



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.

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

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

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47 Patient involvement in OMERACT has been presented as a presumed success and the
48 2002 conference report concluded that “the preliminary success of this forum” was the basis
49 for “continued and possibly expanded patient participation at the next OMERACT
50 meeting”.¹⁶ Two conferences later others perceived the involvement of patients as “indicative
51 of the beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.¹⁷
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3 Since then OMERACT has formulated three principles recognising the essential role of
4 patients in outcome research.¹⁸ First, patients' input is indispensable when defining relevant
5 outcome measures, identifying domains that are important from the perspective of patients,
6 and assessing feasibility of measurement tools. Second, structural involvement of patients
7 during the whole research process provides face validity. Third, OMERACT intends "to
8 ground theoretical discussions in the lived experience of arthritis, and in concepts which can
9 be readily communicated to patients to help with therapeutic decision making".¹⁸
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19 However, the validity of these arguments has never been substantiated by robust evidence
20 for the effectiveness of patient participation and it is not clear whether or how this
21 involvement has influenced methodologies, procedures, attitudes, and research outcomes.
22 Therefore, the objective of this study is to describe and evaluate the contributions made by
23 patients since OMERACT started implementing structural patient participation in its
24 conferences. We review the impact of patients on the research agenda and the development of
25 patient reported outcomes (PRO's) and explore how including patients has influenced the
26 culture and structure of the OMERACT conference through the attitudes, beliefs and
27 perceptions of participants.
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40 **Method**

41 An initial thematic document analysis was carried out including OMERACT conference
42 proceedings and 'grey literature' such as correspondence, invitations, session reports, e-mails
43 and OMERACT policy documents. The review focused on the arguments, reception and
44 evolution of patient involvement in OMERACT conferences and the contributions made by
45 patients.
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53 Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010)
54 using qualitative interviews with representatives of stakeholders. This approach aims to
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3 explore different meanings that participants attribute to patient involvement.¹⁹ It samples all
4 stakeholders and does not seek consensus, but respects different opinions, values and
5 interests. This ensures that no perspective is omitted as the result of an imbalance of power.
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9 The first author held 32 semi-structured interviews before, during and after the
10 conference (table 1) and included senior (n=10) and junior researchers (n=2), representatives
11 of pharmacological industry and regulators (n=2), staff (n=2), new patient participants (n=8)
12 and experienced patient participants (n=8) . The interviewees were invited by e-mail. The
13 patient participants were aware of the purpose of the study through a one page announcement
14 in the patient pack that was distributed two weeks in advance of the conference.
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18 Twenty-eight interviews were recorded, transcribed by an independent secretariat and
19 subjected to a responder check. Three interviews were summarized in a report, one interview
20 took place without protocol and, on request of the interviewee, without recording (PF). One
21 interview was done through Skype (PP). The average duration of the interviews was 52
22 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-
23 four interviews were held in English of which 6 was not the native language. Eight interviews
24 were in Dutch.
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28 The interview protocols were slightly different for professionals, new patients and
29 experienced patients. The topics were derived from the thematic document analysis and pilot
30 interviews with 2 experienced patient participants and 2 researchers (a senior and a junior).
31 The topics dealt with: the expected role of patient participants; their selection, preparation and
32 support; and with the expected or provided contribution to the OMERACT conference.
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52 *Selection of interviewees* - At OMERACT 10 a total of 172 delegates participated, 152
53 professionals and 20 patients. Nine patients attended the conference for the first time.

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56 Selection of interviewees aimed at maximum variation and followed an emergent purposive
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3 sampling approach taking into account stakeholder background, opinion about patient
4 involvement, gender, geographical spread and number of OMERACT conferences attended.
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7 Except for one patient participant from the hosting country, all interviewees were selected by
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9
10 the first and last author. Because of the richness of the interviews, and in order to achieve
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12 saturation, extra interviews were held during the conference and seven other interviews took
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14 place within two weeks after the conference.

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16 Although 2 interviewees were chosen because of their previously reported criticism of
17
18 involving patients, the outcome of the interviews with these professionals was that they
19
20 expressed a change in perception of patient involvement in a more positive way. For this
21
22 reason 2 more interviews with professionals who had expressed critical comments during the
23
24 last conference were arranged. Finally, to ensure the opinions of young investigators, 2
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26 OMERACT Fellows were approached, one undertaking a PhD in translational research and
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28 one post doctoral researcher active in clinical research. In total the perceptions and
29
30 experiences of 16 patient participants and 16 professionals were collected (table 1).
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36 [insert table 1]
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40 *Data-analysis* - A thematic content analysis focused in particular on the reported
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42 contributions attributed to patient participants. Coding of the interviews was done separately
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44 by the first author and an independent second coder (third author) who had never worked with
45
46 active patient involvement before. This resulted in 211 detailed codes that were then
47
48 combined into 27 sub-categories. During several meetings, the project team, representing
49
50 various backgrounds, discussed the codes and subcategories from a variety of perspectives,
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52 and sought natural groupings or categories within the data. Triangulation took place by
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54 synthesizing interview data and results from the document analysis. To increase the relevance
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3 and validity of the analysis and interpretation of the data one of the patients who attended
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5 OMERACT 10 for the first time was invited to join the research team. As a patient research
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7 partner²⁰ she was involved in the coding, data analysis and data interpretation to guarantee
8
9 the patient perspective. To protect the anonymity of the participants all quotes are presented in
10
11 the “she”-form. Quotes of professional researchers are indicated by ‘R’ and those of patient
12
13 research partners (in short: ‘partners’) by ‘P’.

18 Results

22 Document analysis: History of patient involvement at OMERACT

25 OMERACT started in 1992 as an initiative to overcome the problem of widespread and
26
27 inconsistent use of many different outcome measures in rheumatoid arthritis (RA) clinical
28
29 trials. The objective was to improve “the accuracy and responsiveness to change of clinically
30
31 relevant (to patient and clinician) endpoints”.²¹ Rheumatologist from many countries met in
32
33 Maastricht and achieved consensus on a core set of outcomes for RA. The RA core set was
34
35 endorsed by the WHO.²² The initial stand alone conference was sufficiently successful that it
36
37 was followed by conferences in alternate years continuing the discussion and consensus
38
39 building about new core sets for other rheumatic diseases and new measurement instruments.
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43 During the fifth OMERACT conference (2000) participants discussed the concept of a
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45 minimum clinically important difference (MCID). Based on methodological arguments a
46
47 growing interest in patient reported outcomes emerged, culminating in a spontaneous proposal
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49 at the final session to invite patients to the next conference. All participants voted in favour of
50
51 this proposal.²³ The chair of the conference felt confident about the proposal because it had
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53 been discussed in the organising committee before, although no decisions had been taken.
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55 Participants of the MCID module argued that patient perspectives should be explored further
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3 ²⁴ and took responsibility for identifying 11 patients to join OMERACT 6 and to review the
4
5 RA core set.
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8 Our document analysis revealed the unconditional positive reception of patient delegates
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10 at OMERACT conferences, and partners confirmed that concerns regarding their involvement
11
12 were misplaced. They felt their reception was extremely welcoming. “There was a tangible
13
14 feeling of relief and a belief that patients’ views and opinions would be listened to and
15
16 incorporated into the deliberations”.²⁵ Also the organizers were excited and called the patient
17
18 involvement “a tremendous success”.²⁶
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21 Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have
22
23 participated as full delegates with equal voting rights.²⁷ Their role and contributions have
24
25 developed over time. At the first conference (2002) they formed an homogeneous group of
26
27 people with RA with little or no experience in scientific research. The level of involvement in
28
29 the conference in general was relatively low, support was not organized and the number of
30
31 sessions patients attended was limited. Contributions centred on participation in the workshop
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33 discussions about the severity of fatigue and the definition of low disease activity, although
34
35 there was a keynote speech at the opening ceremony.²⁵ In contrast, by OMERACT 11 (2012)
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37 the partners were a heterogeneous group with different rheumatic conditions and different
38
39 levels of experience, competences and cultural background. They received a pre-conference
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41 information pack and were actively supported by a pre-conference dinner, a glossary, training
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43 sessions and a buddy system. They carried out a variety of tasks similar to professionals such
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45 as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout
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47 sessions and preparing consensus statements. Several partners became co-authors of peer-
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49 reviewed publications.
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56 **Interviews: Patient contributions to OMERACT meetings and outcome research**

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3 Interviewees reported a variety of contributions made by partners during the conference
4 where they are an integral part of the deliberative and consensus-building process.¹⁸ These
5 examples are presented below and compared with the document analyses when appropriate.
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7 Because research in the domain of fatigue has been reported as the most illustrative example,
8 the contributions in this area will be described in more detail. Using the methodology
9 described above we identified 5 main categories from the comments made during the
10 interviews with OMERACT participants (Table 2): Contributions to the research agenda; The
11 development of core sets; The development of patient reported outcomes (PRO's); The
12 culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of
13 the challenges that emerged from the interviews.
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27 ***Contributions to the research agenda*** - From the very beginning partners had a significant
28 influence on the research agenda in the field of rheumatology by participating in OMERACT
29 workshops and small group discussions. They identified new outcome domains that are
30 relevant from their perspective.²⁸ The first Patient Perspective Workshop, attended by 11
31 patient participants and 41 professionals, focused on the development of “valid outcome
32 instruments that incorporate the perspective of the patient and to prepare the evidence and
33 arguments for their inclusion in the (RA) core set”²⁹. The preconference paper pointed out the
34 methodological and political challenges: How to elicit and incorporate preferences of patients
35 in RCT's?²⁷ The workshop had been specifically arranged to support the partner
36 contributions including a pre- and post-workshop meeting. The workshop identified
37 subjective experiences of RA, not encompassed in the RA core set but important
38 consequences of the disease: a sense of well being, fatigue, and disturbed sleep.²⁹
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54 After the first conference attended by partners, it became apparent that perspectives of
55 professionals and patients differ and more research was needed to articulate patients
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3 priorities.³⁰⁻³³ Partners emphasized the need for an holistic approach to people with arthritis.²⁵
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5 The acknowledgement of the discordance of perspectives initiated new studies looking into
6
7 the preferences, opinions and experiences of people with rheumatic diseases³⁴⁻³⁶ and
8
9 developing patient-derived core sets.^{37,38} This made participants more aware of the emerging
10
11 patient perspective: “the whole realm of things we haven’t looked at” [RA]. New topics
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13 emerged: remission, pain, flares and foot problems. One interviewee clearly stated that
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15 partners “inspired me for new projects to study the variety in new productivity outcome
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17 measures” [RK].
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22 ***Case-study of fatigue*** - Since 2002 when partners identified new topics for research, studies
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24 have been initiated with firm involvement of partners in the field of sleep disturbances, flares
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26 and well being. The most progress has been made in fatigue and the emergence of fatigue as a
27
28 relevant outcome measure in RA provides an illustrative case history. When asked for the
29
30 greatest benefit of including partners in OMERACT conferences interviewees unanimously
31
32 confirmed that the topic of fatigue would not have been on the research agenda without
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34 partners expressing their concerns about fatigue as an often neglected symptom of their
35
36 disease and without the listening of receptive professionals. One of the partners attending
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38 OMERACT 6 recalled:
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45 “I can’t remember who brought up the subject, but someone mentioned fatigue. And that
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47 was the occasion when one of the other delegates said ‘well, everybody gets tired’. One
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49 patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever
50
51 experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]
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3 Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and
4
5 quantitative research. The first studies investigated the prevalence and severity of fatigue in
6
7 RA and how patients describe their fatigue.³⁹⁻⁴² The next step comprised a systematic review
8
9 of measurement instruments for fatigue⁴³ that explored the rigor of existing measurement
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11 tools and the need to develop patient-derived instruments that are trustworthy, capturing
12
13 concepts and language of patients. Furthermore a standardized visual analogue scale,
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15 opportunities for electronic gathering of data and exploring mechanisms of fatigue that could
16
17 guide researchers in the development of effective interventions, were added to the research
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19 agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a
20
21 consequence of RA, but an independent variable that adds new information to the existing RA
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23 core set.^{44 45} This new perception resulted in the acceptance of fatigue as an important
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25 outcome for clinical trials.^{46 47} Fatigue was subsequently added to the RA core set as a
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27 recommended outcome.
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32 More powerful instruments for measuring fatigue in RA have since been devised and
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34 validated, starting from the perspective of the patients.^{48 49} Nicklin described precisely the role
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36 and contributions of patients in different phases of the development of patient reported
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38 outcome measures for fatigue, including the influence of a patient research partner providing
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40 practical insights into everyday life with RA at every stage of question development.⁵⁰
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42 Patients made significant contributions in pilot interviews by discussing “measurement
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44 properties of wording, time-frame and descriptors” and articulating different meanings of
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46 words like *cope* and *manage*. Their involvement improved the final data collection and has
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48 also been particularly valuable for developing new intervention programs. Outside
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50 OMERACT other researchers initiated similar studies, focusing on the communication
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52 between patients and health professionals in the consultation room.⁵¹ Based on these new
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54 insights intervention studies are now undertaken where fatigue is the primary endpoint^{52 53}.
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3 This case-study of fatigue shows that it took more than a decade to develop effective
4 interventions after identifying a new domain important from the perspective of patients.
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10 The thematic document analysis provided additional evidence for the statement that without
11 patients raising their voice at OMERACT 6 fatigue would not have been high on the research
12 agenda. The issue of fatigue was not new for rheumatologists.⁵⁴⁻⁵⁶ Fatigue was a symptom
13 regularly reported during clinical consultations, but not incorporated in guidelines for
14 monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and
15 incorporated in a disease status questionnaire.⁵⁷ And during OMERACT 3 (1996) delegates
16 carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal
17 diseases. The discussion groups identified outcomes such as pain, depression, anxiety and
18 fatigue as major concerns.⁵⁸ For fatigue eight examples of measurement instruments were
19 given.⁵⁹ However, after this workshop, nothing happened for six years, until patients raised
20 the urgency of fatigue as a serious symptom.
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36 Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only
37 hearing from partners at OMERACT made them change their perception of fatigue as an
38 important outcome:
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45 “Because when I was working in oncology before, during university training, of course
46 we saw that the patients were lying in bed all day and we knew they were exhausted, call
47 that fatigue. But patients with RA, we were ignorant.” [RC]
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54 Another physician, involved in OMERACT from the start:
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3 “We were first discussing on fatigue and to be honest: I never ever had before heard of
4 fatigue being a problem in rheumatology. So it got into my mind and then I got thinking
5 about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a
6 100% positive response. So it was like a coming out, you know. I listened to the patients
7 before but bringing it to a specific topic, that was really what I learned at OMERACT.”
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13 [RA]

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23 ***The development of core sets and patient reported outcomes*** – During the first two
24 conferences including patients the focus of partners’ contributions was on agenda-setting and
25 identifying relevant outcomes for clinical trials. Then, partners started contributing by
26 identifying domains that are relevant for disease-specific core sets for psoriatic arthritis,
27 fibromyalgia, gout and vasculitis. Furthermore they contributed to the development of core
28 sets for methodological or clinical concepts like MCID and remission. Partners have played
29 an important role in the assessment of the feasibility of instruments and core sets, one of the
30 three key components of the OMERACT Filter.⁶⁰
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40 Partners have been helpful in the development of PRO measurement instruments in the
41 field of work productivity, monitoring adverse events, flares and psychosocial interventions
42 such as self-management programs. At the 2010 conference, during the plenary session on
43 flares in RA, one of the partners gave a personal testimony about the devastating impact of the
44 unpredictable nature of RA. A professional in the audience was surprised and reported: “It
45 demonstrates that the disease activity fluctuates more than we can see in our data: Our
46 instruments are more flat, and by the limited frequency of measuring we filter fluctuations
47 out”. [RI]
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3 Although there is a broad consensus that partners have been beneficial in the development
4 of PRO's, several participants also reported the example of a single partner questioning the
5 tolerability of MRI scanning times for people with ankylosing spondylitis. During one of the
6 breakout sessions on imaging she reported not being able to lie still for 35 minutes in an MRI
7 machine. This alerted the researchers to their assumption that only moving hurts and as a
8 consequence the proposed scanning recommendations were adjusted to ensure shorter
9 scanning times. This shows that partners can be helpful in the assessment of the feasibility of
10 technical outcome measures.
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22 Regulators require strong evidence for the effectiveness of new medicines by demonstrating
23 accurately that they reduce structural progression as well as patient important outcomes. By
24 doing both of these, developing standards for high quality imaging techniques and exploring
25 new PRO's and translating them into valid and feasible measures, OMERACT has been
26 extremely advantageous for the negotiations with regulators about the registration and
27 relatively generous reimbursement of new biologic agents:
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38 "I think, to be really honest, the patient involvement process in OMERACT and the
39 changes in outcome measurements and the use of them in the drug tests has made a
40 real difference for so many patients." [RB]
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49 ***The culture of OMERACT***– In spite of the initial unanimous vote to invite patients, some
50 researchers were concerned about changing the layout of the conference:
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3 “My original expectation of a limited contribution was based on fear that patients were
4 not able to transcend their personal experience and to generalize ... new stakeholders
5 often don’t have knowledge about clinometric.” [RE]
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11 In retrospect researchers explained that they deferred to the proposal in order to reflect a core
12 principle of OMERACT of not immediately rejecting new ideas: “To respect and listen rather
13 than just react”. [RA]
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18 Looking back, professional participants were enthusiastic about the advent of partners
19 at the conference: “I was impressed by the very good working flow”. [RC] Participants
20 confirmed that the presence of partners has changed their way of thinking and talking. “They
21 made my blind spot visible” [RK] and another professional reported:
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29 “Now what we have found is, and I changed my view, [be]cause it wasn’t only from
30 OMERACT. As I got to know more and more patients, I realized, this sounds stupid
31 because it’s so obvious but it wasn’t obvious to me, that a patient isn’t their disease. A
32 patient is a person who happens to have a disease. What a big difference. Because if
33 you’re a person that happens to have a disease, then for example you might have
34 incredible skills in an area that might be very useful to move a clinical trial forward.
35 So once I came to that realization then patient involvement becomes an absolutely
36 obvious and integral part of moving forward.” [RA]
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49 Partners improved communication and brought dynamics to the dialogue because they are
50 motivated and constructive, without a personal agenda. At a conference such as OMERACT,
51 where the discussion about methodology may become extremely technical, partners reminded
52 participants of the common goal of the conference by providing a human face of a person
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3 living with the condition day by day. Their presence made participants more explicit about the
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5 objectives of sessions and more explanatory about terms and concepts under discussion.
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7 Together with a reduced use of jargon this 'forced' simplification resulted in fewer
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9 misunderstandings for everyone.
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11 For some professionals the presence of partners complicated the communication.
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13 Some believed that partners slow down the process because they are not familiar with
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15 technical issues. Others felt disinclined to say what they wanted to out of respect to partners
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17 or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and
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19 put her own expertise aside to keep things simple: "Patients didn't sometimes understand the
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21 objective of the research, which hindered us". [RK] One of the partners admitted that "it is a
22
23 thin line between providing input and causing irritation". [PN]
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29 An analysis of the responses of patients attending OMERACT for the first time showed that
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31 new partners experienced a significant learning curve and a variety of personal benefits.⁶¹
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33 Results from this study suggest that in fact all participants learned from the contact with other
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35 stakeholders. During this process participants gained trust, respect and understanding,
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37 reflecting the emergence of relational empowerment.
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42 "Patients were a kind of sparring partner when I entered a relatively new area. That
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44 was fun and did clarify a lot". [RK]
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49 Relational empowerment in the context of health research can be understood as a process in
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51 which traditional doctor-patient relationships transform into equal partnerships enabling
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53 mutual learning processes.⁶² All participants become stronger by sharing knowledge and
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55 responsibilities, and educating and helping each other.
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3 The reported benefits were easiest to identify at the beginning when the level of
4 involvement was still low. They became more diffuse when partners were structurally
5 involved as full and equal collaborators. One interviewee mentioned “a reality check” as an
6 important benefit of partners attending the conference. For professionals it offered the
7 opportunity to check the relevance of the scope of their research: Are we doing the right
8 things according to patients and are we using the right tools and methods? It is a belief of
9 professionals that this kind of feedback is important to legitimize their research and, together
10 with the belief of partners that without this research no innovations will take place, it
11 strengthened the mutual empowerment of both.
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25 ***Consequences outside OMERACT*** – The lessons learnt at OMERACT were noticed by the
26 outside world. Partners returning home after the conference have continued introducing
27 patient participation in local and national research projects or established networks of patient
28 research partners.⁶³⁻⁶⁶ Some delegates published a working framework for incorporating the
29 patient perspective in outcome research.⁶⁷ With the input from several OMERACT
30 participants the European League Against Rheumatism (EULAR) developed
31 recommendations for the inclusion of patient representatives in scientific projects.²⁰
32 Following these recommendations a new patient reported quality of life instrument for RA
33 was developed and validated.^{37 68} Based on the experiences of OMERACT the organizing
34 committee of the 6th International Shared Decision Making conference decided in 2011 to
35 invite patient participants.⁶⁹ In the same year OMERACT delegates, partners as well as
36 professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials
37 (COMET) conference, demonstrating how the OMERACT methodology can be utilized in
38 other disease areas.¹²
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3 ***Remaining challenges emerging from the interviews*** – The role and contribution of patient
4 participants have changed over time and procedures for patient selection and support have
5 been developed in order to identify patient participants who are able to make a difference.
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10 There is still a debate going on whether patients should be selected through strict criteria such
11 as education, communication skills, attitude and familiarity with scientific research. Some
12 argue that an expert meeting like OMERACT needs expert patients who have extended
13 knowledge about methodologies of outcome research, and are able to provide a kind of
14 aggregated patient input. At OMERACT this group represents a minority of delegates, who
15 are reluctant to allocate the same rights and power to partners as to the professionals. The vast
16 majority believes that many patients are able to contribute to an OMERACT conference and
17 emphasizes that a heterogeneous group of partners in age, gender, condition, experience and
18 cultural background are advantageous for the conference. They intend to develop full
19 representative participation in all phases of research by including partners in working group
20 activities between conferences. Finally, some participants point out the potential risks of
21 partners who become too experienced. They appreciate the naive input as a patient, with a
22 minimum of preparation and reflection. They assume that as soon as you start thinking about
23 your contribution, you lose the unique, individual perspective and become a patient-expert
24 who aligns too easily with professionals.
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43 Professionals shared the opinion that partners need training, although they reported
44 different ideas about the content and aims of such training. Experienced partners as well as
45 novice researchers felt that any new participant has to learn the OMERACT objectives,
46 culture and procedures first, before they can become fully productive, mostly at the second or
47 third conference. This accords with the expectations of partners who attended OMERACT for
48 the first time.⁶¹
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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

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3 transition towards patient-oriented health care and health research.⁷⁰⁻⁷² The assumption
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5 however that long term involvement of patients as equal partners guarantees sustainable
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7 inclusion of the patient perspective in outcome research complicates the evaluation process
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9 and makes it difficult to distinguish between expected, perceived and actual contributions.
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11 Many participants, not only partners, but also young researchers and other new-comers, are
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13 ignorant about their own contribution and may not see how their input is reflected in the final
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15 outcomes. Partners reported almost unanimously not being able to confirm substantial
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17 contributions during their participation but they believed they did. More experienced
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19 participants, mostly professionals, were less reluctant in reporting illustrative examples of
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21 patient contributions.
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25 Second, in a dialogue and consensus-based conference such as OMERACT many factors
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27 contribute to the final outcomes. We found that when the level of involvement increased from
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29 consultation to collaboration, it became harder to attribute individual or group contributions in
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31 the final outcomes. Collaboration is a dynamic phenomenon and although the breakout
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33 sessions are the main body of the OMERACT conference, a lot of work is done before, after
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35 and between the formal conference sessions. Since tasks are equally performed by patients as
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37 by any other participant, the dialogue between patients and professionals takes place not only
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39 during the official program, but in all parts of the conference. The corridors are quite
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41 important in this respect. Because neither partners nor professionals act as a representative of
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43 any particular group or constituency it remains difficult to determine the influence of
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45 particular individuals in that process.
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49 Finally, we assume that experiential knowledge, hidden in anecdotal stories, does have an
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51 impact that is often not claimed by patients nor perceived by professionals. Personal
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53 comments are normally not reported because they are not seen as a valuable and valid source
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55 of knowledge⁷³ and yet clear documentation of meetings is required to ensure that patients'
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3 contributions become visible.⁷⁴ Professionals focus on synthesizing data and may not notice
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5 that the dialogue with patients works like a reality check, generates new ideas or changes their
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7 beliefs, behaviour or perception. When partners appear to simply agree with the results
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9 presented at OMERACT it might look as if they do not have any contribution to make, but in
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11 fact they confirm the value of the work under discussion and provide face validity to the
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13 process. It is for this reason that most professionals appreciate the feedback and input from
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15 partners, although not all are aware of this reason. Realizing the importance of such a reality
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17 check is beneficial for the management of realistic expectations: do not expect innovative
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19 ideas, brilliant suggestions and new concepts when inviting partners to join research. Their
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21 contributions are often more subtle and need the attention of a modest and committed
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23 researcher to be noticed.
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27 Despite these limitations, we believe that the results presented in this study are relevant
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29 and valid. Through an approach of responsive evaluation reliable answers have been obtained.
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31 No signals were identified to suggest that interviewees have simply given desirable answers,
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33 or just been friendly to the interviewer. Some interviewees have been rather critical, reporting
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35 several barriers for structural involvement of patients in research, but have always added
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37 constructive suggestions for improvement. Lessons learnt regarding the conditions for
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39 successfully engagement with patients will be published separately.⁷⁵ However, it is
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41 undeniable that within OMERACT there is a growing belief that patient involvement has been
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43 successful and brought a unique added value to the conference. Even those who were
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45 originally among the most skeptical participants now report that they have changed their
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47 perception about the expected contribution of patient research partners.
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51 This study is conducted within the context of a scientific research conference in the field
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53 of rheumatology, a long term somatic condition. Our ability to generalize the findings is
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55 therefore limited and extrapolation to other research contexts or to other conditions should be
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3 done with care. In the COMET-initiative¹² health researchers try to develop a validated
4 methodology for defining core outcome sets for clinical trials in all disease areas and can
5 learn from the experiences of OMERACT.
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12
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16 17 **Competing Interests**

18
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21 received by the authors.
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24 25 **Contributorship**

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27 All authors have been involved in the process of data analysis and interpretation, and have
28 equally contributed to the writing of this manuscript.
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31 32 **Data Sharing**

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34 No additional data are available.
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60

References

1. Keizer B, Bless R. Pilot study on the position of health consumer and patients' organisations in seven EU countries. Den Haag: ZonMW, 2010:54.
2. Involve. People & participation; How to put citizens at the heart of decision-making. London: Beacon Press, 2005.
3. Staley K. Summary Exploring Impact: Public involvement in NHS, public health and social care research. Eastleigh.: INVOLVE, 2009.
4. Wright MT, Roche B, von Unger H, Block M, Gardner B. A call for an international collaboration on participatory research for health. *Health Promot Int* 2010;25(1):115-22.
5. Abma TA. Patients as partners in a health research agenda setting: the feasibility of a participatory methodology. *Eval Health Prof* 2006;29(4):424-39.
6. Abma TA, Nierse C, Widdershoven G. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qualitative Health Research* 2009;19(3):401-15.
7. Abma TA, Broerse JE. Patient participation as dialogue: setting research agendas. *Health Expect* 2010;13(2):160-73.
8. Caron-Flinterman JF, Broerse JE, Bunders JF. The experiential knowledge of patients: a new resource for biomedical research? *Soc Sci Med* 2005;60(11):2575-84.
9. Staniszewska S, Haywood KL, Brett J, Tutton L. Patient and public involvement in patient-reported outcome measures: evolution not revolution. *Patient* 2012;5(2):79-87.
10. Speight J, Barendse SM. FDA guidance on patient reported outcomes. *BMJ* 2010;340:c2921.
11. Rowen D, Carlton J, Brazier JE, Mulhern B, Palfreyman S, Stevens K, et al. FDA on PROMs. Two important points. *BMJ* 2010;341:c5454.
12. Williamson PR, Altman DG, Blazeby JM, Clarke M, Gargon E. The COMET (Core Outcome Measures in Effectiveness Trials) Initiative. *Trials* 2011;12 (Suppl 1)(A70).
13. Clarke M. Standardising outcomes for clinical trials and systematic reviews. *Trials* 2007;8:39.
14. Sinha IP, Smyth RL, Williamson PR. Using the Delphi technique to determine which outcomes to measure in clinical trials: Recommendations for the future based on a systematic review of existing studies. *PLoS Med* 2011;8(1).

- 1
2
3 15. Williamson P, Clarke M. The COMET (Core Outcome Measures in Effectiveness Trials)
4 Initiative: Its Role in Improving Cochrane Reviews. *Cochrane Database Syst Rev*
5 2012;5:ED000041.
6
7
- 8 16. Saag KG. OMERACT 6 brings new perspectives to rheumatology measurement research.
9 *J Rheumatol* 2003;30(4):639-41.
10
- 11 17. Heller JE, Shadick NA. Outcomes in rheumatoid arthritis: incorporating the patient
12 perspective. *Current Opinion in Rheumatology* 2007;19(2):101-05.
13
- 14 18. OMERACT. Guiding principles for patient research partner participation in OMERACT,
15 2011.
16
- 17 19. Abma TA, Widdershoven GAM. Evaluation as a relationally responsible practice. In:
18 Denzin N, Lincoln Y, editors. *Handbook for Qualitative Inquiry*. California: SAGE
19 Publications Ltd., 2011.
20
- 21 20. de Wit MPT, Berlo SE, Aanerud GJ, Aletaha D, Bijlsma JW, Croucher L, et al. European
22 League Against Rheumatism recommendations for the inclusion of patient
23 representatives in scientific projects. *Annals of the rheumatic diseases*
24 2011;70(5):722-6.
25
26
- 27 21. Tugwell P, Boers M. OMERACT conference on outcome measures in rheumatoid arthritis
28 clinical trials: introduction. *J Rheumatol* 1993;20(3):528-30.
29
- 30 22. Brooks PM, Boers M, Tugwell P. OMERACT III: the "ACT" revisited. *Jrn of Rheum*
31 1997;24(4):764-5.
32
- 33 23. Wells G, Anderson J, Beaton D, Bellamy N, Boers M, Bombardier C, et al. Minimal
34 clinically important difference module: summary, recommendations, and research
35 agenda. *J Rheumatol* 2001;28(2):452-4.
36
- 37 24. Kirwan JR, Carr A, Gluck O, Hewlett S. Proposal for inclusion of a 'Patient's
38 perceptions' module in OMERACT VI [unpublished], 2000.
39
- 40 25. Quest E, Aanerud GJ, Kaarud S, Collins S, Leong A, Smedeby B, et al. Patients'
41 perspective. *J Rheumatol* 2003;30(4):884-5.
42
43
- 44 26. Brooks PM, Tugwell P, Strand C, Simon L, Boers M. OMERACT 6: International
45 consensus conference on outcome measures in rheumatology: Introduction. *Jrn of*
46 *Rheum* 2003;30(4):866-7.
47
48
- 49 27. Kirwan J. The patient perspective workshop at OMERACT 6: Rationale and methodology
50 [unpublished preconference paper]. 2002.
51
52
53
54
55
56
57
58
59
60

- 1
2
3 28. Tugwell P, Boers M, Brooks P, Simon L, Strand V, Idzerda L. OMERACT: an
4 international initiative to improve outcome measurement in rheumatology. *Trials*
5 2007;8:38.
6
7
8 29. Kirwan J, Heiberg T, Hewlett S, Hughes R, Kvien T, Ahlmen M, et al. Outcomes from the
9 Patient Perspective Workshop at OMERACT 6. *Journal of Rheumatology*
10 2003;30(4):868-72.
11
12 30. Hewlett SA. Patients and clinicians have different perspectives on outcomes in arthritis. *J*
13 *Rheumatol* 2003;30(4):877-9.
14
15 31. Leeb BF, Sautner J, Leeb BA, Fassel C, Rintelen B. Lack of agreement between patients'
16 and physicians' perspectives of rheumatoid arthritis disease activity changes. *Scand J*
17 *Rheumatol* 2006;35(6):441-6.
18
19 32. Kvien TK, Heiberg T. Patient perspective in outcome assessments--perceptions or
20 something more? *J Rheumatol* 2003;30(4):873-6.
21
22 33. Carr A, Hewlett S, Hughes R, Mitchell H, Ryan S, Carr M, et al. Rheumatology
23 outcomes: the patient's perspective. *Journal of Rheumatology* 2003;30(4):880-3.
24
25 34. Ahlmen M, Nordenskiöld U, Archenholtz B, Thyberg I, Ronnqvist R, Linden L, et al.
26 Rheumatology outcomes: the patient's perspective. A multicentre focus group
27 interview study of Swedish rheumatoid arthritis patients. *Rheumatology (Oxford)*
28 2005;44(1):105-10.
29
30 35. Hewlett S, Carr M, Ryan S, Kirwan J, Richards P, Carr A, et al. Outcomes generated by
31 patients with rheumatoid arthritis: how important are they? *Musculoskeletal Care*
32 2005;3(3):131-42.
33
34 36. Rupp I, Boshuizen HC, Roorda LD, Dinant HJ, Jacobi CE, van den Bos G. Course of
35 patient-reported health outcomes in rheumatoid arthritis: comparison of longitudinal
36 and cross-sectional approaches. *J Rheumatol* 2006;33(2):228-33.
37
38 37. Gossec L, Paternotte S, Aanerud GJ, Balanescu A, Boumpas DT, Carmona L, et al.
39 Finalisation and validation of the rheumatoid arthritis impact of disease score, a
40 patient-derived composite measure of impact of rheumatoid arthritis: a EULAR
41 initiative. *Annals of the Rheumatic Diseases* 2011;70(6):935-42.
42
43 38. Hewlett S, Cockshott Z, Almeida C, Richards P, Lowe R, Greenwood R, et al. Sensitivity
44 to change of the Rheumatoid Arthritis Self-Efficacy scale (RASE) and predictors of
45 change in self-efficacy. *Musculoskeletal Care* 2008;6(1):49-67.
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

- 1
2
3 39. Hewlett S, Cockshott Z, Byron M, Kitchen K, Tipler S, Pope D, et al. Patients' perceptions
4 of fatigue in rheumatoid arthritis: overwhelming, uncontrollable, ignored. *Arthritis*
5 *Rheum* 2005;53(5):697-702.
6
7
8 40. Rupp I, Boshuizen HC, Jacobi CE, Dinant HJ, van den Bos GA. Impact of fatigue on
9 health-related quality of life in rheumatoid arthritis. *Arthritis Rheum* 2004;51(4):578-
10 85.
11
12 41. Repping-Wuts H, Uitterhoeve R, van Riel P, van Achterberg T. Fatigue as experienced by
13 patients with rheumatoid arthritis (RA): A qualitative study. *Intl J Nursing Studies*
14 2008;8(45 (7)):995-102.
15
16
17 42. Nikolaus S, Bode C, Taal E, van de Laar MA. Four different patterns of fatigue in
18 rheumatoid arthritis patients: results of a Q-sort study. *Rheumatology*
19 2010;49(11):2191-9.
20
21
22 43. Hewlett S, Hehir M, Kirwan JR. Measuring fatigue in rheumatoid arthritis: a systematic
23 review of scales in use. *Arthritis & Rheumatism* 2007;57(3):429-39.
24
25
26 44. Kirwan JR, Hewlett S. Patient perspective: reasons and methods for measuring fatigue in
27 rheumatoid arthritis. *J Rheumatol* 2007;34(5):1171-3.
28
29
30 45. Hewlett S, Chalder T, Choy E, Cramp F, Davis B, Dures E, et al. Fatigue in rheumatoid
31 arthritis: time for a conceptual model. *Rheumatology (Oxford)* 2010.
32
33 46. Kirwan JR, Minnock P, Adebajo A, Bresnihan B, Choy E, de Wit M, et al. Patient
34 perspective: fatigue as a recommended patient centered outcome measure in
35 rheumatoid arthritis. *J Rheumatol* 2007;34(5):1174-7.
36
37
38 47. Pincus T, Sokka T. Complexities in the quantitative assessment of patients with rheumatic
39 diseases in clinical trials and clinical care. *Clin Exp Rheumatol* 2005;23(5 Suppl
40 39):S1-9.
41
42
43 48. Nicklin J, Cramp F, Kirwan J, Greenwood R, Urban M, Hewlett S. Measuring fatigue in
44 rheumatoid arthritis: A cross-sectional study to evaluate the Bristol Rheumatoid
45 Arthritis Fatigue Multi-Dimensional questionnaire, visual analog scales, and
46 numerical rating scales. *Arthritis Care Res (Hoboken)* 2010;62(11):1559-68.
47
48
49 49. Nikolaus S, van de Laar MA. Measuring fatigue in rheumatoid arthritis. *Nat Rev*
50 *Rheumatol* 2011;7(10):562-4.
51
52
53 50. Nicklin J, Cramp F, Kirwan J, Urban M, Hewlett S. Collaboration with patients in the
54 design of patient-reported outcome measures: Capturing the experience of fatigue in
55 rheumatoid arthritis. *Arthritis Care & Research* 2010;62(11):1552-8.
56
57
58
59
60

- 1
2
3 51. Repping-Wuts H. Fatigue in patients with rheumatoid arthritis. Nijmegen University, 2009
4 (thesis).
5
6 52. Hewlett S, Ambler N, Almeida C, Cliss A, Hammond A, Kitchen K, et al. Self-
7 management of fatigue in rheumatoid arthritis: a randomised controlled trial of group
8 cognitive-behavioural therapy. *Annals of the rheumatic diseases* 2011;70(6):1060-7.
9
10 53. Dures E, Kitchen K, Almeida C, Ambler N, Cliss A, Hammond A, et al. "They didn't tell
11 us, they made us work it out ourselves": Patient perspectives of a cognitive-
12 behavioural programme for rheumatoid arthritis fatigue. *Arthritis care & research*
13 2011.
14
15 54. Wolfe F, Hawley DJ, Wilson K. The prevalence and meaning of fatigue in rheumatic
16 disease. *The Journal of rheumatology* 1996;23(8):1407-17.
17
18 55. Tack BB. Fatigue in rheumatoid arthritis. Conditions, strategies, and consequences.
19 *Arthritis Care & Research* 1990;3(2):65-70.
20
21 56. Crosby LJ. Factors which contribute to fatigue associated with rheumatoid arthritis. *J Adv*
22 *Nurs* 1991;16(8):974-81.
23
24 57. Garrett S, Jenkinson T, Kennedy LG, Whitelock H, Gaisford P, Calin A. A new approach
25 to defining disease status in ankylosing spondylitis: the Bath Ankylosing Spondylitis
26 Disease Activity Index. *The Journal of rheumatology* 1994;21(12):2286-91.
27
28 58. Newman SP. Psychosocial measures in musculoskeletal trials. *J Rheumatol*
29 1997;24(5):979-84.
30
31 59. Brooks P, McFarlane AC, Newman S, Rasker JJ. Psychosocial measures. *J Rheumatol*
32 1997;24(5):1008-11.
33
34 60. Boers M, Brooks P, Strand CV, Tugwell P. The OMERACT filter for Outcome Measures
35 in Rheumatology. *The Journal of rheumatology* 1998;25(2):198-9.
36
37 61. de Wit MPT, Koelewijn-van Loon MS, Collins S, Abma TA, Kirwan JR. How patients
38 contribute a new voice at a scientific conference: their expectations and experiences at
39 the Outcome Measures in Rheumatology Conference 2010. (submitted) 2012.
40
41 62. Abma TA, Nierse CJ, Widdershoven GA. Patients as partners in responsive research:
42 methodological notions for collaborations in mixed research teams. *Qual Health Res*
43 2009;19(3):401-15.
44
45 63. Kjekken I, Ziegler C, Skrolsvik J, Bagge J, Smedslund G, Tovik A, et al. How to develop
46 patient-centered research: some perspectives based on surveys among people with
47 rheumatic diseases in Scandinavia. *Physical Therapy* 2010;90(3):450-60.
48
49
50
51
52
53
54
55
56
57
58
59
60

- 1
2
3 64. Akerhielm CE. Patient participation in research - A Swedish model. *EULAR congress -*
4 *Session patient priorities in medical & social research [abstract PAR0011].*
5 *Copenhagen, 2009.*
6
7
8 65. Akerhielm CE. Patient research partners - A way to participation and influence in
9 research. *EULAR congress - Session patient information and education [abstract*
10 *PARE0001]. Rome, 2010.*
11
12 66. Hewlett S. Learnings from the Bristol experience - Practicalities of a decade of patient
13 involvement in research on a local level. *EULAR congress - Session patient*
14 *participation in research [abstract SP0095]. Rome: 2010.*
15
16 67. Hewlett S, De Wit M, Richards P, Quest E, Hughes R, Heiberg T, et al. Patients and
17 professionals as research partners: challenges, practicalities, and benefits. *Arthritis &*
18 *Rheumatism* 2006;55(4):676-80.
19
20 68. Gossec L, Dougados M, Rincheval N, Balanescu A, Boumpas DT, Canadello S, et al.
21 Elaboration of the preliminary Rheumatoid Arthritis Impact of Disease (RAID) score:
22 a EULAR initiative. *Annals of the Rheumatic Diseases* 2009;68(11):1680-5.
23
24 69. Stiggelbout AM, Weijden TV, Wit MP, Frosch D, Legare F, Montori VM, et al. Shared
25 decision making: really putting patients at the centre of healthcare. *BMJ*
26 2012;344:e256.
27
28 70. Beresford P. User involvement, research and health inequalities: developing new
29 directions. *Health Soc Care Community* 2007;15(4):306-12.
30
31 71. Ward PR, Thompson J, Barber R, Armitage CJ, Boote JD, Cooper CL, et al. Critical
32 perspectives on 'consumer involvement' in health research: Epistemological
33 dissonance and the know-do gap. *Journal of Sociology* 2009;46(1):63-82.
34
35 72. Lindenmeyer A, Hearnshaw H, Sturt J, Ormerod R, Aitchison G. Assessment of the
36 benefits of user involvement in health research from the Warwick Diabetes Care
37 Research User Group: a qualitative case study. *Health Expectations* 2007;10(3):268-
38 77.
39
40 73. Schipper K. Patient participation & knowledge [thesis]. VU University, 2011.
41
42 74. Jinks C, Ong BN, O'Neill TJ. The Keele community knee pain forum: action research to
43 engage with stakeholders about the prevention of knee pain and disability. *BMC*
44 *Musculoskelet Disord* 2009;10:85.
45
46 75. de Wit MPT, Abma TA, Koelewijn-van Loon M, Collins S, Kirwan J. Facilitating and
47 inhibiting factors for long term involvement of patients in outcome research - Lessons
48 learned from a decade of collaboration at OMERACT conferences. (*submitted*) 2012.
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For peer review only

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 2 3 4 ≥5	5 0 1 4 6	8 3 1 4 0 PA to PF, PO, PP PG to PN
Interview in relation to OMERACT conference	Before During After	2 8 6	1 16 5
Geographical spread	6 countries 2 continents	7 countries 4 continents	
Research Background	10 Senior Researchers 1 Research Fellow 1 Post-doctoral researcher 2 Pharma representatives 2 Staff members		RA to RG, RJ, RK, RY RH RI DA, DD 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: <ul style="list-style-type: none"> • Well being • Fatigue • Sleep disturbance • Flares 	Identification of patient relevant domains to include in core sets for clinical trials: <ul style="list-style-type: none"> • Fibromyalgia • Psoriatic Arthritis • Vasculitis • Gout • MRI • MCID • Remission 	Acceptable, understandable and feasible outcome measures for <ul style="list-style-type: none"> • Monitoring adverse events • Work productivity • Flares • Psychosocial interventions 	<ul style="list-style-type: none"> • Attitudes • Communication • Perceptions • Motivation • Relational empowerment • Personal benefits 	<ul style="list-style-type: none"> • Local initiatives • Local and national networks of partners • EULAR • COMET • ISDM
Reality check				

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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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.

For peer review only

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

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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

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3 Two conferences later others perceived the involvement of patients as “indicative of the
4 beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.¹⁷ Since
5 then OMERACT has formulated three principles recognising the essential role of patients in
6 outcome research.¹⁸ First, patients’ input is indispensable when defining relevant outcome
7 measures, identifying domains that are important from the perspective of patients, and
8 assessing feasibility of measurement tools. Second, structural involvement of patients during
9 the whole research process provides face validity. Third, OMERACT intends “to ground
10 theoretical discussions in the lived experience of arthritis, and in concepts which can be
11 readily communicated to patients to help with therapeutic decision making”.¹⁸
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23 However, the validity of these arguments has never been substantiated by robust evidence
24 for the effectiveness of patient participation in OMERACT and it is not clear whether or how
25 this involvement has influenced methodologies, procedures, attitudes, and research outcomes.
26 Therefore, the objective of this study is to describe and evaluate the contributions made by
27 patients since OMERACT started implementing structural patient participation in its
28 conferences. We review the impact of patients on the research agenda and the development of
29 patient reported outcomes (PRO’s) and explore how including patients has influenced the
30 culture and structure of the OMERACT conference
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43 **Method**

44 Patient participation in research is a new phenomenon and often not reported or reflected on
45 in scientific publications. This lack of written sources in the scientific literature complicates
46 the study of the process and impact of patient participation through a review of relevant
47 literature. A provisory search using Pubmed (March 2010) for the terms “patient
48 participation” OR “patient involvement” OR “user involvement” OR “consumer
49 involvement” AND “OMERACT” did not generate any relevant reference. Therefore we
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3 conducted a content analysis of relevant documents (any written material on the topic of
4
5 patient participation).
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7 Documents are a stable, rich source of contextual information, providing well-grounded
8
9 data on events or situations at low costs. A sound document analysis is rule-bound,
10
11 systematic, following a coding process where raw data are aggregated into units describing
12
13 the content.¹⁹ We included OMERACT conference proceedings as published by The Journal
14
15 of Rheumatology (1992-2010) and 'grey literature' such as correspondence, invitations,
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17 session reports, e-mails and OMERACT policy documents. The review focused on the
18
19 arguments, reception and evolution of patient involvement in OMERACT conferences and the
20
21 contributions made by patients.
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25 Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010)
26
27 using qualitative interviews with representatives of stakeholders. Responsive evaluation is
28
29 grounded in the hermeneutic research tradition and is used by social scientists to interpret
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31 meanings that participants attribute to a phenomenon, here the history and impact of a decade
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33 of patient participation from the perspective of the conference delegates. It samples all
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35 stakeholders and does not seek consensus, but respects the plurality of opinions, values and
36
37 interests. This ensures that no perspective is omitted as the result of an imbalance of power.²⁰
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41 The first author (MW) has been involved in OMERACT since 2002 as a patient
42
43 participant. He has a rheumatic condition and has been educated as a responsive researcher.
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45 Characteristics for a responsive researcher are a multiple partiality and the intent to enhance
46
47 mutual understanding among all stakeholders. The last author (JK) has been involved in
48
49 OMERACT since the first conference (1992) and has been the leader of the patient
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51 perspective workshop between 2002-2012. Having witnessed the involvement of patients
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53 firsthand from the very beginning MW and JK provided useful information to start the
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55 research, yet it also alerted them to critically reflect how this engagement influenced the
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3 research, and how to prevent bias. Therefore two independent experts (TA, MK) were added
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5 to the team. They had no relations with the OMERACT conference and its participants, and
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7 TA acted as peer-debriefer discussing dilemmas and challenging methodological decisions
8
9 with MW.²¹
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11 The first author held 32 semi-structured interviews before, during and after the 10th
12
13 conference (Table 1) and included senior (n=10) and junior researchers (n=2), representatives
14
15 of pharmacological industry and regulators (n=2), conference staff (n=2), new patient
16
17 participants (n=8) and experienced patient participants (n=8). The interviewees were invited
18
19 and informed by e-mail. The patient participants were aware of the purpose of the study
20
21 through a one page announcement in the pre-conference patient pack and were asked for
22
23 informed consent. In the Netherlands no ethical approval is required for non-intrusive
24
25 interviews only.
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29 Twenty-eight interviews were recorded, transcribed by an independent secretariat and
30
31 subjected to a responder check. Three interviews were summarized in a report, one interview
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33 took place without protocol and, on request of the interviewee, without recording (PF). One
34
35 interview was done through Skype (PP). The average duration of the interviews was over 50
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37 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-
38
39 four interviews were held in English of which six was not the native language. Eight
40
41 interviews were in Dutch.
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45 The interview protocols were slightly different for professionals, new patients and
46
47 experienced patients. The topics were not only derived from the document analysis but also
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49 from four pilot interviews and the personal knowledge of MW and JK and the expertise of
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51 TA. The topics dealt with: the expected role of patient participants, their selection, preparation
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53 and support, and the expected or provided contribution to the OMERACT conference.
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55 'Fatigue' was added as a potential probe because publications had already shown that this
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3 topic deserved special attention with regard to our research questions.²²⁻²⁴ Participants with
4
5 long term experience in OMERACT were asked retrospectively to describe their memories of
6
7 the discussions and decisions taken about patient participation before and after 2002. Their
8
9 recollections might be characterized as ‘oral history’.
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14 *Selection of interviewees* - At OMERACT 10 a total of 172 delegates participated, 152
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16 professionals and 20 patients. Nine patients attended the conference for the first time. All
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18 interviewees, except for one patient participant from the hosting country, were selected by
19
20 MW and JK following an emergent purposive sampling approach.²¹ They used a list of
21
22 attendees provided by the congress agency, covering four out of five criteria found to be
23
24 important (stakeholder background, gender, geographical spread and number of OMERACT
25
26 conferences attended). The criterion ‘opinion about patient involvement’ was assessed on the
27
28 basis of authors’ insight of the participant as being ‘positive’ (e.g. contributing to the patient
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30 perspective workshop or involving partners in own activities), ‘indifferent’ or ‘skeptical’ (e.g.
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32 resistant, not collaborating with partners). When it became clear during the process of data
33
34 collection that certain criteria were not well covered new participants were approached till
35
36 maximum variation was realized. For example, two interviewees who were chosen because
37
38 of their previously reported criticism of involving patients, showed a considerable change in
39
40 perception of patient involvement in a positive way. For this reason two more interviews with
41
42 professionals who had expressed critical comments during the last conference were arranged.
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44 Finally, to ensure the opinions of young investigators, two OMERACT Fellows were
45
46 approached, one undertaking a PhD in translational research and one post doctoral researcher
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48 active in clinical research.
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54 Saturation was defined as a repetition of data; theoretical saturation as achieving
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56 sufficiently robust empirical data to support and describe the identified themes and main
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3 categories. Saturation was discussed and agreed within the research team. In total the
4
5 perceptions and experiences of 16 patient participants and 16 professionals were collected
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7 (Table 1).
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11 [insert Table 1]
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16 *Data-analysis* - A thematic content analysis focused in particular on the reported
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18 contributions attributed to patient participants. Coding of the interviews was done separately
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20 by MW and an independent second coder (MK) who had never worked with active patient
21
22 involvement before. This resulted in 211 detailed codes that were then combined into 27 sub-
23
24 categories. During several meetings, the project team, representing various backgrounds,
25
26 discussed the codes and subcategories from a variety of perspectives, and sought natural
27
28 groupings or categories within the data. Triangulation was used in two different meanings:
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30 First, as a means of verifying findings against another source (interview) or another method
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32 (document analysis) and to enhance the validity of the data. Second, as a means to enrich the
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34 data collection and improve the face-validity by synthesizing findings from the document
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36 analysis with the personal memories and experiences of respondents who looked back in time.
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38 By doing so, gaps in the document analysis could be filled in.
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43 The relevance and validity of the analysis and interpretation of the data was increased by the
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45 involvement of an external expert in qualitative health research (TA) as well as by inviting
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47 one of the patients (SC) who attended OMERACT 10 for the first time to join the research
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49 team. As a patient research partner²⁵ she was involved in the coding, data analysis and data
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51 interpretation to guarantee the patient perspective. To protect the anonymity of the
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53 participants all quotes are presented in the “she”-form. Quotes of professional researchers are
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55 indicated by ‘R’ and those of patient research partners (in short: ‘partners’) by ‘P’.
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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 further²⁹ 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

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3 feeling of relief and a belief that patients' views and opinions would be listened to and
4 incorporated into the deliberations".³⁰ Also the organizers were excited and called the patient
5 involvement "a tremendous success".³¹
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10 Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have
11 participated as full delegates with equal voting rights.³² Their role and contributions have
12 developed over time. At the first conference (2002) they formed an homogeneous group of
13 people with RA with little or no experience in scientific research. The level of involvement in
14 the conference in general was relatively low, support was not organized and the number of
15 sessions patients attended was limited. Contributions centred on participation in the workshop
16 discussions about the severity of fatigue and the definition of low disease activity, although
17 there was a keynote speech at the opening ceremony.³⁰ In contrast, by OMERACT 11 (2012)
18 the partners were a heterogeneous group with different rheumatic conditions and different
19 levels of experience, competences and cultural background. They received a pre-conference
20 information pack and were actively supported by a pre-conference dinner, a glossary, training
21 sessions and a buddy system. They carried out a variety of tasks similar to professionals such
22 as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout
23 sessions and preparing consensus statements. Several partners became co-authors of peer-
24 reviewed publications.
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45 **Patient contributions to OMERACT meetings and outcome research**

46 Interviewees reported a variety of contributions made by partners during the conference
47 where they are an integral part of the deliberative and consensus-building process.¹⁸ These
48 examples are presented below and compared with the document analyses when appropriate.
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50 Because research in the domain of fatigue has been reported as the most illustrative example,
51 the contributions in this area will be described in more detail. Using the methodology
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3 described above we identified 5 main categories from the comments made during the
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5 interviews with OMERACT participants (Table 2): Contributions to the research agenda; The
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7 development of core sets; The development of patient reported outcomes (PRO's); The
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9 culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of
10
11 the challenges that emerged from the interviews.
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16 ***Contributions to the research agenda*** - From the very beginning partners had a significant
17
18 influence on the research agenda in the field of rheumatology by participating in OMERACT
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20 workshops and small group discussions. They identified new outcome domains that are
21
22 relevant from their perspective.³³ The first Patient Perspective Workshop, attended by 11
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24 patient participants and 41 professionals, focused on the development of “valid outcome
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26 instruments that incorporate the perspective of the patient and to prepare the evidence and
27
28 arguments for their inclusion in the (RA) core set”²⁴. The preconference paper pointed out the
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30 methodological and political challenges: How to elicit and incorporate preferences of patients
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32 in RCT's? ³² The workshop had been specifically arranged to support the partner
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34 contributions including a pre- and post-workshop meeting. The workshop identified
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36 subjective experiences of RA, not encompassed in the RA core set but important
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38 consequences of the disease: a sense of well being, fatigue, and disturbed sleep.²⁴
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43 After the first conference attended by partners, it became apparent that perspectives of
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45 professionals and patients differ and more research was needed to articulate patients
46
47 priorities.³⁴⁻³⁷ Partners emphasized the need for an holistic approach to people with arthritis.³⁰
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49 The acknowledgement of the discordance of perspectives initiated new studies looking into
50
51 the preferences, opinions and experiences of people with rheumatic diseases ^{22 38 39} and
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53 developing patient-derived core sets.⁴⁰ This made participants more aware of the emerging
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55 patient perspective: “the whole realm of things we haven't looked at” [RA]. New topics
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3 emerged: remission, pain, flares and foot problems. One interviewee clearly stated that
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5 partners “inspired me for new projects to study the variety in new productivity outcome
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7 measures” [RK].
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11 ***Case-study of fatigue*** - Since 2002 when partners identified new topics for research, studies
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13 have been initiated with firm involvement of partners in the field of sleep disturbances, flares
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15 and well being. The most progress has been made in fatigue, and the emergence of fatigue as
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17 a relevant outcome measure in RA provides an illustrative case history. When asked for the
18
19 greatest benefit of including partners in OMERACT conferences interviewees unanimously
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21 confirmed that the topic of fatigue would not have been on the research agenda without
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23 partners expressing their concerns about fatigue as an often neglected symptom of their
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25 disease and without the listening of receptive professionals. One of the partners attending
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27 OMERACT 6 recalled:
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34 “I can’t remember who brought up the subject, but someone mentioned fatigue. And that
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36 was the occasion when one of the other delegates said ‘well, everybody gets tired’. One
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38 patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever
39
40 experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]
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45 Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and
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47 quantitative research. The first studies investigated the prevalence and severity of fatigue in
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49 RA and how patients describe their fatigue.⁴¹⁻⁴⁴ The next step comprised a systematic review
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51 of measurement instruments for fatigue⁴⁵ that explored the rigor of existing measurement
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53 tools and the need to develop patient-derived instruments that are trustworthy, capturing
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55 concepts and language of patients. Furthermore a standardized visual analogue scale,
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3 opportunities for electronic gathering of data and exploring mechanisms of fatigue that could
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5 guide researchers in the development of effective interventions, were added to the research
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7 agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a
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9 consequence of RA, but an independent variable that adds new information to the existing RA
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11 core set.^{46 47} This new perception resulted in the acceptance of fatigue as an important
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13 outcome for clinical trials.^{48 49} Fatigue was subsequently added to the RA core set as a
14
15 recommended outcome.⁵⁰

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

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29 The thematic document analysis provided additional evidence for the statement that without
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31 patients raising their voice at OMERACT 6 fatigue would not have been high on the research
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33 agenda. The issue of fatigue was not new for rheumatologists.⁵⁴⁻⁵⁶ Fatigue was a symptom
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35 regularly reported during clinical consultations, but not incorporated in guidelines for
36
37 monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and
38
39 incorporated in a disease status questionnaire.⁵⁷ And during OMERACT 3 (1996) delegates
40
41 carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal
42
43 diseases. The discussion groups identified outcomes such as pain, depression, anxiety and
44
45 fatigue as major concerns.⁵⁸ For fatigue eight examples of measurement instruments were
46
47 given.⁵⁹ However, after this workshop, nothing happened for six years, until patients raised
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49 the urgency of fatigue as a serious symptom.
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3 Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only
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5 hearing from partners at OMERACT made them change their perception of fatigue as an
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7 important outcome:
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11 “Because when I was working in oncology before, during university training, of course
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13 we saw that the patients were lying in bed all day and we knew they were exhausted, call
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15 that fatigue. But patients with RA, we were ignorant.” [RC]
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21 Another physician, involved in OMERACT from the start:
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25 “We were first discussing on fatigue and to be honest: I never ever had before heard of
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27 fatigue being a problem in rheumatology. So it got into my mind and then I got thinking
28
29 about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a
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31 100% positive response. So it was like a coming out, you know. I listened to the patients
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33 before but bringing it to a specific topic, that was really what I learned at OMERACT.”
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36 [RA]
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41 [Insert Table 2]
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45 ***The development of core outcome measurement sets and patient reported outcomes –***

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47 During the first two conferences including patients the focus of partners’ contributions was on
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49 agenda-setting and identifying relevant outcomes for clinical trials. Then, partners became
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51 gradually involved on different levels in other OMERACT activities, varying from being
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53 consulted (e.g. in a Delphi process) to full collaboration (as partner and as co-author). They
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55 contributed by identifying domains that are relevant for disease-specific core sets for psoriatic
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3 arthritis,⁶⁰ fibromyalgia,^{61 62} gout⁶³ and vasculitis.⁶⁴ Furthermore they contributed to the
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5 development of core outcome measurement sets for methodological or clinical concepts such
6
7 as MCID and remission.⁶⁵ Partners have also played a role in the assessment of the feasibility
8
9 of instruments and core sets, one of the three key components of the OMERACT Filter.⁶⁶
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12 Partners have been helpful in the development of PRO measurement instruments in the
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14 field of work productivity, monitoring adverse events,⁶⁷ flares⁶⁸ and psychosocial
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16 interventions such as self-management programs⁶⁹. At the 2010 conference, during the
17
18 plenary session on flares in RA, one of the partners gave a personal testimony about the
19
20 devastating impact of the unpredictable nature of RA. A professional in the audience was
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22 surprised and reported: “It demonstrates that the disease activity fluctuates more than we can
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24 see in our data: Our instruments are more flat, and by the limited frequency of measuring we
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26 filter fluctuations out”. [RI]
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32 Regulators require strong evidence for the effectiveness of new medicines by demonstrating
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34 accurately that they reduce structural progression as well as patient important outcomes. By
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36 doing both of these, developing standards for high quality imaging techniques and exploring
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38 new PRO’s and translating them into valid and feasible measures, OMERACT has been
39
40 extremely advantageous for the negotiations with regulators about the registration and
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42 relatively generous reimbursement of new biologic agents:
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48 “I think, to be really honest, the patient involvement process in OMERACT and the
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50 changes in outcome measurements and the use of them in the drug tests has made a
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52 real difference for so many patients.” [RB]
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3 *The culture of OMERACT*– In spite of the initial unanimous vote to invite patients, some
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5 researchers were concerned about changing the layout of the conference:
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10 “My original expectation of a limited contribution was based on fear that patients were
11
12 not able to transcend their personal experience and to generalize ... new stakeholders
13
14 often don’t have knowledge about clinometric.” [RE]
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18 In retrospect researchers explained that they deferred to the proposal in order to reflect a core
19
20 principle of OMERACT of not immediately rejecting new ideas: “To respect and listen rather
21
22 than just react”. [RA]
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25 Looking back, the number of professional participants who were in favour of partners
26
27 at the conference slowly increased: “I was impressed by the very good working flow”. [RC]
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29 Participants confirmed that the presence of partners has changed their way of thinking and
30
31 talking. “They made my blind spot visible” [RK] and another professional reported:
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36 “Now what we have found is, and I changed my view, [be]cause it wasn’t only from
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38 OMERACT. As I got to know more and more patients, I realized, this sounds stupid
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40 because it’s so obvious but it wasn’t obvious to me, that a patient isn’t their disease. A
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42 patient is a person who happens to have a disease. What a big difference. Because if
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44 you’re a person that happens to have a disease, then for example you might have
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46 incredible skills in an area that might be very useful to move a clinical trial forward.
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48 So once I came to that realization then patient involvement becomes an absolutely
49
50 obvious and integral part of moving forward.” [RA]
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3 Partners improved communication and brought dynamics to the dialogue because they are
4 motivated and constructive, without a personal agenda. At a conference such as OMERACT,
5 where the discussion about methodology may become extremely technical, partners reminded
6 participants of the common goal of the conference by providing a human face of a person
7 living with the condition day by day. Their presence made participants more explicit about the
8 objectives of sessions and more explanatory about terms and concepts under discussion.
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10 Together with a reduced use of jargon this 'forced' simplification resulted in fewer
11 misunderstandings for everyone.
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20 For some professionals the presence of partners complicated the communication.
21 Some believed that partners slow down the process because they are not familiar with
22 technical issues. Others felt disinclined to say what they wanted to out of respect to partners
23 or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and
24 put her own expertise aside to keep things simple: "Patients didn't sometimes understand the
25 objective of the research, which hindered us". [RK] One of the partners admitted that "it is a
26 thin line between providing input and causing irritation". [PN]
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38 An analysis of the responses of patients attending OMERACT for the first time showed that
39 new partners experienced a significant learning curve and a variety of personal benefits.⁷⁰
40 Results from this study suggest that in fact all participants learned from the contact with other
41 stakeholders. During this process participants gained trust, respect and understanding,
42 reflecting the emergence of relational empowerment:
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51 "Patients were a kind of sparring partner when I entered a relatively new area. That
52 was fun and did clarify a lot". [RK]
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3 Relational empowerment in the context of health research can be understood as a process in
4
5 which traditional doctor-patient relationships transform into equal partnerships enabling
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7 mutual learning processes.⁷¹ All participants become stronger by sharing knowledge and
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9 responsibilities, and educating and helping each other.
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12 The reported benefits were easiest to identify at the beginning when the level of
13
14 involvement was still low. They became more diffuse when partners were structurally
15
16 involved as full and equal collaborators. One interviewee mentioned “a reality check” as an
17
18 important benefit of partners attending the conference. For professionals it offered the
19
20 opportunity to check the relevance of the scope of their research: Are we doing the right
21
22 things according to patients and are we using the right tools and methods? It is a belief of
23
24 professionals that this kind of feedback is important to legitimize their research and, together
25
26 with the belief of partners that without this research no innovations will take place, it
27
28 strengthened the mutual empowerment of both.
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34 ***Consequences outside OMERACT*** – The lessons learnt at OMERACT were noticed by the
35
36 outside world. Partners returning home after the conference have continued introducing
37
38 patient participation in local and national research projects or established networks of patient
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40 research partners.⁷²⁻⁷⁵ Some delegates published a working framework for incorporating the
41
42 patient perspective in outcome research.⁷⁶ With the input from several OMERACT
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44 participants the European League Against Rheumatism (EULAR) developed
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46 recommendations for the inclusion of patient representatives in scientific projects.²⁵
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48 Following these recommendations a new patient reported quality of life instrument for RA
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50 was developed and validated.^{40 77} Based on the experiences of OMERACT the organizing
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52 committee of the 6th International Shared Decision Making conference decided in 2011 to
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54 invite patient participants.⁷⁸ In the same year OMERACT delegates, partners as well as
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3 professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials
4 (COMET) conference, demonstrating how the OMERACT methodology can be utilized in
5 other disease areas.¹²
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11 ***Remaining challenges emerging from the interviews*** – The role and contribution of patient
12 participants have changed over time and procedures for patient selection and support have
13 been developed in order to identify patient participants who are able to make a difference.
14 There is still a debate going on whether patients should be selected through strict criteria such
15 as education, communication skills, attitude and familiarity with scientific research. Some
16 argue that an expert meeting like OMERACT needs expert patients who have extended
17 knowledge about methodologies of outcome research, and are able to provide a kind of
18 aggregated patient input. At OMERACT this group represents a minority of delegates, who
19 are reluctant to allocate the same rights and power to partners as to the professionals. The vast
20 majority believes that many patients are able to contribute to an OMERACT conference and
21 emphasizes that a heterogeneous group of partners in age, gender, condition, experience and
22 cultural background are advantageous for the conference. They intend to develop full
23 representative participation in all phases of research by including partners in working group
24 activities between conferences. Finally, some participants point out the potential risks of
25 partners who become too experienced. They appreciate the naive input as a patient, with a
26 minimum of preparation and reflection. They assume that as soon as you start thinking about
27 your contribution, you lose the unique, individual perspective and become a patient-expert
28 who aligns too easily with professionals.
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52 Professionals shared the opinion that partners need training, although they reported
53 different ideas about the content and aims of such training. Experienced partners as well as
54 novice researchers felt that any new participant has to learn the OMERACT objectives,
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3 culture and procedures first, before they can become fully productive, mostly at the second or
4
5 third conference. This accords with the expectations of partners who attended OMERACT for
6
7 the first time.
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10 11 **Overview of findings**

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14 These results show that a decade of patient involvement has been successful and had a
15
16 significant impact on various aspects of outcome research. Perspectives of patients are
17
18 different from those of health professionals. Broad consensus exists that partners at
19
20 OMERACT have played a vital role in identifying domains relevant from the perspective of
21
22 patients and in developing new PROs such as fatigue, sleep quality, flares and work
23
24 productivity. Especially in the area of fatigue we have shown that patient involvement on
25
26 different levels and in different phases improves the quality of outcome research. By
27
28 combining evidence-based knowledge of researchers and the experiential knowledge of
29
30 patients, a synthesis of both kinds of knowledge has been achieved and documented. The
31
32 benefits are assessment tools that accurately measure what really matters to patients, are
33
34 formulated in understandable language and are user-friendly. Other benefits go beyond
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36 improving clinical outcome research and include improved communication, mutual
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38 empowerment, changed attitudes and substantial consequences outside OMERACT.
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45 **Discussion**

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47 We set out to describe and evaluate the contribution of patients as partners in rheumatology
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49 outcome research, reviewing their impact on the research agenda and the culture and process
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51 of the OMERACT conference. The document analysis provided the recorded facts while the
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53 interviews allowed an exploration of intentions, attitudes and perceived benefits or harms of
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55 patient participation that complements the document analysis. Since validated methodologies
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3 for demonstrating impact of collaboration with patients in the context of research are lacking,
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5 a responsive interview methodology seemed to be a good approach.
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7 Both strengths and limitations of this study relate to the personal experience of the first
8 and last authors as participants in the developing process of patient partner participation.
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10 Having witnessed the OMERACT process, actors and concerns of both the patient community
11 and the research community was advantageous during the development of interview
12 protocols, recruitment and selection of respondents and data analysis. For instance, the
13 knowledge of the opinions of other participants made it possible to achieve maximum
14 variation. Also, the active involvement in the support and training of partners created an
15 adequate awareness of the relevant items to include in the study. The drawbacks of this
16 engagement are the risks of subjectivity, blind spots and over- or under-identification with
17 particular stakeholders. These risks have been addressed by applying strict quality measures
18 for scientific rigor in qualitative, evaluation research.
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31 The composition of the research team purposely included two external experts in
32 qualitative research and a patient research partner, who were actively involved in the coding
33 of interview transcripts and distilling relevant categories for impact, reduced the risk of
34 subjectivity. Bias was avoided by the check-coding procedure in the analysis of the transcripts
35 as at least two researchers independently coded each transcript, after which the whole team
36 discussed the codes until consensus was reached. Saturation was also part of the discussion in
37 the whole team. The inclusion of various stakeholder perspectives prevented one-sidedness.
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39 No signals were identified to suggest that interviewees have simply given desirable answers,
40 or just been friendly to the interviewer. Some interviewees have been rather critical, reporting
41 several barriers for structural involvement of patients in research, but have always added
42 constructive suggestions for improvement. Peer debriefing by an independent colleague (TA)
43 further helped to prevent bias.
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3 Other limitations relate to the difficulties of demonstrating the ‘impact’ of patient
4 involvement.⁹ In OMERACT there is a strong belief that patient participation works, a belief
5 that is nourished by the world-wide transition towards more patient-oriented health care and
6 health research.⁷⁹⁻⁸² The assumption however that long term involvement of patients as equal
7 partners guarantees sustainable inclusion of the patient perspective in outcome research
8 complicates a thorough evaluation and makes it difficult to distinguish between expected,
9 perceived and actual contributions. Many participants, not only partners, but also young
10 researchers and other new-comers, are able to identify their own contribution and may not see
11 how their input is reflected in the final outcomes. Partners reported almost unanimously not
12 being able to confirm substantial contributions during their participation but they believed
13 they did. More experienced participants, mostly professionals, were less reluctant in reporting
14 illustrative examples of patient contributions.

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

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54 A last obstacle for demonstrating the influence of patient participation is the invisibility
55 of experiential knowledge, often hidden in anecdotal stories. It has an impact that is rarely
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3 claimed by patients nor perceived by professionals. Personal comments are normally not
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5 reported because they are not seen as a valuable and valid source of knowledge⁸³ and yet
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7 clear documentation of meetings is required to ensure that patients' contributions become
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9 visible.⁸⁴ Professionals focus on synthesizing data and may not notice that the dialogue with
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11 patients works like a reality check, generates new ideas or changes their beliefs, behaviour or
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13 perception. When partners appear to simply agree with the results presented at OMERACT it
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15 might look as if they do not have any contribution to make, but in fact they confirm the value
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17 of the work under discussion and provide face validity to the process. It is for this reason that
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19 most professionals appreciate the feedback and input from partners, although not all are aware
20
21 of this reason. Realizing the importance of such a reality check is beneficial for the
22
23 management of realistic expectations: do not expect innovative ideas, brilliant suggestions
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25 and new concepts when inviting partners to join research. Their contributions are often more
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27 subtle and need the attention of a modest and committed researcher to be noticed.
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32 Despite these limitations, we believe that the results presented in this study are relevant
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34 and valid. It is undeniable that there is a growing belief that patient involvement has been
35
36 successful and brought a unique added value to the conference. Even those who were
37
38 originally among the most skeptical participants now report that they have changed their
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40 perception about the expected contribution of patient research partners. This study is
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42 conducted within the context of a scientific research conference in the field of rheumatology,
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44 a long term somatic condition. Our ability to generalize the findings is therefore limited and
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46 extrapolation to other research contexts or to other conditions should be done with care.
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References

1. Keizer B, Bless R. Pilot study on the position of health consumer and patients' organisations in seven EU countries. Den Haag: ZonMW, 2010:54.
2. Involve. People & participation; How to put citizens at the heart of decision-making. London: Beacon Press, 2005.
3. Staley K. Summary Exploring Impact: Public involvement in NHS, public health and social care research. Eastleigh.: INVOLVE, 2009.
4. Wright MT, Roche B, von Unger H, Block M, Gardner B. A call for an international collaboration on participatory research for health. *Health Promot Int* 2010;25(1):115-22.
5. Abma TA. Patients as partners in a health research agenda setting: the feasibility of a participatory methodology. *Eval Health Prof* 2006;29(4):424-39.
6. Abma TA, Nierse C, Widdershoven G. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qualitative Health Research* 2009;19(3):401-15.
7. Abma TA, Broerse JE. Patient participation as dialogue: setting research agendas. *Health Expect* 2010;13(2):160-73.
8. Caron-Flinterman JF, Broerse JE, Bunders JF. The experiential knowledge of patients: a new resource for biomedical research? *Soc Sci Med* 2005;60(11):2575-84.
9. Staniszewska S, Haywood KL, Brett J, Tutton L. Patient and public involvement in patient-reported outcome measures: evolution not revolution. *Patient* 2012;5(2):79-87.
10. Speight J, Barendse SM. FDA guidance on patient reported outcomes. *BMJ* 2010;340:c2921.
11. Rowen D, Carlton J, Brazier JE, Mulhern B, Palfreyman S, Stevens K, et al. FDA on PROMs. Two important points. *BMJ* 2010;341:c5454.

- 1
2
3 12. Williamson PR, Altman DG, Blazeby JM, Clarke M, Gargon E. The COMET (Core
4 Outcome Measures in Effectiveness Trials) Initiative. *Trials* 2011;12 (Suppl 1)(A70).
5
6
7 13. Clarke M. Standardising outcomes for clinical trials and systematic reviews. *Trials*
8 2007;8:39.
9
10
11 14. Sinha IP, Smyth RL, Williamson PR. Using the Delphi technique to determine which
12 outcomes to measure in clinical trials: Recommendations for the future based on a
13 systematic review of existing studies. *PLoS Med* 2011;8(1).
14
15
16 15. Williamson P, Clarke M. The COMET (Core Outcome Measures in Effectiveness Trials)
17 Initiative: Its Role in Improving Cochrane Reviews. *Cochrane Database Syst Rev*
18 2012;5:ED000041.
19
20
21 16. Saag KG. OMERACT 6 brings new perspectives to rheumatology measurement research.
22
23
24
25
26
27
28
29 17. Heller JE, Shadick NA. Outcomes in rheumatoid arthritis: incorporating the patient
30 perspective. *Current Opinion in Rheumatology* 2007;19(2):101-05.
31
32
33 18. OMERACT. Guiding principles for patient research partner participation in OMERACT.
34 Accessed: October 18; 2012: Available from: [http://www.omeract.org/pdf/2011-03-
35 27%20Agreed%20Patient%20Participation%20Policy.pdf](http://www.omeract.org/pdf/2011-03-27%20Agreed%20Patient%20Participation%20Policy.pdf).
36
37
38
39 19. Guba EG, Lincoln YS. *Effective evaluation. Improving the usefulness of evaluation results
40 through responsive and naturalistic approaches*. San Francisco: Jossey-Bass
41 Publishers, 1988.
42
43
44
45
46 20. Abma TA, Widdershoven GAM. Evaluation as a relationally responsible practice. In:
47 Denzin N, Lincoln Y, editors. *Handbook for Qualitative Inquiry*. Los Angeles: SAGE
48 Publications Ltd., 2011:669-80.
49
50
51
52 21. Lincoln Y, Guba EG. *Naturalistic inquiry*. Newbury Park: SAGE, 1985.
53
54
55
56
57
58
59
60

- 1
2
3 22. Hewlett S, Carr M, Ryan S, Kirwan J, Richards P, Carr A, et al. Outcomes generated by
4 patients with rheumatoid arthritis: how important are they? *Musculoskeletal Care*
5 2005;3(3):131-42.
6
7
8
9
10 23. Hewlett S, Cockshott Z, Byron M, Kitchen K, Tipler S, Pope D, et al. Patients' perceptions
11 of fatigue in rheumatoid arthritis: overwhelming, uncontrollable, ignored. *Arthritis*
12 *and rheumatism* 2005;53(5):697-702.
13
14
15
16 24. Kirwan J, Heiberg T, Hewlett S, Hughes R, Kvien T, Ahlmen M, et al. Outcomes from the
17 Patient Perspective Workshop at OMERACT 6. *Journal of Rheumatology*
18 2003;30(4):868-72.
19
20
21
22
23 25. de Wit MPT, Berlo SE, Aanerud GJ, Aletaha D, Bijlsma JW, Croucher L, et al. European
24 League Against Rheumatism recommendations for the inclusion of patient
25 representatives in scientific projects. *Annals of the Rheumatic Diseases*
26 2011;70(5):722-6.
27
28
29
30
31
32 26. Tugwell P, Boers M. OMERACT conference on outcome measures in rheumatoid arthritis
33 clinical trials: introduction. *J Rheumatol* 1993;20(3):528-30.
34
35
36
37 27. Brooks PM, Boers M, Tugwell P. OMERACT III: the "ACT" revisited. *Jrn of Rheum*
38 1997;24(4):764-5.
39
40
41
42 28. Wells G, Anderson J, Beaton D, Bellamy N, Boers M, Bombardier C, et al. Minimal
43 clinically important difference module: summary, recommendations, and research
44 agenda. *J Rheumatol* 2001;28(2):452-4.
45
46
47 29. Kirwan JR, Carr A, Gluck O, Hewlett S. Proposal for inclusion of a 'Patient's
48 perceptions' module in OMERACT VI [unpublished], 2000.
49
50
51
52 30. Quest E, Aanerud GJ, Kaarud S, Collins S, Leong A, Smedeby B, et al. Patients'
53 perspective. *J Rheumatol* 2003;30(4):884-5.
54
55
56
57
58
59
60

- 1
2
3 31. Brooks PM, Tugwell P, Strand C, Simon L, Boers M. OMERACT 6: International
4
5 consensus conference on outcome measures in rheumatology: Introduction. *Jrn of*
6
7 *Rheum* 2003;30(4):866-7.
8
9
10 32. Kirwan J. The patient perspective workshop at OMERACT 6: Rationale and methodology
11
12 [unpublished preconference paper]. 2002.
13
14 33. Tugwell P, Boers M, Brooks P, Simon L, Strand V, Idzerda L. OMERACT: an
15
16 international initiative to improve outcome measurement in rheumatology. *Trials*
17
18 2007;8:38.
19
20
21 34. Hewlett SA. Patients and clinicians have different perspectives on outcomes in arthritis. *J*
22
23 *Rheumatol* 2003;30(4):877-9.
24
25 35. Leeb BF, Sautner J, Leeb BA, Fassel C, Rintelen B. Lack of agreement between patients'
26
27 and physicians' perspectives of rheumatoid arthritis disease activity changes. *Scand J*
28
29 *Rheumatol* 2006;35(6):441-6.
30
31
32 36. Kvien TK, Heiberg T. Patient perspective in outcome assessments--perceptions or
33
34 something more? *J Rheumatol* 2003;30(4):873-6.
35
36 37. Carr A, Hewlett S, Hughes R, Mitchell H, Ryan S, Carr M, et al. Rheumatology
37
38 outcomes: the patient's perspective. *Journal of Rheumatology* 2003;30(4):880-3.
39
40 38. Ahlmen M, Nordenskiöld U, Archenholtz B, Thyberg I, Ronnqvist R, Linden L, et al.
41
42 Rheumatology outcomes: the patient's perspective. A multicentre focus group
43
44 interview study of Swedish rheumatoid arthritis patients. *Rheumatology (Oxford)*
45
46 2005;44(1):105-10.
47
48
49 39. Rupp I, Boshuizen HC, Roorda LD, Dinant HJ, Jacobi CE, van den Bos G. Course of
50
51 patient-reported health outcomes in rheumatoid arthritis: comparison of longitudinal
52
53 and cross-sectional approaches. *J Rheumatol* 2006;33(2):228-33.
54
55
56
57
58
59
60

- 1
2
3 40. Gossec L, Paternotte S, Aanerud GJ, Balanescu A, Boumpas DT, Carmona L, et al.
4
5 Finalisation and validation of the rheumatoid arthritis impact of disease score, a
6
7 patient-derived composite measure of impact of rheumatoid arthritis: a EULAR
8
9 initiative. *Annals of the Rheumatic Diseases* 2011;70(6):935-42.
10
11
12 41. Hewlett S, Cockshott Z, Byron M, Kitchen K, Tipler S, Pope D, et al. Patients' perceptions
13
14 of fatigue in rheumatoid arthritis: overwhelming, uncontrollable, ignored. *Arthritis*
15
16 *Rheum* 2005;53(5):697-702.
17
18
19 42. Rupp I, Boshuizen HC, Jacobi CE, Dinant HJ, van den Bos GA. Impact of fatigue on
20
21 health-related quality of life in rheumatoid arthritis. *Arthritis Rheum* 2004;51(4):578-
22
23 85.
24
25
26 43. Repping-Wuts H, Uitterhoeve R, van Riel P, van Achterberg T. Fatigue as experienced by
27
28 patients with rheumatoid arthritis (RA): A qualitative study. *Intl J Nursing Studies*
29
30 2008;8(45 (7)):995-102.
31
32
33 44. Nikolaus S, Bode C, Taal E, van de Laar MA. Four different patterns of fatigue in
34
35 rheumatoid arthritis patients: results of a Q-sort study. *Rheumatology*
36
37 2010;49(11):2191-9.
38
39
40 45. Hewlett S, Hehir M, Kirwan JR. Measuring fatigue in rheumatoid arthritis: a systematic
41
42 review of scales in use. *Arthritis & Rheumatism* 2007;57(3):429-39.
43
44
45 46. Kirwan JR, Hewlett S. Patient perspective: reasons and methods for measuring fatigue in
46
47 rheumatoid arthritis. *J Rheumatol* 2007;34(5):1171-3.
48
49
50 47. Hewlett S, Chalder T, Choy E, Cramp F, Davis B, Dures E, et al. Fatigue in rheumatoid
51
52 arthritis: time for a conceptual model. *Rheumatology (Oxford)* 2010.
53
54
55 48. Kirwan JR, Minnock P, Adebajo A, Bresnihan B, Choy E, de Wit M, et al. Patient
56
57 perspective: fatigue as a recommended patient centered outcome measure in
58
59 rheumatoid arthritis. *J Rheumatol* 2007;34(5):1174-7.
60

- 1
2
3 49. Pincus T, Sokka T. Complexities in the quantitative assessment of patients with rheumatic
4
5 diseases in clinical trials and clinical care. *Clin Exp Rheumatol* 2005;23(5 Suppl
6
7 39):S1-9.
8
9
10 50. Kirwan JR, Minnock P, Adebajo A, Bresnihan B, Choy E, de Wit M, et al. Patient
11
12 perspective: fatigue as a recommended patient centered outcome measure in
13
14 rheumatoid arthritis. *Journal of Rheumatology* 2007;34(5):1174-7.
15
16 51. Nicklin J, Cramp F, Kirwan J, Greenwood R, Urban M, Hewlett S. Measuring fatigue in
17
18 rheumatoid arthritis: A cross-sectional study to evaluate the Bristol Rheumatoid
19
20 Arthritis Fatigue Multi-Dimensional questionnaire, visual analog scales, and
21
22 numerical rating scales. *Arthritis Care Res (Hoboken)* 2010;62(11):1559-68.
23
24 52. Nikolaus S, van de Laar MA. Measuring fatigue in rheumatoid arthritis. *Nat Rev*
25
26 *Rheumatol* 2011;7(10):562-4.
27
28 53. Repping-Wuts H. Fatigue in patients with rheumatoid arthritis (thesis). Nijmegen
29
30 University, 2009.
31
32 54. Wolfe F, Hawley DJ, Wilson K. The prevalence and meaning of fatigue in rheumatic
33
34 disease. *The Journal of rheumatology* 1996;23(8):1407-17.
35
36 55. Tack BB. Fatigue in rheumatoid arthritis. Conditions, strategies, and consequences.
37
38 *Arthritis Care & Research* 1990;3(2):65-70.
39
40 56. Crosby LJ. Factors which contribute to fatigue associated with rheumatoid arthritis. *J Adv*
41
42 *Nurs* 1991;16(8):974-81.
43
44 57. Garrett S, Jenkinson T, Kennedy LG, Whitelock H, Gaisford P, Calin A. A new approach
45
46 to defining disease status in ankylosing spondylitis: the Bath Ankylosing Spondylitis
47
48 Disease Activity Index. *The Journal of rheumatology* 1994;21(12):2286-91.
49
50 58. Newman SP. Psychosocial measures in musculoskeletal trials. *J Rheumatol*
51
52 1997;24(5):979-84.
53
54
55
56
57
58
59
60

- 1
2
3 59. Brooks P, McFarlane AC, Newman S, Rasker JJ. Psychosocial measures. *J Rheumatol*
4
5 1997;24(5):1008-11.
6
7 60. Gladman DD, Mease PJ, Strand V, Healy P, Helliwell PS, Fitzgerald O, et al. Consensus
8
9 on a core set of domains for psoriatic arthritis. *The Journal of rheumatology*
10
11 2007;34(5):1167-70.
12
13 61. Mease P, Arnold LM, Choy EH, Clauw DJ, Crofford LJ, Glass JM, et al. Fibromyalgia
14
15 syndrome module at OMERACT 9: domain construct. *The Journal of rheumatology*
16
17 2009;36(10):2318-29.
18
19 62. Choy EH, Arnold LM, Clauw DJ, Crofford LJ, Glass JM, Simon LS, et al. Content and
20
21 criterion validity of the preliminary core dataset for clinical trials in fibromyalgia
22
23 syndrome. *The Journal of rheumatology* 2009;36(10):2330-4.
24
25 63. Taylor WJ, Schumacher HR, Jr., Baraf HS, Chapman P, Stamp L, Doherty M, et al. A
26
27 modified Delphi exercise to determine the extent of consensus with OMERACT
28
29 outcome domains for studies of acute and chronic gout. *Annals of the rheumatic*
30
31 *diseases* 2008;67(6):888-91.
32
33 64. Merkel PA, Aydin SZ, Boers M, Direskeneli H, Herlyn K, Seo P, et al. The OMERACT
34
35 core set of outcome measures for use in clinical trials of ANCA-associated vasculitis.
36
37 *The Journal of rheumatology* 2011;38(7):1480-6.
38
39 65. van Tuyl LH, Smolen JS, Wells GA, Scholte-Voshaar M, Hoogland W, Boers M. Patient
40
41 perspective on remission in rheumatoid arthritis. *The Journal of rheumatology*
42
43 2011;38(8):1735-8.
44
45 66. Boers M, Brooks P, Strand CV, Tugwell P. The OMERACT filter for Outcome Measures
46
47 in Rheumatology. *The Journal of rheumatology* 1998;25(2):198-9.
48
49 67. Lassere MN, Johnson KR, Boers M, Carlton K, Day RO, de Wit M, et al. Standardized
50
51 assessment of adverse events in rheumatology clinical trials: summary of the
52
53
54
55
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2
3 OMERACT 7 drug safety module update. *Journal of Rheumatology*
4
5 2005;32(10):2037-41.
6
7 68. Bingham CO, 3rd, Alten R, de Wit MP. The importance of patient participation in
8
9 measuring rheumatoid arthritis flares. *Annals of the rheumatic diseases* 2012.
10
11 69. Kristjansson E, Tugwell PS, Wilson AJ, Brooks PM, Driedger SM, Gallois C, et al.
12
13 Development of the effective musculoskeletal consumer scale. *The Journal of*
14
15 *rheumatology* 2007;34(6):1392-400.
16
17 70. de Wit MPT, Koelewijn-van Loon MS, Collins S, Abma TA, Kirwan JR. How patients
18
19 contribute a new voice at a scientific conference: their expectations and experiences at
20
21 the Outcome Measures in Rheumatology Conference 2010. (*forthcoming*) 2012.
22
23 71. Abma TA, Nierse CJ, Widdershoven GA. Patients as partners in responsive research:
24
25 methodological notions for collaborations in mixed research teams. *Qual Health Res*
26
27 2009;19(3):401-15.
28
29 72. Kjekken I, Ziegler C, Skrolsvik J, Bagge J, Smedslund G, Tovik A, et al. How to develop
30
31 patient-centered research: some perspectives based on surveys among people with
32
33 rheumatic diseases in Scandinavia. *Physical Therapy* 2010;90(3):450-60.
34
35 73. Akerhielm CE. Patient participation in research - A Swedish model. *Ann Rheum Dis*
36
37 2009;68(Suppl3):800.
38
39 74. Akerhielm CE. Patient research partners - A way to participation and influence in
40
41 research. *Ann Rheum Dis* 2010;69(Suppl3):753.
42
43 75. Hewlett S. Learnings from the Bristol experience - Practicalities of a decade of patient
44
45 involvement in research on a local level. *Ann Rheum Dis* 2010;69((Suppl3)):29.
46
47 76. Hewlett S, De Wit M, Richards P, Quest E, Hughes R, Heiberg T, et al. Patients and
48
49 professionals as research partners: challenges, practicalities, and benefits. *Arthritis &*
50
51 *Rheumatism* 2006;55(4):676-80.
52
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2
3 77. Gossec L, Dougados M, Rincheval N, Balanescu A, Boumpas DT, Canadello S, et al.
4
5 Elaboration of the preliminary Rheumatoid Arthritis Impact of Disease (RAID) score:
6
7 a EULAR initiative. *Annals of the Rheumatic Diseases* 2009;68(11):1680-5.
8
9
10 78. Stiggelbout AM, Weijden TV, Wit MP, Frosch D, Legare F, Montori VM, et al. Shared
11
12 decision making: really putting patients at the centre of healthcare. *BMJ*
13
14 2012;344:e256.
15
16 79. Beresford P. User involvement, research and health inequalities: developing new
17
18 directions. *Health Soc Care Community* 2007;15(4):306-12.
19
20
21 80. Ward PR, Thompson J, Barber R, Armitage CJ, Boote JD, Cooper CL, et al. Critical
22
23 perspectives on 'consumer involvement' in health research: Epistemological
24
25 dissonance and the know-do gap. *Journal of Sociology* 2009;46(1):63-82.
26
27
28 81. Lindenmeyer A, Hearnshaw H, Sturt J, Ormerod R, Aitchison G. Assessment of the
29
30 benefits of user involvement in health research from the Warwick Diabetes Care
31
32 Research User Group: a qualitative case study. *Health Expectations* 2007;10(3):268-
33
34 77.
35
36 82. Broerse JEW, Elberse JE, Caron-Flinterman JFW, Zweekhorst MBM. Enhancing a
37
38 transition towards a needs-oriented health research system. In: Broerse JEW, Bunders
39
40 JFG, editors. *Transitions in Health Systems: Dealing with persistent problems*.
41
42 Amsterdam: VU University Press, 2010:181-205.
43
44
45 83. Schipper K. Patient participation & knowledge [thesis]. VU University, 2011.
46
47
48 84. Jinks C, Ong BN, O'Neill TJ. The Keele community knee pain forum: action research to
49
50 engage with stakeholders about the prevention of knee pain and disability. *BMC*
51
52 *Musculoskelet Disord* 2009;10:85.
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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	1	5	8
OMERACT conferences attended	2	0	3
	3	1	1
	4	4	4
	≥5	6	0
Interview in relation to	Before	2	1
	During	8	16

} PG to PN

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OMERACT

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

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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: <ul style="list-style-type: none"> • Well being • Fatigue • Sleep disturbance • Flares 	Identification of patient relevant domains to include in core sets for clinical trials: <ul style="list-style-type: none"> • Fibromyalgia • Psoriatic Arthritis • Vasculitis • Gout • MRI • MCID 	Acceptable, understandable and feasible outcome measures for <ul style="list-style-type: none"> • Monitoring adverse events • Work productivity • Flares • Psychosocial interventions 	<ul style="list-style-type: none"> • Attitudes • Communication • Perceptions • Motivation • Relational empowerment • Personal benefits 	<ul style="list-style-type: none"> • Local initiatives • Local and national networks of partners • EULAR • COMET • ISDM

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	<ul style="list-style-type: none"> • Remission 			
Reality check				

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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.

Journal:	<i>BMJ Open</i>
Manuscript ID:	bmjopen-2012-002241.R2
Article Type:	Research
Date Submitted by the Author:	02-Apr-2013
Complete List of Authors:	Wit, Maarten; VU Medical Centre , Metamedica Abma, Tineke; VU Medical Centre, Metamedica Koelewijn-van Loon, Marije; Maastricht University Medical Centre, Department of General Practice Collins, Sarah; Patient Research Partner, Kirwan, John; University of Bristol, Rheumatology
Primary Subject Heading:	Rheumatology
Secondary Subject Heading:	Qualitative research, Patient-centred medicine
Keywords:	HEALTH IMPACT ASSESSMENT, QUALITATIVE RESEARCH, RHEUMATOLOGY

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Manuscripts

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3 **Involving patient research partners has a significant impact on outcomes**
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5 **research: an example from the international OMERACT conferences.**
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11 **Authors:**

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13 Wit, Maarten¹; Abma, Tineke²; Koelewijn-van Loon, Marije²; Collins, Sarah³; Kirwan, John⁴
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42 **Keywords:** HEALTH IMPACT ASSESSMENT , QUALITATIVE RESEARCH,
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44 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.

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- 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.

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

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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

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3 Two conferences later others perceived the involvement of patients as “indicative of the
4 beginning of a paradigm shift in thinking about RA outcomes over the last 5 years”.¹⁷ Since
5 then OMERACT has formulated three principles recognising the essential role of patients in
6 outcome research.¹⁸ First, patients’ input is indispensable when defining relevant outcome
7 measures, identifying domains that are important from the perspective of patients, and
8 assessing feasibility of measurement tools. Second, structural involvement of patients during
9 the whole research process provides face validity. Third, OMERACT intends “to ground
10 theoretical discussions in the lived experience of arthritis, and in concepts which can be
11 readily communicated to patients to help with therapeutic decision making”.¹⁸
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23 However, the validity of these arguments has never been substantiated by robust evidence
24 for the effectiveness of patient participation in OMERACT and it is not clear whether or how
25 this involvement has influenced methodologies, procedures, attitudes, and research outcomes.
26 Therefore, the objective of this study is to describe and evaluate the contributions made by
27 patients since OMERACT started implementing structural patient participation in its
28 conferences. We review the impact of patients on the research agenda and the development of
29 patient reported outcomes (PRO’s) and explore how including patients has influenced the
30 culture and structure of the OMERACT conference
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43 **Method**

44 Patient participation in research is a new phenomenon and often not reported or reflected on
45 in scientific publications. This lack of written sources in the scientific literature complicates
46 the study of the process and impact of patient participation through a review of relevant
47 literature. A provisory search using Pubmed (March 2010) for the terms “patient
48 participation” OR “patient involvement” OR “user involvement” OR “consumer
49 involvement” AND “OMERACT” did not generate any relevant reference. Therefore we
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3 conducted a content analysis of relevant documents (any written material on the topic of
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5 patient participation).
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7 Documents are a stable, rich source of contextual information, providing well-grounded
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9 data on events or situations at low costs. A sound document analysis is rule-bound,
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11 systematic, following a coding process where raw data are aggregated into units describing
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13 the content.¹⁹ We included OMERACT conference proceedings as published by The Journal
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15 of Rheumatology (1992-2010) and 'grey literature' such as correspondence, invitations,
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17 session reports, e-mails and OMERACT policy documents. The review focused on the
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19 arguments, reception and evolution of patient involvement in OMERACT conferences and the
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21 contributions made by patients.
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25 Subsequently a responsive evaluation took place during OMERACT 10 (Malaysia, 2010)
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27 using qualitative interviews with representatives of stakeholders. Responsive evaluation is
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29 grounded in the hermeneutic research tradition and is used by social scientists to interpret
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31 meanings that participants attribute to a phenomenon, here the history and impact of a decade
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33 of patient participation from the perspective of the conference delegates. It samples all
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35 stakeholders and does not seek consensus, but respects the plurality of opinions, values and
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37 interests. This ensures that no perspective is omitted as the result of an imbalance of power.²⁰
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41 The first author (MW) has been involved in OMERACT since 2002 as a patient
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43 participant. He has a rheumatic condition and has been educated as a responsive researcher.
44
45 Characteristics for a responsive researcher are a multiple partiality and the intent to enhance
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47 mutual understanding among all stakeholders. The last author (JK) has been involved in
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49 OMERACT since the first conference (1992) and has been the leader of the patient
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51 perspective workshop between 2002-2012. Having witnessed the involvement of patients
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53 firsthand from the very beginning MW and JK provided useful information to start the
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55 research, yet it also alerted them to critically reflect how this engagement influenced the
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3 research, and how to prevent bias. Therefore two independent experts (TA, MK) were added
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5 to the team. They had no relations with the OMERACT conference and its participants, and
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7 TA acted as peer-debriefer discussing dilemmas and challenging methodological decisions
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9 with MW.²¹
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11 The first author held 32 semi-structured interviews before, during and after the 10th
12
13 conference (Table 1) and included senior (n=10) and junior researchers (n=2), representatives
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15 of pharmacological industry and regulators (n=2), conference staff (n=2), new patient
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17 participants (n=8) and experienced patient participants (n=8). The interviewees were invited
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19 and informed by e-mail. The patient participants were aware of the purpose of the study
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21 through a one page announcement in the pre-conference patient pack and were asked for
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23 informed consent. In the Netherlands no ethical approval is required for non-intrusive
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25 interviews only.
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29 Twenty-eight interviews were recorded, transcribed by an independent secretariat and
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31 subjected to a responder check. Three interviews were summarized in a report, one interview
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33 took place without protocol and, on request of the interviewee, without recording (PF). One
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35 interview was done through Skype (PP). The average duration of the interviews was over 50
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37 minutes, most of them taking place in the humid open lobby of the conference resort. Twenty-
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39 four interviews were held in English of which six was not the native language. Eight
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41 interviews were in Dutch.
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45 The interview protocols were slightly different for professionals, new patients and
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47 experienced patients. The topics were not only derived from the document analysis but also
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49 from four pilot interviews and the personal knowledge of MW and JK and the expertise of
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51 TA. The topics dealt with: the expected role of patient participants, their selection, preparation
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53 and support, and the expected or provided contribution to the OMERACT conference.
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55 'Fatigue' was added as a potential probe because publications had already shown that this
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3 topic deserved special attention with regard to our research questions.²²⁻²⁴ Participants with
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5 long term experience in OMERACT were asked retrospectively to describe their memories of
6
7 the discussions and decisions taken about patient participation before and after 2002. Their
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9 recollections might be characterized as ‘oral history’.
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14 *Selection of interviewees* - At OMERACT 10 a total of 172 delegates participated, 152
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16 professionals and 20 patients. Nine patients attended the conference for the first time. All
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18 interviewees, except for one patient participant from the hosting country, were selected by
19
20 MW and JK following an emergent purposive sampling approach.²¹ They used a list of
21
22 attendees provided by the congress agency, covering four out of five criteria found to be
23
24 important (stakeholder background, gender, geographical spread and number of OMERACT
25
26 conferences attended). The criterion ‘opinion about patient involvement’ was assessed on the
27
28 basis of authors’ insight of the participant as being ‘positive’ (e.g. contributing to the patient
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30 perspective workshop or involving partners in own activities), ‘indifferent’ or ‘skeptical’ (e.g.
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32 resistant, not collaborating with partners). When it became clear during the process of data
33
34 collection that certain criteria were not well covered new participants were approached till
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36 maximum variation was realized. For example, two interviewees who were chosen because
37
38 of their previously reported criticism of involving patients, showed a considerable change in
39
40 perception of patient involvement in a positive way. For this reason two more interviews with
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42 professionals who had expressed critical comments during the last conference were arranged.
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44 Finally, to ensure the opinions of young investigators, two OMERACT Fellows were
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46 approached, one undertaking a PhD in translational research and one post doctoral researcher
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48 active in clinical research.
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54 Saturation was defined as a repetition of data; theoretical saturation as achieving
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56 sufficiently robust empirical data to support and describe the identified themes and main
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3 categories. Saturation was discussed and agreed within the research team. In total the
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5 perceptions and experiences of 16 patient participants and 16 professionals were collected
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7 (Table 1).
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16 *Data-analysis* - A thematic content analysis focused in particular on the reported
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18 contributions attributed to patient participants. Coding of the interviews was done separately
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20 by MW and an independent second coder (MK) who had never worked with active patient
21
22 involvement before. This resulted in 211 detailed codes that were then combined into 27 sub-
23
24 categories. During several meetings, the project team, representing various backgrounds,
25
26 discussed the codes and subcategories from a variety of perspectives, and sought natural
27
28 groupings or categories within the data. Triangulation was used in two different meanings:
29
30 First, as a means of verifying findings against another source (interview) or another method
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32 (document analysis) and to enhance the validity of the data. Second, as a means to enrich the
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34 data collection and improve the face-validity by synthesizing findings from the document
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36 analysis with the personal memories and experiences of respondents who looked back in time.
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38 By doing so, gaps in the document analysis could be filled in.
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43 The relevance and validity of the analysis and interpretation of the data was increased by the
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45 involvement of an external expert in qualitative health research (TA) as well as by inviting
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47 one of the patients (SC) who attended OMERACT 10 for the first time to join the research
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49 team. As a patient research partner²⁵ she was involved in the coding, data analysis and data
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51 interpretation to guarantee the patient perspective. To protect the anonymity of the
52
53 participants all quotes are presented in the “she”-form. Quotes of professional researchers are
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55 indicated by ‘R’ and those of patient research partners (in short: ‘partners’) by ‘P’.
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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 further²⁹ 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

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3 feeling of relief and a belief that patients' views and opinions would be listened to and
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5 incorporated into the deliberations".³⁰ Also the organizers were excited and called the patient
6
7 involvement "a tremendous success".³¹
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10 Between 2002 and 2012 a total of 57 partners with different rheumatic diseases have
11
12 participated as full delegates with equal voting rights.³² Their role and contributions have
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14 developed over time. At the first conference (2002) they formed an homogeneous group of
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16 people with RA with little or no experience in scientific research. The level of involvement in
17
18 the conference in general was relatively low, support was not organized and the number of
19
20 sessions patients attended was limited. Contributions centred on participation in the workshop
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22 discussions about the severity of fatigue and the definition of low disease activity, although
23
24 there was a keynote speech at the opening ceremony.³⁰ In contrast, by OMERACT 11 (2012)
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26 the partners were a heterogeneous group with different rheumatic conditions and different
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28 levels of experience, competences and cultural background. They received a pre-conference
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30 information pack and were actively supported by a pre-conference dinner, a glossary, training
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32 sessions and a buddy system. They carried out a variety of tasks similar to professionals such
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34 as giving plenary presentations, co-chairing breakout sessions, reporting back from breakout
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36 sessions and preparing consensus statements. Several partners became co-authors of peer-
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38 reviewed publications.
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45 **Patient contributions to OMERACT meetings and outcome research**

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47 Interviewees reported a variety of contributions made by partners during the conference
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49 where they are an integral part of the deliberative and consensus-building process.¹⁸ These
50
51 examples are presented below and compared with the document analyses when appropriate.
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53 Because research in the domain of fatigue has been reported as the most illustrative example,
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55 the contributions in this area will be described in more detail. Using the methodology
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3 described above we identified 5 main categories from the comments made during the
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5 interviews with OMERACT participants (Table 2): Contributions to the research agenda; The
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7 development of core sets; The development of patient reported outcomes (PRO's); The
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9 culture of OMERACT; Consequences outside OMERACT. Finally we will highlight some of
10
11 the challenges that emerged from the interviews.
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16 ***Contributions to the research agenda*** - From the very beginning partners had a significant
17
18 influence on the research agenda in the field of rheumatology by participating in OMERACT
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20 workshops and small group discussions. They identified new outcome domains that are
21
22 relevant from their perspective.³³ The first Patient Perspective Workshop, attended by 11
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24 patient participants and 41 professionals, focused on the development of “valid outcome
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26 instruments that incorporate the perspective of the patient and to prepare the evidence and
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28 arguments for their inclusion in the (RA) core set”²⁴. The preconference paper pointed out the
29
30 methodological and political challenges: How to elicit and incorporate preferences of patients
31
32 in RCT's? ³² The workshop had been specifically arranged to support the partner
33
34 contributions including a pre- and post-workshop meeting. The workshop identified
35
36 subjective experiences of RA, not encompassed in the RA core set but important
37
38 consequences of the disease: a sense of well being, fatigue, and disturbed sleep.²⁴
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43 After the first conference attended by partners, it became apparent that perspectives of
44
45 professionals and patients differ and more research was needed to articulate patients
46
47 priorities.³⁴⁻³⁷ Partners emphasized the need for an holistic approach to people with arthritis.³⁰
48
49 The acknowledgement of the discordance of perspectives initiated new studies looking into
50
51 the preferences, opinions and experiences of people with rheumatic diseases ^{22 38 39} and
52
53 developing patient-derived core sets.⁴⁰ This made participants more aware of the emerging
54
55 patient perspective: “the whole realm of things we haven't looked at” [RA]. New topics
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3 emerged: remission, pain, flares and foot problems. One interviewee clearly stated that
4
5 partners “inspired me for new projects to study the variety in new productivity outcome
6
7 measures” [RK].
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10
11 ***Case-study of fatigue*** - Since 2002 when partners identified new topics for research, studies
12
13 have been initiated with firm involvement of partners in the field of sleep disturbances, flares
14
15 and well being. The most progress has been made in fatigue, and the emergence of fatigue as
16
17 a relevant outcome measure in RA provides an illustrative case history. When asked for the
18
19 greatest benefit of including partners in OMERACT conferences interviewees unanimously
20
21 confirmed that the topic of fatigue would not have been on the research agenda without
22
23 partners expressing their concerns about fatigue as an often neglected symptom of their
24
25 disease and without the listening of receptive professionals. One of the partners attending
26
27 OMERACT 6 recalled:
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33
34 “I can’t remember who brought up the subject, but someone mentioned fatigue. And that
35
36 was the occasion when one of the other delegates said ‘well, everybody gets tired’. One
37
38 patient shot to her feet and said ‘no, it’s not, it’s not like anything you’ve ever
39
40 experienced; it’s not tiredness; it’s a complete wipe-out’.” [PM]
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45 Early descriptions of fatigue at OMERACT 6 and 7 led to substantial qualitative and
46
47 quantitative research. The first studies investigated the prevalence and severity of fatigue in
48
49 RA and how patients describe their fatigue.⁴¹⁻⁴⁴ The next step comprised a systematic review
50
51 of measurement instruments for fatigue⁴⁵ that explored the rigor of existing measurement
52
53 tools and the need to develop patient-derived instruments that are trustworthy, capturing
54
55 concepts and language of patients. Furthermore a standardized visual analogue scale,
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3 opportunities for electronic gathering of data and exploring mechanisms of fatigue that could
4
5 guide researchers in the development of effective interventions, were added to the research
6
7 agenda. New data, presented at OMERACT 8 (2006) showed that fatigue is not a
8
9 consequence of RA, but an independent variable that adds new information to the existing RA
10
11 core set.^{46 47} This new perception resulted in the acceptance of fatigue as an important
12
13 outcome for clinical trials.^{48 49} Fatigue was subsequently added to the RA core set as a
14
15 recommended outcome.⁵⁰

16
17
18 More powerful instruments for measuring fatigue in RA have since been devised and
19
20 validated, starting from the perspective of the patients.^{51 52} Outside OMERACT researchers
21
22 initiated similar studies, focusing on the communication between patients and health
23
24 professionals in the consultation room.⁵³

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29 The thematic document analysis provided additional evidence for the statement that without
30
31 patients raising their voice at OMERACT 6 fatigue would not have been high on the research
32
33 agenda. The issue of fatigue was not new for rheumatologists.⁵⁴⁻⁵⁶ Fatigue was a symptom
34
35 regularly reported during clinical consultations, but not incorporated in guidelines for
36
37 monitoring and managing. Fatigue in ankylosing spondylitis was identified by physicians and
38
39 incorporated in a disease status questionnaire.⁵⁷ And during OMERACT 3 (1996) delegates
40
41 carried out a ranking exercise trying to prioritize psychosocial measures in musculoskeletal
42
43 diseases. The discussion groups identified outcomes such as pain, depression, anxiety and
44
45 fatigue as major concerns.⁵⁸ For fatigue eight examples of measurement instruments were
46
47 given.⁵⁹ However, after this workshop, nothing happened for six years, until patients raised
48
49 the urgency of fatigue as a serious symptom.
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3 Retrospectively, professionals admitted they had a blind spot for fatigue in RA and only
4
5 hearing from partners at OMERACT made them change their perception of fatigue as an
6
7 important outcome:
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11 “Because when I was working in oncology before, during university training, of course
12
13 we saw that the patients were lying in bed all day and we knew they were exhausted, call
14
15 that fatigue. But patients with RA, we were ignorant.” [RC]
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21 Another physician, involved in OMERACT from the start:
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25 “We were first discussing on fatigue and to be honest: I never ever had before heard of
26
27 fatigue being a problem in rheumatology. So it got into my mind and then I got thinking
28
29 about it and then, when I was back, I asked patients if they felt fatigue and I got nearly a
30
31 100% positive response. So it was like a coming out, you know. I listened to the patients
32
33 before but bringing it to a specific topic, that was really what I learned at OMERACT.”
34
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36 [RA]
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41 [Insert Table 2]
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45 ***The development of core outcome measurement sets and patient reported outcomes –***
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47 During the first two conferences including patients the focus of partners’ contributions was on
48
49 agenda-setting and identifying relevant outcomes for clinical trials. Then, partners became
50
51 gradually involved on different levels in other OMERACT activities, varying from being
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53 consulted (e.g. in a Delphi process) to full collaboration (as partner and as co-author). They
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55 contributed by identifying domains that are relevant for disease-specific core sets for psoriatic
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3 arthritis,⁶⁰ fibromyalgia,^{61 62} gout⁶³ and vasculitis.⁶⁴ Furthermore they contributed to the
4
5 development of core outcome measurement sets for methodological or clinical concepts such
6
7 as MCID and remission.⁶⁵ Partners have also played a role in the assessment of the feasibility
8
9 of instruments and core sets, one of the three key components of the OMERACT Filter.⁶⁶
10

11
12 Partners have been helpful in the development of PRO measurement instruments in the
13
14 field of work productivity, monitoring adverse events,⁶⁷ flares⁶⁸ and psychosocial
15
16 interventions such as self-management programs⁶⁹. At the 2010 conference, during the
17
18 plenary session on flares in RA, one of the partners gave a personal testimony about the
19
20 devastating impact of the unpredictable nature of RA. A professional in the audience was
21
22 surprised and reported: “It demonstrates that the disease activity fluctuates more than we can
23
24 see in our data: Our instruments are more flat, and by the limited frequency of measuring we
25
26 filter fluctuations out”. [RI]
27
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31
32 Regulators require strong evidence for the effectiveness of new medicines by demonstrating
33
34 accurately that they reduce structural progression as well as patient important outcomes. By
35
36 doing both of these, developing standards for high quality imaging techniques and exploring
37
38 new PRO’s and translating them into valid and feasible measures, OMERACT has been
39
40 extremely advantageous for the negotiations with regulators about the registration and
41
42 relatively generous reimbursement of new biologic agents:
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48 “I think, to be really honest, the patient involvement process in OMERACT and the
49
50 changes in outcome measurements and the use of them in the drug tests has made a
51
52 real difference for so many patients.” [RB]
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3 *The culture of OMERACT*– In spite of the initial unanimous vote to invite patients, some
4
5 researchers were concerned about changing the layout of the conference:
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10 “My original expectation of a limited contribution was based on fear that patients were
11
12 not able to transcend their personal experience and to generalize ... new stakeholders
13
14 often don’t have knowledge about clinometric.” [RE]
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16

17
18 In retrospect researchers explained that they deferred to the proposal in order to reflect a core
19
20 principle of OMERACT of not immediately rejecting new ideas: “To respect and listen rather
21
22 than just react”. [RA]
23
24

25 Looking back, the number of professional participants who were in favour of partners
26
27 at the conference slowly increased: “I was impressed by the very good working flow”. [RC]
28
29 Participants confirmed that the presence of partners has changed their way of thinking and
30
31 talking. “They made my blind spot visible” [RK] and another professional reported:
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35

36 “Now what we have found is, and I changed my view, [be]cause it wasn’t only from
37
38 OMERACT. As I got to know more and more patients, I realized, this sounds stupid
39
40 because it’s so obvious but it wasn’t obvious to me, that a patient isn’t their disease. A
41
42 patient is a person who happens to have a disease. What a big difference. Because if
43
44 you’re a person that happens to have a disease, then for example you might have
45
46 incredible skills in an area that might be very useful to move a clinical trial forward.
47
48 So once I came to that realization then patient involvement becomes an absolutely
49
50 obvious and integral part of moving forward.” [RA]
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3 Partners improved communication and brought dynamics to the dialogue because they are
4 motivated and constructive, without a personal agenda. At a conference such as OMERACT,
5 where the discussion about methodology may become extremely technical, partners reminded
6 participants of the common goal of the conference by providing a human face of a person
7 living with the condition day by day. Their presence made participants more explicit about the
8 objectives of sessions and more explanatory about terms and concepts under discussion.
9
10 Together with a reduced use of jargon this ‘forced’ simplification resulted in fewer
11 misunderstandings for everyone.
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21 For some professionals the presence of partners complicated the communication.
22 Some believed that partners slow down the process because they are not familiar with
23 technical issues. Others felt disinclined to say what they wanted to out of respect to partners
24 or hesitated to criticize them. One researcher felt embarrassed in the presence of partners and
25 put her own expertise aside to keep things simple: “Patients didn’t sometimes understand the
26 objective of the research, which hindered us”. [RK] One of the partners admitted that “it is a
27 thin line between providing input and causing irritation”. [PN]
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38 An analysis of the responses of patients attending OMERACT for the first time showed that
39 new partners experienced a significant learning curve and a variety of personal benefits.⁷⁰
40 Results from this study suggest that in fact all participants learned from the contact with other
41 stakeholders. During this process participants gained trust, respect and understanding,
42 reflecting the emergence of relational empowerment.
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52 “Patients were a kind of sparring partner when I entered a relatively new area. That
53 was fun and did clarify a lot”. [RK]
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3 Relational empowerment in the context of health research can be understood as a process in
4
5 which traditional doctor-patient relationships transform into equal partnerships enabling
6
7 mutual learning processes.⁷¹ All participants become stronger by sharing knowledge and
8
9 responsibilities, and educating and helping each other.
10

11
12 The reported benefits were easiest to identify at the beginning when the level of
13
14 involvement was still low. They became more diffuse when partners were structurally
15
16 involved as full and equal collaborators. One interviewee mentioned “a reality check” as an
17
18 important benefit of partners attending the conference. For professionals it offered the
19
20 opportunity to check the relevance of the scope of their research: Are we doing the right
21
22 things according to patients and are we using the right tools and methods? It is a belief of
23
24 professionals that this kind of feedback is important to legitimize their research and, together
25
26 with the belief of partners that without this research no innovations will take place, it
27
28 strengthened the mutual empowerment of both.
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34 ***Consequences outside OMERACT*** – The lessons learnt at OMERACT were noticed by the
35
36 outside world. Partners returning home after the conference have continued introducing
37
38 patient participation in local and national research projects or established networks of patient
39
40 research partners.⁷²⁻⁷⁵ Some delegates published a working framework for incorporating the
41
42 patient perspective in outcome research.⁷⁶ With the input from several OMERACT
43
44 participants the European League Against Rheumatism (EULAR) developed
45
46 recommendations for the inclusion of patient representatives in scientific projects.²⁵
47
48 Following these recommendations a new patient reported quality of life instrument for RA
49
50 was developed and validated.^{40 77} Based on the experiences of OMERACT the organizing
51
52 committee of the 6th International Shared Decision Making conference decided in 2011 to
53
54 invite patient participants.⁷⁸ In the same year OMERACT delegates, partners as well as
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3 professionals, participated in the 2nd Core Outcome Measures in Effectiveness Trials
4 (COMET) conference, demonstrating how the OMERACT methodology can be utilized in
5
6
7 other disease areas.¹²
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11 ***Remaining challenges emerging from the interviews*** – The role and contribution of patient
12 participants have changed over time and procedures for patient selection and support have
13
14 been developed in order to identify patient participants who are able to make a difference.
15
16 There is still a debate going on whether patients should be selected through strict criteria such
17
18 as education, communication skills, attitude and familiarity with scientific research. Some
19
20 argue that an expert meeting like OMERACT needs expert patients who have extended
21
22 knowledge about methodologies of outcome research, and are able to provide a kind of
23
24 aggregated patient input. At OMERACT this group represents a minority of delegates, who
25
26 are reluctant to allocate the same rights and power to partners as to the professionals. The vast
27
28 majority believes that many patients are able to contribute to an OMERACT conference and
29
30 emphasizes that a heterogeneous group of partners in age, gender, condition, experience and
31
32 cultural background are advantageous for the conference. They intend to develop full
33
34 representative participation in all phases of research by including partners in working group
35
36 activities between conferences. Finally, some participants point out the potential risks of
37
38 partners who become too experienced. They appreciate the naive input as a patient, with a
39
40 minimum of preparation and reflection. They assume that as soon as you start thinking about
41
42 your contribution, you lose the unique, individual perspective and become a patient-expert
43
44 who aligns too easily with professionals.
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52 Professionals shared the opinion that partners need training, although they reported
53
54 different ideas about the content and aims of such training. Experienced partners as well as
55
56 novice researchers felt that any new participant has to learn the OMERACT objectives,
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3 culture and procedures first, before they can become fully productive, mostly at the second or
4
5 third conference. This accords with the expectations of partners who attended OMERACT for
6
7 the first time.
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10 11 12 **Overview of findings**

13
14 These results show that a decade of patient involvement has been successful and had a
15
16 significant impact on various aspects of outcome research. Perspectives of patients are
17
18 different from those of health professionals. Broad consensus exists that partners at
19
20 OMERACT have played a vital role in identifying domains relevant from the perspective of
21
22 patients and in developing new PROs such as fatigue, sleep quality, flares and work
23
24 productivity. Especially in the area of fatigue we have shown that patient involvement on
25
26 different levels and in different phases improves the quality of outcome research. By
27
28 combining evidence-based knowledge of researchers and the experiential knowledge of
29
30 patients, a synthesis of both kinds of knowledge has been achieved and documented. The
31
32 benefits are assessment tools that accurately measure what really matters to patients, are
33
34 formulated in understandable language and are user-friendly. Other benefits go beyond
35
36 improving clinical outcome research and include improved communication, mutual
37
38 empowerment, changed attitudes and substantial consequences outside OMERACT.
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45 **Discussion**

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47 We set out to describe and evaluate the contribution of patients as partners in rheumatology
48
49 outcome research, reviewing their impact on the research agenda and the culture and process
50
51 of the OMERACT conference. The document analysis provided the recorded facts while the
52
53 interviews allowed an exploration of intentions, attitudes and perceived benefits or harms of
54
55 patient participation that complements the document analysis. Since validated methodologies
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3 for demonstrating impact of collaboration with patients in the context of research are lacking,
4
5 a responsive interview methodology seemed to be a good approach.
6

7 Both strengths and limitations of this study relate to the personal experience of the first
8 and last authors as participants in the developing process of patient partner participation.
9
10 Having witnessed the OMERACT process, actors and concerns of both the patient community
11 and the research community was advantageous during the development of interview
12 protocols, recruitment and selection of respondents and data analysis. For instance, the
13 knowledge of the opinions of other participants made it possible to achieve maximum
14 variation. Also, the active involvement in the support and training of partners created an
15 adequate awareness of the relevant items to include in the study. The drawbacks of this
16 engagement are the risks of subjectivity, blind spots and over- or under-identification with
17 particular stakeholders. These risks have been addressed by applying strict quality measures
18 for scientific rigor in qualitative, evaluation research.
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31 The composition of the research team purposely included two external experts in
32 qualitative research and a patient research partner, who were actively involved in the coding
33 of interview transcripts and distilling relevant categories for impact, reduced the risk of
34 subjectivity. Bias was avoided by the check-coding procedure in the analysis of the transcripts
35 as at least two researchers independently coded each transcript, after which the whole team
36 discussed the codes until consensus was reached. Saturation was also part of the discussion in
37 the whole team. The inclusion of various stakeholder perspectives prevented one-sidedness.
38
39 No signals were identified to suggest that interviewees have simply given desirable answers,
40 or just been friendly to the interviewer. Some interviewees have been rather critical, reporting
41 several barriers for structural involvement of patients in research, but have always added
42 constructive suggestions for improvement. Peer debriefing by an independent colleague (TA)
43 further helped to prevent bias.
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3 Other limitations relate to the difficulties of demonstrating the ‘impact’ of patient
4 involvement.⁹ In OMERACT there is a strong belief that patient participation works, a belief
5 that is nourished by the world-wide transition towards more patient-oriented health care and
6 health research.⁷⁹⁻⁸² The assumption however that long term involvement of patients as equal
7 partners guarantees sustainable inclusion of the patient perspective in outcome research
8 complicates a thorough evaluation and makes it difficult to distinguish between expected,
9 perceived and actual contributions. Many participants, not only partners, but also young
10 researchers and other new-comers, are not able to identify their own contribution and may not
11 see how their input is reflected in the final outcomes. Partners reported almost unanimously
12 not being able to confirm substantial contributions during their participation but they believed
13 they did. More experienced participants, mostly professionals, were less reluctant in reporting
14 illustrative examples of patient contributions.
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29 In a dialogue and consensus-based conference such as OMERACT many (f)actors
30 contribute to the final outcomes. A linear causal relation between patient involvement and
31 impact is therefore hard to establish; the processes of involvement are rather influenced by
32 and influencing many (f)actors in a mutually interactive way. We found that when the level of
33 involvement of partners increased from consultation to collaboration, it became harder to
34 solely attribute individual or group contributions to the final outcomes. Because neither
35 partners nor professionals act as a representative of any group or constituency it remains
36 difficult to determine the influence of particular groups or individuals. Participation proved to
37 be a dynamic process, especially when tasks were equally performed by patients and
38 researchers, and when the dialogue between both took not only place during the official
39 sessions, but also in the corridors of the conference. It should be noted that striving toward
40 equality is a normative ideal,⁸³ and fighting inequalities between patients and professionals is
41 and remains an ongoing concern. “Equality” may be seen in two ways: as the formal position
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3 of patients at the conference (as full delegates they had the same voting rights as professional
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5 delegates, they received the same pre-conference materials and had access to all conference
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7 sessions like all other participants) and as equality of partnership or collaboration in terms of
8
9 influence on the decision making process. Given the power inequalities between patients and
10
11 professionals the latter is the greater challenge, but nevertheless some of the documents and
12
13 interviews suggest it has been achieved to some extent. Although patients remain only
14
15 indirectly represented in the executive committee (the highest decision making body), our
16
17 data support the conclusion that a small number of experienced patients achieved an equal
18
19 relationship with researchers in their area of interest. They obtained the competences that
20
21 enabled them to perform all kinds of tasks at the conference similar to professionals, and
22
23 provided input that justified co-authorship of peer-reviewed articles. We did not obtain in-
24
25 depth information about the question to what extent power inequalities between patients and
26
27 researchers still persist but we know from the feedback of all respondents, including some
28
29 fellows and researchers that attended OMERACT for the first time, that some did not feel
30
31 treated equally. To what extent this experience was caused by their status of being a patient or
32
33 by the status of a new participant is still unknown.

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38 A last obstacle for demonstrating the influence of patient participation is the invisibility
39
40 of experiential knowledge, often hidden in anecdotal stories. It has an impact that is rarely
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42 claimed by patients nor perceived by professionals. Personal comments are normally not
43
44 reported because they are not seen as a valuable and valid source of knowledge⁸⁴ and yet
45
46 clear documentation of meetings is required to ensure that patients' contributions become
47
48 visible.⁸⁵ Professionals focus on synthesizing data and may not notice that the dialogue with
49
50 patients works like a reality check, generates new ideas or changes their beliefs, behaviour or
51
52 perception. When partners appear to simply agree with the results presented at OMERACT it
53
54 might look as if they do not have any contribution to make, but in fact they confirm the value
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3 of the work under discussion and provide face validity to the process. It is for this reason that
4
5 most professionals appreciate the feedback and input from partners, although not all are aware
6
7 of this reason. Realizing the importance of such a reality check is beneficial for the
8
9 management of realistic expectations: do not expect innovative ideas, brilliant suggestions
10
11 and new concepts when inviting partners to join research. Their contributions are often more
12
13 subtle and need the attention of a modest and committed researcher to be noticed.
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16 Despite these limitations, we believe that the results presented in this study are relevant
17
18 and valid. It is undeniable that there is a growing belief that patient involvement has been
19
20 successful and brought a unique added value to the conference. Even those who were
21
22 originally among the most skeptical participants now report that they have changed their
23
24 perception about the expected contribution of patient research partners. This study is
25
26 conducted within the context of a scientific research conference in the field of rheumatology,
27
28 a long term somatic condition. Our ability to generalize the findings is therefore limited and
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30 extrapolation to other research contexts or to other conditions should be done with care.
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References

1. Keizer B, Bless R. Pilot study on the position of health consumer and patients' organisations in seven EU countries. Den Haag: ZonMW, 2010:54.
2. Involve. People & participation; How to put citizens at the heart of decision-making. London: Beacon Press, 2005.
3. Staley K. Summary Exploring Impact: Public involvement in NHS, public health and social care research. Eastleigh.: INVOLVE, 2009.
4. Wright MT, Roche B, von Unger H, et al. A call for an international collaboration on participatory research for health. *Health Promot Int* 2010;25(1):115-22.
5. Abma TA. Patients as partners in a health research agenda setting: the feasibility of a participatory methodology. *Eval Health Prof* 2006;29(4):424-39.
6. Abma TA, Nierse C, Widdershoven G. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qualitative Health Research* 2009;19(3):401-15.
7. Abma TA, Broerse JE. Patient participation as dialogue: setting research agendas. *Health Expect* 2010;13(2):160-73.
8. Caron-Flinterman JF, Broerse JE, Bunders JF. The experiential knowledge of patients: a new resource for biomedical research? *Soc Sci Med* 2005;60(11):2575-84.
9. Staniszewska S, Haywood KL, Brett J, et al. Patient and public involvement in patient-reported outcome measures: evolution not revolution. *Patient* 2012;5(2):79-87.
10. Speight J, Barendse SM. FDA guidance on patient reported outcomes. *BMJ* 2010;340:c2921.
11. Rowen D, Carlton J, Brazier JE, et al. FDA on PROMs. Two important points. *BMJ* 2010;341:c5454.
12. Williamson PR, Altman DG, Blazeby JM, et al. The COMET (Core Outcome Measures in Effectiveness Trials) Initiative. *Trials* 2011;12 (Suppl 1)(A70).
13. Clarke M. Standardising outcomes for clinical trials and systematic reviews. *Trials* 2007;8:39.
14. Sinha IP, Smyth RL, Williamson PR. Using the Delphi technique to determine which outcomes to measure in clinical trials: Recommendations for the future based on a systematic review of existing studies. *PLoS Med* 2011;8(1).
15. Williamson P, Clarke M. The COMET (Core Outcome Measures in Effectiveness Trials) Initiative: Its Role in Improving Cochrane Reviews. *Cochrane Database Syst Rev* 2012;5:ED000041.
16. Saag KG. OMERACT 6 brings new perspectives to rheumatology measurement research. *J Rheumatol* 2003;30(4):639-41.
17. Heller JE, Shadick NA. Outcomes in rheumatoid arthritis: incorporating the patient perspective. *Current Opinion in Rheumatology* 2007;19(2):101-05.
18. OMERACT. Guiding principles for patient research partner participation in OMERACT. Accessed: October 18; 2012: Available from: <http://www.omeract.org/pdf/2011-03-27%20Agreed%20Patient%20Participation%20Policy.pdf>.
19. Guba EG, Lincoln YS. *Effective evaluation. Improving the usefulness of evaluation results through responsive and naturalistic approaches*. San Francisco: Jossey-Bass Publishers, 1988.
20. Abma TA, Widdershoven GAM. Evaluation as a relationally responsible practice. In: Denzin N, Lincoln Y, editors. *Handbook for Qualitative Inquiry*. Los Angeles: SAGE Publications Ltd., 2011:669-80.
21. Lincoln Y, Guba EG. *Naturalistic inquiry*. Newbury Park: SAGE, 1985.

22. Hewlett S, Carr M, Ryan S, et al. Outcomes generated by patients with rheumatoid arthritis: how important are they? *Musculoskeletal Care* 2005;3(3):131-42.
23. Hewlett S, Cockshott Z, Byron M, et al. Patients' perceptions of fatigue in rheumatoid arthritis: overwhelming, uncontrollable, ignored. *Arthritis and rheumatism* 2005;53(5):697-702.
24. Kirwan J, Heiberg T, Hewlett S, et al. Outcomes from the Patient Perspective Workshop at OMERACT 6. *Journal of Rheumatology* 2003;30(4):868-72.
25. de Wit MPT, Berlo SE, Aanerud GJ, et al. European League Against Rheumatism recommendations for the inclusion of patient representatives in scientific projects. *Annals of the Rheumatic Diseases* 2011;70(5):722-6.
26. Tugwell P, Boers M. OMERACT conference on outcome measures in rheumatoid arthritis clinical trials: introduction. *J Rheumatol* 1993;20(3):528-30.
27. Brooks PM, Boers M, Tugwell P. OMERACT III: the "ACT" revisited. *Jrn of Rheum* 1997;24(4):764-5.
28. Wells G, Anderson J, Beaton D, et al. Minimal clinically important difference module: summary, recommendations, and research agenda. *J Rheumatol* 2001;28(2):452-4.
29. Kirwan JR, Carr A, Gluck O, et al. Proposal for inclusion of a 'Patient's perceptions' module in OMERACT VI [unpublished], 2000.
30. Quest E, Aanerud GJ, Kaarud S, et al. Patients' perspective. *J Rheumatol* 2003;30(4):884-5.
31. Brooks PM, Tugwell P, Strand C, et al. OMERACT 6: International consensus conference on outcome measures in rheumatology: Introduction. *Jrn of Rheum* 2003;30(4):866-7.
32. Kirwan J. The patient perspective workshop at OMERACT 6: Rationale and methodology [unpublished preconference paper]. 2002.
33. Tugwell P, Boers M, Brooks P, et al. OMERACT: an international initiative to improve outcome measurement in rheumatology. *Trials* 2007;8:38.
34. Hewlett SA. Patients and clinicians have different perspectives on outcomes in arthritis. *J Rheumatol* 2003;30(4):877-9.
35. Leeb BF, Sautner J, Leeb BA, et al. Lack of agreement between patients' and physicians' perspectives of rheumatoid arthritis disease activity changes. *Scand J Rheumatol* 2006;35(6):441-6.
36. Kvien TK, Heiberg T. Patient perspective in outcome assessments--perceptions or something more? *J Rheumatol* 2003;30(4):873-6.
37. Carr A, Hewlett S, Hughes R, et al. Rheumatology outcomes: the patient's perspective. *Journal of Rheumatology* 2003;30(4):880-3.
38. Ahlmen M, Nordenskiold U, Archenholtz B, et al. Rheumatology outcomes: the patient's perspective. A multicentre focus group interview study of Swedish rheumatoid arthritis patients. *Rheumatology (Oxford)* 2005;44(1):105-10.
39. Rupp I, Boshuizen HC, Roorda LD, et al. Course of patient-reported health outcomes in rheumatoid arthritis: comparison of longitudinal and cross-sectional approaches. *J Rheumatol* 2006;33(2):228-33.
40. Gossec L, Paternotte S, Aanerud GJ, et al. Finalisation and validation of the rheumatoid arthritis impact of disease score, a patient-derived composite measure of impact of rheumatoid arthritis: a EULAR initiative. *Annals of the Rheumatic Diseases* 2011;70(6):935-42.
41. Hewlett S, Cockshott Z, Byron M, et al. Patients' perceptions of fatigue in rheumatoid arthritis: overwhelming, uncontrollable, ignored. *Arthritis Rheum* 2005;53(5):697-702.
42. Rupp I, Boshuizen HC, Jacobi CE, et al. Impact of fatigue on health-related quality of life in rheumatoid arthritis. *Arthritis Rheum* 2004;51(4):578-85.

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43. Repping-Wuts H, Uitterhoeve R, van Riel P, et al. Fatigue as experienced by patients with rheumatoid arthritis (RA): A qualitative study. *Intl J Nursing Studies* 2008;8(45(7)):995-102.
44. Nikolaus S, Bode C, Taal E, et al. Four different patterns of fatigue in rheumatoid arthritis patients: results of a Q-sort study. *Rheumatology* 2010;49(11):2191-9.
45. Hewlett S, Hehir M, Kirwan JR. Measuring fatigue in rheumatoid arthritis: a systematic review of scales in use. *Arthritis & Rheumatism* 2007;57(3):429-39.
46. Kirwan JR, Hewlett S. Patient perspective: reasons and methods for measuring fatigue in rheumatoid arthritis. *J Rheumatol* 2007;34(5):1171-3.
47. Hewlett S, Chalder T, Choy E, et al. Fatigue in rheumatoid arthritis: time for a conceptual model. *Rheumatology (Oxford)* 2010.
48. Kirwan JR, Minnock P, Adebajo A, et al. Patient perspective: fatigue as a recommended patient centered outcome measure in rheumatoid arthritis. *J Rheumatol* 2007;34(5):1174-7.
49. Pincus T, Sokka T. Complexities in the quantitative assessment of patients with rheumatic diseases in clinical trials and clinical care. *Clin Exp Rheumatol* 2005;23(5 Suppl 39):S1-9.
50. Kirwan JR, Minnock P, Adebajo A, et al. Patient perspective: fatigue as a recommended patient centered outcome measure in rheumatoid arthritis. *Journal of Rheumatology* 2007;34(5):1174-7.
51. Nicklin J, Cramp F, Kirwan J, et al. Measuring fatigue in rheumatoid arthritis: A cross-sectional study to evaluate the Bristol Rheumatoid Arthritis Fatigue Multi-Dimensional questionnaire, visual analog scales, and numerical rating scales. *Arthritis Care Res (Hoboken)* 2010;62(11):1559-68.
52. Nikolaus S, van de Laar MA. Measuring fatigue in rheumatoid arthritis. *Nat Rev Rheumatol* 2011;7(10):562-4.
53. Repping-Wuts H. Fatigue in patients with rheumatoid arthritis (thesis). Nijmegen University, 2009.
54. Wolfe F, Hawley DJ, Wilson K. The prevalence and meaning of fatigue in rheumatic disease. *The Journal of rheumatology* 1996;23(8):1407-17.
55. Tack BB. Fatigue in rheumatoid arthritis. Conditions, strategies, and consequences. *Arthritis Care & Research* 1990;3(2):65-70.
56. Crosby LJ. Factors which contribute to fatigue associated with rheumatoid arthritis. *J Adv Nurs* 1991;16(8):974-81.
57. Garrett S, Jenkinson T, Kennedy LG, et al. A new approach to defining disease status in ankylosing spondylitis: the Bath Ankylosing Spondylitis Disease Activity Index. *The Journal of rheumatology* 1994;21(12):2286-91.
58. Newman SP. Psychosocial measures in musculoskeletal trials. *J Rheumatol* 1997;24(5):979-84.
59. Brooks P, McFarlane AC, Newman S, et al. Psychosocial measures. *J Rheumatol* 1997;24(5):1008-11.
60. Gladman DD, Mease PJ, Strand V, et al. Consensus on a core set of domains for psoriatic arthritis. *The Journal of rheumatology* 2007;34(5):1167-70.
61. Mease P, Arnold LM, Choy EH, et al. Fibromyalgia syndrome module at OMERACT 9: domain construct. *The Journal of rheumatology* 2009;36(10):2318-29.
62. Choy EH, Arnold LM, Clauw DJ, et al. Content and criterion validity of the preliminary core dataset for clinical trials in fibromyalgia syndrome. *The Journal of rheumatology* 2009;36(10):2330-4.

63. Taylor WJ, Schumacher HR, Jr., Baraf HS, et al. A modified Delphi exercise to determine the extent of consensus with OMERACT outcome domains for studies of acute and chronic gout. *Annals of the rheumatic diseases* 2008;67(6):888-91.
64. Merkel PA, Aydin SZ, Boers M, et al. The OMERACT core set of outcome measures for use in clinical trials of ANCA-associated vasculitis. *The Journal of rheumatology* 2011;38(7):1480-6.
65. van Tuyl LH, Smolen JS, Wells GA, et al. Patient perspective on remission in rheumatoid arthritis. *The Journal of rheumatology* 2011;38(8):1735-8.
66. Boers M, Brooks P, Strand CV, et al. The OMERACT filter for Outcome Measures in Rheumatology. *The Journal of rheumatology* 1998;25(2):198-9.
67. Lassere MN, Johnson KR, Boers M, et al. Standardized assessment of adverse events in rheumatology clinical trials: summary of the OMERACT 7 drug safety module update. *Journal of Rheumatology* 2005;32(10):2037-41.
68. Bingham CO, 3rd, Alten R, de Wit MP. The importance of patient participation in measuring rheumatoid arthritis flares. *Annals of the rheumatic diseases* 2012.
69. Kristjansson E, Tugwell PS, Wilson AJ, et al. Development of the effective musculoskeletal consumer scale. *The Journal of rheumatology* 2007;34(6):1392-400.
70. de Wit MPT, Koelewijn-van Loon MS, Collins S, et al. How patients contribute a new voice at a scientific conference: their expectations and experiences at the Outcome Measures in Rheumatology Conference 2010. (*forthcoming*) 2012.
71. Abma TA, Nierse CJ, Widdershoven GA. Patients as partners in responsive research: methodological notions for collaborations in mixed research teams. *Qual Health Res* 2009;19(3):401-15.
72. Kjekken I, Ziegler C, Skrolsvik J, et al. How to develop patient-centered research: some perspectives based on surveys among people with rheumatic diseases in Scandinavia. *Physical Therapy* 2010;90(3):450-60.
73. Akerhielm CE. Patient participation in research - A Swedish model. *Ann Rheum Dis* 2009;68(Suppl3):800.
74. Akerhielm CE. Patient research partners - A way to participation and influence in research. *Ann Rheum Dis* 2010;69(Suppl3):753.
75. Hewlett S. Learnings from the Bristol experience - Practicalities of a decade of patient involvement in research on a local level. *Ann Rheum Dis* 2010;69((Suppl3)):29.
76. Hewlett S, De Wit M, Richards P, Quest E, Hughes R, Heiberg T, et al. Patients and professionals as research partners: challenges, practicalities, and benefits. *Arthritis & Rheumatism* 2006;55(4):676-80.
77. Gossec L, Dougados M, Rinceval N, et al. Elaboration of the preliminary Rheumatoid Arthritis Impact of Disease (RAID) score: a EULAR initiative. *Annals of the Rheumatic Diseases* 2009;68(11):1680-5.
78. Stiggelbout AM, Weijden TV, Wit MP, et al. Shared decision making: really putting patients at the centre of healthcare. *BMJ* 2012;344:e256.
79. Beresford P. User involvement, research and health inequalities: developing new directions. *Health Soc Care Community* 2007;15(4):306-12.
80. Ward PR, Thompson J, Barber R, et al. Critical perspectives on 'consumer involvement' in health research: Epistemological dissonance and the know-do gap. *Journal of Sociology* 2009;46(1):63-82.
81. Lindenmeyer A, Hearnshaw H, Sturt J, et al. Assessment of the benefits of user involvement in health research from the Warwick Diabetes Care Research User Group: a qualitative case study. *Health Expectations* 2007;10(3):268-77.
82. Broerse JEW, Elberse JE, Caron-Flinterman JFW, et al. Enhancing a transition towards a needs-oriented health research system. In: Broerse JEW, Bunders JFG, editors.

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- Transitions in Health Systems: Dealing with persistent problems.* Amsterdam: VU University Press, 2010:181-205.
83. Habermas J. *Theorie des kommunikativen Handelns. Bd. 1. Handlungsrationalität und gesellschaftliche Rationalisierung* Zweite Auflage 1982 ed. Frankfurt am Main: Suhrkamp, 1981.
84. Schipper K. Patient participation & knowledge [thesis]. VU University, 2011.
85. Jinks C, Ong BN, O'Neill TJ. The Keele community knee pain forum: action research to engage with stakeholders about the prevention of knee pain and disability. *BMC Musculoskelet Disord* 2009;10:85.

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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	1	5	8
OMERACT conferences attended	2	0	3
	3	1	1
	4	4	4
	≥5	6	0
Interview in relation to	Before	2	1
	During	8	16

} PG to PN

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

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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: <ul style="list-style-type: none"> • Well being • Fatigue • Sleep disturbance • Flares 	Identification of patient relevant domains to include in core sets for clinical trials: <ul style="list-style-type: none"> • Fibromyalgia • Psoriatic Arthritis • Vasculitis • Gout • MRI • MCID 	Acceptable, understandable and feasible outcome measures for <ul style="list-style-type: none"> • Monitoring adverse events • Work productivity • Flares • Psychosocial interventions 	<ul style="list-style-type: none"> • Attitudes • Communication • Perceptions • Motivation • Relational empowerment • Personal benefits 	<ul style="list-style-type: none"> • Local initiatives • Local and national networks of partners • EULAR • COMET • ISDM

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	<ul style="list-style-type: none"> • Remission 			
Reality check				

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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