Recommendations for the Involvement of Patient Research Partners (PRP) in OMERACT Working Groups. A Report from the OMERACT 2014 Working Group on PRP

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ABSTRACT. ***Objective.*** Patient participation in research is increasing; however, practical guidelines to enhance this participation are lacking. Specifically within the Outcome Measures in Rheumatology (OMERACT) organization, although patients have participated in OMERACT meetings since 2002, consensus about the procedures for involving patients in working groups has not been formalized. The objective is to develop a set of recommendations regarding patient research partner (PRP) involvement in research working groups.

Methods. We conducted a systematic literature review on recommendations/guidelines of PRP involvement in research; elaborated a structured consensus process involving multiple participants to develop a set of recommendations; and sought endorsement of recommendations by OMERACT.

Results. In the 18 articles included in the literature review, there was general agreement on the broad concepts for recommendations covering PRP involvement in research although they were heterogeneous in detail. Most considered PRP involvement in all phases of research with early engagement, training, and support important, but details on the content were scarce. This review informed a larger consensus-building process regarding PRP inclusion in OMERACT research. Three overarching principles and 8 recommendations were developed, discussed, and refined at OMERACT 2014. The guiding principles were endorsed during the OMERACT plenary session.

Conclusion. These recommendations for PRP involvement in OMERACT research reinforce the importance of patient participation throughout the research process as integral members. Although the applicability of the recommendations in other research contexts should be assessed, the generalizability is expected to be high. Future research should evaluate their implementation and their effect on outcome development. (First Release April 15, 2015; J Rheumatol 2016;43:187–93; doi:10.3899/jrheum.141011)

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Inclusion of the patient perspective in outcome development is an important component of the research process, because the objective is to ultimately improve clinical outcomes for patients. To effectively capture the patient perspective throughout the research process, ongoing and active collaboration between researchers and patients is essential[1].

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From 2002, the Outcome Measures in Rheumatology (OMERACT) initiative started involving patients in research, and the experiences and benefits have been well described[2,3]. Patient involvement in OMERACT work such as the development of clinical outcome measures has evolved. However, a recommended process to facilitate this collaboration has not been formalized[4]. The strong interest around patient inclusion in research in many settings, as exemplified by the Patient-Centered Outcomes Research Institute in the United States[5], by the Core Outcome Measures in Effectiveness Trials in Europe[6], and through the European League against Rheumatism (EULAR)[7], has provided additional impetus toward development of guidelines for including patients in the research process.

To address these needs, the "patient research partner (PRP) involvement in research" working group (WG) was created with the aim to develop recommendations regarding patient involvement throughout the research process for OMERACT projects.

MATERIALS AND METHODS

This WG, with members from Europe, North America, and Australasia, was co-led by 1 PRP (MdW) and 1 rheumatologist/researcher (LG), with a total of 4 rheumatologists/researchers, 4 PRP, and 1 fellow (PPC).

Terminology: The term patient research partner (PRP) is used, although other terms such as patient stakeholder, health care consumer, end-user, or patient advocate have also been proposed. PRP are defined as "persons with a relevant disease who operate as active research team members on an equal basis with professional researchers, adding the benefit of their experiential knowledge to a research project"[7]. PRP are not focus group or research participants, but full members of the research team. It reflects inclusion within the work on an "equal basis," i.e., equal opportunities to participate in research, and in OMERACT activities, including biennial meetings with full voting rights.

Literature Review

A systematic literature search was performed up to September 2013 on existing guidelines for PRP involvement in research. Databases included PubMed, Embase, Cochrane Database of Systematic Reviews, and INVOLVE, a database funded by the National Institute for Health Research to support public involvement in health and social care research (www.invo.org.uk/resource-centre/research-project-database/) using the following keywords: (patient partners or stakeholders or consumer) and (patient participation or community based participatory research or consumer involvement) and (guidelines or recommendations). Hand-searching of references and abstracts (EULAR and American College of Rheumatology meetings from 2010 to 2013) was performed. To obtain unpublished ("gray") literature, Websites relevant to the subject matter were also searched.

Data selection. We selected all publications of consensus documents and/or guidelines relating to patient involvement in research, across medical specialties.

Data retrieval and interpretation. One investigator (PC) selected and extracted the data, dividing documents into 2 levels: (1) direct: actual recommendations, explicit recommendations or guidelines for PRP involvement in research; (2) indirect: descriptive reports of patient involvement in research or opinion papers providing practical guidance or advice (but not consensus guidelines). Extracted information was ordered according to the following broad concepts identified by the WG members: extent of involvement, selection of PRP, support for PRP, communication, and acknowledgment. The analyses were descriptive.

Development of Recommendations

Initial recommendations. Based on both literature review and personal experience, draft recommendations were developed by 2 rheumatologist/researchers and 2 experienced OMERACT PRP with balanced representation from Europe and USA.

Iterative consensus process. During teleconferences and e-mail exchanges over 3 months, draft statements were reviewed and modified by the entire WG. Then, discussion and feedback was obtained on the proposed recommendations during a face-to-face session at the OMERACT 2014 meeting (1.5 h), which included 36 participants: 18 PRP, 10 clinicians/health professionals who self-classified as primarily clinicians, 6 researchers (including clinicians who focused predominantly on research), and 2 pharmaceutical industry representatives. Participants came from United States/Canada (50%) and the United Kingdom (35%), with the remainder (15%) from The Netherlands, Australia, France, Singapore, and Taiwan. Following their feedback, the WG reworded, combined, and reduced the number of statements. Later in the meeting, discussions with an unselected group of OMERACT participants (n = 11) were carried out during one of the OMERACT Filter 2.0 breakout sessions[8]. The recommendations were finalized, and the overarching principles were presented for voting at the final plenary session.

Evaluation Phase

After the OMERACT meeting, the level of agreement on the final set of statements was evaluated through SurveyMonkey, sent once, to all OMERACT 2014 participants (n = 219), without additional reminders. The OMERACT executive committee was also asked for their endorsement.

RESULTS

Literature Review

Of the 550 articles (n = 132 PubMed, n = 232 Embase, n = 180 INVOLVE, n = 84 duplicates) identified, 18 were included (2 from unpublished/"gray" literature). Nine articles were directly relevant while the other 9 were indirectly relevant[1,5,7,9,10,11, 12,13,15,16, 17,18,19,20,21, 22,23,24](Table 1). Nine papers (50%) came from the United Kingdom, 5 from the United States, and 4 from Europe. Medical specialties included oncology (n = 5), diabetes (n = 2), nephrology (n = 2), and rheumatology (n = 2). Six articles were generic in nature. Four articles were actual recommendations (Delphi consensus, n = 2; literature review, n = 2); the other articles were mainly descriptive experiences.

The extent of patient involvement was most frequently mentioned (78%), preferably to be in all phases of the project with early engagement. Support, which included aspects ensuring that the needs of PRP are addressed, such as appropriate location and schedule of meetings, or systems to promote optimal PRP engagement during project discussions, was mentioned in 67% of articles. Selection criteria for PRP were mentioned in a limited number of studies (33%) emphasizing the importance of representation of different patient characteristics and relevant experiential knowledge. Education and training for PRP mainly included providing background information about the project. In addition, formal documentation of the aims of the PRP involvement was recommended at the start of the project, preferably through a direct face-to-face meeting. A mentoring system was suggested in 3 articles, but the exact method was not elabo-

Table 1. Summary of the 18 studies included in the literature review on recommendations for patient involvement in research.

Author, Year	Country*	Source**	Method***	Relevance#	Field^	Generalizability~
Ahmed[9], 2010	USA	Literature	Framework	Direct	Generic	++
Blair[10], 2009	USA	Literature	Literature review	Direct	Geriatric	-
Boote[1], 2006	UK	Literature	Delphi	Direct	Generic	++
De Wit[7], 2011	EU	Literature	Delphi	Direct	Rheum	+
Kent[11], 2013	UK	Gray	Checklist	Direct	Generic	+
Lindenmeyer[12], 2007	UK	Literature	Descriptive	Direct	Diabetes	+
Marsden[13], 2004	UK	Literature	Descriptive	Direct	Cancer	-
PCORI[5], 2013	USA	Gray	Framework	Direct	Generic	++
Staniszewska[15], 2011	UK	Literature	Checklist	Direct	Generic	++
Abma[16], 2009	NED	Literature	Descriptive	Indirect	Renal and disability	-
Caldon[17], 2010	UK	Literature	Descriptive	Indirect	Cancer	+
Guise[18], 2013	USA	Literature	Descriptive	Indirect	Generic	++
Hewlett[19], 2006	EU	Literature	Descriptive	Indirect	Rheum	+
Katz[20], 2012	USA	Literature	Survey	Indirect	Cancer	-
Nierse[21], 2012	NED	Literature	Descriptive	Indirect	Renal	-
Rhodes[22], 2002	UK	Literature	Descriptive	Indirect	Diabetes	-
Stevens[23], 2003	UK	Literature	Descriptive	Indirect	Oncology	+
Wright[24], 2010	UK	Literature	Framework	Indirect	Oncology	+

*Origin of the article. **Where the source was from. ***How the recommendations or guidelines were derived. #Whether the data were directly relevant for recommendations of patient involvement in research or indirectly, i.e., from descriptions or information that may be helpful with recommendations or guidelines. ^The area that the recommendations or data were related to. ~The extent to which the information can be applied to areas outside the field that the information was related to (global assessment by the authors). EU: Europe; NED: the Netherlands.

rated. Effective communication was highlighted in 50% of articles, specifically reporting the importance of open 2-way dialogue between researchers and PRP, using understandable language with regular feedback. Half of the articles covered PRP acknowledgment, through either reimbursement of their travel or time spent. Tokens of appreciation and issues with authorship were mentioned in only 2 studies. Excerpts are available in Appendix 1.

From this review, it was clear that there were limited explicit recommendations in the literature. However, the broad concepts covered in the 18 publications were similar, indicating some level of consensus.

Development of Recommendations

Draft statements included 3 overarching principles and 11 recommendations (reduced to 8 after discussions during the 2 group meetings at OMERACT; Table 2). The final voting in the plenary session resulted in 99% agreement with the overarching principles.

An evaluation phase after the OMERACT 2014 meeting examined the agreement with each individual recommendation. A survey, sent to OMERACT 2014 attendees had a 30% response (66/219), which included 10 respondents who self-identified as PRP (15%), 53 as researchers/professionals (80%), and 3 from the pharmaceutical industry (5%). The recommendations received high levels of agreement (7.9–9.1 on a visual analog scale; Table 2). The final set of recommendations was formally endorsed by the OMERACT executive committee and incorporated in the OMERACT Handbook[25].

Overarching Principles

1. *OMERACT values the experiential knowledge of PRP as critical to outcome research*

The experiential knowledge of patients complements the evidence-based knowledge and clinical expertise of researchers and others. Incorporating the patient perspective is imperative for developing disease-specific core sets and patient-reported outcomes.

2. *Engaging PRP as integral throughout the research process is a fundamental OMERACT principle*

Patients are essential participants in outcomes research. Their involvement over the last decade has provided important value to the OMERACT research agenda and the conduct of outcome research[3]. Patient involvement enables co-ownership over research themes relevant to their own disease experiences and daily lives[1]. The level of involvement varies depending on the scope/type of project (e.g., a statistical project might necessitate less patient involvement).

3. *All OMERACT participants subscribe to the principles of trust, respect, transparency, partnerships, communication, diversity, confidentiality, and colearning with respect to patient involvement in research*

These are general principles of involvement, relevant not only for WG leaders and PRP but for all OMERACT participants. The term "diversity" refers to PRP characteristics, which are further explained in recommendation 3.

Table 2. Overarching principles and recommendations for patient research partner involvement in research projects in OMERACT, and indicative agreement levels for OMERACT participants (n = 66).

Overarching Principles	
1. OMERACT values the experiential knowledge of PRP as critical to outcome research.	
2. Engaging PRP as integral participants throughout the research process is a fundamental OMERACT principle.	
3. All OMERACT participants subscribe to the principles of trust, respect, transparency, partnerships, communication, diversity, confidentiality, and colearning with respect to patient involvement in research.	
Recommendations	Agreement, Numeric scale 0–10, mean (SD)
1. The WG leadership should take responsibility for appropriate representation of the patient perspective in the research project	9.1 (1.3)
2. Each WG should involve at least 2 PRP. An exception may be some projects (e.g., a statistical project) where only 1 PRP may be involved	7.9 (2.4)
3. PRP should be identified based on experiential knowledge and language skills, taking into account their personal interest in the topic	8.3 (1.5)
4. PRP and the WG leadership should discuss the goal of the project and mutual expectations	9.1 (1.7)
5. PRP should be given the opportunity to be involved throughout the research process; the level and timing of involvement may be adapted according to the scope and type of project (e.g., a statistical project)	8.7 (2.1)
6. The WG leadership should provide the PRP with timely and tailored support and information (such as lay summaries) to optimize participation and collaboration throughout the research project	8.9 (1.6)
7. The nature of PRP involvement should be reported throughout the OMERACT process, and at least in the initial research proposal and final reports	8.8 (1.9)
8. Involvement of PRP should be recognized appropriately including cochairing, copresenting, and co-authorship if applicable	8.7 (2.2)

OMERACT: Outcome Measures in Rheumatology; PRP: patient research partner(s); WG: working group.

Detailed examples are provided in the OMERACT Handbook[25] in relation to each of the 8 recommendations.

Recommendations

1. *The WG leadership should take responsibility for appropriate representation of the patient perspective in the research project*

Although the WG leader or a specified member should take primary responsibility for patient involvement, the entire research team plays an active role. PRP involvement throughout research projects is recommended, although patient roles and tasks within an individual project or WG may vary according to the stage or content of the research project. Exceptions are based on discussion with the WG leader, their executive mentor, and the executive patient stream leader.

2. *Each WG should involve at least 2 PRP. An exception may be some projects (e.g., a statistical project), where 1 PRP only may be involved*

WG should obtain the involvement of (usually) 2 or more PRP[7].

PRP are not expected to fund the cost of their participation in OMERACT meetings and related activities. Researchers should plan to support the PRP, for example, by covering expenses to attend meetings, teleconferences, or other incidental expenses.

3. *PRP should be identified based on experiential knowledge and language skills, taking into account their personal interest in the topic*

PRP identification and selection is based on experiential knowledge and language skills, taking into account their personal interest in the topic[16,21,22]. Diversity is an important OMERACT principle. Hence, selection should take into account differences in geography, socioeconomic and cultural contexts, gender, age, disease duration, disease severity, and effect of disease, and potentially other disease, personal, or external characteristics. Potential conflict of interest needs to be disclosed, particularly financial interests that may be affected by the person's involvement[26].

4. *PRP and WG leadership should discuss the goal of the project and mutual expectations*

Mutual goals and expectations are best discussed before the start of a project, during the first contact with the potential PRP[14,19], and should be reviewed regularly. It is desirable to estimate the expected time PRP are required to allocate for the project (e.g., 4 h/month over 6 months)[18], with feasible timelines (e.g., feedback within 2 weeks).

5. *PRP should be given the opportunity to be involved throughout the research process; the level and timing of involvement may be adapted according to the scope and type of project (e.g., a statistical project)*

Generally, PRP should have the opportunity to be involved throughout the research process in the following stages: identifying the research question, reviewing/contributing to

the study design, recruitment, data collection, analysis, and dissemination of the results. PRP should be consulted and should take part in decisions about the implementation of the WG research agenda. Whenever possible, PRP should attend meetings of the WG (e.g., teleconferences)[1,7,9,13,17,19,21], although some PRP may not wish to participate in all phases. The frequency of involvement may differ, depending on the stage of the project: e.g., in core domain selection, frequent involvement may be required whereas there may be less involvement with data mining for discrimination.

6. *The WG leadership should provide PRP with timely and tailored support and information (such as lay summaries) to optimize participation and collaboration throughout the research project*

PRP should receive appropriate, relevant information, e.g., lay summaries, explanation of relevant statistics, research terms, and disease features if appropriate. Open communication is important to all WG members[9,16]. The inclusion of PRP in e-mails, teleconferences, or Web conferences, as well as the OMERACT meeting and other international congresses, is encouraged. E-mails to the research team should include the PRP or a specific patient e-mail should generally be sent at the same frequency.

While some project phases require less patient involvement, a specific PRP e-mail or newsletter at least once a year would be useful. PRP should be offered the choice of what information they would like to receive, relevant for the WG.

Appropriate support at and between meetings includes actions on the part of researchers that encourage and promote PRP to contribute with confidence throughout the research project. Researchers require particular skills to achieve this. Support includes tailored information, debriefings, and encouraging PRP to speak up during meetings[16,18,19,27]. OMERACT has made structural changes to its meetings to support PRP[28]. The term support, here, does not refer to financial subsidies.

7. *The nature of PRP involvement should be reported throughout the OMERACT process, and at least in the initial research proposal and final reports*

Evidence of the effect of PRP involvement is required[15]. WG are expected to report the level of PRP involvement in the initial research proposal and in OMERACT documents such as conference prereading materials.

8. *Involvement of PRP should be recognized appropriately including cochairing, copresenting, and coauthorship if applicable*

Recognition can be ensured by having patients involved in facilitating discussion groups and reporting of results at the meetings. PRP acknowledgment can be indicated at the end of the final research report or as coauthorship if the standard rules are met[7].

DISCUSSION

Through a consensus process and based on an extensive literature review, 3 overarching principles and 8 recommendations for PRP involvement in OMERACT research projects have been developed, providing a practical guide for OMERACT WG that is also potentially useful to other researchers.

There were limitations to consider. Participants in the consensus process may not entirely reflect the whole OMERACT community; however, 99% of voters in the plenary session at OMERACT 2014 concurred with the overarching principles. In addition, these recommendations were initially based on available literature, and broad participation in discussions was encouraged before, during, and after the OMERACT meeting. These recommendations will be dynamic and subject to modification based on their future implementation. Although response to the survey after the OMERACT meeting on the individual recommendations was relatively low at 30%, this was a single mailout, and the responses mirrored that of the consensus process during the meeting.

Many groups advocate involvement of PRP in research although few practical recommendations are available[1,5,6,7]. The problem of representativeness of PRP has often been raised[5,11,23]. We believe PRP represent themselves, and are not expected to represent the entire patient perspective. Through the use of multiple, additional forms of patient participation in the phase of data collection, such as a Delphi exercises, focus group interviews, and surveys, representativeness should be achieved.

Practical support, information, training, and mentoring of PRP should be further addressed and standardized. The model developed by OMERACT appears to be a success[2,27,28] but may not be entirely applicable to other contexts.

Future opportunities include disseminating these recommendations to the wider research community and evaluating their applicability and influence within OMERACT groups and in other settings.

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APPENDIX 1. Literature review excerpts of statements regarding patient involvement in research.

Concept	Excerpt
1. Selection	
Representative	• Recruitment of potential participants to the relevant communities a transparent process that needs to be robust[11] • Suggest going beyond existing support network groups to have a broad based approach to recruitment[23] • All aspects to cover the diversity of participants[5]
Experiential knowledge	• Through informal job interviews, researchers decided whether the experiential knowledge was suitable for the project[16] • No particular education or qualifications required, but informal "job interview" performed by project leader on candidate's personal story and expectation for the project[21] • Have the disease (diabetes), easier to rely on professional referrals for potential participants known to them; however, can introduce bias, i.e., excluding sicker patients, and those who did not speak English[22]
Personal attributes	• Should take account of communication skills, motivation and constructive assertiveness in team setting, no particular education or qualifications required[7]

APPENDIX 1. Continued.

Concept	Excerpt
2. Preparation	
Setting objectives and rules	• PRP receive respect, and all private information from participants remains confidential[9]. • PI explains all aspects of the project using non-technical language before the community partner agrees to participate. All community members have self-determination rights and responsibilities[9]. • State purpose of stakeholder engagement, provide orientation information and materials (including methods of engagement, frequency of activities, project schedule, compensation plan, conflict of interest disclosure requirements)[18]
Training and education	• Increases the expertise of PRP, which might enhance their self-confidence and feelings of security, foster better communication with professional researchers. Should include balance of technical scientific training and also own knowledge base validated, e.g., opportunity to learn as an apprentice, in a climate of support and encouragement[16] • Patient to educate researchers[16] • Need courses for both PRP and researchers[1,16] • Focus on task oriented training, e.g., conducting interviews, to development of a multifaceted program training elders as gerontologists[10] • Induction training course on cancer research and research methodology[17] • PI need to ensure adequate training, although formal training is invaluable, on-the-job training is viewed to be more practical[7] • Training on qualitative and quantitative methods is useful, initial 1-1 meeting with PI with guidelines from INVOLVE[19] • Glossary of research terms[19,21] • Continuing education and training is necessary, receive orientation guidebook and undergo training about basic concepts associated with clinical trials research[12,20] • May require background or training /information if research is complex[13] • Two-day introductory training session[23]
3. Support	
Welfare	• From research PI, but attention to the welfare of the PRP like travel time and duration of meetings. PRP become fatigued easily or lose concentration when working continuously for a long time, therefore negotiations over the planning of breaks, acceptable work periods and schedules. Counselors could be appointed to provide emotional support. Intimate and stable collaborations between a professional researcher and a research partner helped both to express their feelings and cope with tensions[16] • Mentors or mentoring system[1,12,23] • Need a strategy for managing disruptive or dominating stakeholders and for resolving conflicts, conduct icebreaker sessions at in-person activities whenever stakeholders have diverse backgrounds or not already acquainted[17] • From research team[7,12,17]
Feasibility	Meetings close to PRP's homes[21]
4. Acknowledgment	
Financial/token of appreciation	• Reimbursed for travel and indirect costs (e.g. carer)[1,11,12] • Pay by the hour in addition to travel expenses[23] • Small salary acknowledging and expressing appreciation for their efforts, e.g., kidney project PRP offered a contract as a temporary worker at the university (short term project). For the intellectual disability project, arrangement was made with the PRP to prevent reductions in their allowance from the government, and nonmonetary arrangements, e.g., library access was provided[16] • No consensus on direct payment[7] • Appropriate token of appreciation, no consensus on direct payment, honorary contract, access to library, travel bursary, subscription to journal, training opportunities, research institution to develop certificate for contribution[7]
Authorship	• Acknowledged in manuscripts and research reports (with detailing of the contribution)[1] • All papers submitted include acknowledgment of PRP contributions, but not specifically as coauthor[12] • Coauthorship if fulfill ICMJE criteria[7]. Take part in writing report with authorship[21]
5. Phase of involvement	
Extent of involvement	• All phases of research, including ethics, data analysis, and presentation[17,9,13,19,21] • Need to engage in early stage[20,23]
6. Communication	
Language/ communication	• Two-way dialogue, need to create a safe and respectful working environment especially in the beginning[16] • Continuous communication: ongoing communication between the community partner and the PI, bidirectional, PI provide regular progress updates to the community, including community members not directly involved in the research, the community partner informs PI of potential concerns and offers constructive solutions to improve the research. Important in a transparent process for evaluating progress and impact (mutually agreed evaluation strategies)[16]. • Webinars, e-mail, and Web-based prioritization through ranking exercises or Delphi technique[18] • Internet, e-mail, and also early feedback from PI on success of partnership, informal partner groups, newsletters, regular email, occasional meetings[19] • Language should be made so it is understandable by the community[11] • Participants from diverse ethnic backgrounds should have access to findings in their preferred language where feasible[24]
Feedback	• Patient feedback must be provided to ensure accountability and representation[13]

PRP: patient research partner; ICMJE: International Committee of Medical Journal Editors; PI: principal investigator.