

CHALLENGES OF PATIENT PARTICIPATION IN RHEUMATOLOGY RESEARCH

*We need humanities to remind us
what is truly important and why we do what we do.*

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1. Being a patient in a medical system

There is wide spread call for patient centered health care in the western world. Many initiatives to humanize care institutions occur in response to a continuously increasing intrusion of medical technologies, including complex drug regimes. As David Hunter aptly points out: “We struggle with institutions and systems that are no longer fit for purpose”.¹

This process has been described by the German philosopher and sociologist Jürgen Habermas as the institutionalization of the ‘life-world’ by the ‘system world’,² a process that created a supply-driven environment for health care and health research. Professionals are dominating the scope, structure and practices of these systems and they are difficult to change.³ As a consequence people with chronic conditions are forced to adjust their societal role and identity to the expectations of the environment, often leading to loss of work, loss of social relationships and the emergence of new interdependencies with others, family, friends and health professionals.⁴ Talcott Parsons has given some clear features of the ‘sick role’ of patients that are still valid. He emphasizes the helplessness, the lack of knowledge, the lack of (technological) competences and emotional distress.⁵ Patients become dependent on professionals who are supposed to know how the illness can be treated and of a society that has separated medical practice in distinctive roles and instrumental values, and imposes its own normative expectations.

The challenge of the patient journey is to relate to the unbridled medical progress and to preserve a personal sense of identity. Many patients want to retain control over their own lives and, despite their vulnerability and the often unpredictable nature of their disease, stay independent. They want to retain an influence in shaping their environment according to their own needs and values. The call for self-determination can clearly be seen in the western world where new patient roles other than that of passive health consumer have evolved. One new role is that of the informed and autonomous patient who takes responsibility for an active involvement in the decision making process that is centred around well-being rather than only the illness.⁶ This patient treasures autonomy and expects health professionals to provide information, guidance and support that will enable the patient to keep that autonomy for as long as possible.

2. My own stance and locating the thesis within the science traditions

To better understand the opportunities for patients to contribute to more patient oriented health care and health care research this thesis focuses on the experiences and perspectives of both patients and professionals in one particular medical field, namely the area of rheumatology. This is the area where my experiences as a patient with arthritis

are deeply rooted, and from where my journey started. This thesis deals with issues of patient participation relevant to social scientists, medical researchers and patient research partners. My work is deliberately trans-disciplinary using both experiential knowledge as well as expert knowledge from various disciplinary fields. The thesis intersects with the traditions of the medical sciences, the social sciences and the humanities, and accompanying styles of representation. For this reason I briefly review the style of this chapter and that of other parts of this book – a style that might sometimes be different from what some would expect from a ‘*scientific*’ manuscript.

The representation style may swap from the active to the passive tense. It might be that some paragraphs provide information that is known to one group of readers but not to the other. And some chapters are written using the personal pronoun while others are not, dependent on the requirements of a particular science tradition and journal. Sometimes I use the singular first-person when I want to introduce a personal experience or reflection; revealing instead of hiding my identity as patient-researcher behind a neutral and distant narrative. This is an approach often applied in anthropology and sociology that acknowledges that my position as both a patient and a researcher influences the process of inquiring and subsequently the findings and the interpretations.^{7 8} In chapter 9 I will reflect in more detail on the consequences of my dual identity as patient and researcher and the methods I adopted to deal with the potential risks of subjectivity.

Finally, although I regularly use the ‘I’-form, I want to emphasize that this thesis is the result of an intense and ongoing dialogue with prof. Tineke Abma and prof. John Kirwan. Over the years they were actively involved in my journey through the world of research in different roles, even though it was not until 2009 that they officially became my academic mentors to guide this PhD study.

In the last decade the role of patients in rheumatology research has become more important. In this dissertation we will study this collaborative role of patients on four levels: local, national, European and international. One problem is that there is no standard or established methodology for evaluating patient participation. Second, as both patients and their families as well as health care professionals, researchers and policy makers are affected by and relevant for the involvement of patients many stakeholder perspectives should be included in the evaluation of patient participation. There are several strands to evaluation that respond to and involve different stakeholder views. In this thesis we have chosen to use a responsive approach to evaluation because this approach aims to foster a dialogue among stakeholders in order to gain a more informed

and richer understanding of the object of evaluation. Since patient participation affects many stakeholders responsive evaluation seems the most appropriate design.

Essential concepts of the theoretical framework in which this thesis is set are derived from the theory of responsive evaluation and Habermas' theory of communicative action.^{2 9} We will evaluate these concepts in the context of patient participation in rheumatology research. Responsive evaluation is an established scientific methodology that allows the inclusion of multiple perspectives through dialogue. It recognizes the fact that all participants, patients as well as health researchers, should have a say in the evaluation process. This methodology values the dialogue between different stakeholders as an essential vehicle for communication. The aim of the dialogue is to establish mutual understanding, something that can only be achieved, according to Habermas, in the ideal situation where power is symmetrically shared between all stakeholders and all have equal chances to ask questions and to share their views. When applied in different contexts of medical research, this approach may result in findings that, analyzed and interpreted on an aggregated level, may provide a more informed understanding of the phenomenon of patient participation, both in terms of process and impact.

In this chapter I will first introduce the context of rheumatology care (1.3 and 1.4) and rheumatology research (1.5). Then I will elaborate on the relevance of patient participation in rheumatology research (1.6) and outline the challenges for research in the light of the overall literature (1.7). Finally I will present the research objectives (1.8), the methodology of responsive evaluation (1.9) and the four level studies that form the basis of this thesis (1.10).

3. Living with arthritis

Rheumatology covers a broad spectrum of more than 200 musculoskeletal and rheumatic disorders. They can be divided into autoimmune diseases such as rheumatoid arthritis (RA), mechanical joint diseases such as osteoarthritis, and soft tissue diseases, such as fibromyalgia. They all may cause pain, swelling, joint stiffness, fatigue and loss of function, in some cases leading to significant disability.^{10 11} Other features vary from person to person and may include skin or organ involvement. Most rheumatic conditions are progressive, eventually making even simple everyday tasks difficult. Therefore they are accompanied by a considerable decrease of health related quality of life.¹² The costs involved entail a huge economic burden on the health system.^{13 14}

The heterogeneity of signs and symptoms complicates the diagnosis, often causing delays to the start of appropriate treatment.¹⁵ Medical management is different for each condition and often requires a multidisciplinary approach. For many conditions, at the

core of the treatment are medicines including pain killers, non-steroid anti-inflammatory drugs (NSAIDs), corticosteroids and other disease modifying anti-rheumatoid drugs (DMARDs). Some conditions cannot be adequately controlled by drug therapies and require non-pharmacological interventions such as surgery, physical exercises and lifestyle changes. The introduction of biological agents (*bDMARDs*) has substantially changed the landscape of rheumatology for many patients with severe uncontrolled inflammatory conditions, albeit not all. The prognosis of moderate and severe RA as a debilitating disease, leading to functional disability on the long term, can now be reversed into that of a controllable disease that might even result in a state of remission. Yet there are still individuals with rare complications and sensitivities to biologicals who do not benefit from these innovations.

The advent of effective treatments has also generated new concerns, such as the acceptability of high-tech manufactured medicines, the severity of potential side effects and the unknown consequences on the long term. In addition to these sometimes ethical considerations, political and economic concerns emerged about the affordability of expensive agents in an era of economic downturn. Recent papers showed that patients with RA in lower income countries have less access to *bDMARDs* across Europe¹⁶ and have lower health outcomes worldwide.¹⁷ They prominently demonstrate that there are growing inequalities across countries and continents.

In conjunction with the progress of biomedical research, and congruent with the world wide movement towards more patient centred health services, there is a tendency to construct modern ways of disease management in terms of self-management and shared decision making. Here, the authority of the health professional is no longer untouchable. Based on the finding that a *strategic* approach to the management of rheumatic diseases leads to better outcomes,¹⁸ recommendations for ‘tight control’ in which the patient plays an active role have been developed.¹⁹ Unique for this initiative is the intense involvement of patients in the process of developing these recommendations that clearly reflect the increasing wish of many patients to stay autonomous, take responsibility for their own disease and to be part of the decision making process.²⁰ To facilitate self management, adherence and making shared decisions, good patient information and education is necessary. The role of specialized nurses in this area is increasing.

4. Unmet needs in rheumatology care

In a world of advanced health technologies, patients’ identities, values and relationships are increasingly challenged. The so-called patients’ perspective is heterogeneous. This means that not all patients automatically conform to the opportunities and consequences

of new technologies and policies. Rheumatic diseases can have a huge impact on daily life. Long term use of medications can cause many problems and uncertainties. They not only come with sometimes serious adverse events or unexpected interactions with other drugs, they also give rise to many questions about the safety of the drugs on the long run and the occurrence or prevention of other diseases. For young people a child wish, pregnancy or breast feeding may cause serious dilemmas if that means they have to stop medications. Young people often worry about the possible hereditary nature of their disease and about their future abilities to take care of children and family life. The management of the disease may require difficult decisions that go far beyond the choice of drug treatment. Early on or later patients may have to face joint surgery. In the presence of severe erosions or structural damage it is difficult to weigh the pros and cons of orthopedic interventions that have almost always far reaching consequences for daily life.

In addition to the physical symptoms of the disease, patients are confronted with the psychological impact of their disease. These uncertainties relate to fears about the future and the ability to continue working, housekeeping, playing sport or taking part in leisure activities (hobbies). In times of flares people may become depressed as a result of anxiety, diminishing autonomy or unstable relationships. Finally, patients may have to face social consequences such as isolation, loss of income and societal prejudices such as unbelief among other people when the illness is invisible, or mistrust if people cannot understand that symptoms of the disease may vary from day to day. Some patients prefer alternative medicines while others feel forced to try complementary treatment by well meaning advice from friends or family. Not all physicians support the use of alternative treatments which may cause tensions in the patient-doctor relationship. That relationship may also be disturbed by many other reasons such as not feeling understood or taken seriously, lack of attention for important things other than the immediate features of the disease, or a paternalistic attitude.

In finding answers to these questions patients rely in the first place on the expertise and support of health professionals with whom they often cherish a long standing relationship. Trust and continuity of care are important conditions for patient satisfaction, leading to better adherence to treatment recommendations and ultimately to better health outcomes. However, these conditions do not always exist and expectations of patients are not always met. The most common unmet expectation of patients is adequate information.²¹

Unmet needs are often caused by poor communication and ineffective relationships. Especially in times of commercialization of health care and large scale reductions of

health expenditures, there is a growing demand on and obligation assigned to patients to become more responsible for their own health. However, skills for collaboration in the context of health care are often lacking and patients as well as professionals find it difficult to communicate and act differently compared to what they have learned in the past. In contrast to the traditional patient-expert relationship, new forms of collaboration are perceived as a relational responsibility between the patient and the health professional. This is based on the proposition that an enhanced role for patients in health care decisions is essential to optimize clinical outcomes.²²⁻²⁴ For patients who want to keep their independence and are interested to learn to manage their own condition, and for health professionals who see patients as autonomous actors, this creates the challenge of building horizontal relationships based on respect and equality. By a process of 'proto-professionalization' the patient can learn methods of self-management and becoming part of the decision making process. This process is often referred to as 'empowerment'. The health professional will open up the consultation room for alternative ways of providing information. In an open conversation they may discuss the pros and cons of a new treatment option or the need for other changes. The aim of such a dialogue is to achieve a mutual understanding that enables the patient to make well informed decisions.

Despite these intentions, in many relationships the communication is still far from ideal and hampers a sincere dialogue that takes personal values, fears and priorities into account. For patients who have not learned to cope with uncertainties in life or have prejudices about the interests of health professionals, or for health professionals who are not able to depart from traditional expert behavior, it may be difficult to find common ground for a successful relationship.

5. State of the art of rheumatology research

Research in the field of rheumatology is diverse and expanding. The exact cause of many diseases is still unknown. This makes experts believe that, for instance, inflammatory diseases are the result of a combination of multiple factors such as genetics, environment, hormones and the human response to stressful events. Since the arrival of biological agents, more clinical research is undertaken, in many cases sponsored by the pharmaceutical industry. In the Netherlands all academic departments of rheumatology have specialized their research focus with respect to either particular rheumatic conditions or clinical interests such as early diagnosis, work participation or basic research. Funding is obtained through different sources and can come from the pharmaceutical industry, government (nationally or internationally), intermediate bodies and public and private health charities. Most of the funds are invested in basic or clinical

research although there is a growing awareness of the importance of psychosocial and implementation research.

Evidence based medicine in rheumatology research has, as in all other fields of medicine, privileged experimental study designs based on rigorous laboratory testing and methodological sound clinical trials. There is a strong tendency in psychosocial research to use methodologies derived from biomedical research, including often quantitative research designs and highly sophisticated statistical procedures. The increasing complexity of research findings, and the format in which they are published, reflect the distance between the world of science and the daily life of patients.

People are becoming more empowered through the internet and want to be informed about the outcomes of scientific research. From a provisional survey among members of the Dutch arthritis patient magazine (response: n=365), most of the respondents could not tell what had been discovered in the study in which they participated. More than half of the survey participants complain that they have not received any information after completion of the study.²⁵ Decreasing trust in the research community has consequences that go beyond research itself. It not only creates pressure on the recruitment of study participants for trials and the willingness to sponsor scientific research, it also may lead to an increased non-compliance with treatment regimes among patients or push them into the area of alternative medicines. Many publications nowadays highlight the discrepancy between the perspectives of patients and professionals concerning domains that are important to evaluate the quality and effectiveness of health care interventions.²⁶⁻²⁸ These publications show the urgency of the question how to deal with these differences. Evidence is emerging that involving patients in research can bridge this gap and contribute to a more transparent and democratic organization of research and more relevant and broadly agreed research outcomes.^{29 30}

6. Relevance of patient participation in rheumatology research

The opinions, priorities and preferences of patients have become more prominent in the improvement of rheumatology health care. This has been the result of a process of emancipation of the 1970s.³¹ Patients started to establish local self-help groups. Not long thereafter patients became conscious of the fact that advocacy on a national level was necessary to raise awareness of the impact of rheumatic conditions, lobby for better health care services and influence national health policies. In the Netherlands, for instance, in 1976 six regional arthritis patient groups established the Dutch league of arthritis patient associations (Reumapatiëntenbond). On an international level 18 national arthritis organizations joined the European League Against Rheumatism (EULAR) as

full members of the organization with voting rights. They created their own standing committee, unifying 36 patient-led organizations in 2013. In the United States the Arthritis Foundation became an important advocacy body for people with rheumatic diseases. They supported and adopted the first arthritis self-management training ('Challenging Arthritis') that would become the fundament of many other educational health programs for people with chronic conditions.³²

From the start of 2000 the role of patients in improving the quality of health care services became more diverse.³³⁻³⁴ Patients started to contribute to the development of guidelines,³⁵ education of health professionals,³⁶ improvement of health services,^{37,38} and influencing health care strategies.³¹ On different continents patients also tried to actively collaborate with researchers. This process started in Canada where researchers of the Cochrane Musculoskeletal Group already started in 1993 to engage with 'consumers' in the design, conduct and dissemination of systemic reviews in the area of musculoskeletal diseases.³⁹ The arthritis community in Canada has been instrumental in developing a network of consumers that helped the knowledge transfer from the world of scientific research to the individual person with arthritis.

The first mentioning of patients as collaborative partners in research are reported by Sarah Hewlett when she wrote about patients contributing to scientific projects at the rheumatology department of the University of Bristol.³⁴ During later EULAR congresses Hewlett and John Kirwan have looked back at the lessons learned over the last decade.⁴⁰ ⁴¹ The Bristol group has a long tradition in patient participation⁴² and has been leading in introducing the concept of patients as collaborating partners at the Outcome Measures in Rheumatology conferences (OMERACT).⁴³ Since 2002 OMERACT has evolved into a paradigmatic example of the transformation from physician dominated research into collaborative partnerships with patients⁴⁴ and has inspired many other initiatives. Patients leaving OMERACT started to initiate national networks of patient research partners in Sweden^{45,46}, Oslo,^{37,38,47} and the Netherlands,⁴⁸⁻⁵⁰

On a European level EULAR has been groundbreaking in its efforts to integrate the patient perspective in all parts of the organization. In 2007 EULAR facilitated the development of a set of recommendations for the incorporation of patient representatives in scientific projects. These recommendations were accepted in 2010 and published in the year thereafter.⁵¹ EULAR is the only European medical umbrella organization where patients are officially represented in almost all parts of the organization, including the Executive Committee, Scientific Committee and the General Assembly.

In contrast to EULAR the annual congress of the ACR (American College of Rheumatology) does not have a patient program. Patient representatives are not part of

the organization, and the role of patient research partners has not been acknowledged by researchers to the extent that is seen in Canada and the west of Europe. The literature does not provide any information on the involvement of patients in rheumatology research in other continents.

Despite the above mentioned initiatives, in practice patient participation is still often limited to consulting patients as a one off event, depending on the willingness of an individual researcher. Researchers still struggle with many questions around the recruitment and selection of eligible patients: How to distill relevant information out of patient stories and how to guarantee that this information is representative for the entire population? How to involve patient participants in another way than respondent or study participant and how to support them adequately and avoid overburdening? How to provide acknowledgement or compensation and how to deal with ethical issues such as confidentiality? And finally, how should researchers report on all these matters? Because of all these unanswered questions but also for practical reasons researchers often rely on their traditional way of working, and by doing so, insufficiently capturing the patients' perspective.

It is our assumption that the added value of direct patient participation is access to the life world of the patients through their experiential knowledge and the opportunity to integrate this knowledge with that of professionals. It is based on the ethical imperative that patients should have a say in processes that will ultimately influence their own lives and, at the same time, empower them.

7. The concept and challenges of patient participation

There are many initiatives that tried to make health research more patient oriented by active engagement with patients. The arguments to justify these initiatives are threefold⁵²: First, patients have the right to have a say in the development of health research that will affect their daily life. It is not only a fundamental democratic right, it is also the right of tax payers to insist that researchers are transparent and accountable for how they spend public money in the interest of particular target groups. In addition to this moral imperative, there are substantial arguments to believe that the quality and relevance of research outcomes improve when patients become involved. If patients are involved in the identification and prioritization of research themes, in the conduct of the research and in the analysis and dissemination of the collected data, it is expected that the final outcomes will better meet the needs and preferences of patients in their daily life. Finally, the strategic or instrumental argument refers to the use of patient participation to enhance the chances for fund raising or for better implementation of research outcomes.^{39 53 54}

Despite the many initiatives in this area, there are not many publications that have looked at the conditions for effective and sustainable engagement with patients as collaborative partners. When researchers report about the form and effectiveness of participation, it often turns out to be incidental and on a relatively low level such as consultation or ad hoc advice.⁵⁵ Karen Schipper (2012) confirms in her thesis *Patient participation and knowledge*: “no hard figures are available. Involvement as an equal research partner with shared control is still rare and publications on this kind of collaboration are scant”.⁵² She concludes that we actually lack an unequivocal, unambiguous concept of participation in terms of roles, tasks and responsibilities shared between patients and professionals.

The concept of participation originates from the middle of the 20th century, when Sherry Arnstein introduced an hierarchical model for citizen participation that distinguished eight levels of power, ranging from non-participation through consultation, partnership to citizen control.⁵⁶ The lower levels are forms of tokenism while the higher ranks represent increased power and influence.

Although the model has been fundamental in the development of participatory research, it has been criticized for its normative, static and one-dimensional construct. Abma not only questions the normative character of the model by pointing at its potential polarizing impact, but also the lack of consideration of the development and quality of the process of participation.⁵³ In her thesis Janneke Elberse confirms the normative nature of the model by emphasizing that “not every patient aspires to be involved on a high level”.³ She refers to the finding of Francisca Flinterman that it is more important to ensure that the level of participation suits a particular situation.⁵⁷ In a study about participation of older people in residential care homes Vivianne Baur states that the model “does not represent the complexity and multidimensional nature of client participation”.⁵⁸ Jonathan Tritter points out that the model does not provide any suggestions for organizing patient participation in an effective manner.⁵⁹

The above mentioned shortcomings of the model have encouraged the development of more dynamic approaches to participation. The European Patient Partner project⁶⁰ and many of the above mentioned researchers have adjusted the Arnstein model to be used in the context of health care and health research with less normative connotations compared to the original eight rungs.^{55 61} These modified models differ in number of categories, terminology and scope. Some categories entail different levels of influence while others refer to distinct role for patients. Many models emphasize the imbalance in the decision making process and aim to change existing power relations. They promote horizontal relationships between patients and researchers which can be found, according to the authors, in forms of partnership such as co-researcher or patient (research) partner.

Here the asymmetric relationship has been replaced by a relationship that is, according to Habermas, free from unnecessary domination based on tradition or authority only.

The benefits of these new understandings of participation are that they take into account that the desired outcome is often dependent on the quality and dynamics of the relationship between patients and professionals. The above mentioned authors have formulated conditions for genuine partnerships and how these can be fulfilled in practice. An important condition is that patients should be allowed to learn and grow. Over time patients may develop role preferences that meet their individual competences and ambitions.³ Some prefer to act as information-givers, other like to be, and can be, involved in all phases of research and carry out a variety of research tasks. The literature describes tasks such as interviewing fellow patients,⁵² co-moderating focus groups,⁵⁵ analyzing data,⁶² and developing systematic reviews and disseminating results.³⁹ The level of participation may vary and may develop from limited involvement in a single consultation process to structural involvement as co-researcher.⁶²

Other conditions that support a transition towards more patient oriented health research, are acknowledgement of experiential knowledge as a valid source for scientific research, involvement of patients in an early stage, removing restrictions caused by the traditional, rigid research system, development of inclusion strategies, a trans-disciplinary approach of health care research that looks at patients from a holistic perspective and implementation of patient participation as a requirement by funding organizations.^{57 63 64}

In this thesis patient participation is perceived as a dynamic process that needs to be actualized in daily practice rather than a static state of power distribution. We look at participation from the theoretical construct of dialogue and partnership. We believe that patients have not only limitations, but knowledge, values and competences that are relevant for improving health care services and health research. These contributions are often overlooked or trivialized. However, experiential knowledge is not inferior to the scientific knowledge of professionals as illustrated by Karen Schipper in her studies in the field of chronic kidney disease and acquired brain injury.⁵² She has clarified the unique character of experiential knowledge and shown how this individual and subjective knowledge develops into expert knowledge by a process of collaboration between patients and researchers. The experiential knowledge of patients transforms (or better: expands) over time into practical knowledge and finally into expert knowledge. Practical knowledge is perceived here as a shared patient perspective: the often implicit

experiential knowledge that is hidden in authentic patient stories is made explicit and shared with fellow patients. The result is an inter-subjective expert knowledge.

Professionals develop their knowledge in a reversed order compared to patients. First they develop their expert knowledge during their academic training. Then, after entering clinical practice, their knowledge is expanded with practical knowledge and finally with experiential knowledge. This perception of knowledge development does not place experiential knowledge opposite of expert or evidence-based knowledge. It emphasizes the complementary relationship between both sources of knowledge.⁶⁵ In fact, professionals as well as patients possess all three kinds of knowledge, although each in a different composition. When they are brought together in an open dialogue, they can ultimately lead to innovative perspectives and consensual solutions for today's dilemmas. This process is known as "a melting of horizons".⁶⁶ The practical conditions that contribute to this process are complex and not well developed.

8. Main research questions

Implementing patient participation in rheumatology research aims to establish more sustainable and equal relationships between professionals and patients, and will hopefully lead to a better integration of the experiential knowledge of patients in the development of health research. The involvement of patients as collaborative partners is expected to lead to changes in the conduct of research and increase its relevance. This collaborative partnership is not self-evident. How patients and professionals really value the quality of their collaboration, and whether they will succeed in starting a dialogue that will lead to a better mutual understanding and a better integration of the patient perspective in rheumatology research, is still insufficiently known. We also lack convincing scientific evidence about the effectiveness of patient involvement. Our main research question is therefore how patients and professionals can collaborate equally in the context of scientific research. In other words: How can they create conditions for an open and meaningful dialogue in which both groups have equal opportunities to provide their own knowledge and perspectives, leading to enhanced mutual understanding and a change of research practices?

To answer the main research question we conduct a multiple case study in the field of rheumatology. We will study four contexts where patients collaborated actively with researchers, and explore conditions that constitute genuine and sustainable partnerships. The outcome of partnerships between patients and researchers and the issues that emerge during the development of partnerships cannot be predefined. Partnerships may lead to patient involvement in setting the research agenda, the research design or the co-

production of knowledge in research projects. In the longer run partnerships may become sustainable when patients are involved on a more structural basis, and when values for participatory research become part of the research culture. Essential in our approach is the idea that these values and the actual outcomes of the collaboration will be determined by all stakeholders. The process of eliciting or constructing these values and outcomes takes place during the case studies by reflecting on stakeholders' experiences with the new partnerships.

9. Research design: multiple case study

This thesis follows a naturalistic approach of multiple case research which is appropriate to explore and understand the way patient participation and partnerships evolve in a variety of research contexts that are considered unique. According to Robert Stake case study research aims to identify emerging issues that are characteristic for a phenomenon in a specific, bounded context.⁶⁷ The purpose of the research is to better understand the diversity and importance of a phenomenon in a particular case from the perspective of the participants. To that end a case is not studied in isolation but from a holistic point of view, using a variety of research methods. A case study includes not only individual experiences and the meaning that participants give to developments that surround them, but also the context. The aim of a multiple case study is to reveal more general issues and patterns despite the uniqueness of the singular contexts.⁶⁸ Rather than answering a set of predefined research questions, naturalistic case study research will result in a description of issues that are relevant according to the participants in that context, and that may ameliorate our understanding of these issues in other contexts.

The various case studies reported in this thesis are guided by the theory and methodology of 'responsive evaluation' to include multiple stakeholder perspectives and to foster dialogue. As outlined in paragraph 1.2 the evaluation of research strategies or research policies can follow different approaches. In traditional evaluations the assessor evaluates from a distant point of view and acts like a fair judge. The aim is to prove to the commissioners whether their strategic policies have improved a particular practice. The scope and the criteria for the evaluation, often the strategic goals of the organization, are provided by the commissioners. It is the task of the evaluator to formulate indicators that reflects the given criteria. The evaluator subsequently chooses or develops appropriate, often quantitative methods, for measuring the indicators. The stringent application of these methods legitimates the final outcomes that are expected to reveal "the facts", or the "truth" from an external, objectivist point of view. Differences between

the actual findings and the intended outcomes are explained and clarified by referring to causal relationships between the distinct indicators. This approach belongs to a long tradition of positivistic or explanatory research that finds its roots in the sociology of Auguste Comte.

This thesis has been conducted within another tradition. This tradition believes that 'truth' does not exist outside the perceptions of people. Truth is what people agree on at a certain point in time and in a particular context. It is often based on consensus about what the people involved think is a meaningful description of reality. What we call truth or scientific knowledge is the result of a process of collaboration and exchange of information.⁶⁹ This tradition is called social-constructionism⁷⁰ and goes back to the 19th century philosopher Wilhem Dilthey. He tried to make a distinction between the object and methodology of the natural sciences and those of social sciences. The methodologies in the social sciences are focused on the study of complex social relationships and aim at enhancing understanding of these relationships. The purpose of social research is not to explain ('Erklären') but to understand ('Verstehen') the way people give meaning to life-world experiences. Hermeneutic or interpretive approaches are necessary to obtain access to the thoughts, beliefs and experiences of people. In our studies of patient involvement in research we have used one of these interpretative approaches that is called responsive evaluation.⁷¹

Responsive evaluation is a theory and methodology that has been introduced by Bob Stake in the context of evaluating educational programs.⁷² He promoted an approach of evaluation that does not use pre-defined evaluation criteria, but criteria that are derived from all stakeholders involved during the evaluation. The use of qualitative instruments are needed to distill these criteria because participants have to formulate what they expect from a new policy or a new intervention and what they find important to include in the evaluation. This approach was expanded by Egon Guba & Yvonna Lincoln who emphasized the importance of not only consulting multiple stakeholders in the evaluation process, but of doing evaluation research with active involvement of all stakeholders.⁷³ This interactive or participatory approach of evaluation has been adjusted to the context of health programs and health research by Tineke Abma.^{74 75}

Abma's account of responsive evaluation is grounded within narrative psychology⁷⁶ and dialogical ethics.⁷⁷ She emphasizes that stories illuminate how people make sense of their world; stakeholders issues are embedded in stories and go beyond mere information and factual accounts. What really matters to stakeholders and what worries them can be understood when listening to their stories. Abma has expanded the responsive approach with a social-critical frame.^{78 79} Within such a framework evaluation should actively steer

towards the inclusion of marginalized voices. This “ethical” turn is grounded in the empowerment tradition and focuses on the participation of unheard voices providing them a voice and control to shape their own destiny. This participatory framework departs from a democratic tradition in which deliberation and dialogue are the means to strive for social justice and fairness. Central features of dialogue are openness to the perspectives of others, respect, inclusion and engagement.⁸⁰

Philosophically this approach is positioned, as said, within a long tradition of hermeneutic or interpretive research. It takes perspectives of different stakeholders into account and values their experiences, knowledge and opinions. A responsive evaluation is an appropriate approach when there are several stakeholders that may have different interests and perceptions. We used responsive evaluation to raise awareness among stakeholders of the potential value of patient participation and the conditions for its implementation in the field of rheumatology. We also used this approach because it recognizes the value of dialogue as a vehicle for changing existing research practices. Characteristic features of this approach are:

- Multiple partiality of the evaluator with respect for plurality of values and interests, and tolerance for ambiguity. A multiple partiality means engaging simultaneously with both the patient and research community and developing emphatic relationships.
- Multi-stakeholder approach with an interest for the lived experiences and stories of the participants;
- An emergent research design: as stakeholder issues are often unknown before the study starts, the design develops in conversation with the stakeholders;
- Flexible use of methods; The use of methods and concepts derived from a variety of disciplines and dependent on the needs of a particular context;
- A preference for qualitative research strategies to gain an in-depth understanding of the meaning and values of practices, in which the hermeneutic role of the evaluator is recognized;
- Continuous, interactive reflection on the value of practices;
- Not making consensus between stakeholders the primary intent, but a change of practice and mutual understanding, in our case: a greater understanding and appreciation of experiential knowledge of patients, and acknowledgement of their role as collaborating partner;

- The role of the evaluator as facilitator: Creating optimal conditions for interactive dialogues. The relation between the evaluator and the participants is crucial (intersubjective position), including helping participants to articulate their voice, especially of those who are traditionally least heard, preventing subtle mechanisms of exclusion, removing constraining procedures and dealing with dysfunctional power imbalances.

In this thesis there is congruence between the object and the method of the study. Both are grounded in the concept of reflexive dialogue. The concept of dialogue is perceived as an ontological phenomenon. This means that all human communication and behavior can only be understood by the way people talk to each other, share their stories and make validity claims. This communication takes place through a continuous dialogue on all levels. Developments in science, including a phenomenon like patient participation, can be understood by exploring the way different stakeholders communicate with each other or avoid to speak to each other. In the context of our evaluation studies the primary aim was therefore not to achieve consensus on the importance and benefits of patient participation in rheumatology research, but to stimulate the dialogue between patients and researchers and to create mutual understanding of the advantages and risks of patient participation in different research contexts.

While my personal experience may have shepherded me towards an assumption of benefit for patient involvement, the process of enquiry was intended to be open and to allow for the possibility that more harm than good might be the result. Within this purpose I perceived a special role of being a ‘patient researcher’: that of a facilitator creating conditions for a constructive and reflexive dialogue between patients and researchers. This has been achieved, for instance, by developing agenda’s, interviewing stakeholders, providing introductory presentations or moderating team meetings. In accordance with the participatory framework during this process I have paid special attention to the group that has had the weakest voice in the past: that of the patients. It requires alertness for asymmetric relationships to avoid the opinions or priorities of patients passing unnoticed.

Sometimes I performed other roles, such as teacher. The assumption is that participants are willing to contribute to a study when they understand its purpose, are confident that their input will be heard and are able to influence the research design. The main objective was to assess the added value as well as the potential risks of structural involvement of patients in arthritis research from both the perspective of patients and professionals. A recurrent point of consideration was the reluctance of researchers to engage with patients

and the challenge to reduce resistance to change. At the same time we were alert to the risks of overburdening of patients and the dominance of the research system (formalization, professionalization, standardization) that tends to exclude the experiential knowledge of patients and resists forms of active patient participation.

10. Overview of the four case studies

In this thesis I report about four case studies. These cases were selected because they reflect four different levels on which patients have actively contributed to rheumatology research (Table 1):

1. Local: Conducting a doctoral study from the perspective of a patient researcher;
2. National: Monitoring and evaluating the Dutch network of patient research partners (Reumapatiëntenbond);
3. European: Developing European League Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects (EULAR);
4. Worldwide: Evaluating a decade of patient participation in Outcome Measures in Rheumatology conferences (OMERACT).

The added value of a multiple case study design is threefold. First, these four cases have consecutively started to experiment with different forms of patient participation in different contexts (time, place, culture) and are inter-connected. Therefore it is possible to analyze and interpret the dynamic and complex development process of patient participation in the area of rheumatology on four different levels and over a long period of time. By combining data from different sources triangulation will be possible which will result in similar or new information critical to the theme under research and enhance the validity of our final findings. It will enable us to look at the inter-dependencies in this process. Second, it allows a comparison between four roles of patients as collaborative partners in the context of scientific research: In patients acting as conference partners (OMERACT), scientific committee members (EULAR), research partners (Dutch network) and as a patient researcher (this doctoral study). It will be possible to explore what these roles have in common or where they might differ, for example, what are determinants for appropriate selection procedures, training and support. Finally, in all cases patient participation has been organized as a network of peers. It is not one patient who is involved, involvement is embedded in a sustainable structure that guarantees that there are always more patients and more researchers involved.

Table 1. Overview of the four case studies and the applied methodologies

LEVEL OF STUDY	LOCAL	NATIONAL	EUROPEAN	INTERNATIONAL
Context	My own doctoral study	Dutch network of patient research partners	EULAR task force	OMERACT conferences
Chapter	9	2, 3	4	5-8
Patient role evaluated	Patient Researcher	Research partner	Committee member	Conference Participant
Main focus	Reflexive dialogue on the risks and benefits of 'patient researcher' role	Building sustainable partnership relations	Developing recommendations for collaboration	Evaluation of impact and process of structural patient participation
My main role	Reflexive qualitative researcher	Diverse roles of a responsive evaluator	Facilitator	Diverse roles of a responsive evaluator
Data sources	Key reference papers and reflexive dialogues with supervisors	Interviews, focus groups, surveys, emails, meetings and logbook	Literature review, two task force meetings and Delphi	Document review, participant observation and interviews
Methodology	Reflexive dialogues	Responsive evaluation	EULAR standardized operational procedures	Responsive evaluation

Here I should mention that I, as a person with arthritis, have been able to contribute to the establishment of three networks and have witnessed as an insider the process of involvement of patients first hand from the very beginning, over a period of 10 years. This privileged position has been beneficial to conduct these studies. In chapter 9 we will reflect on the challenges of a patient researcher in more detail. Below we will briefly introduce the four case studies.

Monitoring and evaluation of the Dutch arthritis network of patient research partners

In chapter 2 and 3 we will present an evaluation study of the Dutch 'Network of patient research partners'. This network was initiated by the Dutch arthritis patient organization (Reumapatiëntenbond) in 2008. People with a rheumatic condition were selected and prepared to provide the patient perspective in the context of regional research projects. An important part of these preparations were a three-day training course, divided into a two-day introduction meeting at the start, and a follow-up meeting after 6 months. In between patients were expected to follow an apprenticeship with a local research group. The aim of Reumapatiëntenbond was to enhance the patient perspective in Dutch

rheumatology research by educating patients to become volunteers in a network of research partners. At the end of the 3-year project the network consisted of 27 partners at 6 academic research centers.

In chapter 2 we report on the usefulness of the ‘FIRST’ model in the process of developing and implementing structural patient involvement through a network of research partners in the Netherlands. The FIRST model provides practical tools for building collaborative relationships between researchers and patients. In chapter 3 we present the findings of our monitoring and evaluation study that aimed to describe and analyze both the process and results of the network from the perspective of transition theory.

In the case of the Dutch network we followed a formative approach of evaluation and monitoring. This approach is based on the principles of ‘action research’ and provides insights into the process of building sustainable partnerships between patients and professionals on a local level. An emergent research design and the use of mixed methods (qualitative and quantitative instruments) led to actions and recommendations for practical improvements. We did not develop a set of hypotheses, but rather decided regularly to introduce new ways of data collection, depending on the outcomes of former stages. One of the decisions was to formulate endpoints for the evaluation in close collaboration with all stakeholders. During an interim meeting we explored potential criteria for “effectiveness” and “sustainability”. It was also suggested that we collect quantitative data by developing two mirrored surveys among research partners and researchers. In this study the focus lays on evaluating the process. It turned out to be difficult to measure the contributions or added value of patient participants.

EULAR recommendations for the inclusion of patient research partners

EULAR fosters close collaboration between researchers and patients and has recognized the need to provide clear guidance for participants to build structural partnerships in the context of scientific research. For this reason in 2008 EULAR funded a proposal to develop recommendations for patient participation in scientific EULAR projects. Chapter 4 reports the recommendations that came out of this project that was carried out by an international task force of 7 patient experts and 7 professionals. Participants of the Task Force were selected on the basis of their knowledge and experience with patient involvement in research. A number of patient representatives were actively involved in the EULAR Standing Committee of People with Arthritis/Rheumatism in Europe (PARE) and other patient participants had extensive experience in OMERACT. The

professionals were key opinion leaders in the field of rheumatology or researchers with a known interest in patient involvement.

I was a participant in the committee, and we followed the EULAR standardized operational procedures for the development of recommendations⁸¹ and held two heterogeneous face-to-face meetings where all participants discussed over two days the potential opportunities, benefits and conditions for patient participation in EULAR scientific projects. Between the two meetings an extended Delphi method was carried out to collect all suggestions and comments. After the second meeting a wider consultation took place to obtain information on the level of agreement with the recommendations. All participants were treated equally: they received the same background materials, contributed equally to the small group discussions and plenary sessions, and became co-authors of the final peer-reviewed manuscript. The role of the researcher was mainly that of a facilitator.

A responsive evaluation of the inclusion of patients in OMERACT conferences

OMERACT is a five-day, data-driven working conference⁸² attended by a variety of stakeholders, such as epidemiologists, rheumatologists and other health professionals, biostatisticians, and representatives of pharmaceutical industry and health regulators. Since 2002 patients have also been participants in order to bring in the patient perspective. They form 10% of 200 delegates and their role and position were formalized by the Executive Committee in 2011.⁸³ The contributions of patients have developed over time. At the first OMERACT conference the patients formed a homogeneous group of 11 persons with rheumatoid arthritis (RA) without experience in scientific research. As a kind of focus group they provided the experiential knowledge necessary to review the existing RA core set of measurement tools for assessing the effects of treatment from a patient perspective.⁸⁴ During the following conferences the patient group expanded into a heterogeneous group encompassing patients with different competences, knowledge and diseases.^{44 85 86}

Becoming more familiar with the spirit of OMERACT, the language and the working procedures during the conference, the patients started to perform tasks similar to researchers and their role gradually evolved into a more structural collaborative relationship. Nowadays patients take on the role of rapporteur or co-moderator of breakout sessions, presenter at plenary sessions, or co-author on scientific papers. They are also responsible for organizing their own support by appointing buddies for newcomers or organizing patient introduction sessions. Although OMERACT has been cited as a successful initiative for involving patients in research, no rigorous exploration has

been undertaken of the extent to which patients have contributed to the rheumatology research agenda, how they have influenced the OMERACT process and what conditions have contributed to its assumed success. For this reason I performed in 2010 a responsive evaluation of a decade of patient involvement in OMERACT conferences as a substantial part of his PhD. The evaluation includes a systematic document analysis of the process and impact of structural involvement of patients in OMERACT, participant observation during six patient meetings, and 38 qualitative interviews with participants that attended OMERACT 11 (2010) in Kota Kinabalu, Malaysia: 16 patients and 16 professionals. The results are presented in four chapters.

Chapter 5 explores the impact of a decade of patient participation on the process and output of the conference. Here we also explain the methodology of responsive evaluation in more detail. After summarizing the history of patient participation in OMERACT we show that long term engagement with arthritis patients in OMERACT conferences has significantly influenced the OMERACT research agenda and the development of core sets and patient reported outcome measures often used in clinical trials. Patients have identified new domains that are important for patients. Patient involvement has also had a major spin off outside OMERACT. Inspired by the OMERACT experience patient participants started national or local networks of patient research partners and researchers implemented principles of participative research in their own projects. Also beyond rheumatology OMERACT inspired researchers to collaborate with patients as conference partners.

Chapter 6 presents the conditions that have enabled OMERACT to successfully implement patient involvement in its scientific conference. We start by summarizing the challenges of participatory research as reported by researchers from other disease areas. Then we describe and analyze five factors that have been essential to enable patient research partners to contribute effectively to OMERACT. We also report three barriers that prevent participants to optimally utilize the experiential knowledge of patients. Despite the positive examples and experiences of the interviewees, it is also noted that there are still opportunities for improvement.

We assumed that the expectations and experiences of new patient participants could provide us with valuable insights in the process of *initiation*. A separate chapter is devoted to this process of initiation and answers the question how new comers experience their participation and engage with researchers. In chapter 7 we will focus on three different phases associated with important conditions for participation: Introduction and preparation before the conference; moderation and on-site support during the conference; and contributions and personal benefits as perceived by the patients after the

conference. Based on interviews with eight patient delegates that attended OMERACT for the first time, we will describe the process that all newcomers go through to become fully contributive to the conference objectives. Special insight is provided by repeated interviews with three new patients before, during and after the conference, which provide a wealth of information to illustrate the process that can be described as adversarial growth.

Finally, chapter 8 summarizes our overall findings as presented in a separate session at OMERACT 12 (2012) in Pinehurst, North Carolina. There I had the opportunity to introduce the results of the combined review of data derived from the document analysis and the 38 interviews for an audience of interested OMERACT participants.

Reflection on the process of becoming a patient researcher

In chapter 9 I reflect on my personal transition from patient to patient researcher. Here I present a critical appraisal of the particular features that are associated with this role. Together with my supervisors I have analyzed the potential advantages as well as the potential risks that come with this role. These reflective discussions revealed the dangers of over- and under-identification with stakeholders but also the potential usefulness of the theory, principles and role-options of responsive evaluation. They enforced my role as a go-between, connecting the worlds of patients and researchers and enhancing mutual understanding via dialogue. As an interpreter and teacher it was possible to help patients understand the research process and to prepare and empower them for their role as collaborative partner. As a facilitator and Socratic guide it was possible to help researchers to incorporate the patient perspective in their research projects. In practice these roles sometimes conflicted with each other which will be illustrated in two cases from my interviews.

These four case studies, explored and analyzed using the methodology of responsive evaluation, will provide insights into the way structural changes to the research process can help patients, researchers and funders to better support collaborative working with an open eye for potential risks. Though the viewpoints of different stakeholders are sometimes contradictory, common themes emerge that demonstrate the benefits and challenges of patient-researcher collaboration. These are reviewed in chapter 10 and provide evidence on which to formulate recommendations of good practice.

REFERENCES

1. Hunter DJ. Health needs more than health care: the need for a new paradigm. *Eur J Public Health* 2008;18(3):217-9.
2. Habermas J. *Theorie des kommunikativen Handelns. Zur Kritik der funktionalistischen Vernunft* 2nd ed. Frankfurt am Main: Suhrkamp, 1982.
3. Elberse JE. Changing the health research system. Patient participation in health research [dissertation]. Amsterdam: VU University, 2012.
4. de Swaan A. *Het medisch regiem [transl: The medical regime]*. Amsterdam: Meulenhof, 1985 (1984).
5. Kerkhoff AHM. *De samenleving in schema's. Een inleiding in het sociologische denken van Talcott Parsons [transl: Society in patterns. An introduction into the sociology of Talcott Parsons]*. Budel: Damon, 2007.
6. Dahlberg K, Todres L, Galvin K. Lifeworld-led healthcare is more than patient-led care: an existential view of well-being. *Med Health Care Philos* 2009;12(3):265-71.
7. Devereux G. *Angst und Methode in der Verhaltenswissenschaft*. München: Suhrkamp, 1973.
8. Finlay L. "Outing" the researcher: the provenance, process, and practice of reflexivity. *Qualitative health research* 2002;12(4):531-45.
9. Habermas J. *Theorie des kommunikativen Handelns. Handlungsrationality und gesellschaftliche Rationalisierung* 2nd ed. Frankfurt am Main: Suhrkamp, 1982.
10. Hootman JM, Helmick CG, Brady TJ. A public health approach to addressing arthritis in older adults: the most common cause of disability. *Am J Public Health* 2012;102(3):426-33.
11. Woolf AD, Erwin J, March L. The need to address the burden of musculoskeletal conditions. *Best Pract Res Clin Rheumatol* 2012;26(2):183-224.
12. Loza E, Jover JA, Rodriguez L, Carmona L. Multimorbidity: prevalence, effect on quality of life and daily functioning, and variation of this effect when one condition is a rheumatic disease. *Semin Arthritis Rheum* 2009;38(4):312-9.
13. Franke LC, Ament AJ, van de Laar MA, Boonen A, Severens JL. Cost-of-illness of rheumatoid arthritis and ankylosing spondylitis. *Clinical and experimental rheumatology* 2009;27(4 Suppl 55):S118-23.
14. Kobelt G. Economic studies in rheumatology: data, perspectives, challenges. *Rheumatology* 2012;51(2):208-9.
15. Raza K, Stack R, Kumar K, Filer A, Detert J, Bastian H, et al. Delays in assessment of patients with rheumatoid arthritis: variations across Europe. *Annals of the rheumatic diseases* 2011;70(10):1822-5.
16. Putrik P, Ramiro S, Kvien TK, Sokka T, Pavlova M, Uhlig T, et al. Inequities in access to biologic and synthetic DMARDs across 46 European countries. *Annals of the rheumatic diseases* 2013.
17. Sokka T, Kautiainen H, Pincus T, Toloza S, da Rocha Castelar Pinheiro G, Lazovskis J, et al. Disparities in rheumatoid arthritis disease activity according to gross domestic product in 25 countries in the QUEST-RA database. *Annals of the rheumatic diseases* 2009;68(11):1666-72.
18. Schoels M, Knevel R, Aletaha D, Bijlsma JW, Breedveld FC, Boumpas DT, et al. Evidence for

- treating rheumatoid arthritis to target: results of a systematic literature search. *Annals of the rheumatic diseases* 2010;69(4):638-43.
19. Smolen JS, Aletaha D, Bijlsma JW, Breedveld FC, Boumpas D, Burmester G, et al. Treating rheumatoid arthritis to target: recommendations of an international task force. *Annals of the rheumatic diseases* 2010;69(4):631-7.
20. de Wit MPT, Smolen JS, Gossec L, van der Heijde DM. Treating rheumatoid arthritis to target: the patient version of the international recommendations. *Annals of the rheumatic diseases* 2011;70(6):891-5.
21. Rao JK, Weinberger M, Anderson LA, Kroenke K. Predicting reports of unmet expectations among rheumatology patients. *Arthritis and rheumatism* 2004;51(2):215-21.
22. Stevenson FA, Barry CA, Britten N, Barber N, Bradley CP. Doctor-patient communication about drugs: the evidence for shared decision making. *Social science & medicine* 2000;50(6):829-40.
23. Stacey D, Murray MA, Legare F, Sandy D, Menard P, O'Connor A. Decision coaching to support shared decision making: a framework, evidence, and implications for nursing practice, education, and policy. *Worldviews Evid Based Nurs* 2008;5(1):25-35.
24. Stiggelbout AM, Weijden TV, Wit MP, Frosch D, Legare F, Montori VM, et al. Shared decision making: really putting patients at the centre of healthcare. *BMJ* 2012;344:e256.
25. Augustinus L, Kortenhoeven P, Tjong Joe Wai E, de Wit MPT. De opvattingen, ervaringen en verwachtingen van mensen met een reumatische aandoening over wetenschappelijk onderzoek [transl: Perceptions, experiences and expectations of people with a rheumatic disease regarding scientific research]. *Ned Ts voor Reum* 2004;4:63-64.
26. Kvien TK, Heiberg T. Patient perspective in outcome assessments--perceptions or something more? *The Journal of rheumatology* 2003;30(4):873-6.
27. Leeb BF, Sautner J, Leeb BA, Fassl C, Rintelen B. Lack of agreement between patients' and physicians' perspectives of rheumatoid arthritis disease activity changes. *Scand J Rheumatol* 2006;35(6):441-6.
28. Hewlett SA. Patients and clinicians have different perspectives on outcomes in arthritis. *Journal of Rheumatology* 2003;30(4):877-9.
29. Barber R, Boote J, Parry G, Cooper C, Yeeles P. Evaluating the impact of public involvement on research. In: Barnes M, Cotterell P, editors. *Critical Perspectives on User Involvement*. Bristol: The Policy Press, University of Bristol, 2012:217-23.
30. Staley K. Exploring Impact: Public involvement in NHS, public health and social care research. Eastleigh: INVOLVE, 2009:116.
31. Leong AL, Euler-Ziegler L. Patient advocacy and arthritis: moving forward. *Bull World Health Organ* 2004;82(2):115-20.
32. Lorig K, Lubeck D, Kraines RG, Seleznick M, Holman HR. Outcomes of self-help education for patients with arthritis. *Arthritis and rheumatism* 1985;28(6):680-5.
33. de Wit MPT. Entering new area: Patient participation in research, care, education and policies. *Ann Rheum Dis* 2008;67(Suppl II):34.
34. Hewlett S, De Wit M, Richards P, Quest E, Hughes R, Heiberg T, et al. Patients and professionals as research partners: challenges, practicalities, and benefits. *Arthritis & Rheumatism* 2006;55(4):676-80.
35. de Wit MP, Berlo SE, Aanerud GJ, Aletaha D, Bijlsma JW, Croucher L, et al. European League

- Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects. *Annals of the rheumatic diseases* 2011;70(5):722-6.
36. Euller-Ziegler L, Kern-Degiovanni F, Gillet P, Roche M, Ziegler G. Intervention of rheumatoid arthritis patients trainers in a nurse school: a positive evaluation. *Ann Rheum Dis* 2006;65(Suppl II):681.
37. Aanerud GJ, Reinsberg SL, Kjekken I. Experience from four years of involving patient research partners in developing clinical practice and research. *EULAR congress*. London: Ann Rheum Dis, 2011:787.
38. Kjekken I, Aanerud GJ, Reinsberg SL. Involving patient representatives in development and evaluation of clinical care and research. *Ann Rheum Dis* 2009;68(Suppl3):353.
39. Shea B, Santesso N, Qualman A, Heiberg T, Leong A, Judd M, et al. Consumer-driven health care: building partnerships in research. *Health Expectations* 2005;8(4):352-9.
40. Hewlett S. Learnings from the Bristol experience - Practicalities of a decade of patient involvement in research on a local level. *Ann Rheum Dis* 2010;69(Suppl3):29.
41. Kirwan J. Fifteen years of experience with patient involvement in research. What has been achieved? *Ann Rheum Dis* 2012;71(Suppl3):21.
42. Nicklin J, Cramp F, Kirwan J, Urban M, Hewlett S. Collaboration with patients in the design of patient-reported outcome measures: Capturing the experience of fatigue in rheumatoid arthritis. *Arthritis Care & Research* 2010;62(11):1552-8.
43. Kirwan J, Heiberg T, Hewlett S, Hughes R, Kvien T, Ahlmen M, et al. Outcomes from the Patient Perspective Workshop at OMERACT 6. *Journal of Rheumatology* 2003;30(4):868-72.
44. Kirwan JR, Newman S, Tugwell PS, Wells GA, Hewlett S, Idzera L, et al. Progress on incorporating the patient perspective in outcome assessment in rheumatology and the emergence of life impact measures at OMERACT 9. *Journal of Rheumatology* 2009;36(9):2071-6.
45. Akerhielm CE. Patient participation in research – A Swedish model. *Ann Rheum Dis* 2009;68(Suppl3):800.
46. Akerhielm CE. Patient research partners - A way to participation and influence in research. *Ann Rheum Dis* 2010;69(Suppl3):753.
47. Kjekken I, Ziegler C, Skrolsvik J, Bagge J, Smedslund G, Tovik A, et al. How to develop patient-centered research: some perspectives based on surveys among people with rheumatic diseases in Scandinavia. *Physical Therapy* 2010;90(3):450-60.
48. Velthuis H, Schrieke M, de Wit MPT. From passive object to active subject - Preliminary outcomes of the evaluation of the dutch network of patient research partners. *Ann Rheum Dis* 2009;68(Suppl3):793.
49. Elberse JE, de Wit MPT, Velthuis HMA, Dijkmans BAC, Evers AWM, Geenen R, et al. Netwerk Onderzoekspartners in de reumatologie. Getrainde patientenvertegenwoordigers betrokken bij onderzoek [translation: Educated patient representatives involved in research]. *Dutch Journal of Rheumatology* 2009;4:40-4.
50. de Wit MPT, Boonen A, Essers M, Evers AWM, Hoogland W, Meesters J, et al. Onderzoekspartners garanderen patiëntenperspectief: Diversiteit in bijdragen aan onderzoek. *Ned Ts voor Reum*. 2011.
51. de Wit MPT, Berlo SE, Aanerud GJ, Aletaha D, Bijlsma JW, Croucher L, et al. European League Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects. *Annals of the rheumatic diseases* 2011;70(5):722-6.

52. Schipper K. Patient participation & knowledge [dissertation]. Amsterdam: VU University, 2011.
53. Abma TA, Broerse J. Patient participation as dialogue: setting research agendas. *Health Expectations* 2010;13(2):160-73.
54. Thornton H. Patients and health professionals working together to improve clinical research: where are we going? *Eur J Cancer* 2006;42(15):2454-8.
55. Abma TA, Nierse C, Widdershoven G. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qualitative Health Research* 2009;19(3):401-15.
56. Arnstein SR. A ladder of citizen participation. *Journal of the American Planning Association* 1969;35(4):216-24.
57. Caron-Flinterman JF. A new voice in science - Patient participation in decision-making on biomedical research [dissertation]. Amsterdam: VU University, 2005.
58. Baur V. Participation & partnership. Developing the influence of older people in residential care homes [dissertation]. Amsterdam: Vrije Universiteit Amsterdam, 2012.
59. Tritter JQ, McCallum A. The snakes and ladders of user involvement: Moving beyond Arnstein. *Health Policy* 2006;76(2):156-68.
60. PatientPartner. *Patient involvement in clinical research. A guide for patient organisations and patient representatives*. Soest: VSOP, 2011.
61. Abma TA, Broerse JEW. *Zeggenschap in wetenschap, patiëntenparticipatie in theorie en praktijk [Have a say in science. Patient participation in theory and practise]*. Den Haag: Lemma, 2007.
62. Nierse CJ, Schipper K, van Zadelhoff E, van de Griendt J, Abma TA. Collaboration and co-ownership in research: dynamics and dialogues between patient research partners and professional researchers in a research team. *Health Expectations* 2012;15(3):242-54.
63. Elberse JE, Caron-Flinterman JF, Broerse JE. Patient-expert partnerships in research: how to stimulate inclusion of patient perspectives. *Health Expectations* 2010.
64. Broerse JEW, Elberse JE, Caron-Flinterman JFW, Zweekhorst MBM. Enhancing a transition towards a needs-oriented health research system. In: Broerse JEW, Bunders JFG, editors. *Transitions in Health Systems: Dealing with persistent problems*. Amsterdam: VU University Press, 2010:181-205.
65. Kangas I. 'Lay' and "expert": illness knowledge constructions in the sociology of health and illness. *Health: An interdisciplinary journal for the social study of health, illness and medicine* 2002;6(3):301-04.
66. Gadamer H-G. *Wahrheit und Methode. Grundzüge einer philosophischen Hermeneutik*. 2nd ed. Tübingen: Mohr Siebeck Verlag, 1965.
67. Stake RE. Case studies. In: Denzin N, Lincoln YS, editors. *Handbook of qualitative research*. 2 ed. Thousand Oaks, CA: Sage, 2000:435-54.
68. Abma TA, Stake RE. Science of the particular. *Qual Health Res* 2013;in review.
69. Arksey H. Expert and lay participation in the construction of medical knowledge. *Social Health Illn* 1994;16:448-68.
70. Hosking DM, Pluut B. (Re)constructing reflexivity: A relational constructionist approach. *The Qualitative Report* 2010;15(1):59-75.
71. Schwandt TA. Paths to inquiry in the social disciplines: scientific, constructivist, and critical

- theory methodologies. In: Guba EG, editor. *The paradigm dialog*: Sage, 1990:258-76.
72. Stake RE. *Standards-based and responsive evaluations*. Thousand Oaks: Sage Publications, 2004.
73. Guba EG, Lincoln YS. *Fourth generation evaluation*. Beverly Hills: SAGE Publications, 1989.
74. Abma TA. The practice and politics of responsive evaluation. *American journal of evaluation* 2006;27(1):31-43.
75. Abma TA, Widdershoven GAM. Evaluation as a relationally responsible practice. In: Denzin N, Lincoln Y, editors. *Handbook for Qualitative Inquiry*. Los Angeles: SAGE Publications Ltd., 2011:669-80.
76. Abma TA, Widdershoven GA. Sharing stories: narrative and dialogue in responsive nursing evaluation. *Eval Health Prof* 2005;28(1):90-109.
77. Abma TA, Widdershoven GA, Frederiks BJ, van Hooren RH, van Wijmen F, Curfs PL. Dialogical nursing ethics: the quality of freedom restrictions. *Nurs Ethics* 2008;15(6):789-802.
78. Baur VE, Abma TA, Widdershoven GA. Participation of marginalized groups in evaluation: mission impossible? *Evaluation and program planning* 2010;33(3):238-45.
79. Greene JC, Abma TAe. *Responsive evaluation*. San Francisco: Jossey Bass, 2001.
80. Abma T. Opening thoughts. Dialogue in evaluation. *Evaluation* 2001;7(2):155-63.
81. Dougados M, Betteridge N, Burmester GR, Euller-Ziegler L, Guillemin F, Hirvonen J, et al. EULAR standardised operating procedures for the elaboration, evaluation, dissemination, and implementation of recommendations endorsed by the EULAR standing committees. *Annals of the rheumatic diseases* 2004;63(9):1172-6.
82. Tugwell P, Boers M, Brooks P, Simon L, Strand V, Idzerda L. OMERACT: an international initiative to improve outcome measurement in rheumatology. *Trials* 2007;8:38.
83. OMERACT. Guiding principles for patient research partner participation in OMERACT. Accessed: October 18; 2012: Available from: <http://www.omeract.org/pdf/2011-03-27%20Agreed%20Patient%20Participation%20Policy.pdf>.
84. Kirwan J, Heiberg T, Hewlett S, Hughes R, Kvien T, Ahlmen M, et al. Outcomes from the Patient Perspective Workshop at OMERACT 6. *J Rheumatol* 2003;30(4):868-72.
85. Kirwan JR, Ahlmen M, de Wit M, Heiberg T, Hehir M, Hewlett S, et al. Progress since OMERACT 6 on including patient perspective in rheumatoid arthritis outcome assessment. *Journal of Rheumatology* 2005;32(11):2246-9.
86. Kirwan JR, Boonen A, Harrison MJ, Hewlett SE, Wells GA, Singh JA, et al. OMERACT 10 Patient Perspective Virtual Campus: valuing health; measuring outcomes in rheumatoid arthritis fatigue, RA sleep, arthroplasty, and systemic sclerosis; and clinical significance of changes in health. *Journal of Rheumatology* 2011;38(8):1728-34.