

What Has Been the Effect on Trial Outcome Assessments of a Decade of Patient Participation in OMERACT?

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ABSTRACT. *Objective.* Since 2002, 58 patients have participated as collaborating partners in 6 Outcome Measures in Rheumatology (OMERACT) conferences. Little is known about how they engage with researchers and how they have influenced conference outcomes.

Methods. A responsive evaluation was carried out, including a thematic document analysis of conference proceedings and gray literature, participant observation, and 38 interviews with patients and professionals representing research, industry, and regulators. Interview transcripts were subjected to an inductive content analysis.

Results. The role of patients has evolved from a single focus group in 2002 to full integration in all parts of the conference in 2012. Longterm engagement has made a significant change in the scope and conduct of rheumatology research. It has enriched the research agenda by identifying previously neglected outcome domains such as fatigue, sleep disturbances, and flares, and it has contributed to more patient-relevant outcomes in clinical trials. Facilitating factors have been a strong commitment of the leadership, adequate selection procedure, inclusive conference design, interactive and encouraging moderation style, and self-organized support. The intensity of the program and doubts regarding the representativeness of the patient group were still seen as challenges for the future.

Conclusion. Making patient participation an integral part of the vision and procedures of OMERACT has significantly contributed to the success of OMERACT. It has changed the perceptions and beliefs of many participants. Full use of patients' experiential knowledge before and during the conference is still challenging. (J Rheumatol First Release Oct 15 2013; doi:10.3899/jrheum.130816)

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OUTCOME RESEARCH

PATIENT PARTICIPATION

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USER INVOLVEMENT

There is a growing interest in actively engaging with patients in the context of scientific research^{1,2}. Involving patients in the development of patient-reported outcomes (PRO) ensures their relevance, acceptability, and quality^{2,3,4}. OMERACT (Outcome Measures in Rheumatology) has been a pioneer in the field of rheumatology in organizing longterm involvement of patients as colla-

borating partners^{5,6,7,8} in its biannual, worldwide conferences. Since 2002, 58 patients with different rheumatic conditions have participated in 6 conferences with the purpose of providing the patient perspective⁹. Their involvement has been reported as advantageous, a “success,”¹⁰ and “the beginning of a paradigm shift”¹¹. In 2011, OMERACT formalized its policy toward patient participation, recognizing the essential role of patients in outcome research¹². Patients' input is indispensable when identifying domains that are important from the perspective of patients and when assessing feasibility of measurement tools; moreover, it provides face validity to the OMERACT process. It also enables OMERACT 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”¹².

Despite a widespread belief among experienced OMERACT delegates that patient participation is worthwhile, robust evidence for its effectiveness is lacking. Little is known in general about how patients engage with researchers, the specific features that promote collaborative research, and the overall effect and consequences of patient

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involvement. OMERACT, because of its long history of patient participation, provides a unique opportunity to explore these issues by analyzing the effect of patient participation on the nature of its research activities. In addition, the inhibiting and facilitating conditions for this process can be elucidated.

This article contains the preliminary findings of a PhD project as presented by the first author during OMERACT 11 (2012). We describe the combined results of a thematic document analysis of conference proceedings and 38 interviews during OMERACT 10 (May 2010, Malaysia) based on 3 empirical substudies^{12a,12b,12c}.

OMERACT 10 took place over 6 days at a residential conference center and focused on 4 diseases — rheumatoid arthritis (RA), psoriatic arthritis (PsA), ankylosing spondylitis and gout — and on the methodology of choosing domains of interest and instruments appropriate to measuring these domains. As in previous meetings, the program consisted of a mixture of plenary sessions, intense small group discussions, and toward the end, formal presentations and proposals that were voted on to achieve international consensus¹³. Twenty of the 200 places were allocated to patients.

MATERIALS AND METHODS

To assess the process and effect of patient involvement in OMERACT, a responsive evaluation^{14,15} was carried out, starting with a thematic document analysis including scientific literature on patient participation, OMERACT conference proceedings, and gray literature such as correspondence, invitations, session reports, and policy documents. The review

focused on the reception and evolution of patient involvement in OMERACT conferences and the contributions made by patients.

Data collection was based on 38 semistructured interviews (Table 1) before, during, and after OMERACT 10 (Malaysia, 2010), and included researchers (n = 12), representatives of the pharmaceutical industry, regulators, and staff (n = 4), experienced patients (n = 8), and new patients (n = 8). To gain insights into changing perceptions, 3 new patients were interviewed 3 times: before the start of the conference; on the third day; and immediately after the end. Finally the opinions and experiences of 16 professionals and 16 patients were collected (Table 2).

Selection of interviewees aimed at maximum variation and followed an emergent purposive sampling approach taking into account background, opinion about patient involvement, sex, geographical spread, and number of OMERACT conferences attended. A preliminary analysis of the first data revealed an unexpected response shift. The attitude toward patient involvement of 2 professionals had changed from presumed skeptic to supportive. For this reason we identified 2 new interviewees who were known for their critical perceptions. After analyzing the additional transcripts, saturation was achieved.

Broad interview protocols were developed¹⁶ (Table 1) based on the document analysis, 4 pilot interviews, and the first author's personal experience during 3 OMERACT conferences. The protocols differed for professionals, new patients, and experienced patients. The interviews were recorded, transcribed, and subjected to a responder check. One interview took place without protocol and, on request of the interviewee, without recording. The average duration of the interviews was about 50 min. Finally, participant observation took place during 6 patient meetings by the first author.

Interview transcripts were subjected to an inductive content analysis¹⁷. Coding of all interviews was done separately by the first author and an independent health researcher with extensive experience in qualitative research (MK). To increase the relevance and validity of the data analysis, one of the patients who attended OMERACT 10 for the first time (SC) joined the research team. Codes were compared and discussed during several face-to-face meetings, resulting in sets of main categories. To

Table 1. Summary of the interview protocols for new patients, experienced patients, and researchers.

Researchers	Experienced Patient Delegates	New Patient Delegates*
How important is patient participation in OMERACT for you?	What do you remember about your first OMERACT conference?	Can you briefly describe how your rheumatic condition has influenced your personal life?
Can you describe how you see the role of a patient participant at OMERACT?	What makes OMERACT worthwhile to attend for a 2nd/3rd/4th time?	How were you invited for this conference?
For which research phases do you think patient participation is most appropriate?	Which topic did you find most relevant to contribute to and which the least?	How do you see your role at this conference?
What type of research do you think benefits most from patient participation?	How do you see your role at this conference?	Do you have any idea how patients could be of help in studying "remission"?
What do you see as the greatest achievement of patient participation in OMERACT?	How could patients be of any help in studying "remission" and "flares"?	Do you have any idea how patients could contribute to the research on "flares"?
What barriers have you experienced related to incorporating the patient perspective in OMERACT initiatives?	What do you see as the greatest achievement of patient participation in OMERACT?	Do you anticipate any barriers to your participation?
Do you have suggestions how OMERACT could support researchers to optimize patient participation?	What has been supportive for you to participate in OMERACT?	How could OMERACT support you to overcome barriers or to make you feel more confident?
	What barriers have you experienced to contributing fully?	Can you tell me, what are the most important things you hope to achieve by your participation at this OMERACT meeting?
	Do you have suggestions to optimize the input of patients in OMERACT?	

* For new patient delegates we composed 3 versions: 1 protocol for the interview before the conference, 1 for halfway, and 1 for after the conference.

Table 2. Characteristics of interviewees.

Characteristics	Professionals	Patient Research Partners
M:F	12:4	7:9
Professional background or diagnosis	10 practicing rheumatologists 3 fulltime researchers 3 other professionals	10 rheumatoid arthritis 2 vasculitis 2 ankylosing spondylitis 1 fibromyalgia 1 gout
No. of OMERACT conferences attended		
1	5	8
2	0	3
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		
Countries	6	7
Continents	2	4
Research background		
Senior researchers	10	—
Research fellow	1	
Postdoctoral researcher	1	
Pharma representatives	2	
Staff members	2	

reduce the risk of observer bias^{18,19}, all authors, representing different backgrounds, randomly cross-checked the coding and confirmed that there were no significant discordances. Triangulation took place by synthesizing data derived from document analysis, interviews, and participant observation.

RESULTS

Influence of Patient Participation

Widening the research agenda. The main findings are shown in Table 3. There was broad consensus about the significant influence of patients on the research agenda by identifying previously neglected outcome domains such as sleep disturbances, flares, and particularly fatigue^{20,21,22,23}. Increased research in the domain of fatigue has been reported unanimously as the most illustrative result of this process, which would not have occurred without the structural involvement of patients being listened to by receptive researchers. It has led to extensive studies into the nature, influence, and measurement of fatigue in RA^{24,25,26,27,28}. In 2006, fatigue was added to the RA core set as a recommended outcome for clinical trials^{29,30}. More powerful instruments for measuring fatigue have been devised and validated^{31,32} with the active involvement of patients³³. The issue of fatigue was not new for rheumatologists^{34,35,36}. During OMERACT 3 (1996), delegates carried out a ranking exercise where fatigue was already identified as a major concern for patients³⁷. Eight measurement instruments were given for fatigue³⁸. However, after this workshop, there were no other develop-

ments for 6 years. Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only hearing about it from patients at OMERACT 6 made them change their perception of fatigue as an important outcome.

Core sets and outcome measures. Patients have identified domains that are relevant for disease-specific core sets for PsA, fibromyalgia, gout, and vasculitis. Further, they contributed to the development of core sets for clinical concepts such as minimum clinically important difference and remission, and played an important role in the assessment of the feasibility of core sets and outcome measures, one of the key components of the OMERACT filter³⁹. Patients have also been helpful in the development of PRO in the field of work productivity, adverse events, flares, and psychosocial interventions.

Changing perspectives. Longterm engagement with patients has changed the culture of OMERACT and the perceptions of its participants. Initially some researchers were concerned about inviting patients to the conference. They feared that patients were not able to transcend their personal experience, to generalize, and to understand the rigor of the methodological discussions. After a while, when patients started to organize themselves and learned to apply OMERACT procedures, researchers became less reserved and started to see the benefits. They confirmed that it changed their way of thinking and talking. Patients improved communication and brought dynamics to the

Table 3. Main findings of the study.

Effect	Barriers and Facilitators
Longterm involvement of patients as collaborating partners has significantly influenced the OMERACT research agenda	The role of the leadership has been pivotal in implementing structural participation
The identification of fatigue as an underresearched domain and the increased research in this area would not have happened without the direct participation of patient research partners	Strict selection, training, and individualized support of patient research partners are important for successful inclusion of the patient perspective in outcome research
Patient participation in OMERACT has stimulated the inclusion of the patient perspective in the development of core domain sets, outcome measures, and other clinical concepts in rheumatology such as flares and remission	Like other new delegates, patient research partners have to learn the objectives, procedures, and culture of OMERACT before they can fully participate and contribute
Patient participation has been gradually embedded in the vision, culture, and procedures of the OMERACT conferences	The interactive and inclusive conference design, together with a facilitative moderation style, are advantageous for the participation of patients
Patient participants provided a reality check for researchers	The intensity of the program and the academic terminology are important barriers to patients participating
Patient participation in OMERACT has prompted new initiatives in other areas of health research to foster the inclusion of the patient perspective	Resistance to change and skepticism among researchers hinder full implementation of patient participation and increase chances of tokenism Doubts about the representativeness of the patient group still cause reluctance among professionals

dialogue because they were motivated, constructive, and had no personal agenda. When discussions became extremely technical, patients reminded participants of the common goal of the conference by providing the 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.

Patients attending OMERACT for the first time reported significant learning curves and a variety of personal benefits. In fact all participants learned from the contact with other parties. During this process participants gained trust, respect, and understanding. One interviewee mentioned “a reality check” as an important benefit. For professionals, patient participation offered the opportunity to check the relevance and scope of their research: are we doing the right things according to patients and are we using the right tools and methods?

Consequences outside OMERACT. Patient involvement in OMERACT has stimulated more patient-oriented health research in many countries. Patients returning home after the conference have introduced the concept of participatory research in local projects or established networks of patient research partners^{40,41,42,43}. With the input from several OMERACT participants, the European League Against Rheumatism developed recommendations for patient

involvement in scientific projects⁶. Inspired by the experiences of OMERACT, the organizing committee of the 6th International Shared Decision Making conference decided in 2011 to officially invite and facilitate for the first time 4 patient participants⁴⁴. In the same year, OMERACT delegates, patients as well as researchers, participated in the 2nd Core Outcome Measures in Effectiveness Trials (COMET) conference, demonstrating how the OMERACT methodology can be used in other disease areas⁴⁵.

Facilitators

Leadership. The patient role evolved from a single focus group in 2002 to full participation in all parts of the conference in 2012. The longterm commitment of the OMERACT organizing committee has been important for this process. The organizers were intrinsically motivated to foster patient participation in the conference because they believed that patient input is essential to decide on the right measurement tools⁴⁶. The decision to invite patients was not taken lightly, but once OMERACT leaders were determined, they wanted to do it properly. They provided full support to their designated patient participation leader and guaranteed funds for patients to attend the conference. The positive contributions made by patients were regularly acknowledged and stimulated OMERACT in 2006 to formulate basic principles for patient participation. This internal policy document represented a milestone by making patient participation part of the vision of OMERACT,

embedded in a set of operational procedures. In 2011 OMERACT decided on the principle that active involvement of patients would be a prerequisite for working groups suggesting themes in the OMERACT program¹².

Patient selection. Since 2004 OMERACT has aimed for 10% of conference participants to be patients with a mixture of conditions, geographical spread, and OMERACT experience. From the start it was clear that patients were expected not to represent any advocacy group or organization. Although they might be active in patient organizations, they should adopt a strict individual role at OMERACT. To avoid self-selection, recruitment is done through the clinics of participating physicians, who proved to be ideally positioned to identify eligible patients who are able to make a valuable contribution¹². A potential disadvantage of this is the possibility that patients feel unable to decline an invitation to please their consultant or to prevent jeopardizing their care.

Conference design. Constructive engagement with patients was enhanced by the small size of the conference, an open mind among delegates, and the spirit of OMERACT as a consensus-oriented initiative based on the involvement of many parties. From the start, patients have felt “part of the family”⁴⁷. The conference layout and the focus on active interaction and debate in small groups¹⁰ have been facilitators for accepting and incorporating new perspectives. Some of the patients, all of whom were officially invited as full delegates with voting rights and access to all sessions, felt equal to professionals and acted as co-researchers. A majority of the patients saw their role as giving information or as an adviser. In particular, new partners initially felt the same unequal relationship toward professionals as is experienced in clinical practice. The interviews revealed that new researchers and research fellows sometimes also experienced these disparities. However, participants who had attended several conferences confirmed that inequalities disappear over time.

Support. According to the organizers of OMERACT, the first conference with patient involvement was the easiest and most successful because there were no expectations. Partners formed a homogeneous group and the program was not demanding. Although support was limited, patients felt welcome and accepted⁴⁷. During subsequent conferences partners were extremely motivated to learn the OMERACT way and started to organize themselves. They formed a patient liaison group and developed introduction packs, information modules to bring newcomers up to speed, and a glossary in lay language⁴⁸. In 2010 new patients were matched with an experienced patient who took on the role of a buddy. To prevent overburdening, personalized programs were developed that helped patients to pace themselves and to prioritize sessions to attend. Finally, patients have insisted on being involved in the preconference work, saying they felt that early participation in working groups will best prepare

them for worthwhile participation during the conference.

Facilitative style. The breakout discussion sessions form the heart of OMERACT, and the quality of the moderation emerged as of utmost importance for patients to contribute to their full potential. From the perspective of patients, a style of moderation, both interactive and encouraging, that fosters mutual learning through open dialogue was seen as an important facilitator of confidence and greater input. Sometimes patients were confronted with an apparently patronizing attitude of a moderator by whom they felt ignored or disqualified. Language was an underestimated barrier in discussion groups for those whose first language is not English. Some patients tended to hold back when they were afraid of not using the right words or expressions.

Barriers

Overburdening. OMERACT was an overwhelming experience for all patients attending the conference for the first time. Before the conference they felt privileged to be invited but also insufficiently prepared and uncertain about what was expected from them. They anticipated a learning experience and hoped to be able to make a difference. In fact most experienced the conference program as physically and mentally challenging. They had to adjust to the use of medical jargon and scientific terminology, and they were not always familiar with the rules for clinical research. The (intercontinental) travel, the early and late starting times of sessions, and the duration of the conference increased the risk of overburdening. In particular, new patients often participated beyond their physical capacities. Dealing with hierarchical power relations and strongly opinionated professionals was experienced as mentally challenging. A recurring barrier reported by patients was a lack of feedback on provided contributions. At times they felt that their experiential knowledge was not accepted as a valid source for scientific research, nor seen as relevant compared to the expert knowledge of professionals. Experienced patients coped better with the requirements of the conference. After the conference almost all patients described their participation as having been a valuable learning experience. Although new patients presumed that they had not been very productive, they expected their contribution would be more effective at future conferences. Experienced patients as well as researchers confirmed that expectation.

Skepticism. In 2010 a few respondents said they believed that the disadvantages of patient participation outweighed the benefits. They judged the influence of partners out of balance and feared less interest in solving methodology challenges leading to a less diverse research agenda. They said they felt forced to engage with patients in areas of research where they saw little added value, such as imaging and biomarkers. Most of the patients confirmed this opinion and said that they thought they could offer little benefit to domains that were distant from their daily life. They said

they believed that they could contribute most to research focused on their own condition and were uncertain about providing meaningful input during sessions dedicated to other conditions. To a great extent, physicians agreed with this.

Lack of diversity. Institutionalization of patient participation gave rise to a debate regarding the representativeness of the patient group. OMERACT encouraged patients to provide the “naive” dimension of being familiar with the disease although there has been a tendency to raise the selection criteria to recruit better-educated patients. A small number of respondents objected to the participation of patients who did not understand the rigor of scientific research and slowed down the process, thus causing irritation during breakout sessions. They argued that the patient perspective should be obtained during the preconference research work. Other participants emphasized the irreplaceable value of new patients bringing in authentic experiences, crucial for generating new ideas. They reported potential risks of patients becoming too professional, identifying themselves with researchers, and finally losing touch with their peers. Some respondents warned that patients who started acting as advocates might compromise the outcomes of OMERACT as a data-driven conference.

DISCUSSION

Our study has proven that building patient participation structurally into the OMERACT conference program has guaranteed that themes that are important for patients were not only added to the research agenda, but increased a sense of urgency and prompted sustainable collaboration in new research programs. From the example of fatigue we learn

that it takes more than a decade to identify a new domain of interest for patients, develop validated outcome measures, and design intervention trials that target that new domain as a primary outcome. Patient involvement in OMERACT has enriched the research agenda and changed the perceptions of all those involved. Thus patient participation in a research conference is effective and complementary to other structured and deliberate procedures for agenda setting used in other disease areas⁴⁹. In research agenda setting, various models and strategies have been developed with the help of different methods⁵⁰; these may vary from more to less structured approaches, or from one-time consultation to regular advice from beginning to end. We see similarities between OMERACT and the dialogue model for agenda setting with patients, developed in The Netherlands: a stepwise approach for articulating patients’ priorities in homogeneous groups followed by a dialogue with other people involved^{51,52,53,54,55}. The dialogue model strives toward equal partnerships among patients and researchers in which control over the process is shared. In taking part in a dialogue, participants listen, try to understand each other, and find a common ground⁵⁶. Participants engage in a mutual learning process leading to a new perspective that is acceptable and recognizable for all involved⁵⁷. The strength of OMERACT has been the vision that patient participation is an integral part of the conference that requires extensive support and continuous reflection by all.

An important additional finding of our study is that patients and researchers said they feel patients’ knowledge and competences could be even better used. Based on the results of our study and using the 5 categories of the FIRST model (facilitate, identify, respect, support, and train⁵), we

Table 4. Recommendations for improving patient participation at future OMERACT conferences structured according to the FIRST model⁵.

Recommendations	
Facilitate	<ul style="list-style-type: none"> OMERACT should strive for structural involvement of patients in all phases of research, including pre- and post-conference activities Patients should receive personalized programs to prevent overburdening Moderators should create conditions for participation by adapting the objectives and layout of breakout sessions Patient delegates should be involved in the site visit of the accommodation to assess accessibility
Identify	<ul style="list-style-type: none"> Selection criteria for patient research partners attending OMERACT conferences should be made explicit and should take into account the competence of the English language and experience in working with academic researchers OMERACT should strive for a more representative patient group regarding ages, continents, and cultures Recruitment through the clinics of participating researchers should be continued OMERACT should differentiate between types of research regarding the desired level of involvement of patients
Respect	<ul style="list-style-type: none"> More attention is needed for information and acknowledgment after the conference Regular evaluation and reflection on the process of patient participation could improve the implementation and stimulate future research of its conditions and impact
Train	<ul style="list-style-type: none"> Patients need additional training on site to integrate and to achieve a minimum level of understanding OMERACT Training should focus on the expected role and tasks of patients, the topics on the conference program, and only a minimum on biostatistics

FIRST: Facilitate, identify, respect, support, and train.

have produced a list of recommendations that should be taken forward for consideration during future OMERACT conferences (Table 4). We summarize the remaining challenges for OMERACT as: (1) to study how experiential knowledge can be better incorporated in the discussions during breakout sessions. It is known from the literature that patients' views are often excluded unintentionally^{4,58}, and OMERACT should develop more effective inclusion strategies; (2) to facilitate patient participation in working groups that meet between conferences, to better prepare patients for their attendance at the conference; (3) to identify type and phases of research that are likely to benefit more from full involvement of patients; (4) to start reporting on the benefits and limitations of patient involvement; and (5) to guarantee a better representation of patients by inviting patients from continents that are currently absent.

Although we refer to examples outside OMERACT where conference organizers have taken notice of the way OMERACT has organized structural involvement of patients, the transferability of these recommendations may be limited to the specific context in which the data have been collected. They have been formulated by the authors and have not gone through a thorough consensus process. This should be taken forward during future OMERACT conferences. Further, based on the contingency of our findings, we cannot provide a simple template for patient participation in all conferences. Nevertheless, there may be lessons that other researchers can apply to the process of developing patient involvement.

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