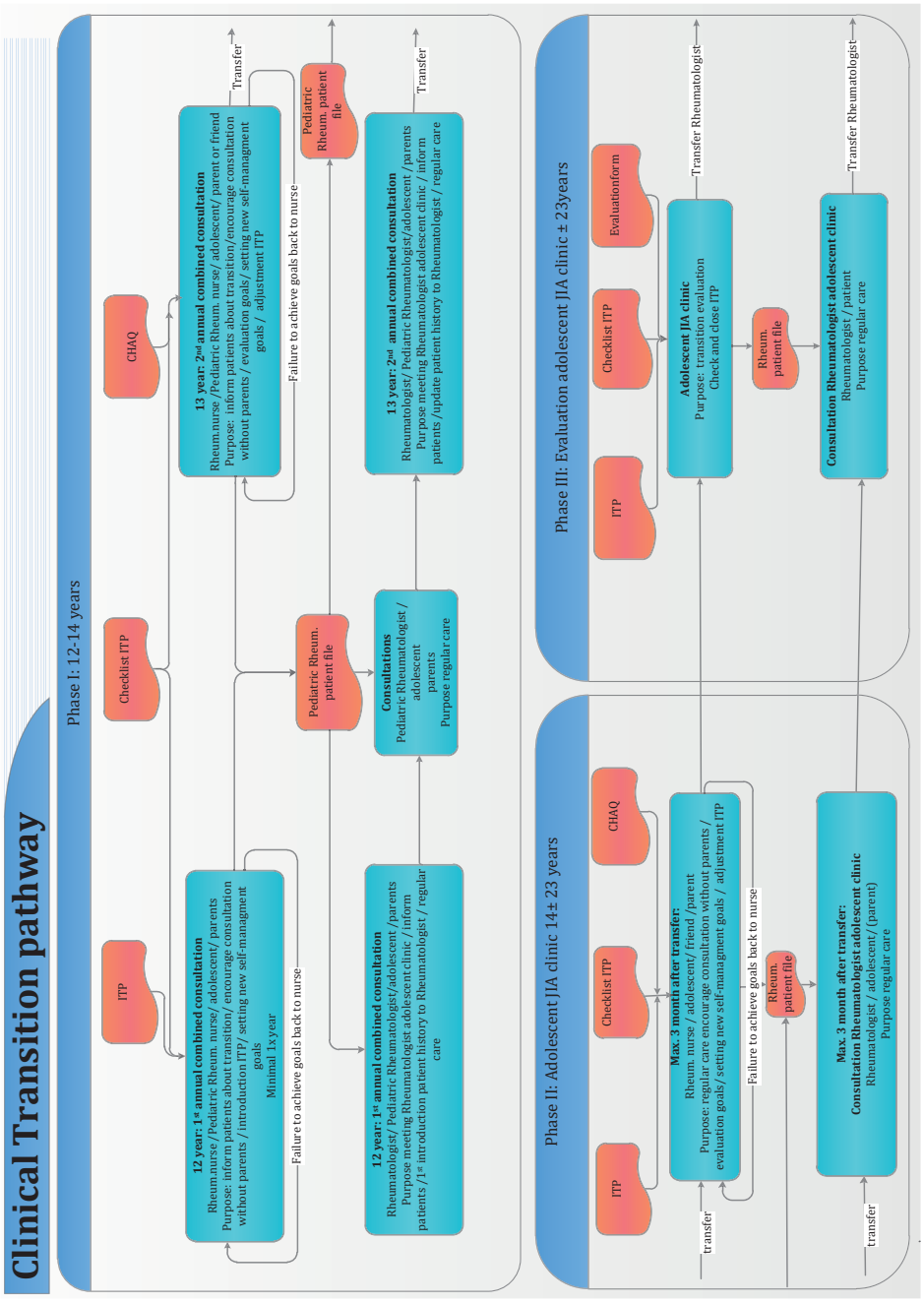

Figure 1 Clinical transition pathway adolescents

CHAPTER 9

General discussion

GENERAL DISCUSSION

The treatment of patients with an inflammatory rheumatic disease (RD) has significantly improved in recent decades by the arrival of the biologicals and novel management strategies, such as treat-to-target and tight-control management (1, 2). The next step, now widely being investigated, is personalised medicine, with regard to both dosage and choice of medication.

Treatment of RD is often medically orientated (cure), while a personalised approach taking into account psychosocial aspects (care) is often lacking. This thesis investigates specific unmet needs of young people (YP) and adults. Special emphasis was placed on the transition process and on self-management (interventions).

EXPLORING UNMET NEEDS IN HEALTHCARE

In chapter 2 we investigated the unmet needs of YP during the process of transition to adult care. It appears that in the current healthcare provision largely does not fit with the YP's experience and life stage. The YP are more interested in topics that are important to them, like sexuality, alcohol or drugs, than in aspects of the disease. These topics were not brought up, however, neither by the health professionals nor by the YP themselves. Regarding the health professionals, they may have had too little knowledge on these topics, may have been thinking that the YP did not want to discuss these, or may for personal reasons (taboo /shame/religion or have been shy to inform after these topics). For that matter, the YP may have been shy to address personal issues in the presence of parents during the consultations. In chapter 8 we showed that by the implementation of an individual transition plan, almost all YP felt that the topics they considered important were covered during consultations. This plan ensures that parents need not always be present during the consultations, which makes it easier to discuss sensitive topics. Although implementation of the individual transition plan proceeded well, practice proved sometimes unruly. For example, even though a plan contained advice on pregnancies and birth control, some YP became pregnant during the transition process under potential harmful medication treatment. Cultural, ethnical or socioeconomic background may have played a role here, as well as simply being an adolescent. The adolescent phase is often accompanied by more risky behavior (3), which may affect general health but could have worse consequences for YP with an RD. Adolescents are less focused on the future consequences of their current behavior. Though the individual transition plan turned out to be very useful, we still need a better understanding of social and cultural background or ethnical differences and their effects on "risky" behavior in patients with juvenile-onset idiopathic arthritis (JIA). This is described in the model of the transition

theory (transition conditions). Transition conditions, like cultural beliefs, can facilitate or hinder the process of transition. Thus, to understand the experience of adolescents during the transition process, it is necessary to uncover the cultural beliefs for reaching successful transition outcomes. This could be a subject for future research.

In adult care, too, personal problems and unmet needs are insufficiently addressed. In chapter 4 we show that fatigue, one of the unmet needs (4) occurs frequently and also persists over time in recently diagnosed patients. If this problem is not addressed, the patient may not be able to deal with the consequences of the RD and thus not be able to effectively self-manage the disease. This may have a negative impact on the transition process and successful transition outcomes; to reach a mastery of the change consequences. Fatigue, pain and disease activity are intermingled and perceived differently by health professionals and patients. We explored the discrepancy with regard to disease activity in chapter 4 and found that patients find it difficult to "cope" with various aspects, such as stress or finding a balance between activity and rest. Patients then may perceive a more active disease compared to the health professional's assessment of the disease activity. If on account of the discrepancy between health professionals' and patients' perspectives the unmet needs are not addressed, this can lead to an unsatisfactory outcome of care, tension and unresolved issues resulting in an disappointing relationship and poor communication between a patient and health professional. Not being able to cope with the disease might also be explained by the unpredictability of the fluctuating inflammatory activity, pain, fatigue and psychological distress. The RD disease process is constantly changing and patients therefore should have different coping strategies available. Furthermore, inadequate coping may be related to the invisibility of the disease to others. Patients must always justify why they cannot do certain activities. All in all, lack of coping skills makes it more difficult to successfully self-manage the disease.

Several steps must be taken to resolve problems. First, when discussing unmet needs in the scope of self-management, and a possible transition, the above-mentioned discrepancy should be acknowledged. Not only patients amplifiers should be taken into account, but also those of the health professional. Health professionals amplifiers may be found in patients being tedious or there is no one size fits all solution and therefore discussing the discrepancy is time consuming. Once the discrepancy has been elucidated, the medical care can be more effectively aligned with the self-management support by communicating with and coaching of the patient. This is an intricate process that starts from a personal problem and requires an individual approach with an individual care plan that provides room for discussion (5,6). This process is indeed time consuming and allows a holistic view on patients. Nurse practitioners have an important role here, as they can identify potential problems, facilitate transition and support patients to self-manage the disease.

IMPROVING CARE BY SELF-MANAGEMENT SUPPORT IN THE PROCESS OF TRANSITION

Transition is defined as "the change from one situation, form, or state to another, which might temporally disrupt normal life and ask for adjustments"(7). During transition, the need for care and support from health professionals may proportionally change. Examples of transition are being healthy to becoming a patient, being in remission to having a flare of the disease, but also the transition from care provided in the pediatric care to the adult care. Health care professionals need to be aware that a patient is in a transition process and therefore should act accordingly by adjusting their own behavior and recommendations to the patient.

In chapter 8 we evaluated the impact of the transition care pathway for YP. This is a complex intervention that consists of several sub-interventions. Implementing a transition care pathway has proven to be effective, but it is not clear what sub-intervention or combination of sub-interventions contributes most to effectiveness. It seems plausible that the individual approach contributes a great deal to a successful transition in the context of self-management. The individual approach implies setting personal goals. This is an important aspect of self-management that possibly reinforces the patient's intrinsic motivation, which is considered the most determining factor for strengthening self-management (8).

All adolescents with an RD pass the complete transition care pathway. This process is embedded in daily care and provides continued, structured and effective care. It ends naturally when the adolescent has reached the adult age. This transition care pathway is a time-consuming intervention. It might be possible to offer this in parts, with not all sub-interventions included. Then, however, problems may be missed and the patient is more likely to drop out from care – no longer feeling connected with the health professional. Our intervention stressed that offering the complete intervention is important because it reduces the drop-out rate.

During transition to adult care the adolescent is intensively supported to self-manage the disease in daily life without disrupting effect on medical drug treatment. Since the transition process is related with growing up, this encompasses a long period, where care is appropriate for each age phase. For adult patients a transition process can be less extensive. Nonetheless, it can be more difficult to identify the start of the transition process as this is unique for every patient. For the health professionals it requires a recognition and understanding of the nature of transition and the patterns of response to change, so they can adequately support patients. They must promote health and wellbeing prior to, during and at the end of the change event. Only then the health professional is able to make valuable contribution to support patients in recognising their own needs and making informed choices for effective self-management (9).

During the complete transition process the emphasis must be on actively involving both the young and adult patients in their own health care and strengthening self-management.

IMPROVING CARE BY SELF-MONITORING AS A PART OF SELF-MANAGEMENT

As patients with an RD may go through a process of transition, they have to apply different self-management strategies to cope with the RD. Self-monitoring of the disease can be of help here (10). This is seen as a self-evident aspect within the concept of self-management (11), allowing patients to get more control over the disease, search for help when necessary and to better integrate the RD into daily life.

However, self-monitoring in RD appears to be complex. In chapter 6 it was shown that it is currently not possible to self-monitor the disease activity by using patient reported outcomes. There was a discrepancy in the perceived disease activity between patients and health professionals, as additional complaints influence a patient's perception of having an active disease, as seen in chapter 4. A review of the literature showed that using patient self-report joint counts obtained by a text and/or mannequin format, also resulted in a discrepancy of the perceived disease activity between health professionals and patients, especially for swollen joints (12). Thus, self-monitoring one's disease activity is conceptually an effective way to support self-management, but is currently difficult to fit this concept into the care of patients with an RD. A better understanding is needed on how to incorporate self-monitoring in the care provision.

Self-monitoring of other aspects of the RD than the disease activity might be more effective. These could include, for example pain, fatigue, physical activity or disturbed night's sleep, which are aspects patients consider important. Patients can be taught to find the most useful support for themselves if problems arise in one or more of these areas.

These ways of self-monitoring will help patients to get more grip on and insight in the disease burden caused by the RD. If causes and consequences become more clear, it will be easier to interpret signs and symptoms of RD and help to better distinguish features belonging to RD from those originating from other aspects in their lives. This in the end might result in less discrepancy in perceiving the disease activity.

New technologies such as the "quantified self" approach can be helpful for self-monitoring and are easy applicable in daily life. The quantified self-approach implies engaging in self-tracking all kind of information about yourself e.g. biological, physical, and behavioral (13). This allows recognising patterns and becoming more aware of one's own behavior, which might have a positive effect because patients can act on it.

IMPROVING CARE BY SELF-MANAGEMENT SUPPORT WITH THE ASSISTANCE OF AN RD-APP

Other new technologies such as health related apps might contribute in assisting self-management. We developed an RD-app and evaluated this as a self-management tool in chapter 7. Although a validated self-management questionnaire showed no difference after three months of app-using, the majority of users would recommend this RD-app to peers and almost half of the patients experienced more grip on the RD by using the RD-app.

A possible explanation for not experiencing a change of the perception of more grip on their RD might be that these patients were convinced they already had sufficient grip. Most participants in this study had an established disease and probably had previously received self-management support. Still, it is also possible this intervention is inadequate and that additional support is needed. Another reason for patients not to perceive an improvement on perception of self-management may be already full adaption to the RD in daily life, without feeling the necessity for reaching an even higher level of self-management.

A low percentage actually used the RD-app. Actually using of the app apparently also depends on expectations prior to this intervention (chapter 7). Expectations influence one's motivation and, therefore, having positive expectations of the RD-app might result in stronger motivation to engage in effective self-management support. Intrinsic motivation is an important aspect of self-management (8). Thus, it might be useful to raise expectations on the app's value by communicating more positively. Furthermore the app could be expand with tools with "quantified self" functions, such as physical activity, optimizing the night's rest up to achieving goals.

METHODOLOGICAL CONSIDERATIONS

This thesis aimed to explore preferences and unmet needs and investigated how we can improve care by providing self-management support interventions. Self-management support is a complex process and consists of different components. Apart from the limitations described in each separate study, some overarching methodological considerations are addressed below.

Validity

It is not clear what outcomes of self-management support interventions are the most appropriate and how the construct of self-management should be measured which is most in line with the perception of the patients. We used different outcomes to investi-

gate the interventions described in the thesis, varying from process outcomes, such as drop-out from care to self-management outcomes (specific self-management questionnaires). It may be possible that the concept of self-management is not well-founded in the used questionnaire and does not correspond accurately with the sense of grip on the disease as perceived by the patients. Patient reported outcomes should therefore provide information about the preferences of patients. Self-management might not be a proven "fact", but a patient's own perception of grip on the disease, which was not represented in the used self-management measures. This was also seen in another study, which evaluated an intensive exercise program for rheumatoid arthritis patients. The commonly used questionnaire for functional ability (HAQ) showed no significant difference, whereas the questionnaire measuring the changes important to the individual patients (McMaster Toronto Arthritis Patient Preference Disability Questionnaire) did change (14). We recommend to at least use one single question and to rate the answer using a VAS or simply yes /no; "Do you think you can manage the disease by yourself?". A combination of outcomes is more preferable, so we can combine not only the preferences, but also the process outcomes.

Generalizability

Almost all patients participating in the studies were recruited from an university medical centre. For this reason generalizability of all results must be interpreted with some caution. Patients treated in a university centre are more likely to have more complex diseases and comorbidities, which can affect the perceived disease burden. Self-management and self-monitoring may therefore be more difficult, and patients may need more or different self-management support. This does not necessarily apply to less complex patients. Patients with less complex RD may be sufficiently helped with only little support, like an RD-app.

Selection bias

For both the transition care pathway and the RD-app the self-managements questionnaires showed positive ratings. We have no knowledge, however, about patients' self-management needs prior to the intervention. Respondents who were motivated to participate might already have embraced the concept of self-management and obtained the skills to effectively self-manage the disease. This might explain the relatively high scores on, for example, the self-management questionnaire and the SE scale (chapter 7 & 8).

Design considerations

The transition care pathway had beneficial effects on drop-out from care and self-management behaviour. This was established in an uncontrolled study however. When

comparing frequencies of drop out with historical data -obtained in the same hospital, with the same health professionals and participants with the same age - higher drop out frequencies were seen. Previous studies from others also showed higher drop-out rates (15,16). Our population is a multicultural population, with lower socio-economic status, where you would expect a high drop-out from care.

RECOMMENDATIONS FOR CLINICAL PRACTICE AND FOR IMPROVING THE ROLE OF THE NP

The nurse practitioner is a professional who offers care and cure in an integrated manner to promote the continuity and quality of both nursing care and medical treatment from the patient's perspective. He or she focuses on supporting self-management with the aim of sustaining or improving a patient's daily functioning in relation to health and disease and quality of life. The support has to be easy accessible since self-management needs can change over time, particularly when patients go through a transition phase.

In this thesis we showed that there is not one self-management solution that fits all patients. To improve care in relation to transition and self-management we proposed the following recommendations:

1. *Health professionals must be aware of possible transition in patients with an RD.*
   In addition to identifying a state of transitions, the needs for self-management support during this transition have to be addressed.
2. *For each patient it is necessary to gauge whether support on self-management is needed.*
   This need for support must be assessed regularly because patients' needs can change over time. A good follow-up is important to ensure continuity of care.
3. *There must be an awareness of the discrepancy between health professionals' and patients' perceptions of the disease activity, and which must be addressed.*
4. *To ensure adequate care, an individual care plan should be drawn up for each patient who needs self-management support.*
   This individual care plan, which is focussed on individual needs, values, preferences and life context and is aligned with the medical plan, describes the realistic achievable goals set by the patient himself or herself. Setting one's own goals helps to increase the chances of success, thereby increasing the intrinsic motivation. Patients must be guided in setting and achieving the goals.
5. *The RD-app must be extended with other tools to support self-management, such as the "quantified self".*
6. *The nurse practitioner should be more effectively involved in the daily care for patients with a chronic disease.*

RECOMMENDATIONS FOR FUTURE RESEARCH

In the concept of self-management, patients are expected to transform from passive patients to more active partners. We need to know more about the individual needs and skills for on self-management.

Self-management is a complex concept in which many different factors and many different outcomes are at play. An important factor is the patient's own perception of effective self-management.

Further research is needed to address these items and the following questions can be formulated:

- What is the ultimate outcome of effective self-management according to the patient?
- What personal and disease-specific characteristics and socio-economic status are associated with self-management needs?
- What personal and disease-specific characteristics and socio-economic status are associated with successful self-management?
- What is the most effective way to self-monitoring the disease?
- How can we improve the RD-app to make it more applicable for self-management support for patients with an RD?
- What training program is the most suitable for NPs in view of assisting patients in self-management?

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CHAPTER 10

Summary

Samenvatting

SUMMARY

This thesis aimed to gain more insight information on the ins and outs of the self-management of patients with a rheumatic disease. Chapter 1 contains a general introduction on self-management and transition in different age categories. The main questions of this thesis were: 1) What do patients with an RD prefer with regard to self-management support and what are the unmet needs that might impede self-management (support)?; and 2) What self-management support interventions can improve the care for adult and young patients with an RD?

In chapter 2 we explored how young people with JIA (juvenile idiopathic arthritis) and their parent(s) experience care and unmet needs during preparation for the upcoming transfer to adult department. We conducted a survey and developed a clinical transition pathway based on evidence from the literature and the unmet needs of the patients and their parents. Thirty-two youngsters and 33 parents filled out questionnaires. These revealed that current clinical care does not meet the needs of young people with JIAs during the transition process. Unmet needs existed because of a lack of discussion of relevant topics, neglected self-management skills training and insufficient knowledge about the differences in care between the children's and adult departments. Moreover, parents did not receive guidance for the child's transition process, which led to feelings of anxiety for the upcoming transfer to adult care. Implementing an evidence-based clinical transition pathway may improve the transition process by providing holistic care and a focus on the development of the young person's self-management skills.

It is important to incorporate the patient perspective in the self-management support. The purpose of the study presented in chapter 3 was to identify what kind of support outpatients with rheumatic disorders need and who they would like to receive support from. To this aim we used the model of 'SMS needs'. A qualitative design using focus group and face-to-face interviews with a variety of outpatients with a rheumatic disorder was applied. Twenty patients participated in two focus groups and six interviews. We could conclude that self-management was primarily seen as the patient's own task, but patients still appreciated support to help achieve this. They wanted to be seen as experienced experts of living with a rheumatic disorder. Hence they do need professionals' self-management support that coaches them in developing problem-solving skills for managing the medical, emotional and social challenges experienced in dealing with a rheumatic disorder in daily life.

Fatigue is an unmet need with a large impact on quality of life, and it is still unmanageable for many patients. Therefore we investigated fatigue in a multi-centre trial (chapter 4). Almost half of all patients had high fatigue levels at baseline, which decreased slightly over time, with 43% fatigued patients after one year follow-up. On group level, predicting fatigue after 12 months resulted in an AUC of 0.89 with only fatigue in the

model. Multivariable analyses revealed that depression and coping were associated with developing fatigue over time. So, despite a strict treat-to-target strategy, fatigue remained an overall problem during the first year of treatment. Fatigue status is mainly predicted by its previous status. But, in subgroups a small additional effect of depression was seen. Monitoring fatigue and depression may be important in managing fatigue.

Patients and health professionals do not always equally rate the patient's disease activity. This is an undesirable discrepancy, as it may affect the patient's satisfaction, adherence to treatment and outcome. We conducted focus group interviews (Chapter 5) to study this discrepancy in depth. A total of 29 patients participated. Summarizing the data of the focus groups resulted in the emergence of seven themes explaining why the patient view of the disease activity could differ from that of the treating physician. The seven themes that according to patients were relevant to high disease activity are the following: perceived stress, balancing activities and rest, medication intake, social stress, relationship with professionals, comorbidity and physical fitness. The way in which patients coped with these themes in demanding situations may be the overarching theme.

Chapter 6 describes the results of a study on self-monitoring. This activity is an important aspect of self-management as it allows predicting one's own disease activity on the basis of self-reported questionnaires Patient-reported outcome measures (PROMs) are valuable tools in the clinic to guide treatment in addition to disease activity measures performed by the physician. For this study, patients with RA have completed monthly questionnaires (RADIA, HAQ and VAS fatigue) and were checked 3 monthly for disease activity (DAS28) by the nurse specialist or the rheumatologist. It was found that both the DAS28 and the questionnaires fluctuated at individual level, at group level it remained stable. Using the score and evolvement of RADAI, HAQ and VAS fatigue over time to predict future DAS28 moderate to high disease activity resulted in a moderate performance (sensitivity 0.61, specificity 0.75).

To support patients' self-management, we developed and implemented a smartphone-application (RD-app). In chapter 7 we evaluated this RD-app with a prospective before-after study. Both the baseline and the follow-up surveys were completed by 397 participants. Eventually, 114 participants used the RD-app after 3 months. Forty-two percent of these app-users felt that use of the RD-App had helped to get more grip on the disease. Hence, the self-management scale did not changed after 3 months. The RD-app seemed to be beneficial for participants in their need to get more grip on the disease by receiving tips, information on exercises and gaining insight in self-reported disease activity. Participants with positive expectations of the RD-app for getting grip on the disease were more likely to use the RD-app.

A quantitative cross-sectional observational study was performed to evaluate the clinical transition pathway (chapter 8). One hundred and fifty-four adolescents with juvenile-onset RD who transferred to the adult department via the clinical transition

pathway between 2009-2015 were included. In 2012 we created a dedicated adolescent JIA-clinic located at the adult rheumatology department. Seventy-six adolescents transferred to this clinic and 78 to the adult clinic. The primary outcomes were drop-out from care and satisfaction with the transition. Furthermore, to the adolescents completed questionnaires concerning self-management skills. This study showed that the implementation of the clinical transition pathway has led to a substantial improvement of patient care during the transitional process – leading to a lower drop-out rate and more satisfaction with transition. High scores on the self-reported self-efficacy scale suggest that these youngsters have achieved sufficient skills and are confident to successfully manage their disease.

Chapter 9 provides a reflection of the findings of all studies in this thesis, followed by methodological limitations. This chapter ends with recommendations for clinical practice and future research.

NEDERLANDSE SAMENVATTING

Dit proefschrift levert een bijdrage aan het inzicht van zelfmanagement bij patiënten met een reumatische aandoening in transitie. In hoofdstuk 1 geven we een introductie op de begrippen zelfmanagement en transitie in verschillende leeftijdscategorieën.

De hoofdvragen van dit proefschrift zijn:

- Wat zijn de voorkeuren van de patiënten met een reumatische aandoening met betrekking tot zelfmanagement ondersteuning en wat zijn hierbij de (onvervulde) behoeften die zelfmanagement (ondersteuning) mogelijk kunnen belemmeren?
- Welke interventies ter ondersteuning van het zelfmanagement kunnen leiden tot een verbetering in de zorg voor zowel jongeren als volwassen patiënten met een reumatische aandoening?

In hoofdstuk 2 hebben we onderzocht hoe jongeren met een reumatische aandoening de zorg hebben ervaren tijdens het transitieproces van de kinder naar de volwassen polikliniek. Hiertoe heeft er een survey plaatsgevonden onder jongeren die onder behandeling waren in het Sophia kinderziekenhuis. Tevens is er onderzoek gedaan onder de ouders van de jongeren. Tweeëndertig jongeren en drieëndertig ouders hebben deelgenomen aan dit onderzoek. Dit resulteerde in de volgende bevindingen; de zorg was onvoldoende passend bij de behoefte van zowel de jongeren als de ouders, belangrijke onderwerpen volgens de jongeren werden niet besproken, er werd geen zelfmanagement ondersteuning gegeven, er was onvoldoende kennis over de verschillen tussen de kinder- en de volwassenenpolikliniek. Ouders werden niet begeleid in dit proces, hetgeen leidde tot gevoelens van angst voor de transfer. In vervolg op deze bevindingen is er een transitie zorgpad ontwikkeld, met als doel een verbetering van de zorg in het transitie proces waarbij holistische zorg en de focus op zelfmanagement ondersteuning op de voorgrond staat.

Het is belangrijk om te zoeken naar ondersteuning die patiënten nodig hebben in het managen van de ziekte. Het doel van hoofdstuk 3 was om te ontdekken welke soort zelfmanagement ondersteuning patiënten met een reumatische ziekte nodig hadden. Om dit te onderzoeken is gebruik gemaakt van het "SMS needs model". Een kwalitatief onderzoek met focusgroepen en face-to-face interviews zijn toegepast. In de focusgroepen hebben 20 patiënten deelgenomen en bij de interviews 6 patiënten. Geconcludeerd kan worden dat patiënten zelfmanagement primair als hun eigen taak zien, maar hulp daarbij wel waarderen. Zij willen graag als expert worden gezien. De hulp die de patiënten van de professional verwachten betreffende zelfmanagement ondersteuning ligt op het terrein van het coachen in het ontwikkelen van probleem oplossende technieken met betrekking tot medische, emotionele en sociale uitdagingen die zij ervaren in het dagelijks leven.

Het niet onderkennen van vermoeidheid is een onvervulde behoefte in de zorg die een grote impact heeft op het dagelijks leven van patiënten met een reumatische aandoening en die moeilijk te managen is. Daarom hebben we vermoeidheid onderzocht bij patiënten die net de diagnose reumatoïde artritis (RA) hadden gekregen (hoofdstuk 4). Uit dit onderzoek blijkt dat bijna de helft van alle onderzochte patiënten last had van vermoeidheid op baseline. Deze vermoeidheid verminderde maar weinig over de tijd, met 43% vermoeide patiënten na 1 jaar. Op groepsniveau resulteerde het voorspellen van vermoeidheid na 12 maanden in een area under the curve van 0.89, met alleen vermoeidheid in het model. Daarnaast liet de multivariate analyse zien dat zowel depressie als coping een rol spelen in de ontwikkeling van vermoeidheid bij patiënten die bij aanvang van de studie niet vermoeid waren. Concluderend bleek dat ondanks een strikte behandeling met een "treat-to-target" strategie, vermoeidheid een probleem blijft gedurende het eerste jaar na diagnose. Het monitoren van vermoeidheid en depressie lijkt belangrijk te zijn om de vermoeidheid te managen.

Patiënten en health professionals beoordelen de ziekte activiteit van de RA vaak verschillend. Dit verschil is niet wenselijk omdat dit mogelijk invloed kan hebben op de tevredenheid en therapietrouw van de patiënten. Met behulp van focusgroepen is deze discrepantie tussen health professionals en patiënten onderzocht (hoofdstuk 5). In totaal hebben 29 patiënten deelgenomen aan deze studie. Wij hebben 7 thema's kunnen ontdekken die volgens de patiënten gerelateerd zijn aan het ervaren van een hoge ziekte activiteit. De voor de patiënten relevante thema's zijn de waargenomen stress, balans tussen activiteit en rust, medicatie, sociale stress, relatie met health professionals, co morbiditeit en lichamelijke activiteit. De manier waarop patiënten in veeleisende situaties met deze thema's omgingen, kan het overkoepelende thema zijn.

In hoofdstuk 6 beschrijven wij de resultaten van een studie met betrekking tot het zelf monitoren van de ziekteactiviteit. Zelfmonitoren wordt als belangrijk element binnen zelfmanagement gezien. Zelf gerapporteerde vragenlijsten zijn een waardevolle aanvulling in de kliniek, die aanvullend kunnen zijn op de door de health professionals onderzochte ziekte activiteit. Voor deze studie hebben patiënten met RA maandelijks online vragenlijsten ingevuld (RADIA, HAQ en VAS vermoeidheid) en zijn zij drie maandelijks gecontroleerd op de ziekteactiviteit (DAS28) door de verpleegkundig specialist of de reumatoloog. Geconstateerd is dat zowel de DAS28 als de uitkomsten van de vragenlijsten op individueel niveau fluctueerden, op groep niveau bleef het stabiel. Met dit onderzoek hebben we kunnen concluderen dat de combinatie van zowel de score, als de evolutie van de uitkomsten van de HAQ, RADAI en de VAS vermoeidheid over de tijd, de ziekteactiviteit niet goed genoeg te voorspellen is (sensitiviteit 0.61, specificiteit 0.75) met deze vragenlijsten.

Om patiënten te ondersteunen in het zelfmanagement is een reuma-app ontwikkeld. In hoofdstuk 7 is deze reuma-app geëvalueerd met een vóór en na meting. Van de 397

patiënten met een reumatische aandoening die hebben deelgenomen aan dit onderzoek, hebben 114 patiënten de reuma app gebruikt. 42% van deze app gebruikers hebben aangegeven dat de reuma app heeft bijgedragen tot het gevoel van meer grip op de reuma. Echter de gevalideerde zelfmanagement vragenlijst liet dit verschil niet zien. Redenen die patiënten aangaven voor het hebben van meer grip op de reumatische aandoening door het gebruik van de reuma-app zijn het ontvangen van persoonlijke tips, informatie over oefeningen voor de gewrichten en inzicht in het verloop van de zelf gerapporteerde activiteit. Deelnemers die vooraf positieve verwachtingen hadden over de reuma-app hebben de app ook daadwerkelijk meer gebruikt.

Om het in hoofdstuk 1 ontwikkelde transitie zorgpad te evalueren is er een cross-sectionele kwantitatieve studie uitgevoerd (hoofdstuk 8). Tussen 2009-2015 hebben 154 jongeren met een juveniele reumatische aandoening het transitie zorgpad doorlopen. In 2012 is er een speciale adolescenten kliniek opgericht, gelokaliseerd op de volwassen polikliniek. Van de 154 jongeren zijn er 76 naar de speciale adolescenten kliniek gegaan en 78 naar de volwassen kliniek. De primaire uitkomstmaten waren de drop-out van zorg en de tevredenheid met de transitie. Daarnaast zijn de zelfmanagement vaardigheden bestudeerd. Deze studie laat zien dat het implementeren van het transitie zorgpad heeft geleid tot een verbetering in de zorg, met lage percentages van drop-out en hoge scores op de tevredenheid met de transitie zorg. Hoge uitkomsten op de zelf-effectiviteit score lijsten suggereren dat de jongeren voldoende zelfmanagement vaardigheden hebben ontwikkeld om de ziekte succesvol te managen.

Hoofdstuk 9 geeft een reflectie op de bevindingen van dit proefschrift, gevolgd door de methodologische beperkingen van deze thesis. Dit hoofdstuk wordt afgesloten met aanbevelingen en onderzoeksvragen voor de toekomst.

ADDENDUM

Dankwoord

PhD portfolio

About the author

List of publications

DANKWOORD

Het is zover, er staat een datum voor mijn promotie, 5 december 2018.

Promoveren is meer dan het schrijven van een proefschrift. Het is een traject van rennen, vliegen, stilstaan, duiken, vallen en weer opstaan. Het is persoonlijke groei. Het schrijven is nu voorbij en ik kan mij richten op het verdedigen en het terugkijken met het dankwoord. Want een promotietraject doorloop je nooit alleen. Ik heb veel steun gehad van veel mensen. Met dit dankwoord probeer ik iedereen te bedanken.

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Dit proefschrift is opgedragen aan To en Piet

PHD PORTFOLIO

1. PhD training	Year	ECTS
General courses		
Masterclass research for nurse practitioners	2011	1
NIHES Bio statistical Methods I: Basic Principles	2012	5.7
Basic principles didactics	2015	1
Basic qualification examination	2016	3
Basic Designing education	2017	3
Seminars and workshops		
Annual congress ACR, Atlanta	2010	1
Annual congress ACR, Washington	2012	1
Annual congress ACR, San Francisco	2015	1
Annual congress NVR	2012	0.5
Annual congress NVR	2013	0.5
Annual congress NVR	2017	0.5
Annual congress NVR	2018	0.5
Annual congress V&VN VS	2015	0.5
Annual congress V&VN VS	2016	0.5
Annual congress ACR, Washington	2016	1
Annual congress Eular, Amsterdam 2018	2018	1
10th ICN NP/APN Conference, Rotterdam	2018	0.5
National presentations		
Poster presentation NVR, Increased Fatigue does not herald a flare among patients with RA, Arnhem	2011	0.5
Poster presentation NVR, Discrepancies between disease activity and disease burden, Arnhem	2012	0.5
Presentation NVR, Reuma app, Arnhem	2018	0.5
Presentation conferentie transitie zorg, Utrecht	2018	0.5
International presentations		
Poster presentation, ACR Atlanta, One monthly patient reported disability, disease activity and fatigue shows more fluctuation then expected based on the 3-monthly clinical evaluation by DAS-28	2010	0.5
Poster op Eular, London, 2011, Increased Fatigue does not herald a flare among patients with RA	2011	0.5
Poster presentation ACR, Washington, 2012Discrepancies between disease activity and disease burden.	2012	0.5
Poster presentation, San Francisco, 2015, The prediction of fatigue in early rheumatoid arthritis patients	2015	0.5
Poster presentation ACR, Washington, 2016 Meeting the Needs of Adolescents with Autoimmune Diseases, the Development of a Clinical Transition Pathway	2016	0.5

Oral presentation ACR, Washington, 2016 Evaluation of a Clinical Transition Pathway for Adolescents with Autoimmune Diseases	2016	0.5
Other		
Research Meetings department Rheumatology	2012-2014	0.5
Erasmus MC Nurse Practitioner Lectures	2012-2018	1
2. Teaching activities		
Teacher University of applied science, MANP, Utrecht, student coach	2013-2014	4
Teacher University of applied science, MANP, Utrecht, student coach	2014-2015	4
Teacher University of applied science, MANP, Utrecht, student coach	2015-2016	4
Teacher University of applied science, MANP, Utrecht, student coach	2016-2017	4
Supervising Master's theses, MANP, Utrecht	2014-2015	3
Supervising Master's theses, MANP, Utrecht	2015-2016	3
Nurse teacher of MANP student, Rotterdam	2017-2018	0.5
Grants		
Implementation grant, Nuts Ohra, 2009, € 100.000	2009	
Research grant EBCN (evidence based care for nurses, 2012, € 50.000	2012	
Research grant EBCN (evidence based care for nurses, 2015, € 50.000	2015	
Professional membership Dutch Society of nurse specialist (V&VN-VS) Dutch Society rheumatology (V&VN Reumatologie)		

ABOUT THE AUTHOR

Margot Walter was born on November 29th 1965 in Zwijndrecht, The Netherlands. In 1984 she graduated from her high school and moved to Breda. She was qualified as a Registered Nurse in 1988, followed by obtaining her certification for pediatric nursing in 1989. She worked as a pediatric registered nurse in several outpatient clinics knowable Diaconessen hospital and Sophia Children's hospital. Thereafter, she worked as a head nurse in night shifts in a nursing home combined with a job at the office of her parents where she worked as an accounting professional. In 2003 she started to work at the department of rheumatology as a research nurse. Next, in 2006 she started her Master in Advanced Nursing Practice (MANP) and completed this in 2008. During the last year of her master she developed a transition program for young patients with any rheumatic disease based on the unmet needs of the adolescents and their parents. Ever since, she works as a nurse practitioner in the field rheumatology for adult patients at the Erasmus University Medical Center in Rotterdam, with a special interest for adolescents with a rheumatic disease where patients care is combined with research. Besides her job as a Nurse practitioner, she additionally became a teacher in 2013 at the University of applied science at the department of the Master of advanced nursing practice.

Margot is married to Arjen van Kaam and together they have four children; Lisa, Wouter, Josefien and Femke.

LIST OF PUBLICATIONS

Successful implementation of a clinical transition pathway for adolescents with juvenile-onset rheumatic and musculoskeletal diseases.
Walter M, Kamphuis S, van Pelt P, de Vroed A,Hazes JMW.
Pediatr Rheumatol Online J. 2018 Aug 3;16(1):50. doi: 10.1186/s12969-018-0268-3.

Fatigue in early, intensively treated and tight-controlled rheumatoid arthritis patients is frequent and persistent: a prospective study.
Walter MJM, Kuijper TM, Hazes JMW, Weel AE, Luime JJ.
Rheumatol Int. 2018 Jul 16. doi: 10.1007/s00296-018-4102-5. [Epub ahead of print]

Development of a clinical transition pathway for adolescents in the Netherlands.
Walter M, Hazes JM, Dolhain RJ, van Pelt P, van Dijk A, Kamphuis S.
Nurs Child Young People. 2017 Nov 7;29(9):37-43. doi: 10.7748/ncyp.2017.e932.

What support is needed to self-manage a rheumatic disorder: a qualitative study.
Been-Dahmen JM, Walter MJ, Dwarswaard J, Hazes JM, van Staa A, Ista E.
BMC Musculoskelet Disord. 2017 Feb 16;18(1):84. doi: 10.1186/s12891-017-1440-5.

Focus group interviews reveal reasons for differences in the perception of disease activity in rheumatoid arthritis.
Walter MJ, Van't Spijker A, Pasma A, Hazes JM, Luime JJ.
Qual Life Res. 2017 Feb;26(2):291-298. doi: 10.1007/s11136-016-1369-4. Epub 2016 Jul 21.

Is tightly controlled disease activity possible with online patient-reported outcomes?
Walter MJ, Mohd Din SH, Hazes JM, Lesaffre E, Barendregt PJ, Luime JJ.
J Rheumatol. 2014 Apr;41(4):640-7. doi: 10.3899/jrheum.130174. Epub 2014 Feb 15.

Facilitators and barriers to adherence in the initiation phase of Disease-modifying Antirheumatic Drug (DMARD) use in patients with arthritis who recently started their first DMARD treatment.
Pasma A, van 't Spijker A, Luime JJ, Walter MJ, Busschbach JJ, Hazes JM.
J Rheumatol. 2015 Mar;42(3):379-85. doi: 10.3899/jrheum.140693. Epub 2014 Dec 15.

De verpleegkundig specialist in de reumatologie: nurse practitioner nieuwe stijl
J.A. Schoemaker-Delsing, M.Walter. 2010|3 Nederlands Tijdschrift voor Reumatologie