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CHANGING RESEARCH PRACTICES THROUGH DIALOGUE.

A CRITICAL ANALYSIS OF PATIENT PARTICIPATION IN RHEUMATOLOGY RESEARCH

The powerful research methods of qualitative scientists
allow us to transcend anecdote and its risks
and to capture and learn from the evidence of the singular.

Rita Charon[1]

Maarten P.T. de Wit
John Kirwan
Tineke A. Abma

1. Introduction

There is a worldwide transition taking place in health research from supply driven to more need-oriented research. The traditional belief that the role of patients is limited to that of study participant is diminishing in favour of a more active engagement with patients as collaborative partners.[2] There is a slowly growing confidence that including patients as partners in research can bridge the gap between the life-world of patients and the medical system. It is expected that opening up the experiential knowledge of patients to researchers will increase the legitimacy and transparency of medical research and enhance co-ownership of patients over the research process.[3]

In the literature many challenges regarding patient participation are reported: Patients experience pseudo-participation and difficulties in achieving equal relationships with researchers; And for researchers patient participation is easier said than done. The role of patients as research partners is not yet well-developed nor accepted by the main actors in the field. Patient participation might lead to conflicting expectations and interests among participants, sometimes even resistance among researchers to changing existing practices. Scepticists have argued that not all patients aspire to become active in research,[4] which might be true for many. Others have shown that patients are willing to contribute to research although they are not always aware of their added value and opportunities.[5]

From our multiple case study it has become clear that the biggest challenge for patient research partners as well as for researchers is to engage in dialogue and create sustainable partnerships that assure adequate access to the experiential knowledge of patients. Earlier studies have demonstrated that lower forms of participation such as incidental consultation often fail to fully capture the patient perspective.[5-7] We believe that this can be explained by the absence of dialogue, a critical factor for co-production of knowledge. In our four studies we have focused on the conditions for partnerships and sustainable participation of patients in the role of collaborative partners (Table 1). Sustainable involvement, as explained in chapters 1 and 6, means that the initiative to engage with patients is not incidental and dependent on the goodwill of one or two individuals, but is a long-term commitment to create partnerships on a structural basis and to make values of participatory research part of the vision, structure and practices of the organization.

In chapter 2 and 3 we have concentrated on the establishment of a network of patient research partners by the Dutch national patient organization (Reumapatiëntenbond). During a 2 year monitoring and evaluation study we evaluated the usefulness of the FIRST model for establishing structural partnerships. This framework was developed in 2006 and provides practical advice for researchers. Members of the network provided

the patient perspective in a variety of research projects at 6 academic rheumatology centres.

Table 1

Outline of case studies of four patient roles as collaborative partners in scientific research investigated as part of this thesis.

PATIENT ROLE	CONTRIBUTION	METHOD OF PARTICIPATION	PHASE OF INVOLVEMENT	REQUIRED COMPETENCES	PREPERATION & SUPPORT
Committee member	Monitoring and co-producing	Equal participation in committee meetings	All phases	Social skills (communicator)	Training opportunities; Moderate support
Conference partner	Providing patient expertise	Collaborating as full delegate	During the conference	Communication skills	Pre-reading material, full support
Patient research partner	Providing support and patient expertise	Member of the research team	Active, equal involvement in all phases	Reflexive skills Basic knowledge of research	Maximum preparation, maximal support
Patient researcher	Patient expertise & professional research skills	Conducting research	Active involvement in all phases	Professional research skills and reflexive skills	Professional education, minimal support

Chapter 4 presented eight recommendations for participation of patients as committee members in scientific projects. These were developed by a team of seven experienced patient research partners and seven professionals on behalf of the European League Against Rheumatism (EULAR). The recommendations are currently followed by all EULAR task force leaders and are an example of institutionalization of participatory research.

   In the third case study we have focused on the role of patients as conference partners in Outcome Measures in Rheumatology (OMERACT). These international research conferences have been held every second year from their inception in 1992. Since 2002 patients have been invited to provide the patient perspective and have continued to do so till today. OMERACT offered us a unique opportunity to study the supportive and constraining conditions for patients participation over a longer period of time through responsive evaluation. In chapter 5 we presented the impact of a decade of patient participation on the research agenda and on other outcomes of the conference. Chapter 6 provided more detailed information about the facilitators and barriers of patient participation and in chapter 7 we have studied the experiences of patient research partners attending OMERACT for the first time in more detail. In chapter 8 we summarized and

integrated our findings from the previous three chapters in a set of recommendations for improvement.

In chapter 9, the fourth case-study, I have reflected on my own role as a patient researcher conducting a doctoral study. Here I described my role as a facilitator and analyzed two cases that illustrated the precariousness of patient participation in practice.

Implementing values of participatory research is a dynamic process where actors gradually establish new relationships. The traditional professional-patient relationship is based on a medical definition of illness as a deficit. The disease and the limitations are central and the role of the patient is passive receiver of health care. The main driver for scientific research is to find a cure for the disease or repair the deficit (medical engineering). Nowadays this approach of doing *to* and doing *for* doesn't fit the required new approach of doing *with*.[2] For this the actors need to enter a dialogue in which they are able to express themselves and to influence the process. To create opportunities that are inclusive and where all actors have a say, trustworthy relationships should be developed. All involved need time to get acquainted with each other and to adjust to the new approach of collaboration. In the absence of routines on how to introduce, support, sustain and evaluate patient participation, there is room for experimenting. Through the study of four case studies that have experimented with participatory research, we have been able to investigate our main research question from chapter 1: How can patients and professionals collaborate equally in the context of scientific research? The objective of this chapter is to present the main findings of the analysis, interpretation and triangulation of data derived from the multiple case studies. We will critically interpret these findings using Habermas' theory of communicative action[8 9] and the theoretical framework of responsive evaluation. In consonance with our research question we will elaborate on the conditions for establishing an open and meaningful dialogue in which all stakeholders have equal opportunities to provide their own knowledge and perspectives, leading to enhanced mutual understanding and a change of research practices.

Outline
This chapter is structured as follows: We start with a brief review of the theory of Habermas (section 2) followed by the main lessons learned from the four case studies (section 3). In par. 3.1 we describe the impact of patient participation on the research agenda in the case of OMERACT where patients successfully brought the topic of fatigue to the attention of researchers. In par. 3.2 we elaborate on the importance of the individual learning curve that patient research partners go through when they enter the world of

research. In par. 3.3 we concentrate on the conditions for researchers to prepare for collaboration and genuine dialogues with patient research partners. Finally we present in par. 3.4 the process of mutual learning as an important condition for co-production and establishing sustainable partnerships. To better understand the dynamics of developing new partnerships and creating opportunities for a constraint-free communication, we critically reflect on the particular issues as they occurred in our case studies through the theory of communicative action (section 4). After summarizing the conditions for collaboration with patient research partners, we will focus specifically on the value of responsive evaluation in the context of participatory research (section 5). After highlighting the strengths and limitations of this study (section 6) we will elaborate on implications for further research and for research practices (section 7).

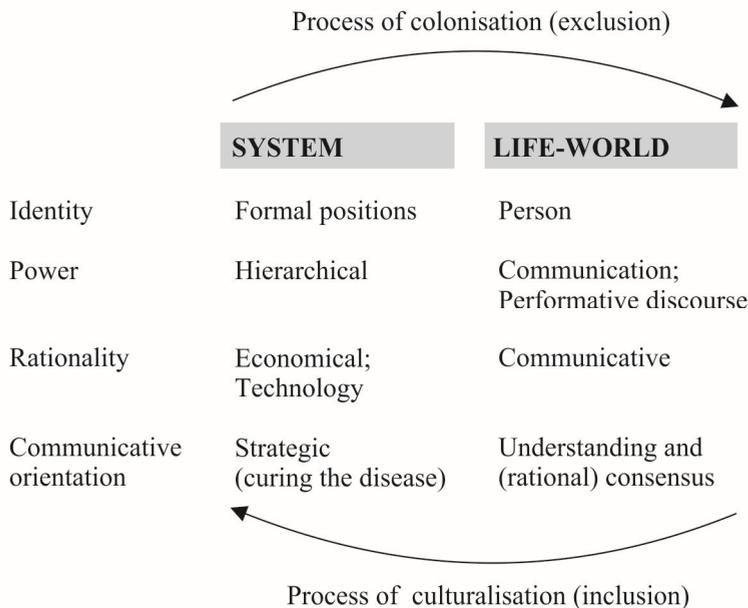## 2.   Habermas' theory of communicative action

Jürgen Habermas (1929) is one of the greatest German philosophers of the 20th century. He is a late representative of Critical Theory and the well-known Frankfurt School and writes in the tradition of Dilthey, Husserl and Heidegger. Habermas' magnum opus 'Theory of communicative action' (1981), presented in two volumes, provides a useful model to interpret the development of patient participation in modern society. It emphasizes the importance of the interaction between people in the production of knowledge. Communicative action is the opposite of strategic action and can be defined as collaborative action undertaken by individuals based upon mutual deliberation and argumentation. The quality of the deliberation between different stakeholders about their validity claims is key in Habermas' definition of reason and rationality. The ideal situation of argumentative speech is characterized by the absence of communication blockades and the mutual search for understanding and rational consensus. Crucial in the context of research is the absence of hierarchical relationships and the use of language that facilitates all stakeholders to understand the arguments and values that support the validity claims. Only when all stakeholders have equal rights to express their thoughts and opinions, a genuine dialogue will be possible in which participants will be able to contest validity claims and to criticize them through deliberate argumentation. For this reason we believe that the theory of Habermas will help us to better understand the integration process of different kinds of knowledge, including the experiential knowledge of patients, and how such a dialogue can be facilitated.

   In his second volume Habermas constructs a two-level concept of modern society that integrates the domains of the life-world and the system world. In his view modern society has resulted in an imbalance between both worlds, meaning that the system components

economy and technology penetrate all aspects of daily life, repressing chances for communicative action in the life-world. When we apply this model on the theme of this dissertation, the developments in the research system show that decisions are not driven by the motives and values of the participants, but often by political and economic powers, such as health authorities, insurance companies and the pharmaceutical industry. Habermas speaks of strategic action ("Erfolgsorientiertes Handeln") and describes the supremacy of the system as a process of colonization of the life-world (Figure 1). In this modernization process the connection between the medical system and the life-world of patients has become problematic. New technologies in the biomedical system emerged that developed their own dynamics and started to permeate the life-world of patients.

Experiential knowledge provides insights in the way patients experience the impact of a chronic illness on everyday life, what their struggles are and how they cope with and endure the influence of the biomedical system. This system does not automatically address the concerns and desires of patients. It concentrates on the pathogenesis of the disease and tries to cure while remaining unaware of the more existential, practical and social questions of patients, and in doing so, may limit the opportunities to alleviate the consequences of illness.[10] Restoring the relationship between experiential knowledge and

Figure 1 Habermas' distinction between system and life-world.

	SYSTEM	LIFE-WORLD
Identity	Formal positions	Person
Power	Hierarchical	Communication; Performative discourse
Rationality	Economical; Technology	Communicative
Communicative orientation	Strategic (curing the disease)	Understanding and (rational) consensus

Process of colonisation (exclusion)

Process of culturalisation (inclusion)

other kinds of medical or expert knowledge is pivotal for patients to regain trust and relative autonomy within the health care system. The question is whether the medical and research system is willing and able to open up their practice and to start listening to patients, changing practices and accepting a new role of patients as responsible and collaborative partners.

Culturalization

We will use this concept to analyze how the experiential knowledge of patients has been abandoned by the world of medical research and how patient participation may contribute to a reverse process of culturalization of the world of research. The term culturalization has been coined by the Dutch philosopher Harry Kunneman to emphasize the interference between both systems.[11][12] Patients and professionals are to a certain extend able to transform the research system by introducing life-world experiences and communicative action into the system world. The question is: Can structural engagement with patients through a process of dialogue provide access to the experiential knowledge of patients that cannot be obtained in any other way? Will it contribute to an enhanced understanding of the patient perspective and stimulate a process of culturalization of the medical research system?

According to Habermas the ideal of emancipation and a constraint-free communication (herrschaftsfreihe Kommunikation) cannot be achieved in practice. It is a normative horizon that provides participants with a clear orientation for action. For this reason we believe that the theory of Habermas converges with the methodology of responsive evaluation. Both theories are grounded in a democratic and participatory framework and start from a critical stance to modernity and colonization of the life-world. They both value multi-stakeholder involvement (including the imperative to attend to the plurality of values and interests), the role of reflexive dialogue and the aim of mutual understanding.

3.   Main findings

3.1 Patient participation works

To what extent has patient participation enhanced more patient oriented research? Looking at the case study of the OMERACT conferences we found that patient participation has changed the process and results of outcome research in the field of rheumatology. In chapter 5 we described how in 2002 patients participated in OMERACT for the first time and took part in a workshop to assess the existing

rheumatoid arthritis (RA) core-set. They identified fatigue, well-being and sleep disturbances as neglected domains in health research. Patients entered a genuine dialogue with researchers explaining that the existing core-set was too limited, focused too much on physical signs and symptoms and insufficiently on the impact of the disease on daily life. Fatigue was almost unanimously reported by patients as an overwhelming symptom that interferes with all aspects of life and is almost always ignored in clinical practice. The researchers participating in the workshop listened to the patient stories and took the message home to explore the phenomenon in all of its dimensions.

The results have been illustrative. Many studies have been published on the impact of fatigue, how patient describe fatigue, what it means to them, how it can be assessed, what it adds to the RA core-set, and how it can be treated. Many OMERACT delegates have confirmed that the interest in this topic would not have occurred without patients attending the conference and telling researchers of the immense influence of fatigue in their lives. Conference delegates had discussed the importance of fatigue several years before, but without any follow-up. Only when patients asked researchers to take fatigue serious, it's urgency became apparent. In 2008 fatigue was added as a recommended outcome to the RA core-set to be used in all RA clinical trials.

3.2 Adversarial growth of patients

Based on the positive experiences with patient participation in 2002 and because of the continuous commitment of the OMERACT leadership, patients stayed involved in subsequent conferences. Chapter 5 has shown that for patients their participation in these conferences was often a rewarding activity. By interactions with their peers and the collaboration with professionals they went through a process of empowerment in which they learned more about their own disease and about the methodology and language used in health research. They built an identity as research partners and gained self-confidence and a better understanding of their own situation. This enabled them to speak in a more relaxed way about their personal experiences in small group discussions as well as to present a transcending perspective of living with a chronic condition in other contexts. These findings confirmed experiences of patient research partners in other disease areas.[5] [13] As a consequence of the individual learning curve the contributions of patients changed over time and became richer and more diverse.

In chapter 6 we have shown that the way research partners became acquainted with scientific research was not only a positive experience, it was also often an ambivalent experience that can be described as a process of socialization. Partners reported that in the beginning they did not know what was expected of them. They were uncertain about

how their involvement could be useful in the context of health research and felt uncomfortable. They did not know how to prepare for their participation. After a while they became aware of the importance of understanding the rules and procedures for doing rigorous scientific research and were eager to learn common routines. They started to know what was expected of them, although they still did not know how to collaborate with professionals and how they could bring in their experiential knowledge to the discussions.

At a later stage, they developed basic skills in the methodology of research and learned what is important from the perspective of researchers. They dared to raise their voice and learned to deal with risks of overburdening, power imbalances and difficult jargon. They were also able to contribute to the training of new patients and accept the role of buddy. In the end for some partners their competence to work in a research environment became second nature. The purpose to bring in experiential knowledge and supporting other participants had been internalized within the attitude and behaviour of the partner. This process of empowerment and obtaining skills and the ability to provide the patient perspective in accordance with requirements of scientific research can also be referred to as a process of proto-professionalization.

Specially from the repeated episode interviews with patient delegates attending OMERACT for the first time (chapter 8) we learned that new patients felt incompetent to meet the presumed expectations of professionals and doubted whether they had contributed much to the conference. Professionals confirmed that also new patients make important contributions although it is harder for them to distinguish between perceived contribution and actual contribution.

Learning to understand the objectives and procedures of OMERACT and participating in breakout sessions was a sometimes daunting experience. Their overall feedback however was that of a worthwhile experience and the expectation that they would be more productive at a next conference. Participants confirmed that you cannot prepare for this conference other than give yourself over to something new, going on a learning expedition.

That expedition might be described as a process of *adversarial growth*. This term stems from psycho-traumatology and means that patients grow and acquire positive experiences of competence in a painful process of confrontation and perceived failure.[14-16] In the context of research, socialization means: acquiring the ability to construe meaningful patient roles and an identity as research partner based on encounters with not only satisfying experiences of competence and appreciation, but also discouraging and sometimes even disempowering events. The example of the patient who felt totally

disqualified by a professor (chapter 9) illustrates the ambiguity of the process of empowerment. Part of this process is the need to learn to withstand sometimes disruptive attitudes and hierarchical barriers that hinder effective collaboration.

The evaluation study of the Dutch network (chapters 2 and 3) confirmed these findings. All patients looked on their participation as a learning curve, although often not a linear process. Patients reported not only the benefits of collaboration but also the inconveniences. From the outside it may look like patient participation is an advantageous and satisfying activity for everyone, but it became clear that at times participation can also be frustrating. New patients entered an unknown world that was often challenging. Patients reported many difficulties such as not understanding the medical terminology or research jargon, lack of support, over-burdening, poor moderation of sessions, and scepticism and resistance to change among researchers. For as long as the new relationships were not well established, patients had to withstand the sometimes defensive or laconic attitude of the researcher and learn on their own how their experiential knowledge could be made beneficial for the research project.

3.3 Don't forget the professional!
In the context of the Dutch network researchers struggled with several dilemmas to establish sustainable relationships such as identifying eligible research partners, formulating expectations of the tasks and role of patients, calculating a budget for expenses, communicating adequately, creating time to explain complex concepts to partners and acknowledging their contributions. We found many examples of partners criticizing items in already validated questionnaires that made researchers almost desperate. When it became apparent that the researchers were not able to work differently as they designed and performed research projects and seemed incapable of accepting patients as collaborative partners, they started to question the usefulness of training patient research partners. In response to the lack of clear role definitions, researchers tended to raise criteria for selection: Research partners should have an academic degree and understand that it is not possible to change or replace items in existing questionnaires.

The case story of the critical OMERACT researcher (chapter 9) illustrated that some researchers believed that only high educated patient representatives are able to contribute in a meaningful manner. Other researchers plead for more education of methods and statistics to enable research partners to better understand the scientific process. A majority of the researchers seldom questioned their own competences to engage with patients. For many their identity as a researcher, based on the traditional appreciation of expert knowledge and academic authority, seemed difficult to change. All case studies

showed that participatory research requires a change of attitude by researchers and additional instruction, support and encouragement. We found that researchers who wanted to invest in patient participation did not know how to start.

From our inquiries we learned that the call for comprehensive training and strict selection of patients was often grounded in a lack of skills and knowledge of participatory research among researchers, and the pressure and restrictions coming from the research system. The efforts that researchers made to involve patients were often not rewarded and sometimes even discouraged by superiors. We also found some researchers had difficulties in starting to work with partners with an open mind. In their perception of evidence-based medicine (EBM), there was hardly any place for the particularity of patient stories. In our study of the Dutch network we concluded that not only patients needed training and support, but also professionals. They needed time, guidance and encouragement to adjust to the role of a participatory researcher and to meet patients in a genuine dialogue. If superiors did not back up these attempts by professionals, they felt unsupported. The pressure to perform and deliver scientific output, the deadlines of scientific projects and submission procedures, and the shortage of time were experienced as hindering factors for such a dialogue.

The evaluation of the FIRST model (chapter 2) taught us the relevance of acknowledging the reciprocity of this model. We concluded that a change of practice requires a pro-active facilitation of researchers, a role that should be allocated preferably to professionals higher in the organizational hierarchy (principal investigators; managers). This responsibility can be distinguished from the researchers´ role of direct engagement with patient research partners and would provide adequate support.

Also within EULAR the lack of a practical framework to support researchers and task force leaders formed the argument to develop recommendations for the inclusion of research partners in scientific projects (chapter 4). In practice the recommendations have helped researchers to recruit and involve research partners from an early stage.

3.4 Mutual learning process

In our studies we identified that some experiments with direct and long term involvement of patients as collaborative partners resulted in a genuine dialogue between patients and researchers. In the case of EULAR we found that through an open exchange of stories, arguments and personal experiences, patients and researchers were able to have a genuine dialogue about the values, benefits and conditions for patient participation in scientific projects. This fruitful dialogue resulted in a broad consensus on a set of recommendations for participatory research (chapter 4). In the case of OMERACT we found that

facilitating structural engagement with patients can initiate learning processes that enhance understanding of the values and interests of all stakeholders and ultimately change the conduct and output of outcome research (chapter 5 and 6). In both cases the experiential knowledge of patients was valued and included. Through our analysis we observed that participants who succeeded in entering such a genuine dialogue experienced a mutual learning process that turned out to be advantageous for the entire research process as well as for all individual participants.

4.   Reflection on the main findings

4.1 Fatigue as a life-world issue raising 'slow questions'

The identification of fatigue as an important determinant of quality of life emerged directly from the life-world experiences of patients attending the OMERACT conference in 2002. A genuine dialogue facilitated a process in which researchers seriously listened to stories, based on everyday experiences of patients. These stories could not be told at previous conferences because patients were not there. Till then researchers were not aware of the importance of fatigue, and if the topic arose, it was put aside because it did not fit the existing medical framework of diagnosis and treatment: It could not be measured; it was perceived as highly subjective; and there was nothing that could be done to 'cure' fatigue. The medical model had no answers in response to the devastating experience of fatigue among rheumatology patients. Fatigue is highly uncontrollable from the viewpoint of a medical model, so medical research rather prefers to concentrate on more controllable aspects of the disease.

The symptom of fatigue illustrates the existing friction between life-world expectations and the imperatives of a rigid health care system. In daily life people with rheumatic diseases often feel exhausted despite the prescribed medications. At all times they have to take the impact of their disease on their energy levels into account. Patients are not able to do everything that they want, they have to make choices and give up social and leisure activities – not always understood by colleagues and others who cannot 'see' the limitation. Fatigue is a complex phenomenon that patients find hard to explain to health professionals, especially when they feel that professionals cannot handle fatigue in clinical practice. Patients suppress their complaints, accepting fatigue as something that will never go away rather than bringing it up again.

These existential and social concerns form the heart of experiential knowledge and belong to the life-world. Patients' experiences should therefore be respected and acknowledged by the health care and research systems. The neglect of the impact of

fatigue on the social life of many people with a chronic condition might metaphorically illustrate the colonization of the life-world by a system that is based on speed and production. The issue of fatigue creates what Kunneman calls 'slow questions'; questions that cannot be answered immediately, but require reflection. In that regard the fatigue issue conflicts with the medical model. The system world works like a spotlight and focuses on a few selected issues that can be easily operationalized. Fatigue is typically an issue that cannot be easily brought under control.

The stories of several patients who attended OMERACT caught the attention of the researchers. In a safe atmosphere where patients had the chance to tell their disease history, and experiencing that physicians were really interested and did not interrupt or correct them, made patients felt welcomed and listened to. There was time to discuss issues relevant to patients. The researchers participated on the same footing which resulted in a genuine dialogue, an exchange of thoughts, fears, ideas and expectations without hierarchical barriers.

Patient participation in OMERACT has shown that it is possible to bring back the existential concerns of patients in medical research. The first dialogue between patients and researchers in 2002 prompted a long term interest in the study of fatigue, resulting in an official recommendation to accept fatigue as a standard outcome for clinical trials. Although the inclusion of fatigue in the RA core-set might be perceived as an example of instrumentalization to fit the research system, more important for patients is the fact that it has resulted in a major change of medical practice. Fatigue has become an important symptom to discuss in the consultation room. The example of fatigue demonstrates the potential value of patient participation as a powerful strategy of culturalization of the medical care and medical research system: Patients can develop new identities that are based on collaboration and giving instead of waiting and receiving. As collaborative partners, patients become contributors to society. They feel satisfied that they have helped to improve health research by identifying new domains that are relevant to patients. They feel useful again and appreciate new friendships with other patients as researchers. Also researchers might develop new identities that are no longer based on strategic action but on meeting patients as persons. Their mutual efforts are aimed at opening up the life-world of patients and bring experiential knowledge into health research to obtain communicative rationality. This transformation process did not start automatically. Many challenges had to be overcome.

4.2 Experiential knowledge versus proto-professionalization

In this dissertation a variety of barriers to participation has been reported such as the use of scientific and medical jargon, the risks of pseudo-participation, overburdening, exclusion, alienation and loss of the patient perspective. All our case studies have shown some examples of tokenism, cases where research partners were strategically used to fulfil criteria of funding organizations or for other extrinsic reasons. In cases where researchers tried sincerely to engage with patients, institutional obstacles prevented the establishment of sustainable and effective relationships.

If we critically analyze the process of socialization we become aware of the significant impact of the research system on the learning curve of patient research partners. We have introduced this process as adversarial growth and colonization of the life-world: Bureaucratic procedures of large research institutes or rigid interpretations of EBM force researchers to act strategically within the system without opportunities to engage with patients on the basis of communicative action. Also, the call by some researchers for stricter selection criteria of research partners reinforced the uncertainty of patients, who in response tried to do better by asking for more background information, developing a glossary and training sessions, or organizing forms of peer support.

From a traditional perspective of scientific research patients are perceived as incompetent and passive receivers of health care because they are not able to understand the language and complexity of modern scientific research. This perception means that the experiential knowledge of patients is regarded as invalid for use in scientific research. Researchers assess patient stories within a dominant discourse that tells us that there is no place for experiential knowledge in clinical research. Patients, who already feel vulnerable about telling their personal story, receive unintended and unconsciously the message not to speak about their personal life-world experiences, and to try to adjust to the standards and values of the scientific community. The prevailing scientific discourse can make patients timid and unconfident by disqualifying their unique expertise. They internalize the belief that their existential, practical and social concerns are not relevant in the context of scientific research and attribute their failure to provide the patient perspective to their own incompetence. In fact they don't realize that the system makes them feel incompetent because within that system values such as hierarchical power, efficiency and tight deadlines rule.

In our case studies more time was spent on educating patients than on educating researchers. A potential problem is that some educational programs for patients, propagated by researchers, seem to be set up to guarantee that partners better suit the requirements of the system. Even when the official purpose is to increase self-confidence,

the actual impact might be regularly the opposite. Patients might learn how to raise their voice, but the question is whether this voice still reflects the authentic patient perspective. The phenomenon of proto-professionalization reveals the system pressure to ascertain that patients meet the expectations of researchers and that their responses fit the validated instruments. If experiential knowledge (unintentionally) gets lost in the training of patients, if patients are so well trained that they indeed fit the medical system, then one might question whether the potential of participation does not get lost as well. In the case of OMERACT patients were told that they do not need to know how to calculate statistics or design an experiment, but they do need to know how to speak up and contribute their points. Here system-oriented education was purposefully avoided. Participation and experiential knowledge are closely aligned; the experiential knowledge – like the stories about fatigue – has the potential to raise 'slow questions' and introduce new issues and considerations that cannot be easily dealt with within the current medical system and research. It is precisely the precarious nature of experiential knowledge that needs to be cherished instead of polished away.

4.3 Patients running the risk of feeling alienated

What is the impact of the socialization of research partners on the nature and value of experiential knowledge? This question became a recurrent issue in all our studies. The learning process of patient research partners often aims at incorporating the values and concepts of EBM in its most narrow and imperfect definition. Not many researchers recall the true description of EBM as "the integration of best research evidence with clinical expertise and patient values".[17] The downside of proto-professionalization is the risk of alienation. Patients become detached from their own stories and their own life-world. They may lose touch with fellow patients which dilute the unique added value of their experiential knowledge. We also showed that losing the authenticity and appreciation of experiential knowledge may provoke 'adversarial growth'.

Becoming a research partner is for many patients a painful process, caused by the overwhelming amount of bio-medical jargon, statistical concepts and lack of understanding of patient issues. This process has recently been published in the personal narrative of Truus Teunissen.[18]

Even when patients are involved in the development of patient-reported outcomes, they encounter feelings of uselessness when the discussion focuses on the psychometric properties of the instruments. Although this feeling never goes away, a majority of patients have learned to accept that they do not have to understand all the methodological issues in order to have a valuable input. Part of the socialization is recognizing the

importance of doing sound scientific research using appropriate methods without understanding their meaning: "…that they were looking for words from me, and not fully understanding the numbers…", as one of our patient partners rightly said. Becoming conscious of the unique contribution that patients can provide and that experiential knowledge is frequently missed in research projects, is often motivating the partners to continue.

In projects where the added value of patients was experienced, participants demonstrated their competence to become partners in a process of co-creation, each adding a unique value of knowledge that was meaningful in the context of research. Here patient participation became an effective strategy to preserve the life-world of patients.

4.4 Researchers caught up in the research system

Our studies showed that researchers are also confronted with many challenges, often caused by environmental factors intrinsic to existing research systems. Scepticism to patient participation is not only a matter of the knowledge and attitude of individual researchers, but also related to norms about good science and the hierarchy of knowledge. The perceived inferiority of experiential knowledge is one important barrier to engage with patients.[19] The system does not stimulate researchers to adjust their attitude and behaviour to the requirements of communicative action.[20] Listening to patients' stories, taking time to build a relationship of trust, and collaborating during the whole research project is difficult in a system that is regulated by the values of efficiency, competition and output. Patient participation is not impossible under such conditions but certainly causes frictions and tensions. Handling these tensions requires a great deal of courage and creativity of individual researchers, and support from fellow researchers and leaders. Without such support researchers can also feel lost.

Patient participation is precarious because it can suddenly diminish or be criticized. We found examples of lack of support from superiors as well as examples where the support of the leadership had been instrumental for guaranteeing genuine dialogues with research partners. Patient participation is also precarious because it confronts researchers with time consuming demands and the risk of delays. Sufficient time is an essential condition for engaging with patients, but time is also scarce. The system hardly allows nor rewards researchers to study patient issues in the broader context of the life-world. It expects researchers to look from a strategic point of view at participation, to use methods that focus on immediate needs, and to publish quickly. Researchers might also face a call by patients to become part of the decision making process, which may make researchers cautious as this entails sharing power and control over the research.

In the introduction we have referred to the potential benefit of participatory research as enhancing co-ownership by patients over the research process. Patient participation might make medical research more transparent and endorses the democratic right of patients to have a say in developments that might ultimately influence their daily life-world. The position of patients as partners in research is currently still minor and often contested, and such doubts may function as an obstacle for establishing sustainable relationships with researchers. For this reason we have emphasized the importance of the patient role as collaborating partner, defined in chapter 4 as an equal member of a research team. Patient participation is needed because patients' experiential knowledge is rarely incorporated as a valid source for scientific research.

One may question how the underlying principles of equality, co-ownership, patients' rights and democracy link to the responsibility and professional autonomy of the researcher. Patient participation has increasingly become obligatory in Western countries with funding agencies requiring such involvement. Yet researchers are still in a position to decide on patient participation. It is the researcher who invites patients to join the team, and the researcher can decide when to stop patient involvement. The initiative is still in the hands of researchers. Should and can this be changed? In the UK and elsewhere there is a movement for user-led research, started by patients who are no longer satisfied with non-participation or tokenistic involvement.[21] Such groups no longer want to respond to current research practices and projects, but want to have influence and control over the research agenda. If researchers do not respond to the increased desire to become co-researchers with equal decision making power, this counter-movement may grow. This challenges researchers to find the right balance between these tensions of democracy and scientific rigor.

When we spoke about equality in the context of research, it referred to the right of patients to be heard in the prioritization of research topics: Are researchers enquiring what really matters to patients? Are they listening and taking patients seriously in a dialogical exchange? This is an essential communicative imperative in both Habermas' theory and the methodology of responsive evaluation. Patients have a stake in scientific research and should have equal opportunities to present and defend their own validity claims. We found in our empirical studies that patients were able to identify circumstances in which they did not felt heard or where their opportunities to speak up were decreased as the result of power imbalances, ignorance or personal disinterest. Here it is the responsibility of the researcher to ensure that all perspectives are generously integrated in the research process. Equality means equal opportunities rather than equal positions. Equality does not mean that researchers have less responsibility for the

integrity of their work or lose professional autonomy. It means that they are expected to take their role as facilitator more serious and initiate open dialogues in which patients, and all other stakeholders, feel accepted as collaborative partners and feel sufficiently confident to contribute their patient perspective.

Finally, equality of patient research partners in a research team does not mean that they have to act as professionals or bring in professional knowledge. That would divert research partners away from contributing personal experience of disease in the context of the research project. As we have described in the introduction (chapter 1) the primary purpose of a research partner in the team is creating a better understanding of the life-world of patients. Researchers should develop the appropriate social and inter-personal skills to support, motivate and acknowledge the patient perspective. Researchers should have both technical knowledge of the research area and competencies to develop links with patient research partners. Ideally, they might want to make the inclusion of the patient perspective a shared responsibility of the research team, although the final decision about the adequate level and form of patient involvement will remain the responsibility of the principal researcher and the research partner.

Our findings justify the conclusion that many researchers are operating in a system unable to create conditions for an open dialogue with patients on an equal footing and without communicative blockades. Medical experts are often unable to see the absence of such conditions as they have internalized the values and rationality inherent to the system. They rely on their formal position and formal academic training, and find it hard in such context to meet as persons with a name and face. The inclination of researchers to emphasize the need of educating research partners to become more effective actually confirms their own orientation towards continuing the medical research system. As a result the opportunities for engaging in an open dialogue with patients get lost. Rather than developing an open attitude towards patients and using understandable language, listening and asking questions, researchers act in line with the strategic orientation to formalize the education and formulate job descriptions for patient research partners.

Only a minority of researchers developed the skills to reflect successfully on the challenges of participatory research. They became more aware of the existing difficulties and found ways to address them. They were open to the perspective of patients, accepted the sometimes impeding impact of participation and gave up their scepticism or resistance when they had positive experiences. We learned from the reports of partners that these researchers were finally able to build successful relationships with partners and became inspiring role models for (young) colleagues.

4.5 Relational empowerment

The four case studies taught us the importance of peer support for research partners and the dynamics of mutual learning processes. These helped us to better understand the emergence of relational empowerment[22]. We have defined patient participation as a process of dialogue. In the ideal situation we found that participants left their established positions and became open to the perspective, values and arguments of the other; not in their role as patients and researchers, but as human beings. When they got to know each other and were able to respect the competences, interests and limitations of the other, projects evolved successfully. This was certainly the case in the EULAR project. Having proportional representation of patients and researchers in the taskforce as well as in the Delphi, and facilitated by a patient researcher, created the optimal circumstances of dialogue and consensus on the recommendations for the inclusion of patients in research projects (chapter 4).

   In those situations personal learning curves merged into a process of mutual empowerment in which participants retained their own identity but gained more understanding for the perspective of others. In these encounters relational empowerment could be observed in a shift from strategic to communicative action by all the participants. They entered an open dialogue in which validity claims were exchanged in the absence of hierarchical relationships. Reflection took place upon the values, arguments and interests of the various stakeholders but also upon the language used to express the different arguments; Not only those validity claims that are perceived as 'truth' (facts; evidence), but also normative values and subjective experiences were subjected to this deliberate reflection. The domains of truth, values and experiences are defined by Habermas as potential issues for argumentative communication. When these domains are thematized and criticized in a systematic and rigorous way, and in a constraint-free environment, the result will be a form of intersubjective rationality (rational consensus). This result might help in enhancing mutual understanding and relational empowerment as important conditions for restoring the balance between system world and life-world.

5. **Conditions favouring patient participation in research and the value of responsive evaluation**

We found that responsive evaluation was an adequate methodology for studying the process and impact of patient participation in OMERACT conferences and the Dutch

network of patient research partners. The key characteristics of responsive evaluation converged with those of implementing structural patient participation in practices of rheumatology research. These characteristics can be summarized as engaging with all stakeholders in a constraint-free dialogue with the purpose of enhancing mutual understanding and fostering mutual learning processes. For this reason responsive evaluation is not only a good methodology to evaluate patient participation, it also supports the implementation of patient participation because of the need to engage in a dialogue with all stakeholders and to support relational empowerment. Finally, we found that, as illustrated in chapter 9, responsive evaluation is a useful approach to guide and inspire patients with the ambition of becoming a patient researcher (chapter 9). A patient researcher, complying with a multiple partiality, adds a unique value to responsive research. Risks of subjectivity can be minimized by applying existing research quality control measures such as practicing reflexivity.

The evaluation of our four case studies revealed conditions that are supportive for implementing patient participation and achieving relational empowerment. First, establishing conditions for a constraint-free dialogue where people can meet each other on the basis of communicative action instead of strategic interests and formal positions. The dialogue should create room for patient stories and refrain from the instrumental emphasis on treating the disease. Dialogue on life-world issues that really matter to patients and reflection on the precarious nature of participation flourishes when there is a relationship of trust. Patients and researchers need to feel safe in each other's company to bring in experiential knowledge, to show their vulnerability and to deal with issues that are contentious.

Given the importance of a relationship of trust as a condition for a genuine and reflexive dialogue (on the collaboration) between researchers and patients, structural involvement of patients in research is favourable over ad hoc initiatives. Involving patient research partners structurally in research sensitizes researchers to the values and beliefs of patients and increases sustainable inclusion of the patient perspective on the long run. Patient participation should become an integral part of the vision, strategy and procedures of research institutes in order to support researchers and patients starting up a collaboration surrounded with the tensions flowing from the friction between system world and life world. There is no simple recipe for successful participation. Patient participation is context bound and requires a situational and emergent strategy, often including multiple forms and levels of involvement, and above all systematic reflection on the collaboration in an atmosphere of trust.[23] [24] The role of collaborating research

partner seems ideal for achieving sustainability because it respects the reciprocity of the relationship.

We found several examples of self-organization of peer support and training among research partners. As a consequence of the reciprocity of the relationship, patients started to develop a shared responsibility for the process. At one of the universities of the Dutch network we have reported the initiative of a local group of partners who appointed a network coordinator (chapter 3). In OMERACT patients learned step by step to communicate with researchers, to search for information and jointly took responsibility for organizing their own support (chapter 6). These examples of self-organization empowered not only the individual partners, but strengthened also the mutual relationship with professionals. Self-organization of patients requires care and attention from researchers and should be encouraged and endorsed by the leadership.

Fostering mutual learning processes will enhance relational empowerment and may result in an increased respect for the integration of different sources of knowledge, a synthesis of perspectives without one triumphing over the other. With Gadamer we have referred to this outcome as a melting of horizons.[25] A more equal distribution of control over the research process is likely to be achieved and can be assessed as a valuable outcome of this process. Relational empowerment means that all invest equally, that the learning curves of both parties are intertwined and that everyone profits from the collaboration.[24]

Finally, as indicated, continuous reflection on the process and results of patient participation is needed. Although patient research partners and researchers grow and develop in their ability to interact and collaborate with each other, they should exchange mutual expectations at the start of a project and agree on a form and level of participation. Everyone may feel more comfortable with such an arrangement, and it may make the start of the process easier. However, it may not, as we have seen in our studies, encompass the actual interaction as it finally turns out. Therefore along the way the level and form of participation and collaboration needs to be subject of evaluation and renegotiation.

During and at the end of the project the question should be answered whether the communicative space has enabled all participants to speak up freely and contribute to a fair dialogue about different kind of validity claims. We found that a responsive evaluator, and in our case-studies a patient researcher, could be an ideal facilitator to guide this dialogue in the context of medical research. The purpose of the facilitator should be to signal symptoms of pseudo-participation (colonization) and to stimulate and enhance the process of culturalization of scientific projects and programs. In chapter 9 I

have described how along the way I have learned the principles and language of scientific research as well as the issues of patients. Being familiar with both communities enabled me to become a go-between and to develop this role of facilitator.

6   Strengths and limitations

This dissertation provides useful knowledge on the role of patients as collaborative partners in four different research contexts in one disease area: rheumatology. It has enabled us to study the barriers and facilitators of effective forms of structural patient participation and the potential and actual outcomes of that process. We have been able to involve a variety of stakeholders and to compare the views and thoughts of professionals with patient research partners. In the introduction chapter I have presented my insiders' perspective in the four case studies, both as a patient and a researcher, as a privilege that has been advantageous to conduct this PhD. Having contributed to the establishment of three networks of patient research partners and having witnessed the process of involvement of patients first hand over a period of 10 years, gave me access to key opinion leaders as well as all other kinds of stakeholders. They were all willing to contribute to this study and have done so in different ways.

One limitation to these studies was that I did not speak to patients who dropped out early from our case studies. They might have stopped their involvement for various reasons that could have given us a more comprehensive understanding of their motivation, expectations and experiences. Another limitation is the fact that in the case of OMERACT we have concentrated on the event of the conference and not on the pre- and post-conference activities. More recently patient research partners have become more involved in these phases. One of the outcomes of conducting this study is the increased expectation of participants that a more intense involvement is beneficial for the research process and for the development of genuine dialogues. We have not been able to look into this within this study itself.

Finally, work was undertaken in the area of rheumatology, a chronic disease where patients are highly dependent on a good relationship with their health professional whom they often meet on a regular basis. It is not clear to what extent this is a determining feature of the findings of this study and therefore whether these insights would be different for other disease areas such as life-threatening diseases or mental health care. The issue of generalizability or transferability in qualitative research is not based on statistical generalization but works toward 'naturalistic generalization' based on pattern matching. It is therefore difficult to formulate more general statements that go beyond the context of our four case studies. We hope that others may recognize patterns on the

basis of our 'thick description' of the meaning of patient participation and transfer them to other contexts.

7 Implications for further research and for research practices

7.1 Implications for further research

Future research of participation processes will benefit from the use of other qualitative methods as described in this dissertation. Where we based our responsive evaluations mainly on the use of document analysis, interviews, focus groups and a survey, more extensive participant observation and ethnographic research will provide more insights in not only the verbal but also the non-verbal interactions in daily research practices. These methods are adequate to study the difference between what people say and what people do. It will teach us more about the impact of communicative action and strategic action on including and excluding patient validity claims (life-world experiences) in research processes. This research should also include patient research partners who end their involvement early in the course of a project.

Also the role of the responsive evaluator or patient researcher as facilitator is worth exploring further. What skills and competences, and perhaps personal characteristics, are needed to create an open dialogue and to stimulate mutual learning processes? That role is not only relevant in the context of OMERACT conferences, but also in local projects and in working groups that engage with patients between research conferences or committee meetings. Finally, we need more research of the role of the researchers. What happens when researchers engage with patients as collaborative partners? How can researchers contribute to the culturalization of medical research and what is needed to resolve institutional barriers in this respect? Additional research should also look into adequate forms of education of future researchers to prepare them better for collaboration with patient research partners.

7.2 Implications for patient participation in research practices

We hope that the findings of this dissertation will support patient research partners and researchers, and encourage them to engage in establishing new partnerships. For researchers it is important to learn that their position is different compared to that of patients. This creates a moral responsibility. They need to become aware of the ambivalent process of socialization that many patient research partners go through before they become confident about their role and contributions.

As emphasized in the FIRST model the facilitative role of researchers is key.[26] They need to create supportive conditions for participative research. It is a challenge to support the learning curve of patients without losing the authenticity and experience-base of their stories. Providing only technical training might have a disempowering effect on patients.[24] Education of partners should aim at increasing self-confidence and helping partners to understand of the colonizing impact of the research system. Researchers should acknowledge the value of the patient perspective and accept the responsibility of making an effort to gain access to this life-world perspective. A genuine dialogue provides the opportunity to listen to patient stories and to learn more about their values and their existential, social and practical concerns without framing these concerns within the traditional hierarchy of scientific sources of knowledge. Involving patients as collaborative partners in genuine dialogues (without institutionalization of this new patient role in research) might prevent the colonization of the life-world of patients and contribute to the culturalization of medical research.

Making time for patient participation is an important factor for success. Communicative action, while having many advantages as discussed earlier, requires more time than the traditional system allows. Time is needed to become sufficiently acquainted with each other. Therefore the commitment should be made for a longer period of time. Structural involvement does not mean that the partnership should be formalized, but that there is time to build trust and confidence and, as we have seen in OMERACT, to enhance mutual learning processes and relational empowerment.

Researchers could become champions and change agents and further develop their role as a facilitator, encouraging other researchers to overcome their reluctance and enabling patient research partners to interact with researchers in constraint-free dialogues. The leadership should make sure that system barriers for researchers and patients are removed. This requires continuous reflection and feedback on the process and results of collaboration. Appropriate acknowledgement of the contributions of all involved encourages participants to continue. A structural approach of participation provides opportunities for such reflection that is necessary to address the precarious nature of this process.

The education of future researchers can be improved on the basis of the EULAR recommendations and on the evaluation of the FIRST model. Researchers should become better prepared for their role as a facilitator in participatory research projects and acknowledge the value of patient participation as an important strategy to culturalize the medical research system. Medical education should include recommendations regarding the selection, support and facilitation of patient research partners and stimulate on open

mind towards the life-world experiences and expectations of patients. Although we realize that it will not be easy to culturalize the education of researchers, the case study considering how fatigue became an outcome measure in OMERACT shows it is possible to have a major and substantive effect, and so it is clearly worthwhile striving for a better balance between system and life world.

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