SHARED DECISION-MAKING IN RHEUMATOLOGY:

WHAT MATTERS TO PATIENTS?

Ingrid Nota

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CONTENTS

Chapter 1

General introduction

In recent decades, the relationship between clinicians and their patients has increasingly been emphasized and viewed as essential within the health care system. Consequently, patient-centred care and patient involvement are considered vital elements of high quality health care and modern medicine (1-5) and are increasingly recognized by governments across the world at the health policy level (6). An essential activity of patient-centred care is shared decision-making (SDM) (5). In SDM, the clinician and patient collaborate to understand the patient's situation and to determine how best to address it.

Research in SDM has mainly focused on the general population, acute care settings or one-time decisions, such as screening or surgery (7). The increasing recognition of SDM and a lack of knowledge on this topic in chronic care, triggered the initiation of this project to research and enhance patient participation in medical decision-making in the field of rheumatology, as described in this thesis. This chapter introduces the thesis that resulted from this SDM project, and touches upon the history and theoretical framework of patient involvement in medical decision-making. This chapter then discusses the role of patient decision aids and how best to develop these interventions, and finally presents an outline of this dissertation.

SHIFT FROM PHYSICIAN-CENTRED CARE TO PATIENT-CENTRED CARE

Although from the time of Hippocrates, guidelines have existed that stipulated how doctors should communicate with their patients (8), patient involvement in medical decision-making was considered undesirable in ancient Greece. At that time, clinicians were divine and at the centre of the system; they had exclusive access to knowledge and should, therefore, direct patients' care. One of the clinician's primary tasks was to establish hope and trust in the treatment. They believed that any disclosure of diagnosis, prognosis or possible adverse effects of treatment might harm this trust. It was, therefore, advised that clinicians conceal most information from their patients, and manipulation and deception were acceptable techniques for doing so (9).

Hope and trust in the treatment are still key-elements of health care, however, nowadays we believe that educated and empowered patients will better manage their disease and adhere to treatment. This shift towards patient-centred care started with patients acquiring the basic right for self-determination and consent before initiation of an intervention. This right became popular in the Western world at

the beginning of the 20th century as a consequence of the prevailing broader-based civil rights movements for equality before the law (9-12). After World War II, this right was expanded to include the right to be informed about possible risks and side effects of the proposed medical intervention, i.e. the right to *informed* consent. Patients in the United States of America were the first to acquire this right in 1957, with many countries following suit in the years after (9-13). In the Netherlands, the right to informed consent was established in 1994 (*Wet op de geneeskundige behandelingsovereenkomst*, WGBO) (14). This process of patient empowerment has since been catalysed by the rapid growth of the Internet, which offers convenient access to a wide range of health information. Given these more recent developments, patient involvement in medical decision-making was inevitable.

THEORETICAL FRAMEWORK: MODELS OF MEDICAL DECISION-MAKING

The phrase "sharing of decision-making" was used for the first time in 1972 by Veatch (15) in his paper "Models for Ethical Medicine in a Revolutionary Age: What physician-patient roles foster the most ethical relationship?". Yet, the concept of SDM did not appear in the research literature till 1997 with the landmark paper of Charles, Gafni and Whelan (16): "Shared Decision-making in the medical encounter: what does it mean? (Or: it takes at least two to tango)". In this paper and its follow-up (17), Charles and colleagues argue that models of medical decision-making can be regarded as a continuum with two extremes –'paternalistic decision-making' and 'informed decision-making'. In between is 'shared decision-making'. Table 1 presents the key characteristics of each model.

Table 1.1. *Models of treatment decision-making*[a]

Analytical stages		Models				
		Paternalistic	*(in between approaches)*	**Shared**	*(in between approaches)*	**Informed**
Information exchange	Flow	One way (largely)		Two way		One way (largely)
	Direction	Physician→patient		Physician⇄patient		Physician→patient
	Type	Medical		Medical and personal		Medical
	Amount[b]	Minimum, legally required		All relevant for decision-making		All relevant for decision-making
Deliberation		Physician alone or with other physicians		Physician and patient (plus potential others)		Patient (plus potential others)
Deciding on treatment to implement		Physicians		Physician and patient		Patient

Derived from the 1999 paper "Decision-making in the physician-patient encounter: Revisiting the shared treatment decision-making model" by Charles, Gafni, & Whelan (17).

[a.] Simplified illustration for an encounter focusing on the case of a (treating) physician-patient dyad.

[b.] Minimum required

On the one side of the continuum is the synthetic paternalistic model, which considers the clinician to be the rational guardian of the patient who is in need of caring custody (16, 18). The clinician reports only selected information – the minimum required by law – to the patient and chooses the intervention he/she considers best for this patient. (Of course, the clinician acts in the best interest of the patient and bases his/her decisions on the best available clinical evidence.) The information exchange is largely one-way – from the clinician to the patient. As a result, the patient has a passive dependent role upon the clinician, who is the expert, and the patient's preferences are not elicited (16).

On the other side of the continuum is the informed decision-making model. This model considers the patient to be fully autonomous. As with the paternalistic model, the information exchange is largely one-way. However in this informed model, the clinician informs the patient about *all* treatment options and their risks and benefits. The patient then weighs the options and decides which option reflects both his/her preferences and the best evidence available (16). However, this model is not suitable for most patients. Firstly, deciding autonomously about treatments is often a complex process that includes many factors. This process becomes especially difficult when stress and emotions cloud the patient's judgement. Secondly, research has shown that patients generally have a high need for information, but many do not feel the need to autonomously make the final decision (19-22).

In between both models is the shared decision-making (SDM) model, which considers the decision-making process to be a collaboration between clinician and patient. In a two-way information exchange, medical knowledge and personal values of the patient are shared and balanced. Together the clinician and patient discuss the options, elicit preferences and come to an agreement regarding the decision (or agree to disagree) (16, 17). SDM is recommended particularly for preference-sensitive medical decisions (23). In preference-sensitive decisions, two or more equivalent treatment options are available or consequences of the treatment have major implications for the patient's daily life. Consequently, the best decision largely depends on the patient's informed preferences regarding existing treatment options and the patient's personal value of risks, barriers and benefits.

SHARED DECISION-MAKING

Benefits of shared decision-making

SDM is considered beneficial for various reasons. Firstly, as previously discussed, patient participation in medical decision-making is an essential part of obtaining informed consent, which is ethically correct as well as mandatory by law in a growing

number of countries (13). Secondly, patients increasingly report a desire to engage in medical decision-making (24). Finally, according to a recent systematic review, when patients have participated in SDM, they are more likely to enjoy better affective-cognitive outcomes, such as less decisional conflict and improved knowledge, satisfaction with care and trust in the treatment and clinician (25). Furthermore, some studies have shown that interventions to promote SDM can lead to improvements in behavioural and health outcomes, such as self-management, adherence, coping behaviour, symptom reduction and quality of life (26-30).

Barriers to shared decision-making

While the benefits of patient involvement in care and medical decision-making have been frequently reported, SDM is not widely adopted in mainstream clinical practice (31, 32). Several barriers to SDM exist, both clinician-related and patient-related. Barriers mentioned by clinicians are a lack of time, feeling unprepared to involve patients in medical decisions and low confidence in their ability to communicate risks effectively. In addition, not all clinicians are convinced of the benefits of SDM (33-35). Patient-reported barriers for SDM are their lack of awareness about having a choice, low confidence in their capacity to participate, a self-perceived lack of knowledge, low health literacy, low numeracy skills and uncertainty about what questions to ask (36-41).

Furthermore, although overtime patients have increasingly desired to participate in their medical decision-making (24), not all patients wish to be similarly involved (42). According to a systematic review including 115 studies, in 63% of the studies the majority of patients preferred sharing decisions with clinicians (24). This leaves a sizable minority of patients who do not wish to be involved in their medical decision-making. Whether patients' reluctance to engage in the decision-making process is truly a lack of desire to be involved as opposed to a lack of self-efficacy (41), it remains challenging for clinicians to tailor the level of patient involvement to the desire of each individual patient.

Shared decision-making in practice

SDM is a complex concept. To provide clinicians guidance about how to accomplish SDM in routine practice, Elwyn and colleagues developed a three-step deliberation process model for clinical practice (43-45), which is illustrated in Figure 1.1. In this deliberation process, patients become aware of a choice, learn about their options and consider 'what matters most to them', to finally agree upon an informed shared decision. This process may require more than one clinical contact – not necessarily face-to-face – and may include the use of decision support tools and discussion with others (e.g. family members, friends, next of kin).

The first step focuses on patient's awareness that: a choice exists, the choice includes preference sensitive aspects, and his/her participation is desirable *(team talk)*. This step can be initiated by either the clinician or the patient. Then, the clinician informs the patient about all treatment options and their potential harms and benefits, taking into account the best clinical evidence available. Together they compare alternatives *(option talk)*. For the patient, awareness of options leads to the development of initial preferences. The patient is supported to further explore these initial preferences and his/her concerns and priorities regarding these options to arrive at informed preferences. Patient decision aids (PtDAs) can help patients during this process. Finally, in the third step, in a thoughtful dialogue, these preferences are elicited and integrated into subsequent actions (*decision talk*).

This process model describes several key elements to help the clinician implement SDM in daily clinical practice. However, as mentioned in a recent viewpoint paper: "these elements are not items on a checklist or instructions on a recipe, but rather iterative and interactive steps in a conversational dance" (46). Consequently, continual dialogue must be taken into account when implementing and measuring SDM.

Team Talk Explain the intention to collaborate and support deliberation
Option Talk Compare alternatives
Decision Talk Elicit preferences and integrate into subsequent actions

Figure 1.1. *A shared decision-making model (45)*

INTERVENTIONS TO PROMOTE SHARED DECISION-MAKING

To support SDM in practice and overcome the previously mentioned barriers, many interventions have been developed. These include training programmes for clinicians, large marketing campaigns for the general public and numerous patient decision aids (PtDAs) that prepare patients in making medical decisions in collaboration with their clinician. Worldwide, more than fifty SDM-training programmes for clinicians

have been developed, varying in learning objectives, duration, and teaching materials (47). Most training programmes aim to educate clinicians, and include topics such as what SDM entails in daily clinical care as well as workshops to practice SDM communication skills. However, the effectiveness of such programmes has yet to be properly assessed (47, 48).

To raise awareness and educate the general public about SDM, targeted marketing campaigns have been implemented, such as the MAGIC (Making Good Decisions In Collaboration) program. The MAGIC program encourages patients to 'ask 3 questions': 'What are my options?', 'What are the possible benefits and risks?', and 'How can we make a decision together that is right for me?' Both professionals and patients have responded positively to this campaign, but they have also indicated that additional actions are needed to activate patients' participation (49).

Finally, various sorts of patient decision aids (PtDAs) have been and are being developed. PtDAs are commonly defined as tools that prepare patients to make informed value-based decisions together with their clinician. PtDAs specifically state the decision being considered and stress the relevance of SDM. PtDAs come in many forms. They can be brief enough to be used during the clinical encounter or they can include detailed information to be used before or after consulting the clinician. They can also be of any type, but are most commonly pamphlets, videos or web-based tools (30). While synthetic patient educational materials often only include one treatment option and commonly are provided after the decision has been made, PtDAs provide information about all treatment options and help patients to elicit and express their preferences, concerns and priorities regarding these options (50-52). PtDAs are not a substitute for clinician consultation. Instead, they guide the SDM process, enable patients to become active, informed participants and strengthen the clinician-patient partnership.

Currently, two decades of research has established the positive effect of using PtDAs. A recent systematic review included 115 randomized controlled trials of PtDAs for many different decisions and conditions (30). The findings showed that when patients use PtDAs, they improve their knowledge of their options and feel more informed and clear about what matters most to them. Furthermore, patients using PtDAs more often have accurate expectations of possible benefits and harms of their options and more readily participate in decision-making. Patients who used PtDAs that included an exercise to help them clarify what matters most to them were more likely to reach decisions that were consistent with their values. PtDAs do not worsen health outcomes and people using them are not less satisfied. However, it is still unclear the effect PtDAs have on adherence (30).

Despite the proliferation in the development and evaluation of PtDAs showing the effectiveness of these interventions (30), very few to none are implemented in

daily clinical care and their influence, when used in routine workflows, has yet to be determined (31). Despite such a positive evaluation in the research setting, it remains unclear why PtDAs are not adopted in daily clinical practice. Therefore, it is essential to assess the barriers for implementation during development. Patients, health professionals and policy decision-makers can provide valuable insights regarding this matter, and their involvement during development is, therefore, key (31, 53).

DEVELOPMENT OF PATIENT DECISION AIDS: THE IPDAS DEVELOPMENT PROCESS MODEL

For over a decade, policy makers have driven the development and implementation of PtDAs in daily clinical care. However, as previously mentioned, widespread adoption has not yet occurred. One way to boost PtDA adoption is to assure clinicians and patients that these tools have a sufficient level of quality. With this aim, in 2006, the International Patient Decision Aid Standards (IPDAS) Collaboration (established in 2003; (50), developed a quality criteria framework (51). This framework contains 64 criteria regarding the content, development, implementation and evaluation of PtDAs. The quality framework addresses the following twelve domains: (1) instigating a systematic and transparent development process; (2) providing information about options; (3) presenting probabilities; (4) clarifying and expressing values; (5) using patient stories; (6) guiding or coaching in deliberation and communication; (7) disclosing conflicts of interest; (8) delivering patient decision aids on the Internet; (9) balancing the presentation of options; (10) using plain language; (11) basing information on up-to-date scientific evidence; and (12) establishing effectiveness.

The first domain addresses the relevance of a systematic and transparent process for developing PtDAs and the importance of involving key stakeholders, especially patients and health professionals (50, 51). To be able to develop a PtDA that supports patients to think about personal values and prepare them to participate in decision-making, it is essential to understand patients' considerations, how they would like to be supported, and the best way to enable their participation in the decision-making process. Clinicians, on the other hand, need to be involved in the development process to provide guidance on how to integrate the PtDA in daily clinical practice. Clinicians also need to approve the content of the PtDA. The scientific literature, however, rarely discusses in detail how PtDAs are developed; and more specifically, the actual operationalization of user involvement is lacking. Many studies do not describe how and when users were involved during PtDA development (53). This first IPDAS criterion often seems to be more of a symbolic

statement or used to describe the participation of users (specifically patients) during evaluation of a PtDA.

In response to this omission, the IPDAS Collaboration has recently updated their evidence base with a PtDA development process model that places more emphasis on user involvement (52, 53). This process model provides a stepwise approach to the careful and systematic development and implementation of PtDAs that involves patients and health professionals at several key phases (see Figure 1.2). The six steps of the IPDAS process model are: (1) determining the scope of the PtDA, (2) eliciting user needs to determine the design of the PtDA, (3) developing the prototype, (4) usability testing and redesigning, (5) feasibility testing and redesigning, and (6) implementing and further evaluating the final version of the PtDA. This process is overseen by a multidisciplinary steering group.

Although this new comprehensive model provides an overview of the entire development process, it does not provide guidance on how to best involve patients and health professionals or specify which research methods to use. The authors, therefore, urged PtDA developers to complement the IPDAS development process model with other guidelines, such as a user-centred design approach (53). In a user-centred design, specific research methods are used to consult with potential users relatively early within the developmental timeframe (54, 55). This approach allows developers to adopt and implement user-centred input, resulting in the product more adequately fulfilling users' needs and, consequently, positively effecting user satisfaction (54, 55). Despite their potential to highly contribute to the successful implementation of a PtDA in clinical practice, neither the IPDAS development process model nor user-centred design methods have been applied in PtDA development (53).

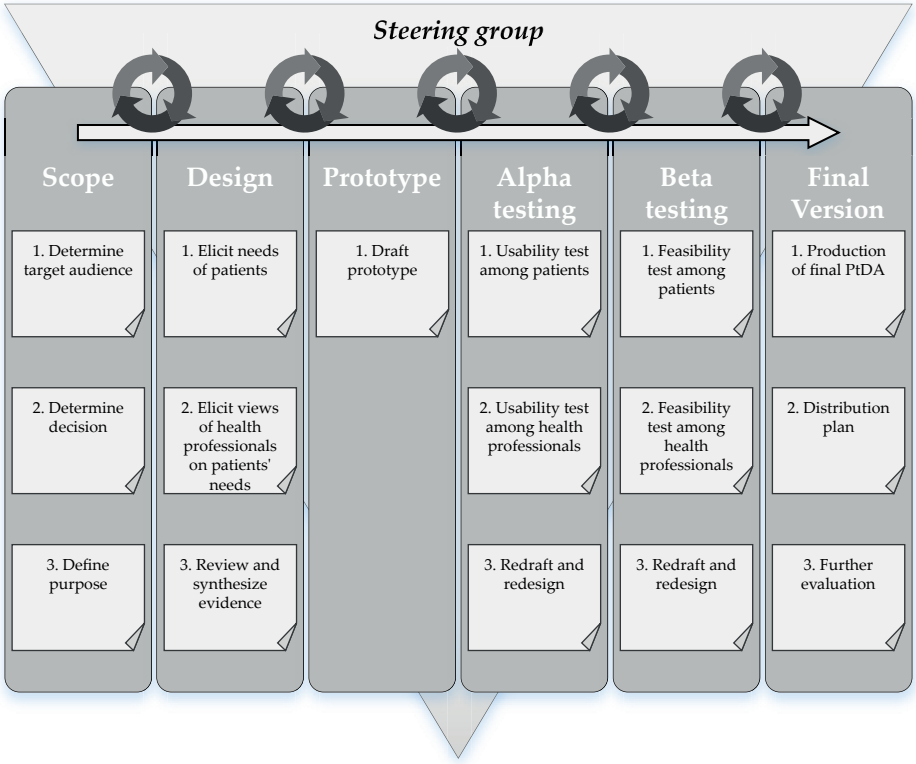


Figure 1.2. *IPDAS development process model (53).*

SHARED DECISION-MAKING IN RHEUMATOLOGY

SDM seems particularly suitable in the context of chronic diseases where decisions are often long term and are more likely to require a more active patient role in carrying out the decision (i.e. self-management and adherence). Chronic care decisions are also often taken over a longer time period, which naturally offers more of an opportunity for shared decision-making between the clinician and patient (56, 57). This thesis specifically focusses on SDM in rheumatology.

The broader category of rheumatic diseases is an umbrella term for more than a 100 various types of musculoskeletal disorders. One subcategory is chronic inflammatory arthritis, which includes rheumatoid arthritis, psoriatic arthritis and ankylosing spondylitis (amongst others). These forms of inflammatory arthritis are chronic, progressive and autoimmune, and characterized by inflammation of the joints and

often other tissues. Symptoms include pain, tender and swollen joints, stiffness, functional limitations and fatigue, and may also be in combination with other symptoms, such as skin rashes. In the long term, joints and other tissues may become permanently damaged. Though inflammatory arthritis is incurable, treatments exist to manage the symptoms and to decrease disease activity in order to minimize or even prevent permanent damage. Early diagnosis and aggressive treatment is critical for the management of inflammatory arthritis. Additionally, successful management of chronic diseases relies heavily on patients' self-management.

In the last decade, decision-making in rheumatology has become increasingly complex. Remission of disease activity is the goal and may be achieved by using disease-modifying anti-rheumatic drugs (DMARDs) to control the disease and to relieve or reverse symptoms (58-60). Many new DMARDs have become available, and the range of possible treatments has increased tremendously. The new DMARDs have proven their efficacy, but also often have serious implications for patients' lives due to the manner of administration, the need for continued monitoring, and/or the risk of serious side effects (58-60). Because of the large influence the symptoms and side effects can have on patients' wellbeing and daily lives, it is vital that patients' preferences are included in treatment decision-making. Moreover, because patients' and rheumatologists' beliefs about illness and treatment may differ (85, 86) and success of treatment largely depends on a patient's willingness to adhere to the medication, the choice of treatment needs to be based on a shared decision between the patient and rheumatologist. While developing treatment recommendations for inflammatory arthritis, the European League against Rheumatism (EULAR) international task force even emphasised that "decision-sharing by patient and rheumatologist is of such overwhelming importance that it should spearhead the recommendations" (58, 61).

Despite the fact that SDM seems to be of the utmost importance in rheumatology, relatively little studies have addressed this topic. It is known that patients with rheumatic diseases have a high need for information (26, 62-64), but only a few studies have examined these patients' desire to become involved in decision-making regarding their treatment (22, 63-65). These studies have showed that a large number of patients want to be involved in medical decision-making, yet the percentages varied from 42% to 83% across studies (22, 63-65). The observed variation may be due to differences in patient populations, the way of questioning and the type of medical decision to be made. Moreover, as most of these studies have used quantitative designs, the focus was predominantly on the amount of preferred influence, rather than on the patients' motives for the preferred type of decision-making. Finally, little

is known about the use and impact of PtDAs in the care for patients with rheumatic diseases. Only two studies on PtDAs about DMARDs have recently been reported with preliminary, though promising, results (30, 66).

OUTLINE OF THIS DISSERTATION

The project presented in this dissertation was developed with the primarily aim to gain knowledge of patients' perspective of SDM and the potential role of a web-based PtDA in the setting of rheumatology care. The secondary aim was to examine the feasibility and value of developing a PtDA in co-creation with health professionals and patients. The third aim was to evaluate the impact of the developed PtDA on patients' involvement in medical decision-making. The IPDAS development process model (53) guided the development and evaluation of this PtDA. As part of the stepwise user-centred development of the PtDA, we conducted a series of studies. The following paragraphs outline the aims of all conducted studies and their applied methods.

This dissertation begins by presenting two studies which were conducted to obtain more knowledge about patients' perceptions of SDM in rheumatology. **Chapter 2** describes a qualitative study that used in-depth semi-structured interviews with 29 patients diagnosed with rheumatoid arthritis. This study was conducted to gain knowledge about patients' motives for (not) wanting to be involved in medical decision-making and the factors that hinder or promote patient involvement. To examine patients' preferences and experiences in a more quantitative way, we conducted a cross-sectional survey, as described in **Chapter 3**. This study focused on medical decision-making in rheumatology in general, but also distinguished between various common decisions related to the use of DMARDs. Furthermore, this study examined the concordance between preferred and perceived roles, how patients' involvement is related to satisfaction about decision-making and which factors are associated with preferred roles, perceived roles and concordance.

Chapter 4 describes the exploration of patients' considerations, worries and questions when deciding about DMARDs. We also determined the information patients desired to have in order to participate in the decision-making process. In depth face-to-face interviews were conducted with 32 patients who had recently consulted their rheumatologists and discussed initiating DMARDs. The findings of this study helped determine what information the PtDA needed to provide.

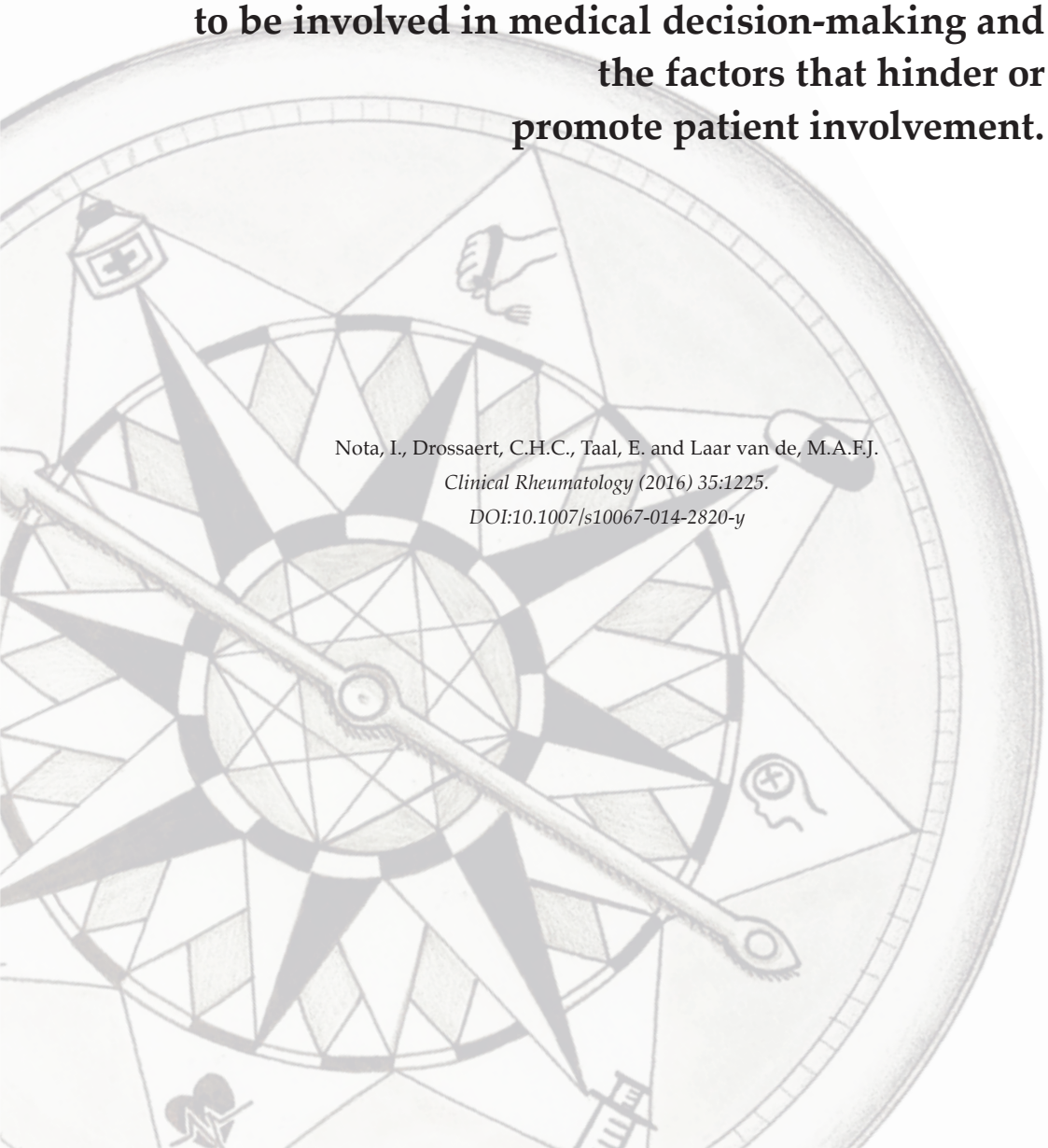
Chapter 5 provides an overview of the developmental process of the PtDA and includes results of needs assessments and usability studies among both patients and health professionals. In this chapter, we evaluate the feasibility and value of the IPDAS development process model in combination with user-centred design methods. To provide the reader of this dissertation an idea of the appearance of the web-based PtDA, all the components of the PtDA are presented as an **intermezzo**.

Chapter 6 describes the study that was conducted to evaluate use, appreciation and effect of the PtDA. The primary outcome measure of the study was the impact of the PtDA on patients' perceived role in medical decision-making, in comparison to usual care. Secondary outcome measures comprised satisfaction with the decision-making process and the decision, beliefs about medication, adherence to the medication and trust in the physician.

To conclude, **Chapter 7** presents the summary and general discussion of the studies described in this dissertation. We reflect on the results of the studies and our chosen research methods. Implications for clinical practice and future research are also discussed in this chapter.

Chapter 2

Arthritis patients' motives for (not) wanting to be involved in medical decision-making and the factors that hinder or promote patient involvement.



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ABSTRACT

To gain insight into arthritis patients' motives for (not) wanting to be involved in medical decision-making (MDM) and the factors that hinder or promote patient involvement. In-depth semi-structured interviews were conducted with 29 patients suffering from Rheumatoid Arthritis (RA). Many patients perceived the questions about involvement in MDM as difficult, mostly because they were unaware of having a choice. Shared decision-making (SDM) was generally preferred, but the preferred level of involvement varied between and within individuals. Preference regarding involvement may vary according to the type of treatment and the severity of the complaints. A considerable group of respondents would have liked more participation than they had experienced in the past. Perceived barriers could be divided into doctor-related (e.g. a paternalistic attitude), patient-related (e.g. lack of knowledge) and context-related (e.g. too little time to decide) factors. This study demonstrates the complexity of predicting patients' preferences regarding involvement in MDM: most RA patients prefer SDM, but their preference may vary according to the situation they are in and the extent to which they experience barriers in getting more involved. Unawareness of having a choice is still a major barrier for patient participation. The attending physician seems to have an important role as facilitator in enhancing patient participation by raising awareness and offering options, but implementing SDM is a shared responsibility; all parties need to be involved and educated.

INTRODUCTION

In recent years, patients have been increasingly encouraged to take up an active role in managing their health and in medical decision-making (MDM). Patient empowerment, participation, involvement and shared decision-making (SDM) are frequently used concepts in this context (63, 67-71). Patient involvement in MDM is considered to be a patient's right (13), but it has also been positively associated with satisfaction with care, self-management, coping behaviour and adherence (26-29).

While the benefits of participating in care and MDM have been widely reported, some studies have shown that not all patients want to be actively involved (19, 21, 22, 63, 64, 72-77). Other studies have identified barriers for involvement in MDM. Unawareness of having a choice, low confidence in the capacity to participate, a perceived lack of knowledge and uncertainty about which questions to ask are among the barriers mentioned by patients (36-40).

Patients' preferences regarding involvement in MDM have been explored in some depth for irreversible decisions like screening or surgery (19, 24, 74-76, 78, 79), but are less well known for decisions concerning chronic health problems, such as arthritis, where the doctor-patient relationship is potentially a long-term one (56). In managing arthritis, the decision-making process has become increasingly complex due to the rapid development of new disease modifying anti-rheumatic drugs (DMARDs). Including patients' preferences in these decisions is important as these medications often have serious implications for patients' lives due to the way of administration, the need for continued monitoring, and/or the risk of serious side effects. Moreover, because the success of treatment largely depends on a patient's willingness to adhere to the medication, the treatment needs to closely fit in with the patient's values and lifestyle.

To date, only a few studies have examined patients' preferences regarding involvement in treatment decision-making in rheumatology. These studies showed that a large number of patients want to be involved in shared decision-making, yet the percentages varied from 42% to 83% across studies (22, 63-65). Differences in patient populations, in the way of questioning and in the type of medical decision to be taken may be responsible for the observed variation. Moreover, as most of these studies have used quantitative designs, the focus was predominantly on the amount of preferred influence, rather than on the patients' motives for the preferred type of decision-making. More knowledge about patients' motives for (not) wanting to participate in MDM and the factors that hinder or promote participation can make it easier for health-care professionals to pursue the preferred level of patient involvement. The aim of this study was to gain insight into rheumatic patients'

notions about involvement in MDM, by using a qualitative study design using in-depth interviews.

METHODS

Recruitment of respondents

Patients were recruited from two hospitals in the Netherlands: Medisch Spectrum Twente (MST) and Ziekenhuisgroep Twente (ZGT). Patients diagnosed with rheumatoid arthritis (RA) scheduled to have an appointment with the rheumatologist were preselected by one of the researchers. Rheumatologists were instructed to invite all these preselected patients to participate in the study if they had the ability to complete an interview in Dutch without assistance. After having been informed about the aim and procedure of the study, patients were asked to sign an informed consent form. Thirty patients initially consented to participate in the study, and one respondent cancelled the appointment due to being too ill.

Procedures

Semi-structured interviews were conducted by the first author (IN). The interviews, which lasted approximately sixty minutes each, were audiotaped and took place at patients' homes or at the university. First, respondents were asked to describe the decision-making process of a recent medical decision related to the treatment of their RA that was highly important to them. Next, respondents were asked if they had participated in the decision-making process – and if so, how – or, if this was not the case, if and how they would have preferred to have participated in the decision-making process. Subsequently, the Control Preferences Scale (CPS) (80) was used to grade the level of preferred and perceived participation. The CPS consists of five cards portraying five different roles patients can assume in treatment decision-making, each role being described by a statement and a cartoon. Respondents were asked to pick one that portrayed their preferred role and motivate their choice. If they preferred to actively participate in MDM, they were asked whether they had been able to take on their preferred role during the recent decision-making process – and if this was not the case, which role they perceived to have had. In addition, respondents were asked which factors facilitated or hindered their participation. Patients were also invited to elaborate on their preferences and barriers and facilitators for participation in other treatment decisions related to their RA. The last five interviews identified no significant new themes, indicating that data saturation had occurred.

Data analysis

Data were analysed using inductive analyses. This means that the patterns, themes and categories of analysis arose from the data (81). After verbatim transcription of the audiotaped interviews, two analysts (IN and CHD) independently read ten of the transcripts several times to familiarize themselves with the data. They identified emerging themes and selected relevant quotations (sentences or small paragraphs). Then, the two analysts compared and discussed their findings to develop a thematic framework. Relevant quotations were selected from the rest of the transcripts by the first author. These quotations were then grouped by three researchers (IN, CHD and ET) independently using the thematic framework. After every ten interviews, the researchers met to discuss their findings until consensus was reached. Themes were refined and subthemes were determined until a final thematic framework was developed. All quotations provided in this article were reviewed by a translator.

FINDINGS

Respondent characteristics

A total of 19 women and 10 men all suffering from rheumatoid arthritis were recruited from nine different rheumatologists. The average age of the respondents was 56 years (range = 17-74 years) and most respondents had a low or medium level of education (n=15 and 9, respectively). The majority of respondents was not employed (n=22). The average disease duration was 8 years (range = 0-38 years). The current medication of patients was either synthetic DMARDs (n=20) or biologic DMARDs combined with methotrexate (n=9).

A difficult concept

Overall, respondents appeared to have some difficulty in determining their experiences with and preference regarding involvement in MDM. They frequently hesitated in providing an answer or changed their answer during the interview. Many statements were qualified by "I think…" or "Maybe…" and several respondents mentioned literally: "Those are difficult questions." Other patients mentioned that they had never thought about it: "I never thought about that, but after having this conversation with you I am going to ask more questions." [Male, 66 years] or that they felt they did not have a choice: "My involvement? Did I have a choice?" [Male, 44 years] Some patients had difficulties conceptualizing patient involvement in MDM and gave somewhat ambiguous answers. For example, one respondent stated: "We do that together. He prescribes the medicine and I take it. […] That's the way it is. I don't know how else to explain it." [Female, 69 years] Someone else

was convinced that the doctor made the decision: "The rheumatologist made that decision." But shortly after, she showed to have (obliviously) influenced the decision and decision-making process: "And he was very much aware of the fact that I did not want prednisolone." [Female, 41 years]

Patients' preferences regarding involvement in MDM

Patients' preference to let the doctor decide

Despite considering it a difficult question, most patients were able to indicate their preference for participation regarding involvement in MDM. A small but considerable group of respondents (n=8) preferred the doctor to decide about which treatment to initiate. Trust in their doctor and valuing the expertise of the doctor were the main reasons for preferring not to be actively involved in MDM, as illustrated by this quotation: "I think highly of the medical profession. I trust them." [Male, 64 years]. Patients who valued the expertise of the doctor mentioned that being well informed, being listened to and having their problems taken seriously were important prerequisites for satisfaction with this form of decision-making: "She decides, but I insist that she takes it… takes me seriously." [Female, 61 years]

Patients' preference to decide mostly by themselves

Only a few respondents (n=3) wanted to decide mostly by themselves. One patient stated that she herself feels her symptoms best: "Well, for example, if I get side effects, then I believe I should be the one to decide whether or not to continue taking the medication, because I feel my body best." [Female, 62 years] Another patient wanted to be involved because she wanted to evaluate the consequences the decision would have for his personal situation: "I have a family and I do not want to be hospitalised for a few months. I weigh up the pros and cons, I decide that." [Female, 41 years] Finally, one patient simply wanted to be in control: "I am in control over my own body. If there is a decision at stake, I decide by myself. I do not need anybody else to help me." [Female, 74 years]

Patients' preferences for shared decision-making

Most respondents (n=17) preferred shared decision-making (SDM), because it reflects a good relationship with the doctor, as illustrated by this quotation: "I want to share in the decision-making process. That he listens carefully to what you have to say and that you listen to his arguments as well. And that you can say anything, even small things, without feeling a bore. That's when you have a good relationship." [Female, 60 years] Other reasons for this preference were mostly a combination of the aforementioned reasons for preferring the doctor to decide and the reasons to

decide by themselves. For example, many respondents valued the expertise of the doctor highly, but wanted to be a part of the decision-making process because they themselves feel their symptoms best, wanted to have some level of control or wanted to critically evaluate the impact the doctor's advice would have on their personal situation and discuss this. The following quotation illustrates this last issue: "I want to share in the decision-making process. As a patient, you should follow the doctor's advice, you should not say it is nonsense, you cannot do that, but I do critically evaluate his advice. […] And if I do not agree or have questions, well, then I discuss this with him." [Male, 56 years]

Some patients were attracted to the notion of shared responsibility for the treatment: "It is about you, you are responsible for your own body, but because you do not have the knowledge, you also depend on the doctor, so he needs to be responsible as well. So you share the decision-making." [Male, 50 years] However, others did not agree with the shared responsibility. Although they did prefer SDM, they wanted the doctor to be responsible for the outcome of the treatment. "He is the expert and, in the end, it's his responsibility. He is the one who is truly responsible, but we decide together." [Female, 54 years]

Patients' preferences regarding involvement vary according to the circumstances

Some respondents noted that their preference regarding involvement in medical decision-making depends on the occasion. They mentioned that their preference may depend on the type of treatment and the severity of their complaints (Box 2.1). With regards to the type of treatment, decisions that came up were about surgery, medication (starting, stopping, changing the dosage or way of administration), physiotherapy, psychological support and diet. There was no clear pattern to be able to predict *how* certain decisions or circumstances would influence patients' preferences regarding involvement. For example, when comparing decisions regarding surgery with decisions regarding medication, some respondents preferred more involvement in deciding about surgery, whereas other respondents preferred more involvement regarding decisions about medicines. To provide another example, some respondents stated they preferred more involvement in deciding about changing the dosage - as opposed to deciding what medicines to take -, whereas others preferred more involvement regarding the decision what medicines to take - as opposed to deciding about changing the dosage. Regarding the severity of the complaints, there were respondents who preferred more personal involvement if the severity of their complaints increased, whereas others preferred to leave treatment decisions more to their doctor in such cases.

Box 2.1. *Common rationales regarding circumstances that may change respondents' role preferences*

Type of treatment

"With medication, you often know what will happen. Surgery is often much more radical to me: Then you need stop your medication, you need to be hospitalised, you just feel much worse. [...] If the time comes that a surgery is necessary, then the doctor can make that decision. Not me." [Female, 41 years]

"Well, with medication, […] you always have something to say about it, because you do not have to take them anymore if you do not want to. But If she tells me about a surgery, [...] I would say I would first like to wait a little longer and think about it. But that, to me, is of a different order than medication." [Female, 61 years]

"Starting [medication]. Because the medication can be quite intense, it is very important to me to think about it: Do I want this? And if you are already using medication, and your dosage needs to be increased, then… I cannot decide myself if the dosage needs to be changed or not. That is a doctor's task." [Female, 62 years]

"The way of administration is more personal than increasing or decreasing the dosage. Starting to inject yourself is more personal than starting to take tablets." [Female, 41 years]

"When starting medication I prefer to share in the decision-making process. Increasing the dosage is something I want to decide myself, as I'm the one who can best determine how severe my pain is. And the doctor decides if the dosage needs to be decreased, because he/she understands what my blood level results mean. If I would need psychological support, I would make that decision myself. And with regards to a decision about diet, I prefer to go to a naturopath, because that is better suited to my eating habits and way of living." [Female, 56 years]

"I don't have knowledge of medication, but I do have an opinion about physiotherapy." [Female, 60 years]

Severity of complaints

"It also depends on how you feel. Actually. If you feel fine, you think: Say whatever you want, but I do not need it, and if you do not feel so good, then I gratefully take the advice." [Female, 41 years]

"When you get so many physical complaints, you start to think: action needs to be taken. But I do believe that you need to talk with your doctor about the right solution for you personally and what should be done. [...] And information should also be provided about the medication, the pros and cons." [Female, 62 years]

"Last year I was in so much pain. My knees were killing me. I called the doctor and like a drug addict I begged for an injection. Normally I wait until the next check-up and the blood level results, but now I took control." [Female, 54 years]

Perceived involvement in MDM

When asking respondents about how they perceived their involvement in MDM so far, most respondents stated they had experienced either shared decision-making (n=13) or the doctor making the decision(s) (n=15). One respondent perceived to have decided by herself. Overall, it seemed patients wanted more participation than they perceived.

Barriers to get involved in MDM

Some patients who preferred to have a more active role in MDM perceived barriers in getting involved in the decision-making process. An overview of all identified barriers is provided in table 2.1. The perceived barriers can be doctor or patient related, but can also be contextual. Examples of doctor-related barriers perceived by patients are (a) the doctor not appearing to take the patient's problems seriously and the patient not knowing how to respond and thus freezing; (b) the patient not being able to participate in the medical decision-making process, because of the doctor

not acknowledging their role in the decision-making, to be seen from the fact that he/she does not offer alternatives or immediately rejects the patient's questions or suggestions; (c) patients not being adequately informed (respondents stated to have received either too little, too much or too complex information).

On the patient's side, some patients lacked awareness about treatment alternatives or the possibility to choose. Other respondents mentioned that they experienced a lack of knowledge or that they did not want to delay the treatment and therefore chose to let the doctor decide. Others stated that their lack of assertiveness hindered their participation. A few respondents mentioned that they were not yet ready to accept the diagnosis and therefore found it hard to participate in MDM. Finally, some patients mentioned that they purposively held back certain information from their doctor (about visiting another health-care professional, taking complementary or alternative medicine or about not supporting treatment). According to patients themselves, this does not necessarily have to be a barrier for patient participation, but it may influence the collaboration and interaction between doctor and patient.

We also identified contextual barriers, such as 'too little time to decide' or the study protocols, which leave little room for an informed choice. In those circumstances, respondents often felt they had no choice.

Facilitators for participation in MDM

Respondents were asked which factors facilitated or would have facilitated their participation in MDM. An overview of the facilitators identified is provided in table 2.2. The results show that many facilitators for patient participation are the opposite of the reported barriers. For example, patients feel they can more easily participate in the decision-making process when they are explicitly invited to do so, when they are taken seriously and being listened to, when the doctor is open to answering questions, and when he/she explains well and offers alternative options. A good doctor-patient relationship with mutual respect, an open style of communication and trust is often seen as a great facilitator for patient participation. Certain characteristics of the patients themselves can also make it easier to participate: if the patient is assertive and not reserved about asking questions, the patient can more easily participate in the decision-making process. Other facilitators are contextual, such as time to think things over, the availability of information to read at home and the opportunity to ask someone from the hospital or clinic questions. These contextual facilitators are important, because many patients have questions that arise after the consultation (at home), when they process all the information.

Table 2.1. *Perceived barriers to patient participation*

Themes	Quotations
Doctor-related barriers	
Doctor does not listen/take patient seriously	*"I told him: 'I am very tired, though.' And he said: 'Yes, half the Dutch population is tired.' […] Then I briefly froze. With such an answer, you feel like you're a bit of a complainer." [Female, 45 years]*
Doctor does not recognise role of patient	
offers no alternatives	*"If you do not agree, you say so. But if he then explains it and there are no alternatives, well, then there is nothing to choose." [Male, 70 years]*
immediately rejects the patient's questions or suggestions	*"I want to quit that poison, but he says: 'you can't.'" [Male, 66 years]*
Doctor does not provide adequate information	
gives too little information	*"He is very good at his job, but I have to ask him everything, for example when I don't agree with something he says. I really need to drag it out of him." [Female, 74 years]*
uses difficult language	*"Then they start using those difficult words at the hospital and I think: never mind, I don't want to hear it anymore." [Male, 50 years]*
gives too much information	*"When I was younger they gave me medication and occasionally I heard what it was, but I don't think they properly explained what the side effects were or could be. Now they do, but now it is tough, because I sometimes get medication which makes me think: do I dare to start using this?" [Female, 62 years]*
Patient-related barriers	
Patient is not aware of alternatives or possibility to choose	*"But it [an alternative] has to be out there somewhere and the doctor needs to tell me." Did you ask for an alternative? "No, I did not. And I did not know I could, either. If the doctor says it's effective, then I think: You know best." [Female, 69 years]*
Patient lacks knowledge	*"Of course many decisions are made for you, because you yourself cannot… I did not know anything about this when it all started." [Female, 17 years]*
Patient does not want to delay treatment	*"Medication is prescribed. You want to become well again, so you cannot say: "I do not want it." [Male, 50 years]*
Patient lacks assertiveness	
does not dare to speak up to the doctor	*"I feel I do what the doctor says very quickly. No nagging, just do as he/she tells you. That is nonsense, I know that. You should keep on asking for clarification until you are satisfied." [Female, 45 years]*

Table 2.1. Perceived barriers to patient participation (continued)

Themes	Quotations
is reserved in asking questions or does not know which questions to ask	*"I believe we are partly to blame for that, too, because we do not keep on asking. And then, at home, you have all those questions. You think about them, but actually you should just immediately ask the doctor any questions you might have." [Male, 66 years]*
Patient does not yet accept diagnosis	*"I want to be involved, but […] it takes so much effort to deal with [the diagnosis], so I really wanted to hide it all the time." [Female, 57 years]*
Patient holds back information	*"Sometimes I take less, but he does not know. And I do not feel any different." [Male, 66 years]* *"I did not tell the rheumatologist, because… they do not acknowledge alternative medicine." [Male, 50 years]*
Contextual barriers	
Too little time to decide	*"It [starting to use methotrexate] is quite a radical decision. So, er, yes, that [the time given to decide] was a bit short." [Female, 41 years]*
Study protocol leaves no room for alternative options	*"You really do not have a choice in that respect. There is a [study] protocol, and it is not like I can use other medication." [Female, 17 years]*

Table 2.2. *Facilitators for patient participation*

Themes	Quotations
Doctor-related facilitators	
Doctor invites patient explicitly to participate in MDM	*"We decide everything together. She just asks what I think about it. Then I say that it is extremely hard for me to make that decision. But then she explains […] what is the best thing to do, what she thinks the options are and how and why she came to these options. And if I want time to think things over, that is possible." [Female, 62 years]*
Doctor takes patient seriously, listens to him/her and is open to answering questions	*"The choice is also up to you. There is room to ask questions, address doubts. That is very pleasant. That is different to how it was before, to how it used to be." [Female, 39 years]*
Doctor offers alternative options	*"He explains all the options and then you can decide what is best for you. He suggests searching the internet, talking about it at home, and then you yourself should just tell him what you want. But he recommends one he… thinks is best, but you do not have to accept that." [Female, 41 years]*
Doctor explains well	*"Well, then they thoroughly explain how it [the medication] works and what the consequences are." [Male, 56 years]*
Good doctor-patient relationship	
Mutual respect	*"I always feel that there is mutual respect, understanding, when I talk with him. He listens to me. I do not feel like I am talking to a doctor. We are equals. That is why the decision was easier to make." [Female, 54 years]*
Patient trusts doctor	*"I trust her. That is why we can do it together. And I have always had that feeling, to do it together, also with the nurse." [Female, 60 years]*
Open style of communication	*"We are always very open in our communication towards each other. And he knows what we have to offer and we know what he has to offer. [...] It works very well." [Male, 70 years]*
Patient-related facilitators	
Patient is assertive and is not reserved about asking questions	*"I think that you yourself have to take a little initiative with these things. Do I want to know all the side effects, or do I know enough?" [Female, 54 years]*
Contextual facilitators	
There is time to think things over	*"But, yes, at least there was time in between. I liked that. I did not need to make a decision right away [anti-TNF protocol]." [Female, 56 years]*
Hospital or clinic is available for questions	*"I also talked a lot with the rheumatologist's nurse. I called her as well. She was always available for questions and things like that." [Female, 62 years]*
Patient gets information to read at home (leaflet, website, etc.)	*"I was already being prepared. She gave me an information leaflet to read at home. So then […] I had time to think about it and a few days later, during the instruction, I recognised them [the injections] from a picture. She did not have to explain that much and I was less afraid." [Female, 61 years]*

DISCUSSION AND CONCLUSION

By understanding patients' motives for (not) wanting to participate in MDM and the factors that hinder or promote their participation, we can make it easier for health-care professionals to pursue the preferred level of individual patient involvement. Our findings are consistent with previous studies which also showed that many RA patients prefer SDM (22, 63-65) and that preference regarding involvement varies within and between individuals (19). Our qualitative study revealed some interesting findings and demonstrates the complexity of factors influencing (the preference regarding) patient involvement.

Many patients participating in our study had obvious difficulties in determining their preference regarding involvement in MDM, because they had never actively considered it, had problems conceptualizing patient participation, or felt they had no choice. Unawareness of having a choice is a known barrier for patient participation (36-39) and previous studies have shown that patients are more motivated for SDM after being informed about the possibilities and benefits of it (40, 82-85). Therefore, we recommend initiatives to inform and educate patients about SDM to be more specifically aimed at increasing patients' awareness of having a choice.

These difficulties expressed by respondents when indicating their preference for participation regarding involvement in MDM, are also reflected in the ambiguous answers some respondents gave. Several studies have reported that patients have difficulties with conceptualizing their role in MDM (80, 86-90) and some report that this may have to do with different interpretations of the CPS labels (86-88, 91). These studies suggest that the CPS may conflate several concepts like the complexity of preferred patient involvement, information seeking preferences, and doctor's ability to engage in shared decision making.

Some patients who preferred SDM were especially attracted to the notion of shared responsibility, whereas others did not agree with that. Many other studies have shown that patients want information about their medical condition and the different treatment options without necessarily having to make the final treatment decisions (16, 17, 19, 63, 92-96). Our results go one step further: although some patients preferred to share in the decision-making process or even preferred autonomous decision-making, they wanted the doctor to be responsible for the outcome of the treatment. This shows that patients may feel responsible for the decision about the treatment, because they value to be given treatment options to evaluate the impact they may have on their life, but because they do not have the medical knowledge and have to rely on the expertise of the doctor, they see the doctor as the person to be responsible for the outcome of the treatment. When involving patients in MDM,

doctors will need to make explicit to patients that participating in MDM is not a derogation of responsibility.

As with other conditions, some of our respondents preferred to leave the decision-making up to their doctor. Reasons for this preference included trusting the doctor and valuing the doctor's expertise. This finding is consistent with those of Kraetschmer et al. (97) and Fraenkel et al. (38), who found an inverse relationship between the preference to take on an active role in decision-making and trust. They suggest that patients may fail to recognise the potential value of their own input in situations where they have complete trust in their physician. Alternatively, they suggest that patients who trust their physician may believe him/her to understand their values and to know what is best for them. We believe that patients' preferences regarding involvement in MDM need to be respected. However, if patients prefer to leave the decision-making to their doctor, the patients' input should still be acknowledged, the doctor closely fitting in the chosen treatment with the patients' values and lifestyle. This patient-centred way of communication may again increase trust and adherence to the decisions made (98, 99).

Our study revealed that patients' preferences regarding involvement in MDM may change over time, depending on the severity of the complaints and the type of decision. Previous studies have shown that preference for involvement may decrease as the severity of the complaints increases (19, 36, 37, 93, 100). Our data, however, suggest that for some patients, increased severity of health problems actually increases the preference for involvement. With regards to the type of decision, prior studies have found that participants prefer more active roles in the decision-making process where minor illnesses, behavioural decisions, major surgeries or decisions that require medical knowledge are concerned (70). In our study, however, we found no clear pattern of how certain types of decision affected patients' preferences in this respect. This means that these factors are hard to use when predicting patients' preferences regarding involvement in the decision-making process. It is necessary to further examine these complex relationships between severity of health problems and the type of treatment on the one hand and preference regarding involvement on the other. We recommend health-care professionals to assess a patient's individual preference with every decision at stake. Person perception training (101) may enhance the professional's accuracy in perceiving and understanding a patient's preference in particular situations.

Although it is essential to know if patients want to participate, it is as much important to know if they can. A considerable group of patients in our study would have

liked more participation than they had experienced in past MDM. As with studies conducted for other conditions (36-39), we identified barriers in patient involvement related to the doctor, the patient and the circumstances. Doctor-related barriers mostly concern communication skills and a paternalistic attitude. Known patient-related barriers in patient involvement are a lack of knowledge, lack of awareness of having a choice and a lack of assertiveness. Our study also revealed a barrier on the patient's side that, to our knowledge, has not been reported in previous studies about barriers in patient participation. Patients sometimes hold back information, which may, according to the patients themselves, negatively influence the doctor-patient relationship and the decision-making process. Other patients emphasised that an open style of communication and mutual respect are important facilitators for a good doctor-patient relationship. As a two-way information exchange is a prerequisite for SDM - according to the definition (16, 17)-, holding back information may inhibit SDM. Another interesting barrier on the patients' side is patients not wanting to delay the treatment and thus letting the doctor decide. Salt and Peden (102) reported that desperation or hope for the relief of symptoms were the foundation for deciding to take medications for RA, but it has not previously been reported as a barrier for patient participation. In sum, many barriers are related to communication on both the doctor's and the patient's side.

These communication-related barriers may be overcome with education and support of both doctors and patients. According to a recent report, more than fifty SDM-training programmes for health-care professionals have been developed worldwide, varying in learning objectives, duration, and teaching materials (47). For practical reasons, most programmes are accessible to doctors only, and the effectiveness of such programmes has not yet been properly assessed (47, 48).

With regards to the education of patients, patient decision aids (PtDA's) that offer balanced and reliable information about all treatment options and that help patients examine their personal values, worries, doubts and questions regarding these treatment options may be helpful (103). Integrating PtDA's in the health-care system may raise awareness for patient participation in MDM and could help educate patients about asking the right questions and doctors about offering more than one option and recognising the patient's role in MDM. However, according to a recent systematic review providing knowledge to patients and encouraging them to think about personal values is not enough. The authors concluded that, to participate in SDM, patients need knowledge (about treatment options available and of personal preferences and goals) *and* power (i.e. the believed ability to use this knowledge to influence decision-making in the encounter with the doctor) (39). Education should therefore also be focused on changing attitudes of both doctors and patients to

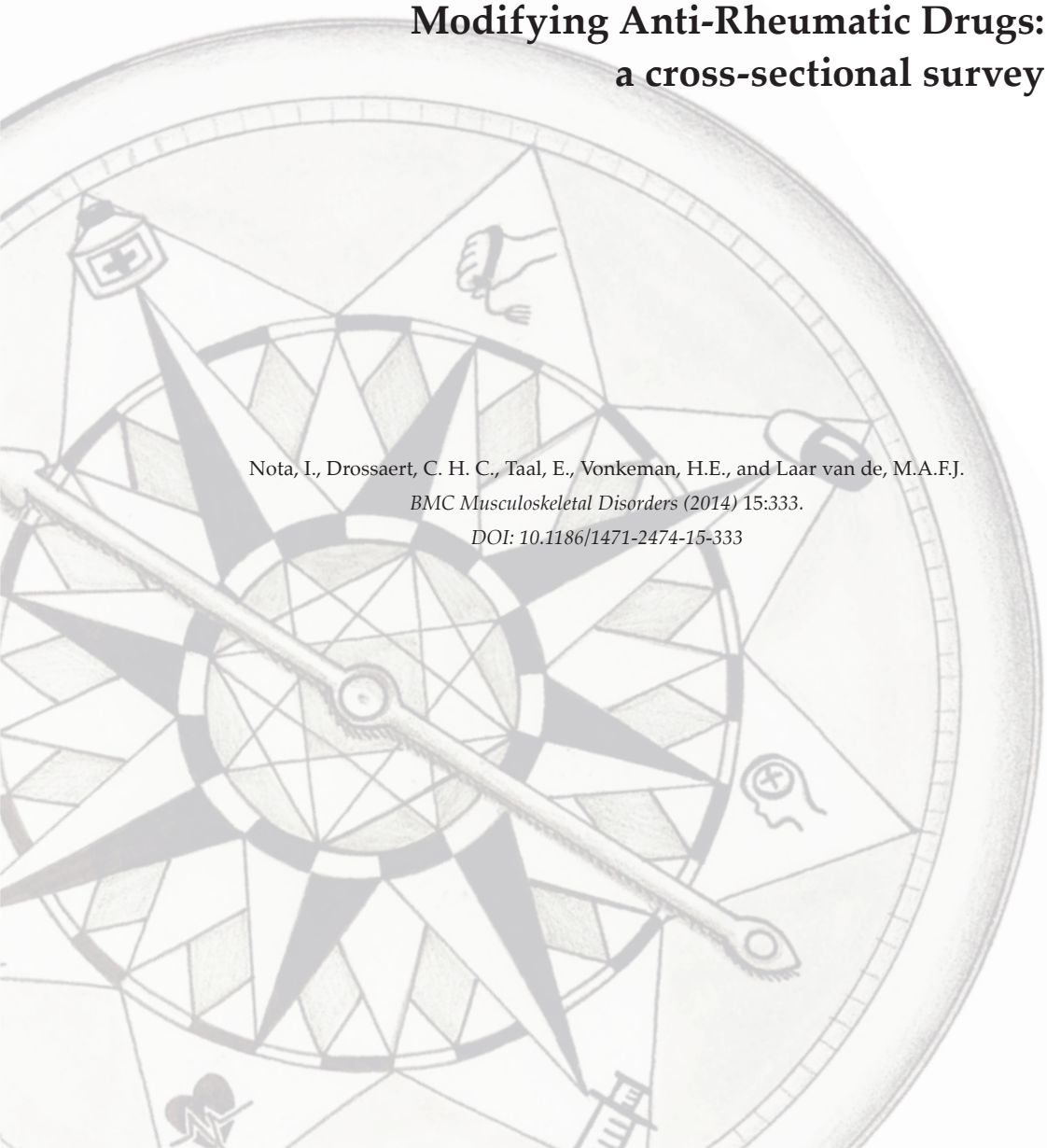
overcome the power imbalance between doctors and patients in medical decision-making.

There are some limitations to this study that need to be considered. Firstly, the participants in this study were recruited from two hospitals. Although these hospitals are large hospitals covering both urban and rural areas, this might limit the generalizability of the results. However, we have no reason to believe that patients from the eastern parts of the Netherlands, where this study was conducted, think differently about participation in MDM than patients from other Dutch regions. Future quantitative studies are needed to replicate and expand our results. Secondly, although we tried to prevent selection bias by preselecting patients diagnosed with RA before they consulted their rheumatologist, we cannot guarantee it did not occur. Thirdly, this was a retrospective study in which patients were asked to reflect on a recent medical decision, but sometimes that decision occurred weeks or months prior to the interview. However, what is potentially lost by these limitations was gained by allowing respondents to tell their own story.

Despite these limitations, our findings provide important practical information and recommendations for future research. It seems that recent attempts from the Dutch government to improve patient-centred care and SDM (104) (e.g. by developing PtDA's and quality indicators) have not yet been successful. We believe that the physician can have an important role as facilitator in enhancing patient participation in MDM, but implementing SDM is a shared responsibility; all parties need to be involved and educated. Physicians need to be aware of the fact that preferences regarding participation may vary both between and within individuals. They need to mention and explain all treatment options available and invite patients explicitly to participate in every treatment decision. Even if there is only one possible treatment option available, patients still have a choice – that is, to initiate or not – and they need to be asked about their opinion, worries, doubts and questions. With regards to the patient, more initiatives need to be taken that are directly aimed at patients to make them aware of the possibility to participate in MDM and of the potential value of their input (39). To support shared decision-making, the development and implementation of PtDA's using a holistic approach, which encounters the needs of all stakeholders (patients and health professionals) and the integration in the health care system, can be of great value. For future research, we recommend a quantitative and longitudinal study to show how patients' preferences regarding participation in rheumatology care may change over time - the patient during this time establishing a long-term relationship with the health-care professionals, - and how these preferences are related to the type of treatment and the severity of the complaints.

Chapter 3

Patient participation in decisions about Disease Modifying Anti-Rheumatic Drugs: a cross-sectional survey



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ABSTRACT

Involvement of patients in decision-making about medication is currently being advocated. This study examined (the concordance between) inflammatory arthritis patients' preferred and perceived involvement in decision-making in general, and in four specific decisions about Disease-Modifying Anti-Rheumatic Drugs (DMARDs). Furthermore, this study examined how patients' involvement is related to satisfaction about decision-making and which factors are related to preferred roles, perceived roles and concordance. Using a cross-sectional survey, 894 patients diagnosed with Rheumatoid Arthritis, Psoriatic Arthritis or Ankylosing Spondylitis were sent a questionnaire which focused on medical decisions in general and on four specific decisions: (a) starting with a synthetic DMARD; (b) starting to inject methotrexate; (c) starting a biologic DMARD; and (d) decreasing or stopping a DMARD. For each decision preferred and perceived involvement in decision-making was assessed using the Control Preference Scale. Concordance was calculated by subtracting the scores for perceived role from scores for the preferred role. Furthermore, satisfaction with the decision process and socio-demographic, health-related, patient-related and physician-related variables were assessed. The response rate was 58%. For all decisions, most patients (59%-63%) preferred Shared Decision-Making (SDM). SDM was perceived frequently (26%-55%) and patients' preferences were met in 54% of the respondents. Yet, in some specific decisions, 26% to 54% of patients would have liked more participation. Perceiving less participation than preferred was associated with less satisfaction with the decision-process, but perceiving more participation than preferred was not. Our results did not reveal any meaningful models to predict preferred or perceived participation in decision-making in general or with reference to specific decisions about DMARDs. Most arthritis patients prefer to be involved in decisions about their medication and SDM is perceived frequently. Yet, in some specific decisions patient participation can be further improved. Patients especially prefer more participation in decision-making regarding starting a first synthetic DMARD, which occurs most commonly in newly diagnosed patients. Whereas perceiving too little participation was associated with decreased satisfaction, perceiving too much participation was not. Therefore, rheumatologists should urge patients to participate in every medical decision.

INTRODUCTION

Medication use is central to the management of rheumatic diseases and medication adherence is essential for the success of the treatment. Syntheticly, decisions about medication have been viewed from a paternalistic perspective where the prescriber makes decisions based on medical knowledge and the patient either complies or does not comply with the prescribed regime (also defined as clinician-led decision-making).

Currently, involvement of patients in decision-making about medication is being advocated. Patient involvement in decision-making is considered beneficial for various reasons. First, the patient's agreement with the choice of treatment is important since the patient's cooperation in carrying out the treatment is essential (69). Secondly, Shared Decision-Making (SDM) is assumed to lead to improvement in health outcomes, such as health status, self-management, adherence, coping behavior and satisfaction with care (26-29), especially in chronic diseases (56). Finally, patients have the right to self-determination and should thereby be empowered by information about their diagnosis, treatment options and prognosis to make treatment decisions that correspond with their preferences and values (13).

Whereas patient participation is considered to be important and beneficial, SDM can be difficult to achieve for both doctors and patients. Doctors are often reluctant or unprepared to involve patients in medical decisions (33, 34). Some barriers mentioned by doctors are lack of time and low confidence in their ability to communicate risks effectively (35). Patients also experience barriers, such as unawareness of having a choice, low confidence to participate, a belief of having a lack of knowledge and uncertainty about which questions to ask (37, 38).

Furthermore, not all patients want to be actively involved in medical decision-making. Results of previous studies concerning patients' preferences regarding participation in treatment decisions show high variability (19, 74-77), including the field of rheumatology (22, 63-65). Although, there is extensive literature that has examined factors (socio-demographics, health-related, patient-related and physician-related) that predict patients' preferences regarding involvement, results are inconclusive and it remains difficult to explain or predict patient preferences (19, 64, 65, 72, 73, 80, 93, 105). Garfield, Francis and Smith (105) suggest that preference regarding involvement might vary per type of decision. Moreover, role preference may change over time and change as health status changes (19, 37, 65, 77, 93, 100). Thus, to pursue concordance between patients' preferred and actual role in decision-making, it is essential to study patients' preferences regarding involvement and to discriminate between specific decisions.

Compared to decisions in acute care, decisions in chronic care are more likely to need an active patient role in executing the decision (57). In rheumatology, decisions about medication reoccur during the process of the disease and are likely to be revised and reversed. However, the latter does not make it easier to make a decision. Treatment decisions have become increasingly complex due to the many new available Disease-Modifying Anti-Rheumatic Drugs (DMARDs). These drugs vary with respect to approximate time to benefit, side effects and risks, dosage, and route of administration.

Four specific decisions regarding DMARDs are particularly relevant: (a) starting with synthetic DMARDs; (b) starting to inject methotrexate; (c) starting a biologic DMARD; and (d) decreasing or stopping a DMARD. Guidelines strongly recommend early intervention with a synthetic DMARD (58, 106, 107). The recommended synthetic DMARD of first choice is methotrexate, which can be administered orally or by subcutaneous injection. In case of intolerance or disfavour for methotrexate, other synthetic DMARDs are good alternatives. In the Netherlands, therapy with a biologic DMARD can only be prescribed to patients with at least moderate disease activity and in whom treatment with at least 2 synthetic DMARDs has failed. The decision to decrease or stop medication occurs when the disease is in remission or when side effects are presented.

Although it seems the management of inflammatory arthritis is strongly protocolled, involving patients in decision-making about DMARDs is important, as some of the DMARDs can have serious side effects and the route of administration (orally, subcutaneous injection or intravenous injection) may have a large impact on patients' daily lives. Thus, to choose the best treatment is a process concerning clinical aspects, but also patients' preferences need to be considered. After all, these decisions require an active patient role in carrying out the decision and adherence is essential for the success of the treatment.

More knowledge about inflammatory arthritis patients' preferred level of involvement could lead to rheumatologists and other caregivers anticipating on this and make it easier to pursue the preferred level of patient involvement. We expect patients to be more satisfied with the decision-process if concordance is reached between the preferred and perceived level of participation. Whereas a few studies have examined inflammatory arthritis patients' preferred and/or perceived role in medical decision-making in general, to the best of our knowledge there is no data comparing patients' preferred and perceived role in specific decisions. Therefore, this study focused on inflammatory arthritis patients' preferred and perceived participation in various decisions related to the use of DMARDs. We studied the concordance between preferred and perceived roles and the perceived satisfaction

about the decision-making process. Furthermore, we examined which factors (socio-demographic, health-related, patient-related and physician-related) are associated with the preferred and perceived roles.

METHODS

Sample and setting

We focused our cross-sectional survey on patients with rheumatic diseases who were likely to use DMARDs: patients diagnosed with Rheumatoid Arthritis (RA), Psoriatic Arthritis (PsA) or Ankylosing Spondylitis (AS). Patients were recruited from two hospitals in the Netherlands: Medisch Spectrum Twente (MST) and Ziekenhuisgroep Twente (ZGT). Patients were selected with use of the electronic hospital records. First, a random sample of 965 patients (500 from MST, 465 from ZGT) who met the following criteria was selected: (a) consulted their rheumatologist in the past year; and (b) were diagnosed with RA, PsA or AS. The list of selected patients was then discussed with the treating rheumatologist. Based upon this, 71 patients were excluded because either the patient (1) was deceased, (2) had an incorrect diagnosis registered in the electronic hospital record, or (3) was not able to complete a Dutch written questionnaire (subjective interpretation by the rheumatologist). In total 894 eligible patients were sent a questionnaire by mail, accompanied by a letter of invitation from their rheumatologist and an informed consent form. The patients were asked to return the filled out questionnaires and the informed consent form to the University of Twente using a prepaid envelope. After three weeks a reminder was sent.

The study did not need approval of the ethical review board according to the Dutch Medical Research Involving Human Subjects Act (WMO); only (non-intervention) studies with a high burden for patients have to be reviewed.

Measures

Standardized scales were used as much as possible. If there was no Dutch scale available, scales were translated using the forward-backward procedure (108).

The questionnaire contained 65 questions and focused on medical decisions in general and on four specific decisions: (a) starting with a synthetic DMARD; (b) starting to inject methotrexate; (c) starting a biologic DMARD; and (d) decreasing or stopping medication. To make it easier for patients to remember the decisions addressed in the questionnaire, a short description of each decision was given including purpose, route of administration and generic and brand medicine names (see Box 3.1). For each decision patients were asked (1) whether they had ever faced the decision, and if relevant, patients were asked to think of the first time they faced

the decision. Then they were asked (2) what role they had perceived, (3) what the outcome of the decision had been (e.g. starting or not starting with the suggested medication), and (4) if they were satisfied with the decision-making process. Subsequently, all patients (including patients who had never faced the decision) were asked what role they preferred to have. Furthermore, socio-demographic, health-related, physician-related and patient-related variables were questioned.

Box 3.1. *Description of decisions as provided in questionnaire (translated from Dutch)*

Starting synthetic anti-rheumatic drugs
The following questions concern starting synthetic anti-rheumatic drugs, also called synthetic DMARDs. These drugs can reduce joint damage. They decrease disease activity: they ease pain and rigor and on the long term prevent further joint damage.
Examples: methotrexate (Emthexate®, Ledertrexate®), sulfasalazine (Salazopyrine®); gold (Tauredo®, Ridaura®), hydroxychloroquine (Plaquenil®), penicillamine (Gerodyl®), azathioprine (Imuran®), ciclosporine (Neoral®) and leflunomide (Arava®).

Starting to inject
Medication can be administered in various ways. Most drugs are administered orally as tablets. Another way is by subcutaneous injection. Methotrexate (Emthexate®, Ledertrexate®) is available as tablet, but can also be administered by subcutaneous injection. The following questions concern starting subcutaneous methotrexate injections.
Beware: these questions only concern methotrexate and not other drugs that may be administered by subcutaneous injection.

Starting biologic anti-rheumatic drugs
The following questions concern starting biologic anti-rheumatic drugs, also called biologic DMARDs. Biologic DMARDs are administered by subcutaneous injection or directly into a vein. Biologic DMARDs aim to reduce arthritis by inhibiting mediators of inflammation, such as TNF and Interleukine-1.
Examples: Adalimumab (Humira®), Etanercept (Enbrel®), Infliximab (Remicade®), Anakinra (Kineret®).

Decreasing or stopping anti-rheumatic drugs
For various reasons medication can be decreased or even stopped. This may be due to side effects or because you are doing so well that the dosage may be decreased. The following questions concern decreasing or stopping anti-rheumatic drugs.
Beware: these questions only concern your anti-rheumatic drugs.

Preferred and perceived participation and concordance. Preferred and perceived roles in medical decision-making were assessed with the 'Control Preference Scale' (CPS) (109) adapted by Garfield, et al. (65). Questions about the perceived role started with "In your opinion, who decided to …"; questions about the preferred role started with "If you are informed about the benefits and risks, who should finally decide about …". Response categories were: "The rheumatologist" (1), "Mostly the rheumatologist" (2), "The rheumatologist and me together" (3), "Mostly me" (4), and "Me alone" (5). Further, we recalculated the CPS scores to 3 levels: doctor (1-2), shared (3) and patient (4-5), as validated by Degner (109). Concordance was calculated by subtracting the

original perceived CPS scores from the original preferred CPS scores. The results ranged from -4 to 4 and were then coded into 3 levels: too little participation (<0), enough participation (0) and too much participation (>0).

Satisfaction. For each specific decision, satisfaction with the decision-making process was assessed with one item 'How satisfied are you about how this decision was made?' using a five-point Likert scale (very unsatisfied (0) – very satisfied (4)).

Socio-demographic variables included sex, age, marital status, education (low, medium, high), income (low, medium and high) and work status (employed vs. unemployed, volunteer, student, retired, or homemaker).

Health related variables included diagnosis (RA, PsA or AS), time since diagnosis (<1 year, 1-5 years, 5-10 years, or >10 years) and health-related quality of life. Health-related quality of life was assessed with the SF-12, version 2 (110). Standardized scores were calculated for the physical and mental well-being varying from 0 (poor well-being) to 100 (excellent well-being), with a mean of 50 and a standard deviation of 10 in the general population of the United States (110).

Patient-related variables. Need for information was assessed with a subscale of the Autonomy Preference Index (API) (93). The API consists of 8 items and patients respond on a five-point Likert scale. Response choices range from 0 (strongly disagree) to 4 (strongly agree). Sum scores were linearly adjusted to range from 0 (no need for information) to 100 (strongest possible need for information). Internal consistency was adequate (Cronbach's α=0.66).

Patients' self-efficacy in obtaining medical information and attention to their medical concerns by physicians was assessed using the 'Perceived Efficacy In Patient-Physician Interaction' (PEPPI) scale (111). The PEPPI consists of 10 items, each beginning with 'How confident are you in your ability to…' and using response options 1 (not at all confident) to 5 (very confident). Scores on the 10 items were added for each patient to acquire a total score, with higher scores indicating more self-efficacy. Internal consistency was good (Cronbach's α=0.91).

Physician-related variables. Characteristics of consultations with the rheumatologist included three variables: 1) frequency of visits in the last year (once a year, 2-4 times a year, more than 4 times a year), 2) having a regular rheumatologist ('How often do you consult the same rheumatologist') using a five-point Likert scale (always – never) and 3) duration of the relationship with the rheumatologist (in years).

Perceived trust in the physician and emotional support from the physician were assessed with 2 subscales of the 'Cologne Patient Questionnaire' (CPQ) (112, 113). Perceived trust in the physician was measured with 3 items. Patients' evaluation of emotional support was assessed with 4 items. Response choices range from 1 (strongly disagree) to 4 (strongly agree). Scale scores were computed for each scale by the mean of the items. Both scales range from 1 to 4 with a lower score indicating

lower trust or lower emotional support. Internal consistency of both scales were good (Cronbach's α=0.93 and α=0.85, respectively).

Prior to inclusion, we performed a pilot test among patients (n=8) to assess the readability and acceptability of time to complete the questionnaire. The test showed that the questionnaire took about 20-25 minutes to complete, which was acceptable according to the participants. Minor textual adjustments were made following the results of the pilot test.

Statistical analysis

To detect differences in the distributions of (concordance between) preferred and perceived participation across decisions, chi-square tests were performed. Chi-square tests were also used to detect differences in the distribution of preferred participation between respondents who had faced the decision versus respondents who had not faced the decision.

The Kruskal Wallis test was used to compare differences in satisfaction between groups with different levels of perceived participation and with different levels of concordance. Next, a post hoc Mann-Whitney U test with Bonferroni correction was used to test which groups were significantly different from each other.

To examine which factors are associated with preferred role and perceived role we performed multivariate binary logistic regression analyses. We predicted the preference for and perception of shared decision-making compared to clinician-led decision-making. The relationship with patient-led decision-making has not been analysed because of the too small numbers of patients preferring or perceiving this (frequencies ranging from 6 to 76, depending on the type of decision) relative to the number of predictors (n=13). We included the following predictors: age, sex, education, employment, years since diagnosis, physical and mental well-being, self-efficacy in patient-provider interaction, need for information, frequency of visits in the last year, duration of relationship with the rheumatologist, trust in physician and emotional support of physician.

RESULTS

Patient characteristics

We received 519 completed questionnaires (response rate 58%). The sample of respondents was heterogeneous in regard to socio-demographic and health related variables (table 3.1). The mean score for physical wellbeing was 39, somewhat lower than that of the US general population (50), but similar to that of the US RA population (110). The mean score for mental wellbeing was similar to that in the general population.

Most respondents visited their rheumatologist 2-4 times per year (n=344; 67.5%) and saw the same rheumatologist at almost every visit (n=501; 97%).

Table 3.1. *Demographic, health-related, physician-related and patient-related characteristics (n = 519)**

Variables	Categories	Value
Socio-demographic variables		
Age, years		56 ± 12
Women, no. (%)		285 (59)
Married/living with a partner, no. (%)		391 (82)
Education, no. (%)	Low (<12 years) Medium (12 – 16 years) High (>16 years)	155 (33) 220 (47) 94 (20)
Family income, no. (%)	Low (< €28.500/year) Medium (€28.500 - €34.000/year) High (> €34.000/year)	114 (31) 112 (31) 139 (38)
Fulltime and part time employed, no. (%)		198 (45)
Health-related variables		
Diagnosis (n,%)	Rheumatoid Arthritis Psoriatic Arthritis Ankylosing Spondylitis	307 (63) 120 (25) 58 (12)
Years since diagnosis, no. (%)	<1 1-5 6-10 >10	19 (5) 82 (21) 159 (40) 139 (35)
Well-being (SF-12) (range 0-100)	Physical Mental	39 ± 10 49 ± 10
Patient-related		
Self-efficacy in patient-provider interaction (PEPPI) (range 10-50)		39.9 ± 4.2
Need for information (API) (range 0-100)		71.7 ± 10.3
Physician-related variables		
Frequency of visits in the last year, no. (%)	once a year 2-4 times a year >4 times a year	75 (14.7) 344 (67.5) 89 (17.5)
Duration of relationship with rheumatologist (years)		7 (7)
Almost every visit the same rheumatologist, no. (%)		501 (97%)
Trust in physician (CPQ) (range 1-4)		3.48 ± .49
Emotional support of physician (CPQ) (range 1-4)		3.13 ± .49

** Values are the mean ± SD (range) unless otherwise indicated.*

SF12 = 12-Item Short Form Health Survey; CPQ = Cologne Preference Questionnaire; PEPPI = Perceived Efficacy in Patient-Provider Interaction; API = Autonomy Preference Index.

Concordance of preferred and perceived participation

Across decisions, most respondents (59-63%) preferred to share decisions about their treatment with their doctor, though a small but considerable group wanted the doctor to decide (table 3.2). We found no significant differences between respondents who had faced the decision versus respondents who had not faced the decision. A small, though statistically significant (Chi-Square = 15.22; df = 6; P = 0.02) difference in the distributions of preferred participation was found between the four decisions: regarding the decision whether or not to start injecting MTX, relatively more respondents preferred to decide by themselves (15% versus 9-11% for the other decisions). The distributions of role preference did not significantly differ between the other three decisions.

With regard to the perceived roles, the majority felt that decisions were often made by doctor and patient together (table 3.2). Yet, a considerable number of patients felt that ultimately the doctor made the final decision. We found a significant difference between the distributions of perceived participation between the four decisions (Chi-Square = 139.56; df = 6; P < 0.001). Some decisions stand out: 72% of the respondents perceived that the decision to start using a synthetic DMARD was made by the doctor alone, as opposed to 38% – 44% for the other decisions. On the other hand, for the decision to start injecting methotrexate or to decrease or stop medication, a considerable number of patients felt they had made the decision by themselves (17% and 24%, respectively).

Table 3.2. *Preferred and perceived role in medical decision-making*

Decision	**Preferred role**[1]				**Perceived role**[1]			
	Doctor (1)	Shared (2)	Patient (3)	Valid N	Doctor (1)	Shared (2)	Patient (3)	Valid N
MDM in general	31%	61%	8%	504	43%	55%	1%	506
Synthetic DMARD	32%	59%	10%	491	72%	26%	2%	368
Injecting MTX	25%	60%	15%	466	43%	40%	17%	162
Biologic agent	26%	63%	11%	471	44%	50%	6%	149
Decrease/stop	30%	61%	9%	489	38%	38%	24%	314

MDM = Medical Decision-making; DMARD = Disease Modifying Anti-Rheumatic Drug; MTX = methotrexate.

[1] *Data of perceived role included respondents who had ever faced the decision; data of preferred role included all respondents.*

Table 3.3 shows the data on concordance of the preferred and perceived roles. For 43% - 62% of the patients, a match was established between the preferred and perceived roles. A considerable group (26% - 54%) perceived "too little" participation, compared to their preference. Again, there was considerable and significant variation between decisions (Chi-Square = 120.99; df = 6; P < 0.001): more than half of the respondents perceived too little participation with the decision to start using a synthetic DMARD and almost one third perceived too much participation in deciding to decrease or stop their medication.

Table 3.3. *Concordance between preferred and perceived role*

	Too little participation	Enough participation	Too much participation
MDM in general (n=496)	29%	61%	10%
Synthetic DMARD (n=330)	54%	43%	4%
Injecting MTX (n=137)	29%	56%	14%
Biologic agent (n=129)	30%	62%	8%
Decrease/stop (n=303)	26%	46%	28%

MDM = Medical Decision-making; DMARD = Disease Modifying Anti-Rheumatic Drugs; MTX = methotrexate.

Satisfaction with the decision process

Most respondents (83% – 89%) felt "satisfied" or "very satisfied" with the decision process in general and for each specific decision. There were however significant differences in satisfaction between the three levels of perceived participation (doctor, shared, patient) (table 3.4). For most decisions, patients were more satisfied when they participated in decision-making.

Regarding the relationship between satisfaction and concordance, we expected that patients who achieved concordance ("enough participation") would be more satisfied than those who perceived "too little" or "too much" participation. Our results indeed revealed that, for most decisions, patients who perceived "too little" participation were significantly less satisfied. Yet, getting "too much" participation did not decrease satisfaction. Overall, our results suggest that perceiving "too much participation" is not related to less satisfaction.

Table 3.4. *Satisfaction with the decision process[1] by perceived role and by concordance*

	Perceived role				Concordance			
	Doctor Mean (SD)	**Shared** Mean (SD)	**Patient** Mean (SD)	P[2]	**Too little participation** Mean (SD)	**Enough participation** Mean (SD)	**Too much participation** Mean (SD)	P[3]
MDM in general (N=502)	4.0[a] (0.8) (N=218)	4.2[a] (0.7) (N=278)	4.0 (0.6) (N=6)	.04*	3.9[ab] (0.8) (N=142)	4.2[a] (0.7) (N=302)	4.4[b] (0.5) (N=52)	.00**
Synthetic DMARD (N=332)	3.9[a] (0.7) (N=234)	4.2[a] (0.6) (N=90)	4.3 (0.5) (N=8)	.00**	3.8[ab] (0.6) (N=177)	4.1[a] (0.6) (N=141)	4.3[b] (0.7) (N=12)	.00**
Injecting MTX (N=137)	3.8[a] (0.9) (N=53)	4.2[a] (0.6) (N=59)	4.0 (0.7) (N=25)	.02*	3.7[ab] (0.8) (N=41)	4.1[a] (0.7) (N=76)	4.3[b] (0.7) (N=20)	.00**
Biologic agent (N=131)	4.0 (0.8) (N=58)	4.3 (0.6) (N=67)	4.2 (1.6) (N=6)	.16	4.1 (0.9) (N=39)	4.3 (0.7) (N=80)	3.8 (1.2) (N=10)	.31
Decrease/stop (N=304)	3.9[a] (0.8) (N=115)	4.1[ab] (0.6) (N=115)	3.8[b] (0.8) (N=74)	.00**	3.8[a] (0.8) (N=80)	4.1[ab] (0.7) (N=139)	3.8[b] (0.8) (N=84)	.00**

MDM = Medical Decision-making; DMARD = Disease Modifying Anti-Rheumatic Drug; MTX = methotrexate.

[1] ranging from 0 – 4 in which higher scores indicate more satisfaction.

[2] p-levels for differences between doctor, shared and patient, tested with the Kruskal-Wallis test.

[3] p-levels for differences between too little, enough and too much participation, tested with the Kruskal-Wallis test.

[a or b] Distributions are significant different from each other (post hoc test with Mann Whitney with Bonferroni correction)

** Significant on the .05 level*

*** Significant on the .01 level*

Factors associated with preferred and perceived roles

When analyzing factors associated with the preference for and perception of SDM compared to clinician-led decision-making, only a few significant weak relationships were found in the multivariate binary logistic regression analysis. For the preference for SDM in general medical decision-making only age (OR = 0.960; 95% CI 0.935 - 0.985; p = 0.002) and education (OR = 1.462; 95% CI 1.007 - 2.121; p = 0.046) were significant predictors; meaning that younger and higher educated patients more often prefer SDM than clinician-led decision-making. However, the goodness of fit of the total model was small (Nagelkerke Pseudo $R^2 = 0.102$; p = 0.028). No significant relationships were found between preferred role and any of the other variables included in the regression analysis.

For the perception of SDM in general medical decision-making only physical wellbeing (OR = 0.973; 95% CI 0.949 - 0.998; p = 0.036) and emotional support (OR = 2.232; 95% CI 1.172 - 4.251; p = 0.015) were significant predictors; meaning that patients with physical problems and who perceive more emotional support from their attending physician more often perceive SDM than clinician-led decision-making. However again, the goodness of fit of the total model was small (Nagelkerke Pseudo $R^2 = 0.130$; p = 0.001). None of the other variables that were included in the regression analysis were significantly related to the perception of SDM.

We also analyzed factors associated with preference for and perception of SDM for specific decisions (data not shown), but no clear pattern arose and relationships were small (R^2's for all models < 0.14).

DISCUSSION

Our study shows that the majority of patients with RA, PsA and AS prefer to share decisions about medication, although a small, but significant group still wants the doctor to decide. These results are in line with other studies in rheumatology (22, 63-65) and other chronic diseases (24).

To the best of our knowledge, our study is the first in the field of rheumatology that examined the concordance between preferred and perceived roles. Our study shows that, in rheumatologic outpatient care, Shared Decision-Making is perceived frequently and patients' preferences are met in over half of the patients. However, the amount of concordance varied significantly between decisions; a considerable group (on average 34%) still wanted more participation than they perceived. These results are comparable to studies examining other conditions, such as cancer and

asthma where concordance levels varying from 34% - 66% have been reported (36, 75, 79, 80, 100, 114).

We also examined the relationship between concordance in patient participation and patients' satisfaction. We expected that patients would be less satisfied with the decision process if they perceived either too little or too much participation. However, our results suggested that patients are only less satisfied if they perceive too little participation. If patients perceived more participation than preferred, they were still highly satisfied. Although many studies have shown that SDM can improve satisfaction (26-29, 56), to our knowledge it has not been previously reported that offering a greater than preferred level of participation is not related to diminished satisfaction, but offering too little is. These findings implicate that patients should be invited to participate in medication decision-making by their rheumatologist at all times.

Previous studies on patient involvement in medical decision-making have mostly looked at decision-making in general. Our study discriminated between medical decision-making in general and four specific decisions that are common in rheumatology. Contrary to our expectations, we found no relevant variety in role preference between these decisions. It seems to be that role preference in decisions about medication for rheumatic diseases is rather stable.

Although we did not find any relevant differences in the distribution of role preferences between the four decisions, we did find differences in the distribution of perceived role and concordance. Two decisions stand out: the decision to start using a synthetic DMARD and the decision to decrease or stop a DMARD. With this first decision, the majority of patients (72%) perceived that the doctor decided and in contrast to the other decisions, more than half of the patients (54%) did not achieve their preferred level of participation. An explanation for this finding might be that in the setting of starting a synthetic DMARD for the first time there is lack of awareness of choice and too little time for patients to participate. The decision to start a synthetic DMARD for the first time is a decision that occurs most commonly in newly diagnosed patients. The current guidelines for early arthritis recommend starting with aggressive treatment as soon as possible, with methotrexate being the recommended drug of first choice. It is plausible that patients initially only receive one treatment recommendation and are not aware of alternative treatment options. Upon receiving the diagnosis, the patient needs to process a lot of information (about the influence of this chronic disease on daily life, starting aggressive treatment, etc.) in a short time. Not being aware of having a choice, little time, and/or an overload of information may be a barrier for patient involvement (37, 38). In clinical practice, extra time (to think and to create awareness of choice) needs to be considered when

dealing with newly diagnosed patients that need to make decisions about starting with a synthetic DMARD. Additionally, patients need to be urged to participate to arrive at a decision concordant with their values. These actions may not only enhance patients' satisfaction, it may also increase patients' self-efficacy in being adherent to medication use (115-118).

The decision to decrease or stop medication stands out because, in contrast to the other decisions, relatively many respondents (24%) felt they had made this decision by themselves. Moreover, a large group perceived too much participation in this decision. The decision to decrease or stop medication is different from the other decisions, because it occurs when the disease is in remission or when side effects are presented. It is possible that patients are more strongly invited to participate in the decision to decrease or stop their medication, because it is more preference sensitive. Previous studies have shown that patients fear returning symptoms (in the case of remission) or unknown side effects (when changing therapy) (119, 120). More research is necessary to clarify why patients feel they are too much involved in the decision to decrease or stop medication. These studies should discriminate between decisions to decrease or stop medication in case of remission or to decrease or stop medication when side effects are presented.

The final aim of this study was to examine for each decision which factors were associated with preferred and perceived roles. Although we assessed many possible variables, our results did not reveal any meaningful models to predict preferred or perceived participation in decision-making in general or with reference to specific decisions about DMARDs. In rheumatology, only a few studies have examined associated factors for preferred and perceived roles and those results are inconclusive (22, 63-65, 121). For example, female gender was significantly associated with higher preferences for involvement in decision-making in one study (63), but not others (22, 65). Likewise, younger age has been reported as a significant predictor of preference for involvement (22, 63, 65), but in our data we only found weak correlations and the results varied per decision. As far as we know, only two previous studies have examined associations with perceived involvement in rheumatology (26, 121). Although this study found several significant associated factors, the presented odds ratios for high involvement were low or with a high confidence interval (indicating a low level of precision of the odds ratio) (122). Results of studies using other populations were also inconclusive (36, 78, 114, 123-126). Our findings imply that it remains difficult to identify subgroups that are more in need of being involved. However, as our results revealed that too much participation is not related to diminished satisfaction, we can recommend that caregivers facilitate patient participation in all of their patients. Therefore we suggest training in SDM should be emphasized in educational programs of rheumatologists.

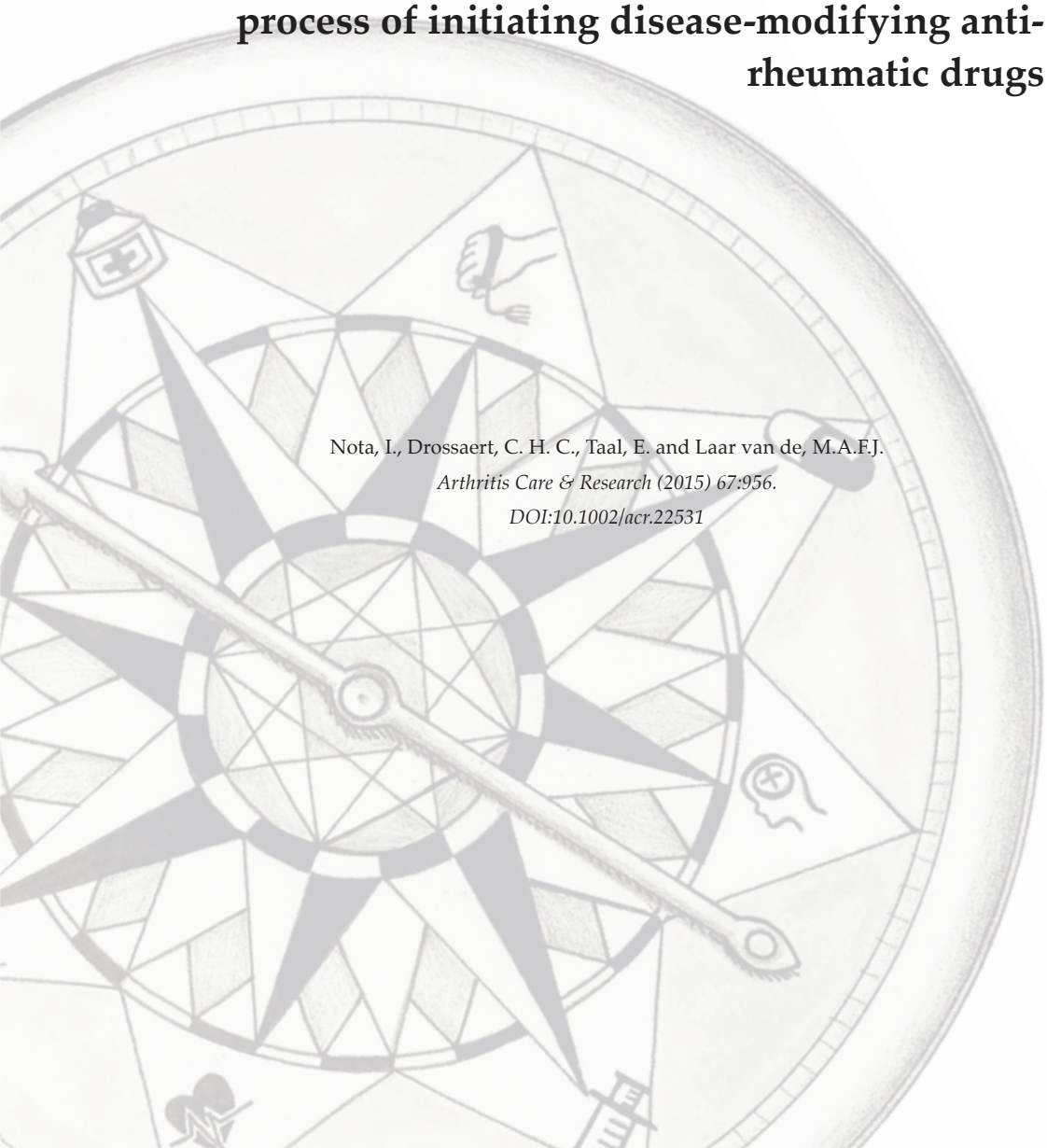
A strength of this study is its large representative sample of patients to examine preferred roles, perceived roles, concordance and satisfaction in various decisions regarding medication use in rheumatology. Due to some limitations of the study, some caution is necessary when interpreting our results. First, due to sizable non-response, our results might be slightly biased. Although we had a response rate of 58%, selection bias might have occurred. It is possible that patients who have no need of participating in medical decision-making, are less interested in (responding to questionnaires about) patient participation. Second, due to limited resources we chose to conduct a retrospective study and therefore it is possible that recall bias occurred. We questioned patients about the first time they faced these decisions. Some of these decisions may have occurred years before the study. Even though we did not find any significant differences in preferred and perceived role between patients with a long (>1 year) and short (<1 year) illness duration, the limitation of possible recall bias remains. We therefore recommend a prospective study which questions patients at the time of the decision. Third, no patient representatives were included in our research group as has been recently recommended (127). Patient representatives can provide valuable suggestions about which aspects to include in the questionnaires and the interpretation of results. Yet, as the current study is part of a larger project to develop a Patient Decision Aid (PtDA) for anti-rheumatic drugs in which patients were repeatedly and extensively involved in various research and design activities, we feel that we have included patient perspectives to at least some extend.

CONCLUSION.

In conclusion our study shows that arthritis patients appreciate being involved in decisions about their medication and that shared decision-making is perceived frequently in rheumatology outpatient care. Yet, patient participation can be further improved, particularly in decision-making about starting a synthetic DMARD for the first time. As our results revealed that too much participation is not related to diminished satisfaction, we recommend assessing patients' preferred and perceived role in medical decision-making regularly and invite patients to participate in every decision. Moreover, we recommend rheumatologists and other caregivers to consider extra time for patients to create awareness of choice and to process all the information, especially when dealing with newly diagnosed patients that need to make decisions about initiating a synthetic DMARD.

Chapter 4

Patients' considerations in the decision-making process of initiating disease-modifying anti-rheumatic drugs



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ABSTRACT

This study was conducted to explore what considerations patients have when deciding about disease-modifying anti-rheumatic drugs (DMARDs) and what information patients need to participate in the decision-making process. In-depth face-to-face interviews were conducted with 32 inflammatory arthritis patients who recently consulted their rheumatologist and discussed initiating DMARDs. Beliefs in the necessity of DMARDs, either for relief of symptoms or prevention of future joint damage, were reasons to initiate DMARDs. Furthermore, trust in the rheumatologist and the healthcare system was important in this respect. Patients expressed many concerns about initiating DMARDS. These related to the perceived aggressive and harmful nature of DMARDs, potential (or unknown) side effects, influence on fertility and pregnancy, combination with other medicines, time to benefit and manner of administration. Participants also worried about the future: about long term medication use and the feeling of dependency, and, -if this medicine proved to be ineffective-, about the risks of future treatments and running out of options. To decrease this uncertainty, participants wanted to be informed about multiple treatment options, both current and future. They did not only want clinical information, but also information on how the medications could affect their daily lives. Health education should inform patients about multiple treatment options, for the current time being as well as for the future. It should enable patients to compare treatments with regards to both clinical aspects as well as possible consequences for their daily lives.

INTRODUCTION

In recent years, significant progress has been made in treating inflammatory arthritis. Current guidelines allow a variety of substances, including non-steroidal anti-inflammatory drugs (NSAIDs), corticosteroids, synthetic and biologic disease-modifying anti-rheumatic drugs (sDMARDs and biologic DMARDs) (58, 106). These treatment approaches vary in time to benefit, dosage and mode of application (e.g. oral, subcutaneous, intravenous), potential side effects and risks (e.g. toxicity), and costs (58, 106). With the continuing introduction of new treatment strategies, patients as well as rheumatologists face increasingly complex decisions about how, when and what to initiate. Choosing the best treatment is a process concerning both clinical aspects and patients' preferences.

Nowadays, patients' involvement in decision-making about medication is advocated (58), because it is both considered a patient's right (13) and associated with positive effects on patients' satisfaction with healthcare, self-management, coping behaviour and adherence (26-29). In rheumatology, patients have a high need for information (26, 62-64) and most want to participate in decision-making (22, 63-65, 128). Furthermore, patient participation is important because studies show that patients' and rheumatologists' beliefs about illness and treatment may differ. This includes how they rank the potential benefits and side effects of available treatment options and how they prioritize long-term outcomes (120, 129-139). Therefore high quality patient-physician communication about the choice of treatment is important.

Patient Decision Aids (PtDA's) are tools designed and implemented to prepare patients to participate in making specific and deliberated choices among healthcare options (50-52, 140, 141). PtDA's differ from standard health education in various ways. Firstly, PtDA's specifically state the decision being considered and they emphasize the importance of patients' role in decision-making. Secondly, PtDA's describe multiple treatment options (instead of one). Finally, they help patients to get insight into their personal values and preferences (52, 103). According to a recent systematic review, PtDA's have shown to be effective for many different decisions and conditions (103). PtDA's can improve patients' knowledge about options, risk perceptions, their feeling of being informed and being certain about what matters to them (103). Furthermore, evidence indicates that PtDA's improve patient-doctor communication (103). Patients using PtDA's are more likely to reach decisions that are consistent with their personal values (103). In rheumatology, PtDA's are relatively new; only a few studies on PtDA's about DMARDs have been reported (30, 66).

To develop a PtDA that supports patients to think about personal values and prepare them to participate in decision-making, it is essential to understand how patients

decide about DMARDs. A number of studies described how arthritis patients decide about DMARDs (102, 120, 137, 139, 142). However, these studies were either retrospective (120), scenario based (137, 139) or focused on adherence (102, 142).

A theoretical framework often used to understand patients' considerations about medication is the Necessity/Concerns Framework (NCF) (143). The NCF states that two beliefs about medication are important: (a) the perceived necessity for the medicine to maintain or improve their current and future health and (b) the perceived concerns about potential adverse effects of using the medicine. This theory has been developed and used extensively to understand (non)adherence to medication, but may also be helpful to understand patients' beliefs and considerations before initiating medication.

This qualitative study aims to deepen the understanding of patients' considerations when deciding about DMARDs and what information patients need to participate in the decision-making process. The results of this study have been used to develop a PtDA for initiating DMARDs.

METHODS

Participant selection and recruitment

This study focused on patients diagnosed with Rheumatoid Arthritis (RA), Ankylosing Spondylitis (AS) or Psoriatic Arthritis (PsA), who recently (< 1 month ago) consulted their rheumatologist and discussed initiating DMARDS. The participants were required to speak Dutch. During the study period (September 2011 until March 2012) all patients who visited the clinic and fulfilled the inclusion criteria were informed about the study by their rheumatologist and were asked to give permission for contacting them. In total 34 patients were contacted by the researcher; 33 patients consented to be interviewed. Eventually, one participant cancelled the appointment without providing a reason. The remaining 32 interviews were held within 1 month from the consultation with their rheumatologist. No approval of the ethical review board according to the Dutch Medical Research Involving Human Subjects Act (WMO) was needed; only (non-intervention) studies with a high impact for patients have to be reviewed.

Procedure

In-depth face-to-face interviews were conducted by the first author (IN). The interviews were audio recorded and lasted between 45 and 120 minutes (average time 66 minutes). Depending on the patient's preference interviews took place at

patients' homes or at the university. Patients were informed that the results would be reported anonymously and that they could withdraw from the study at any time. Before the interview, participants signed an informed consent form. We used a semi-structured interview method where the order of questions followed the natural progression of the conversation (144). The first part of the interview assessed patients' considerations, questions, concerns and information needs when deciding about DMARDs. The second part assessed patients' need for a PtDA about initiating DMARDs and patients' preferences for its content and design; these results will be reported elsewhere. The first two interviews were used for intermediate evaluation of the interview guide. Minor adjustments were made to ease the usability and question 3 was added. The final interview guide can be seen in box 4.1.

Box 4.1. *Interview guide (translated from Dutch)*

1. First of all, I would be interested to hearing about what happened since you have had your diagnosis.
   - Onset of illness, previous medication and effects and side effects.
2. You have recently discussed with your rheumatologist to initiate <name of drugs>. Could you tell me something about that?
   - What thoughts did you have about it? (considerations, expectations, concerns, doubts, questions)
   - What information did you receive, from whom and what was your opinion about the information?
   - After the consultation, did you have any questions that were left unanswered?
   - Were you satisfied with the level of information?
   - Was any information lacking?
   - Did you search for additional information? (resources and opinion about the information)
   - Did you decide to initiate <name of drugs>? Why (not)?
3. You have recently initiated <name of drugs>, what do you know now about the medication that you would have liked to know before you started to use this medication? (if applicable)
4. Demographic data
   - Age, gender, education, employment, marital status.

Analysis

Interviews were transcribed verbatim and analysed using ATLAS/ti 7.1, a qualitative analysis software application which allows to overview the codes, link statements and visualize connections between codes (145). Firstly, the analysts (IN and CHCD) read the transcripts to familiarize themselves with the content. Then, one analyst (IN) selected relevant fragments (sentences or small paragraphs) related to both main themes: (a) patients' considerations when deciding about DMARDs and (b) need for information and satisfaction with obtained information. Both analysts mutually independently further categorized the fragments. To segment and reassemble the data, they used the principle of constant comparison (146) and a process of open coding, followed by axial coding and selective coding (146, 147) until a final thematic framework was developed for each theme. Finally, the Necessity/

Concerns Framework (NCF) (143) was, amongst others, used to interpret the data. The individual findings were compared and analysed until consensus was reached. After analysing 27 of the 32 interviews no meaningful new information was found in the last three interviews, hence the analysts concluded that saturation occurred. This was confirmed by the final five interviews. All quotes in this article were literally translated from Dutch to English by a professional translator.

RESULTS

Participant characteristics

In total 26 women and 6 men participated. Some participants (N=5) discussed initiating DMARDs with their rheumatologist for the first time. Others (N=27) already used sDMARDs or bDMARDS before. They discussed changing to another DMARD. Table 4.1 lists demographics and characteristics. Figure 4.1 shows the status of the decision at the time of inclusion and the time of the interview.

Table 4.1. *Participant demographics and characteristics.*

	sDMARDs[1] (N=16)	bDMARDs[2] (N=16)	Total (N=32)
Socio-demographic variables			
Women, n. (%)	11 (69)	15 (94)	26 (81)
Age, average (range)	58 (31-76)	50 (25-82)	54 (25-82)
Married/living with a partner, n. (%)	14 (88)	14 (88)	28 (88)
Education			
Low (< 12 years), n. (%)	5 (31)	2 (13)	7 (22)
Medium (12 – 16 years), n. (%)	7 (44)	13 (81)	20 (62)
High (> 16 years), n. (%)	4 (25)	1 (6)	5 (16)
Full-time and part-time employed, n. (%)	9 (56)	9 (56)	18 (56)
Health-related variables			
Diagnosis			
Rheumatoid arthritis, n. (%)	14 (88)	14 (88)	28 (89)
Ankylosing spondylitis, n. (%)	1 (6)	2 (12)	3 (9)
Psoriatic arthritis, n. (%)	1 (6)	0 (0)	1 (3)
Illness duration, average in years (range)	5.6 (1-40)	2.0 (0-15)	7.8 (0-40)
Number of previous DMARDs, median (range)	1 (0-3)	2 (2-7)	2 (0-7)

[1] *sDMARDs = synthetic disease-modifying anti-rheumatic drugs*

[2] *bDMARDs = biologic disease-modifying anti-rheumatic drugs*

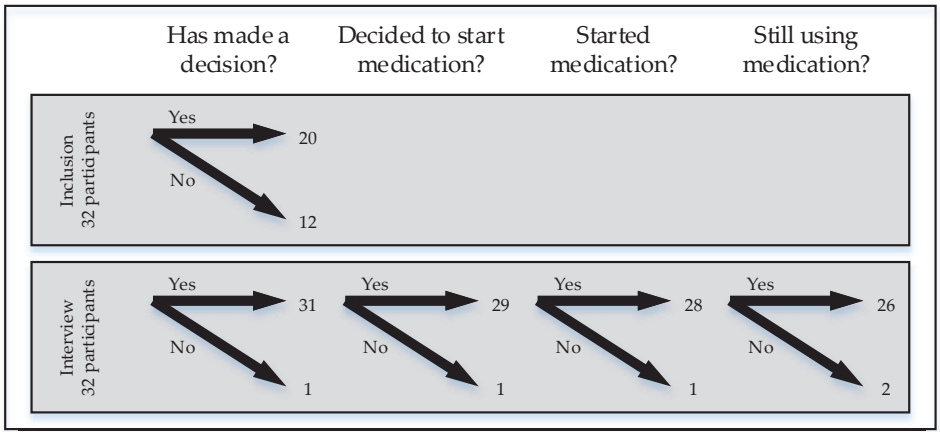


Figure 4.2: *Status of decision at time of inclusion and interview*

Patients' considerations and concerns

We asked participants for their considerations, questions and concerns regarding initiating DMARDs. Answers were grouped in three considerations in favour of and two considerations against initiating DMARDs, which are presented in table 4.2 and 4.3, respectively.

Considerations in favour of initiating DMARDs

'Necessity', the first category, was based on the NCF. Necessity was often associated with a need for relief of symptoms. Some participants deliberately chose to initiate the medication to ease their complaints. For example, some stated that they valued current quality of life over potential future adverse effects. Others were so desperate in need to ease their complaints (e.g. pain), that they consented immediately to the proposed medication: *"I would have accepted anything"*. In other cases the perceived necessity was less associated with direct symptom relief, but rather with prevention of future joint damage.

'Trust', the second category, exceeds the necessity component of the NCF. Some participants completely relied on their rheumatologist or the health care system. They did not have any considerations or concerns about initiating DMARDs and just started the prescribed medication without reviewing other options.

'Relative benefits' is the third and final category. It relates to benefits the drug may have compared to other options. Participants mentioned several benefits influencing their decision to initiate the proposed medication, especially lower risks of side effects, less frequent and friendlier manner of drug administration.

Table 4.2. *Patients' considerations in favour of initiating (different) DMARDs*

Category	Subcategory	Quotes (translated from Dutch)
Necessity	Symptom relief	*The pain. You want to get rid of the pain. I've already had so many painkillers. You just want to try something else.*
		I need to use it now. I want to have a good time, so the stuff just needs to work now! And well, what's it like when I'm 65, we'll see about that when I get there. That is something to worry about later, at least I'd be able to tell I've had a good life.
		Yes, so I'll accept anything. As long as it gets better.
	Prevention of joint damage	*I once knew someone with the same problems. [...] But that person was already in an advanced stage, had new joints, was given an artificial hip and an artificial knee. But that person was of my age and [...] knowing that, I sort of thought, well, now I must act before it gets any worse.*
Trust	Trust in physician	*So I presume they know what they're doing. No, in good spirits that we'll start, no. And let's see if it helps.*
	Trust in health care system	*I still have a little faith in the health care system.*
Relative benefits	Lower risk of side effects compared to other drugs	*Because of less… It had side effects though, but less…*
	Less frequent drug administration compared to other drugs	*And that means not having to take all those drugs each day, since I only have to take this injection once in a fortnight.*

Considerations against initiating DMARDs

The first category, 'concerns', originates from the NCF. Participants mentioned numerous concerns about initiating DMARDs. Many perceived DMARDs in general as aggressive and harmful medicines, making them doubt the necessity. More specifically, participants worried about both the number of potential side effects as well as specific side effects. The latter included the risk of a weaker immune system, the loss of hair, loss of vision or developing cancer. Some of the available DMARDs are relatively new drugs where robust long term safety and efficacy data lacks. This makes many participants feel like a guinea pig. Younger participants also tended to worry about the influence on fertility and/or pregnancy. Some unexpectedly had to think about having children in the upcoming years. Participants also worried about the combination with other medicines, the time to benefit and the manner of drug administration. Notably, many had concerns about future treatment of their arthritis, for example: *"What if this drug does not work?"* There may be a difference between sDMARDs and bDMARDs. Participants facing the decision to initiate sDMARDs worried about the side effects of both the current proposed drug as well as future options. As they assumed that doctors will first propose the drug with the lowest risks, they had concerns about what kind of risks future treatments will bring: "*If*

this is the drug they start with [methotrexate], what will be the side effects of the next drug?" Participants facing the decision to initiate bDMARDs especially worried about the availability of future treatments, because they perceived initiating bDMARDs as the 'final option'. Finally, some worried about the long term use and dependency.

Table 4.3. *Patients' considerations against initiating (different) DMARDs*

Category	Subcategory	Quotes (translated from Dutch)
Concerns	Aggressive and harmful nature of medicine	*Is this really necessary? I mean, it's not just an ordinary painkiller that you get prescribed. It is quite a strong medicine.*
		I had hoped that it wasn't necessary [to start with bDMARDs] and that I could go on much further with the methotrexate.
	Risks for side effects	*Yeah, when I read all that, immediately I won't touch it, there are so many side effects.*
		The long term risks. Well, they're the ones that really stick out and of which not much information is given.
		What I feared the most when taking this drug was that it has an impact on the immune system. I really find that very hard to accept.
		Well, and then I said, I only want to get better and not … serve as a guinea pig.
	Influence on fertility and pregnancy	*I was confronted with the question if I had any desire to have children. I mean, we haven't considered the option yet. We've only just started our relationship.*
	Combination with other medicines	*And then I had to take all of that. And I think: "Oh, I'll be loaded with drugs. How will they all go together?"*
	Time to benefit	*I ask myself: "Is it going to work?" You just don't know right away. You have to wait.*
	Manner of drug administration	*Yes, he did discuss injections and the like, but I'd rather not, I'd rather not yet do that.*
	Future treatment	*I was also thinking, if this is the drug they start with [methotrexate], what will be the side effects of the next drug?*
		Once you start with this [bDMARDs], you can't go any further. [...] With methotrexate you know that there's always something stronger, in case it doesn't work anymore, you can still move on. And now you know, well, that's it and there's nothing stronger anymore, [So I wonder] has the moment arrived that I should start with such drugs?
		And yes, the only thing is, for how long should you take it? And I just would like to know what comes next, for how long can you continue? Can I ever stop taking this?
Emotional impact	Amount and number of drugs	*The more medication I take, the more I must face the facts of having a number of conditions, I really am seriously ill.*
	Severity of drug	*My first response [to the bDMARDs via infusion] was: "Oh my, we've gone down one step further." To put it like that.*

The second category, 'emotional impact', exceeds the concerns component of the NCF. Some participants considering DMARDs experienced an emotional impact; they realized they are seriously ill. First, having to take many (different) or much drugs (volume, frequency, type), made some participants feel more ill and therefore reluctant to initiate DMARDs: *"The more medication you take, […] the more ill you feel. Maybe even more than you really are."* Second, some patients' experienced an emotional impact because of the perceived severity of the proposed medication. Certain drugs, especially bDMARDs, were associated with aggravation of the disease. As this quote illustrates: *"This [initiating bDMARDs] is the next step. […] Now it gets serious. […] Because you now are 'privileged' for initiating this category of drugs."*

Need for information and satisfaction with obtained information

The need for information varied highly. Some wanted to know every detail: *"I always get down to the details. Why? I don't know. I need to know what I'm taking and what to expect"* and *"I read everything. I know I shouldn't, because I only start to worry, but still…"* Others were satisfied with the provided information: *"For me it's actually quite plain. I just don't feel like going into that much further than I have to, until you start thinking you should do something. I think that, er… knowing too much, er… works obstructive, I follow my feelings. [...] if you have little information, there's little to worry about."*

We asked participants about the obtained information; from whom and how they valued it. Participants mentioned different sources of information, and most sources got both positive and negative remarks (table 4.4). Whereas many patients were satisfied with the information provided by health professionals, sometimes the difference between the proposed options was not clear, especially concerning bDMARDs. Others found it difficult to comprehend contradictive information from different caregivers. Some participants had searched additional information on the internet about the proposed treatment options. This was mostly satisfying, as it confirmed the information the doctor provided. Some also read stories of peers. Not all valued that type of information as they could not relate to these experiences. After deciding which treatment to initiate, participants received an information leaflet from the pharmacist and the package insert from the manufacturing pharmaceutical company. Relating to bDMARDs, the manufacturing pharmaceutical company often provided additional material, such as books, CD-ROMs, DVDs, etc.

Table 4.4. *Opinions about information received through various sources*

Source	Positive quotes (translated from Dutch)	Negative quotes (translated from Dutch)
Doctor / Nurse	*Very good explanation on the injections. [...] she not only showed it, but made me try it myself. That way, you know you're doing it correctly.*	*I could choose, but the difference [between the biologic injections] is not entirely clear to me. Except for the frequency of applying.*
	The pros and cons were discussed. And the side effects. [...] And whenever I've got any question, I can always ask or call.	*The rheumatology nurse, the substitute rheumatologist and my own rheumatologist all gave contradictive information. That wasn't easy.*
Leaflet from hospital	*So they gave me this booklet and just like I said, at such a moment you are caught off guard, so I liked reading about it all over.*	*I was given five leaflets and could make a choice. I read the documents. Well, they didn't contain much for a patient. It's no use. And then I said: "You choose." The leaflets really don't say much.*
Internet	*Well, what I saw, like I just said, was the same information I got from the rheumatologist, so I assume that's what I can rely on.*	
	I think it's just a feeling. [...] Whenever I think, oh well, this can't be true, I don't read [that website] any further. Then I'll return to reliable sites. And generally speaking, most of the doctor's and hospital's sites are.	
Online support groups	*Yeah, and then I think, well yes, it's a bit more personal, a bit more realistic or so. You think, why yes, they really must have written it themselves.*	*I don't need anyone else. I'll find it out my own way. If I have to hear other people's. No, thanks. I would be happy to help, though. But I'm doing this for myself, and not in a group.*
	It is of course nicely anonymous, so everyone is really very open.	*Yes, it can be hard what people experience, I always have the idea that people who are doing not so fine write these things.*
		You feel and experience it yourself and that can be completely different for someone else. Like pain levels, they can be different for everyone.
Pharmacy	*I always ask if I can combine it with all those other tablets, [...] but the pharmacist takes that into account as well.*	*And why do they say at the pharmacist's that you cannot take this drug in combination with naproxen, so why is it prescribed then? That's funny.*

Table 4.4. *Continued*

Source	Positive quotes (translated from Dutch)	Negative quotes (translated from Dutch)
Package insert	*Yes, I mean the one in the box, the patient information leaflet, it is very informative, but not the one the rheumatologist gave. That one is actually quite scanty.*	*No. and then I think, er …, yes, er… that is the same for all manufacturers of such medicines. They just want cover themselves against legal actions, so they put in anything, so I think, I am not interested in that, am I?*
		When you start reading about the side effects a lot and dig into them, then you'll have it. It might not be the case, but somewhere inside you there's this little voice saying: "Oh, this is what I feel, so it must be this or that, because it's in the leaflet."
Pharmaceutical company (only relevant for patients who decided to initiate bDMARDs[1, 2])	*Yes, you always get these very extensive leaflets [with the bDMARDs I like that, because they [the doctors] tell you all sorts of things and you don't remember most of it and er…, yes, you can go through a lot again in them.*	*Then you get your first batch of Humira with a mega large file containing all sorts of information: booklets and leaflets, plasticized and pens and covers to keep the stuff in. So I think, well, isn't that a bit exaggerated [...] I thought the rheumatologists booklet gave sufficient information.*

[1] *bDMARDs = biologic disease-modifying anti-rheumatic drugs*
[2] *In the Netherlands advertising of drugs aimed directly at patients is prohibited.*

Most participants initiated the proposed medication. We asked them what information they would have liked to have before deciding to use the medication. Some mentioned they did not lack anything, while others gave clear examples. Many stated that they had discussed only one treatment option and would have liked to know more about other available treatments: *"If they would have told me [about the wide range of options], then I would not have been so insecure. [...] about, well [...] what awaits me."* Knowledge about the wide range of possible treatments might have reduced concerns, because some feared "running out of options". Others wanted to compare treatment options with respect to potential benefits and risks and the consequences for daily life (e.g. influence on driving and alcohol consumption). As is illustrated by these quotes:

> *"What I missed? [...] If he prescribes me methotrexate […] maybe there are ten other types of drugs with fewer side effects. But I don't know the names."*

> *"The package insert contains millions of things the doctor didn't say. They said [...] three things that very often occur: nausea, fatigue and er… nothing else. And the package insert says [...] a million of other things. That you can be restless, that your sexual life may get worse, that sort of things [...]. I would have found that useful to*

know, to consider taking this drug or another, because who would like their sexual life to deteriorate?"

"After taking the drip you're not allowed to drive. That's difficult for me. Well, difficult. It takes a bit of planning and arranging. But you must consider it and I would really have liked to know that when considering the options."

"I like drinking a glass of red wine at night, but you can't have that for three days, can you. Well, I took just a small glass. But on the day before, the day of the treatment and the day after, you absolutely shouldn't. You really have to consider that. I'd like to have known before."

Most participants were satisfied about the provided instructions for use, especially for the injections. However, some would have liked more information, especially when multiple drugs were prescribed with a complex time schedule for administration (e.g. methotrexate, folic acid and a bDMARD). They would have liked to receive more help implementing it in their daily lives. Additionally, some would have liked more assistance determining the start of the cycle of administration, since side effects disturbed their daily routine. These (e.g. nausea, vomiting, malaise, fatigue, and chills) occur often shortly after administrating the drugs. The following quote illustrates this: *"Taking methotrexate [which is administered once a week] makes me very nauseous. Then we adjusted the scheme and considered the best time for taking it. I deliberately did not choose to take it before the weekend starts, because suppose you keep being troubled by the side effects - I'd not want that over the weekend. [...] I would have liked assistance with that sooner."*

DISCUSSION

This qualitative study explored what considerations patients had when deciding about DMARDs and what information patients need to participate in the decision-making process.

The NCF (143) was used to structure patients' beliefs and considerations when deciding about DMARDs. In our population necessity of DMARDs was often associated with direct relief of symptoms and sometimes with prevention of future joint damage. Yet, many patients considered DMARDs in general as aggressive and harmful, making some doubt the necessity. This tension between necessity and negative beliefs about DMARDs has been described before (120, 142, 148, 149). DMARDs have been described by patients as essential for managing arthritis, but

also as 'powerful', 'strong' and 'toxic' (142). Some reports show that patients are unwilling to take risks and are reluctant to change treatment even when disease activity is high (120, 148); fear of loss of control over arthritis and the fear of side effects are major concerns. However, other studies have shown that patients are willing to initiate aggressive treatment in full awareness of the potential side effects (137). It seems important to discuss this tension with patients. Rheumatologists should inform patients about the (long term) goals of the treatment, invite patients to share any concerns and address potential misunderstandings about DMARDs.

Some considerations in favour of initiating DMARDs exceeded the considerations about necessity. First, trust in physician and/or the health care system were mentioned as a strong motivator to initiate DMARDs. Sometimes even without reviewing the pros and cons. This is in line with other studies (102, 137, 139, 150). Although trust is essentially a good thing, it should not limit patients to review the pros and cons. If patients completely rely on the expertise of their rheumatologist, they may fail to recognise the potential value of their own input (38). Since rheumatologists' and patients' beliefs about treatment differ (120, 129-139), the input of patients seems important. Moreover, patients' adherence to treatment might increase if it matches their preferences. Second, benefits of the medicine compared to other drugs were often mentioned in favour of initiating a particular DMARD. These benefits may relate to a lower risk of side effects or to features that concern the potential impact on daily life, such as a less frequent and/or friendlier manner of administration. Future studies examining reasons for (not) starting certain drugs should include these relative benefits and the influence of trust in physician.

Patients' concerns were related to the perceived aggressive and harmful nature of the medicine, potential side effects, influence on fertility and pregnancy, time to benefit, combinations with other medicines and manner of administration. Most interestingly, patients also worried about future treatments. With regards to sDMARDs, patients worried about what kind of risks future treatments would bring, as they assumed that their rheumatologist would start with proposing the 'mildest' drug. With regards to bDMARDs patients often worried that this is the 'final option'. This may be due to the Dutch stepwise treatment approach, which states that therapy with a bDMARD can only be prescribed to patients with at least moderate disease activity and in whom treatment with at least two sDMARDs has failed. Finally, some patients were concerned if they would ever be able to stop using DMARDs once they started.

Previous studies showed that arthritis patients worry about the future, especially about the long term medication use and their prognosis, due to the unpredictable course and the varying disease activity (151-154). To our knowledge no previous

studies have reported patients' concerns about the availability and potential risks of future treatments. Therefore patients need to be informed about all current and future treatments and their risks. Furthermore, patients have concerns about long term medication use and fear dependency. Patients might benefit from information about recent developments regarding tapering DMARDs when persistent and long term remission has occurred (58). It may be helpful to have a personal long-term treatment decision plan that lists and explains current and future available treatments and also highlights the possibility to taper medication in case of persistent and long term remission.

Some experienced an emotional impact when considering DMARDs, because it made them realize they were seriously ill. Therefore they were reluctant to start taking the medication. It has been found that patients with chronic illness may experience an ongoing process of negotiation between resistance against medication and acceptance of their diagnosis (155). Then it is essential to ask what it means to initiate this treatment and to evaluate alternatives that correspond better with patients' values.

Most patients were satisfied with the information they obtained. However, some indicated that they had missed information on how medication could affect their daily lives or how to minimize the impact of the administration schedule on their daily lives. The need to stay in control and the need for practical information is in line with studies showing that patients prioritize treatment outcomes related to quality of life (129-134, 138). To increase patients' involvement in medical decision-making, it is essential to not only inform patients about clinical elements of the treatment, but also about the impact on lifestyle, control and comfort.

The NCF, mostly used to explain adherence, proved useful for understanding patients' considerations about initiating DMARDs. Patients mainly weigh the necessity and concerns when considering initiating DMARDs. However, concerns should not only be assessed in relation to clinical elements of the treatment, but also to the impact on lifestyle, control and comfort. Furthermore, NCF should be extended with elements as 'trust' and 'relative benefits'. Finally, attention should be paid to the emotional impact, such as the 'realizing being seriously ill'.

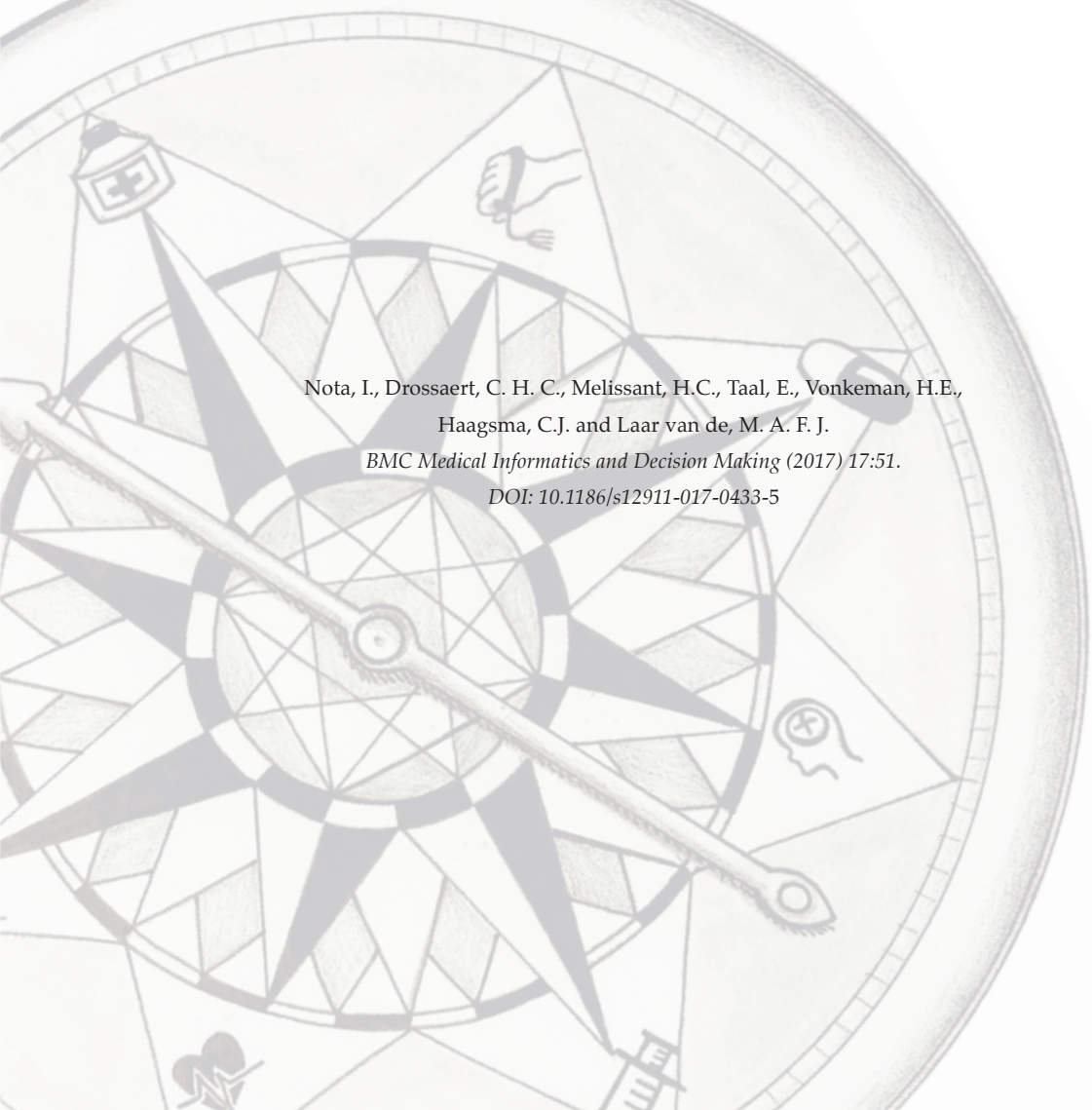
The results of this study were used to develop a PtDA which includes all available DMARDs. It provides the opportunity to compare DMARDs with regards to both clinical information as well as practical information with possible consequences for daily life. The PtDA has a flexible information system to fulfil the needs of most users without overwhelming others. Furthermore, it includes value clarification exercises that acknowledges questions and concerns, and supports patients to communicate their values with their rheumatologist.

Our results must be interpreted in view of the limitations of the study design. The participants in this study were recruited from two hospitals, thereby questioning the generalizability of the results. Our sample contained only a few patients who decided not to initiate the drugs. Whereas qualitative data has the advantage to gain new insights, it does not permit measurement of the impact of each of these factors on decision-making. More quantitative studies are needed to confirm our results.

From this study we conclude that patients should be informed about multiple treatments both current and future. Information should enable patients to compare treatments with regards to both clinical aspects as well as possible consequences for their daily lives.

Chapter 5

Development of a web-based patient decision aid for initiating disease modifying anti-rheumatic drugs using user-centred design methods



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ABSTRACT

A main element of patient-centred care, Patient Decision Aids (PtDAs) facilitate shared decision-making (SDM). A recent update of the International Patient Decision Aids Standards (IPDAS) emphasised patient involvement during PtDA development, but omitted a methodology for doing so. This article reports on the value of user-centred design methods for the development of a PtDA that aims to support inflammatory arthritis patients in their choice between disease modifying anti-rheumatic drugs (DMARDs). The IPDAS development process model in combination with user-centred design methods were applied. The process was overseen by an eight-member multidisciplinary steering group. Patients and health professionals were iteratively consulted. Qualitative in-depth interviews combined with rapid prototyping were conducted with patients to assess their needs for specific functionality, content and design of the PtDA. Group meetings with health professionals were organized to assess patients' needs and to determine how the PtDA should be integrated into individual patient pathways. The current literature was reviewed to determine the clinical evidence to include in the PtDA. To evaluate usability among patients, they were observed using the PtDA while thinking aloud and then interviewed.

The combination of patient interviews with rapid prototyping revealed that patients wanted to compare multiple DMARDs both for their clinical aspects and implications for daily life. Health professionals mainly wanted to refer patients to a reliable, easily adjustable source of information about DMARDs. A web-based PtDA was constructed consisting of four parts: 1) general information about SDM, inflammatory arthritis and DMARDs; 2) an application to compare particular DMARDs; 3) value clarification exercises; and 4) a printed summary of patients' notes, preferences, worries and questions that they could bring to discuss with their rheumatologist.

The study demonstrated that user-centred design methods can be of great value for the development of PtDAs. The early, iterative involvement of patients and health professionals was helpful in developing a novel user-friendly PtDA that allowed patients to choose between DMARDs. The PtDA fits the values of all stakeholders and easily integrates with the patient pathway and daily workflow of health professionals. This collaborative designed PtDA may improve SDM and patient participation in arthritis care.

INTRODUCTION

Shared decision-making (SDM) is one of the main activities of patient-centred care (156, 157). It involves the exchange of information and negotiation between the clinician and the patient to agree on the best way to medically proceed for the individual patient (17). Often the decision-making process is complex - especially when preference sensitive aspects are involved. Various interventions have been developed to facilitate SDM.

Patient Decision Aids (PtDAs) are intended to support patients in making specific and deliberated choices among healthcare options (50-52). In contrast to more general health education materials (e.g. information leaflets), PtDAs specifically state the decision being considered and stress the relevance of a SDM process (50-52). Furthermore, PtDAs provide information on all available treatment options and help patients clarify what matters to them regarding these treatment options (50-52). A systematic review recently revealed that, for many different decisions and conditions, PtDAs can improve patients' knowledge about options, risk perceptions, feelings of being informed and being certain about what matters to them (30). Furthermore, with the use of PtDAs, patients more often reach decisions that are consistent with their personal values (30). Finally, PtDAs can improve patient-doctor communication (30).

The International Patient Decision Aids Standards (IPDAS) Collaboration states that the development of PtDAs should be systematic and include consultations of patients and health professionals (50-52). However, many studies of PtDA development projects do not report on having involved patients during their development (53). In response to this omission, the IPDAS' evidence base has recently been updated to include a development process model that places more emphasis on patient involvement during PtDA development (52, 53). This process model provides a step-wise approach to careful and systematic development, evaluation and implementation of PtDAs. Although this new comprehensive model provides an overview of the entire development process, it does not provide guidance on *how* to best involve patients and health professionals nor which research methods to use. The authors, therefore, urged PtDA developers to complement the IPDAS development process model with other guidelines, such as a user-centred design approach (53). In a user-centred design, specific research methods are used to consult with potential users relatively early within the developmental timeframe (54, 55). This approach allows developers to adopt and implement user-centred input, resulting in the product more adequately fulfilling users' needs and, consequently, positively affecting user satisfaction (54, 55).

While patients with rheumatic diseases often face long-term treatment decisions, only a few studies have been reported on PtDAs for initiating disease modifying anti-rheumatic drug (DMARD) therapy (30, 66, 158, 159). DMARDs are the core element of the management of inflammatory arthritis in order to control the disease process and to relieve or reverse symptoms (58-60). DMARDs form two major classes: synthetic chemical compounds (sDMARDs) and biologic agents (bDMARDs). With regard to DMARDs, the decision-making process has become increasingly complex, as numerous therapeutic options are available. In addition, new treatment strategies are rapidly evolving, but without sufficient information on differential efficacy and safety (58-60). When weighing the options, elements to consider include treatment efficacy, approximate time-to-benefit, possible side effects, current and future risks, cost-effectiveness, route of administration and impact on daily life. Because this complex decision-making process concerns both clinical and preference-sensitive aspects, the choice of treatment needs to be based on a shared decision between the patient and rheumatologist.

The reported PtDAs for initiating DMARDs mainly focus on the decision whether to initiate one specific DMARD or a particular class of DMARDs (30, 66, 159). Yet, our previous study showed that patients would like to be informed about multiple specific DMARDs (160). In fact, previous research has shown that patients with a rheumatic disease are often less informed and less involved in decision-making than they would prefer (22, 26, 62-65, 128, 160). SDM barriers reported by patients include being unaware of having a choice, lack of medical knowledge and a power imbalance in the doctor-patient relationship (39, 160).

In order to fulfil this need of patients with rheumatic disease, the aim of this study was to develop a tool that could compare multiple specific DMARDs. This paper describes the development of a web-based PtDA with use of the IPDAS development process model and user-centred design methods. The PtDA is intended for inflammatory arthritis patients who face the decision whether to initiate DMARDs.

METHODS

To develop the PtDA, the IPDAS development process model was used in combination with user-centred design methods. As illustrated in Figure 5.1, the IPDAS process model provides a careful and systematic step-wise approach to develop PtDAs that are user-tested and open to scrutiny (53). The current study focused on the first four steps of the IPDAS process model: 'scope,' 'design,' 'prototype development' and 'alpha testing,' which are further described below.

The process was overseen by a multidisciplinary steering group, consisting of three rheumatologists (including one epidemiologist), a rheumatology specialist nurse, three experts in SDM and health psychology, and an experienced web designer.

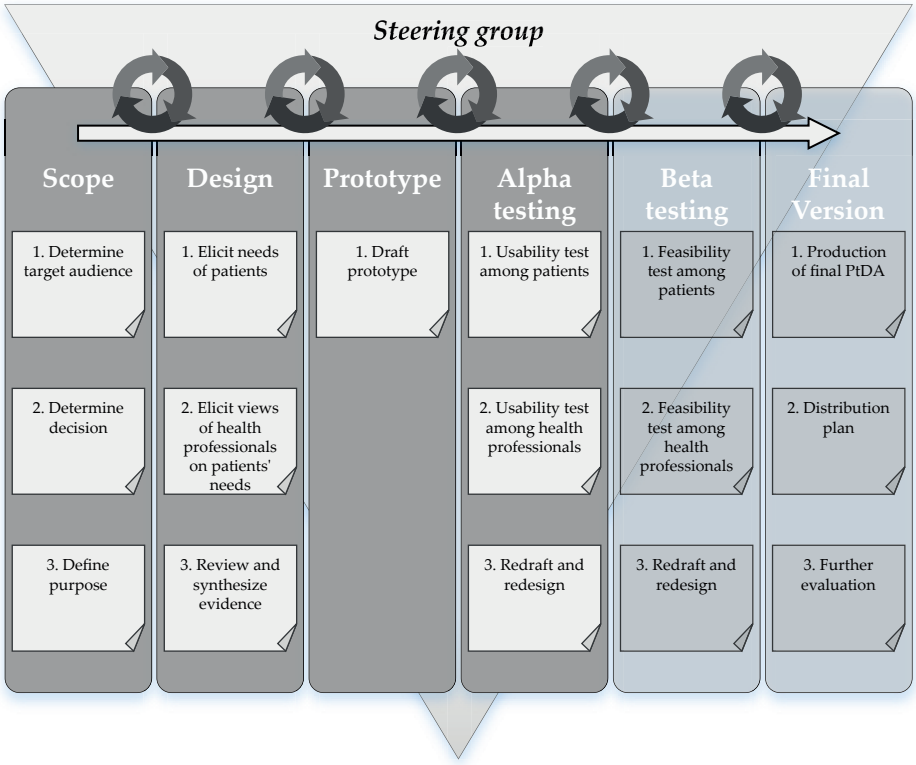


Figure 5.1. *The IPDAS Development Process Model (53)*

Scope

Because our previous studies showed a need for patient participation and information about multiple specific DMARDs (128, 160), the steering group decided to develop a PtDA for patients diagnosed with Rheumatoid Arthritis (RA), Ankylosing Spondylitis (AS) or Psoriatic Arthritis (PsA) who face the decision to initiate (a different) DMARD.

Design

Needs assessment among patients (Design 1)

In-depth semi-structured face-to-face interviews were conducted (by IN) to assess patients' needs for functionalities, content and design of the PtDA. We recruited 32 patients diagnosed with RA, AS or PsA, who recently (< 1 month ago) consulted their rheumatologist and discussed initiating a (different) DMARD.

The interviews lasted between 45 and 120 minutes, were recorded and consisted of two parts. The first part of the interview explored what considerations, worries and questions patients had when deciding about DMARDs and what information patients needed in order to participate in the decision-making process. These findings helped determine what information the PtDA needed to provide. This part of the interview has been reported elsewhere (161).

The second part of the interview focused on patients' needs regarding the functionalities, content and design of a PtDA for initiating DMARD therapy. To introduce the concept of a PtDA, we gave a general description. As a picture is worth a thousand words, rapid prototyping (162) was conducted to assess the usefulness of several potential PtDA features. A prototype was drafted with use of the software application Evolus Pencil 1.2 (163). Previous developed PtDAs (e.g. (164, 165)) and the stepwise model for SDM developed by Elwyn and others (43) were used as an inspiration for the steps to guide patients through the decision-making process (i.e. acknowledging a decision needs to be made, gaining knowledge about options, preference eliciting and preparation for the decision talk).

The prototype included an innovative application to compare medications. This application was a direct response to findings from previous studies in which patients' expressed the need for information about multiple options (160) and was inspired by commercial web-applications that allow consumers to compare various product features. The prototype was printed on paper and appears in Additional File 1 (in Dutch). Each page in Additional File 1 is a copy of the 10 prototype screens in the same order in which they would appear online.

The interviewer and patients walked through the paper prototype and discussed usability issues and additional needs (regarding functionalities, content as well as design). Patients' remarks were written on the paper prototype and later analysed alongside the analysis of the audio recordings.

Interviews were transcribed verbatim and analysed using Atlas.ti 7.1 (166), a qualitative analysis software application which allows researchers to overview the codes, link statements and visualize connections between themes (145). Furthermore, this software can also integrate pictures - in this case, the paper prototype with written remarks of the participants. The analysts (IN, CHCD and HCM) mutually

independently analysed the data using the principle of constant comparison (146) and an iterative process of deductive and inductive analysis. First, all quotes were (deductively) categorized into needs for functionalities, content and design. These quotes were then further analysed using a process of open coding (inductive analysis), followed by axial and selective coding (deductive analysis) (146, 147). During this process the analysts preserved the voice of the patients. After each phase, the individual findings were compared and analysed until consensus was reached. Finally, in close collaboration with the web designer, the analysts translated the needs (in the voice of the patients) into a list with requirements.

Needs assessment among health professionals (Design 2)

In accordance with user-centred design theories, all stakeholders need to be consulted during development (54, 55). To comply with this requirement, all rheumatologists (n=11), specialized nurses (n=3) and rheumatology nurses (n=4) of the two participating hospitals were invited to participate in a semi-structured group interview. They were asked to give their expert opinion on functionalities, content, design and distribution of a PtDA for initiating DMARD therapy.

Firstly, health professionals were asked to indicate what information patients needed to know before being able to make a decision and how this information should be presented. Then, the paper prototype was presented and participants were asked to express their expert opinion regarding its functionalities, content and design. After that, the results of the needs assessment among patients were presented, and participating health professionals were asked to reflect on them.

Secondly, to be able to determine how to distribute the PtDA and how to best integrate the PtDA into clinical practice, the health professionals were asked to outline the patient pathway and to discuss when and how to refer patients to the PtDA (setting and timing). The patient pathway was outlined by following the steps of regular patients from their appointment to discuss initiating DMARDs with the rheumatologist to taking their first DMARD dosage. Patient pathways were outlined for newly diagnosed patients as well as for patients with a longer history of RA, AP or PsA.

During the meeting, we explored the range of opinions and aimed for consensus on the health professionals' needs for the PtDA. Notes were taken by two members of the steering group (IN and HCM). Similar to the analysis of the needs assessment among patients, the analysts (IN and HCM) mutually independently analysed the notes using the principle of constant comparison (146) and a combination of deductive and inductive analysis. The notes were first classified into the needs for functionalities, content, design and distribution/implementation (deductive analysis). Then, the notes were inductively analysed with a process of open coding,

which resulted in the categories arising from the data. The coding ended with deductive analyses (axial coding and selective coding) (146, 147). After each phase, the findings of the analysts were compared and further analysed and discussed until consensus was reached. Then, the needs (in the voice of the health professionals) were translated into a list of PtDA requirements for content, design and distribution/ implementation. To confirm whether we translated the needs correctly, the list was sent by email to the health professionals, all subsequently agreed on the listed items.

Evidence review (Design 3)

The background information and clinical evidence included in the PtDA were based on the needs assessments among patients and health professionals, availability and quality of the evidence. We reviewed current international guidelines on the management of RA, AS and PsA (58-60) which provide recommendations on general aspects of treatment, mostly on group drug levels (e.g. sDMARDs and bDMARDs). Furthermore, we reviewed medication information leaflets from the participating hospitals, the local pharmacists, the Dutch Arthritis Association (167), the "Farmacotherapeutisch Kompas" (a Dutch database that encompasses independent information on all drugs available in The Netherlands) (168), and the information pharmaceutical companies provide to health professionals and patients.

Working prototype

Based upon the needs assessments with the various stakeholders (patients, health professionals and the steering group), the IPDAS criteria (51, 52) and the evidence review, the paper prototype of the PtDA was redrafted and redesigned. First, the steering group developed a plan for integrating the PtDA into the patient pathway, then the PtDA was redesigned and programmed into a working prototype. Additional File 2 shows the redrafting process of the PtDA screen that enabled patients to compare DMARDs.

Alpha testing

Usability test among patients (Alpha testing 1)

A usability study was conducted with the working prototype of the PtDA. Patients with RA, PsA, or SA were recruited from the "Patient Research Partners"-Panel of the Arthritis Centre Twente and via the two participating hospitals.

Data were collected by observing the patients' usage of the PtDA and semi-structured interviews conducted during and after usage by two of the authors (IN and HCM). First, participants were asked about their demographics, their perceived health status on a scale ranging from 1 indicating 'poor health' to 10 for 'excellent

health,' their history with regard to decision-making about DMARDs and their experiences with online health information. They were then presented a scenario describing a possible decision (closely matching their history with decision-making about DMARDs) and a brief description of the rheumatologist's consultation that referred them to the PtDA. They were subsequently given a referral card containing the internet address and the treatment options suggested by the rheumatologist and were assigned to visit the PtDA website. While using the PtDA, participants were asked to think aloud (169, 170). When visiting the homepage, they were interrupted briefly and questioned about their expectations of the website. After observation of their free usage of the PtDA, the semi-structured interview started which included questions about *perceived usefulness, perceived ease of use, attitude towards using* and *intention to use*. These elements are based on the Technology Acceptance Model (TAM) (171, 172). The sessions lasted between 45 and 97 minutes.

We used Morae 3.3.0 (173), a software application for usability testing, to record the performance task and the interview. This programme records a video of the user (including sound), screen activity and system events (including mouse clicks, web page changes and onscreen text).

After completing all sessions, two analysts (IN and HCM) selected relevant written remarks of participants, watched the videos of the usage and interviews and made notes on their observations.. The analysis mainly focused on the correspondence between the structure of the website and the cognitive steps the users followed. The notes were linked to specific pages of the website (e.g. the homepage or the page enabling comparison of medication) and to the topic of discussion (e.g. structure, navigation, content, format, and colour). This resulted in a list of positive remarks as well as of points for generally improving the PtDA and each screen in particular.

Usability test among health professionals (Alpha testing 2)

To evaluate usability from the health professionals' perspective, all rheumatologists (n=11), specialized nurses (n=3) and rheumatology nurses (n=4) of the two participating hospitals were invited to participate in another semi-structured group interview. The aim of this usability study was focused on acceptability and compatibility of the PtDA and the current process of medical decision-making. This approach was taken since health professionals would not directly be using the website, but instead referring patients to the website. Health professionals would then need to interact with patients once they had used the website.

During the meeting, the working prototype of the PtDA was presented (by IN) and the health professionals were asked to give their opinion on the content and design of every screen. Notes were taken by a member of the steering group (HCM). After the presentation, the group was asked to test the working prototype

individually, write down their remarks and reflect on *perceived usefulness, perceived ease of use, intention to use* and *compatibility* with the current process of medical decision-making. These elements are based on the Technology Acceptance Model (TAM) (171, 172).

Two analysts (IN and HCM) mutually independently read all the notes and, using inductive analysis, coded relevant remarks of the participants. The analysis mainly focused on compatibility with the current process of medical decision-making and intention to use. This resulted in a list of positive remarks as well as elements that could be improved.

Redraft and redesign of PtDA (Alpha testing 3)

Based on the results of the usability tests, an iterative draft-review-revise process by the steering group was conducted until the PtDA reached content and format 'saturation' (i.e. all points for improvement were accounted for). Overall, no major adjustments were conducted, and hence the steering group decided to forego another usability study.

RESULTS

Design

Needs assessment among patients (Design 1)

In total 26 women and 6 men participated, with an average age of 54 years. Most participants (62%) had completed 12–16 years of education and were currently employed (56%). Some participants (n=5) had discussed initiating their first DMARD with their rheumatologist. Others (n=27) were already using sDMARDs or bDMARDS and had discussed changing to another DMARD with their rheumatologist.

The first part of the interview aimed at deepening our understanding of patients' considerations when deciding about DMARDs and what information patients need to participate in the decision-making process. The results of this part of the interview have been published elsewhere (161); but briefly, patients felt the need for a complete overview of treatment options. Results also showed that before deciding about DMARDs, arthritis patients wanted information with regard to both clinical features (e.g. aim and working mechanism, time-to-benefit, manner of administration, potential side effects and risks, influence on fertility and pregnancy) as well as possible consequences for their daily lives (e.g. restrictions on driving a vehicle and alcohol consumption and how to fit the treatment schedule into their daily lives).

Finally, patients mentioned many concerns and questions that could be incorporated into the value clarification exercises (i.e. the lists of common worries and questions).

The second part of the interview introduced the concept of a PtDA. In general, participants were positive about implementing a PtDA for choosing between DMARDs. In line with our previous research (128, 160), a few participants did not feel the need for a PtDA, because they did not want to participate in medical decision-making or because they felt the current information was sufficient. For example: *"I do find that easy, to just leave [the decisions] behind at the doctor's"* and *"The information given by the rheumatologist is good enough for me."* Most participants liked the idea because it would be a reliable source of information, to help them prepare for the decision-making consultation. This is illustrated by the following quotations:

> *"This way you can keep [all the information] together, without having to look for it. And when your doctor refers you to it, well, then it has to be trustworthy."*

> *"In this way you can really prepare yourself for the decision-making consultation, having some idea beforehand of what to expect. If you go to the rheumatologist without having the slightest notion, then, after some time, there still are questions you might not have asked."*

Furthermore, a PtDA would fit patients' need to be informed about multiple treatment options for their present situation as well as for the future. To quote one participant:

> *"When you consult your doctor, it is only a ten-minute-conversation, ending with a prescription that you think is alright – and when it actually helps, it is alright indeed. Yet there are perhaps many other possibilities, with less side-effects or smaller doses …, and the doctor will certainly not explain all of them ... This tool enables us to have insights into all the possibilities, to work in a structural way and say, 'All right, this is where you are, and from here you can go either in this or in that direction … And when this does not work, you can go in that other direction."*

Reviewing the paper prototype, most participants liked that the PtDA provided insights into all available medication options. However, some had their doubts. In The Netherlands, therapy with a biologic DMARD is reimbursed for patients with at least moderate disease activity for whom treatment with at least two synthetic DMARDs has failed. Some participants felt that it could be frustrating to receive information about medication for which they were not (yet) eligible: *"It is frustrating*

indeed when something is not available, because [the insurance company] considers it to be too expensive." Some of them suggested to solve this by tailoring the appropriate options to the individual patient.

Almost all participants liked the opportunity to compare treatment options for different features in a structured manner prior to the decision, as illustrated by the following quotation:

> *"In this way you get a clearer idea about different kinds of medicines. Normally you receive only information about the drug you start with, like, 'This is what you have to take, so there you go'. This does not leave you with a clear idea about whether another medicine in the same category is perhaps more suitable. I think that this other approach does help to sort it all out a little better."*

Participants also appreciated that the prototype provided, besides clinical benefits and risks, practical information with possible implications for patients' daily lives. They suggested a variety of categories that should be added in the side-by-side comparison, including: restrictions for nutrition and alcohol, storage instructions, influence on daily routine and guidelines for traveling. Most participants did not value the personal stories of peers that were included for each DMARD because *"every patient is different," "they will probably be actors"* or *"that is not reliable information and does not belong on such a website."*

All participants liked that the information was provided in portions; the paper prototype suggested that supplemental information could be obtained via links that would unfold elements of the webpage. Some of the information in the paper prototype was provided using pictograms, pictures or videos. Although some pictograms needed clarification, many respondents asked if we could add more pictograms and decrease the amount of text. To further reduce the amount of text, some participants suggested tailoring the content of the information (e.g. risks) to the individual patient.

The pictures and videos illustrating the administration of the treatments were appreciated by most respondents. Such illustrations can decrease uncertainty and anxiety, especially when the medication requires the administration of injections or infusions. To quote two participants:

> *"It really was a relief to see that injection needle, which was quite different from what I expected. So I believe that when people see such short instructive films, they can be better prepared for [their treatment]."*

"If you watch [such a short film], you know beforehand what you are getting yourself into."

With regard to the proposed value clarification exercises, most thought this would be helpful to their decision-making process. Some participants mentioned that thinking about their preferences would support them in participating in decision-making. One participant hypothesized that *"this may increase patients' feelings of being in control"*. Many participants also appreciated the lists of worries and questions. They felt that the lists acknowledged that it is normal to have these worries and questions and thought that it would support them to express these feelings and questions during their next consultation with the rheumatologist. For example, one participant said: *"The question I had is one of those addressed here [on the list], so it doesn't seem to be such a strange question, but one I can ask without any fear."*

Participants also expressed that they would like to bring the summary containing their notes, preferences, worries and questions to their consultation as a reminder and to increase their confidence in their ability to participate in medical decision-making. To quote one participant: *"Everything is really more focused, like, 'I see you have prepared yourself and that you still have the following questions, so let us start with those'."*

Most participants expressed that they intended to use the PtDA, especially if it was available at home which would allow them to use it at their ease. A few mentioned that they would like to use the PtDA, but were afraid it would take too much time and effort to go through all the steps. Finally, a couple of participants did not like the PtDA to be computer-based, because they felt that they lacked sufficient computer skills or did not have access to a computer/internet.

The needs elicited from this study were translated into requirements of the PtDA, as presented in Box 5.1.

Needs assessment among health professionals (Design 2)

Ten rheumatologists, two specialized nurses and two rheumatology nurses were present at the group meeting. Most health professionals were eager to implement a PtDA into their practice, considering it an innovative way to inform patients. Their primary reason for adoption was to be able to refer patients to a website with reliable health information. However, some health professionals were sceptical at first. They thought the PtDA would be time-consuming without adding value to the current information leaflets from the hospital. However, when they learned (from the results of the needs assessment among patients) that many patients desired more information than they currently received and that they would like to be able to compare DMARD

options with regard to clinical elements as well as possible consequences for their daily lives, the health professionals understood the added value of a PtDA.

The health professionals discussed when and how to refer patients to the PtDA by outlining patients' pathways. All of the health professionals worried that patients might become overwhelmed when informed about all available DMARDs. Therefore, all agreed that patients should be referred to the PtDA only after having consulted their rheumatologist or specialized nurse who would first provide them with a personal recommendation for appropriate options for medication. The health professionals disagreed on whether patients should be able to see all options for medication or only the ones that were personally recommended. After some discussion, consensus was reached; patients should receive a clear personal recommendation in writing (preferably digital), but should be free to also read information about other DMARD options in the PtDA. The health professionals were asked if they had specific requirements for the PtDA. Some mentioned they would like to read patients' preferences, worries and questions before the encounter, but others felt that this would be a violation of privacy and that it was the patients' right to decide what to share with their health professional. Another requirement of the health professionals was to be able to easily add new drugs to the PtDA because of the rapid development of new DMARDs. Box 5.2 presents an overview of the requirements based on the needs assessment among health professionals.

With regard to the needs of the steering group, one additional item was included in the list of PtDA's requirements. For the purpose of evaluating the usage of the PtDA, the steering group wanted the website of the PtDA to log anonymous user data (navigation and input).

Evidence review (Design 3)

When reviewing the clinical evidence, we discovered there was insufficient evidence on differential efficacy and safety. Therefore, it was difficult to use the available clinical information for detailed comparisons of DMARDs. Not only was information unequally available for each drug, but it was also conflicting. In such cases we based the information in our PtDA on consensus between the rheumatologists of the participating hospitals (n=11).

Two informal group meetings with all rheumatologists, a specialized nurse and three members of the steering group (IN, HV and ML) were organized to discuss the clinical evidence. Taking into account the information needs of the patients, the rheumatologists decided, in general, what information: 1) must be disclosed to all patients, 2) should be provided as supplemental information for patients who desire additional information, and 3) need not be included at all. With this information, we

were able to develop a flexible information system which would fulfil the needs of most users without overwhelming others. During the second meeting, the unclear and conflicting information was presented and discussed until consensus was reached among all rheumatologists. The final texts were checked by five rheumatologists.

Working prototype

Based upon the results of the previous studies and the evidence review, we developed a working prototype and a plan for integration of the PtDA into the patient pathway (illustrated in Figure 5.2). According to this plan, the patient and rheumatologist have an initial conversation about starting a (different) DMARD. During this conversation, the rheumatologist refers the patient to the web-based PtDA along with a referral card. On this referral card, the rheumatologist indicates the DMARDs appropriate for the patient at this specific moment. The referral card states the internet address of the PtDA, and the patient is encouraged to access the PtDA at home.

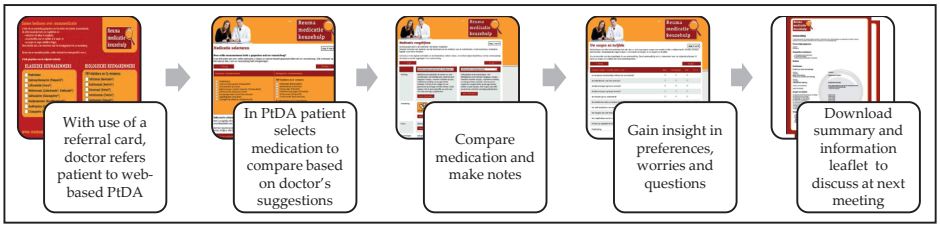


Figure 5.2. *Process of the Patient Decision Aid*

The working prototype of the website consisted of two components: general information and the PtDA itself. The component with general information addressed SDM, emphasized the importance of the patient's role in medical decision-making, and provided general information about inflammatory arthritis and DMARDs. The component with the PtDA (see Figure 5.2) consisted of an application to compare selected DMARDs side-by-side. Elements that were compared included: target and working mechanism of the medication, manner of administration, approximate time-to-benefit, risks of side effects, follow-up process, combination with other drugs, fertility/pregnancy, consequences of continuing or stopping the DMARD, drug marketing history, restrictions and warnings for nutrition and alcohol consumption, and impact on daily life (e.g. storage, daily routine, traveling). In order to fulfil the high need for information of most users while not overwhelming others, the PtDA has a flexible information system - supplemental information about the medication can be obtained via links that unfold certain elements. Furthermore, the working prototype included a digital notebook and value clarification exercises to gain insight

into one's own preferences, worries and questions. The notes and exercise responses were compiled in a summary. This summary could be downloaded, printed and later used during the patient's next consultation with the rheumatologist.

Box 5.1: *Requirements of PtDA based on patients' needs*

Functional requirements

- PtDA encourages patients to participate in medical decision-making.
- PtDA provides overview of (all) available options for medication.
- PtDA provides the opportunity to compare options for medication.
- PtDA supports patients to gain insight into their preferences, worries and questions regarding medication.
- PtDA urges patients to express their preferences, worries and questions about initiating medication.

Requirements for content, design and distribution

- The PtDA includes information about the decision-making process, SDM and the importance of the role of the patient.
- Available options for medication are listed to provide an overview.
- Medication can be compared for:
  - Clinical aspects: aim and working mechanism of medication, manner of administration, time to benefit, risks for side effects, follow-up process, combination with other drugs, influence on fertility/pregnancy, continuing or stopping medication and history of medication.
  - Possible implications for daily life: restrictions for nutrition and alcohol, storage instructions, influence on daily routine and guidelines for traveling.
- Tailoring:
  - Appropriate treatment options are tailored to individual patient.
  - Content of information is tailored to individual patient based on gender.
  - Content of information is tailored to individual patient based on desire to have children.
  - Content of information is tailored to individual patient based on risk profile.*
- Information in PtDA is easy to read:
  - Pictograms are used as much as possible to decrease amount of text.
  - Pictures and videos are used to provide insight into administration of medication.
  - Information is written in plain language with links to definitions.
  - Information is provided in portions; amount and complexity of information can be adapted to individual needs.
- PtDA provides the opportunity to give value to specific treatment options and features of the specific treatment options.
- PtDA includes exercises to gain insight into patients' preferences, worries and questions.
- PtDA provides a summary of patients' notes, preferences, worries and questions which can be saved and printed.
- Can be used at home (i.e. outside the hospital).
- Does not take more than 30 minutes to complete (on average).

** Not realized in final PtDA*

PtDA = Patient Decision Aid; SDM = Shared Decision-Making

Box 5.2: *Requirements of PtDA based on health professionals' needs*

Functional requirements

- PtDA provides health information on inflammatory arthritis and DMARDs.
- PtDA provides health professionals insight into patients' preferences, worries and questions about DMARDs.

Requirements for content, design and distribution

- The provided information is based on the highest level of evidence available.
- The PtDA is an extension of existing information patients receive (added value).
- The PtDA meets the criteria set by laws regulating patient education and informed consent.
- Referral to the PtDA is accompanied by a clear personal recommendation for appropriate options for medication in writing

  … preferably digital.*
- Information about DMARDs other than those personally recommended by the rheumatologist to the patient is freely available.
- The PtDA provides patients the opportunity to share their preferences, worries and questions about DMARDs with their health professionals.

  … by digitally sending these insights to the health professional before the consultation.*
- The PtDA easily integrates with the existing daily work process / patient pathway.
- The PtDA is not time-consuming for health professionals.
- The PtDA is easily adjustable for newly developed DMARDs.

** Not realized in final PtDA*
PtDA = Patient Decision Aid; DMARD = Disease Modifying Anti-Rheumatic Drug

Some wished-for attributes of the PtDA were not realized due to their technical complexity, time and financial limitations, and/or privacy issues (see Boxes 5.1 and 5.2). The steering group weighed the needs of both patients and health professionals. For example, many patients expressed the need to have an overview of all available DMARDs, while others only want information about medication that is personally recommended to them. In order to not overwhelm patients, health professionals concluded that patients should receive a clear personal treatment recommendation in writing -preferably digital-, but should be free to also read information about other DMARD options in the PtDA. A solution would be to tailor the PtDA to the information needs of each patient. This would require patients to register online before accessing the PtDA. However, this solution raises privacy concerns and would withhold some patients to use the PtDA. Therefore the steering group decided to use the referral card – It provides patients with an overview of treatment options combined with a clear recommendation from the rheumatologist. When patients access the web-based PtDA, they are asked to select the medication that was recommended by their rheumatologists, but the information on other available medication options is freely accessible.

Tailoring the information to the patients individual risk profile proved too time consuming and out of budget. Also, sending the summary of the patient's notes,

preferences, worries and questions directly to the health professional was not implemented because of privacy concerns.

Alpha Testing

Usability test among patients (Alpha testing 1)

A total of 5 women and 5 men participated in the usability study, with an average age of 55 years (range 31–85 years). The participants were heterogeneous with regard to their educational status and current employment (1 participant finished primary education, 4 achieved intermediate education and 5 achieved higher education; 6 participants were employed). They were also heterogeneous regarding their disease-related internet use. On average, the participants each spent 11 hours per week online (range 2–35 hours per week). All had at least searched once online for information about arthritis, treatments and health services. Most (n=8) had ordered their medication online, but only a few (n=2) had used interactive health websites (e.g. online consultation).

All participants were diagnosed with RA. At the moment of the usability test, 4 participants experienced a poor health status (score <5). Some respondents (n=4) had been diagnosed in the past year and had only discussed initiating DMARDs once or twice (only sDMARDs). Others (n=6) had a longer disease duration and had previously decided to initiate sDMARDs as well as bDMARDs.

When visiting the homepage, most participants mentioned the working prototype had a clear structure and professional appearance. When asked about their expectations, some expected the PtDA to result in a treatment recommendation, but most correctly expected the PtDA to give them the opportunity to compare DMARDs.

During our observations of patients' free usage and walkthrough of the working prototype, we discovered some significant barriers to usability. Firstly, we discovered that the referral card was not easy to use; the card was printed on both sides, one side for the sDMARDs and the other for the bDMARDs. Not all participants noticed this and, therefore, did not have an overview of all DMARDs supposedly ticked by the rheumatologist. Secondly, we discovered early in the study (after three observations) that participants had difficulties navigating the screen that allowed them to compare DMARDs. All three participants did not know what to do, stopped and asked for help. We asked the three participants for tips to improve navigation, and, based on their suggestions, we made a paper prototype of the revised screen. We added this paper prototype during the usability test with subsequent participants (n=7). Most of them liked the revised page and thought it would be easier to use. Thirdly, we observed that after completing the comparison of DMARDs, some participants

felt they finished the PtDA. They were not aware that more steps followed. This was because the button for the next step was not prominently visible. We asked participants to suggest a better location for the button and how to highlight the next steps of the PtDA. Finally, a few small programming errors were found.

The interviews largely confirmed the results of the observations; overall, participants mentioned that the working prototype was easy to use and the information easy to read. The PtDA was perceived as useful with regard to comparing DMARDs side-by-side; gaining insights into preferences, worries and questions; and having all the information on one reliable location, all of which might support their decision-making process, as illustrated by the following quotations:

> *"The tool to compare medication is very useful – every aspect [of the medication] can be compared side by side."*

> *"They <the value clarification exercises> help you prepare <for the next consultation>. I think I would be able to ask better questions and I would feel less insecure."*
> *"All information is in one place, a reliable source, so you do not have to search anymore."*

> *"This helps me to think systematically about my options."*

Furthermore, many participants appreciated the pictures and videos visualizing the manner of administrating the medications. Most suggestions were directed at clarifying the PtDA steps and improving the navigation on the screen that enabled the comparison of DMARDs. Minor remarks included clarifying some specific content and decreasing the amount of text. With regard to intention to use, most participants said that they would use it, some would not, but all would recommend it to others.

Usability test among health professionals (Alpha testing 2)

Nine rheumatologists, one specialized nurse and two rheumatology nurses attended the group meeting. Overall, all health professionals appreciated the clear structure of the website and the clarity of the text. Similar to the results of the usability test among patients, some health professionals perceived the navigation on the screen that enabled comparison of DMARDs to be rather complex. Other screens were perceived as easy to use. Most health professionals believed that the PtDA would be very useful, especially to gain insight into patients' preferences, worries and questions and to discuss these topics with them. A few remained sceptical about the added value (see our previous discussion of the needs assessment among health

professionals), but were willing to try using it. All health professionals thought the PtDA would be highly compatible with the regular process of medical decision-making and easily implemented.

Redraft and redesign of PtDA (Alpha testing 3)

Based on the results of the usability tests, the steering group discussed which adjustments needed to be implemented. The referral card was adjusted to print on one side only, allowing a clear overview of all treatment options and the personal recommendation of the rheumatologist for appropriate medication. To highlight the steps of the PtDA, we altered some of the text on the website and added an instructional video (see Additional File 4). The navigation on the screen that enabled comparison of DMARDs was adjusted according to the recommendations of the participants. We did not increase the amount of medications to compare side by side because the font would then become too little to read. Some buttons were relocated, and several programming errors were fixed. Overall, no major adjustments were necessary. Additional File 2 shows how the page that enabled comparison of DMARDs was redrafted from paper prototype to working prototype and the final version. The card rheumatologists use to refer patients to the PtDA can be found in Additional File 3. Additional File 4 contains the instructional video of the PtDA and provides a clear representation of the PtDA and its functionalities.

DISCUSSION

We have described in detail the development of a PtDA for patients with inflammatory arthritis that helps them to choose between DMARDs. This PtDA was developed using the IPDAS development process model (53) and user-centred design methods (54, 55). Based upon the needs assessments of both patients and health professionals, we constructed a web-based PtDA consisting of the following parts: 1) general information about SDM, inflammatory arthritis and DMARDs; 2) an application to compare specific DMARDs attributes; 3) exercises to gain insight into the patient's preferences, worries and questions; and 4) a printed summary of the patient's notes, preferences, worries and questions to be discussed with the rheumatologist at the next consultation. The results of the alpha tests revealed that the developed PtDA largely satisfied the needs of both patients and health professionals and thus has the potential of being a valuable tool for patients who need to choose between DMARDs.

The overall process of development was satisfactory. The IPDAS development process model is relatively new and has yet to be substantially tested. Nevertheless, this process model proved to be systematic and helpful to our iterative development

of the PtDA as well as compatible with user-centred design methods. In addition, the user-centred design methods proved to be helpful in gaining valuable insights into different stakeholders' needs with regard to the PtDAs content and design and how it should be integrated into daily practice.

Firstly, rapid prototyping (i.e. the use of paper prototypes) proved to be of additional value to the needs assessment interviews. Patients (but also clinicians) often have difficulty conceptualizing what a PtDA is and how it should look and function, which might limit them in expressing their needs. With the use of rapid prototyping, it was easier for users to express their wishes and needs and to give critical input. For this reason, we recommend using rapid prototyping in the development process of future PtDAs.

Secondly, according to the IPDAS development process model, health professionals' perceptions of patients' needs for information and decision support should be assessed. We recommend conducting this step after having elucidated the patients' needs. In our study we intentionally conducted the study first among patients and presented the results of this study during the session with the health professionals. By doing so, the results of the needs assessments among patients were largely confirmed. But perhaps more essentially, this procedure proved to be effective in creating support among more sceptical health professionals for the development of the PtDA. Health professionals who initially questioned the added value of a PtDA had less misgivings and were more willing to use it.

Thirdly, we recommend not only asking health professionals about their perception of patients' needs, but also asking them about their own needs and thoughts on implementing a PtDA into their practice. Their practical and expert knowledge on the decision-making process can be of great value for the integration of a PtDA into the patient pathway and daily workflow of health professionals, and consequently enhance the adoption and implementation of the PtDA. The adoption and implementation of PtDAs using a referral model (i.e. health professionals inviting eligible patients to use the PtDA) is often challenged by indifference on the part of health professionals (31). This indifference may stem from a lack of confidence in the content of the PtDAs and concerns about disruption of established workflows (31). Our PtDA is still being successfully used after conclusion of the project (after beta testing), and newly developed DMARDs have since been added by the health professionals. This indicates that the iterative and extensive involvement of health professionals and the acknowledgement of their needs for the PtDA were important in creating ownership.

Finally, as the scope of the internet grows, PtDAs will be more and more computerized and web-based. These formats may offer many opportunities, not only for rapid adjustments of the PtDAs, but also for studying usage and usage

behaviour in detail. For instance, the amount of log-ins, page-views, and time spent on the PtDA could be logged, but also patterns of usage (e.g. How do users navigate? Which elements and combination of elements are often used? When do users drop-out?) and users' input (e.g. selected preferences, worries and questions) (174, 175). This information could be used to gain more insight into users' (evolving) needs and improve the PtDAs usage, usability and impact. Therefore, we recommend adding researchers to the stakeholders of web-based PtDAs and advising researchers to include logging anonymous user data as a requirement for the PtDA.

Compared to most previously reported PtDAs, the PtDA in this study encompasses many treatment options (176). Although patients have the right to be informed about all treatment options (13) and one of the quality domains of the IPDAS is to provide all options to patients (51, 52), we had a valuable discussion with the health professionals about whether to give patients access to all available medication options. Our previous studies (160, 161) showed that patients not only worry about the side effects and potential risks of their current or proposed treatment, but also had significant worries about the risks of future treatments and about 'running out of options,' should the proposed medication fail to work. To decrease this uncertainty, patients expressed a need to have an overview of all available options, for the time being as well as for the future. However, patients will most certainly be overwhelmed by all the different options and their pros and cons. To guide patients through this plethora of options, we chose to provide them in writing (the referral card) a clear personal recommendation of their most appropriate medication options. To respect their needs and rights, we also provided an overview of all other medication options and gave patients access to this information as well. This format may also be suitable for PtDAs that address multiple treatment options for other conditions, such as asthma or diabetes.

Previously developed PtDAs for initiating DMARDs mainly differ in the amount of treatment options that are included and how it is integrated in the patient pathway and distributed to patients. Most of these PtDAs focus on the decision whether to initiate one specific DMARD or a particular class of DMARDs, are to be used outside the clinical encounter and the plan for distribution is often unclear (30, 66, 159). Only one other PtDA includes all DMARD options and a clear plan for integration in daily clinical care. It consists of a card deck to be used during the medical encounter and is developed for patients with limited health literacy or limited English language proficiency (158).

One limitation of the currently developed PtDA is that it does not present outcome probabilities. This is due to the large number of treatment options included and the lack of evidence on differential efficacy and safety. Presenting outcome probabilities is a quality domain of the IPDAS (51, 52). Not presenting outcome probabilities limits the comparison of treatment options. However, the PtDA does present the negative and positive features in equal detail and a structured manner of the available treatment options for clinical and practical elements as well as the possible implications for daily life. This way of presenting outcomes was regarded as useful by the patients.

Furthermore, it should be noted that some wished-for attributes of the PtDA were not realized due to their technical complexity, limited time and finances, and privacy issues. A few (elderly) patients stated they did not want a computerized version of the PtDA because they feared that they lacked sufficient computer skills or did not have access to a computer/internet. Since it was only a few patients who stated this and because of limited time and finances, we did not develop a paper and pencil PtDA. However, we chose to acknowledge this need by having a computer available for patient use in the hospital and having a nurse guide the patient through the PtDA decision-making process.

The content of the PtDA is now tailored to the individual based on gender and desire to have children, but not on risk profile. With the insufficient evidence on differential efficacy and safety of DMARDs, this attribute remains a challenge for future research.

Due to time and financial limitations, it was also infeasible to develop a digital referral to the PtDA accompanied by a personal recommendation for appropriate medication options. Nevertheless, it is technically interesting to digitalize this process and may even improve the uptake. Sending the summary with the patients' notes, preferences, worries and questions directly to the health professional was also not realized because it raised privacy issues. Perhaps in the future, the PtDA could incorporate an option that would allow patients to send their summary to their health professional. Such additional attributes might also help increase the users' uptake.

Compared to the majority of PtDA developments, our PtDA substantially attempted to include all stakeholders. However, only small groups of participants were involved and all (patients and health professionals) were recruited from two hospitals. Although the number of participants in all our steps actually match the recommended numbers (see (162, 177, 178)), our results may not be generalizable. When further developing or distributing this PtDA, attention should be paid to involve a larger and wider group of users.

Additionally, we have not compared and assessed our development of a web-based PtDA using the IPDAS process model and user-centred design methods with the development a web-based PtDA in a different way. To assess this, two web-based PtDAs need to be developed using different methods, but using the same ideas or content as the starting point. In our view, this seems unfeasible and undesirable. From our study, however, we can say that our methodology did allow us to clarify needs and we were able to adapt the PtDA to these needs.

Finally, in this article we have not addressed evaluating the effectiveness or impact of the PtDA. To evaluate whether the PtDA is successful in improving patient participation and supporting SDM, we have conducted a post-test only study with a historical comparison group (beta testing). The results are published elsewhere (179) and described in Chapter 6.

CONCLUSION

By combining the IPDAS development process model with user-centred design methods, patients and health professionals contributed to the development of a novel web-based PtDA. This PtDA aims to support arthritis patients in their choice between DMARDs after they have received suggestions for appropriate treatment options from their rheumatologist. We have successfully demonstrated that user-centred design methods were helpful in developing a user-friendly application and creating support for the adoption of the PtDA. With use of these methods, the PtDA fits the values of all stakeholders and easily integrates with the patient pathway and daily workflow of health professionals. It is our expectation that this design approach may ease the uptake of PtDAs.

ADDITIONAL FILES

All additional files can be downloaded via: https://bmcmedinformdecismak.biomedcentral.com/articles/10.1186/s12911-017-0433-5

Additional file 1: All paper prototypes used in the needs assessment. Description: Images of all the paper prototypes used to assess needs.

Additional file 2: Design process from paper prototype to working prototype and final version. Description: Screenshots of the page that allows patients to compare DMARDs, illustrating the design process from paper prototype to working prototype and final version.

Additional file 3: Card to refer to the Patient Decision Aid. Description: Image of the card rheumatologists use to refer patients to the Patient Decision Aid.

Additional file 4: Instructional video of the Patient Decision Aid. Description: Instructional video of the Patient Decision Aid (in Dutch).

Intermezzo

This intermezzo provides an overview of the content and some screenshots of the final version of the PtDA. The PtDA is targeted at patients with inflammatory arthritis who face the decision to initiate a (different) DMARD. Figure 1 illustrates how it is embedded in the patient pathway. The patient and rheumatologist have an initial conversation about starting a (different) DMARD. During this conversation, the rheumatologist refers the patient to the web-based PtDA along with a referral card. The referral card states the Internet address of the PtDA, and the patient is encouraged to access the PtDA at home. The PtDA aims to support patients to compare DMARDs and elicit preferences, worries and questions regarding the decision to initiate a (different) DMARD. In the next consultation, the patient and rheumatologist can discuss the insights gained from using the PtDA and come to a shared decision.

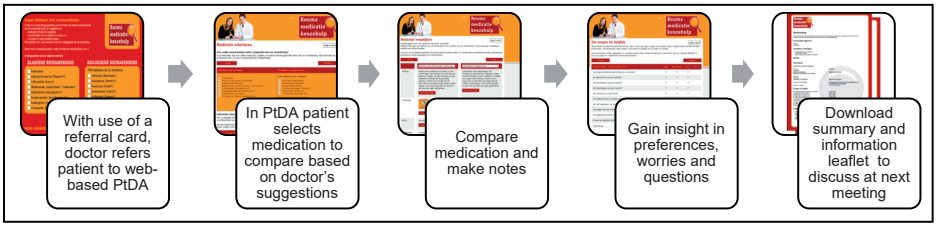


Figure 1: *Integration of PtDA in patient pathway*

THE REFERRAL CARD

After having an initial conversation about starting a (different) DMARD, the rheumatologist refers the patient to the web-based PtDA along with a referral card. On this referral card, the rheumatologist indicates the DMARDs appropriate for the patient at this specific moment. The referral card states the internet address of the PtDA, and the patient is encouraged to access the PtDA at home.

Title: Deciding together about DMARDs.

Short text indicating purpose of PtDA:
- Compare medication
- Weigh pros and cons
- Elicit worries and questions

After completion the insights can be discussed with rheumatologist to come to a shared decision.

From the lists with DMARDs, the rheumatologist can tick appropriate medication.

Referral to Internet address of PtDA.

Figure 2: *The referral card*

Table 1: *Structure of the web-based patient decision aid (PtDA)*

	Focus	**Key components**
General information		
Welcome	Focus on shortly describing aim and structure of the website.	Relevance of SDM, description of target group, goal of PtDA.
Shared decision-making	Focus on educating patients on SDM, the added value of patient participation and how the PtDA might be helpful.	Relevance of SDM, process of SDM, preparing for a SDM conversation (decision talk), function of PtDA.
Inflammatory arthritis	Focus on educating patients about their diagnosis, its symptoms and consequences.	Inflammatory arthritis, rheumatoid arthritis, psoriatic arthritis, ankylosing spondylitis.
Medication	Focus on general information about medication to control symptoms of inflammatory arthritis. The PtDA focuses on DMARDs, however this page offers also some information on various types of painkillers.	Simple analgesic drugs, nonsteroidal anti-inflammatory drugs (NSAIDs), synthetic DMARDs, biologic DMARDs.
Effectiveness of medication	Focus on educating patients about how to measure and influence the effectiveness of DMARDs.	General aims of DMARDs, how to influence the effectiveness, how to measure effectiveness.
Side effects of medication	Focus on educating patients about side effects, how to recognize and deal with them.	Definition of a side effect, how to deal with side effects, additional checkups in the hospital.
Science and your treatment plan	Focus on educating patients about the relation between the results of science, medical guidelines and a personal treatment plan.	Aims and process of research on effectiveness and safety of medication, development and function of medical guidelines, personal treatment plan.

Table 1: Structure of the web-based patient decision aid (continued)

	Focus	Key components
Patient Decision Aid		
Instruction PtDA	Instruction on goal and steps of the PtDA.	Instructional video, written instruction.
Start PtDA	Patients are asked to select the medication that the rheumatologist ticked on the referral card. Plus, patients are asked to answer some questions in order to tailor the information to the personal situation.	Selection of medication to compare, answer questions to tailor information.
Comparing medication	This application helps patients to compare medication side to side. Elements to compare include both clinical aspects as well as practical implications. Patients can also make notes.	Comparison of clinical aspects, comparison of practical implications, digital notebook.
Eliciting preferences	This application helps patients to gain insight in their preferences.	Questions about preference for manner of administration, frequency of administration, preliminary preference for DMARD.
Worries and doubts	This application helps patients to gain insight in their worries and doubts about initiating DMARDs.	A list with common worries and doubts, possibility to add a note.
Questions	This application helps patients to gain insight in remaining questions about initiating DMARDs.	A list with common questions, possibility to add a question.
Summary	Patients can download a summary that compiles the patient's notes and exercise responses. This summary be used during the patient's next consultation with the rheumatologist.	Summary with patients' notes and exercise responses.

THE WEB-BASED PATIENT DECISION-AID

The website consists of two components: general information and the PtDA itself, as illustrated in Table 1. The component with general information addresses SDM, emphasizes the importance of the patient's role in medical decision-making, and provides general information about inflammatory arthritis and DMARDs. The component with the PtDA consists of 1) an application to compare particular DMARDs; 2) value clarification exercises; and 3) a printed summary of patients' notes, preferences, worries and questions that patients can bring to discuss with their rheumatologist. The PtDA can be accesses via (http://www.reumamedicatiekeuzehulp.nl).

Homepage

The homepage describes shortly the relevance of SDM, the aims of the PtDA and the content and structure of the website (see Figure 3). The two components of the website, general information and the PtDA, are highlighted by the red blocks on the left side of the screen.

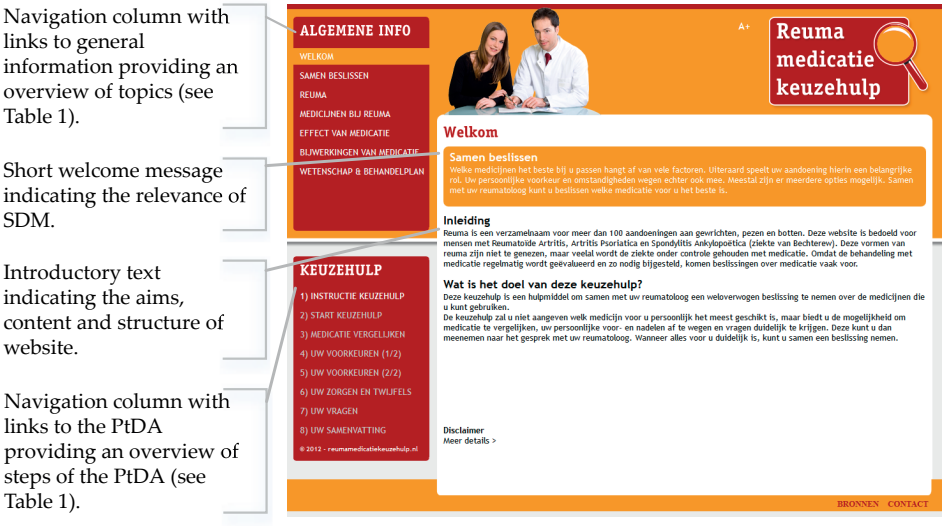


Figure 3: *Screenshot of homepage of PtDA*

Instruction page of PtDA

Prior to starting the PtDA, written instructions and an instructional video are provided to inform patients about the aims and steps of the PtDA and how to use it.

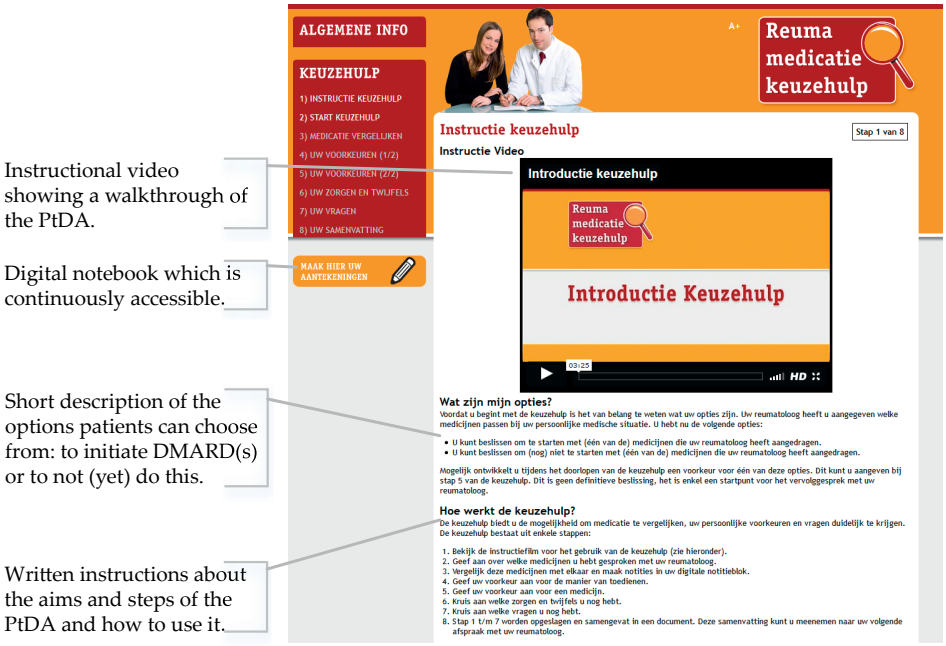


Figure 4: *Screenshot of instruction page of PtDA*

Start of PtDA

The first step of the PtDA is to select DMARDs to compare based on the suggestions the rheumatologist ticked on the referral card. By answering a few questions, patients can tailor the content of the PtDA based on their gender and desire to have children. If they choose to not answer these questions they will receive all information. Figure 5 illustrates this page.

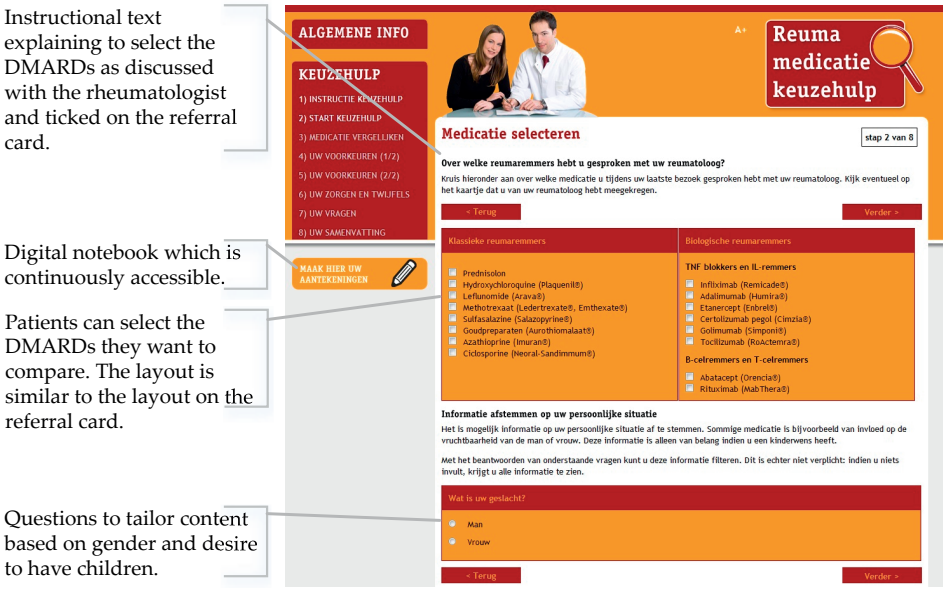


Figure 5: *Screenshot of start of PtDA where patients select DMARDs to compare*

Application to compare DMARDs

This application allows patients to compare selected DMARDs side-by-side. Figure 6 presents a screenshot of this application. Elements that can be compared include: target and working mechanism of the medication, manner of administration, approximate time-to-benefit, risks of side effects, follow-up process, combination with other drugs, fertility / pregnancy, consequences of continuing or stopping the DMARD, drug marketing history, restrictions and warnings for nutrition and alcohol consumption, and impact on daily life (e.g. storage, daily routine, traveling).

In order to fulfil the high need for information of most users while not overwhelming others, the PtDA has a flexible information system - supplemental information about the medication can be obtained via links that unfold certain elements. Furthermore, a digital notebook is provided. Notes are continually accessible and automatically saved as part of the summary which can be downloaded and printed at the final step of the PtDA.

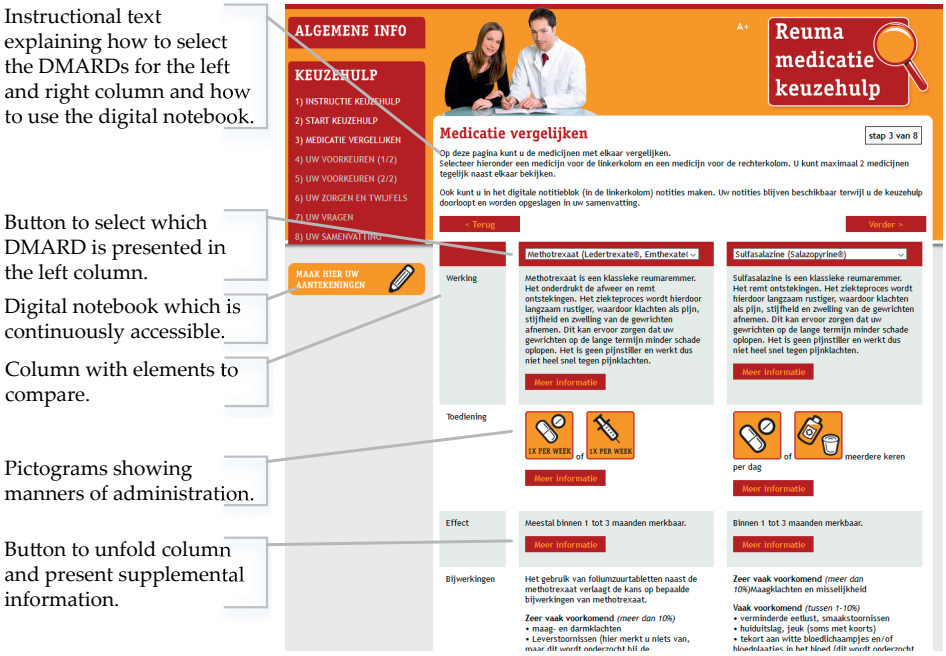


Figure 6: *Screenshot of application to compare DMARDs*

Value clarification exercises

The value clarification exercises consist of a couple of questions to elicit preferences, worries and questions. The questions about preferences focus on preference for manner and frequency of administration (see Figure 7) and preference for a particular DMARD. When answering these questions, the PtDA automatically presents which DMARD(s) fits these preferences.

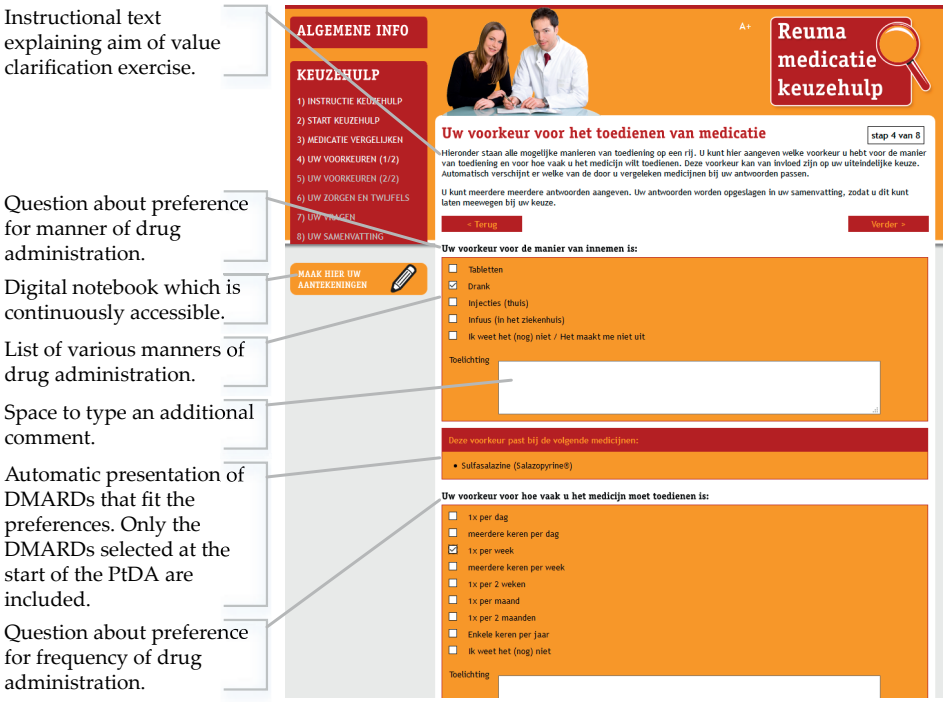


Figure 7: *Screenshot of page with a value clarification exercise to elicit patient's preference for manner and frequency of administration.*

To elicit patients' worries about initiating a (different) DMARD, a list with common worries is used. For each item patients grade their level of concern (see Figure 8). From a list with common questions, patients can select the questions they want to ask their rheumatologist. These lists are based on interviews we had with patients (see Chapter 4). Patients can also add their concerns and questions to the list. The results of the value clarification exercises are automatically saved as part of the summary which can be downloaded and printed at the end of the PtDA.

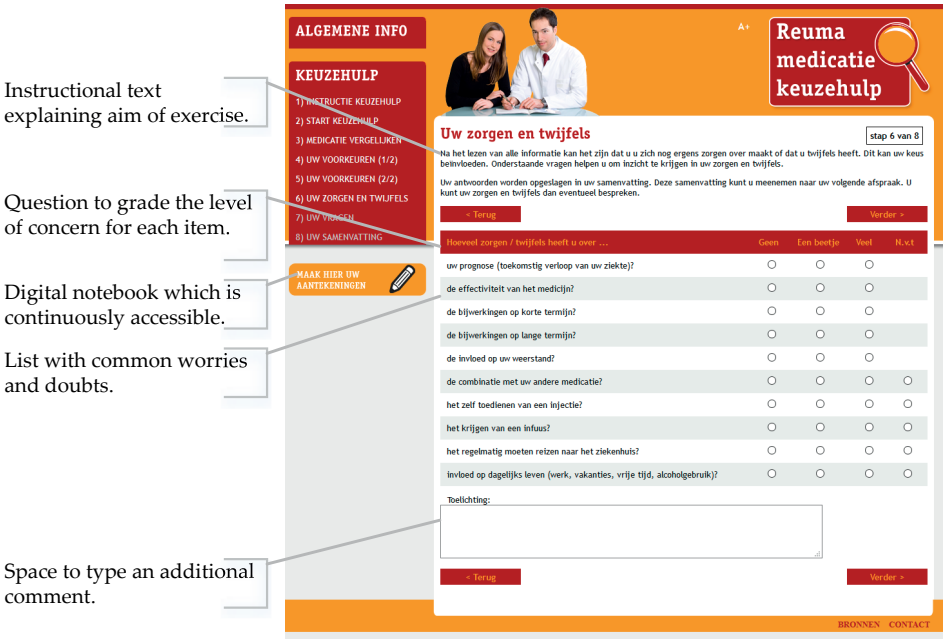


Figure 8: *Screenshot of page with a value clarification exercise to elicit patient's worries and doubts about initiating DMARDs.*

The summary

The final step of the PtDA is to download the summary compiling patient's notes, preferences, worries and questions. The summary can also be send to an email address. Patients can print or save this summary and discuss with their rheumatologist during the next consultation. On this page also leaflets of DMARDs can be downloaded. Figure 9 presents a screenshot of this page and Figure 10 the summary.

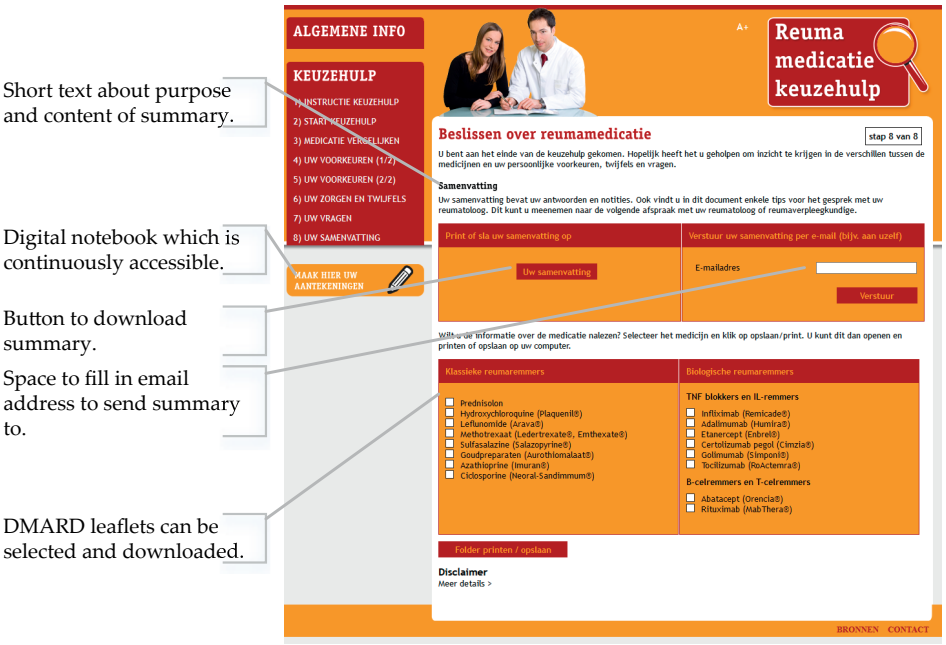


Figure 9: *Screenshot of page where summary and information leaflets can be downloaded.*

Short text about purpose and content of summary.

Personal information that was used to tailor content (e.g. gender and desire to have children).

DMARDs that were selected at the start of the PtDA.

Notes from digital notebook.

Preferences for manner and frequency of drug administration.

Preference to initiate treatment or not, and if yes which DMARD.

List with worries and doubts and their graded level of concern.

List with selected questions.

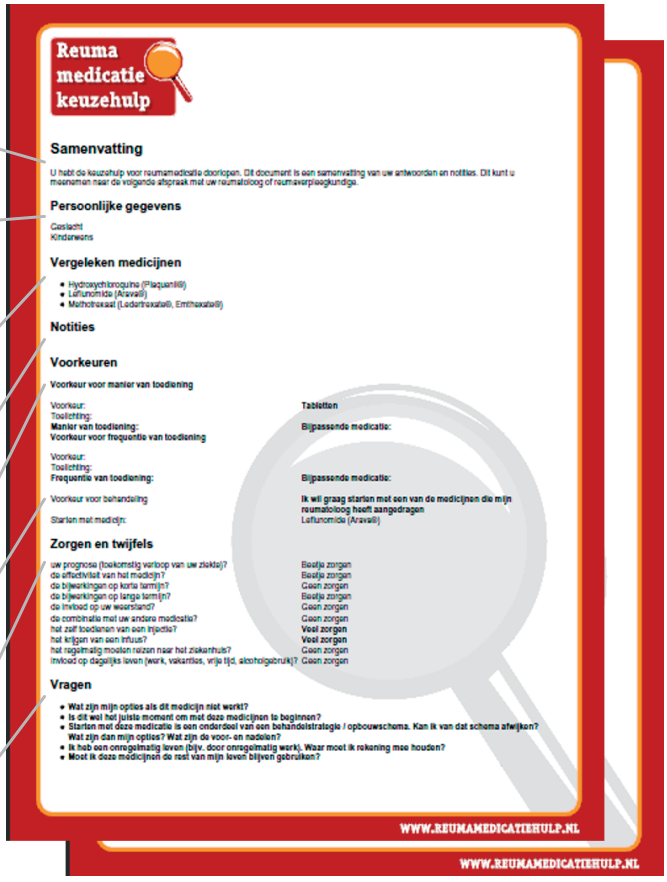
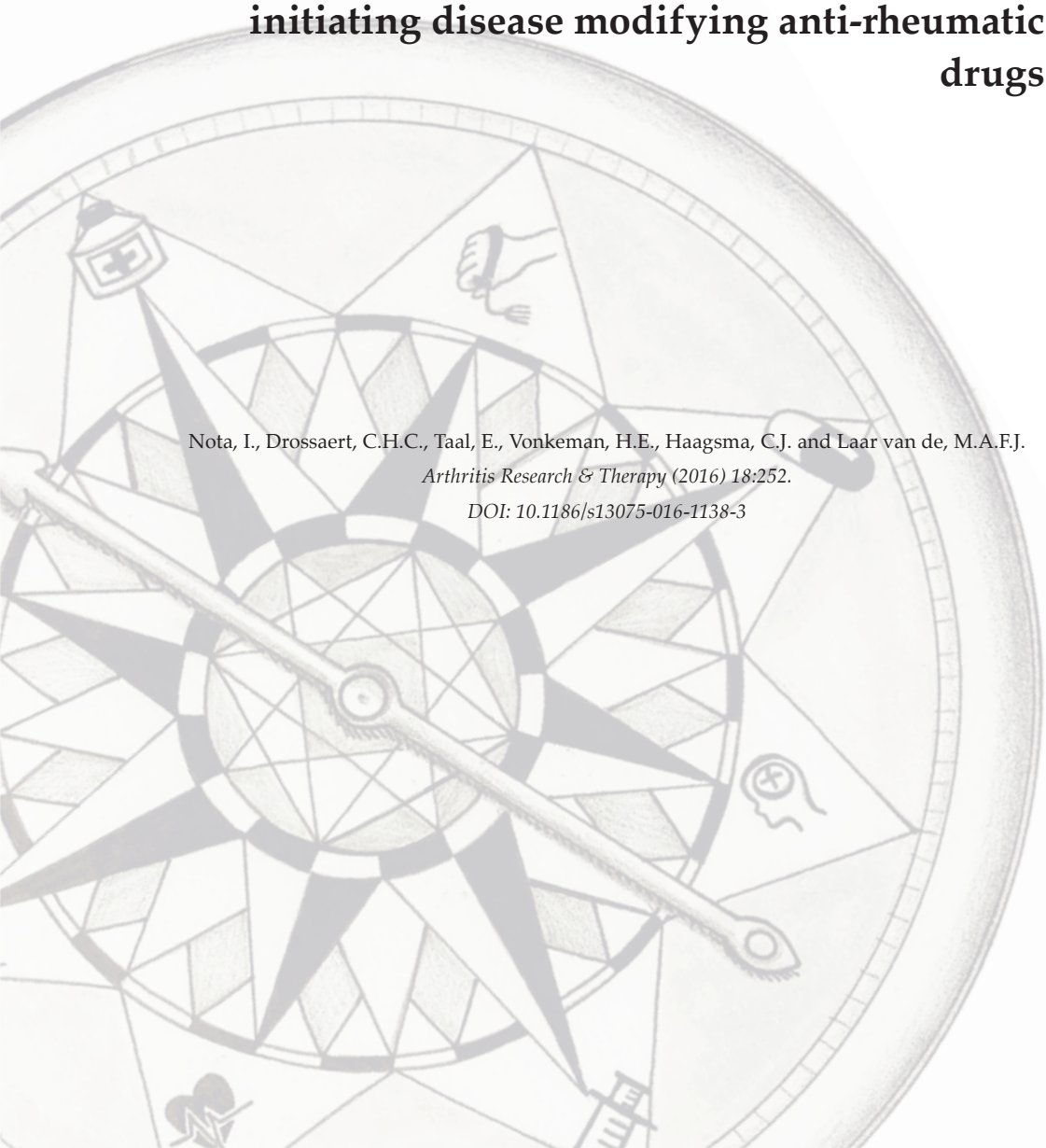


Figure 10: *Screenshot of summary compiling patient's notes, preferences, worries and questions.*

Chapter 6

Evaluation of a patient decision aid for initiating disease modifying anti-rheumatic drugs



Nota, I., Drossaert, C.H.C., Taal, E., Vonkeman, H.E., Haagsma, C.J. and Laar van de, M.A.F.J.
Arthritis Research & Therapy (2016) 18:252.
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ABSTRACT

According to international guidelines, treatment of inflammatory arthritis should be based on a shared decision between the patient and the rheumatologist. Furthermore, patients with inflammatory arthritis have a high information need and want to be more actively involved in medical decision-making. To facilitate shared decision-making and to support patients in choosing between disease modifying anti-rheumatic drugs (DMARDs), a web-based Patient Decision Aid (PtDA) was developed. A study was conducted to evaluate use, appreciation and effect of this PtDA. A post-test only study with a historical comparison group was conducted. In a two-year period, all patients diagnosed with Rheumatoid Arthritis, Psoriatic Arthritis or Ankylosing Spondylitis who were facing the decision to start a (different) DMARD were invited to participate. In the first year, patients received standard information (comparison group). In the second year, patients were referred to the PtDA (intervention group). In both groups, a questionnaire was sent four weeks after consulting the rheumatologist. Patient characteristics included socio-demographic, health-related and preference-related variables. Process measures regarded use and appraisal of the PtDA (intervention group only). The primary outcome measure was patients' perceived role in medical decision-making. Secondary outcome measures comprised satisfaction with the decision-making process and the decision, beliefs about medication, adherence to medication and trust in physician. We received 158/232 (68%) questionnaires from the comparison group and 123/200 (61%) from the intervention group. The PtDA was used by 69/123 (57%) of patients in the intervention group. Patients that used the PtDA highly appreciated it and perceived it as easy to use and helpful. Relative to the comparison group, patients in the intervention group perceived a more active role in medical decision-making and decisions were more in line with patients' personal preferences. Other outcomes showed no significant difference between the two groups. The web-based PtDA was appreciated highly and perceived as helpful for decision-making. Implementation of the PtDA in rheumatology practice was associated with a significant higher proportion of patients perceiving an active role in medical decision-making and decisions were more in line with patients' personal preferences. The PtDA can be a valuable aid in improving patient participation in decision-making about DMARDs.

INTRODUCTION

In recent years, several studies have shown that patients with inflammatory arthritis have a high information need and want to be more actively involved in medical decision-making (22, 26, 62-65, 128, 160). Medical decisions in this population focus primarily on the management of the disease with synthetic and biologic disease modifying anti-rheumatic drugs (sDMARDs and bDMARDs). When weighing the options, elements to consider include treatment efficacy, approximate time to benefit, possible side effects, current and future risks, cost-effectiveness, route of administration and impact on daily life. Given the preference sensitive elements of these treatment options, treatment of inflammatory arthritis should be based on a shared decision between the patient and the rheumatologist (58-60).

While desirable, implementing shared decision-making (SDM) in daily clinical practice is challenging. Patients often find it hard to recognise that a decision needs to be made and difficult to actively participate in the process to come to an informed values-based decision (39, 160). Physicians, on the other hand, may be uncomfortable with patient-involvement due to a lack of time, self-efficacy or skills (31).

To facilitate SDM and to support patients in making treatment decisions, Patient Decision Aids (PtDA's) have been developed for a wide variety of conditions and treatments (30). PtDA's make the decision being considered explicit, describe all available treatment options and their pros and cons, and help patients to consider the options from a personal perspective (51, 52).

PtDA's have repeatedly shown to have a positive impact on patients' knowledge about options, accurate risk perceptions, and feelings of being informed (30). Moreover, PtDA's have improved patients' involvement in medical decision-making and lead to decisions that are more in line with patients' personal preferences (30). Furthermore, PtDA's sometimes have a positive impact on patients' satisfaction with decision-making, anxiety, adherence or health outcomes (30). Although these effects are not likely to be very different, in rheumatology, only a few studies on PtDA's have been reported and their effects have not yet been thoroughly determined (30, 66, 158).

With the objective of supporting SDM in rheumatology, we developed a web based PtDA for initiating sDMARDs and bDMARDs. We have previously described the systematic development of this PtDA (180) using the development process model of the International Patient Decision Aids Standards (IPDAS) (53) combined with user-centred design methods (54, 55).

We conducted a study to evaluate the use, appreciation and effect of the PtDA. The study focused on answering the following research questions: 1) how

many patients use the PtDA; 2) what are determinants of use; 3) how do patients appreciate the PtDA; and finally, 4) in comparison to usual care, what is the effect of the PtDA on patients' perceived role in medical decision-making, satisfaction with the decision and decision-making process, beliefs about medication, adherence and trust in the physician. The primary outcome of the study was the impact of the PtDA on patients' perceived role in medical decision-making, in comparison to usual care. We also examined use of the PtDA by patients, determinants of use and patients' appreciation of the PtDA. Furthermore, we explored the impact on satisfaction with the decision and the decision-making process, beliefs about medication, adherence to medication, and trust in the physician.

METHODS

Description of the PtDA and its integration in clinical practice

The PtDA is intended for patients diagnosed with Rheumatoid Arthritis (RA), Ankylosing Spondylitis (AS) or Psoriatic Arthritis (PsA) who face the decision to initiate a DMARD or change to a different DMARD. Based on previous work (160, 161), the tool was designed to enable patients to compare multiple DMARDs with regard to both clinical information as well as practical information with possible consequences for daily life. Furthermore, it aims to support patients in determining treatment preferences, worries and questions and to help patients to express these feelings and questions to the health professionals.

Ideally, the PtDA is integrated in the patient pathway. First, the rheumatologist and patient have an initial conversation about starting a (different) DMARD. During this conversation the rheumatologist refers the patient to the web-based PtDA with use of a card. On this referral card the rheumatologist ticks the DMARDs appropriate for this individual patient. The referral card also states the internet address of the PtDA. After the conversation, the patient can use the PtDA at home. The web-based PtDA consists of various parts: 1) general information about shared decision-making, inflammatory arthritis (RA, AS and PsA) and DMARDs; 2) an application to compare the particular DMARDs ticked on the card by the rheumatologist; 3) exercises to gain insight into preferences, worries and questions; 4) a printed summary with the patients' notes, preferences, worries and questions to be discussed with the rheumatologist at the next consultation. The PtDA is in Dutch, but can be visited via www.reumamedicatiekeuzehulp.nl.

Study design

A post-test only study with non-equivalent groups was conducted. In this design two nonrandomized groups are compared in a post-test design. In this study, a historical comparison group was used, i.e. the comparison group preceded the intervention group in time.

The study covered a two-year period. In the first year, patients received standard information (comparison group). This standard information consisted of a one page information leaflet briefly describing the DMARD under consideration. It described the intended effect, possible interactions with other medicines, the manner of administration, the follow-up process, a short list with common and important side effects, and possible impact on fertility, pregnancy and breastfeeding. In the second year, patients were referred to the PtDA (intervention group), as described earlier. A questionnaire was sent four weeks after inclusion.

Patients and procedure

Patients were recruited from two large teaching hospitals in the Netherlands: Medisch Spectrum Twente and Ziekenhuisgroep Twente. Both participating hospitals work according to shared standard operating procedures on how to provide treatment information, which are in line with national guidelines. This practice is uniform across the 6 rheumatologists in each hospital.

All consecutive patients diagnosed with RA, PsA or AS who visited one of the clinics and discussed initiating a (different) DMARD were informed about the study by their rheumatologist (both years included the same rheumatologists) and asked to give permission for the researcher to contact them. Patients who participated in the comparison group were excluded from participation in the intervention group by the researcher (IN).

Patients who agreed to participate (232 patients in the first year; 200 patients in the second year) were sent the questionnaire by mail, four weeks after the consultation. The questionnaire was accompanied by a letter from their rheumatologist and an informed consent form. The letter stated the decision and the treatment options as discussed at time of inclusion. Patients were asked to return the completed questionnaires and informed consent form to the university using a prepaid return envelope. After three weeks a reminder was sent to those who had not yet returned the questionnaire.

Measurements

The questionnaire contained questions on patient characteristics, process measures and outcome measures. Below the measurements and the statistics for internal consistency we calculated are described for each measure. Standardized scales were

used as much as possible. If there was no Dutch scale available, scales were translated using the forward-backward procedure (108).

Patient characteristics

Patient characteristics included socio-demographic, health-related and preference-related variables and treatment options as suggested by the rheumatologist.

Socio-demographics included gender, age, marital status, education level and work status.

Health-related variables included diagnosis, time since diagnosis, pain and physical function. Pain was assessed as arthritis related pain in the prior week with a 0 to 10 numerical rating scale. To measure physical function we used the 10-item Health Assessment Questionnaire version 2 (HAQ-II) (181). Mean scores range from 0 (minimal loss of function) to 3 (completely disabled) (Cronbach's α=0.92).

Preference-related variables included role preference in decision-making about DMARDs and need for information. Role preference was assessed using the 'Control Preferences Scale' (CPS) (109) adapted by Garfield et al. (65). Patients were asked: "If you are informed about the benefits and risks, who should finally decide about initiating DMARDs?" and could respond on a 5-point scale: 1 (the rheumatologist), 2 (mostly the rheumatologist), 3 (the rheumatologist and me together), 4 (mostly me), and 5 (me alone). The answers were summarized into the values 1 ((mostly) doctor), 2 (shared) and 3 ((mostly) patient), as validated by Degner et al. (109). Need for information was measured with a 4-item subscale 'need for clarification of medical facts' of the Cologne Patient Questionnaire (CPQ) (112, 113). Mean scores range from 1 to 5 with a higher score indicating higher need for information (Cronbach's α=0.83).

Process measures

Process measures regarded use and appraisal of the PtDA (intervention group only).

Use of the PtDA was assessed by asking respondents if they had 1) received the referral-card, 2) received explanation about the PtDA and 3) hd visited the PtDA-website. Reasons for not visiting the PtDA-website were also assessed. Users of the PtDA were asked which tasks they performed on the PtDA-website. Response options are specified in table 2.

Appraisal of the PtDA was assessed with constructs including subjective impact of the PtDA (5 items; Cronbach's α=0.84), perceived usefulness (8 items; Cronbach's α=0.88), ease of use (4 items; Cronbach's α=0.87), attractiveness (2 items; Cronbach's α=0.97), and attitude towards future use (2 items; Cronbach's α=0.92). The latter four constructs are based on the Technology Acceptance Model (TAM) (171, 172). Statements regarded the PtDA in general and specific elements. Items and response

options are specified in table 4. Mean construct scores range from 1 to 5 with higher scores reflecting higher appraisal. Finally, respondents were asked to rate the overall quality of the PtDA in a range from 0 to 10.

Outcome measures

Our primary outcome measure was patients' perceived role in medical decision-making. Secondary outcome measures comprised satisfaction with the decision and decision-making process. Other secondary outcome measures comprised beliefs about medication, adherence and trust in the physician. All outcome measures specifically focused on the decision that patients discussed with their rheumatologist at time of inclusion. Rheumatologists registered which treatment options they suggested.

Perceived role in medical decision-making was assessed with the Control Preferences Scale (CPS) (65, 109). This measure was also used to assess patients' preferred role. To assess perceived role, patients were asked "In your opinion, who finally made this decision?" Patients could respond on a 5-point scale (see above). Scores were summarized into the values 1 ((mostly) doctor), 2 (shared) and 3 ((mostly) patient).

Satisfaction with the decision and decision-making process was assessed with 6 scales: satisfaction with participation, satisfaction with amount of received information, informed choice, decision control, satisfaction-uncertainty and consistency with personal values. The satisfaction with participation scale was developed for this study by the researchers and also consists of five items: "My rheumatologist asked me my opinion on this decision"; "I expressed my opinion on this decision"; "There was enough time for questions"; "I was able to express my questions, worries and doubts" and "My questions were answered". Mean scale scores range from 1 to 5 with a higher score indicating higher levels of satisfaction with participation (Cronbach's α=0.71).

Satisfaction with amount of received information was assessed with items based on the Satisfaction with Information about Medicines Scale (SIMS) (182). Respondents rated the amount of information received. It originally includes seventeen items (i.e. information topics), we added four items based on the Dutch legal standards for informed consent: the dosage, the frequency of administration, storage and storage life. Response options are: 0 (no, far too little), 1 (no, little too little) and 2 (yes, sufficient) which were recoded into: 0 (no) and 1 (yes). The sum scores range from 0 to 21 with higher scores indicating a higher degree of overall satisfaction with the amount of information received (Cronbach's α=0.91).

The Dutch Decision Evaluation Scales (DES) (183) were used to assess [A] informed choice: patient's perception of the quality of the received information (5 items α=0.86), [B] decision control: patient's perceived level of control over the

decision in terms of feelings of regret, anxiety and deciding under pressure (5 items, α=0.62), and [C] satisfaction-uncertainty: the extent to which a patient is satisfied or still has doubts about the decision (5 items, α=0.56). Scale scores range from 1 to 5 with higher scores indicating higher levels of informed choice, decision control and higher satisfaction (less uncertainty).

The consistency with personal values scale is a two-item subscale of the Satisfaction With Decision (SWD) scale (184) and measures whether the decision meets personal preferences (Cronbach's α=0.79). Mean scale scores for both scales range from 1 to 5 with a higher score indicating higher consistency.

Other secondary outcome measures comprised patients' beliefs about DMARDs, adherence and trust in the physician. Patients' beliefs about DMARDs was assessed with the Beliefs about Medicines Questionnaire (BMQ) (185). The BMQ includes two 5-item subscales assessing patients' beliefs about the necessity of medication and their concerns about it. Sum scores for both scales range from 5 to 25 with higher scores indicating stronger beliefs (Cronbach's α=0.83 and α=0.73, respectively).

Adherence was measured in participants who self-administered their DMARD. Participants who had help from a caregiver or who went to the clinic for administration (e.g. intravenous therapy) were excluded from the analysis. We used the 8-item Morisky Medication Adherence (MMA) scale (186). Sum scores range from 0 to 8 with higher scores representing better adherent behaviour (Cronbach's α=0.77).

Trust in the physician was assessed with a 3-item subscale of the Cologne Patient Questionnaire (CPQ) (112, 113). Mean scale scores range from 1 to 5 with a higher score indicating higher trust (Cronbach's α=0.90).

Treatment options suggested by the rheumatologist were registered at time of inclusion by the rheumatologist after obtaining the patients' consent. We counted the number of suggested options and grouped it into "one option" or "more than one option". If combination therapy was suggested (e.g. methotrexate combined with hydroxychloroquine or methotrexate combined with adalimumab) and no alternative options were presented, it was coded as one option.

Pilot test

Prior to inclusion, we performed a pilot test among patients (n=10) to assess the readability of the questionnaire and acceptability of the time it takes to complete the questionnaire. The test showed that the questionnaire took about 30 minutes to complete, which was acceptable according to the participants. Minor textual adjustments were made following the results of the pilot test.

Statistical analysis

The Statistical Package for the Social Sciences (version 21.0 IBM SPSS Inc, Chicago, IL, USA) (187) was used to perform all analyses. Pearson chi-square tests (for categorical variables) and t-tests (for continuous variables) were performed to compare characteristics of the comparison group and the intervention group, to examine which factors were associated with use of the PtDA and to evaluate the impact of the PtDA.

RESULTS

Patient characteristics

The patients in the comparison group returned 158/232 questionnaires (response rate 68%), from the intervention group we received 123/200 (response rate 61%). Within both the comparison group and the intervention group there were no significant differences between respondents and non-respondents with regard to age, gender, diagnosis and amount of options suggested by the rheumatologist.

The comparison group and intervention group did not differ with regard to socio-demographic, health-related and preference-related variables, except for marital status (table 6.1). In both groups mean age was about 55 years, most respondents were women and most were diagnosed with RA. Both groups responded having a high need for medical information and most participants preferred shared decision-making.

Table 6.1. *Patient-related characteristics (N=281)*

Variables	Categories / Range	Comparison group (N=158)	Intervention group (N=123)	P[a]
Socio-demographic variables				
Age, years		54 ± 15 (158)	55 ± 13 (123)	n.s.[b]
Gender, % (n)	Women	65% (102)	61% (75)	
	Men	35% (56)	39% (48)	n.s.
Marital status, % (n)	Married / living with partner	78% (121)	89% (109)	
	Not married / Living alone	22% (34)	11% (14)	.02
Education, % (n)	Low	26% (41)	30% (37)	
	Medium	52% (81)	50% (61)	
	High	22% (34)	20% (25)	n.s.
Work status, % (n)	Employed / studying	67% (82)	67% (58)	
	Not employed / not studying	33% (41)	33% (29)	n.s.
Health-related variables				
Diagnosis, % (n)	Rheumatoid Arthritis	76% (108)	81% (91)	
	Psoriatic Arthritis	19% (27)	13% (15)	
	Ankylosing Spondylitis	5% (7)	6% (7)	n.s.
Years since diagnosis, % (n)	<1	37% (58)	25% (31)	
	1-5	34% (52)	41% (50)	
	6-10	10% (16)	12% (14)	
	>10	19% (29)	22% (27)	n.s.
Pain (NRS)	Range 0-10	4,7 ± 2,4 (158)	4,7 ± 2,5 (123)	n.s.[b]
Physical function (HAQ-II)	range 0-3	2.17 ± 0.57 (155)	2.13 ± 0.57 (120)	n.s.[b]
Preference-related variables				
Preferred role in decision-making (CPS), % (n)	(Mostly) doctor	15% (23)	8% (10)	
	Shared	76% (119)	77% (94)	
	(Mostly) patient	9% (14)	15% (18)	n.s.
Need for information (CPQ)	range 1-5	4.4 ± 0.8 (154)	4.5 ± 0.7 (121)	n.s.[b]

Note. Values are the mean ± standard deviation (n) unless otherwise indicated. Percentages do not include missing cases.

n.s. = not significant p>0.05; NRS = numerical rating scale; HAQ-II= Health Assessment Questionnaire, version 2 (181); CPS = Control Preference Scale (65, 109); CPQ = Cologne Preference Questionnaire (112, 113).

[a] *Tested with Pearson chi-square tests unless otherwise indicated.*

[b] *Tested with t-test.*

Use of the PtDA

The PtDA was used by 69/123 (57%) of respondents in the intervention group (table 6.2). Many of the non-users (23/53 (43%) of non-users; which is 19% of all respondents in the intervention group) mentioned that they had not received a referral card or could not remember having received one. Other reasons for not visiting the PtDA-website were not having an internet connection, having troubles finding the website, no interest and lack of time.

Of the PtDA-website visitors, 65/69 (94%) read the general information and 61/69 (90%) compared two or more DMARDs. The exercises to gain insight into preferences, doubts & questions were performed by 26/69 (38%) of the users. Furthermore, 31/69 (47%) of the users saved or printed an information leaflet. The summary with user's notes, preferences, doubts and questions was read by 50/69 (75%), shown to others by 24/69 (38%), saved or printed by 34/69 (52%) and taken to their next appointment with the rheumatologist by 18/69 (28%) of the users.

Determinants of use of the PtDA by patients

When exploring determinants of use of the PtDA, a few significant differences were found between users and non-users (table 6.3). Users were significantly younger and higher educated. There were no associations between PtDA-use and gender, marital status and employment. Nor was use associated with any of the health-related or preference-related factors. The number of options suggested by the rheumatologist was significantly associated with use of the PtDA; patients who were offered more than one treatment option, were more likely to use the PtDA than those who were offered only one.

Table 6.2. *Use of patient decision aid (N=123)*

Tasks	**% (n)**
Visited the PtDA-website	57% (69)
Reasons for not visiting the PtDA-website[a]	
Did not receive referral to the PtDA-website or cannot remember	19% (23)
No internet	7% (8)
Could not find PtDA-website	6% (7)
Website did not work	1% (1)
Not interested	6% (7)
No time	6% (7)
Missing	(1)
Received explanation about PtDA	
Yes	69% (85)
No	22% (27)
Cannot remember	9% (11)
Tasks performed on the PtDA-website (visitors only; n=69)[b]	
Read general information	94% (65)
Compared two or more DMARDs	90% (61)
Made notes in the digital notebook	16% (11)
Performed exercises about preferences, worries & questions	38% (26)
Saved or printed an information leaflet	47% (31)
Read the summary	75% (50)
Showed the summary to others	38% (24)
Saved or printed the summary	52% (34)
Took the summary to their next appointment with the rheumatologist	28% (18)

Note. Percentages do not include missing cases. PtDA = Patient Decision Aid. DMARD = Disease Modifying Anti-Rheumatic Drug.

[a] For reasons for not visiting the website, percentages are taken from the total population in the intervention group (n=123).

[b] For tasks performed, percentages are taken from visitors only (n=69).

Table 6.3. *Determinants of use of the Patient Decision Aid (N=123)*

Variables	Categories / Range	Users PtDA (N=69)	Non-users PtDA (N=53)	P[a]
Socio-demographic variables				
Age, years		52 ± 13 (69)	58 ± 12 (53)	.003[b]
Gender, % (n)	Women	57% (39)	68% (36)	
	Men	43% (30)	32% (17)	n.s.
Marital status, % (n)	Married/living with partner	87% (60)	91% (48)	
	Not married/Living alone	13% (9)	9% (5)	n.s.
Education, % (n)	Low	20% (14)	43% (23)	
	Medium	51% (35)	47% (25)	
	High	29% (20)	9% (5)	.004
Employment, % (n)	Employed/studying	65% (33)	69% (24)	
	Not employed/studying	35% (18)	31% (11)	n.s.
Health-related variables				
Diagnosis, % (n)	Rheumatoid Arthritis	81% (52)	80% (39)	
	Psoriatic Arthritis	13% (8)	14% (7)	
	Ankylosing Spondylitis	6% (4)	6% (3)	n.s.
Years since diagnosis, % (n)	<1	19% (13)	32% (17)	
	1-5	43% (29)	40% (21)	
	6-10	13% (9)	9% (5)	
	>10	25% (17)	19% (10)	n.s.
Pain (NRS)	Range 0-10	4.7 ± 2.5 (69)	4.8 ± 2.8 (53)	n.s.[b]
Physical function (HAQ-II)	range 0-3	2.14 ± 0.70 (69)	2.10 ± 0.71 (53)	n.s.[b]
Preference-related variables				
Preferred role in decision-making (CPS), % (n)	(Mostly) doctor	9% (6)	8% (4)	
	Shared	78% (54)	75% (39)	
	(Mostly) patient	13% (9)	17% (9)	n.s.
Need for information (CPQ)	range 1-5	4.6 ± 0.6 (69)	4.5 ± 0.7 (121)	n.s.[b]
Number of treatment options suggested by rheumatologist, % (n)	1 option	38% (23)	73% (36)	
	>1 option	62% (38)	27% (13)	.000

Note. Values are the mean ± standard deviation (n) unless otherwise indicated. Percentages do not include missing cases.
PtDA = Patient Decision Aid; n.s. = not significant p>0.05; NRS = numerical rating scale; HAQ-II= Health Assessment Questionnaire, version 2 (181); CPS = Control Preference Scale (65, 109); CPQ = Cologne Preference Questionnaire (112, 113).
[a] Tested with Pearson chi-square tests unless otherwise indicated.

[b]Tested with t-test.

Appraisal of the PtDA

Overall, users were very positive about the PtDA-website (table 6.4). Many respondents indicated that they learned a lot from it. They also indicated that it contained new information, helped to gain insight into preferences, worries and questions, helped to discuss things with their rheumatologist and helped with making a decision about the medication. They perceived it to be very useful, easy to use, easy to understand and attractive. The general information, the specific pharmaceutical information and the comparison of DMARDs were perceived as most useful. Furthermore, most participants intended to use the PtDA again in the future and would recommend the PtDA to others. The overall quality of the PtDA-website received a grade 7.7 on a scale from 0 - 10.

Impact of the PtDA

Relative to the comparison group, patients in the intervention group perceived significantly less often that the doctor decided and more often that they made the final decision about initiating DMARDs (table 6.5). With regard to the secondary outcome measures, we found that patients in the intervention group regarded the decision to be significantly more consistent with their personal preferences than patients in the comparison group. Finally, more participants in the intervention group were offered more than one medication option compared to patients in the comparison group, 46% vs 12%; $p<0.05$, respectively. For all other variables no significant differences were found between the groups.

Table 6.4 *Appraisal of the Patient Decision Aid (N=69)*

	Mostly Disagree (1 point)	Neutral (3 points)	Mostly Agree (5 points)	Mean ± SD
Subjective impact of the PtDA				
I learned a lot from the PtDA	11%	17%	71%	
The PtDA contained new information	7%	23%	70%	
The PtDA helped me to gain insight into my preferences, worries and questions	9%	22%	70%	
The PtDA helped me in making a decision about medication	11%	18%	70%	
The PtDA helped me in discussing my preferences, worries and questions with my rheumatologist	11%	28%	60%	
Total impact				3.8 ± 0.9
Perceived usefulness				
The PtDA in general was useful	7%	5%	88%	
Reading the general information was useful	2%	5%	94%	
The specific information about DMARDs was useful	4%	3%	94%	
Comparing DMARDs was useful	4%	8%	89%	
Making notes in the digital notebook was useful	22%	51%	28%	
The exercises about preferences and worries were useful	11%	29%	59%	
The list of frequently asked questions was useful	12%	32%	56%	
Total usefulness				4.0 ± 0.7
Perceived ease of use				
The website is easy to use	5%	3%	92%	
The information is easy to understand	2%	6%	92%	
The time the PtDA takes to finish is acceptable	5%	5%	90%	
The structure of the website is logical	10%	11%	79%	
Total ease of use				4.5 ± 0.8

Table 6.4 *Appraisal of the Patient Decision Aid (N=69) (continued)*

	Mostly Disagree (1 point)	Neutral (3 points)	Mostly Agree (5 points)	Mean ± SD
Attractiveness				
The colour of the website is pleasant	2%	17%	81%	
The font on the website is pleasant	4%	9%	87%	
Total attractiveness				4.4 ± 0.9
Attitude towards future use				
I would use the PtDA again in the future	5%	3%	91%	
I would recommend the PtDA to others	2%	7%	89%	
Total attitude				4.6 ± 0.9
Overall grade regarding the quality of the PtDA (range 0-10)				7.7 ± 0.9

Note. Percentages do not include missing cases. PtDA = Patient Decision Aid. DMARD = Disease Modifying Anti-Rheumatic Drug.

Table 6.5 *Impact of the Patient Decision Aid (N=281)*

	Comparison group N=158	**Intervention group N=123**	**P[a]**
Perceived role in decision-making *(CPS), % (n)*			
(Mostly) doctor	25% (39)	14% (17)	
Shared	70% (111)	73% (90)	
(Mostly) patient	5% (8)	13% (16)	.01 [b]
Satisfaction with decision and decision-making process			
Satisfaction with participation (range 1-5)	4.6 ± 0.6 (144)	4.6 ± 0.6 (115)	n.s.
Satisfaction with received information (range 0-21)	15.7 ± 4.9 (130)	15.3 ± 5.7 (106)	n.s.
Informed choice (DES) (range 1-5)	4.2 ± 1.0 (145)	4.3 ± 0.9 (115)	n.s.
Decision-control (DES) (range 1-5)	4.6 ± 0.5 (146)	4.6 ± 0.7 (114)	n.s.
Satisfaction-Uncertainty (DES) (range 1-5)[c]	4.0 ± 0.8 (147)	4.1 ± 0.7 (116)	n.s.
Consistency with personal values (SWD)	4.2 ± 1.0 (148)	4.5 ± 0.8 (112)	.02
Other categories			
Beliefs about medication - Necessity (range 5-25)	18.6 ± 4.5 (137)	19.6 ± 4.6 (87)	n.s.
Beliefs about medication - Concerns (range 5-25)	13.8 ± 4.1 (136)	12.9 ± 4.9 (90)	n.s.
Medication Adherence (MMAS) (range 0-8)[d]	7.2 ± 1.4 (129)	7.2 ± 1.4 (102)	n.s.
Trust in Physician (CPQ) (range 1-5)	4.8 ± 0.5 (155)	4.8 ± 0.4 (120)	n.s.
Number of treatment options suggested by rheumatologist, % (n)			
1 option	88% (137)	54% (60)	
>1 option	12% (18)	46% (51)	.000

** Values are the mean ± standard deviation (n) unless otherwise indicated.*
n.s. = not significant p>0.05; CPS = Control Preference Scale (65, 109); DES = Decision Evaluation Scales (183); SWD = Satisfaction With Decision scale (184); BMQ = Beliefs about Medication Questionnaire (185); MMAS = Morisky Medication Adherence Scale (186); CPQ = Cologne Preference Questionnaire (112, 113).
[a] Tested with t-test unless otherwise indicated.
[b] Tested with Pearson chi-square test.
[c] Higher scores indicate less uncertainty and higher satisfaction
[d] Adherence not assessed in patients that get professional assistance with administrating injections or go to the hospital for intravenous medication.

DISCUSSION

This study was conducted to evaluate the use, appraisal and impact of a PtDA for initiating DMARDs in patients with rheumatic diseases. The PtDA is designed to improve patient participation by supporting patients in determining treatment preferences, worries and questions and by endorsing them to express these feelings and questions to their health professionals. The study demonstrated that patients perceived the PtDA as very helpful in the decision-making process. Our primary research question focused on the impact of the PtDA on perceived role in medical decision-making. Relative to the comparison group, patients in the intervention group perceived a more active role in medical decision-making. Furthermore, decisions were more in line with patients' personal preferences. We found no differences between groups in satisfaction with the decision-making process, beliefs about medication, adherence or trust in the physician. However, this may be due to ceiling effects or limited psychometric quality of some of the instruments used (188). Generally, our results are in line with the impact of other PtDA's, as was shown in a recent systematic review (30).

By developing the PtDA in co-creation with patients and health professionals we aimed to develop a user-friendly PtDA that closely fits the needs of all users and consequently eased adoption and implementation. This study demonstrated that patients appreciated the PtDA highly and perceived it as useful, usable and helpful in the decision-making process.

The PtDA was used by 57% of the patients in the intervention group who had returned the questionnaire. Users were mostly younger and higher educated patients. Compared to other studies on PtDA's in routine practice as well as in clinical trials, our patient user rates are high. In other routine practice studies, patients' use of PtDA's varied between 25% and 37% (189-191). Clinical trials report much higher patient user rates, varying from 49% to 85% (192-194).

A recent systematic review suggests that adoption and implementation of PtDA's using a referral model (i.e. health professionals inviting eligible patients to use the PtDA) is often challenged by indifference on the part of health professionals (31). This indifference may stem from a lack of confidence in the content of PtDA's and concerns about disruption of established workflows (31). However, we believe that the relatively high percentage of patients that used the PtDA in our study may partly be explained by the active referral by the rheumatologists. Although we did not specifically assess factors enhancing system adoption in this study, we believe that the high referral and using rates could be attributed to the iterative and extensive involvement of patients and health professionals during the development process (180). To determine how to further increase the referral rates to the PtDA, we recently

conducted a focus group study with health professionals, the results of which are currently being analysed. Likewise, to further increase patients' PtDA use it should be investigated how the tool can be further adapted to the needs of older and lower educated patients.

Notably, our results showed a significant difference between the comparison group and the intervention group regarding the number of treatment options offered by the rheumatologist. This might have biased the findings because when patients are offered more than one option -or when options are more explicitly discussed-, patients might (automatically) feel that their role in medical decision-making is larger. However, the question remains why the amount of offered options registered by the rheumatologists was higher in the second period of the study (intervention group). There have been no apparent changes in availability of DMARDs and the way rheumatologists were asked to register the offered options was identical in both periods. Interviewing some of the participating rheumatologists revealed that the referral card and PtDA may have prompted the rheumatologists to more explicitly discuss options with patients and consequently more accurately register the offered options for the study.

Unfortunately, we cannot compare the effect on number of offered options to results of other PtDA's, because this is not a common measure in PtDA evaluations. Previously studied PtDA's have largely focused on decisions on whether or not to initiate a treatment or on choosing between a predefined limited number of treatment options. Widely studied examples include decisions like "Should I have chemo-therapy for early stage breast cancer?" and "Should I have breast-conserving surgery or a mastectomy for early stage breast cancer?" In rheumatology, previous PtDA's focused on the decision on whether or not to initiate one specific DMARD or a particular class of DMARDs (30, 66). Compared to these previously studied PtDA's, our PtDA encompasses many different treatment options. To reduce the potentially overwhelming number of choices and to eliminate all inappropriate options, we chose to let the rheumatologist preselect which DMARDs are appropriate choices for the individual patient at that specific moment. To our knowledge, this innovative flexible referral model has not previously been studied.

The main strength of this study is its virtual implementation of a PtDA in daily clinical practice. However, due to limitations inherent to the study design, some caution is needed when interpreting our results. Firstly, the post-test only study with a non-equivalent historical comparison group is susceptible to the internal validity of selection; any prior differences between the groups may have affected the outcome of the study. Yet, despite this limitation we chose deliberately for this study design in order to reduce contamination effects. If patients would have been randomized to a condition, PtDA or standard information, physicians would have

been exposed to both conditions simultaneously, which might have influenced their behaviour. Secondly, although we included many variables in our study, it remains difficult to control for all confounding variables. We realize that in this study we did not evaluate merely the effect of the PtDA. Introducing the PtDA, obviously, affected the health care system and the daily workflow of health professionals. Therefore, some caution is needed with causal interpretations of our results. Finally, due to non-response, our results might have been biased. It is likely that patients who have no need for participating in medical decision-making or in using a PtDA, are less interested in responding to a questionnaire about this subject.

Future multi-centre randomized trials need to be conducted to further study the impact of this PtDA and to compare the impact of this PtDA with other SDM interventions. A longitudinal study is needed to reveal what the impact is on the number of sessions and on cost-effectiveness. Furthermore, more research is needed to determine how to involve lower educated patients and patients in different age groups in medical decision-making.

CONCLUSION

This study was conducted to evaluate use, appraisal and impact of a PtDA regarding initiating DMARDs. The PtDA was used by the majority of the respondents, was appreciated highly and was perceived as helpful in the decision-making process. Relative to the comparison group, patients perceived a more active role in medical decision-making and felt the final choice to be more consistent with their personal preferences. From this study we can conclude that this PtDA can be a valuable aid in improving patient participation in medical decision-making about DMARDs.

Chapter 7

Summary and general discussion

Patients with inflammatory arthritis see their health professionals less than one hour per year. The rest of the time they must cope, on their own and in their own environment, with the erratic symptoms and side effects of treatment. These circumstances make it essential that patients are involved in their care and that patients' preferences are included in treatment decision-making. The choice of treatment should be based on a shared decision between the patient and rheumatologist (58, 61). This means that, in collaboration, the patient and rheumatologist choose the best treatment by weighing and balancing the medical knowledge and patient's values. To support the Shared Decision-Making (SDM) process and prepare patients to make medical decisions in collaboration with their clinician, Patient Decision Aids (PtDAs) can be used (30). Most commonly appearing as pamphlets, videos or web-based tools (30), PtDAs come in many forms, but all specifically state the decision being considered and stress the relevance of SDM. They can be brief enough to be used during the clinical encounter or they can include detailed information to be used before or after consulting the clinician.

The overarching aims of the project described in this dissertation were: (1) to gain knowledge of patients' perspectives on SDM and the potential role of a web-based PtDA in the setting of rheumatology care, (2) to examine the feasibility and value of developing a PtDA in co-creation with health professionals and patients, and (3) to evaluate the impact of the developed PtDA on patients' involvement in medical decision-making. In this final chapter, we will summarize the findings of our studies and reflect on our project. We conclude with practical implications and directions for future research that were an outcome of these reflections.

SHARED DECISION-MAKING IN RHEUMATOLOGY: WHAT MATTERS TO PATIENTS?

7

SDM is seen as a key element of high quality modern medicine, and its research has extensively focused on the general population, acute care settings and for one-time decisions, for example, governing screening or surgery. However, little is known about this topic in the rheumatology setting (7). Our studies, which we described in Chapters 2 and 3 of this dissertation, addressed this gap. With the use of qualitative and quantitative research methods, we gained knowledge about patients' perspectives of SDM in rheumatology. The results showed that, in rheumatology care. The majority of patients prefer to be involved in medical decision-making. Yet, our interviews (see Chapter 2) showed that many patients found it difficult to determine their preference regarding this subject, because they had never actively considered

it, had problems conceptualizing patient participation, or were unaware of having a choice. Furthermore, we found that patients' preferred level of involvement varied between *and* within individuals – depending on the type of decision and the severity of their complaints.

After we examined patients' experiences of medical decision-making, our studies revealed that SDM is frequently perceived in rheumatology outpatient care. Yet, there is room for improvement. A considerable group of patients (26% – 54%, depending on the type of decision) would have liked more participation than they had experienced. Newly diagnosed patients who faced the decision to initiate a synthetic disease-modifying anti-rheumatic drug (DMARD; 54%) especially desired more participation in decision-making (see Chapter 3). DMARDs are used to control the disease and to relieve or reverse rheumatic symptoms. Perceived barriers for patient participation could be divided into three factors: doctor-related (e.g. a paternalistic attitude), patient-related (e.g. lack of awareness of having a choice and lack of medical knowledge) and context-related (e.g. too little time to decide; see Chapter 2).

In summary, patients with inflammatory arthritis often want to participate but are frequently unable to do so to the extend they desire. In order to participate and overcome the barriers they experience, patients need knowledge, skills and personal power (157). Regarding the *knowledge*, they need knowledge about the disease and symptoms, treatment options and possible outcomes. Besides this, they need knowledge about SDM and the relevance of patient participation in medical decision-making. Required *skills* are related to health literacy (e.g. searching and reviewing medical information) and decision-making (e.g. eliciting one's own needs and preferences, communicating worries and questions, and deliberating options). Finally, they need *power* to believe in their capacity to influence the treatment decision-making (i.e. self-efficacy). This includes factors such as: believing that they have permission to participate and ask questions, having confidence in the value of their own knowledge and ability to acquire medical knowledge, and self-efficacy to use decision-making skills. To support patients in decision-making about DMARDs on all these three levels (knowledge, skills and power), we chose to develop a PtDA.

DEVELOPING A PATIENT DECISION AID WITH A USER-CENTRED DESIGN APPROACH

Although PtDAs are increasingly being developed and their evaluations have shown great potential (30), widespread adoption has not yet occurred (31). The reasons

behind this are still unclear, although many explanations have been proposed. From a patient viewpoint, possible reasons are that patients may be unaware of the existence of these tools, which corresponds with patients being unaware of having a choice, as described in Chapter 2. In addition, there may be usability issues or the goals of the PtDA may not adequately match the aims of the patients (195). Because utilization of PtDAs by patients often relies on health professionals referring patients to these tools, health professionals have a key role in the adoption process. Barriers for adoption from the health professional viewpoint are a lack of confidence in the content of PtDAs and concerns about disruption of established workflows (31). Similar to their patients, health professionals may also be unaware of the existence of these tools. From an organizational viewpoint, the limited adoption of PtDAs can be explained by the fact that many do not comply with other daily clinical care processes (31).

All these stakeholders – patients, health professionals and policy makers– can have diverse values and interests, which can facilitate or hamper the uptake and implementation of PtDAs. Therefore, it is essential to take into account the needs of the main stakeholders during PtDA development. Even though the importance of stakeholder involvement during development of e-health applications like PtDAs is gaining ground (53, 195), it is unclear whether and how this happens in practice. Most studies of PtDAs only report about the use, satisfaction and effects of the PtDA, but neglect to examine the developmental process (53, 103). Those rare studies that do examine the process, for the most part, just mention *which* stakeholders were involved and fail to describe *how* and *when* the stakeholders were involved, nor what the results or encountered obstacles were. This lack of information about the developmental process of PtDAs is unfortunate. Without an understanding of how and when to involve stakeholders, the improving the development and ultimately the implementation of future PtDAs can remain severely limited.

To address this gap in previous studies, we wanted to specifically involve users in our project. To do so, we chose to use the IPDAS development process model (53) – a novel stepwise approach to develop PtDAs that emphasises involving patients and health professionals in the process. Although this new comprehensive model provides an overview of the entire development process, it does not provide methods on *how* to best involve patients and health professionals. Therefore, we complemented the IPDAS development process model with methods derived from user-centred design (UCD) (54, 55), which we extensively described in Chapter 5.

For this project, user involvement started relatively early within the developmental timeframe and was operationalized in multiple ways. First, qualitative in-depth interviews were conducted with patients. The interviews consisted of two parts. As described in Chapter 4, during the first part, patients were asked about

their considerations and informational needs when deciding about DMARDs. In the second part of the interviews, the concept of a PtDA was introduced and patients were asked for their opinions about the value of such a tool and their wishes for the content and design. The results of this part are described in Chapter 5. The latter part was conducted with use of the UCD-method rapid prototyping (i.e. the use of paper prototypes) to assess patients' needs for specific content, functionalities and design of the PtDA.

The results of our studies revealed that patients felt the need for a complete overview of treatment options instead of only the option(s) suggested by the rheumatologist. Results also showed that patients wanted not only information with regard to clinical features (e.g. aim and working mechanism, time-to-benefit, manner of administration, potential side effects and risks, influence on fertility and pregnancy), but also information about possible consequences for their daily lives (e.g. restrictions on driving a vehicle and alcohol consumption and how to fit the treatment schedule into their daily lives).

With use of the paper prototype, patients critically reviewed our initial idea for the PtDA. While generally positive, patients were also able to provide us with useful insights for improvement. For example, patients were very positive about the list of the DMARDs' practical implications and, at the same time, were able to suggest additional items for a variety of categories. Another example is that most patients did not value the personal stories of peers and, therefore, these were excluded from the design.

After the qualitative in-depth interviews with patients, we organized group meetings with health professionals, as described in Chapter 5. In these meetings, health professionals were consulted for their perspectives on patients' decisional and informational needs and to determine the content of the PtDA. These meetings also aimed to determine the best way to embed the tool into daily clinical care. Especially insightful was our assessment of the perceptions held by health professionals about their patients' needs for information and decision support, particularly when compared to the data we had previously collected during the patients' interviews. While patients mentioned a desire for an overview of treatment options, health professionals were worried about overwhelming them. The result of our discussing this patient need with the health professionals was the development of a solution that supports the needs of both parties – which we called the 'referral card'. This referral card lists all DMARDs and the rheumatologist can indicate the DMARDs appropriate for the patient at any specific time. This solution provides patients with an overview of treatment options and rheumatologists the opportunity to guide patients through the plethora of options. Moreover, this co-creation and discussion created support among more sceptical health professionals for the development

and implementation of the PtDA. Health professionals who initially questioned the added value of a PtDA had less misgivings and were more willing to use it.

Finally, users were involved in usability testing of the PtDA (see Chapter 5). Based upon the results of the needs assessments, a working prototype of the PtDA was developed. While using this working prototype in real-time, both patients and health professionals were consulted to evaluate its usability. Consequently, they were able to help us identify and solve significant barriers to its usability.

This developmental process resulted in an innovative web-based PtDA consisting of four parts: (1) general information about SDM, inflammatory arthritis and DMARDs; (2) an application to compare particular DMARDs; (3) personal value clarification exercises; and (4) a printed summary of the user/patient's notes, preferences, worries and questions to bring to the rheumatologist to discuss. Screenshots of the PtDA are presented in the intermezzo of this dissertation.

The combination of the systematic IPDAS development process model and user-centred design methods were helpful for developing and implementing the current PtDA for two main reasons. Firstly, this approach led to a PtDA that met the needs of both the patients and health professionals, which helped to assure that it was integrated into the patient pathway and daily clinical care. Even though it was sometimes challenging to balance the stakeholders' needs and requirements alongside programming feasibility and available resources, this method allowed us to clarify user needs for content, design and distribution of the PtDA and incorporate these needs into the design. We feel that without the extensive involvement of users, many problems would only have come to light during the PtDA evaluation – or after – and it would have been more difficult to accommodate them. We, therefore, recommend that users should be extensively involved at an early stage in future PtDA development.

Secondly, the user-centred design methods we applied (i.e. rapid prototyping and think-aloud usability testing) proved to be valuable during the needs assessment and usability testing. Patients (but also clinicians) often have difficulty conceptualizing what a PtDA is and how it might look and function, which can limit them in expressing their needs. With the use of rapid prototyping (i.e. the use of paper prototypes during needs assessment), it was easier for them to express their wishes and needs and to give critical input. During usability testing, it can be challenging for researchers to interpret observed user behaviour. When participants express their thoughts aloud while using the PtDA, insights into patients' experiences can be gained at a detailed level. With this detailed understanding of patients' experience using the PtDA, our data proved to be rich as we were able to combine these user insights with our observation of their usage, the recording of screens, and a post-usage interview. To our knowledge, these methods are rarely used in PtDA development.

Therefore, we recommend using rapid prototyping and think-aloud usability testing in the development process of future PtDAs.

In conclusion, this project successfully demonstrated that user-centred design methods can be combined with the IPDAS process model to yield complementary results that inform the design and support adoption. It is our expectation that this design approach may ease the uptake of future PtDAs, and we, therefore, strongly recommend using such an elaborate stepwise approach in future PtDA development projects.

EVALUATION OF A PATIENT DECISION AID IN RHEUMATOLOGY

In the final phase of our project, the PtDA was evaluated among patients and health professionals, as described below.

Evaluation among patients

As described in Chapter 6, patients participated in a post-test only study. This study was conducted among 432 patients, of which 232 patients were in a historical comparison group and received care as usual and 200 patients were in the intervention group and received a referral to the PtDA. The results revealed that the PtDA was used by 57% of the respondents in the intervention group (a majority of 69 out of 123). Results of this study also demonstrated that we succeeded in fulfilling patients' needs: patients highly appreciated the PtDA and perceived it as useful, easy-to-use and helpful in the decision-making process. Comparison of the two groups showed that patients in the intervention group perceived a more active role in medical decision-making. Patients in the intervention group also perceived decisions to be more in line with their personal preferences. We found no differences between the groups with regard to satisfaction with the decision-making process, beliefs about medication, adherence, or trust in the physician. In general, these results correspond to the impact of other PtDAs, as was demonstrated in a recent systematic review (30).

Even though the PtDA was used by the majority of the respondents and was highly appreciated, room for improvement remains. For instance, the PtDA was mainly used by younger and highly educated patients. Although this is compatible with results of previous reported PtDAs, it should be investigated how the tool can be further adapted to the needs of older and lower educated patients. Additionally, some elements of the PtDA were used less than expected. Examples are the value clarification exercises and the summary. The value clarification exercises aimed to help patients clarify their preferences, worries and questions regarding initiation of

their medication. The summary is a document that lists patients' notes, preferences, worries and questions and can be downloaded and printed at the end of the PtDA. During the needs assessment and usability study, many participants expressed their appreciation for both elements of the PtDA (see Chapter 5). They felt that the exercises acknowledged that it is normal to have worries and questions. Patients thought that these exercises would also support them to express their concerns during their next consultation with their rheumatologist. They also mentioned that they would like to bring the summary to their consultation as a reminder and that the summary could help increase their confidence in their ability to participate in medical decision-making. During the evaluation of the PtDA, it became clear, however, that patients did not use these elements as much (see Chapter 6). At this moment, we do not know why patients did not use these elements as expected, despite their appreciation of these particular functionalities during the needs assessment. A recent study has shown that the main reason provided for non-completion of value clarification exercises was that patients had already made a decision (196). It is worthwhile to investigate whether their findings are applicable to our issue.

From the differences between the two groups, we can conclude that this PtDA *can* be a valuable aid in improving patients' perceived participation in medical decision-making about DMARDs. However, it must be noted that the differences between the groups were modest, and possible explanations for this may be ceiling effects or the limited quality of some of the psychometric instruments used (188). Nevertheless, when assessing the self-indicated impact of the PtDA, our results show a more positive effect. Patients who used the PtDA indicated that it helped them to gain insight into preferences, worries and questions, discuss things with their rheumatologist and make a decision about the medication.

Evaluation among health professionals

The PtDA was also evaluated among health professionals. All health professionals working at the rheumatology departments of the two hospitals where the PtDA was implemented were invited to participate in a focus group study (unpublished data; not reported in this dissertation). Participants included seven rheumatologists, four rheumatologists in training, one nurse practitioner and two rheumatology nurses. The study focused on use, appreciation, perceived impact, and future usage of the PtDA. This study revealed that health professionals commonly refer to the PtDA when patients have a high need for information and when there are equipotent treatment options. They also refer to the PtDA at times when the chance is high that the patient will need to change DMARDs in the near future.

Results showed that health professionals felt some situations are less suitable for referring patients to the PtDA. For example, some patients have consented to be

treated according to a research protocol. Some health professionals felt that in those cases there is no choice to make; other health professionals disagreed and felt that a patient always has a choice – even when they consented to be treated by a research protocol. Other situations where health professionals are less inclined to refer to the PtDA is when patients' language proficiency is insufficient, when patients previously have expressed that they did not want to participate in medical decision-making, and in decisions that require acute care. Finally, health professionals mentioned that they hesitate to refer newly diagnosed patients to the PtDA because they do not want to overwhelm them.

Health professionals highly appreciated the PtDA (average grade of 7.5 on a scale from 1 – 10) and noted various changes in their experience of the clinical encounter when patients had used the PtDA. Some health professionals felt that the decision-making takes more time and often requires an extra consultation. However, they also mentioned that this is not necessarily undesirable since the quality of the conversations often improves. For example, they mentioned that after having used the PtDA, patients asked more focused/explicit questions. Health professionals felt that this was because patients became aware of having a choice and were better informed and prepared. Some also mentioned that they learned about what mattered most to their patients, which in turn positively affected the patient-doctor partnership.

The majority of the health care participants clearly intended to refer future patients to the PtDA. Of the 14 participants, 9 definitely intended to refer future patients to the PtDA, 2 were unsure and 3 did not answer the question. These results are supported with current findings. Almost three years after conclusion of the project, the PtDA is still being successfully used and newly developed DMARDs have since been added to the PtDA by the health professionals, indicating that the PtDA sufficiently fits their needs.

In summary, both patients and health professionals highly appreciated the PtDA and have expressed mainly positive experiences with the tool. Therefore, we feel justified to recommend further implementation of the PtDA in other institutions.

PRACTICAL IMPLICATIONS AND SUGGESTIONS FOR FUTURE DIRECTIONS

The project described in this dissertation aimed to advance patient SMD participation in rheumatology with use of a web-based PtDA that was developed in close collaboration with patients and health professionals. We have translated the lessons learned from this project into practical implications and challenges for the future. In

this section we discuss how the PtDA can be further implemented, evaluated and adapted. In addition, we offer recommendations and address the challenges for future PtDA development and SDM research.

FURTHER DEVELOPMENT AND IMPLEMENTATION OF THE PTDA

To further develop the current PtDA, some of the wished-for attributes could be realized. These attributes were initially not implemented due to their technical complexity, limited time and finances, and privacy issues. A few elderly patients stated during the needs assessment that they did not want a computerized version of the PtDA because they feared that they lacked sufficient computer skills or did not have access to a computer/the Internet. Since it was only a few patients who stated this and because of limited time and finances, we did not develop a paper and pencil PtDA. Instead we chose to acknowledge this need by having a computer available for patient use in the hospital and guiding the patient through the PtDA decision-making process with the aid of a nurse. Since we conducted the needs assessment a few years ago, it might be worthwhile to explore whether patients still have this need.

A few patients suggested further tailoring of the content of the PtDA to the risk profile of the patient. In addition, health professionals suggested full digitalizatioin of the PtDA. This would imply additional functionality such as a digital referral to the PtDA (accompanied by a recommendation for appropriate DMARDs) and the ability for patients to send the summary with their notes, preferences, worries and questions to their health professional prior to the decision-making consultation. These suggestions from both patients and health professionals are technically interesting and may even improve the uptake of the tool. One approach might be to link the PtDA to the electronic patient record (EPR). However, one must first seriously consider the related ethical issues such as privacy. Another consideration is the insufficient evidence on differential efficacy and the safety of DMARDs; consequently, further tailoring of the content of the PtDA to the risk profile also remains a challenge for future research.

Based on the positive evaluation by patients and health professionals, it would be of interest to implement the PtDA in other rheumatology outpatient clinics. Since the PtDA has been tailored to the needs of the current users, it should be investigated whether the PtDA fits the needs of other users. With the expectation that the needs of the patients will not differ much, we feel that the main focus of this study should

be on the PtDA's implementation in the new institutions and its integration into the daily workflow of the health professionals working in those institutions.

In addition, it may be interesting to adapt the PtDA to other treatment decisions. Compared to most previously reported (web-based) PtDAs, the PtDA in this project encompasses many treatment options (176). The format we used to address this issue can be of interest to those researchers developing future web-based PtDAs. We chose to let rheumatologists refer patients to the PtDA and write a personal recommendation for appropriate medication options. The web-based PtDA, however, lists all medication options available out of respect for the patients' informational needs and rights. Patients are asked to select the medication that was recommended by their rheumatologists, but the information on other available medication options is freely accessible. This format may also be suitable for PtDAs that address other conditions, such as asthma or diabetes, that also have many available treatment options.

FURTHER EVALUATION OF THE PTDA

The focus of our evaluation of the impact of the PtDA on patient participation was from the patient and health professional perspective. Even though these perspectives are valuable, they only measure how patients and health professionals *feel* about their role in medical decision-making. In order to assess how the PtDA actually influences the decision-making dialogue between the patient and health professional, an observer perspective is necessary. A longitudinal study might reveal how SDM influences the clinician-patient partnership over time. In an ideal situation, one wants to follow newly diagnosed patients and their rheumatologists and evaluate their collaboration while the relationship evolves. It would then be interesting to take into account the patient, clinician and observer perspective. Recently, multiple initiatives have been taken to develop instruments that measure SDM processes from a dyadic or even a triadic approach (188, 197).

Our evaluation study showed that, despite our efforts to involve patients of various ages and educational levels during our PtDA development, the programme was mainly used by younger and highly educated patients. It would be interesting to explore why lower educated and elderly patients did not use the PtDA and how the PtDA might be adapted to their needs.

Additionally the results about usage showed that some elements of the PtDA were not used as much as we expected (i.e. the value clarification exercises and summary), despite these elements being highly appreciated during the needs assessment and usability study. To gain more knowledge about PtDA usage, it would

be interesting to analyse anonymous user data. For instance, researchers could log the amount of users' log-ins, page-views, and time spent on the PtDA, as well as patterns of usage (e.g. How do users navigate? Which elements and combination of elements are often used? When do users drop out?) and users' input (e.g. selected preferences, worries and questions). This might provide valuable knowledge about patients' (evolving) needs for PtDAs, which can be then used to adapt the PtDA and further improve its uptake, usage, usability and impact.

FURTHER DEVELOPMENT OF THE IPDAS PROCESS MODEL FOR PTDA DEVELOPMENT

The newly developed IPDAS development process model (53), which was empirically tested in this project, emphasises user involvement during PtDA development. However, due to a lack of evidence on best practices, the model omits a methodology for doing so. Our project shows that a combination of the IPDAS process model and user-centred design methods can yield complementary results that inform the design and support adoption. It is our expectation that this design approach may ease the uptake of future PtDAs, and we, therefore, strongly recommend including these methods in a new version of the IPDAS process model.

IMPROVE UPTAKE OF PTDAS

As mentioned in the introductory chapter of this dissertation, widespread adoption of PtDAs has not yet occurred (31). Why PtDAs are not adopted in daily clinical practice after positive evaluations in research settings, is still unclear (53). This project has shown that involving health professionals and patients during the initial stages of PtDA development can provide an opportunity to remove barriers against its adoption and implementation. Consequently, we recommend future PtDA developers also apply this approach. Furthermore, to enable the dissemination of learning experiences, developers of PtDAs should report on their development process and PtDA implementation, including the critical factors of its success and failure. In these reports, the focus should be on both quantitative outcomes (e.g. referral/usage rates) and qualitative outcomes (e.g. patients' and health professionals' experiences).

FUTURE SDM RESEARCH

Our project has demonstrated the complexity of factors influencing patient involvement and patients' preference regarding this topic. Consistent with previous research (19), the results from one of our qualitative studies showed that patients' preferences for involvement in medical decision-making can vary over time. For future research, we therefore recommend a quantitative and longitudinal study to show how patients' preferences regarding participation in rheumatology/chronic care may change over time. Changing preferences with regard to patient participation have been studied previously in end-of-life decisions (198, 199), but according to our knowledge this has not been studied in chronic care where patients establish a long term relationship with their health care professionals.

An intriguing outcome of our evaluation study was the difference between the impact of the PtDA measured by comparing the two groups as opposed to the self-indicated impact. The self-indicated impact of the PtDA showed a much more positive effect on patient participation compared to impact measured by the comparison of groups. From these results, one might wonder what is the best way to measure patient participation and SDM. Measurement of SDM is challenging due to several factors, as described by Scholl and colleagues in their systematic review about how to best measure SDM (188). First, one must differentiate between the measurement of the decisional process (e.g. choice awareness, information giving, deliberation, preference elicitation), outcome (e.g. satisfaction, regret) and surrounding elements of SDM (e.g. role preference, self-efficacy, trust in physician, health beliefs). Second, given the complexity and lack of conceptual clarity of what constitutes SDM, there are, to date, no general applicable primary measurement tools or standard outcome measures for SDM. Often the available instruments have not been properly tested for their psychometric qualities. Most of the instruments show satisfactory to excellent reliability, however, validity and sensitivity has not been sufficiently investigated. Finally, one has to decide from what perspective one wants to measure SDM, i.e. from the perspective of the patient, clinician or observer. Most instruments are self-reporting scales that assess the patient's perspective. To improve SDM research, guidelines should be developed that help researchers choose the right SDM measures for various situations. SDM experts, patients and health professionals should be involved in formulating these guidelines.

SDM is the target of educational and quality programs in many countries around the world. The Dutch government also took initiatives to improve patient-centred care and SDM through developing PtDAs, quality indicators and training programs ((104). Yet, even today, many patients are unaware that they have the possibility to participate in medical decision-making and often have difficulty conceptualizing

what this means. Since patients are also unaware of the existence of PtDAs, which have yet to be widely adopted, more initiatives need to be taken to make patients aware of their possibility to participate in medical decision-making and of the potential value of their input (39). Hospitals, for example, may implement the MAGIC (Making Good Decisions In Collaboration) program and its 'Ask 3 questions' approach as described in Chapter 1. This approach expressively encourages patients who attend outpatient clinics to ask their professionals questions (49). While most SDM interventions focus on the interaction between patient and clinician, another area to explore is how other health professionals (e.g. nurses) might become involved in the patient's decision-making. For example, nurses can frequently play an important supportive role in helping patients elicit preferences, weigh the pros and cons of their treatment options and prepare for the decision-making dialogue with the clinician. Their potential role is an interesting subject for future SDM research.

CONCLUSION

In conclusion, the project described in this dissertation has resulted in the successful development of a PtDA that has demonstrated its high potential to be a valuable aid in improving patient participation in rheumatology care. The supportive studies described in this dissertation provide valuable knowledge about patients' perceptions of SDM in the field of rheumatology. We have successfully verified that the IPDAS development process model can be combined with user-centred design methods, and this combination of methods was helpful in developing a user-friendly novel application and creating support for the adoption of the PtDA. By using these methods, the PtDA was able to fit the values of all the stakeholders and easily integrate into the patient pathway and daily workflow of the health professionals. Currently, our PtDA is still being successfully used and has been updated after completion of the project, a clear indication of the substantive value of our developed PtDA and developmental approach.

Dutch Summary | Samenvatting

Patiënten met gewrichtsontstekingsreuma zien hun zorgverleners ongeveer een uur per jaar. De rest van de tijd dienen zij zelf om te gaan met de grillige symptomen en bijwerkingen van de behandeling – zelfstandig en in hun eigen omgeving. Daarom is het essentieel dat patiënten betrokken zijn bij hun zorg en dat hun kennis, ervaringen en inzichten worden meegenomen bij het nemen van medische beslissingen. De keus voor een interventie zou het resultaat moeten zijn van gedeelde besluitvorming waarin zowel de patiënt als de zorgverlener(s) betrokken zijn (58, 61).

Gedeelde besluitvorming is tegenwoordig een breed gedragen ideaal en een essentieel kenmerk van kwaliteitszorg. In gedeelde besluitvorming komen de zorgverlener en patiënt samen tot een beslissing op basis van een open uitwisseling van kennis, ervaringen en inzichten. Keuzehulpen kunnen het proces van gedeelde besluitvorming ondersteunen. Ze ondersteunen de patiënt bij het inzichtelijk maken en communiceren van voorkeuren, zorgen en prioriteiten om zo een actieve, geïnformeerde deelnemer van het besluitvormingsproces te worden (50-52).

Onderzoek naar gedeelde besluitvorming heeft zich voornamelijk gericht op de algemene populatie, acute zorg en eenmalige beslissingen (bijv. screening of operaties) (7). Ook zijn keuzehulpen voorhanden voor uiteenlopende medische beslissingen en hebben zij hun effectiviteit bewezen (30). Er is echter nog maar weinig bekend over gedeelde besluitvorming en de rol van keuzehulpen in de chronische zorg. Dit was de aanleiding om te starten met een project rondom dit onderwerp in de reumatologie, zoals beschreven in dit proefschrift. Het eerste doel van dit project was inzicht verkrijgen in de gewenste en ervaren rol van reumapatiënten bij het nemen van medische beslissingen. Het tweede doel van dit project was het ontwikkelen van een keuzehulp in samenspraak met patiënten en zorgverleners. Het derde en laatste doel van dit project was om het effect van de keuzehulp op patiënt participatie te evalueren. Deze samenvatting biedt een overzicht van de resultaten van dit project zoals beschreven in dit proefschrift.

GEDEELDE BESLUITVORMING: WENSEN EN ERVARINGEN VAN PATIËNTEN

De studies beschreven in **Hoofdstuk 2 en 3** bieden inzicht in hoe reumapatiënten denken over hun betrokkenheid bij medische beslissingen. We hebben gebruik gemaakt van kwalitatieve en kwantitatieve onderzoeksmethoden. Voor het onderzoek beschreven in **Hoofdstuk 2** hebben we 29 patiënten geïnterviewd en gevraagd naar hun motieven om (niet) te willen participeren in medische besluitvorming en welke factoren hun gewenste en ervaren participatie beïnvloedden. Uit dit onderzoek bleek dat veel patiënten het moeilijk vonden om voorkeursrol te bepalen, omdat

ze hierover nog nooit nagedacht hadden, omdat ze moeite hadden om hun rol te conceptualiseren, of omdat ze zich niet bewust waren dat ze een keus hadden. Verder bleek dat de voorkeur voor participatie zou kunnen variëren door bijvoorbeeld de ernst van de klachten of de aard van de interventie.

Wanneer we patiënten vroegen naar hun *ervaren participatie* bij beslissingen over de reumabehandeling, vertelden de meeste patiënten dat beslissingen veelal ófwel in samenspraak met hun reumatoloog werden genomen, ófwel (voornamelijk) door de reumatoloog. Een enkeling had ervaren dat ze voornamelijk zelf hadden besloten. Een groep patiënten had graag meer inspraak gewild bij de besluitvorming. Ervaren barrières voor participatie waren gerelateerd aan de patiënt zelf, de zorgverlener of de context. Respondenten gaven bijvoorbeeld aan dat hun inspraak belemmerd werd omdat ze niet wisten welke vragen ze moesten stellen (patiënt-gerelateerd), omdat er maar één optie werd aangeboden (dokter-gerelateerd), of omdat er te weinig tijd was om te beslissen (context-gerelateerd). Al deze factoren konden patiënten het gevoel geven geen keus te hebben.

Deze, in hoofdstuk 2 beschreven, kwalitatieve studie leverde ons waardevolle informatie op over de behoefte aan patiënt participatie en de barrières die overkomen dienen te worden, waaronder de onbekendheid van dit concept. Om meer inzicht te krijgen in de wensen en ervaringen van een bredere groep patiënten met gewrichtsontstekingsreuma, werd een kwantitatieve studie uitgevoerd, welke is beschreven in **Hoofdstuk 3**. Ten behoeve van dit onderzoek vulden 519 patiënten een vragenlijst in. Het doel van deze studie was om op een meer kwantitatieve manier te onderzoeken welke rol patiënten wensen en ervaren, en wat de overeenstemming tussen deze twee is. Deze studie richtte zich op medische besluitvorming in het algemeen en ten aanzien van vier specifieke beslissingen over reumamedicatie: 1) starten met een traditionele reumamedicatie, 2) starten met het injecteren van Methotrexaat, 3) starten met een biologische reumamedicatie, en 4) afbouwen of stoppen met een reumamedicatie. Voor iedere beslissing werd gevraagd naar de gewenste en ervaren rol. Met deze informatie kon de overeenstemming tussen gewenste en ervaren rol berekend worden. Ook richtte deze studie zich op de tevredenheid van patiënten over het besluitvormingsproces en factoren die mogelijk gerelateerd zijn aan de gewenste en ervaren rol en de overeenstemming hiertussen.

Uit de resultaten over de gewenste rol bleek dat de meeste patiënten (59%-63%, afhankelijk van de beslissing) de voorkeur had om samen met hun reumatoloog medische beslissingen te nemen. Een aanzienlijk deel (25%-32%) wilde graag dat de reumatoloog de beslissing nam en een kleine, maar significante groep (8%-15%) wenste volledig autonoom te beslissen. Op basis van de resultaten uit de interviewstudie beschreven in Hoofdstuk 2 hadden we verwacht dat er tussen de

vier specifieke beslissingen variatie zou zijn ten aanzien van de gewenste rol, maar dit was niet het geval.

De resultaten over de ervaren rol lieten zien dat gedeelde besluitvorming frequent (26%-55%) werd ervaren en de ervaren rol kwam veelal (43%-62%) overeen met de gewenste rol. Echter, bij een paar specifieke beslissingen had een aanzienlijke groep patiënten (26%–54%) meer inspraak gewenst. Dit betreft in het bijzonder de beslissing om te starten met traditionele reumamedicatie – een beslissing die veelal aan de orde is bij nieuw gediagnosticeerde patiënten.

Ook bleek uit dit onderzoek dat patiënten minder tevreden over de besluitvorming waren wanneer zij minder inspraak hadden gehad dan gewenst. Echter, hun tevredenheid werd niet negatief beïnvloed als zij méér inspraak hadden gehad dan gewenst.

Uit deze onderzoeken kunnen we concluderen dat de meeste patiënten met gewrichtsontstekingsreuma graag betrokken zijn bij medische beslissingen en dat zij frequent gedeelde besluitvorming ervaren. Echter, er is ruimte voor verbetering – veel patiënten zijn zich niet bewust dat er een keuze is en dat zij een belangrijke rol kunnen vervullen. Anderen ervaren barrières om te participeren op het door hen gewenste niveau. Vooral bij de beslissing om te starten met traditionele reumamedicatie is er winst te behalen. Op basis van deze behoefte is er besloten een keuzehulp te ontwikkelen om patiënt participatie te vergroten bij beslissingen over reumamedicatie.

ONTWIKKELING VAN DE KEUZEHULP MET GEBRUIK VAN USER-CENTRED DESIGN METHODIEK

Ondanks de snelle toename van het aantal beschikbare keuzehulpen en hun bewezen effectiviteit, worden keuzehulpen nog maar weinig toegepast in de dagelijkse praktijk (31). De reden hiervan is nog onduidelijk. Mogelijk sluiten bestaande keuzehulpen onvoldoende aan bij de dagelijkse praktijkvoering of bij de behoeften en wensen van de eindgebruikers (zorgverleners en patiënten). Om implementatie te bevorderen is een systematisch en iteratief ontwikkelproces waarbij eindgebruikers op verschillende momenten betrokken worden essentieel.

In dit project hebben we gebruik gemaakt van het IPDAS ontwikkelingsmodel (53) – een nieuw stapsgewijs procesmodel wat nadruk legt op het belang van het betrekken van zorgverleners en patiënten bij de ontwikkeling en evaluatie van keuzehulpen. Ook al biedt dit model een overzicht over het gehele ontwikkelingsproces, het biedt geen methoden *hoe* zorgverleners en patiënten het best betrokken kunnen

worden. Daarom hebben we dit procesmodel gecomplementeerd met methodiek van *user-centred design (UCD)* (54, 55). Hoofdstukken 4 en 5 hebben betrekking op de ontwikkeling en evaluatie van de keuzehulp. Hoofdstuk 4 beschrijft onze exploratie naar overwegingen, zorgen en vragen die patiënten hebben wanneer ze voor de keus staan te starten met reumamedicatie. Inzicht hierin was nodig om de inhoud, de functies en het ontwerp van de keuzehulp te kunnen bepalen. Hoofdstuk 5 biedt een overzicht van het totale ontwikkelingsproces van de keuzehulp.

In ons project waren toekomstige gebruikers van de keuzehulp betrokken vanaf het begin van het ontwikkelingsproces. We hebben gebruik gemaakt van diverse methodes. Eerst hebben we interviews gehouden met 32 patiënten met gewrichtsontstekingsreuma die recent met hun reumatoloog gesproken hadden over het starten met reumamedicatie. Deze interviews bestonden uit twee delen. Zoals beschreven in **Hoofdstuk** 4, had het eerste deel als doel om inzicht te krijgen in de overwegingen en informatiebehoefte van patiënten wanneer ze voor de keus staan te starten met reumamedicatie. Uit de resultaten bleek dat patiënten behoefte hadden aan een overzicht van behandelopties (reumamedicatie). Ook bleek dat patiënten niet alleen informatie wilden over klinische aspecten van de medicatie, zoals werking, bijwerkingen en invloed op fertiliteit en zwangerschap, maar ook behoefte hadden aan meer praktische informatie en de potentiele invloed op het dagelijks leven – zoals restricties t.a.v. autorijden, alcoholconsumptie en hoe de toediening van de medicatie in te passen in het dagelijks leven.

Tijdens het tweede deel van de interviews werd het concept 'keuzehulp' geïntroduceerd. Patiënten werden gevraagd naar hun mening over de mogelijke waarde van dit hulpmiddel en hun wensen voor de inhoud en vormgeving. Hierbij is gebruik gemaakt van een papieren prototype, ook wel *rapid-prototyping* genoemd - een veelgebruikte UCD-methode. We hebben voor deze methode gekozen omdat gedeelde besluitvorming en keuzehulpen bij veel patiënten een onbekend concept is. Op deze manier spreekt het concept wat meer tot de verbeelding. De resultaten van dit deel van de interviews zijn beschreven in **Hoofdstuk 5**.

Met behulp van het papieren prototype konden patiënten een kritische blik werpen op ons initiële idee voor de keuzehulp. Patiënten waren over het algemeen vrij positief, maar gaven ook inzicht in punten voor verbetering. Zo waren patiënten bijvoorbeeld erg enthousiast over de lijst met praktische informatie over het gebruik van reumamedicatie en gaven zij diverse suggesties voor categorieën die aan deze lijst konden worden toegevoegd. Zo bleek ook dat veel patiënten geen behoefte hadden aan persoonlijke verhalen van lotgenoten en zijn deze uit het ontwerp gehaald.

Na dit behoeftenonderzoek met patiënten hebben we een onderzoek met reumatologen en reumaverpleegkundigen uitgevoerd (beschreven in **Hoofdstuk**

5). We hebben hen tijdens een semigestructureerd groepsinterview gevraagd naar hun deskundig oordeel over informatiebehoefte van patiënten en naar hun ideeën ten aanzien van de inhoud en vorm van de keuzehulp. Ook wilden we met deze bijeenkomsten bepalen hoe de keuzehulp het best kon worden ingepast in de dagelijkse praktijk.

Vooral de discussie rondom de informatiebehoefte van patiënten was inzichtelijk. Hieruit bleek dat terwijl patiënten behoefte hadden aan een overzicht van behandelopties en het kunnen vergelijken van reumamedicatie, de zorgverleners bang waren om patiënten te overspoelen met informatie. Het ter tafel brengen van dit verschil, resulteerde in de ontwikkeling van een oplossing die voldeed aan de behoeften van beide partijen – de verwijskaart. Deze verwijskaart bevat een lijst met alle beschikbare reumamedicatie en de reumatoloog kan hierop aangeven welke reumamedicatie voor de patiënt op dat moment geschikt zijn. Deze oplossing biedt de patiënten een overzicht van behandelopties en de zorgverleners de mogelijkheid om de patiënt te begeleiden door de stroom van informatie. Bovendien leidde deze co-creatie en discussie tot steun van zorgverleners die in eerste instantie ietwat sceptisch waren over de ontwikkeling en implementatie van de keuzehulp.

Gebaseerd op de resultaten van deze twee behoeftenonderzoeken en een literatuurstudie is het papieren prototype vertaald in een werkend prototype van de keuzehulp. Dit prototype is voorgelegd aan patiënten en zorgverleners om gebruikersgemak te evalueren, wat geholpen heeft bij het identificeren en verhelpen van diverse gebruikersongemakken (zie ook **Hoofdstuk 5)**.

Door de combinatie van het IPDAS ontwikkelingsmodel met UCD methoden, hebben patiënten en zorgverleners bijgedragen aan de ontwikkeling van een innovatieve online keuzehulp. Deze benadering bracht waardevolle inzichten in de behoeftes van de eindgebruikers die meegenomen zijn bij de verdere ontwikkeling van de keuzehulp.

De uiteindelijke keuzehulp bestaat uit: (1) algemene informatie over gedeelde besluitvorming, gewrichtsontstekingsreuma en reumamedicatie; (2) een applicatie om reumamedicatie te vergelijken; (3) een applicatie om voorkeuren, zorgen en twijfels inzichtelijk te maken; en (4) een te downloaden samenvatting van de notities, voorkeuren, zorgen, twijfels en vragen van de patiënt, welke meegebracht kan worden naar het volgende consult met de reumatoloog. Een overzicht van de inhoud en een aantal screenshots van de definitieve versie van de reumamedicatie keuzehulp (http://www.reumamedicatiekeuzehulp.nl) is te vinden in het **intermezzo** van dit proefschrift.

EVALUATIE VAN DE KEUZEHULP

Tijdens de laatste fase van ons project is het gebruik, de waardering en de effectiviteit van de keuzehulp op patiënt participatie geëvalueerd met behulp van een *post-test only* studie, zoals beschreven in **Hoofdstuk 6**. Deze studie is uitgevoerd onder 432 patiënten met gewrichtsontstekingsreuma. Gedurende twee jaar zijn alle patiënten die voor de keus stonden om te starten met (andere) reumamedicatie uitgenodigd voor deelname aan een vragenlijstonderzoek. Het eerste jaar ontving men standaard informatie over reumamedicatie (controlegroep; n=158). Het tweede jaar werd men door de reumatoloog verwezen naar de keuzehulp (interventiegroep; n=123).

Uit de resultaten bleek dat 57% van de respondenten uit de interventiegroep de keuzehulp heeft gebruikt. Jongere en hoger opgeleide mensen hebben de keuzehulp vaker gebruikt. De gebruikers vonden de keuzehulp nuttig, gebruiksvriendelijk en makkelijk te begrijpen. Verder waren gebruikers van mening dat de keuzehulp nieuwe informatie bevatte (70%), dat ze veel geleerd hadden (71%), dat de keuzehulp inzicht gaf in persoonlijke zorgen, twijfels, voorkeuren en vragen (70%), dat de keuzehulp geholpen had bij het gesprek (60%) en het maken van de beslissing (70%). In vergelijking met de controlegroep, hadden patiënten uit de interventiegroep een actievere rol ervaren bij het beslissen over starten met reumamedicatie. Ook ervaarden patiënten in deze groep vaker dat beslissingen in lijn waren met hun voorkeuren. Er werden geen verschillen tussen de groepen gevonden ten aanzien van tevredenheid, overtuigingen over medicatie, therapietrouw of vertrouwen in de zorgverlener. Hieruit concluderen we dat onze keuzehulp een waardevol hulpmiddel kan zijn om patiënten meer te betrekken bij beslissingen over reumamedicatie.

DISCUSSIE

Zoals in **Hoofdstuk 7** beschreven, kunnen we concluderen dat dit project heeft geresulteerd in de ontwikkeling van een keuzehulp met potentie om patiënt participatie in de reumatologie te verhogen. De ondersteunende studies beschreven in dit proefschrift bieden nieuwe inzichten in de perceptie van patiënten over gedeelde besluitvorming in de reumatologie. Daarnaast heeft dit project gedemonstreerd dat het IPDAS ontwikkelingsmodel gecombineerd kan worden met UCD methoden. Deze benadering was waardevol om te komen tot een gebruiksvriendelijke innovatieve applicatie en steun voor adoptie en implementatie van de keuzehulp. Met behulp van deze benadering sluit de keuzehulp aan bij de behoeften van alle eindgebruikers en de dagelijkse praktijk.

Hoewel dit project zich heeft gericht op patiënten met gewrichtsontstekingsreuma die voor de keus staan te starten met reumamedicatie, denken wij dat het ontwerp van de keuzehulp ook voor andere doelgroepen/beslissingen interessant zou kunnen zijn. Vergeleken met eerder ontwikkelde keuzehulpen, bevat deze keuzehulp veel behandelopties (176). De verwijskaart biedt de patiënten een overzicht van behandelopties en de zorgverleners de mogelijkheid om de patiënt te begeleiden door de stroom van informatie. Ook de interactieve applicatie voor vergelijk van medicatie kan een interessant ontwerp zijn voor de ontwikkeling van andere keuzehulpen. Daarnaast is vervolgonderzoek nodig naar het gebruik van de keuzehulp. Uit de evaluatiestudie bleek dat sommige elementen van de keuzehulp minder gebruikt waren dan verwacht. Deze elementen werden tijdens het behoeftenonderzoek en gebruiksonderzoek juist hoog gewaardeerd. Om meer inzicht te krijgen in het gebruik en de (veranderde) behoeften van patiënten zou het wenselijk zijn als log data zou worden geanalyseerd. De resultaten hiervan zouden gebruikt kunnen worden om de keuzehulp verder te ontwikkelen en de adoptie, het gebruik en de mogelijke impact te verbeteren.

De evaluatiestudies in dit project hebben zich gericht op de impact van de keuzehulp vanuit het perspectief van de patiënt en de zorgverlener. Ook al zijn deze perspectieven enorm waardevol, ze bieden alleen inzicht in hoe de patiënt en de zorgverlener hun rol in het beslissingsproces *ervaren*. Om op een meer objectieve wijze te evalueren hoe de keuzehulp het beslissingsproces beïnvloedt is het perspectief van een waarnemer nodig. Een longitudinale observatie studie zou zelfs inzicht kunnen bieden in hoe gedeelde besluitvorming de relatie tussen zorgverlener en patiënt veranderd over tijd.

Patiëntgerichte zorg, patiënt participatie en gedeelde besluitvorming staan hoog op de agenda van beleidsprogramma's voor verbetering van de zorg. Ook in Nederland zijn diverse initiatieven genomen. Echter, uit onze resultaten van onze onderzoeken blijkt dat veel reumapatiënten zich niet bewust zijn van de mogelijkheid om te participeren in medische besluitvorming. Ook vinden zij het moeilijk hun rol hierin te conceptualiseren en van het bestaan van keuzehulpen had menigeen nog nooit gehoord. We verwachten niet dat dit anders is bij andere aandoeningen. Keuzehulpen en algemene trainingsprogramma's kunnen hierbij helpen. Naast het implementeren en integreren van keuzehulpen in de dagelijkse zorg dienen ook algemene trainingsprogramma's gericht op bewustwording van het belang van patient participatie gestart te worden. Zorginstellingen zouden bijvoorbeeld het MAGIC (Making Good Decisions In Collaboration) programma en het 'Stel 3 vragen' campagne kunnen initiëren. Deze campagne stimuleert patiënten om vragen te stellen aan hun zorgverleners en op deze manier betrokken te raken bij het beslisproces (49). Daarnaast is de rol van andere zorgverleners dan de arts

vaak onderbelicht in onderzoek naar gedeelde besluitvorming. Verpleegkundigen zouden bijvoorbeeld een belangrijke ondersteunende rol kunnen spelen bij het implementeren van keuzehulpen. Zo zouden ze ook samen met de patiënt de voorkeuren, zorgen en vragen in kaart kunnen brengen, voor- en nadelen van de behandelopties afwegen en de patiënt voorbereiden op het gesprek met de arts. Hun potentiele rol is een interessant onderwerp voor verder onderzoek.

Samenvattend heeft het onderzoek in dit proefschrift een bijdrage geleverd aan het inzichtelijk maken van de perceptie van patiënten op gedeelde besluitvorming in de reumatologie en de factoren die dit stimuleren en belemmeren. Een innovatieve keuzehulp is ontwikkeld die patiënten ondersteunt bij het inzichtelijk maken van voorkeuren, zorgen en twijfels zodat zij samen met hun reumatoloog een weloverwogen beslissing kunnen nemen over het gebruik van reumamedicatie. Deze keuzehulp wordt door zowel patiënten als zorgverleners als prettig en nuttig ervaren. Op dit moment (2017) wordt de keuzehulp nog steeds gebruikt en is sinds afronding van het project al meerdere malen vernieuwd met nieuwe medicatie, wat een indicatie is voor de potentiele waarde van deze keuzehulp en de gekozen strategie voor ontwikkeling.

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About the author

Ingrid Nota was born in Appelscha, The Netherlands, on December 10, 1983. In 2002 she finished secondary education at the Stellingwerf College in Oosterwolde. In 2007 she received her bachelor degree in occupational therapy and started working as an occupational therapist specialized in rheumatology at the University Medical Centre Groningen (UMCG). Parallel to her work at the UMCG, she started the master program Health Sciences / Health Care Management at the University of Twente. Ingrid received her Master of Science degree in January 2009 and immediately started working as a PhD candidate at the department Psychology, Health and Technology of the University of Twente. Her research focused on the perspective of patients on shared decision-making in the setting of rheumatology care and the development of a patient decision aid (PtDA) in co-creation with patients and health professionals. As part of the stepwise user-centred development of the PtDA a series of studies was conducted, which are described in this dissertation. Ingrid currently works as a lecturer at the University College Twente.

List of publications

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