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CLINICAL SCIENCE

Patient research partner involvement in rheumatology research: a systematic literature review informing the 2023 updated EULAR recommendations for the involvement of patient research partners

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ABSTRACT

Background Patient research partners (PRPs) are people with a disease who collaborate in a research team as partners. The aim of this systematic literature review (SLR) was to assess barriers and facilitators to PRP involvement in rheumatology research.

Methods The SLR was conducted in PubMed/Medline for articles on PRP involvement in rheumatology research, published between 2017 and 2023; websites were also searched in rheumatology and other specialties. Data were extracted regarding the definition of PRPs, their role and added value, as well as barriers and facilitators to PRP involvement. The quality of the articles was assessed. Quantitative data were analysed descriptively, and principles of thematic content analysis was applied to qualitative data.

Results Of 1016 publications, 53 articles were included; the majority of these studies were qualitative studies (26%), opinion articles (21%), meeting reports (17%) and mixed-methods studies (11%). Roles of PRPs ranged from research partners to patient advocates, advisors and patient reviewers. PRPs were reported/advised to be involved early in the project (32% of articles) and in all research phases (30%), from the conception stage to the implementation of research findings. The main barriers were challenges in communication and support for both PRPs and researchers. Facilitators of PRP involvement included more than one PRP per project, training of PRPs and researchers, a supportive environment for PRPs (including adequate communication, acknowledgement and compensation of PRPs) and the presence of a PRP coordinator.

Conclusion This SLR identified barriers and facilitators to PRP involvement, and was key to updating the European Alliance of Associations for Rheumatology recommendations for PRP–researcher collaboration based on scientific evidence.

INTRODUCTION

Patient research partners (PRPs) are described as individuals living with a health condition who ‘provide input to research, through active collaboration as equal partners with researchers’.¹ Their involvement is essential to make research more patient centred, for instance, by capturing outcomes

WHAT IS ALREADY KNOWN ON THIS TOPIC

- ⇒ Patient research partners (PRPs) are increasingly integrated into medical research, particularly in rheumatology.
- ⇒ Major global health organisations recognise the central role of PRPs’ involvement in research.
- ⇒ Previous recommendations have guided researchers and PRPs to build collaborative relations but lack a strong evidence base.

WHAT THIS STUDY ADDS

- ⇒ This systematic literature review provides for the first time a comprehensive overview of the emerging role of PRPs in rheumatology research, emphasising their expanding roles, contributions and the value they bring to the research process.
- ⇒ The review identified key barriers to PRP involvement, ranging from personal factors to challenges in training, communication and collaboration, and also identified strategies to enhance PRP involvement.
- ⇒ Early and sustained involvement of PRPs, as well as a supportive environment and effective communication, were found to be essential to enhance the relevance and impact of PRP contribution to research.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

- ⇒ Recognising and addressing the barriers to PRP involvement can lead to better support for PRPs, enhancing their involvement in research.
- ⇒ Some facilitators identified include involvement of PRPs since the early stages of research, a supportive environment for PRPs and encouraging researchers to adopt more flexible strategies and behaviours to maximise the benefit of PRP involvement.
- ⇒ This literature review informed European Alliance of Associations for Rheumatology recommendations, highlighting the importance of active collaboration, training, mutual respect, and transparent communication between PRPs and researchers.

that matter to patients. Over the past two decades, the magnitude of PRP involvement and their roles in research has grown substantially.^{2–8} Patients have transitioned from passive subjects to active collaborators and equal partners, bringing their unique perspectives and valuable insights to the forefront of medical research.⁵ This change has not only profoundly modified research practices but has also underscored the integral role PRPs play in shaping the future of medical practice.⁹ The importance of PRP involvement in research has become widely recognised as an essential component of high-quality patient care, highlighted by organisations such as the WHO⁴ and European Medicine Agency (EMA),¹⁰ and is acknowledged across various medical specialties.^{11–13}

In rheumatology, this paradigm shift has been significant. In 2011, the European Alliance of Associations for Rheumatology (EULAR) developed recommendations for the involvement of patient representatives in scientific projects based on expert opinion.¹⁴ These recommendations marked a pivotal step, setting the stage for the involvement of PRPs in research projects. Since then, these EULAR recommendations have guided other organisations such as Outcome Measures in Rheumatology (OMERACT), Group for Research and Assessment of Psoriasis and Psoriatic Arthritis (GRAPPA) and the Foundation for Research in Rheumatology (FOREUM), to recognise the important role of PRPs or to develop their own guidelines for collaborative research.^{15–18}

As the landscape of patient involvement in research evolves, the literature has witnessed a great surge in data and studies concerning PRP involvement.^{2 4 5 15 19–25} These studies not only shed light on the benefits of PRP participation but also highlighted the challenges encountered in this collaborative effort and solutions proposed to overcome barriers.^{21 22 25–29}

In 2022, EULAR decided to update the 2011 recommendations for PRP involvement in research, focusing specifically on PRPs in the context of chronic conditions.¹⁴ In accordance with the EULAR standardised operational procedures (SOPs) supporting this update, we conducted a systematic literature review (SLR) to inform the EULAR taskforce.

METHODS

To support the update of the EULAR recommendations, we conducted in 2023 an SLR that encompassed both qualitative and semiquantitative analyses of recent publications in rheumatology, with the goal of identifying factors that affect PRP involvement, including barriers and facilitators.

Literature search

The SLR aimed to identify publications reporting PRP involvement in rheumatology research published between 1 January 2017 and 1 January 2023. We searched the electronic database PubMed MEDLINE using the terms “patient research partner”, “patient expert”, “patient and public involvement (PPI)”, their synonyms and related concepts. Details of the search terms and search strategy can be found in online supplemental table 1. Two authors (KA, LG) independently assessed the title, abstract and keywords of every publication identified. In the event of disagreement between the reviewers, disparities were discussed and resolved. Additionally, we performed a scoping review of databases to assess PRP involvement and explored six websites from rheumatology: OMERACT, GRAPPA, American College of Rheumatology, EULAR, FOREUM and Osteoarthritis Research Society International. We also searched 2 regulator websites: Food and Drug Administration and EMA, and 10 websites of

three selected specialties recognised for significant PRP involvement: cardiology, oncology, endocrinology (diabetes) (online supplemental table 2). A specific search was done in two websites focusing on patient and public involvement: INVOLVE UK by the National Institute for Health Research and Education that empowers (European Patients’ Academy on Therapeutic Innovation), and in orphan diseases to answer specific research questions about training, involvement in grant applications and remuneration of PRPs (online supplemental table 3).

The scope of the literature search was defined by the EULAR taskforce steering group,¹ and addressed 11 specific research questions (Box 1).

Inclusion and exclusion criteria

We included all types of articles reporting PRP involvement in all types of research, including trials and observational studies, qualitative studies, mixed-methods studies and reports of meetings, opinion papers and reviews. We did not exclude published articles from any country, aiming to enhance the generalisability of our findings. Recommendations and guidelines on PRPs were also analysed and were used as supportive information. Articles not focused on rheumatology research or not bringing any information on PRPs (ie, not answering one or more research questions), as well as not in English, were excluded. Articles only mentioning PRPs or their involvement, without providing any details (eg, on their roles, contributions or barriers/facilitators), were excluded as well. Articles with duplicate information (ie, multiple publications reporting on a single study) were excluded if they did not provide additional information relevant to our research questions.

We also identified relevant articles by hand search of the references cited in the included studies, extending the inclusion period to the date of publication of the previous recommendations (2011–2023).

Data extraction

Data collection encompassed both quantitative and qualitative data, addressing various aspects of PRP involvement and providing answers to our research questions (Box 1). Data were extracted and checked independently by two authors (KA and MdW). Discrepancies were resolved by discussion among the core team (KA, MdW, PS, LG).

Quality assessment

Papers were assessed for quality only if they reported original data. Review papers, recommendation papers, opinion papers, case studies, study protocols, report papers and qualitative studies not primarily focused on PRPs were excluded from quality assessment. Given the diversity of study types, we used the Critical Appraisal Skills Programme (CASP) checklist for qualitative studies, literature reviews and cross-sectional studies as described in the EULAR SOI.^{30 31} This tool, originally developed for qualitative studies, assesses elements such as the clarity of research aims, appropriateness of methodology, suitability of the research design, adequacy of data collection and clarity in reporting outcomes. For mixed-methods studies, we used the Mixed Methods Appraisal Tool (MMAT, a critical appraisal tool that is designed for the appraisal stage of systematic mixed-studies reviews, that is, reviews that include qualitative, quantitative and mixed-methods studies³² (see online supplemental tables 4 and 5 for quality assessment). To facilitate interpretation, an overall quality assessment for the level of evidence (LoE) was conducted by evaluating the number of items on the

Box 1 Research topics included in the systematic literature review

1. Definition of patient research partners (PRPs)
⇒ *How to define a PRP? Is the current definition of PRPs still adequate?*
2. Roles and activities undertaken by PRPs
⇒ *What are the roles and activities of PRPs in rheumatic musculoskeletal disease research?*
3. Benefits and added value of PRP involvement for PRPs themselves, researchers, the research itself
⇒ *What is the added value of PRPs in different types of research and groups?*
4. Types of scientific projects that involved PRPs and the stages of the projects in which they participated
⇒ *What types of projects are (or should) PRPs (be) involved in?*
⇒ *What phases of a project are (or should) PRPs (be) involved in?*
5. Selection and recruitment processes for PRPs
⇒ *How are (or should) PRPs (be) recruited and selected?*
⇒ *How many PRPs are (or should be) involved in the research?*
6. Insights into the experiences and feedback provided by PRPs
⇒ *What are the PRP feedback and experiences, in terms of facilitators and barriers to PRP involvement?*
⇒ *How can we improve the PRP experience and involvement overall?*
7. Roles of a coordinator for PRPs in research
⇒ *Are PRP facilitators involved, if so how, and is it useful?*
⇒ *Is a facilitator/PRP coordinator recommended?*
⇒ *What is the reported usefulness of a facilitator?*
8. Training provided to PRPs or researchers
⇒ *Do the PRPs involved have a specific training (previously/ during the study)?*
⇒ *How should researchers be educated, trained, supported to enhance PRP involvement?*
9. Evaluation and monitoring related to PRP involvement
⇒ *How should PRP involvement be monitored or evaluated? At which time points and by whom?*
⇒ *How should PRP involvement evaluation/monitoring be reported?*
10. Recognition, compensation and acknowledgement of PRPs during their involvement in a scientific project
⇒ *How should PRP involvement be recognised and acknowledged?*
⇒ *Is (should) compensation (be) proposed?*
11. Barriers encountered and proposed solutions to enhance PRP involvement
⇒ *What are the barriers encountered during PRP involvement?*
⇒ *Which strategies and contextual factors enable optimal engagement of PRPs?*

score checklist and on the key items. Subsequently, the authors reached a consensus on classification of the articles' quality as high, medium or low quality.

This SLR was not considered appropriate by PROSPERO for registration due to the mixed-methods study analyses involved.

Patient and public involvement

This SLR study is the result of a co-production of three PRPs (MdW, CZ, HB) and five researchers, all being members of the EULAR steering committee responsible for updating the EULAR

recommendations on PRP involvement.¹ The three PRPs actively contributed to all meetings and discussions within the steering committee. They were involved at the early stage of formulating the research questions until reviewing and agreeing on the final manuscript. They were also actively engaged in planning dissemination and implementation of the study findings within the wider community and patient associations. The recruitment of the PRPs was coordinated by one of the PRPs (MdW), the convenor of the project.

Analysis

For quantitative data, a descriptive analysis of findings is reported, including characteristics of studies (study design, population, country, study objectives), characteristics of PRPs, selection process of PRPs, type of involvement, phases of the research where their involvement occurred, with numbers and percentages using frequency tables and charts.

The number of PRPs involved in the studies was quantified using two distinct methods: first, coauthorship count: direct examination of the research articles' authorship lists. PRPs were identified based on explicit mentioning of their role as 'PRP' or other specific identification. Second, participation count: this approach assessed the number of broader involvements of PRPs in activities of the research project. For instance, in a GRAPPA meeting report, the number of PRPs who actively participated was counted.⁸

Qualitative data were analysed according to the principles of thematic content analysis (more details in online supplemental table 6).³³ The results were discussed within the EULAR task-force,¹ and any disagreements on the interpretation of the findings were resolved by a consensus of the core group (MdW, LG, PS, KA).

RESULTS

Search strategy

The SLR yielded a total of 1016 records of which 941 (92.6%) were excluded based on titles and abstracts. We conducted a full-text screening of 75 papers and 46 (61.3%) were included. The main reasons for exclusion were papers not related to rheumatology, lacking reports of PRP involvement in research, being irrelevant to our research questions, or being duplicates or conference abstracts (figure 1). Additionally, 7 papers were identified by hand search, resulting in a total number of 53 included articles.

Quality assessment (LoE) of the papers

Nineteen articles were assessed for quality using the predefined scores according to the study type. Overall, 79% (15 of 19) were classified as high quality, 11% (2 of 19) as medium quality and 11% as low quality (online supplemental table 4).

Study characteristics

The included studies were qualitative studies (14 of 53, 26%), opinion articles (11 of 53, 21%), meeting reports (9 of 53, 17%), mixed-methods studies (6 of 53, 11%), recommendation articles (4 of 53, 8%), reviews (SLR or scoping review; 3 of 53, 6%), cross-sectional (2 of 53, 4%), case studies (2 of 53, 4%), observational (1 of 53, 2%) and study protocol (1 of 53, 2%) (online supplemental tables 4 and 5).

Overall, 62% were published in rheumatology journals. Geographically, most of the studies were from Europe (50%), followed by North America (31%).

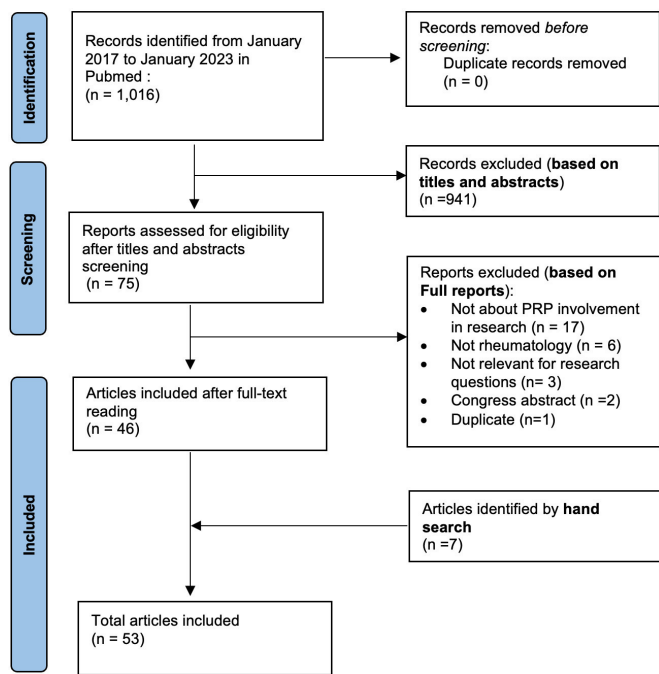


Figure 1 Flow chart of selected article search on PRP involvement in rheumatology research. PRP, patient research partner.

Identification of barriers encountered and proposed solutions to enhance PRP involvement

Barriers to PRP involvement (table 1 and online supplemental table 7) included emotional and personal factors, communication and relationship challenges, inadequate training and

support, difficulties in the research process and pace, as well as collaboration and engagement issues.^{2-4 21 22 24-27 34-42} Effective strategies to enhance PRP involvement (table 1) included early involvement, a supportive environment, effective communication and trust, and providing support and training for PRPs and researchers.^{7 21 22 26 29 38 40 43 44}

Definitions of PRP

Among the 53 included papers, 62% provided a definition of PRP. Importantly, a significant portion (30%) of these papers^{4 6 15 26 27 34-36 45 46} adopted the 2011 EULAR definition of PRP 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 any phase of the project’.¹⁴ These papers consistently emphasised the importance of active involvement and fostering equal partnerships between PRP and researchers.

Additionally, seven papers (13%) expanded upon this definition by incorporating informal caregivers into the PRP definition,^{20 28 37 38 47-49} known as persons, usually family members, who provide unpaid care to someone with whom they share a personal relationship.

The roles and activities of PRPs

The roles and activities of PRPs covered a wide spectrum, extending from research partners to patient advocates, advisory roles and participation as patient reviewers (as detailed in table 2 and online supplemental table 8). Their contributions encompassed a diverse range of activities, including providing input in guideline development, shaping research agendas, and actively advocating in scientific and clinical committees.

Table 1 Barriers and strategies to enhance PRP involvement in rheumatology research

Concept	Barriers	Strategies to enhance patient involvement
Emotional and personal factors	<ul style="list-style-type: none"> ▶ Emotional burden ▶ Fatigue ▶ Need to accommodate PRP needs (physical and knowledge, level of involvement) ▶ Lack of trust ▶ Time and budget constraints 	<ul style="list-style-type: none"> ▶ Provide a supportive environment ▶ Provide flexibility and accessible accommodation ▶ Allocate adequate resources ▶ Practice active listening ▶ Recognise PRP contributions
Communication and relationship	<ul style="list-style-type: none"> ▶ Feeling unheard ▶ Power imbalance ▶ Inconsistent and poor communication ▶ Loss of confidentiality ▶ Use of medical jargon 	<ul style="list-style-type: none"> ▶ Appoint a coordinator to facilitate PRP involvement ▶ Clarify patient roles and objectives ▶ Exchange mutual expectations early at project initiation ▶ Avoid complex medical terminology ▶ Ensure open and transparent communication ▶ Ensure feedback and trust
Training and support	<ul style="list-style-type: none"> ▶ Lack of awareness about PRP involvement among researchers ▶ Overburdening of PRPs ▶ Inadequate training and support of PRP ▶ Lack of resources and compensation of PRP 	<ul style="list-style-type: none"> ▶ Increase awareness about importance of PRP involvement ▶ Allocate resources for patient engagement ▶ Provide proper training tailored to PRP needs ▶ Provide training and support to researchers ▶ Appropriate recognition of PRP ▶ Encourage PRPs to ask questions and express needs ▶ Communicate workloads
Research process and pace	<ul style="list-style-type: none"> ▶ Challenges to recruit PRPs ▶ Time commitment for researchers ▶ Anxiety about delays in projects ▶ Higher demands on resources ▶ Forced changes in working practice 	<ul style="list-style-type: none"> ▶ Address recruitment challenges ▶ Establish realistic timelines ▶ Manage researcher and PRP time commitments ▶ Build trust through open communication and demonstrate commitment to research progress
Collaboration and engagement	<ul style="list-style-type: none"> ▶ Lack of PRP diversity and representativeness challenges ▶ Discrepancies in views ▶ Uncertainty in incorporating patient experiences ▶ Risk of sharing data prior to peer review 	<ul style="list-style-type: none"> ▶ Discuss representativeness and diversity of PRPs within research team ▶ Involve PRPs from project inception and clearly define their roles upfront ▶ Discuss and address discrepancies ▶ Co-create PRP contracts to ensure confidentiality

PRP, patient research partner.

Table 2 Activities and roles of PRPs

Areas of involvement	Activities and roles of PRPs
Development/design	<ul style="list-style-type: none"> ▶ Guidelines development,²² including voting on draft recommendations²⁴ ▶ PRO development^{5,21} ▶ Grant application³ ▶ Survey/questionnaire development^{2,3} ▶ Interview guide⁶⁶ ▶ Tools (educational or others)^{3,4,57} ▶ Patient-facing materials (develop patient information sheets, pamphlets, glossary, distribution and use of booklet, website, poster)^{3,6,42} ▶ Developing or reviewing (recommendations) lay summaries^{4,5}
Leadership	▶ Establishing a patient association ⁵
Co-leadership role	<ul style="list-style-type: none"> ▶ Member of steering committee⁷ (eg, EULAR, FOREUM, GRAPPA⁶¹) ▶ Member of working groups (eg, GRAPPA⁸)
Coauthorship	<ul style="list-style-type: none"> ▶ Writing research papers (coauthoring)³ ▶ Writing research protocol⁵⁶
Education	<ul style="list-style-type: none"> ▶ Trainer at a training programme⁶ ▶ Mentoring less-experienced PRPs³⁸
Planning	<ul style="list-style-type: none"> ▶ Strategic planning^{42,47} ▶ Event planning: organise, communicate⁵² ▶ Interview schedules³
Facilitation	<ul style="list-style-type: none"> ▶ Session at a conference⁵³ ▶ Held sessions in a meeting⁶⁸
Reviewer	<ul style="list-style-type: none"> ▶ Grant application² ▶ Research projects (ongoing)^{5,58} ▶ Summaries of scientific publications⁵
Recruitment	▶ Support recruitment of PRPs (to a project or association) ^{3,6,67}
Evaluation	▶ Training programme ⁶
Participation	<ul style="list-style-type: none"> ▶ In dissemination of research results^{3,7} ▶ Attendance of conferences and scientific meetings^{3,40} ▶ In meetings², discussions⁶⁶ ▶ In quality of care improvement programmes^{51,60}

EULAR, European Alliance of Associations for Rheumatology; FOREUM, Foundation for Research in Rheumatology; GRAPPA, Group for Research and Assessment of Psoriasis and Psoriatic Arthritis; PRO, patient-reported outcome; PRPs, patient research partners.

The added value of PRP involvement

The literature reported that PRPs added significant value across various aspects of research (table 3). Specifically, 53% of the articles indicated that PRP involvement brought benefits for the PRP themselves, that is, better understanding of their medical condition, acquisition of practical skills, improved comprehension of the research process and increased self-confidence.^{2,21,25,36,39} Furthermore, 26% of the articles highlighted advantages for the research process, that is, heightened relevance of the research, enhancement of its overall impact and enrichment of the results by adding experiential knowledge.^{2,7,21,25,29,36,38,39,45} The positive impact on researchers, reported in 15% of the articles, encompassed deeper insights into research priorities, increased motivation, innovative ideas, awareness of the impact of their work, a comprehensive approach to addressing patients' needs and improved communication in lay language (table 3).^{2,21,25,34,36,38,40} The added value of PRP involvement was also reported as advantageous for the wider community by enhancing the acceptance of research that prioritises community benefits.^{2,21,25,36}

Types of research that involved PRPs

PRPs were actively involved in a wide range of scientific projects, including basic, translational and clinical research.⁵⁰ Although

Table 3 Articles reporting on added value of PRP involvement in research for PRPs, for researchers and for the research

Added value for:		Percentage of articles	Number of articles (N=53)
PRPs	Better knowledge of disease	30	8
	Better knowledge of research	19	5
	Acquisition of practical skills	15	4
	Confidence	15	4
	All	Total: 51	27
Researchers	Better understanding of research priorities and needs	19	7
	Increased motivation and focus	9	3
	Gain of novel perspectives and ideas	9	3
	Real-life implication of their work	9	4
	Attaining a more holistic view of patients with RMD	9	3
	Better use of lay language	6	2
	All	Total: 15	8
Research	Enhancement of the relevance of research	50	7
	Improve the impact of the research	21	3
	Bring experiential knowledge to research	21	3
	All	Total: 26	14

PRP, patient research partner; RMD, rheumatic musculoskeletal disease.

the benefits of PRP involvement were less apparent in basic and translational research, some researchers and PRPs recognised the substantial advantages of collaborative partnerships in this area.^{3,25,34} A scoping review highlighted the benefits of PRP engagement in preclinical research, including enhanced understanding of basic science research for PRPs, broadened perspectives for researchers, and positive influence on study questions and methods, along with fostering mutual learning, new collaborations, and improved research quality and efficiency.⁴⁰ One study reported that researchers were committed to finding more meaningful ways to integrate PRPs into basic scientific research and dissemination of the project results.³ Strategies to enhance PRP involvement (ie, training, support, PRP-focused tasks) were also reported.³

Research phases in which PRP participated

Early involvement of PRPs in the research was reported or recommended in 32% of the included articles, emphasising engaging PRPs from the inception of a research project.^{2,19-22,27-29,34,36-38,43,45,47,51} This early engagement was reported to enable PRPs to actively shape research questions and methodologies in line with their priorities. Additionally, 30% of the articles stressed the importance of PRPs' continuous participation throughout all research stages (table 4).^{4,15,21,22,26,35,43,52-54}

Number of PRPs

The number of PRPs involved in research is shown in online supplemental figure 1. When considering the coauthorship lists, the majority of articles clearly specified the name and identity of PRPs; subsequently, the number of PRPs involved in the writing and reviewing of the article could be easily deduced. Yet, in 19% of cases, the identification of a coauthor as a PRP was unclear. In cases where PRP involvement was explicitly highlighted by coauthorship, 34% of the articles included one or two PRPs per project, 17% of articles included three or four PRPs, and 25% of articles involved more than five PRPs. Notably, single-centre

Table 4 Articles reporting or recommending PRP involvement in different phases of the research project

Phases of the research project	Number of articles (n) reporting or recommending PRP involvement n (%) (n total=53)
Conception of the study and research question	9 (47)
Study design and planning	9 (47)
Patient inclusion in the study	9 (47)
Data collection	8 (45)
Data analysis	8 (45)
Data interpretation	11 (51)
Dissemination	15 (58)
Implementation	4 (38)
Evaluation	3 (6)
All phases	16 (32)

PRP, patient research partner.

studies commonly involved one or two PRPs as coauthors. One study, which engaged four PRPs, found this number to be beneficial due to the diverse perspectives they brought.⁴⁵ Larger-scale international consortia projects recruited a higher number of PRPs, with around six PRPs being identified as an effective group size for facilitating participation and decision-making.²

On the other hand, when reporting all PRP involvement and activities in a research project, 36% of the articles reported a number of PRP higher than nine (online supplemental figure 1). Therefore, the number of PRPs involved in research can be higher than the number of PRPs mentioned as coauthors.

Selection and recruitment processes for PRPs

The selection process of PRPs was reported in 34% of articles (figure 2). PRP selection criteria were mainly language proficiency (11%), research knowledge (6%), disease diagnosis (9%), communication skills and constructive assertiveness (9%), motivation (8%), educational background (6%), experiential knowledge and expertise (6%) as well as travel capability (4%).^{2 3 15 19 21 23 24 27 34 35 55-58} Recruitment methods for PRPs were diverse, relying on patient organisations, marketing companies, rheumatology associations, social media, community outreach, clinic visits, personal connections with patients or researchers, word-of-mouth referrals and volunteering.^{2 21 34 38 41 44 53 59} Furthermore, 28% of studies emphasised the importance of clarifying patient roles through clear goal-setting and exchanging mutual expectations early in the project

initiation phase.^{15 19-21 27 29 36 42 45 47} Additionally, 28% of studies highlighted the need for inclusivity and diverse representation in PRP recruitment.^{2 4 15 35 41 42 52}

Creating a supportive environment for PRPs

A supportive environment for PRPs was reported to depend on several key principles (table 1)^{4 19 20 25 36 42 52 53 60}: ensuring a balanced and manageable workload that respects PRP abilities, providing adequate resources and time for PRP involvement, offering support to overcome language barriers, promoting flexibility and offering accessible accommodation to participate in meetings and scientific conferences.^{7 21 38 40 43 44} Equal relationships and co-leadership between PRPs and researchers were cited in several papers as crucial, emphasising mutual respect, trust, and open, transparent communication.^{7 15 19} Building strong team communication, and establishing informal personal relationships between PRPs and researchers were also found to be important factors to enhance collaboration.^{20 38 47} Regular feedback and discussions about the quality of collaboration, combined with ongoing adjustments to meet the needs and preferences of PRPs, were proposed in two papers.^{34 45}

Roles of a PRP coordinator

A PRP ‘coordinator’ was defined in some papers, as an individual or a role within a research team responsible for facilitating and supporting the collaboration between researchers and PRPs.^{2 20 25 47 61} The presence of a PRP coordinator was reported or advised in 29% of the included articles.^{2 3 19 28 34 35 40 42 44 48 61} PRP coordinators were reported to be helpful in facilitating effective communication among PRPs, researchers and stakeholders, aligning expectations, organising logistics, moderating group discussions, providing ongoing education and support, and assisting in the recruitment and selection of PRPs in projects (table 5).^{2 20 25 35 36 42 47} This role was reported to be taken by a member of the research team, a PRP or a designated person within a patient organisation or academic institution.^{2 38}

Training of researchers

We found that 34% of the included articles included in the SLR reported or advised training or education of researchers.^{4 7 19 21 25 28 29 38-40 44} Researchers could receive training concerning various aspects of working with PRPs (table 6).

Training of PRPs

Educating and training PRPs was proposed in many papers to enhance the quality of their collaboration with researchers. Notably, nearly half of the publications emphasised the

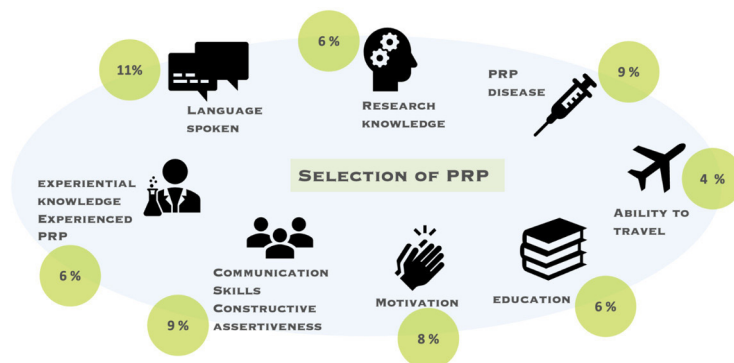
**Figure 2** The selection criteria of PRPs reported in the studies. PRP, patient research partner.

Table 5 Potential roles of a PRP coordinator

Competencies	Roles of PRP coordinator
Communication	<ul style="list-style-type: none"> ▶ Enable effective communication among patients, researchers and other stakeholders while maintaining neutrality ▶ Clarify diverse perspectives, strengths and weaknesses ▶ Ensure equal and impartial participation ▶ Foster positive social relations between PRPs and researchers ▶ Monitor and evaluate PRP strategies
Project coordination and expectation alignment	<ul style="list-style-type: none"> ▶ Mediate discussions to align expectations across all parties ▶ Ensure clarity on roles and expectations ▶ Organise logistics around PRP involvement ▶ Assist at any stage of the research regarding PRP involvement
Facilitate research discussions	<ul style="list-style-type: none"> ▶ Moderate (small) group discussions effectively ▶ Encourage deeper discussions through probing questions
Continuity and education	<ul style="list-style-type: none"> ▶ Provide on-demand patient engagement education ▶ Maintain training and support continuity for PRPs and researchers
Selection of PRP	<ul style="list-style-type: none"> ▶ Select and invite PRPs for project participation ▶ Assist in PRP group development
PRP, patient research partner.	

importance of training, with 21% recommending it and 25% providing it.^{25 28 29 35 37 45 51 62-64} PRP training and support included various aspects (table 6). Training of PRPs was reported to foster well-prepared and empowered PRPs ready to engage effectively in research collaborations.^{22 26 29}

Evaluation and monitoring related to PRP involvement

Around 21% of the included publications recommended or reported some form of evaluation,^{3 4 15 19-21 25 34 35} with 28% collecting feedback from PRPs on their involvement. Regular discussions and evaluations of the quality and impact of PRPs' collaboration and contributions were reported to enhance understanding, satisfaction and impact, allowing for adjustments and improvements as needed.^{4 5 37 60} Some tools were reported for monitoring such as the Patient-Centered Outcomes Research Institute conceptual framework, an evaluative framework for research engagement,¹⁹ surveys to evaluate the impact of PRPs in the project,^{3 26} the Public Involvement Impact Assessment Framework Guidance,⁵³ and the Guidance for Reporting Involvement of Patients and the Public.^{25 34}

Recognition, compensation and acknowledgement of PRPs

Recognising, compensating and acknowledging the contributions of PRPs during their involvement in a scientific project

Table 6 Reported training content for researchers and PRPs

Soft skills	Training content for researchers	Training content for PRPs
General training	<ul style="list-style-type: none"> ▶ Provide guidance on how to engage lay audiences in research 	<ul style="list-style-type: none"> ▶ Guidance and support from experienced PRPs¹⁹ ▶ Individual support and training provided upon request²⁵
Communication skills training	<ul style="list-style-type: none"> ▶ Exercises to improve communicating research findings to non-scientists ▶ Strategies to avoid jargon and define terms for better understanding of research discussions by PRPs ▶ Practising communication skills: respectful dialogue, active listening and knowledge checks for successful PRP engagement 	<ul style="list-style-type: none"> ▶ Communication skills training⁴⁵
Education	<ul style="list-style-type: none"> ▶ Understanding the advantages of involving PRP in research ▶ Understanding collaborative research principles 	<ul style="list-style-type: none"> ▶ Training courses on their own⁴⁵ ▶ One-on-one training with mentors³⁸ ▶ Individual support and training tailored to needs of the individual or specific research activities^{4 15 24} ▶ Intensive training programme in research for PRPs, for example, 2-day training as a PRP in research⁶³ ▶ Preparatory course for PRPs, for example, special training by the German League against Rheumatism⁴⁵ ▶ Continued education opportunities for PRPs, for example, courses through EULAR School of Rheumatology^{5 69} ▶ Educational webinars, workshops and feedback sessions, for example, OMERACT,⁵ EUPATI
Resources	<ul style="list-style-type: none"> ▶ Resources available on websites for researchers 	<ul style="list-style-type: none"> ▶ Online training on patient participation in research⁶² ▶ Reference materials for PRP involvement in research, for example, EULAR patient involvement reference cards^{51 70} ▶ Glossaries for research terminology, for example, glossaries on NIH and CARRA websites³⁸ ▶ Glossary and lay summaries of sessions in conferences or prior to meetings,²⁵ for example, OMERACT¹⁹ ▶ Extensive library of engagement training resources, for example, PCORI engagement training materials³⁸ ▶ Comprehensive training manual for PRPs,³⁷ for example, EUPATI
Confidence building	<ul style="list-style-type: none"> ▶ Building confidence in engaging young individuals in research activities 	–
Engagement of PRPs	<ul style="list-style-type: none"> ▶ Practising role-playing for the first introduction meetings between PRPs and researchers ▶ Identification of opportunities to increase PRP involvement 	–
CARRA, Childhood Arthritis and Rheumatology Research Alliance; EULAR, European Alliance of Associations for Rheumatology; EUPATI, European Patients' Academy on Therapeutic Innovation; NIH, National Institute of Health; OMERACT, Outcome Measures in Rheumatology; PCORI, Patient-Centered Outcomes Research Institute; PRP, patient research partner.		

were reported to be essential components of equal and meaningful partnerships.²⁷

In the context of recognition, coauthorship was cited as proof of PRP involvement and equality in research collaborations.^{5 39} The SLR revealed a growing trend in recognising PRPs through coauthorship in 68% of articles,^{2-6 8 15 19-21 23-26 28 29 34 36-40 42 43 45 47-50 52 54 56 58 60 63 65} and acknowledgement in 45% of articles.^{3 6 7 25 27 28 34 37 43-45 48 51 53 56-61 63 65-67}

Compensation refers to the payment of salary, wages, honorarium, fees or allowances for the time commitment and expertise of PRPs; this is different from reimbursing PRPs for expenses (eg, travel expenses and accommodation).⁴⁹ Non-compensation for PRPs was reported as a limitation and challenge for their effective involvement.⁴ While PRPs can opt out of payment, several papers reported that researchers should consider compensation in their budget planning.^{2 39 49} Some articles advised that institutions should simplify processes for fair PRP payment, and funders should enable researchers to allocate resources for PRP involvement.⁵

DISCUSSION

The role of PRPs in rheumatology research has significantly expanded over recent years. The findings of this SLR underscore the important roles and contributions of PRPs in research projects, and the added value of PRP involvement, not only in clinical research, but also in basic, translational, registry and longitudinal observational studies. This review also highlighted current challenges and barriers, and pulled together proposals of strategies to overcome them.

The exact definition and roles of PRPs remain unclear for some researchers. A wide proportion of the reviewed studies had adopted the 2011 EULAR definition of PRP which reflects the global acknowledgement of the importance of PRP involvement in rheumatology research and the need for specific recommendations.¹⁴ PRPs hold a crucial position in recognising and actively integrating the patient perspective, their voice and needs into research decision-making processes. Diverse roles and activities were undertaken by PRPs in this SLR, from research partners to patient advocates, reflecting the many ways PRPs can contribute. Their involvement, as evident in recent papers shaping research priorities, guideline development, and scientific and clinical committees, suggests a trend towards more inclusive and patient-centred research practices.

Our review revealed specific barriers and challenges in communication, training, research processes and collaboration. These challenges highlight difficulties in communication and relationship dynamics during research, the necessity for training and support for both PRPs and researchers, concerns about the research process and its pace, and obstacles in PRP collaboration, including issues of recognition and diversity. Inclusivity and diversity are important topics for future research. To address these challenges effectively, targeted strategies such as fostering open communication, creating a supportive environment, ensuring early and sustained involvement, using a PRP coordinator and providing appropriate training and support for PRPs and researchers are crucial. These findings underscore the ongoing need for refining and implementing these strategies to enhance PRP involvement more efficiently.²⁶

A key observation from the SLR is the importance of early and sustained PRP involvement in research projects. Engaging PRPs from the research project's inception ensures that research questions and methodologies are aligned with patients' priorities and perspectives right from the start. Sustained involvement

further reinforces the trust and collaboration between PRPs and researchers, leading to research outcomes that are more relevant and impactful. The OMERACT recommendations proposed that the level and timing of PRP involvement should vary based on the scope and type of project, emphasising adaptability as a key factor for successful involvement.¹⁵

Evaluation and monitoring are also integral aspects of PRP involvement. This ongoing reflection and feedback process is vital for fostering effective and meaningful PRP involvement in research. Recognition, compensation and acknowledgement of PRPs stand as key elements for fostering a meaningful partnership. Coauthorship serves not only to document the PRP's contribution but also reinforces the idea of collaborative research. Of note, we observed disparity between the involvement of PRPs in research activities versus their acknowledgement as coauthors. This disparity may arise from some PRPs not prioritising or desiring coauthorship, or being unable to participate in producing and writing a research paper due to health-related challenges such as disease flare-ups or fatigue. In ensuring equitable recognition, a collective effort is essential to guarantee that PRPs receive due acknowledgement and compensation for their valuable contributions to scientific research.

Our study has strengths and weaknesses. One important strength of this SLR is that the findings will equip researchers, healthcare professionals and other stakeholders with evidence-based solutions to improve PRP involvement in medical research. To this end, the findings have supported the process of updating the EULAR recommendations for PRP involvement and made them more evidence based.¹ Another strength is the obtention of a more nuanced understanding of the challenges and complexities surrounding PRP involvement in rheumatology research. Furthermore, our study stands out for its comprehensive approach, analysing a broad spectrum of study types, including quantitative and qualitative studies, reviews, opinion pieces and information from websites. The inclusion of various rheumatic musculoskeletal disease conditions, encompassing both paediatric and adult populations, enhances the robustness of our findings. Another notable strength lies in the co-production of this work by three PRPs. The project was initiated and led by a PRP (MdW) who gave the work direction, participated in article screening, article analysis, overall interpretation and manuscript writing. The two other PRPs brought important insights into PRP roles, facilitators and barriers.

A limitation of the study might be the heterogeneity of the included papers. Because of the expected limited reporting of PRP involvement in rheumatology research, we decided to include a diversity of papers in the SLR, varying from qualitative studies, case studies and original research papers to conference reports and opinion articles. This heterogeneity did not allow for any form of meta-analysis, nor for identifying themes that would benefit individual groups of PRPs such as people with rare diseases, children or young adults, or people with different cultural or ethnic backgrounds. Furthermore, quality assessments could not be uniformly applied across all study types. It is important to note that the traditional evidence hierarchy may not be applicable to this SLR, given the expected absence of randomised controlled studies. Despite this, certain papers were assessed to be of high quality of evidence within their respective study types. While the systematic approach ensured a comprehensive gathering of data, there might be relevant grey literature or non-English-language publications that were not included. Another limitation might be the time period of the last 6 years, including data from articles published between January 2017 and January 2023. This time frame was chosen to reflect studies

performed after the 2011 EULAR recommendations were published, taking into account the implementation time gap.¹⁴ Furthermore, the chosen time span resulted in 53 articles which was deemed sufficient for gathering relevant data related to our research questions.

In conclusion, this SLR identified numerous publications reporting on PRP involvement in rheumatology research. Most authors reported that PRP involvement not only enriches the research process but also ensures that research outcomes are more relevant, meaningful and patient centred. However, for this involvement to be genuinely effective, it is essential to address the barriers and challenges that PRPs and researchers are facing. By updating the EULAR 2011 recommendations, based on the findings of this SLR, we can look forward to a future where research is more inclusive, collaborative, and aligned with patient needs and perspectives.

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REFERENCES

- de Wit M, Aouad K, Elhai M, *et al.* EULAR updated recommendations for the involvement of patient research partners in rheumatology research. 2023.
- Taylor J, Dekker S, Jurg D, *et al.* Making the patient voice heard in a research consortium: experiences from an EU project (IMI-APPROACH). *Res Involv Engagem* 2021;7:24.
- Birch R, Simons G, Wähämaa H, *et al.* Development and formative evaluation of patient research partner involvement in a multi-disciplinary European translational research project. *Res Involv Engagem* 2020;6:6.
- de Wit M, Cooper C, Tugwell P, *et al.* Practical guidance for engaging patients in health research, treatment guidelines and regulatory processes: results of an expert group meeting organized by the World Health Organization (WHO) and the European society for clinical and economic aspects of osteoporosis, osteoarthritis and musculoskeletal diseases (ESCEO). *Aging Clin Exp Res* 2019;31:905–15.
- de Wit M, Adebajo A. Unique role of rheumatology in establishing collaborative relationships in research. Past, present and future of patient engagement. *Ann Rheum Dis* 2019;78:293–6.
- Schöpf AC, Schlöf M, Amos T, *et al.* Development and formative evaluation of a communication skills training program for persons with rheumatic and musculoskeletal diseases. *Health Commun* 2019;34:680–8.
- Pollock J, Raza K, Pratt AG, *et al.* Patient and researcher perspectives on facilitating patient and public involvement in rheumatology research. *Musculoskeletal Care* 2017;15:395–9.
- O'Sullivan DP, Steinkoenig I. GRAPPA patient research partner network: update to the GRAPPA 2020 annual meeting. *J Rheumatol* 2021;97:64.
- Nikiphorou E, Alunno A, Carmona L, *et al.* Patient-physician collaboration in rheumatology: a necessity. *RMD Open* 2017;3:e000499.
- EMA. Getting involved. 2018. Available: <https://www.ema.europa.eu/en/partners-networks/patients-consumers/getting-involved> [Accessed 18 Sep 2023].
- Costa Alencar AB, Selig WKD, Geissler J, *et al.* Adopting recommendations for implementing patient involvement in cancer research: a Funder's approach. *Res Involv Engagem* 2023;9:6.
- ESC patient engagement. Available: <https://www.escardio.org/The-ESC/What-we-do/esc-patient-engagement> [Accessed 18 Sep 2023].
- Diabetes UK. Patient and public involvement (PPI) in your study. Available: <https://www.diabetes.org.uk/research-for-researchers/apply-for-a-grant/help-with-involving-participants> [Accessed 18 Sep 2023].
- de Wit MPT, Berlo SE, Aanerud GJ, *et al.* European League against rheumatism recommendations for the inclusion of patient representatives in scientific projects. *Ann Rheum Dis* 2011;70:722–6.
- Cheung PP, de Wit M, Bingham CO 3rd, *et al.* Recommendations for the involvement of patient research partners (PRP) in OMERACT working groups. A report from the OMERACT 2014 working group on PRP. *J Rheumatol* 2016;43:187–93.
- OMERACT patient research partner network. Available: <https://omeractprnetwork.org/> [Accessed 18 Sep 2023].
- FOREUM – involving PRP. Available: https://www.foreum.org/involving_prp_cfm [Accessed 18 Sep 2023].
- GRAPPA Network. Patient research partners. Available: <https://www.grappanetwork.org/prp-network/> [Accessed 9 Oct 2023].
- Kirwan JR, de Wit M, Frank L, *et al.* Emerging guidelines for patient engagement in research. *Value in Health* 2017;20:481–6.
- Belton J, Hoens A, Scott A, *et al.* Patients as partners in research: it's the right thing to do. *J Orthop Sports Phys Ther* 2019;49:623–6.
- Goel N. Enhancing patient research partner engagement: research in psoriatic arthritis. *Best Pract Res Clin Rheumatol* 2021;35:101685.
- Mikdashi J. The meaningful role of patients, and other stakeholders in clinical practice guideline development. *Rheum Dis Clin North Am* 2022;48:691–703.
- de Wit M, Abma T, Koelewijn-van Loon M, *et al.* Involving patient research partners has a significant impact on outcomes research: a responsive evaluation of the International OMERACT conferences. *BMJ Open* 2013;3:e002241.
- de Wit M, Abma T, Koelewijn-Van Loon M, *et al.* Facilitating and inhibiting factors for long-term involvement of patients at outcome conferences—lessons learnt from a decade of collaboration in OMERACT: a qualitative study. *BMJ Open* 2013;3:e003311.

- 25 de Wit MPT, Koenders MI, Neijland Y, *et al.* Patient involvement in basic rheumatology research at Nijmegen: a three year's responsive evaluation of added value, pitfalls and conditions for success. *BMC Rheumatol* 2022;6:66.
- 26 Studenic P, Sekhon M, Carmona L, *et al.* Unmet need for patient involvement in rheumatology registries and observational studies: a mixed methods study. *RMD Open* 2022;8:e002472.
- 27 Goel N. Conducting research in psoriatic arthritis: the emerging role of patient research partners. *Rheumatology (Oxford)* 2020;59:i47–55.
- 28 Costello W, Dorris E. Laying the groundwork: building relationships for public and patient involvement in pre-clinical paediatric research. *Health Expect* 2020;23:96–105.
- 29 Tunis SR, Maxwell LJ, Graham ID, *et al.* Engaging stakeholders and promoting uptake of OMERACT core outcome instrument SETS. *J Rheumatol* 2017;44:1551–9.
- 30 EULAR. Project grant application. Available: <https://www.eular.org/project-grant-application> [Accessed 9 Oct 2023].
- 31 Long HA, French DP, Brooks JM. Optimising the value of the critical appraisal skills programme (CASP) tool for quality appraisal in qualitative evidence synthesis. *Research Methods in Medicine & Health Sciences* 2020;1:31–42.
- 32 Hong QN, Pluye P, Fàbregues S, *et al.* Improving the content validity of the mixed methods appraisal tool: a modified E-Delphi study. *J Clin Epidemiol* 2019;111:49–59.
- 33 Hsieh H-F, Shannon SE. Three approaches to qualitative content analysis. *Qual Health Res* 2005;15:1277–88.
- 34 de Souza S, Johansson EC, Karlfeldt S, *et al.* Patient and public involvement in an international rheumatology translational research project: an evaluation. *BMC Rheumatol* 2022;6:83.
- 35 de Wit M, Kirwan JR, Tugwell P, *et al.* Successful stepwise development of patient research partnership: 14 years' experience of actions and consequences in outcome measures in rheumatology (OMERACT). *Patient* 2017;10:141–52.
- 36 de Wit M, Campbell W, FitzGerald O, *et al.* Patient participation in psoriasis and psoriatic arthritis outcome research: a report from the GRAPPA 2013 annual meeting. *J Rheumatol* 2014;41:1206–11.
- 37 Carr ECJ, Patel JN, Ortiz MM, *et al.* Co-design of a patient experience survey for arthritis central intake: an example of meaningful patient engagement in healthcare design. *BMC Health Serv Res* 2019;19:355.
- 38 Del Gaizo V, Kohlheim M. Patient engagement in pediatric rheumatology research. *Rheum Dis Clin North Am* 2022;48:1–13.
- 39 Haribhai-Thompson J, Dalbeth N, Stewart S, *et al.* Involving people with lived experience as partners in musculoskeletal research: lessons from a survey of Aotearoa/New Zealand musculoskeletal researchers. *J Orthop Sports Phys Ther* 2022;52:307–11.
- 40 Fox G, Fergusson DA, Daham Z, *et al.* Patient engagement in preclinical laboratory research: a scoping review. *EBioMedicine* 2021;70:103484.
- 41 Golenya R, Chloros GD, Panteli M, *et al.* How to improve diversity in patient and public involvement. *Br J Hosp Med (Lond)* 2021;82:1–8.
- 42 de Wit M, Campbell W, Coates LC, *et al.* Let's talk about inclusion: a report on patient research partner involvement in the GRAPPA 2015 annual meeting. *J Rheumatol* 2016;43:970–3.
- 43 de Wit M, Guillemin F, Grimm S, *et al.* Patient engagement in health technology assessment (HTA) and the regulatory process: what about rheumatology? *RMD Open* 2020;6:e001286.
- 44 Parsons S, Thomson W, Cresswell K, *et al.* What do young people with rheumatic conditions in the UK think about research involvement? A qualitative study. *Pediatr Rheumatol Online J* 2018;16:35.
- 45 Schöpf-Lazzarino AC, Böhm P, Garske U, *et al.* Involving patients as research partners exemplified by the development and evaluation of a communication-skills training programme (KOKOS-Rheuma). *Z Rheumatol* 2021;80:132–9.
- 46 Wang H, Stewart S, Darlow B, *et al.* Patient research partner involvement in rheumatology clinical trials: analysis of journal articles 2016–2020. *Ann Rheum Dis* 2021;80:1095–6.
- 47 Young K, Kaminstein D, Olivos A, *et al.* Patient involvement in medical research: what patients and physicians learn from each other. *Orphanet J Rare Dis* 2019;14:21.
- 48 Leese J, Macdonald G, Kerr S, *et al.* 'Adding another spinning plate to an already busy life'. benefits and risks in patient partner-researcher relationships: a qualitative study of patient partners' experiences in a Canadian health research setting. *BMJ Open* 2018;8:e022154.
- 49 Richards DP, Cobey KD, Proulx L, *et al.* Identifying potential barriers and solutions to patient partner compensation (payment) in research. *Res Involv Engagem* 2022;8:7.
- 50 Schoemaker CG, Richards DP, de Wit M. Matching researchers' needs and patients' contributions: practical tips for meaningful patient engagement from the field of rheumatology. *Ann Rheum Dis* 2023;82:312–5.
- 51 Morin SN, Djekic-Ivankovic M, Funnell L, *et al.* Patient engagement in clinical guidelines development: input from >1000 members of the Canadian osteoporosis patient network. *Osteoporos Int* 2020;31:867–74.
- 52 Leese J, Kerr S, McKinnon A, *et al.* Evolving patient-researcher collaboration: an illustrative case study of a patient-led knowledge translation event. *J Particip Med* 2017;9:e13.
- 53 Esen E, Gnanenthiran S, Lunt L, *et al.* The your rheum story: involvement of young people in rheumatology research. *BMC Rheumatol* 2022;6:43.
- 54 Van der Elst K, De Cock D, Bangels L, *et al.* 'More than just Chitchat': a qualitative study concerning the need and potential format of a peer mentor programme for patients with early rheumatoid arthritis. *RMD Open* 2021;7:e001795.
- 55 O'Sullivan DP, Steinkoenig I. GRAPPA patient research partner network: update to the GRAPPA 2020 annual meeting. *J Rheumatol Suppl* 2021;97:64.
- 56 Gossec L, de Wit M, Kiltz U, *et al.* A patient-derived and patient-reported outcome measure for assessing psoriatic arthritis: elaboration and preliminary validation of the psoriatic arthritis impact of disease (Psaid) questionnaire, a 13-country EULAR initiative. *Ann Rheum Dis* 2014;73:1012–9.
- 57 Elliott RS, Taylor E, Ainsworth J, *et al.* Improving communication of the concept of 'treat-to target' in childhood lupus: a public and patient (PPI) engagement project involving children and young people. *BMC Rheumatol* 2022;6:69.
- 58 Lyng KD, Larsen JB, Birnie KA, *et al.* Participatory research: a priority setting partnership for chronic musculoskeletal pain in Denmark. *Scand J Pain* 2023;23:402–15.
- 59 Goodman SM, Miller AS, Turgunbaev M, *et al.* Clinical practice guidelines: incorporating input from a patient panel. *Arthritis Care Res (Hoboken)* 2017;69:1125–30.
- 60 Shoop-Worrall SJW, Cresswell K, Bolger I, *et al.* Nothing about us without us: involving patient collaborators for machine learning applications in rheumatology. *Ann Rheum Dis* 2021;80:1505–10.
- 61 Jongma KR, Milota MM. Establishing a multistakeholder research agenda: lessons learned from a James LIND alliance partnership. *BMJ Open* 2022;12:e059006.
- 62 Schoemaker CG, Armbrust W, Swart JF, *et al.* Dutch juvenile idiopathic arthritis patients, carers and clinicians create a research agenda together following the James LIND alliance method: a study protocol. *Pediatr Rheumatol Online J* 2018;16:57.
- 63 Goel N, O'Sullivan D, de Wit M, *et al.* The patient research partner network matures: a report from the GRAPPA 2017 annual meeting. *J Rheumatol Suppl* 2018;94:52–3.
- 64 Pauling JD, Frech TM, Domsic RT, *et al.* Patient participation in patient-reported outcome instrument development in systemic sclerosis. *Clin Exp Rheumatol* 2017;35 Suppl 106:184–92.
- 65 Goel N, O'Sullivan D, Steinkoenig I, *et al.* Tackling patient centricity: a report from the GRAPPA 2016 annual meeting. *J Rheumatol* 2017;44:703–5.
- 66 Bywall KS, Esbensen BA, Lason M, *et al.* Functional capacity vs side effects: treatment attributes to consider when Individualising treatment for patients with rheumatoid arthritis. *Clin Rheumatol* 2022;41:695–704.
- 67 Brett J, Staniszewska S, Simeria I, *et al.* Reaching consensus on reporting patient and public involvement (PPI) in research: methods and lessons learned from the development of reporting guidelines. *BMJ Open* 2017;7:e016948.
- 68 Helliwell PS, Gladman DD, Gottlieb AB. Prologue: 2016 annual meeting of the group for research and assessment of psoriasis and psoriatic arthritis (GRAPPA). *J Rheumatol* 2017;44:658–60.
- 69 PARE-PRP-2326. Available: <https://esor.eular.org/enrol/index.php?id=398> [Accessed 16 Oct 2023].
- 70 EULAR. PARE patient research partners. Available: <https://www.eular.org/pare-patient-research-partners> [Accessed 16 Oct 2023].

Online supplementary material

Supplementary Table 1. Search strategy used for PRP involvement in rheumatology research

Keywords	Search strategy
Patient research partners	("patient research partner"[All Fields] OR "patient research partners"[All Fields] OR "patient research"[All Fields] OR "patient partner"[All Fields] OR "patient partners"[All Fields] OR "patient participation"[All Fields] OR "patient involvement"[All Fields] OR "patient and public involvement"[All Fields] OR "patients and public involvement"[All Fields] OR "user involvement"[All Fields] OR "users involvement"[All Fields] OR "lay member"[All Fields] OR "lay members"[All Fields] OR "patient advisory group"[All Fields] OR "patient engagement"[All Fields] OR "patient organisation"[All Fields] OR "patient organisations"[All Fields] OR "patient organization"[All Fields] OR "patient advisor"[All Fields] OR "patient advisors"[All Fields] OR "patient advocacy"[All Fields] OR "patient advocate"[All Fields] OR "patient expert"[All Fields] OR "patient experts"[All Fields] OR "participatory research" [All Fields])
Rheumatology	AND ((rheumat* OR (rheumatology[MeSH Terms]) OR ("rheumatic diseases"[MeSH Terms]) OR (rheumatic disease[MeSH Terms]) OR (arthritis[MeSH Terms]) OR (musculoskeletal disease[MeSH Terms]) OR (musculoskeletal diseases[MeSH Terms]))
Date	AND (2017:2023[pdat])
Language	AND (English[Language]).

Supplementary Table 2: Overview of PRP Involvement across Rheumatology and other Key Specialties Websites.

Websites	Specialty	Key findings on PRP involvement
EULAR www.eular.org/pare-patient-research-partners	Rheumatology	<ul style="list-style-type: none"> Strongly endorses the involvement of PRPs in research projects related to RMDs. Advocates for the establishment and use of the PARE network, offering guiding documents to facilitate PRP inclusion in research endeavors.
GRAPPA www.grappanetwork.org/prp-network/	Rheumatology	<ul style="list-style-type: none"> Established a PRP Network in 2012, formalized by 2017, to actively include patient perspectives in the development of guidelines and recommendations for psoriatic disease. Produced a "Patient's Guide to Treatments for Psoriatic Arthritis," authored by members of GRAPPA's PRP team, aimed at both patients and healthcare professionals. Collaborates with organizations like OMERACT and IDEOM to further incorporate patient perspectives in research and guidelines.
OMERACT www.omeractprpnetwork.org	Rheumatology	<ul style="list-style-type: none"> Prioritizes the experiential knowledge of PRPs, explicitly valuing their contributions to research. Provides comprehensive recommendations for PRP involvement, including identification based on experiential knowledge and language skills, and discusses methods to support PRPs, like offering tailored information and meeting invitations.
ACR www.rheumatology.org	Rheumatology	<ul style="list-style-type: none"> Runs a Patient Perspectives program and ensures that PRPs are involved in the formulation of clinical guidelines and recommendations. Not explicit information about PRP involvement found on their website.
FOREUM www.foreum.org/involving_prp_of_m	Rheumatology	<ul style="list-style-type: none"> Incorporates PRPs in its Executive and Scientific committees. Strongly recommends the participation of PRPs in clinical research projects and suggests referring to EULAR documents for best practices, aiming to improve the relevance, quality, and validity of research.
European society of cardiology www.escardio.org/The-ESC/What-we-do/esc-patient-engagement	Cardiology	<ul style="list-style-type: none"> Establishment of the 2018 ESC patient forum with involvement of patients representatives : <ul style="list-style-type: none"> - in training of physicians - in education of patients - in development of guidelines

		<ul style="list-style-type: none"> • Participation of patient representative in meetings (workshops for patients, participation in congress as speakers and co-chairs) • Patient representatives are co-authors and reviewers of papers.
British cardiovascular society www.britishcardiosociet y.org/about/research	Cardiology	<ul style="list-style-type: none"> • Establishment of the BHF Clinical research Collaborative • Promotion of PPI in grant application
European Society for Medical Oncology www.esmo.org	Oncology	<ul style="list-style-type: none"> • Establishment of the patient advocacy track • Involvement of patient representatives in education of the patients • Participation of patient representatives in meetings/congresses
Diabetes UK www.diabetes.org.uk/profession als/resources/shared-practice/patient-and-public-involvement	Endocrinology	<ul style="list-style-type: none"> • Promotion of PPI in grant application • Development of guidelines for researchers to involve PPI • Review of the applications by the grant advisory panel
Food and Drug Administration (FDA) https://www.fda.gov/patients/learn-about-fda-patient-engagement	Regulatory agency	<ul style="list-style-type: none"> • Patient focused drug development (PFDD) • Patient representative program • Patient Engagement Advisory Committee (PEAC) and Patient Engagement Collaborative (PEC) • Patient listening session program
European Medicines Agency (EMA) Patients and consumers European Medicines Agency (europa.eu)	Regulatory agency	<ul style="list-style-type: none"> • Comprehensive framework for engagement • Collaboration with both individual patient experts and patient organisations. • Patients' and consumer working party • Public engagement department • Patients are members of the management board and scientific committees • Training for patient participants is provided by a comprehensive set of videos and in-house one-day training sessions.

ACR: American College of Rheumatology, EMA: European Medicines Agency, EULAR: European Alliance of Rheumatology Associations, FDA: Food and Drug Administration, FOREUM: Foundation for Research in Rheumatology, GRAPPA: Group for Research and Assessment of Psoriasis and Psoriatic Arthritis, OMERACT: Outcome Measures in Rheumatology, OARSI: Osteoarthritis Research Society International, NIHR: National Institute for Health Research.

The following websites were searched but did not provide any information on the involvement of PRPs: OARSI, American heart association, American society of clinical oncology, American diabetes association, European Foundation for the Study of Diabetes, British Society for Paediatric endocrinology and diabetes.

Supplementary Table 3. Specific website searches in patient and public involvement about training, involvement in grants and remuneration of PRPs.

INVOLVE UK guidelines (by NIHR) www.invo.org.uk	Patient and public involvement	<ul style="list-style-type: none"> • Development of guidelines to involve PPI in research projects
EUPATI www.eupati.eu	Patient and public involvement	<ul style="list-style-type: none"> • EUPATI Fundamentals: training (co-designed and co-delivered by patients and pharmaceutical experts) about Patient Engagement addressed to professionals in academia and pharmaceutical industry • EUPATI training for patients and patient representatives. • Two EUPATI Patient Expert Training Courses (14 months each)
Short guide on patient partnerships in rare disease research projects https://www.ejprarediseases.org/our-actions-and-services/patients-in-research/	Orphan diseases	<ul style="list-style-type: none"> • Development of a guide to include PPI in research projects • Suggestion that a PRP should be ideally affiliated to a patient organisation or a patient group.

EUPATI: European Patients' Academy on Therapeutic Innovation, PPI (Patient and public involvement), UK: United Kingdom.

Supplementary Table 4. A total of 53 papers included in the Systematic literature review.

First author (name)	Year of publication	Title	Journal	Study type	Quality assessment
Studenic P et al [26]	2022	Unmet need for patient involvement in rheumatology registries and observational studies: a mixed methods study.	RMD open	Mixed methods	MMAT: High quality
Haribhai-Thompson J et al [40]	2022	Involving People with Lived Experience as Partners in Musculoskeletal Research: Lessons From a Survey of Aotearoa/New Zealand Musculoskeletal Researchers.	The Journal of orthopaedic and sports physical therapy	Cross-sectional	CASP: High quality
de Wit M et al [25]	2022	Patient involvement in basic rheumatology research at Nijmegen: a three year's responsive evaluation of added value, pitfalls and conditions for success.	BMC rheumatology	Mixed methods	MMAT: High quality
de Souza S et al [35]	2022	Patient and public involvement in an international rheumatology translational research project: an evaluation.	BMC rheumatology	Qualitative	CASP: High quality
Elliott RS et al [58]	2022	Improving communication of the concept of 'treat-to target' in childhood lupus: a public and patient (PPI) engagement project involving children and young people.	BMC rheumatology	Mixed methods	MMAT: High quality
Fox et al [41]	2021	Patient engagement in preclinical laboratory research: A scoping review	EBioMedicine	Scoping review	CASP: High quality
Costello W et al [28]	2020	Laying the groundwork: Building relationships for public and patient involvement in pre-clinical paediatric research.	Health expectations : an international journal of public participation in health care and health policy	Qualitative	CASP: High quality
Birch et al [3]	2020	Development and formative evaluation of patient research partner involvement in a multi-disciplinary European translational research project	BMC Research involvement and engagement	Mixed methods	MMAT: High quality

Morin SN et al [52]	2020	Patient engagement in clinical guidelines development: input from >â€%1000 members of the Canadian Osteoporosis Patient Network.	Osteoporosis international : a journal established as result of cooperation between the European Foundation for Osteoporosis and the National Osteoporosis Foundation of the USA	Mixed methods	MMAT: High quality
Young K et al [48]	2019	Patient involvement in medical research: what patients and physicians learn from each other.	BMC, Orphanet journal of rare diseases	Qualitative	CASP: High quality
Parsons S et al [45]	2018	What do young people with rheumatic conditions in the UK think about research involvement? A qualitative study.	Pediatric rheumatology online journal	Qualitative	CASP: High quality
Leese J et al [49]	2018	Adding another spinning plate to an already busy life'. Benefits and risks in patient partner-researcher relationships: a qualitative study of patient partners' experiences in a Canadian health research setting	BMJ open	Qualitative	CASP: High quality
Gossec L et al [57]	2014	A patient-derived and patient-reported outcome measure for assessing psoriatic arthritis: elaboration and preliminary validation of the Psoriatic Arthritis Impact of Disease (PsAID) questionnaire, a 13-country EULAR initiative	ARD	Cross-sectional	CASP: high quality
de Wit M et al [23]	2013	Involving patient research partners has a significant impact on outcomes research: a responsive evaluation of the international OMERACT conferences.	BMJ open	Qualitative	CASP: high quality
de Wit M et al [24]	2013	Facilitating and inhibiting factors for long-term involvement of patients at outcome conferences-- lessons learnt from a decade of collaboration in OMERACT: a qualitative study.	BMJ open	Qualitative	CASP: high quality
Lyng KD et al [59]	2022	Participatory research: a Priority Setting Partnership for chronic musculoskeletal pain in Denmark.	Scandinavian journal of pain	Mixed methods	MMAT: Medium quality

Carr ECJ et al [38]	2019	Co-design of a patient experience survey for arthritis central intake: an example of meaningful patient engagement in healthcare design.	BMC health services research	Qualitative	CASP: Medium quality
Wang H et al [47]	2021	Patient research partner involvement in rheumatology clinical trials: analysis of journal articles 2016-2020.	Annals of the rheumatic diseases	Systematic literature review	CASP: Low quality
Pauling JD et al [65]	2017	Patient participation in patient-reported outcome instrument development in systemic sclerosis.	Clinical and experimental rheumatology	Systematic literature review	CASP: Low quality
Schoemaker CG et al [51]	2023	Matching researchers' needs and patients' contributions: practical tips for meaningful patient engagement from the field of rheumatology.	Annals of the rheumatic diseases	Qualitative	NS
Richards D et al [50]	2022	Identifying potential barriers and solutions to patient partner compensation (payment) in research	BMC Research involvement and engagement	Qualitative	NS
Del Gaizo V et al [39]	2022	Patient Engagement in Pediatric Rheumatology Research.	Rheumatic diseases clinics of North America	Opinion article	NS
Mikdashi J [22]	2022	The Meaningful Role of Patients, and Other Stakeholders in Clinical Practice Guideline Development.	Rheumatic diseases clinics of North America	Opinion article	NS
Jongsma KR et al [62]	2022	Establishing a multistakeholder research agenda: lessons learned from a James Lind Alliance Partnership.	BMJ open	Opinion article	NS
Bywall KS et al [67]	2022	Functional capacity vs side effects: treatment attributes to consider when individualising treatment for patients with rheumatoid arthritis.	Clinical rheumatology	Qualitative not focused on PRP	NS
Ecem Esen et al [54]	2022	The Your Rheum story: involvement of young people in rheumatology research	BMC rheumatology	Case study	NS
Goel [21]	2021	Enhancing patient research partner engagement: Research in psoriatic arthritis	Best Practice & Research Clinical Rheumatology	Opinion article	NS
Schöpf-Lazzarino AC et al [46]	2021	Involving patients as research partners exemplified by the development and evaluation of a communication-skills	Zeitschrift für Rheumatologie	Case study	NS

		training programme (KOKOS-Rheuma).			
Van der Elst K et al [55]	2021	More than just chitchat': a qualitative study concerning the need and potential format of a peer mentor programme for patients with early rheumatoid arthritis.	RMD open	Qualitative, not focused on PRP	NS
Shoop-Worrall SJW et al [61]	2021	Nothing about us without us: involving patient collaborators for machine learning applications in rheumatology.	Annals of the rheumatic diseases	Opinion article	NS
O'Sullivan DP et al [56]	2021	GRAPPA Patient Research Partner Network: Update to the GRAPPA 2020 Annual Meeting.	The Journal of rheumatology	Meeting report	NS
Golenya R et al [42]	2021	How to improve diversity in patient and public involvement.	British journal of hospital medicine (London, England : 2005)	Opinion article	NS
Taylor J et al [2]	2021	Making the patient voice heard in a research consortium: experiences from an EU project (IMI-APPROACH).	Research involvement and engagement	Case study	NS
Goel [27]	2020	Conducting research in psoriatic arthritis: the emerging role of patient research partners	Rheumatology (Oxford, England)	Opinion article	NS
de Wit M et al [44]	2020	Patient engagement in health technology assessment (HTA) and the regulatory process: what about rheumatology?	RMD open	Opinion article	NS
de Wit M et al [4]	2019	Practical guidance for engaging patients in health research, treatment guidelines and regulatory processes: results of an expert group meeting organized by the World Health Organization (WHO) and the European Society for Clinical and Economic Aspects of Osteoporosis, Osteoarthritis and Musculoskeletal Diseases (ESCEO).	Aging clinical and experimental research	Recommendations	NS
de Wit M et al [5]	2019	Unique role of rheumatology in establishing collaborative relationships in research.	Annals of the rheumatic diseases	Opinion article	NS

		Past, present and future of patient engagement.			
Schopf AC et al [6]	2019	Development and Formative Evaluation of a Communication Skills Training Program for Persons with Rheumatic and Musculoskeletal Diseases.	Health communication	Case study	NS
Belton J et al [20]	2019	Patients as Partners in Research: It's the Right Thing to Do.	The Journal of orthopaedic and sports physical therapy	Opinion article	NS
Schoemaker CG et al [63]	2018	Dutch juvenile idiopathic arthritis patients, carers and clinicians create a research agenda together following the James Lind Alliance method: a study protocol.	Pediatric rheumatology online journal	Study protocol	NS
Goel N et al [64]	2018	The Patient Research Partner Network Matures: A Report from the GRAPPA 2017 Annual Meeting.	The Journal of rheumatology.	Meeting report	NS
Helliwell PS et al [69]	2017	Prologue: 2016 Annual Meeting of the Group for Research and Assessment of Psoriasis and Psoriatic Arthritis (GRAPPA).	The Journal of rheumatology	Meeting report	NS
Goel N et al [66]	2017	Tackling Patient Centricity: A Report from the GRAPPA 2016 Annual Meeting.	The Journal of rheumatology	Meeting report	NS
Goodman SM et al [60]	2017	Clinical Practice Guidelines: Incorporating Input From a Patient Panel.	Arthritis care & research	Report	NS
de Wit M et al [36]	2017	Successful Stepwise Development of Patient Research Partnership: 14 Years' Experience of Actions and Consequences in Outcome Measures in Rheumatology (OMERACT).	The patient	Opinion article	NS
Pollock J et al [7]	2017	Patient and researcher perspectives on facilitating patient and public involvement in rheumatology research.	Musculoskeletal care	Meeting report	NS
Kirwan JR et al [19]	2017	Emerging Guidelines for Patient Engagement in Research.	Value in health: the journal of the International Society for Pharmacoeconomics and Outcomes Research	Recommendations	NS
Brett J et al [68]	2017	Reaching consensus on reporting patient and public involvement (PPI) in	BMJ open	Recommendations	NS

		research: methods and lessons learned from the development of reporting guidelines.			
Leese J et al [53]	2017	Evolving Patient-Researcher Collaboration: An Illustrative Case Study of a Patient-Led Knowledge Translation Event.	Journal of participatory medicine	Meeting report	NS
Tunis SR et al [29]	2017	Engaging Stakeholders and Promoting Uptake of OMERACT Core Outcome Instrument Sets.	The Journal of rheumatology	Meeting report	NS
Cheung PP et al [15]	2016	Recommendations for the Involvement of Patient Research Partners (PRP) in OMERACT Working Groups. A Report from the OMERACT 2014 Working Group on PRP.	The Journal of rheumatology	Recommendations	NS
de Wit M et al [43]	2016	Let's Talk about Inclusion: A Report on Patient Research Partner Involvement in the GRAPPA 2015 Annual Meeting.	The Journal of rheumatology	Meeting report	NS
de Wit M et al [37]	2014	Patient participation in psoriasis and psoriatic arthritis outcome research: a report from the GRAPPA 2013 Annual Meeting.	The Journal of rheumatology	Meeting report	NS

NS: Not scored.

Cross-sectional observational studies were assessed using the STROBE evaluation form; mixed methods studies using the mixed methods appraisal tool (MMAT); qualitative studies using the CASP checklist and reviews (SLR and scoping reviews) using the PRISMA. Qualitative studies without a focus on PRP were not assessed for quality (n=2).

Supplementary Table 5. Type of studies included in the SLR.

Type of study	n (%)	Quality assessment
Qualitative	12 (23%)	Yes*
Mixed methods	6 (11%)	Yes
Review (SLR or scoping)	3 (6%)	Yes
Cross-sectional	2(4%)	Yes
Opinion	11(21%)	No
Report	10 (19%)	No
Recommendation	4(8%)	No
Case study	4(8%)	No
Study protocol	1(2%)	No

*excluding 2 qualitative articles not focused on PRPs.

Supplementary Table 6. Analysis of qualitative data according to the principles of thematic content analysis.

The qualitative analysis involved extracting pertinent information from the text of each study and developing a coding scheme to categorize the approaches to patient involvement. Extracted data were then organized into conceptual categories.

	Extracted data ordered in conceptual categories
a	a narrative description detailing the roles, activities, added values, experiences, and feedback from PRP in the included studies
b	a narrative overview of the PRP selection process, the types of training provided, and the recognition they received
c	a narrative account of the researcher's role and the training they underwent
d	a narrative overview of the potential PRP coordinator's role, along with details on the evaluation and monitoring of PRP involvement
e	a synthesis of the identified barriers and facilitators to patient involvement, encompassing contextual factors and the strategies used within the included studies

Supplementary Table 7. Key excerpts on barriers and strategies to enhance PRP involvement.

Concept	Barriers	Excerpts on barriers	Strategies to Enhance Patient Involvement	Excerpts on strategies
Emotional and Personal Factors	<ul style="list-style-type: none"> Emotional burden Fatigue Need to accommodate PRP needs (physical and knowledge, level of involvement) Lack of trust Time and budget constraints 	<p><i>“Resources constraints may limit the capability to train and accommodate the needs of the involved patients.” [22]</i></p>	<ul style="list-style-type: none"> Provide a supportive environment Provide flexibility and accessible accommodation Allocate adequate resources Practice active listening Recognize PRP contributions 	<p><i>“Creating a safe space where patient partners and researchers feel comfortable to collaborate” [41]</i></p> <p><i>“This recognition and appreciation for mutual learning and respect built on the established research partnership, and made reflecting on the challenges more comfortable.”[53]</i></p>
Communication and Relationship	<ul style="list-style-type: none"> Feeling unheard Power imbalance Inconsistent and poor communication Loss of confidentiality Use of medical jargon 	<p><i>“Lack of genuine engagement feeling may also arise from inadequate patient team diversity and the absence of patient coleadership and power sharing, and not leaving rooms for discussions or allowing time for authentic partnership”. [22]</i></p> <p><i>“The use of medical jargon and the nature of some of the discussion topics such as ethics applications may make it difficult for patients and other stakeholders to understand and follow what is being discussed” [22]</i></p>	<ul style="list-style-type: none"> Appoint a coordinator to facilitate PRP involvement Clarify patient roles and objectives Avoid complex medical terminology Exchange mutual expectations early at project initiation. Ensure open and transparent communication Ensure feedback and trust 	<p><i>“The assigned PC coordinator should take the lead in setting up meetings and ensuring that infrastructures such as video calling technology are accessible to PC members. In addition, the PC coordinator should, at the start of the project, mediate discussions to align expectations from all parties involved.” [2]</i></p> <p><i>“In describing the responsibilities and practical tasks undertaken, values and ethical considerations (eg, mutuality, understanding, respect and diversity) that underpin patient engagement in research are revealed (...)”[53]</i></p>
Training and Support	<ul style="list-style-type: none"> Lack of awareness about PRP involvement among researchers 	<p><i>“Lack of researcher training opportunities to guide meaningful</i></p>	<ul style="list-style-type: none"> Increase awareness about importance of PRP involvement 	<p><i>“(...) importance of providing training and educational resources to support and enhance patient</i></p>

	<ul style="list-style-type: none"> • Overburdening of PRPs • Inadequate training and support of PRP • Lack of resources and compensation of PRP 	<p><i>patient engagement". [41]</i></p> <p><i>"Insufficient researcher resources to support patient partners including time and budget restrictions" [41]</i></p>	<ul style="list-style-type: none"> • Allocate resources for patient engagement • Provide proper training tailored to PRP needs • Provide training and support to researchers • Appropriate recognition of PRP • Encourage PRPs to ask questions and express needs • Communicate workloads 	<p><i>involvement in research."</i>[26]</p> <p><i>"Three studies reported offering training sessions for researchers to facilitate patient engagement, which included exercises to improve communicating research to non-scientists." [41]</i></p>
Research Process and Pace	<ul style="list-style-type: none"> • Challenges to recruit PRPs • Time commitment for researchers • Anxiety about delays in projects • Higher demands on resources • Forced changes in working practice 	<p><i>"(...) working together in a joint intellectual effort (...) meant delays to the scheduled release of promotional materials, and contributed additional unanticipated hours that had not been bracketed into already busy schedules"[53]</i></p>	<ul style="list-style-type: none"> • Address recruitment challenges • Establish realistic timelines • Manage researcher and PRP time commitments • Build trust through open communication and demonstrate commitment to research progress 	<p><i>"It is desirable to estimate the expected time PRP are required to allocate for the project (e.g., 4 h/month over 6 months), with feasible timelines (e.g., feedback within 2 weeks)."[15]</i></p>
Collaboration and Engagement	<ul style="list-style-type: none"> • Lack of PRP diversity and representativeness challenges • Discrepancies in views • Uncertainty in incorporating patient experiences • Risk of sharing data prior to peer-review 	<p><i>"Aside from the challenges of identifying appropriate PRP, research teams may struggle with the lack of awareness of the need and impact of the PRP role, identification of diverse PRP (...)"[21]</i></p>	<ul style="list-style-type: none"> • Discuss representativeness and diversity of PRPs within research team • Involve PRPs from project inception and clearly define their roles upfront • Discuss and address discrepancies • Co-create PRP-contracts to ensure confidentiality 	<p><i>"Engagement of the patients early in the decision process is critical (...)"[22]</i></p> <p><i>"Whatever the nature of a project, it is essential that the patient voice is represented in early discussions when formulating the concept and idea."[2]</i></p> <p><i>"Goals and expectations should be discussed in the early stages of the project"[27]</i></p>

Supplementary Table 8. Key excerpts on activities and roles of PRPs.

Areas of involvement	Activities and roles of PRPs	Excerpts from the articles
Development/ Design	<ul style="list-style-type: none"> • Guidelines development [22], including voting on draft recommendations [24] • PRO development [5,21] • Grant application [3] • Survey / questionnaire development [2,3] • Interview guide [67] • Tools (educational or others) [3,4,58] • Patient facing materials (develop patient information sheets, pamphlets, glossary, distribution and use of booklet, website, poster) [3,6,43] • Developing or reviewing (recommendations) lay summaries [4,5] 	<p>“...patient engagement must continue if there is a public reason for the topic reconsideration and whether and when the guidelines require updates apart from new evidence.” [22]</p> <p>“The involvement of patients is (...) most tangible in the development of its international management recommendations for rheumatic diseases. According to its standardised operational procedures, involvement of PRPs is pivotal in every phase of the development project.” [5]</p> <p>“PRPs were involved in decisions regarding design of interview and focus group protocols, analysis of transcripts, draft language of items(...). This strategy of patient involvement ensures that PROs are grounded in patient data, have face and content validity and are comprehensive.” [5]</p> <p>“(...) full PRP participation in accordance with the EULAR recommendations has been the development of two patient-derived impacts of disease scores for rheumatoid arthritis and psoriatic arthritis. (elaboration and validation of composite indices)” [5]</p> <p>“Since 2012, EULAR develops public summaries of all its management recommendations that are freely available on their website. These summaries are produced with the active involvement of PRPs who take responsibility for checking relevance, comprehension and completeness. They also assist in reviewing summaries of scientific publications of the Annals of Rheumatology. Finally, three PRPs wrote a lay version of the GRAPPA treatment recommendations for psoriatic arthritis.” [5]</p> <p>“PRPs contributed actively to a number of research activities over the 4 year project, it included:</p> <ul style="list-style-type: none"> • Attending and contributing to annual scientific meetings and regular teleconferences • Development of a glossary resource (WP1–4) • Contributing to a meta-synthesis of qualitative literature on public perceptions of predictive testing [13, 14] (WP4) • Contributing to the development of interview schedules (including question setting) and the interpretation of qualitative data [12, 46] (WP4) • Contributing to the development of informational resources for those at risk (WP4) • Evaluating a web-based platform for the communication of risk information (WP4) • Developing a questionnaire for patients undergoing a lymph node biopsy procedure (WP3) • Developing informational resources for patients about RA such as the ‘Metaphor Project’ (a collaboration between Eva C Johansson (PRP) and Dr. Heidi Wähämaa) • Exploring the communication of scientific concepts via the use of metaphors and visual representations • Contributing to the project website (e.g. providing news items and reports, creating subtitles for videos) (WP4)

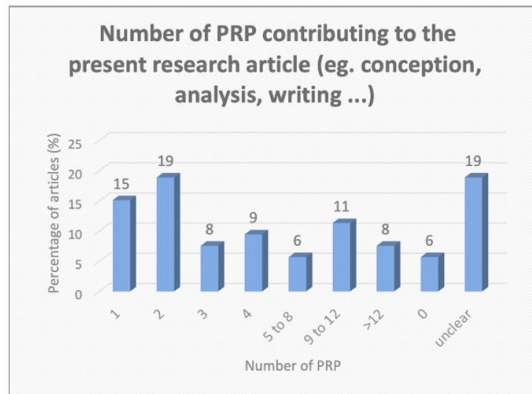
		<ul style="list-style-type: none"> • Developing lay summaries of EuroTEAM methods and findings (e.g. lay summary of metabolomics in EuroTEAM) (WP2) • Developing posters for dissemination at the European League Against Rheumatism (EULAR) Congress (WP1–4)” [3]
Leadership	<ul style="list-style-type: none"> • Establishing a patient association [5] 	<p>“Creaky Joints, a patient organisation founded by a person with RA, was one of the collaborating partners. (...) Creaky Joints also became a successful applicant of the second project with the objective of developing strong partnerships with trialists to conduct patient-centred comparative effectiveness research.” [5]</p>
Co-leadership role	<ul style="list-style-type: none"> • Member of steering committee [7] (eg. EULAR, FOREUM [17], GRAPPA [62]) • Member of working groups (eg. GRAPPA [8]) 	<p>“Offering advice as a member of a project steering group” [7]</p> <p>“The Glasgow Patient Involvement in Rheumatology Research (PIRR) group is in the early stages of its development with patient representation on clinical trial steering groups” [7]</p> <p>“Within the GRAPPA–Outcome Measures in Rheumatology (OMERACT) Core Set group, 3 PRPs worked on the steering committee, as well as additional PRPs were involved in the working subgroups; particularly in the Physical Function Working Subgroup.” [8]</p>
Coauthorship	<ul style="list-style-type: none"> • Writing research papers (co-authoring) [3] • Writing research protocol [57] 	<p>“Coauthorship is a recognition of the contributions made by patients, and is the ultimate proof of equal and meaningful partnerships”. [5]</p> <p>“getting involved in (...) writing book chapters and magazine articles; (...) writing conference abstracts, and contributing to academic papers” [54]</p>
Education	<ul style="list-style-type: none"> • Trainer at a training program [6] • Mentoring less-experienced PRPs [39] 	<p>“one full-day course was conducted by two patient PRPs and some trainings were conducted by a trained lay person and a patient PRP.” [6]</p> <p>“(...) patients and caregivers who have experience being engaged on a study team can effectively serve as mentors for patient families new to the role of research partner who can greatly benefit from their experiences.” [39]</p>
Planning	<ul style="list-style-type: none"> • Strategic planning [43,48] • Event planning: organize, communicate [53] • Interview schedules[3] 	<p>“PRP also created their own governance document and PRP handbook (outlining how the PRP network works within GRAPPA to fulfill expectations and achieve its own goals, including ethical considerations), approved by the GRAPPA executive committee”[21]</p>
Facilitation	<ul style="list-style-type: none"> • Session at a conference[54] • Held sessions in a meeting [69] 	<p>“...key youth development opportunities taken up by the young people include facilitating a session at a national rheumatology conference, presenting at 9 other conferences (7 national, 2 international)” [54]</p> <p>“(...) co-presenting at conferences” [54]</p>
Reviewer	<ul style="list-style-type: none"> • Grant application [2] • Research projects (ongoing) [5,59] • Summaries of scientific publications [5] 	<p>“Reviewing grant applications is an effective way for patients to influence research agendas. From its inception, the Foundation for Research in Rheumatology involves PRPs in its governing bodies and in the review of research proposals. Members of the Consumer Advisory Group (CAG) of the Australian and New Zealand Musculoskeletal Clinical Trials network operate as PRPs and review all trials from a patient’s perspective. Clinical trialists need to respond to their feedback before they are endorsed by the network.” [5]</p> <p>“They also assist in reviewing summaries of scientific publications of the Annals of Rheumatology.” [5]</p>

Recruitment	<ul style="list-style-type: none"> • Support recruitment of PRPs (to a project or association). [3] [6] [68] 	<p><i>"The patient partners, along with other patient organisations and charities, recruited nearly half of all participants for the Delphi survey" [68]</i></p> <p><i>"Additional PRPs were recruited via clinician researchers and PRPs' own networks, resulting in a panel of 9 PRPs" [3]</i></p>
Evaluation	<ul style="list-style-type: none"> • Training program [6] 	<p><i>" (...) the trainers (including PRPs) completed an evaluation form containing closed and open questions asking about the conduction of the training course as well as for suggestions regarding the preparation of the trainers in addition to the training itself." [6]</i></p>
Participation	<ul style="list-style-type: none"> • In dissemination of research results [3,7] • Attendance of conferences and scientific meetings [3,41] • In meetings[2], discussions [67] • In quality of care improvement programs [52,61] 	<p><i>"Patient representatives may as well post drafts of evidence summaries and conclusion for the public comments, which may improve guidelines awareness and implementation." [22]</i></p> <p><i>"Within GRAPPA, (...) the patient role has become more formalized, with patients attending the 2013 annual meeting and each subsequent annual meeting as PRP." [21]</i></p> <p><i>"...involved as patient-partners in peer-reviewed research and quality of care improvement programs." [52]</i></p>

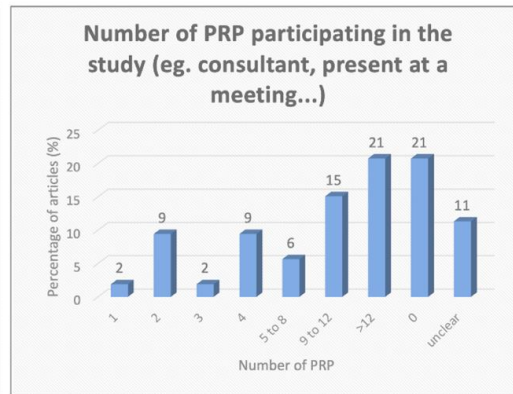
GRAPPA: Group for Research and Assessment of Psoriasis and Psoriatic Arthritis; PRO: Patient Reported Outcomes.

Supplementary Figure 1. Number of PRPs contributing or participating in the study.

A. Number of PRPs stated as coauthors



B. Number of PRPs contributing to the research project



A. Number of PRPs stated as coauthors**B. Number of PRPs contributing to the research project**