

Outcomes from the Patient Perspective Workshop at OMERACT 6

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ABSTRACT. The objective of the Patient Perspective Workshop at OMERACT 6 was to address the question of assessing the outcomes of intervention in rheumatoid arthritis (RA) from the perspective of those who experience the disease themselves. This was done by reviewing the current state of research in the area, identifying the requirements for the development of valid instruments, delineating a research agenda that can attain these requirements, and motivating participants to undertake the appropriate research. Through a series of meetings and discussion sessions a research agenda emerged that includes: exploring subjective experiences of RA identified by patients as important but not encompassed within the current “core set” of outcome measures (such as a sense of well being, fatigue, and disturbed sleep); clarifying terminology; and empowering patients to be more effective partners in outcomes research. These were supported by the OMERACT plenary session. Specific actions were required by both patient participants and organizers to ensure the nature of the conference, its focus and method of working were understood, and that the patient participants were sufficiently confident to make their contribution. (*J Rheumatol* 2003;30:868–72)

Key Indexing Terms:

RANDOMIZED CONTROLLED TRIAL
OUTCOME AND PROCESS ASSESSMENT

RHEUMATOID ARTHRITIS
PATIENT PARTICIPATION

The Patient Perspective Workshop at OMERACT 6 addressed the question of assessing the outcomes of intervention in rheumatoid arthritis (RA) from the perspective of those who experience the disease themselves. This approach arose from the discussions about the “minimum clinically important difference” in an outcome measure, raised at

OMERACT 5 in 2000¹. Patient Perspective Workshop participants included 11 patients from 7 countries, 5 organizing group members, and 41 other participants from those attending the OMERACT 6 meeting. The workshop (Table 1) consisted of 3 formal sessions each of 2 hours, working group meetings between and after the formal sessions, and an unscheduled meeting of the patient participants.

This report has been prepared by the organizing group and the facilitators and reporters from the discussion groups in the workshop, and briefly describes the outcome of the various sessions, and sets out the issues and research agenda identified and the decisions endorsed by the final OMERACT plenary session.

Preamble Meeting

In preparation for the main workshop a preamble meeting briefly reviewed the process by which the workshop had been convened. Recent research on the inclusion of the patient perspective in outcome research², together with a preliminary review of the results of a series of focus groups looking at outcome from a patient’s perspective³ were reviewed. Outcome assessment in RA has been highly dependent on the physician assessing the patient. This varies considerably between clinicians, such that judgments of change in response to therapy could be diametrically opposed (Figure 1)⁴. The rheumatology community responded to the need to standardize outcome assessments by inventing the “core set” of outcome measures (Table 2)^{5–7} and adopting the “OMERACT Filter” of truth, discrimination, and feasibility⁸ against which to assess the validity of

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Table 1. Structure of the Patient Perspective Workshop

Session	Objectives	Participants
Preamble meeting	Identify key questions	Patient participants, organizing group, and active researchers in this area
Workshop	Review the current state of research in area Identify the requirements for developing valid instruments Delineate a research agenda which can attain these requirements Motivate participants to undertake the appropriate research	Patient participants, organizing group, and OMERACT participants who registered for the workshop
Post-workshop meeting	Detailed assessment of feedback and formulation of research agenda	Patient participants, organizing group, and active researchers in this area
Patient participants' meeting	To discuss content of patient participants' report	Patient participants
OMERACT plenary session	To present and seek endorsement of final conclusions	All OMERACT participants

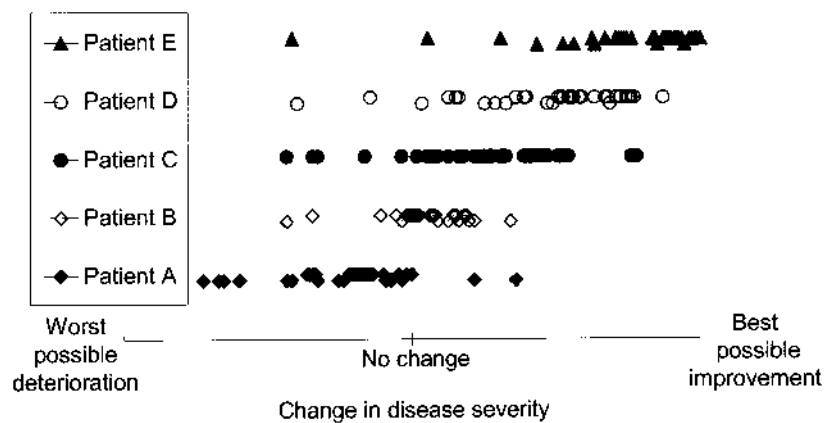


Figure 1. The opinion of 38 rheumatologists on the change in disease status of 5 patients.

specific measurement instruments. Measures were included by a consensus of doctors and methodological scientists working in the field. Subsequently, regulatory authorities and pharmaceutical companies have adopted the core set for assessing new treatments.

More recently there have been attempts to define threshold changes of significance in a combination of variables (Disease Activity Score⁹, American College of Rheumatology Response Criteria¹⁰), and the minimum clinically important difference (MCID) for changes in individual outcomes or combinations of outcome measures¹. These activities have been undertaken primarily by rheumatologists, but the perspective of patients themselves has not been directly included in the process of consensus and decision making. Patients can be involved in outcomes research in a variety of ways (Table 3). In this instance, their personal knowledge of the disease, together with a developing understanding of the nature of the OMERACT process, forms a basis on which to question and possibly expand the current notions of outcome in RA.

The overall aim, therefore, was to develop valid outcome instruments that incorporate the perspective of the patient and to prepare the evidence and arguments for their inclusion in the core set of outcome measures in RA. The objectives of the workshop were to review the current state of research in the area, identify the requirements for the development of valid instruments, delineate a research agenda that can attain these requirements, and motivate participants to undertake the appropriate research. The first session served to identify and prioritize issues and key questions that would be appropriate for the full workshop discussion. Following discussion groups and a report-back session, the questions in Table 4 were selected.

Main Workshop Discussions

In preparation for the workshop discussion groups, members were first reminded of the genesis of the workshop (see above). A patient participant summarized her own experience of the effects of RA and how she evaluated the state of her disease. Evidence that patients and clinicians hold

Table 2. The “core set” of outcome measures currently used in rheumatoid arthritis.

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- Pain
 - Tender joints
 - Swollen joints
 - Patient’s overall opinion
 - Clinician’s overall opinion
 - Function
 - Radiographs
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Table 3. Ways the patient’s perspective can be included in research.

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- Identify research questions
 - Undertake research
 - Identify outcomes
 - Define clinically important changes
 - Build patient perspective into outcomes
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differing views of the importance of various outcome assessments was presented¹¹, and the main questions to be addressed by the workshop discussion groups (Table 4) were introduced. Each question was addressed by 2 groups, which consisted of a chair, a rapporteur, 2 patient participants, and up to 6 other participants. At the feedback session for all participants, each question was addressed in turn, first hearing the comments from the 2 groups, then turning to a general discussion. At the end there was an opportunity for participants to raise any other matters. Points to emerge from the discussions and feedback session were recorded on flip charts or overhead projectors, and taken forward to the post-workshop meeting (the “postamble”).

Postamble Meeting

Organizers, patient participants, discussion group chairs, rapporteurs, and others with a specific research interest in this area gathered the following day to draw together all the issues that emerged at the workshop and to organize them into a coherent presentation for the final OMERACT plenary session. In relation to the aims of the workshop, the review of research in the area concluded that many changes in society and in rheumatology over the last 20 years may require a reevaluation of instruments, their terminology, and the range of symptoms covered; there also was recognition

of the need for a wider review than had so far been conducted. Considering the need to develop valid instruments, the Patient Perspective Workshop agreed on the need to apply the OMERACT Filter⁸, but felt that outcomes identified by patients but currently outside the filter required development. In some areas there is a need to “listen and interpret” before making assumptions about patient views on outcomes. The Patient Perspective Workshop formulated a suggested research agenda covering 3 areas for exploration and development: novel outcomes and approaches to assessment, terminology and current knowledge, and the role of the patient. These are presented more fully in Table 5.

Research Agenda

Novel outcomes and approaches to assessment. A number of subjective experiences of RA are not encompassed within the current core set of outcome measures, but were identified by patients as important consequences of their disease. These include a sense of well being, fatigue, and disturbed sleep, but further work is required to develop and explore this list, and to develop valid measurement instruments. Cross-cultural comparisons will be required and further work to incorporate the notion of the impact of particular symptoms on the lives of individual patients. Further, methods of collecting data, particularly in a more continuous way, should be explored in partnership with patients and might include some feedback from the measurement technique into patient care.

Terminology and current knowledge. It was clear that there are many uses for and interpretations of words and phrases such as “patient-centered” and even “disability.” Opportunities for greater understanding of outcome assessment would follow from a combined patient and professional approach to the definition of terms and a clarification of their usage. Direct qualitative research will be required to clarify specific terminologies to be used as assessment tools, such as a clear description of “fatigue” as an outcome of RA, rather than a description of general tiredness. A full literature review could then use agreed-on terminology for describing what is currently established in this area.

The role of the patient. For patients to contribute fully to the inclusion of their perspective into outcome assessment, they would require sufficient understanding and expertise in the field. How this can be best achieved and how their contri-

Table 4. Issues taken to discussion at the main workshop.

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- Would continuous measurement provide a basis for within-patient comparisons over time, and how can this be achieved efficiently?
 - What mechanisms can we devise to continue patient involvement in the process?
 - Which factors predict variation in patient preferences? How can these be incorporated into measurement?
 - How can we produce a glossary of appropriate terms?
 - How can we establish which outcomes are relevant to patients? Are there some we can already say are known to be important?
 - Are there variations in patient perspective according to age, disease duration, or other items?
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Table 5. OMERACT 6 Patient Perspective Workshop research agenda.

Identify novel outcomes and instruments of relevance	
“Well-being”/“fatigue”/sleep pattern	Standardized patient diaries
Low disease activity state	Use of information technology for repeated measurement
Check in several countries (cultures)	Patient feedback from questionnaires
Weighting for priorities or impact	
Terminology and current knowledge	
Glossary	Literature review
• List of terms to define	• Systematic
• Review existing definitions	• Linked to glossary development
• Professional/patient glossary review group	• Publishable in itself
• Work particularly on fatigue/well being as related to RA	• Leading to Cochrane reviews of appropriate outcomes
	Register of current work
The role of the patient	
Developing patient expertise	Patients as writers/editors
• Specific educational and communication support	• Plain language editing
• Mechanisms of dissemination and representation	• Defining research agenda in patient terms
• OMERACT patient panel	Patient review group for current measures
	• Relevance
	• Linguistics
	• Comprehension

tribution can be effectively disseminated and used to influence the publishing process are all areas of legitimate concern and experimentation.

Plenary Proposals

In common with other OMERACT 6 workshops and modules, a list of proposals was presented to the final plenary session for brief debate and voting. The workshop was careful to offer proposals that OMERACT members themselves could implement, so that endorsement would help to drive forward action in this area of outcomes research. By large majorities (80–95%) the OMERACT participants as a whole agreed:

- research to include the patient perspective in outcome assessment should be carried forward
- patients themselves should increase and facilitate patient involvement in research
- patient participation is an integral part of OMERACT activities
- an OMERACT patient advisory group should be established

Observations from the Organizing Group

Directly involving patients as integral participants in the OMERACT conference presented many challenges. Although not all challenges were fully overcome on this first attempt, the nature of the meeting was fundamentally changed for the better by including the Patient Perspective Workshop. Effort was required by both patient participants and organizers to ensure the nature of the conference and its focus and method of working were understood, and that

patient participants were sufficiently confident to make their contribution. The sight of one patient participant eagerly requesting the microphone at the final plenary session to correct an observation made by one senior research worker in the area showed this had been achieved, at least by the end of the proceedings. Further, patient participants decided to write their own report on their view of the meeting¹².

New ideas were generated from the patient participants, and the OMERACT movement as a whole has clearly recognized the advantage of this. The identification of potential patient participants on this occasion was through the personal contacts of organizing group members and their colleagues, which helped to ensure that people were invited who could make a contribution in unfamiliar circumstances. There remains the potential in the future to include patients from a wider range of backgrounds, in an attempt to obtain representative opinion. However, the success of patient involvement will rely on finding participants who can demonstrate a special interest in moving the science of outcome measurement forward. These may not necessarily be the same group who act as patient advocates for resources in RA treatment, and further thought should be given to the criteria for identifying and selecting OMERACT patient participants.

REFERENCES

1. Wells G, Anderson J, Beaton D, et al. Minimal clinically important difference module: summary, recommendations, and research agenda. *J Rheumatol* 2001;28:452-4.
2. Kvien T, Heiberg T. Patient perspective in outcome assessments — perceptions or something more? *J Rheumatol* 2003;30:873-6.
3. Carr A, Hewlett S, Hughes R, Mitchell H, Ryan S, Carr M, Kirwan

- J. Rheumatology outcomes: the patient's perspective. *J Rheumatol* 2003;30:880-3.
4. Kirwan JR. Minimum clinically important difference: the crock of gold at the end of the rainbow? *J Rheumatol* 2001;28:439-44.
 5. Tugwell P, Boers M, for the OMERACT Committee. Developing consensus on preliminary efficacy endpoints for RA clinical trials. *J Rheumatol* 1993;20:555-6.
 6. Boers M, Tugwell P, Felson DT, et al. World Health Organization and International League of Associations for Rheumatology core endpoints for symptom modifying antirheumatic drugs in rheumatoid arthritis clinical trials. *J Rheumatol* 1994;21 Suppl 41:86-9.
 7. Felson DT, Anderson JJ, Boers M, et al. The American College of Rheumatology preliminary core set of disease activity measures for rheumatoid arthritis clinical trials. The Committee on Outcome Measures in Rheumatoid Arthritis Clinical Trials. *Arthritis Rheum* 1993;36:729-40.
 8. Boers M, Brooks P, Strand CV, Tugwell P. The OMERACT filter for outcome measures in rheumatology. *J Rheumatol* 1998;25:198-9.
 9. van der Heijde DM, van't Hof MA, van Riel PL, et al. Judging disease activity in clinical practice in rheumatoid arthritis: first step in the development of a disease activity score. *Ann Rheum Dis* 1990;49:916-20.
 10. Felson DT, Anderson JJ, Boers M, et al. American College of Rheumatology preliminary definition of improvement in rheumatoid arthritis. *Arthritis Rheum* 1995;38:727-35
 11. Hewlett S. Patients and clinicians have different perspectives on outcomes in arthritis. *J Rheumatol* 2003;30:877-9.
 12. Quest E. Patients' perspective. *J Rheumatol* 2003;30:884-5.