Involving patient research partners has a significant impact on outcomes research: an example from the international OMERACT conferences.

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Article Summary

Article focus

- Since 2002 patients participate as collaborative partners in the biannual conference on Outcome Measures in Rheumatology (OMERACT).
- Although the contribution of patients has constantly been praised and prompted a wide-spread call for scientific publications on the impact of engaging with patients, no systematic obtained evidence has been published to support the idea that structural involvement of patients in research is beneficial.
- Our qualitative study reports the combined results of a thematic document analysis and 38 semi structured interviews with all stakeholders including researchers, patient participants and representatives from pharmaceutical industry and international regulators.

Key messages

- Long term engagement with arthritis patients in OMERACT conferences has significantly influenced outcome research in the field of rheumatology.
- Patients have successfully contributed to the research agenda of OMERACT by identifying new domains that are important for patients, and provided the patient perspective in the development of core outcome sets and the development of patient reported outcome measures.
- This study demonstrates that patients can play a valuable role as collaborative partners in a scientific conference on outcome research.

Strengths and limitations of this study

- Triangulation of the combined review of the document analysis and interviews, together with the active involvement of all team members representing a variety of perspectives in the phase of data analysis and interpretation, have enhanced the rigor of the study.
- The unique context of the OMERACT conferences limits the generalizability of the study results and makes comparable evaluation studies recommendable.

ABSTRACT

Objective – To assess the impact of patients as international research partners by analysis of the inclusion of patients in OMERACT conferences and how this has changed the scope and conduct of outcomes research.

Methods – A thematic content analysis of Outcome Measures in Rheumatology (OMERACT) internal documents, publications and conference proceedings, followed by interviews with 16 patient participants and 16 professional participants from research, pharmaceutical industry and regulatory backgrounds.

Results – The role of patients evolved over 10 years from a single patient focus group to full participation in all areas of the meeting and inclusion in research groups meeting between conferences. Five main categories of impact emerged: widening the research agenda; including patient relevant outcomes in core sets; enhancing patient reported instruments; changing the culture of OMERACT; and consequences outside OMERACT. Patient participants identified previously neglected outcome domains such as fatigue, sleep disturbances and flares which prompted collaborative working on new programmes of research. Specific benefits and challenges for patients and professionals were identified, such as personal fulfilment, widening of research interests, difficulties in establishing equal partnerships, and concerns about loss of research rigour.

Conclusions – Including patients as partners in an international research initiative changes its focus and way of working. For OMERACT it has resulted in new developments in the research agenda and the use of more patient relevant outcomes in clinical trials. These collaborations have changed perceptions and beliefs and led to wider patient involvement as partners in research.

Incorporating the patient perspective in health care research is strongly promoted by policy makers,[1-4] funding bodies and international regulators. Many theoretical benefits from patient involvement in research have been reported, [5-8] such as improving the relevance of research questions, improving recruitment of study participants, and increasing chances for funding and dissemination of results. In addition there is an increasing recognition of the essential role of patients in outcome research.[9]. The FDA has made patient involvement mandatory in the process of the development of patient reported outcome measures[10 11] and in the context of COMET (Core Outcome Measures in Effectiveness Trials) patient contributions are seen as crucial in defining domains that are relevant to include in core outcome sets for clinical trials.[12] Development of core outcome sets might lead to less variety of incomparable and inappropriate outcome measures, more patient oriented endpoints and less bias by selective reporting of only positive or statistically relevant outcomes.[13] Core outcome sets may ease the work of systematic reviewers in synthesizing the results of multiple studies.[14 15] The question is however whether these theoretical benefits of patient involvement in outcome research makes any difference in practice?

The international group Outcome Measures in Rheumatology (OMERACT), which defines core outcome sets in rheumatic diseases, first included patient participants at its 6th bi-annual conference in 2002 and has continued to do so. This provides an opportunity to analyse the consequences and address the important question of whether patient participation has resulted in any demonstrable impact on the nature of its research activity.

Patient involvement in OMERACT has been presented as a presumed success and the 2002 conference report concluded that “the preliminary success of this forum” was the basis for “continued and possibly expanded patient participation at the next OMERACT meeting”.[16] Two conferences later others perceived the involvement of patients as “indicative of the beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.[17]

Since then OMERACT has formulated three principles recognising the essential role of patients in outcome research.[18] First, patients' input is indispensible when defining relevant outcome measures, identifying domains that are important from the perspective of patients, and assessing feasibility of measurement tools. Second, structural involvement of patients during the whole research process provides face validity. Third, OMERACT intends "to ground theoretical discussions in the lived experience of arthritis, and in concepts which can be readily communicated to patients to help with therapeutic decision making".[18]

However, the validity of these arguments has never been substantiated by robust evidence for the effectiveness of patient participation and it is not clear whether or how this involvement has influenced methodologies, procedures, attitudes, and research outcomes. Therefore, the objective of this study is to describe and evaluate the contributions made by patients since OMERACT started implementing structural patient participation in its conferences. We review the impact of patients on the research agenda and the development of patient reported outcomes (PRO's) and explore how including patients has influenced the culture and structure of the OMERACT conference through the attitudes, beliefs and perceptions of participants.

Method

An initial thematic document analysis was carried out including OMERACT conference proceedings and 'grey literature' such as correspondence, invitations, session reports, e-mails and OMERACT policy documents. The review focused on the arguments, reception and evolution of patient involvement in OMERACT conferences and the contributions made by patients.

Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010) using qualitative interviews with representatives of stakeholders. This approach aims to explore different meanings that participants attribute to patient involvement.[19] It samples all stakeholders and does not seek consensus, but respects different opinions, values and interests. This ensures that no perspective is omitted as the result of an imbalance of power.

The first author held 32 semi-structured interviews before, during and after the conference (table 1) and included senior (n=10) and junior researchers (n=2), representatives of pharmacological industry and regulators (n=2), staff (n=2), new patient participants (n=8) and experienced patient participants (n=8) . The interviewees were invited by e-mail. The patient participants were aware of the purpose of the study through a one page announcement in the patient pack that was distributed two weeks in advance of the conference.

Twenty-eight interviews were recorded, transcribed by an independent secretariat and subjected to a responder check. Three interviews were summarized in a report, one interview took place without protocol and, on request of the interviewee, without recording (PF). One interview was done through Skype (PP). The average duration of the interviews was 52 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-four interviews were held in English of which 6 was not the native language. Eight interviews were in Dutch.

The interview protocols were slightly different for professionals, new patients and experienced patients. The topics were derived from the thematic document analysis and pilot interviews with 2 experienced patient participants and 2 researchers (a senior and a junior). The topics dealt with: the expected role of patient participants; their selection, preparation and support; and with the expected or provided contribution to the OMERACT conference.

Selection of interviewees - At OMERACT 10 a total of 172 delegates participated, 152 professionals and 20 patients. Nine patients attended the conference for the first time. Selection of interviewees aimed at maximum variation and followed an emergent purposive sampling approach taking into account stakeholder background, opinion about patient involvement, gender, geographical spread and number of OMERACT conferences attended. Except for one patient participant from the hosting country, all interviewees were selected by the first and last author. Because of the richness of the interviews, and in order to achieve saturation, extra interviews were held during the conference and seven other interviews took place within two weeks after the conference.

Although 2 interviewees were chosen because of their previously reported criticism of involving patients, the outcome of the interviews with these professionals was that they expressed a change in perception of patient involvement in a more positive way. For this reason 2 more interviews with professionals who had expressed critical comments during the last conference were arranged. Finally, to ensure the opinions of young investigators, 2 OMERACT Fellows were approached, one undertaking a PhD in translational research and one post doctoral researcher active in clinical research. In total the perceptions and experiences of 16 patient participants and 16 professionals were collected (table 1).

[insert table 1]

Data-analysis - A thematic content analysis focused in particular on the reported contributions attributed to patient participants. Coding of the interviews was done separately by the first author and an independent second coder (third author) who had never worked with active patient involvement before. This resulted in 211 detailed codes that were then combined into 27 sub-categories. During several meetings, the project team, representing various backgrounds, discussed the codes and subcategories from a variety of perspectives, and sought natural groupings or categories within the data. Triangulation took place by synthesizing interview data and results from the document analysis. To increase the relevance and validity of the analysis and interpretation of the data one of the patients who attended OMERACT 10 for the first time was invited to join the research team. As a patient research partner [20] she was involved in the coding, data analysis and data interpretation to guarantee the patient perspective. To protect the anonymity of the participants all quotes are presented in the “she”-form. Quotes of professional researchers are indicated by ‘R’ and those of patient research partners (in short: ‘partners’) by ‘P’.

Results

Document analysis: History of patient involvement at OMERACT

OMERACT started in 1992 as an initiative to overcome the problem of widespread and inconsistent use of many different outcome measures in rheumatoid arthritis (RA) clinical trials. The objective was to improve “the accuracy and responsiveness to change of clinically relevant (to patient and clinician) endpoints”.[21] Rheumatologist from many countries met in Maastricht and achieved consensus on a core set of outcomes for RA. The RA core set was endorsed by the WHO.[22] The initial stand alone conference was sufficiently successful that it was followed by conferences in alternate years continuing the discussion and consensus building about new core sets for other rheumatic diseases and new measurement instruments.

During the fifth OMERACT conference (2000) participants discussed the concept of a minimum clinically important difference (MCID). Based on methodological arguments a growing interest in patient reported outcomes emerged, culminating in a spontaneous proposal at the final session to invite patients to the next conference. All participants voted in favour of this proposal.[23] The chair of the conference felt confident about the proposal because it had been discussed in the organising committee before, although no decisions had been taken. Participants of the MCID module argued that patient perspectives should be explored further

[24] and took responsibility for identifying 11 patients to join OMERACT 6 and to review the RA core set.

Our document analysis revealed the unconditional positive reception of patient delegates at OMERACT conferences, and partners confirmed that concerns regarding their involvement were misplaced. They felt their reception was extremely welcoming. “There was a tangible feeling of relief and a belief that patients’ views and opinions would be listened to and incorporated into the deliberations”.[25] Also the organizers were excited and called the patient involvement “a tremendous success”.[26]

Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have participated as full delegates with equal voting rights.[27] Their role and contributions have developed over time. At the first conference (2002) they formed an homogeneous group of people with RA with little or no experience in scientific research. The level of involvement in the conference in general was relatively low, support was not organized and the number of sessions patients attended was limited. Contributions centred on participation in the workshop discussions about the severity of fatigue and the definition of low disease activity, although there was a keynote speech at the opening ceremony.[25] In contrast, by OMERACT 11 (2012) the partners were a heterogeneous group with different rheumatic conditions and different levels of experience, competences and cultural background. They received a pre-conference information pack and were actively supported by a pre-conference dinner, a glossary, training sessions and a buddy system. They carried out a variety of tasks similar to professionals such as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout sessions and preparing consensus statements. Several partners became co-authors of peer-reviewed publications.

Interviews: Patient contributions to OMERACT meetings and outcome research

Interviewees reported a variety of contributions made by partners during the conference where they are an integral part of the deliberative and consensus-building process.[18] These examples are presented below and compared with the document analyses when appropriate. Because research in the domain of fatigue has been reported as the most illustrative example, the contributions in this area will be described in more detail. Using the methodology described above we identified 5 main categories from the comments made during the interviews with OMERACT participants (Table 2): Contributions to the research agenda; The development of core sets; The development of patient reported outcomes (PRO's); The culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of the challenges that emerged from the interviews.

Contributions to the research agenda - From the very beginning partners had a significant influence on the research agenda in the field of rheumatology by participating in OMERACT workshops and small group discussions. They identified new outcome domains that are relevant from their perspective.[28] The first Patient Perspective Workshop, attended by 11 patient participants and 41 professionals, focused on the development of "valid outcome instruments that incorporate the perspective of the patient and to prepare the evidence and arguments for their inclusion in the (RA) core set" [29]. The preconference paper pointed out the methodological and political challenges: How to elicit and incorporate preferences of patients in RCT's? [27] The workshop had been specifically arranged to support the partner contributions including a pre- and post-workshop meeting. The workshop identified subjective experiences of RA, not encompassed in the RA core set but important consequences of the disease: a sense of well being, fatigue, and disturbed sleep.[29]

After the first conference attended by partners, it became apparent that perspectives of professionals and patients differ and more research was needed to articulate patients priorities.[30-33] Partners emphasized the need for an holistic approach to people with arthritis.[25] The acknowledgement of the discordance of perspectives initiated new studies looking into the preferences, opinions and experiences of people with rheumatic diseases [34-36] and developing patient-derived core sets.[37 38] This made participants more aware of the emerging patient perspective: “the whole realm of things we haven’t looked at” [RA]. New topics emerged: remission, pain, flares and foot problems. One interviewee clearly stated that partners “inspired me for new projects to study the variety in new productivity outcome measures” [RK].

Case-study of fatigue - Since 2002 when partners identified new topics for research, studies have been initiated with firm involvement of partners in the field of sleep disturbances, flares and well being. The most progress has been made in fatigue and the emergence of fatigue as a relevant outcome measure in RA provides an illustrative case history. When asked for the greatest benefit of including partners in OMERACT conferences interviewees unanimously confirmed that the topic of fatigue would not have been on the research agenda without partners expressing their concerns about fatigue as an often neglected symptom of their disease and without the listening of receptive professionals. One of the partners attending OMERACT 6 recalled:

> “I can’t remember who brought up the subject, but someone mentioned fatigue. And that was the occasion when one of the other delegates said ‘well, everybody gets tired’. One patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]

Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and quantitative research. The first studies investigated the prevalence and severity of fatigue in RA and how patients describe their fatigue.[39-42]. The next step comprised a systematic review of measurement instruments for fatigue [43] that explored the rigor of existing measurement tools and the need to develop patient-derived instruments that are trustworthy, capturing concepts and language of patients. Furthermore a standardized visual analogue scale, opportunities for electronic gathering of data and exploring mechanisms of fatigue that could guide researchers in the development of effective interventions, were added to the research agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a consequence of RA, but an independent variable that adds new information to the existing RA core set.[44 45] This new perception resulted in the acceptance of fatigue as an important outcome for clinical trials.[46 47] Fatigue was subsequently added to the RA core set as a recommended outcome.

More powerful instruments for measuring fatigue in RA have since been devised and validated, starting from the perspective of the patients.[48 49] Nicklin described precisely the role and contributions of patients in different phases of the development of patient reported outcome measures for fatigue, including the influence of a patient research partner providing practical insights into everyday life with RA at every stage of question development.[50] Patients made significant contributions in pilot interviews by discussing “measurement properties of wording, time-frame and descriptors” and articulating different meanings of words like *cope* and *manage*. Their involvement improved the final data collection and has also been particularly valuable for developing new intervention programs. Outside OMERACT other researchers initiated similar studies, focusing on the communication between patients and health professionals in the consultation room.[51] Based on these new insights intervention studies are now undertaken where fatigue is the primary endpoint[52 53].

This case-study of fatigue shows that it took more than a decade to develop effective interventions after identifying a new domain important from the perspective of patients.

The thematic document analysis provided additional evidence for the statement that without patients raising their voice at OMERACT 6 fatigue would not have been high on the research agenda. The issue of fatigue was not new for rheumatologists.[54-56] Fatigue was a symptom regularly reported during clinical consultations, but not incorporated in guidelines for monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and incorporated in a disease status questionnaire.[57] And during OMERACT 3 (1996) delegates carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal diseases. The discussion groups identified outcomes such as pain, depression, anxiety and fatigue as major concerns.[58] For fatigue eight examples of measurement instruments were given.[59]. However, after this workshop, nothing happened for six years, until patients raised the urgency of fatigue as a serious symptom.

Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only hearing from partners at OMERACT made them change their perception of fatigue as an important outcome:

> “Because when I was working in oncology before, during university training, of course we saw that the patients were lying in bed all day and we knew they were exhausted, call that fatigue. But patients with RA, we were ignorant.” [RC]

Another physician, involved in OMERACT from the start:

> "We were first discussing on fatigue and to be honest: I never ever had before heard of fatigue being a problem in rheumatology. So it got into my mind and then I got thinking about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a 100% positive response. So it was like a coming out, you know. I listened to the patients before but bringing it to a specific topic, that was really what I learned at OMERACT." [RA]

[Insert table 2]

The development of core sets and patient reported outcomes – During the first two conferences including patients the focus of partners' contributions was on agenda-setting and identifying relevant outcomes for clinical trials. Then, partners started contributing by identifying domains that are relevant for disease-specific core sets for psoriatic arthritis, fibromyalgia, gout and vasculitis. Furthermore they contributed to the development of core sets for methodological or clinical concepts like MCID and remission. Partners have played an important role in the assessment of the feasibility of instruments and core sets, one of the three key components of the OMERACT Filter.[60]

Partners have been helpful in the development of PRO measurement instruments in the field of work productivity, monitoring adverse events, flares and psychosocial interventions such as self-management programs. At the 2010 conference, during the plenary session on flares in RA, one of the partners gave a personal testimony about the devastating impact of the unpredictable nature of RA. A professional in the audience was surprised and reported: "It demonstrates that the disease activity fluctuates more than we can see in our data: Our instruments are more flat, and by the limited frequency of measuring we filter fluctuations out".[RI]

Although there is a broad consensus that partners have been beneficial in the development of PRO's, several participants also reported the example of a single partner questioning the tolerability of MRI scanning times for people with ankylosing spondylitis. During one of the breakout sessions on imaging she reported not being able to lie still for 35 minutes in an MRI machine. This alerted the researchers to their assumption that only moving hurts and as a consequence the proposed scanning recommendations were adjusted to ensure shorter scanning times. This shows that partners can be helpful in the assessment of the feasibility of technical outcome measures.

Regulators require strong evidence for the effectiveness of new medicines by demonstrating accurately that they reduce structural progression as well as patient important outcomes. By doing both of these, developing standards for high quality imaging techniques and exploring new PRO's and translating them into valid and feasible measures, OMERACT has been extremely advantageous for the negotiations with regulators about the registration and relatively generous reimbursement of new biologic agents:

> "I think, to be really honest, the patient involvement process in OMERACT and the changes in outcome measurements and the use of them in the drug tests has made a real difference for so many patients." [RB]

The culture of OMERACT– In spite of the initial unanimous vote to invite patients, some researchers were concerned about changing the layout of the conference:

> "My original expectation of a limited contribution was based on fear that patients were not able to transcend their personal experience and to generalize … new stakeholders often don't have knowledge about clinometric." [RE]

In retrospect researchers explained that they deferred to the proposal in order to reflect a core principle of OMERACT of not immediately rejecting new ideas: "To respect and listen rather than just react".[RA]

Looking back, professional participants were enthusiastic about the advent of partners at the conference: "I was impressed by the very good working flow".[RC] Participants confirmed that the presence of partners has changed their way of thinking and talking. "They made my blind spot visible" [RK] and another professional reported:

> "Now what we have found is, and I changed my view, [be]cause it wasn't only from OMERACT. As I got to know more and more patients, I realized, this sounds stupid because it's so obvious but it wasn't obvious to me, that a patient isn't their disease. A patient is a person who happens to have a disease. What a big difference. Because if you're a person that happens to have a disease, then for example you might have incredible skills in an area that might be very useful to move a clinical trial forward. So once I came to that realization then patient involvement becomes an absolutely obvious and integral part of moving forward." [RA]

Partners improved communication and brought dynamics to the dialogue because they are motivated and constructive, without a personal agenda. At a conference such as OMERACT, where the discussion about methodology may become extremely technical, partners reminded participants of the common goal of the conference by providing a human face of a person living with the condition day by day. Their presence made participants more explicit about the objectives of sessions and more explanatory about terms and concepts under discussion. Together with a reduced use of jargon this ‘forced’ simplification resulted in fewer misunderstandings for everyone.

For some professionals the presence of partners complicated the communication. Some believed that partners slow down the process because they are not familiar with technical issues. Others felt disinclined to say what they wanted to out of respect to partners or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and put her own expertise aside to keep things simple: “Patients didn’t sometimes understand the objective of the research, which hindered us”. [RK] One of the partners admitted that “it is a thin line between providing input and causing irritation”. [PN]

An analysis of the responses of patients attending OMERACT for the first time showed that new partners experienced a significant learning curve and a variety of personal benefits.[61] Results from this study suggest that in fact all participants learned from the contact with other stakeholders. During this process participants gained trust, respect and understanding, reflecting the emergence of relational empowerment:

> “Patients were a kind of sparring partner when I entered a relatively new area. That was fun and did clarify a lot”. [RK]

Relational empowerment in the context of health research can be understood as a process in which traditional doctor-patient relationships transform into equal partnerships enabling mutual learning processes.[62] All participants become stronger by sharing knowledge and responsibilities, and educating and helping each other.

The reported benefits were easiest to identify at the beginning when the level of involvement was still low. They became more diffuse when partners were structurally involved as full and equal collaborators. One interviewee mentioned “a reality check” as an important benefit of partners attending the conference. For professionals it offered the opportunity to check the relevance of the scope of their research: Are we doing the right things according to patients and are we using the right tools and methods? It is a belief of professionals that this kind of feedback is important to legitimize their research and, together with the belief of partners that without this research no innovations will take place, it strengthened the mutual empowerment of both.

Consequences outside OMERACT – The lessons learnt at OMERACT were noticed by the outside world. Partners returning home after the conference have continued introducing patient participation in local and national research projects or established networks of patient research partners.[63-66] Some delegates published a working framework for incorporating the patient perspective in outcome research.[67] With the input from several OMERACT participants the European League Against Rheumatism (EULAR) developed recommendations for the inclusion of patient representatives in scientific projects.[20] Following these recommendations a new patient reported quality of life instrument for RA was developed and validated.[37 68] Based on the experiences of OMERACT the organizing committee of the 6th International Shared Decision Making conference decided in 2011 to invite patient participants.[69] In the same year OMERACT delegates, partners as well as professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials (COMET) conference, demonstrating how the OMERACT methodology can be utilized in other disease areas.[12]

Remaining challenges emerging from the interviews – The role and contribution of patient participants have changed over time and procedures for patient selection and support have been developed in order to identify patient participants who are able to make a difference. There is still a debate going on whether patients should be selected through strict criteria such as education, communication skills, attitude and familiarity with scientific research. Some argue that an expert meeting like OMERACT needs expert patients who have extended knowledge about methodologies of outcome research, and are able to provide a kind of aggregated patient input. At OMERACT this group represents a minority of delegates, who are reluctant to allocate the same rights and power to partners as to the professionals. The vast majority believes that many patients are able to contribute to an OMERACT conference and emphasizes that a heterogeneous group of partners in age, gender, condition, experience and cultural background are advantageous for the conference. They intend to develop full representative participation in all phases of research by including partners in working group activities between conferences. Finally, some participants point out the potential risks of partners who become too experienced. They appreciate the naive input as a patient, with a minimum of preparation and reflection. They assume that as soon as you start thinking about your contribution, you lose the unique, individual perspective and become a patient-expert who aligns too easily with professionals.

Professionals shared the opinion that partners need training, although they reported different ideas about the content and aims of such training. Experienced partners as well as novice researchers felt that any new participant has to learn the OMERACT objectives, culture and procedures first, before they can become fully productive, mostly at the second or third conference. This accords with the expectations of partners who attended OMERACT for the first time.[61]

Overview of findings

These results show that a decade of patient involvement has been successful and had a significant impact on various aspects of outcome research. Perspectives of patients are different from those of health professionals. Broad consensus exists that partners at OMERACT have played a vital role in identifying domains relevant from the perspective of patients and in developing new PROs such as fatigue, sleep quality, flares and work productivity. Especially in the area of fatigue we have shown that patient involvement on different levels and in different phases improves the quality of outcome research. By combining evidence-based knowledge of researchers and the experiential knowledge of patients, a synthesis of both kinds of knowledge has been achieved and documented. The benefits are assessment tools that accurately measure what really matters to patients, are formulated in understandable language and are user-friendly. Other benefits go beyond improving clinical outcome research and include improved communication, mutual empowerment, changed attitudes and substantial consequences outside OMERACT.

Discussion

We set out to describe and evaluate the contribution of patients as partners in outcome research, reviewing their impact on the research agenda in rheumatology and the culture and process of the OMERACT conference. The document analysis provided the recorded facts while the interviews allowed an exploration of intentions and attitudes. Since validated methodologies for measuring impact of including new stakeholders in the context of research are lacking, a responsive interview methodology seemed to be a good approach. However, there are several limitations involved in this methodology. First, this responsive study presents an ‘insider’ perspective of patient contributions. There is a strong belief within OMERACT that patient participation works, a belief that is nourished by the world-wide transition towards patient-oriented health care and health research.[70-72]. The assumption however that long term involvement of patients as equal partners guarantees sustainable inclusion of the patient perspective in outcome research complicates the evaluation process and makes it difficult to distinguish between expected, perceived and actual contributions. Many participants, not only partners, but also young researchers and other new-comers, are ignorant about their own contribution and may not see how their input is reflected in the final outcomes. Partners reported almost unanimously not being able to confirm substantial contributions during their participation but they believed they did. More experienced participants, mostly professionals, were less reluctant in reporting illustrative examples of patient contributions.

Second, in a dialogue and consensus-based conference such as OMERACT many factors contribute to the final outcomes. We found that when the level of involvement increased from consultation to collaboration, it became harder to attribute individual or group contributions in the final outcomes. Collaboration is a dynamic phenomenon and although the breakout sessions are the main body of the OMERACT conference, a lot of work is done before, after and between the formal conference sessions. Since tasks are equally performed by patients as by any other participant, the dialogue between patients and professionals takes place not only during the official program, but in all parts of the conference. The corridors are quite important in this respect. Because neither partners nor professionals act as a representative of any particular group or constituency it remains difficult to determine the influence of particular individuals in that process.

Finally, we assume that experiential knowledge, hidden in anecdotal stories, does have an impact that is often not claimed by patients nor perceived by professionals. Personal comments are normally not reported because they are not seen as a valuable and valid source of knowledge [73] and yet clear documentation of meetings is required to ensure that patients' contributions become visible.[74] Professionals focus on synthesizing data and may not notice that the dialogue with patients works like a reality check, generates new ideas or changes their beliefs, behaviour or perception. When partners appear to simply agree with the results presented at OMERACT it might look as if they do not have any contribution to make, but in fact they confirm the value of the work under discussion and provide face validity to the process. It is for this reason that most professionals appreciate the feedback and input from partners, although not all are aware of this reason. Realizing the importance of such a reality check is beneficial for the management of realistic expectations: do not expect innovative ideas, brilliant suggestions and new concepts when inviting partners to join research. Their contributions are often more subtle and need the attention of a modest and committed researcher to be noticed.

Despite these limitations, we believe that the results presented in this study are relevant and valid. Through an approach of responsive evaluation reliable answers have been obtained. No signals were identified to suggest that interviewees have simply given desirable answers, or just been friendly to the interviewer. Some interviewees have been rather critical, reporting several barriers for structural involvement of patients in research, but have always added constructive suggestions for improvement. Lessons learnt regarding the conditions for successfully engagement with patients will be published separately.[75] However, it is undeniable that within OMERACT there is a growing belief that patient involvement has been successful and brought a unique added value to the conference. Even those who were originally among the most skeptical participants now report that they have changed their perception about the expected contribution of patient research partners.

This study is conducted within the context of a scientific research conference in the field of rheumatology, a long term somatic condition. Our ability to generalize the findings is therefore limited and extrapolation to other research contexts or to other conditions should be done with care. In the COMET-initiative [12] health researchers try to develop a validated methodology for defining core outcome sets for clinical trials in all disease areas and can learn from the experiences of OMERACT.

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Competing Interests

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Contributorship

All authors have been involved in the process of data analysis and interpretation, and have equally contributed to the writing of this manuscript.

Data Sharing

No additional data are available.

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Table 1 Characteristics interviewees

		Professionals	Patient Research Partners	Interview Code
Sex (M:F)		12 : 4	7 : 9	
Professional background or Diagnosis		10 practicing rheumatologists 3 full time researchers 3 other professionals	10 rheumatoid arthritis 2 vasculitis 2 ankylosing spondylitis 1 fibromyalgia 1 gout	
Number of OMERACT conferences attended	1	5	8	PA to PF, PO, PP
	2	0	3	PG to PN
	3	1	1	
	4	4	4	
	≥5	6	0	
Interview in relation to OMERACT conference	Before	2	1	
	During	8	16	
	After	6	5	
Geographical spread		6 countries 2 continents	7 countries 4 continents	
Research Background		10 Senior Researchers		RA to RG, RJ, RK, RY
		1 Research Fellow		RH
		1 Post-doctoral researcher		RI
		2 Pharma representatives		DA, DD
		2 Staff members		DB, DC

Table 2

Main and sub-categories from the analysis of patient contributions to OMERACT meetings and outcome research since 2002

<table>
<tr><th colspan="5">IMPACT OF A DECADE OF PATIENT INVOLVEMENT IN OMERACT</th></tr>
<tr><th>Research Agenda</th><th>Outcome core sets</th><th>Patient reported outcomes</th><th>Culture of OMERACT</th><th>Consequences outside OMERACT</th></tr>
<tr><td>Generating challenging ideas
Identification of patient-relevant research topics:
• Well being
• Fatigue
• Sleep disturbance
• Flares</td><td>Identification of patient relevant domains to include in core sets for clinical trials:
• Fibromyalgia
• Psoriatic Arthritis
• Vasculitis
• Gout
• MRI
• MCID
• Remission</td><td>Acceptable, understandable and feasible outcome measures for
• Monitoring adverse events
• Work productivity
• Flares
• Psychosocial interventions</td><td rowspan="2">• Attitudes
• Communication
• Perceptions
• Motivation
• Relational empowerment
• Personal benefits</td><td rowspan="2">• Local initiatives
• Local and national networks of partners
• EULAR
• COMET
• ISDM</td></tr>
<tr><td colspan="3">Reality check</td></tr>
</table>

Abbreviations

COMET	Core Outcome Measures in Effectiveness Trials
EULAR	European League Against Rheumatism
ISDM	International Shared Decision Making
MCID	Minimal Clinically Important Difference
MRI	Magnetic Resonance Imaging
OMERACT	Outcome Measurement in Rheumatology
PRO	Patient Reported Outcome
RA	Rheumatoid Arthritis

Involving patient research partners has a significant impact on outcomes research: an example from the international OMERACT conferences.

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Primary Subject Heading:	Rheumatology
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Article Summary

Article focus

- Since 2002 patients have participated as collaborative partners in the biannual conference on Outcome Measures in Rheumatology (OMERACT).
- Although the contribution of patients has been praised and there is a wide-spread call for scientific publications on the impact of engaging with patients, no systematically obtained evidence has been published to support the idea that structural involvement of patients in research conferences is beneficial.
- Our qualitative study reports the combined results of a thematic document analysis and 32 semi structured interviews with all stakeholders including researchers, patient participants and representatives from pharmaceutical industry and international regulators.

Key messages

- Long term engagement with arthritis patients in OMERACT conferences has significantly influenced outcome research in the field of rheumatology.
- Patients have successfully contributed to the research agenda of OMERACT by identifying new domains that are important for patients, and provided the patient perspective in the development of core outcome measurement sets and the development of patient reported outcome measures.

Strengths and limitations of this study

- Triangulation of the combined review of the document analysis and interviews, together with the active involvement of all team members representing a variety of perspectives in the phase of data analysis and interpretation, have enhanced the validity of the study.

- The unique context of the OMERACT conferences may limit the generalizability of the results so comparable evaluation studies in other conference formats would be worthwhile.

Incorporating the patient perspective in health care research is strongly promoted by policy makers,[1-4] funding bodies and international regulators. Many theoretical benefits from patient involvement in research have been reported, [5-8] such as improving the relevance of research questions, improving recruitment of study participants, and increasing chances for funding and dissemination of results. In addition there is an increasing recognition of the essential role of patients in outcome research.[9] The USA Food and Drug Administration (FDA) has made patient involvement mandatory in the process of the development of patient reported outcome measures[10 11] and in the context of COMET (Core Outcome Measures in Effectiveness Trials) patient contributions are seen as crucial in defining domains that are relevant to include in core outcome measurement sets for clinical trials.[12] Development of core outcome sets might lead to less variety of incomparable and inappropriate outcome measures, more patient oriented endpoints and less bias by selective reporting of only positive or statistically relevant outcomes.[13] Core outcome measurement sets may ease the work of systematic reviewers in synthesizing the results of multiple studies.[14 15] The question is however whether these theoretical benefits of patient involvement in outcome research make any difference in practice.

The international group Outcome Measures in Rheumatology (OMERACT), which defines core outcome measurement sets in rheumatic diseases, first included patient participants at its 6th bi-annual conference in 2002 and has continued to do so. This provides an opportunity to analyse the consequences and address the important question of whether patient participation has resulted in any demonstrable impact on the nature of its research activity.

Patient involvement in OMERACT has been presented as beneficial and the 2002 conference report concluded that “the preliminary success of this forum” was the basis for “continued and possibly expanded patient participation at the next OMERACT meeting”.[16]

Two conferences later others perceived the involvement of patients as “indicative of the beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.[17] Since then OMERACT has formulated three principles recognising the essential role of patients in outcome research.[18] First, patients’ input is indispensable when defining relevant outcome measures, identifying domains that are important from the perspective of patients, and assessing feasibility of measurement tools. Second, structural involvement of patients during the whole research process provides face validity. Third, OMERACT intends “to ground theoretical discussions in the lived experience of arthritis, and in concepts which can be readily communicated to patients to help with therapeutic decision making”.[18]

However, the validity of these arguments has never been substantiated by robust evidence for the effectiveness of patient participation in OMERACT and it is not clear whether or how this involvement has influenced methodologies, procedures, attitudes, and research outcomes. Therefore, the objective of this study is to describe and evaluate the contributions made by patients since OMERACT started implementing structural patient participation in its conferences. We review the impact of patients on the research agenda and the development of patient reported outcomes (PRO’s) and explore how including patients has influenced the culture and structure of the OMERACT conference

Method

Patient participation in research is a new phenomenon and often not reported or reflected on in scientific publications. This lack of written sources in the scientific literature complicates the study of the process and impact of patient participation through a review of relevant literature. A provisory search using Pubmed (March 2010) for the terms “patient participation” OR “patient involvement” OR “user involvement” OR “consumer involvement” AND “OMERACT” did not generate any relevant reference. Therefore we conducted a content analysis of relevant documents (any written material on the topic of patient participation).

Documents are a stable, rich source of contextual information, providing well-grounded data on events or situations at low costs. A sound document analysis is rule-bound, systematic, following a coding process where raw data are aggregated into units describing the content.[19] We included OMERACT conference proceedings as published by The Journal of Rheumatology (1992-2010) and 'grey literature' such as correspondence, invitations, session reports, e-mails and OMERACT policy documents. The review focused on the arguments, reception and evolution of patient involvement in OMERACT conferences and the contributions made by patients.

Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010) using qualitative interviews with representatives of stakeholders. Responsive evaluation is grounded in the hermeneutic research tradition and is used by social scientists to interpret meanings that participants attribute to a phenomenon, here the history and impact of a decade of patient participation from the perspective of the conference delegates. It samples all stakeholders and does not seek consensus, but respects the plurality of opinions, values and interests. This ensures that no perspective is omitted as the result of an imbalance of power.[20]

The first author (MW) has been involved in OMERACT since 2002 as a patient participant. He has a rheumatic condition and has been educated as a responsive researcher. Characteristics for a responsive researcher are a multiple partiality and the intent to enhance mutual understanding among all stakeholders. The last author (JK) has been involved in OMERACT since the first conference (1992) and has been the leader of the patient perspective workshop between 2002-2012. Having witnessed the involvement of patients firsthand from the very beginning MW and JK provided useful information to start the research, yet it also alerted them to critically reflect how this engagement influenced the research, and how to prevent bias. Therefore two independent experts (TA, MK) were added to the team. They had no relations with the OMERACT conference and its participants, and TA acted as peer-debriefer discussing dilemmas and challenging methodological decisions with MW.[21]

The first author held 32 semi-structured interviews before, during and after the 10th conference (Table 1) and included senior (n=10) and junior researchers (n=2), representatives of pharmacological industry and regulators (n=2), conference staff (n=2), new patient participants (n=8) and experienced patient participants (n=8). The interviewees were invited and informed by e-mail. The patient participants were aware of the purpose of the study through a one page announcement in the pre-conference patient pack and were asked for informed consent. In the Netherlands no ethical approval is required for non-intrusive interviews only.

Twenty-eight interviews were recorded, transcribed by an independent secretariat and subjected to a responder check. Three interviews were summarized in a report, one interview took place without protocol and, on request of the interviewee, without recording (PF). One interview was done through Skype (PP). The average duration of the interviews was over 50 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-four interviews were held in English of which six was not the native language. Eight interviews were in Dutch.

The interview protocols were slightly different for professionals, new patients and experienced patients. The topics were not only derived from the document analysis but also from four pilot interviews and the personal knowledge of MW and JK and the expertise of TA. The topics dealt with: the expected role of patient participants, their selection, preparation and support, and the expected or provided contribution to the OMERACT conference. ‘Fatigue’ was added as a potential probe because publications had already shown that this topic deserved special attention with regard to our research questions.[22-24] Participants with long term experience in OMERACT were asked retrospectively to describe their memories of the discussions and decisions taken about patient participation before and after 2002. Their recollections might be characterized as 'oral history'.

Selection of interviewees - At OMERACT 10 a total of 172 delegates participated, 152 professionals and 20 patients. Nine patients attended the conference for the first time. All interviewees, except for one patient participant from the hosting country, were selected by MW and JK following an emergent purposive sampling approach.[21] They used a list of attendees provided by the congress agency, covering four out of five criteria found to be important (stakeholder background, gender, geographical spread and number of OMERACT conferences attended). The criterion 'opinion about patient involvement' was assessed on the basis of authors' insight of the participant as being 'positive' (e.g. contributing to the patient perspective workshop or involving partners in own activities), 'indifferent' or 'skeptical' (e.g. resistant, not collaborating with partners). When it became clear during the process of data collection that certain criteria were not well covered new participants were approached till maximum variation was realized. For example, two interviewees who were chosen because of their previously reported criticism of involving patients, showed a considerable change in perception of patient involvement in a positive way. For this reason two more interviews with professionals who had expressed critical comments during the last conference were arranged. Finally, to ensure the opinions of young investigators, two OMERACT Fellows were approached, one undertaking a PhD in translational research and one post doctoral researcher active in clinical research.

Saturation was defined as a repetition of data; theoretical saturation as achieving sufficiently robust empirical data to support and describe the identified themes and main categories. Saturation was discussed and agreed within the research team. In total the perceptions and experiences of 16 patient participants and 16 professionals were collected (Table 1).

[insert Table 1]

Data-analysis - A thematic content analysis focused in particular on the reported contributions attributed to patient participants. Coding of the interviews was done separately by MW and an independent second coder (MK) who had never worked with active patient involvement before. This resulted in 211 detailed codes that were then combined into 27 sub-categories. During several meetings, the project team, representing various backgrounds, discussed the codes and subcategories from a variety of perspectives, and sought natural groupings or categories within the data. Triangulation was used in two different meanings: First, as a means of verifying findings against another source (interview) or another method (document analysis) and to enhance the validity of the data. Second, as a means to enrich the data collection and improve the face-validity by synthesizing findings from the document analysis with the personal memories and experiences of respondents who looked back in time. By doing so, gaps in the document analysis could be filled in.

The relevance and validity of the analysis and interpretation of the data was increased by the involvement of an external expert in qualitative health research (TA) as well as by inviting one of the patients (SC) who attended OMERACT 10 for the first time to join the research team. As a patient research partner[25] she was involved in the coding, data analysis and data interpretation to guarantee the patient perspective. To protect the anonymity of the participants all quotes are presented in the “she”-form. Quotes of professional researchers are indicated by ‘R’ and those of patient research partners (in short: ‘partners’) by ‘P’.

Results

History of patient involvement at OMERACT

OMERACT started in 1992 as an initiative to overcome the problem of widespread and inconsistent use of many different outcome measures in rheumatoid arthritis (RA) clinical trials. The objective was to improve “the accuracy and responsiveness to change of clinically relevant (to patient and clinician) endpoints”.[26] Rheumatologist from many countries met in Maastricht and achieved consensus on a core set of outcomes for RA. The RA core set was endorsed by the WHO.[27] The initial stand alone conference was sufficiently successful that it was followed by conferences in alternate years continuing the discussion and consensus building about new core sets for other rheumatic diseases and new measurement instruments.

During the fifth OMERACT conference (2000) participants discussed the concept of a minimum clinically important difference (MCID). Based on methodological arguments a growing interest in patient reported outcomes emerged, culminating in a spontaneous proposal at the final session to invite patients to the next conference. All participants voted in favour of this proposal.[28] The chair of the conference felt confident about the proposal because it had been discussed in the organising committee before, although no decisions had been taken. Participants of the MCID module argued that patient perspectives should be explored further [29] and took responsibility for identifying 11 patients to join OMERACT 6 and to review the RA core set.

Our document analysis revealed the unconditional positive reception of patient delegates at OMERACT conferences, and partners confirmed that concerns regarding their involvement were misplaced. They felt their reception was extremely welcoming. “There was a tangible feeling of relief and a belief that patients' views and opinions would be listened to and incorporated into the deliberations".[30] Also the organizers were excited and called the patient involvement "a tremendous success".[31]

Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have participated as full delegates with equal voting rights.[32] Their role and contributions have developed over time. At the first conference (2002) they formed an homogeneous group of people with RA with little or no experience in scientific research. The level of involvement in the conference in general was relatively low, support was not organized and the number of sessions patients attended was limited. Contributions centred on participation in the workshop discussions about the severity of fatigue and the definition of low disease activity, although there was a keynote speech at the opening ceremony.[30] In contrast, by OMERACT 11 (2012) the partners were a heterogeneous group with different rheumatic conditions and different levels of experience, competences and cultural background. They received a pre-conference information pack and were actively supported by a pre-conference dinner, a glossary, training sessions and a buddy system. They carried out a variety of tasks similar to professionals such as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout sessions and preparing consensus statements. Several partners became co-authors of peer-reviewed publications.

Patient contributions to OMERACT meetings and outcome research

Interviewees reported a variety of contributions made by partners during the conference where they are an integral part of the deliberative and consensus-building process.[18] These examples are presented below and compared with the document analyses when appropriate. Because research in the domain of fatigue has been reported as the most illustrative example, the contributions in this area will be described in more detail. Using the methodology described above we identified 5 main categories from the comments made during the interviews with OMERACT participants (Table 2): Contributions to the research agenda; The development of core sets; The development of patient reported outcomes (PRO's); The culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of the challenges that emerged from the interviews.

Contributions to the research agenda - From the very beginning partners had a significant influence on the research agenda in the field of rheumatology by participating in OMERACT workshops and small group discussions. They identified new outcome domains that are relevant from their perspective.[33] The first Patient Perspective Workshop, attended by 11 patient participants and 41 professionals, focused on the development of "valid outcome instruments that incorporate the perspective of the patient and to prepare the evidence and arguments for their inclusion in the (RA) core set" [24]. The preconference paper pointed out the methodological and political challenges: How to elicit and incorporate preferences of patients in RCT's? [32] The workshop had been specifically arranged to support the partner contributions including a pre- and post-workshop meeting. The workshop identified subjective experiences of RA, not encompassed in the RA core set but important consequences of the disease: a sense of well being, fatigue, and disturbed sleep.[24]

After the first conference attended by partners, it became apparent that perspectives of professionals and patients differ and more research was needed to articulate patients priorities.[34-37] Partners emphasized the need for an holistic approach to people with arthritis.[30] The acknowledgement of the discordance of perspectives initiated new studies looking into the preferences, opinions and experiences of people with rheumatic diseases [22 38 39] and developing patient-derived core sets.[40] This made participants more aware of the emerging patient perspective: "the whole realm of things we haven't looked at" [RA]. New topics emerged: remission, pain, flares and foot problems. One interviewee clearly stated that partners “inspired me for new projects to study the variety in new productivity outcome measures” [RK].

Case-study of fatigue - Since 2002 when partners identified new topics for research, studies have been initiated with firm involvement of partners in the field of sleep disturbances, flares and well being. The most progress has been made in fatigue, and the emergence of fatigue as a relevant outcome measure in RA provides an illustrative case history. When asked for the greatest benefit of including partners in OMERACT conferences interviewees unanimously confirmed that the topic of fatigue would not have been on the research agenda without partners expressing their concerns about fatigue as an often neglected symptom of their disease and without the listening of receptive professionals. One of the partners attending OMERACT 6 recalled:

> “I can’t remember who brought up the subject, but someone mentioned fatigue. And that was the occasion when one of the other delegates said ‘well, everybody gets tired’. One patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]

Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and quantitative research. The first studies investigated the prevalence and severity of fatigue in RA and how patients describe their fatigue.[41-44]. The next step comprised a systematic review of measurement instruments for fatigue [45] that explored the rigor of existing measurement tools and the need to develop patient-derived instruments that are trustworthy, capturing concepts and language of patients. Furthermore a standardized visual analogue scale, opportunities for electronic gathering of data and exploring mechanisms of fatigue that could guide researchers in the development of effective interventions, were added to the research agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a consequence of RA, but an independent variable that adds new information to the existing RA core set.[46 47] This new perception resulted in the acceptance of fatigue as an important outcome for clinical trials.[48 49] Fatigue was subsequently added to the RA core set as a recommended outcome.[50]

More powerful instruments for measuring fatigue in RA have since been devised and validated, starting from the perspective of the patients.[51 52] Outside OMERACT researchers initiated similar studies, focusing on the communication between patients and health professionals in the consultation room.[53]

The thematic document analysis provided additional evidence for the statement that without patients raising their voice at OMERACT 6 fatigue would not have been high on the research agenda. The issue of fatigue was not new for rheumatologists.[54-56] Fatigue was a symptom regularly reported during clinical consultations, but not incorporated in guidelines for monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and incorporated in a disease status questionnaire.[57] And during OMERACT 3 (1996) delegates carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal diseases. The discussion groups identified outcomes such as pain, depression, anxiety and fatigue as major concerns.[58] For fatigue eight examples of measurement instruments were given.[59]. However, after this workshop, nothing happened for six years, until patients raised the urgency of fatigue as a serious symptom.

Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only hearing from partners at OMERACT made them change their perception of fatigue as an important outcome:

> "Because when I was working in oncology before, during university training, of course we saw that the patients were lying in bed all day and we knew they were exhausted, call that fatigue. But patients with RA, we were ignorant." [RC]

Another physician, involved in OMERACT from the start:

> "We were first discussing on fatigue and to be honest: I never ever had before heard of fatigue being a problem in rheumatology. So it got into my mind and then I got thinking about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a 100% positive response. So it was like a coming out, you know. I listened to the patients before but bringing it to a specific topic, that was really what I learned at OMERACT." [RA]

[Insert Table 2]

The development of core outcome measurement sets and patient reported outcomes – During the first two conferences including patients the focus of partners' contributions was on agenda-setting and identifying relevant outcomes for clinical trials. Then, partners became gradually involved on different levels in other OMERACT activities, varying from being consulted (e.g. in a Delphi process) to full collaboration (as partner and as co-author). They contributed by identifying domains that are relevant for disease-specific core sets for psoriatic arthritis,[60] fibromyalgia,[61 62] gout[63] and vasculitis.[64] Furthermore they contributed to the development of core outcome measurement sets for methodological or clinical concepts such as MCID and remission.[65] Partners have also played a role in the assessment of the feasibility of instruments and core sets, one of the three key components of the OMERACT Filter.[66]

Partners have been helpful in the development of PRO measurement instruments in the field of work productivity, monitoring adverse events,[67] flares[68] and psychosocial interventions such as self-management programs[69]. At the 2010 conference, during the plenary session on flares in RA, one of the partners gave a personal testimony about the devastating impact of the unpredictable nature of RA. A professional in the audience was surprised and reported: "It demonstrates that the disease activity fluctuates more than we can see in our data: Our instruments are more flat, and by the limited frequency of measuring we filter fluctuations out".[RI]

Regulators require strong evidence for the effectiveness of new medicines by demonstrating accurately that they reduce structural progression as well as patient important outcomes. By doing both of these, developing standards for high quality imaging techniques and exploring new PRO's and translating them into valid and feasible measures, OMERACT has been extremely advantageous for the negotiations with regulators about the registration and relatively generous reimbursement of new biologic agents:

> "I think, to be really honest, the patient involvement process in OMERACT and the changes in outcome measurements and the use of them in the drug tests has made a real difference for so many patients." [RB]

The culture of OMERACT– In spite of the initial unanimous vote to invite patients, some researchers were concerned about changing the layout of the conference:

> “My original expectation of a limited contribution was based on fear that patients were not able to transcend their personal experience and to generalize … new stakeholders often don’t have knowledge about clinometric.” [RE]

In retrospect researchers explained that they deferred to the proposal in order to reflect a core principle of OMERACT of not immediately rejecting new ideas: “To respect and listen rather than just react”.[RA]

Looking back, the number of professional participants who were in favour of partners at the conference slowly increased: “I was impressed by the very good working flow”.[RC] Participants confirmed that the presence of partners has changed their way of thinking and talking. “They made my blind spot visible” [RK] and another professional reported:

> “Now what we have found is, and I changed my view, [be]cause it wasn’t only from OMERACT. As I got to know more and more patients, I realized, this sounds stupid because it’s so obvious but it wasn’t obvious to me, that a patient isn’t their disease. A patient is a person who happens to have a disease. What a big difference. Because if you’re a person that happens to have a disease, then for example you might have incredible skills in an area that might be very useful to move a clinical trial forward. So once I came to that realization then patient involvement becomes an absolutely obvious and integral part of moving forward.” [RA]

Partners improved communication and brought dynamics to the dialogue because they are motivated and constructive, without a personal agenda. At a conference such as OMERACT, where the discussion about methodology may become extremely technical, partners reminded participants of the common goal of the conference by providing a human face of a person living with the condition day by day. Their presence made participants more explicit about the objectives of sessions and more explanatory about terms and concepts under discussion. Together with a reduced use of jargon this 'forced' simplification resulted in fewer misunderstandings for everyone.

For some professionals the presence of partners complicated the communication. Some believed that partners slow down the process because they are not familiar with technical issues. Others felt disinclined to say what they wanted to out of respect to partners or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and put her own expertise aside to keep things simple: "Patients didn't sometimes understand the objective of the research, which hindered us". [RK] One of the partners admitted that "it is a thin line between providing input and causing irritation". [PN]

An analysis of the responses of patients attending OMERACT for the first time showed that new partners experienced a significant learning curve and a variety of personal benefits.[70] Results from this study suggest that in fact all participants learned from the contact with other stakeholders. During this process participants gained trust, respect and understanding, reflecting the emergence of relational empowerment:

> "Patients were a kind of sparring partner when I entered a relatively new area. That was fun and did clarify a lot". [RK]

Relational empowerment in the context of health research can be understood as a process in which traditional doctor-patient relationships transform into equal partnerships enabling mutual learning processes.[71] All participants become stronger by sharing knowledge and responsibilities, and educating and helping each other.

The reported benefits were easiest to identify at the beginning when the level of involvement was still low. They became more diffuse when partners were structurally involved as full and equal collaborators. One interviewee mentioned “a reality check” as an important benefit of partners attending the conference. For professionals it offered the opportunity to check the relevance of the scope of their research: Are we doing the right things according to patients and are we using the right tools and methods? It is a belief of professionals that this kind of feedback is important to legitimize their research and, together with the belief of partners that without this research no innovations will take place, it strengthened the mutual empowerment of both.

Consequences outside OMERACT – The lessons learnt at OMERACT were noticed by the outside world. Partners returning home after the conference have continued introducing patient participation in local and national research projects or established networks of patient research partners.[72-75] Some delegates published a working framework for incorporating the patient perspective in outcome research.[76] With the input from several OMERACT participants the European League Against Rheumatism (EULAR) developed recommendations for the inclusion of patient representatives in scientific projects.[25] Following these recommendations a new patient reported quality of life instrument for RA was developed and validated.[40 77] Based on the experiences of OMERACT the organizing committee of the 6th International Shared Decision Making conference decided in 2011 to invite patient participants.[78] In the same year OMERACT delegates, partners as well as professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials (COMET) conference, demonstrating how the OMERACT methodology can be utilized in other disease areas.[12]

Remaining challenges emerging from the interviews – The role and contribution of patient participants have changed over time and procedures for patient selection and support have been developed in order to identify patient participants who are able to make a difference. There is still a debate going on whether patients should be selected through strict criteria such as education, communication skills, attitude and familiarity with scientific research. Some argue that an expert meeting like OMERACT needs expert patients who have extended knowledge about methodologies of outcome research, and are able to provide a kind of aggregated patient input. At OMERACT this group represents a minority of delegates, who are reluctant to allocate the same rights and power to partners as to the professionals. The vast majority believes that many patients are able to contribute to an OMERACT conference and emphasizes that a heterogeneous group of partners in age, gender, condition, experience and cultural background are advantageous for the conference. They intend to develop full representative participation in all phases of research by including partners in working group activities between conferences. Finally, some participants point out the potential risks of partners who become too experienced. They appreciate the naive input as a patient, with a minimum of preparation and reflection. They assume that as soon as you start thinking about your contribution, you lose the unique, individual perspective and become a patient-expert who aligns too easily with professionals.

Professionals shared the opinion that partners need training, although they reported different ideas about the content and aims of such training. Experienced partners as well as novice researchers felt that any new participant has to learn the OMERACT objectives, culture and procedures first, before they can become fully productive, mostly at the second or third conference. This accords with the expectations of partners who attended OMERACT for the first time.

Overview of findings

These results show that a decade of patient involvement has been successful and had a significant impact on various aspects of outcome research. Perspectives of patients are different from those of health professionals. Broad consensus exists that partners at OMERACT have played a vital role in identifying domains relevant from the perspective of patients and in developing new PROs such as fatigue, sleep quality, flares and work productivity. Especially in the area of fatigue we have shown that patient involvement on different levels and in different phases improves the quality of outcome research. By combining evidence-based knowledge of researchers and the experiential knowledge of patients, a synthesis of both kinds of knowledge has been achieved and documented. The benefits are assessment tools that accurately measure what really matters to patients, are formulated in understandable language and are user-friendly. Other benefits go beyond improving clinical outcome research and include improved communication, mutual empowerment, changed attitudes and substantial consequences outside OMERACT.

Discussion

We set out to describe and evaluate the contribution of patients as partners in rheumatology outcome research, reviewing their impact on the research agenda and the culture and process of the OMERACT conference. The document analysis provided the recorded facts while the interviews allowed an exploration of intentions, attitudes and perceived benefits or harms of patient participation that complements the document analysis. Since validated methodologies for demonstrating impact of collaboration with patients in the context of research are lacking, a responsive interview methodology seemed to be a good approach.

Both strengths and limitations of this study relate to the personal experience of the first and last authors as participants in the developing process of patient partner participation. Having witnessed the OMERACT process, actors and concerns of both the patient community and the research community was advantageous during the development of interview protocols, recruitment and selection of respondents and data analysis. For instance, the knowledge of the opinions of other participants made it possible to achieve maximum variation. Also, the active involvement in the support and training of partners created an adequate awareness of the relevant items to include in the study. The drawbacks of this engagement are the risks of subjectivity, blind spots and over- or under-identification with particular stakeholders. These risks have been addressed by applying strict quality measures for scientific rigor in qualitative, evaluation research.

The composition of the research team purposely included two external experts in qualitative research and a patient research partner, who were actively involved in the coding of interview transcripts and distilling relevant categories for impact, reduced the risk of subjectivity. Bias was avoided by the check-coding procedure in the analysis of the transcripts as at least two researchers independently coded each transcript, after which the whole team discussed the codes until consensus was reached. Saturation was also part of the discussion in the whole team. The inclusion of various stakeholder perspectives prevented one-sidedness. No signals were identified to suggest that interviewees have simply given desirable answers, or just been friendly to the interviewer. Some interviewees have been rather critical, reporting several barriers for structural involvement of patients in research, but have always added constructive suggestions for improvement. Peer debriefing by an independent colleague (TA) further helped to prevent bias.

Other limitations relate to the difficulties of demonstrating the ‘impact’ of patient involvement.[9] In OMERACT there is a strong belief that patient participation works, a belief that is nourished by the world-wide transition towards more patient-oriented health care and health research.[79-82] The assumption however that long term involvement of patients as equal partners guarantees sustainable inclusion of the patient perspective in outcome research complicates a thorough evaluation and makes it difficult to distinguish between expected, perceived and actual contributions. Many participants, not only partners, but also young researchers and other new-comers, are able to identify their own contribution and may not see how their input is reflected in the final outcomes. Partners reported almost unanimously not being able to confirm substantial contributions during their participation but they believed they did. More experienced participants, mostly professionals, were less reluctant in reporting illustrative examples of patient contributions.

In a dialogue and consensus-based conference such as OMERACT many (f)actors contribute to the final outcomes. A linear causal relation between patient involvement and impact is therefore hard to establish; the processes of involvement are rather influenced by and influencing many (f)actors in a mutually interactive way. We found that when the level of involvement of partners increased from consultation to collaboration, it became harder to solely attribute individual or group contributions to the final outcomes. Because neither partners nor professionals act as a representative of any group or constituency it remains difficult to determine the influence of particular groups or individuals. Participation proved to be a dynamic process, especially when tasks were equally performed by patients and researchers, and when the dialogue between both took not only place during the official sessions, but also in the corridors of the conference.

A last obstacle for demonstrating the influence of patient participation is the invisibility of experiential knowledge, often hidden in anecdotal stories. It has an impact that is rarely claimed by patients nor perceived by professionals. Personal comments are normally not reported because they are not seen as a valuable and valid source of knowledge [83] and yet clear documentation of meetings is required to ensure that patients' contributions become visible.[84] Professionals focus on synthesizing data and may not notice that the dialogue with patients works like a reality check, generates new ideas or changes their beliefs, behaviour or perception. When partners appear to simply agree with the results presented at OMERACT it might look as if they do not have any contribution to make, but in fact they confirm the value of the work under discussion and provide face validity to the process. It is for this reason that most professionals appreciate the feedback and input from partners, although not all are aware of this reason. Realizing the importance of such a reality check is beneficial for the management of realistic expectations: do not expect innovative ideas, brilliant suggestions and new concepts when inviting partners to join research. Their contributions are often more subtle and need the attention of a modest and committed researcher to be noticed.

Despite these limitations, we believe that the results presented in this study are relevant and valid. It is undeniable that there is a growing belief that patient involvement has been successful and brought a unique added value to the conference. Even those who were originally among the most skeptical participants now report that they have changed their perception about the expected contribution of patient research partners. This study is conducted within the context of a scientific research conference in the field of rheumatology, a long term somatic condition. Our ability to generalize the findings is therefore limited and extrapolation to other research contexts or to other conditions should be done with care.

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Table 1 Characteristics interviewees

		Professionals	Patient Research Partners	Interview Code
Sex (M:F)		12 : 4	7 : 9	
Professional background or Diagnosis		10 practicing rheumatologists 3 full time researchers 3 other professionals	10 rheumatoid arthritis 2 vasculitis 2 ankylosing spondylitis 1 fibromyalgia 1 gout	
Number of OMERACT conferences attended	1	5	8	PA to PF, PO, PP
	2	0	3	PG to PN
	3	1	1	
	4	4	4	
	≥5	6	0	
Interview in relation to	Before	2	1	
	During	8	16	

OMERACT conference	After	6	5	
Geographical spread		6 countries 2 continents	7 countries 4 continents	
Research Background		10 Senior Researchers		RA to RG, RJ, RK, RY
		1 Research Fellow		RH
		1 Post-doctoral researcher		RI
		2 Pharma representatives		DA, DD
		2 Staff members		DB, DC

Table 2

Main and sub-categories from the analysis of patient contributions to OMERACT meetings and outcome research since 2002

IMPACT OF A DECADE OF PATIENT INVOLVEMENT IN OMERACT				
Research Agenda	Outcome core sets	Patient reported outcomes	Culture of OMERACT	Consequences outside OMERACT
Generating challenging ideas Identification of patient-relevant research topics: • Well being • Fatigue • Sleep disturbance • Flares	Identification of patient relevant domains to include in core sets for clinical trials: • Fibromyalgia • Psoriatic Arthritis • Vasculitis • Gout • MRI • MCID	Acceptable, understandable and feasible outcome measures for • Monitoring adverse events • Work productivity • Flares • Psychosocial interventions	• Attitudes • Communication • Perceptions • Motivation • Relational empowerment • Personal benefits	• Local initiatives • Local and national networks of partners • EULAR • COMET • ISDM

	• Remission			
Reality check				

Abbreviations

COMET	Core Outcome Measures in Effectiveness Trials
EULAR	European League Against Rheumatism
FDA	USA Food and Drug Administration
ISDM	International Shared Decision Making
MCID	Minimal Clinically Important Difference
MRI	Magnetic Resonance Imaging
OMERACT	Outcome Measurement in Rheumatology
PRO	Patient Reported Outcome
RA	Rheumatoid Arthritis

Involving patient research partners has a significant impact on outcomes research: an example from the international OMERACT conferences.

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Keywords:	HEALTH IMPACT ASSESSMENT, QUALITATIVE RESEARCH, RHEUMATOLOGY

Involving patient research partners has a significant impact on outcomes research: an example from the international OMERACT conferences.

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Keywords: HEALTH IMPACT ASSESSMENT , QUALITATIVE RESEARCH, RHEUMATOLOGY

Article Summary

Article focus

- Since 2002 patients have participated as collaborative partners in the biannual conference on Outcome Measures in Rheumatology (OMERACT).
- Although the contribution of patients has been praised and there is a wide-spread call for scientific publications on the impact of engaging with patients, no systematically obtained evidence has been published to support the idea that structural involvement of patients in research conferences is beneficial.
- Our qualitative study reports the combined results of a thematic document analysis and 32 semi structured interviews with all stakeholders including researchers, patient participants and representatives from pharmaceutical industry and international regulators.

Key messages

- Long term engagement with arthritis patients in OMERACT conferences has significantly influenced outcome research in the field of rheumatology.
- Patients have successfully contributed to the research agenda of OMERACT by identifying new domains that are important for patients, and provided the patient perspective in the development of core outcome measurement sets and the development of patient reported outcome measures.

Strengths and limitations of this study

- Triangulation of the combined review of the document analysis and interviews, together with the active involvement of all team members representing a variety of perspectives in the phase of data analysis and interpretation, have enhanced the validity of the study.

- The unique context of the OMERACT conferences may limit the generalizability of the results so comparable evaluation studies in other conference formats would be worthwhile.

Incorporating the patient perspective in health care research is strongly promoted by policy makers,[1-4] funding bodies and international regulators. Many theoretical benefits from patient involvement in research have been reported, [5-8] such as improving the relevance of research questions, improving recruitment of study participants, and increasing chances for funding and dissemination of results. In addition there is an increasing recognition of the essential role of patients in outcome research.[9] The USA Food and Drug Administration (FDA) has made patient involvement mandatory in the process of the development of patient reported outcome measures[10 11] and in the context of COMET (Core Outcome Measures in Effectiveness Trials) patient contributions are seen as crucial in defining domains that are relevant to include in core outcome measurement sets for clinical trials.[12] Development of core outcome sets might lead to less variety of incomparable and inappropriate outcome measures, more patient oriented endpoints and less bias by selective reporting of only positive or statistically relevant outcomes.[13] Core outcome measurement sets may ease the work of systematic reviewers in synthesizing the results of multiple studies.[14 15] The question is however whether these theoretical benefits of patient involvement in outcome research make any difference in practice.

The international group Outcome Measures in Rheumatology (OMERACT), which defines core outcome measurement sets in rheumatic diseases, first included patient participants at its 6th bi-annual conference in 2002 and has continued to do so. This provides an opportunity to analyse the consequences and address the important question of whether patient participation has resulted in any demonstrable impact on the nature of its research activity.

Patient involvement in OMERACT has been presented as beneficial and the 2002 conference report concluded that “the preliminary success of this forum” was the basis for “continued and possibly expanded patient participation at the next OMERACT meeting”.[16]

Two conferences later others perceived the involvement of patients as “indicative of the beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.[17] Since then OMERACT has formulated three principles recognising the essential role of patients in outcome research.[18] First, patients’ input is indispensable when defining relevant outcome measures, identifying domains that are important from the perspective of patients, and assessing feasibility of measurement tools. Second, structural involvement of patients during the whole research process provides face validity. Third, OMERACT intends “to ground theoretical discussions in the lived experience of arthritis, and in concepts which can be readily communicated to patients to help with therapeutic decision making”.[18]

However, the validity of these arguments has never been substantiated by robust evidence for the effectiveness of patient participation in OMERACT and it is not clear whether or how this involvement has influenced methodologies, procedures, attitudes, and research outcomes. Therefore, the objective of this study is to describe and evaluate the contributions made by patients since OMERACT started implementing structural patient participation in its conferences. We review the impact of patients on the research agenda and the development of patient reported outcomes (PRO’s) and explore how including patients has influenced the culture and structure of the OMERACT conference

Method

Patient participation in research is a new phenomenon and often not reported or reflected on in scientific publications. This lack of written sources in the scientific literature complicates the study of the process and impact of patient participation through a review of relevant literature. A provisory search using Pubmed (March 2010) for the terms “patient participation” OR “patient involvement” OR “user involvement” OR “consumer involvement” AND “OMERACT” did not generate any relevant reference. Therefore we conducted a content analysis of relevant documents (any written material on the topic of patient participation).

Documents are a stable, rich source of contextual information, providing well-grounded data on events or situations at low costs. A sound document analysis is rule-bound, systematic, following a coding process where raw data are aggregated into units describing the content.[19] We included OMERACT conference proceedings as published by The Journal of Rheumatology (1992-2010) and 'grey literature' such as correspondence, invitations, session reports, e-mails and OMERACT policy documents. The review focused on the arguments, reception and evolution of patient involvement in OMERACT conferences and the contributions made by patients.

Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010) using qualitative interviews with representatives of stakeholders. Responsive evaluation is grounded in the hermeneutic research tradition and is used by social scientists to interpret meanings that participants attribute to a phenomenon, here the history and impact of a decade of patient participation from the perspective of the conference delegates. It samples all stakeholders and does not seek consensus, but respects the plurality of opinions, values and interests. This ensures that no perspective is omitted as the result of an imbalance of power.[20]

The first author (MW) has been involved in OMERACT since 2002 as a patient participant. He has a rheumatic condition and has been educated as a responsive researcher. Characteristics for a responsive researcher are a multiple partiality and the intent to enhance mutual understanding among all stakeholders. The last author (JK) has been involved in OMERACT since the first conference (1992) and has been the leader of the patient perspective workshop between 2002-2012. Having witnessed the involvement of patients firsthand from the very beginning MW and JK provided useful information to start the research, yet it also alerted them to critically reflect how this engagement influenced the research, and how to prevent bias. Therefore two independent experts (TA, MK) were added to the team. They had no relations with the OMERACT conference and its participants, and TA acted as peer-debriefer discussing dilemmas and challenging methodological decisions with MW.[21]

The first author held 32 semi-structured interviews before, during and after the 10th conference (Table 1) and included senior (n=10) and junior researchers (n=2), representatives of pharmacological industry and regulators (n=2), conference staff (n=2), new patient participants (n=8) and experienced patient participants (n=8). The interviewees were invited and informed by e-mail. The patient participants were aware of the purpose of the study through a one page announcement in the pre-conference patient pack and were asked for informed consent. In the Netherlands no ethical approval is required for non-intrusive interviews only.

Twenty-eight interviews were recorded, transcribed by an independent secretariat and subjected to a responder check. Three interviews were summarized in a report, one interview took place without protocol and, on request of the interviewee, without recording (PF). One interview was done through Skype (PP). The average duration of the interviews was over 50 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-four interviews were held in English of which six was not the native language. Eight interviews were in Dutch.

The interview protocols were slightly different for professionals, new patients and experienced patients. The topics were not only derived from the document analysis but also from four pilot interviews and the personal knowledge of MW and JK and the expertise of TA. The topics dealt with: the expected role of patient participants, their selection, preparation and support, and the expected or provided contribution to the OMERACT conference. ‘Fatigue’ was added as a potential probe because publications had already shown that this topic deserved special attention with regard to our research questions.[22-24] Participants with long term experience in OMERACT were asked retrospectively to describe their memories of the discussions and decisions taken about patient participation before and after 2002. Their recollections might be characterized as 'oral history'.

Selection of interviewees - At OMERACT 10 a total of 172 delegates participated, 152 professionals and 20 patients. Nine patients attended the conference for the first time. All interviewees, except for one patient participant from the hosting country, were selected by MW and JK following an emergent purposive sampling approach.[21] They used a list of attendees provided by the congress agency, covering four out of five criteria found to be important (stakeholder background, gender, geographical spread and number of OMERACT conferences attended). The criterion 'opinion about patient involvement' was assessed on the basis of authors' insight of the participant as being 'positive' (e.g. contributing to the patient perspective workshop or involving partners in own activities), 'indifferent' or 'skeptical' (e.g. resistant, not collaborating with partners). When it became clear during the process of data collection that certain criteria were not well covered new participants were approached till maximum variation was realized. For example, two interviewees who were chosen because of their previously reported criticism of involving patients, showed a considerable change in perception of patient involvement in a positive way. For this reason two more interviews with professionals who had expressed critical comments during the last conference were arranged. Finally, to ensure the opinions of young investigators, two OMERACT Fellows were approached, one undertaking a PhD in translational research and one post doctoral researcher active in clinical research.

Saturation was defined as a repetition of data; theoretical saturation as achieving sufficiently robust empirical data to support and describe the identified themes and main categories. Saturation was discussed and agreed within the research team. In total the perceptions and experiences of 16 patient participants and 16 professionals were collected (Table 1).

[insert Table 1]

Data-analysis - A thematic content analysis focused in particular on the reported contributions attributed to patient participants. Coding of the interviews was done separately by MW and an independent second coder (MK) who had never worked with active patient involvement before. This resulted in 211 detailed codes that were then combined into 27 sub-categories. During several meetings, the project team, representing various backgrounds, discussed the codes and subcategories from a variety of perspectives, and sought natural groupings or categories within the data. Triangulation was used in two different meanings: First, as a means of verifying findings against another source (interview) or another method (document analysis) and to enhance the validity of the data. Second, as a means to enrich the data collection and improve the face-validity by synthesizing findings from the document analysis with the personal memories and experiences of respondents who looked back in time. By doing so, gaps in the document analysis could be filled in.

The relevance and validity of the analysis and interpretation of the data was increased by the involvement of an external expert in qualitative health research (TA) as well as by inviting one of the patients (SC) who attended OMERACT 10 for the first time to join the research team. As a patient research partner[25] she was involved in the coding, data analysis and data interpretation to guarantee the patient perspective. To protect the anonymity of the participants all quotes are presented in the "she"-form. Quotes of professional researchers are indicated by 'R' and those of patient research partners (in short: 'partners') by 'P'.

Results

History of patient involvement at OMERACT

OMERACT started in 1992 as an initiative to overcome the problem of widespread and inconsistent use of many different outcome measures in rheumatoid arthritis (RA) clinical trials. The objective was to improve “the accuracy and responsiveness to change of clinically relevant (to patient and clinician) endpoints”.[26] Rheumatologist from many countries met in Maastricht and achieved consensus on a core set of outcomes for RA. The RA core set was endorsed by the WHO.[27] The initial stand alone conference was sufficiently successful that it was followed by conferences in alternate years continuing the discussion and consensus building about new core sets for other rheumatic diseases and new measurement instruments.

During the fifth OMERACT conference (2000) participants discussed the concept of a minimum clinically important difference (MCID). Based on methodological arguments a growing interest in patient reported outcomes emerged, culminating in a spontaneous proposal at the final session to invite patients to the next conference. All participants voted in favour of this proposal.[28] The chair of the conference felt confident about the proposal because it had been discussed in the organising committee before, although no decisions had been taken. Participants of the MCID module argued that patient perspectives should be explored further [29] and took responsibility for identifying 11 patients to join OMERACT 6 and to review the RA core set.

Our document analysis revealed the unconditional positive reception of patient delegates at OMERACT conferences, and partners confirmed that concerns regarding their involvement were misplaced. They felt their reception was extremely welcoming. “There was a tangible feeling of relief and a belief that patients' views and opinions would be listened to and incorporated into the deliberations".[30] Also the organizers were excited and called the patient involvement "a tremendous success".[31]

Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have participated as full delegates with equal voting rights.[32] Their role and contributions have developed over time. At the first conference (2002) they formed an homogeneous group of people with RA with little or no experience in scientific research. The level of involvement in the conference in general was relatively low, support was not organized and the number of sessions patients attended was limited. Contributions centred on participation in the workshop discussions about the severity of fatigue and the definition of low disease activity, although there was a keynote speech at the opening ceremony.[30] In contrast, by OMERACT 11 (2012) the partners were a heterogeneous group with different rheumatic conditions and different levels of experience, competences and cultural background. They received a pre-conference information pack and were actively supported by a pre-conference dinner, a glossary, training sessions and a buddy system. They carried out a variety of tasks similar to professionals such as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout sessions and preparing consensus statements. Several partners became co-authors of peer-reviewed publications.

Patient contributions to OMERACT meetings and outcome research

Interviewees reported a variety of contributions made by partners during the conference where they are an integral part of the deliberative and consensus-building process.[18] These examples are presented below and compared with the document analyses when appropriate. Because research in the domain of fatigue has been reported as the most illustrative example, the contributions in this area will be described in more detail. Using the methodology described above we identified 5 main categories from the comments made during the interviews with OMERACT participants (Table 2): Contributions to the research agenda; The development of core sets; The development of patient reported outcomes (PRO's); The culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of the challenges that emerged from the interviews.

Contributions to the research agenda **-** From the very beginning partners had a significant influence on the research agenda in the field of rheumatology by participating in OMERACT workshops and small group discussions. They identified new outcome domains that are relevant from their perspective.[33] The first Patient Perspective Workshop, attended by 11 patient participants and 41 professionals, focused on the development of "valid outcome instruments that incorporate the perspective of the patient and to prepare the evidence and arguments for their inclusion in the (RA) core set" [24]. The preconference paper pointed out the methodological and political challenges: How to elicit and incorporate preferences of patients in RCT's? [32] The workshop had been specifically arranged to support the partner contributions including a pre- and post-workshop meeting. The workshop identified subjective experiences of RA, not encompassed in the RA core set but important consequences of the disease: a sense of well being, fatigue, and disturbed sleep.[24]

After the first conference attended by partners, it became apparent that perspectives of professionals and patients differ and more research was needed to articulate patients priorities.[34-37] Partners emphasized the need for an holistic approach to people with arthritis.[30] The acknowledgement of the discordance of perspectives initiated new studies looking into the preferences, opinions and experiences of people with rheumatic diseases [22 38 39] and developing patient-derived core sets.[40] This made participants more aware of the emerging patient perspective: "the whole realm of things we haven't looked at" [RA]. New topics emerged: remission, pain, flares and foot problems. One interviewee clearly stated that partners “inspired me for new projects to study the variety in new productivity outcome measures” [RK].

Case-study of fatigue - Since 2002 when partners identified new topics for research, studies have been initiated with firm involvement of partners in the field of sleep disturbances, flares and well being. The most progress has been made in fatigue, and the emergence of fatigue as a relevant outcome measure in RA provides an illustrative case history. When asked for the greatest benefit of including partners in OMERACT conferences interviewees unanimously confirmed that the topic of fatigue would not have been on the research agenda without partners expressing their concerns about fatigue as an often neglected symptom of their disease and without the listening of receptive professionals. One of the partners attending OMERACT 6 recalled:

> “I can’t remember who brought up the subject, but someone mentioned fatigue. And that was the occasion when one of the other delegates said ‘well, everybody gets tired’. One patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]

Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and quantitative research. The first studies investigated the prevalence and severity of fatigue in RA and how patients describe their fatigue.[41-44]. The next step comprised a systematic review of measurement instruments for fatigue [45] that explored the rigor of existing measurement tools and the need to develop patient-derived instruments that are trustworthy, capturing concepts and language of patients. Furthermore a standardized visual analogue scale, opportunities for electronic gathering of data and exploring mechanisms of fatigue that could guide researchers in the development of effective interventions, were added to the research agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a consequence of RA, but an independent variable that adds new information to the existing RA core set.[46 47] This new perception resulted in the acceptance of fatigue as an important outcome for clinical trials.[48 49] Fatigue was subsequently added to the RA core set as a recommended outcome.[50]

More powerful instruments for measuring fatigue in RA have since been devised and validated, starting from the perspective of the patients.[51 52] Outside OMERACT researchers initiated similar studies, focusing on the communication between patients and health professionals in the consultation room.[53]

The thematic document analysis provided additional evidence for the statement that without patients raising their voice at OMERACT 6 fatigue would not have been high on the research agenda. The issue of fatigue was not new for rheumatologists.[54-56] Fatigue was a symptom regularly reported during clinical consultations, but not incorporated in guidelines for monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and incorporated in a disease status questionnaire.[57] And during OMERACT 3 (1996) delegates carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal diseases. The discussion groups identified outcomes such as pain, depression, anxiety and fatigue as major concerns.[58] For fatigue eight examples of measurement instruments were given.[59]. However, after this workshop, nothing happened for six years, until patients raised the urgency of fatigue as a serious symptom.

Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only hearing from partners at OMERACT made them change their perception of fatigue as an important outcome:

> “Because when I was working in oncology before, during university training, of course we saw that the patients were lying in bed all day and we knew they were exhausted, call that fatigue. But patients with RA, we were ignorant.” [RC]

Another physician, involved in OMERACT from the start:

> “We were first discussing on fatigue and to be honest: I never ever had before heard of fatigue being a problem in rheumatology. So it got into my mind and then I got thinking about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a 100% positive response. So it was like a coming out, you know. I listened to the patients before but bringing it to a specific topic, that was really what I learned at OMERACT.” [RA]

[Insert Table 2]

The development of core outcome measurement sets and patient reported outcomes – During the first two conferences including patients the focus of partners’ contributions was on agenda-setting and identifying relevant outcomes for clinical trials. Then, partners became gradually involved on different levels in other OMERACT activities, varying from being consulted (e.g. in a Delphi process) to full collaboration (as partner and as co-author). They contributed by identifying domains that are relevant for disease-specific core sets for psoriatic arthritis,[60] fibromyalgia,[61 62] gout[63] and vasculitis.[64] Furthermore they contributed to the development of core outcome measurement sets for methodological or clinical concepts such as MCID and remission.[65] Partners have also played a role in the assessment of the feasibility of instruments and core sets, one of the three key components of the OMERACT Filter.[66]

Partners have been helpful in the development of PRO measurement instruments in the field of work productivity, monitoring adverse events,[67] flares[68] and psychosocial interventions such as self-management programs[69]. At the 2010 conference, during the plenary session on flares in RA, one of the partners gave a personal testimony about the devastating impact of the unpredictable nature of RA. A professional in the audience was surprised and reported: “It demonstrates that the disease activity fluctuates more than we can see in our data: Our instruments are more flat, and by the limited frequency of measuring we filter fluctuations out”.[RI]

Regulators require strong evidence for the effectiveness of new medicines by demonstrating accurately that they reduce structural progression as well as patient important outcomes. By doing both of these, developing standards for high quality imaging techniques and exploring new PRO’s and translating them into valid and feasible measures, OMERACT has been extremely advantageous for the negotiations with regulators about the registration and relatively generous reimbursement of new biologic agents:

> “I think, to be really honest, the patient involvement process in OMERACT and the changes in outcome measurements and the use of them in the drug tests has made a real difference for so many patients.” [RB]

The culture of OMERACT– In spite of the initial unanimous vote to invite patients, some researchers were concerned about changing the layout of the conference:

> "My original expectation of a limited contribution was based on fear that patients were not able to transcend their personal experience and to generalize … new stakeholders often don't have knowledge about clinometric." [RE]

In retrospect researchers explained that they deferred to the proposal in order to reflect a core principle of OMERACT of not immediately rejecting new ideas: "To respect and listen rather than just react".[RA]

Looking back, the number of professional participants who were in favour of partners at the conference slowly increased: "I was impressed by the very good working flow".[RC] Participants confirmed that the presence of partners has changed their way of thinking and talking. "They made my blind spot visible" [RK] and another professional reported:

> "Now what we have found is, and I changed my view, [be]cause it wasn't only from OMERACT. As I got to know more and more patients, I realized, this sounds stupid because it's so obvious but it wasn't obvious to me, that a patient isn't their disease. A patient is a person who happens to have a disease. What a big difference. Because if you're a person that happens to have a disease, then for example you might have incredible skills in an area that might be very useful to move a clinical trial forward. So once I came to that realization then patient involvement becomes an absolutely obvious and integral part of moving forward." [RA]

Partners improved communication and brought dynamics to the dialogue because they are motivated and constructive, without a personal agenda. At a conference such as OMERACT, where the discussion about methodology may become extremely technical, partners reminded participants of the common goal of the conference by providing a human face of a person living with the condition day by day. Their presence made participants more explicit about the objectives of sessions and more explanatory about terms and concepts under discussion. Together with a reduced use of jargon this 'forced' simplification resulted in fewer misunderstandings for everyone.

For some professionals the presence of partners complicated the communication. Some believed that partners slow down the process because they are not familiar with technical issues. Others felt disinclined to say what they wanted to out of respect to partners or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and put her own expertise aside to keep things simple: "Patients didn't sometimes understand the objective of the research, which hindered us". [RK] One of the partners admitted that "it is a thin line between providing input and causing irritation". [PN]

An analysis of the responses of patients attending OMERACT for the first time showed that new partners experienced a significant learning curve and a variety of personal benefits.[70] Results from this study suggest that in fact all participants learned from the contact with other stakeholders. During this process participants gained trust, respect and understanding, reflecting the emergence of relational empowerment:

> "Patients were a kind of sparring partner when I entered a relatively new area. That was fun and did clarify a lot". [RK]

Relational empowerment in the context of health research can be understood as a process in which traditional doctor-patient relationships transform into equal partnerships enabling mutual learning processes.[71] All participants become stronger by sharing knowledge and responsibilities, and educating and helping each other.

The reported benefits were easiest to identify at the beginning when the level of involvement was still low. They became more diffuse when partners were structurally involved as full and equal collaborators. One interviewee mentioned “a reality check” as an important benefit of partners attending the conference. For professionals it offered the opportunity to check the relevance of the scope of their research: Are we doing the right things according to patients and are we using the right tools and methods? It is a belief of professionals that this kind of feedback is important to legitimize their research and, together with the belief of partners that without this research no innovations will take place, it strengthened the mutual empowerment of both.

Consequences outside OMERACT – The lessons learnt at OMERACT were noticed by the outside world. Partners returning home after the conference have continued introducing patient participation in local and national research projects or established networks of patient research partners.[72-75] Some delegates published a working framework for incorporating the patient perspective in outcome research.[76] With the input from several OMERACT participants the European League Against Rheumatism (EULAR) developed recommendations for the inclusion of patient representatives in scientific projects.[25] Following these recommendations a new patient reported quality of life instrument for RA was developed and validated.[40 77] Based on the experiences of OMERACT the organizing committee of the 6th International Shared Decision Making conference decided in 2011 to invite patient participants.[78] In the same year OMERACT delegates, partners as well as professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials (COMET) conference, demonstrating how the OMERACT methodology can be utilized in other disease areas.[12]

Remaining challenges emerging from the interviews – The role and contribution of patient participants have changed over time and procedures for patient selection and support have been developed in order to identify patient participants who are able to make a difference. There is still a debate going on whether patients should be selected through strict criteria such as education, communication skills, attitude and familiarity with scientific research. Some argue that an expert meeting like OMERACT needs expert patients who have extended knowledge about methodologies of outcome research, and are able to provide a kind of aggregated patient input. At OMERACT this group represents a minority of delegates, who are reluctant to allocate the same rights and power to partners as to the professionals. The vast majority believes that many patients are able to contribute to an OMERACT conference and emphasizes that a heterogeneous group of partners in age, gender, condition, experience and cultural background are advantageous for the conference. They intend to develop full representative participation in all phases of research by including partners in working group activities between conferences. Finally, some participants point out the potential risks of partners who become too experienced. They appreciate the naive input as a patient, with a minimum of preparation and reflection. They assume that as soon as you start thinking about your contribution, you lose the unique, individual perspective and become a patient-expert who aligns too easily with professionals.

Professionals shared the opinion that partners need training, although they reported different ideas about the content and aims of such training. Experienced partners as well as novice researchers felt that any new participant has to learn the OMERACT objectives, culture and procedures first, before they can become fully productive, mostly at the second or third conference. This accords with the expectations of partners who attended OMERACT for the first time.

Overview of findings

These results show that a decade of patient involvement has been successful and had a significant impact on various aspects of outcome research. Perspectives of patients are different from those of health professionals. Broad consensus exists that partners at OMERACT have played a vital role in identifying domains relevant from the perspective of patients and in developing new PROs such as fatigue, sleep quality, flares and work productivity. Especially in the area of fatigue we have shown that patient involvement on different levels and in different phases improves the quality of outcome research. By combining evidence-based knowledge of researchers and the experiential knowledge of patients, a synthesis of both kinds of knowledge has been achieved and documented. The benefits are assessment tools that accurately measure what really matters to patients, are formulated in understandable language and are user-friendly. Other benefits go beyond improving clinical outcome research and include improved communication, mutual empowerment, changed attitudes and substantial consequences outside OMERACT.

Discussion

We set out to describe and evaluate the contribution of patients as partners in rheumatology outcome research, reviewing their impact on the research agenda and the culture and process of the OMERACT conference. The document analysis provided the recorded facts while the interviews allowed an exploration of intentions, attitudes and perceived benefits or harms of patient participation that complements the document analysis. Since validated methodologies for demonstrating impact of collaboration with patients in the context of research are lacking, a responsive interview methodology seemed to be a good approach.

Both strengths and limitations of this study relate to the personal experience of the first and last authors as participants in the developing process of patient partner participation. Having witnessed the OMERACT process, actors and concerns of both the patient community and the research community was advantageous during the development of interview protocols, recruitment and selection of respondents and data analysis. For instance, the knowledge of the opinions of other participants made it possible to achieve maximum variation. Also, the active involvement in the support and training of partners created an adequate awareness of the relevant items to include in the study. The drawbacks of this engagement are the risks of subjectivity, blind spots and over- or under-identification with particular stakeholders. These risks have been addressed by applying strict quality measures for scientific rigor in qualitative, evaluation research.

The composition of the research team purposely included two external experts in qualitative research and a patient research partner, who were actively involved in the coding of interview transcripts and distilling relevant categories for impact, reduced the risk of subjectivity. Bias was avoided by the check-coding procedure in the analysis of the transcripts as at least two researchers independently coded each transcript, after which the whole team discussed the codes until consensus was reached. Saturation was also part of the discussion in the whole team. The inclusion of various stakeholder perspectives prevented one-sidedness. No signals were identified to suggest that interviewees have simply given desirable answers, or just been friendly to the interviewer. Some interviewees have been rather critical, reporting several barriers for structural involvement of patients in research, but have always added constructive suggestions for improvement. Peer debriefing by an independent colleague (TA) further helped to prevent bias.

Other limitations relate to the difficulties of demonstrating the ‘impact’ of patient involvement.[9] In OMERACT there is a strong belief that patient participation works, a belief that is nourished by the world-wide transition towards more patient-oriented health care and health research.[79-82] The assumption however that long term involvement of patients as equal partners guarantees sustainable inclusion of the patient perspective in outcome research complicates a thorough evaluation and makes it difficult to distinguish between expected, perceived and actual contributions. Many participants, not only partners, but also young researchers and other new-comers, are not able to identify their own contribution and may not see how their input is reflected in the final outcomes. Partners reported almost unanimously not being able to confirm substantial contributions during their participation but they believed they did. More experienced participants, mostly professionals, were less reluctant in reporting illustrative examples of patient contributions.

In a dialogue and consensus-based conference such as OMERACT many (f)actors contribute to the final outcomes. A linear causal relation between patient involvement and impact is therefore hard to establish; the processes of involvement are rather influenced by and influencing many (f)actors in a mutually interactive way. We found that when the level of involvement of partners increased from consultation to collaboration, it became harder to solely attribute individual or group contributions to the final outcomes. Because neither partners nor professionals act as a representative of any group or constituency it remains difficult to determine the influence of particular groups or individuals. Participation proved to be a dynamic process, especially when tasks were equally performed by patients and researchers, and when the dialogue between both took not only place during the official sessions, but also in the corridors of the conference. It should be noted that striving toward equality is a normative ideal,[83] and fighting inequalities between patients and professionals is and remains an ongoing concern. “Equality” may be seen in two ways: as the formal position of patients at the conference (as full delegates they had the same voting rights as professional delegates, they received the same pre-conference materials and had access to all conference sessions like all other participants) and as equality of partnership or collaboration in terms of influence on the decision making process. Given the power inequalities between patients and professionals the latter is the greater challenge, but nevertheless some of the documents and interviews suggest it has been achieved to some extent. Although patients remain only indirectly represented in the executive committee (the highest decision making body), our data support the conclusion that a small number of experienced patients achieved an equal relationship with researchers in their area of interest. They obtained the competences that enabled them to perform all kinds of tasks at the conference similar to professionals, and provided input that justified co-authorship of peer-reviewed articles. We did not obtain in-depth information about the question to what extent power inequalities between patients and researchers still persist but we know from the feedback of all respondents, including some fellows and researchers that attended OMERACT for the first time, that some did not feel treated equally. To what extent this experience was caused by their status of being a patient or by the status of a new participant is still unknown.

A last obstacle for demonstrating the influence of patient participation is the invisibility of experiential knowledge, often hidden in anecdotal stories. It has an impact that is rarely claimed by patients nor perceived by professionals. Personal comments are normally not reported because they are not seen as a valuable and valid source of knowledge [84] and yet clear documentation of meetings is required to ensure that patients' contributions become visible.[85] Professionals focus on synthesizing data and may not notice that the dialogue with patients works like a reality check, generates new ideas or changes their beliefs, behaviour or perception. When partners appear to simply agree with the results presented at OMERACT it might look as if they do not have any contribution to make, but in fact they confirm the value of the work under discussion and provide face validity to the process. It is for this reason that most professionals appreciate the feedback and input from partners, although not all are aware of this reason. Realizing the importance of such a reality check is beneficial for the management of realistic expectations: do not expect innovative ideas, brilliant suggestions and new concepts when inviting partners to join research. Their contributions are often more subtle and need the attention of a modest and committed researcher to be noticed.

Despite these limitations, we believe that the results presented in this study are relevant and valid. It is undeniable that there is a growing belief that patient involvement has been successful and brought a unique added value to the conference. Even those who were originally among the most skeptical participants now report that they have changed their perception about the expected contribution of patient research partners. This study is conducted within the context of a scientific research conference in the field of rheumatology, a long term somatic condition. Our ability to generalize the findings is therefore limited and extrapolation to other research contexts or to other conditions should be done with care.

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Table 1 Characteristics interviewees

		Professionals	Patient Research Partners	Interview Code
Sex (M:F)		12 : 4	7 : 9	
Professional background or Diagnosis		10 practicing rheumatologists 3 full time researchers 3 other professionals	10 rheumatoid arthritis 2 vasculitis 2 ankylosing spondylitis 1 fibromyalgia 1 gout	
Number of OMERACT conferences attended	1	5	8	PA to PF, PO, PP
	2	0	3	PG to PN
	3	1	1	
	4	4	4	
	≥5	6	0	
Interview in relation to	Before	2	1	
	During	8	16	

OMERACT conference	After	6	5	
Geographical spread		6 countries 2 continents	7 countries 4 continents	
Research Background		10 Senior Researchers		RA to RG, RJ, RK, RY
		1 Research Fellow		RH
		1 Post-doctoral researcher		RI
		2 Pharma representatives		DA, DD
		2 Staff members		DB, DC

Table 2

Main and sub-categories from the analysis of patient contributions to OMERACT meetings and outcome research since 2002

IMPACT OF A DECADE OF PATIENT INVOLVEMENT IN OMERACT				
Research Agenda	Outcome core sets	Patient reported outcomes	Culture of OMERACT	Consequences outside OMERACT
Generating challenging ideas Identification of patient-relevant research topics: • Well being • Fatigue • Sleep disturbance • Flares	Identification of patient relevant domains to include in core sets for clinical trials: • Fibromyalgia • Psoriatic Arthritis • Vasculitis • Gout • MRI • MCID	Acceptable, understandable and feasible outcome measures for • Monitoring adverse events • Work productivity • Flares • Psychosocial interventions	• Attitudes • Communication • Perceptions • Motivation • Relational empowerment • Personal benefits	• Local initiatives • Local and national networks of partners • EULAR • COMET • ISDM

	• Remission			
Reality check				

Abbreviations

COMET	Core Outcome Measures in Effectiveness Trials
EULAR	European League Against Rheumatism
FDA	USA Food and Drug Administration
ISDM	International Shared Decision Making
MCID	Minimal Clinically Important Difference
MRI	Magnetic Resonance Imaging
OMERACT	Outcome Measurement in Rheumatology
PRO	Patient Reported Outcome
RA	Rheumatoid Arthritis