

BMJ Open The James Lind Alliance process approach: scoping review

Agnete Nygaard,^{1,2} Liv Halvorsrud,¹ Siv Linnerud,² Ellen Karine Grov,¹ Astrid Bergland¹

To cite: Nygaard A, Halvorsrud L, Linnerud S, *et al.* The James Lind Alliance process approach: scoping review. *BMJ Open* 2019;**9**:e027473. doi:10.1136/bmjopen-2018-027473

► Prepublication history for this paper is available online. To view these files, please visit the journal online (<http://dx.doi.org/10.1136/bmjopen-2018-027473>).

Received 05 November 2018
Revised 11 July 2019
Accepted 16 July 2019

ABSTRACT

Objective To summarise study descriptions of the James Lind Alliance (JLA) approach to the priority setting partnership (PSP) process and how this process is used to identify uncertainties and to develop lists of top 10 priorities.

Design Scoping review.

Data sources The Embase, Medline (Ovid), PubMed, CINAHL and the Cochrane Library as of October 2018.

Study selection All studies reporting the use of JLA process steps and the development of a list of top 10 priorities, with adult participants aged 18 years.

Data extraction A data extraction sheet was created to collect demographic details, study aims, sample and patient group details, PSP details (eg, stakeholders), lists of top 10 priorities, descriptions of JLA facilitator roles and the PSP stages followed. Individual and comparative appraisals were discussed among the scoping review authors until agreement was reached.

Results Database searches yielded 431 potentially relevant studies published in 2010–2018, of which 37 met the inclusion criteria. JLA process participants were patients, carers and clinicians, aged 18 years, who had experience with the study-relevant diagnoses. All studies reported having a steering group, although partners and stakeholders were described differently across studies. The number of JLA PSP process steps varied from four to eight. Uncertainties were typically collected via an online survey hosted on, or linked to, the PSP website. The number of submitted uncertainties varied across studies, from 323 submitted by 58 participants to 8227 submitted by 2587 participants.

Conclusions JLA-based PSP makes a useful contribution to identifying research questions. Through this process, patients, carers and clinicians work together to identify and prioritise unanswered uncertainties. However, representation of those with different health conditions depends on their having the capacity and resources to participate. No studies reported difficulties in developing their top 10 priorities.

INTRODUCTION

Over the past decade, patient and public involvement (PPI) has been highlighted worldwide in both health research agendas and the development of next-step research projects.¹ PPI has been defined as ‘experimenting with’ as opposed to ‘experimenting on’ patients or the public.² PPI allows patients

Strengths and limitations of this study

- This is the first scoping review of published studies using the James Lind Alliance (JLA) approach available with involvement of patients, carers and the public in the setting the research agenda.
- The weakest voices often lack representation, which could limit the generalisability of these priorities to these populations.
- Because a scoping review approach was used, the quality of the articles was not assessed prior to inclusion.
- We were not in contact with the JLA Coordinating Centre and search in all relevant literature, such as grey literature and studies, which do not described all steps of the JLA process, might have limited our results.
- A limitation of this scoping review was our inclusion of only English-language articles.

to actively contribute, through discussion, to decision-making regarding research design, acceptability, relevance, conduct and governance from study conception to dissemination.³ However, PPI may also involve active data collection, analysis and dissemination.⁴

Researchers have noted that involving healthcare service users, the public and patients improves research quality, relevance, implementation and cost-effectiveness; it also improves researchers’ understanding of and insight into the medical and social conditions they are studying,^{1,5} although such evidence is still relatively limited.⁴

The James Lind Alliance (JLA) is a UK-based non-profit initiative that was established in 2004. The JLA process is focused on bringing patients, carers and clinicians together, on an equal basis, in a priority setting partnership (PSP) to define and prioritise uncertainties relating to a specific condition.⁶ Hall *et al*⁷ note that the JLA aims to raise awareness among research funding groups about what matters most to both patients and clinicians, in order to ensure that clinical research is both relevant and beneficial to end users. According to the *JLA Guidebook*,⁶



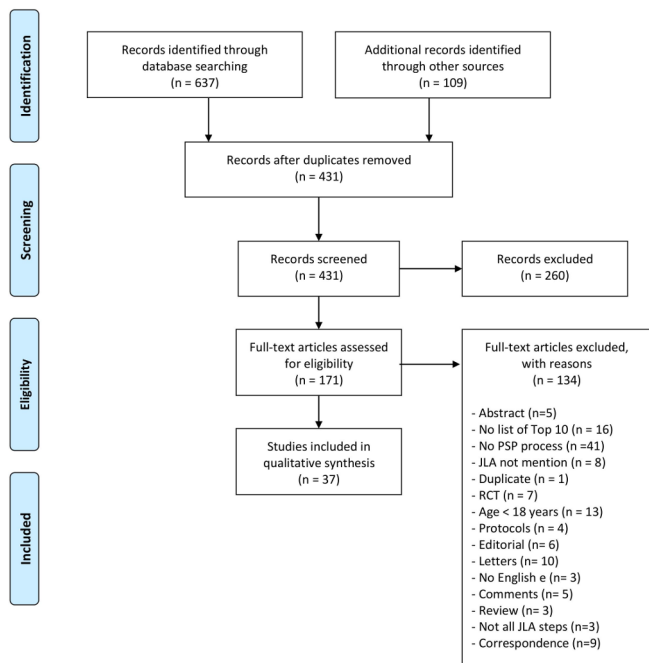
© Author(s) (or their employer(s)) 2019. Re-use permitted under CC BY-NC. No commercial re-use. See rights and permissions. Published by BMJ.

¹Faculty of Health Sciences, Oslo Metropolitan University, Oslo, Norway

²Center for Development of Institutional and Home Care, Lørenskog, Akershus

Correspondence to

Agnete Nygaard;
s98209@oslomet.no



From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Med* 6(7): e1000097. doi:10.1371/journal.pmed1000097

For more information, visit www.prisma-statement.org.

Figure 1 Flow diagram. JLA, James Lind Alliance; PSP, priority setting partnership; RCT, randomised controlled trial.

uncertainties and how to prioritise these are key features of the JLA process. The process begins by defining unanswered questions (ie, ‘uncertainties’) about the effects of treatment and healthcare—questions that cannot be adequately answered based on existing research evidence, such as reliable, up-to-date systematic reviews—and then prioritises the uncertainties based on their importance. The most recent version of the *JLA Guidebook* explains that many PSPs interpret the definition of treatment uncertainties broadly. They may interpret ‘treatments’ to include interventions such as care, support and diagnosis. This approach has been an important development and one that helps the JLA adapt to the changing health and care landscapes, as well as to the changing needs of its users.⁶

The JLA provides facilitation and guidance in the identification and prioritisation processes. This process forms part of a widening approach to PPI in research. The characteristics of the PSP process are (1) setting up a steering group to supervise all aspects of the study; (2) establishing a PSP; (3) assembling potential research questions; (4) processing, categorising, and summarising those research questions; and (5) determining the top 10 research priorities through an interim process and a final priority setting workshop using respondent ranking and consensus discussion. To ensure that all voices in the workshop are heard, the JLA supports an adapted nominal group technique (NGT) for PSPs when choosing their priorities. NGT is a well-established and well-documented approach to decision-making.⁶

Table 1 Criteria for inclusion and exclusion

Inclusion criteria	Exclusion criteria
<ul style="list-style-type: none"> ▶ All steps from JLA ▶ List of top 10 priorities ▶ Adults (aged >18 years or older) 	<ul style="list-style-type: none"> ▶ Unpublished literature ▶ Articles not written in English ▶ PSP without JLA ▶ JLA without PSP ▶ Protocols ▶ Errata ▶ Editorial ▶ Thesis ▶ Comments ▶ Review ▶ Guidelines ▶ Randomised controlled trials

JLA, James Lind Alliance; PSP, priority setting partnership.

To our knowledge, there is a gap in existing research given that no review has yet been published describing how the JLA approach is used to establish steering groups, set up PSPs, gather uncertainties, summarise uncertainties and determine the lists of top 10 priorities. Thus, the objective of this scoping review was to summarise study descriptions of the JLA approach to the PSP process, and how this process is used to identify uncertainties and develop lists of top 10 priorities.

- ▶ How do the studies describe the characteristics of the PSPs and, elaborating on aspects, how have they operationalised the JLA methods?
- ▶ How do the studies describe involvement of different user groups?
- ▶ What processes are used to gather and verify uncertainties?

METHODS

Identifying relevant studies

A systematic search was conducted up to October 2018 using five databases: Embase, Medline (Ovid), PubMed, CINAHL and the Cochrane Library. The search strategy in each database was «james lind*» OR «priorit* setting partnership*». We also searched in JLA website. This search identified 746 records and 431 potentially relevant citations. After removing duplicates and screening titles and abstracts based on our inclusion and exclusion criteria, the full text of 171 studies was examined in greater detail. A total of 37 studies met all criteria for review and were subsequently investigated. These numbers were verified by a university librarian (see flowchart, [figure 1](#)).

Selecting relevant studies

A prescreening process included reviewing the search results and excluding all articles that were not research studies, that were unavailable in full text or that clearly did not involve the JLA PSP approach. At least two authors screened the remaining articles using the inclusion and exclusion criteria presented in [table 1](#).

Charting data

A data extraction sheet was created to collect studies' demographic details, aims, samples and patient groups. The sheet was used to collect methodological details about the studies' PSPs, including descriptions of stakeholders, lists of top 10 priorities, descriptions of the roles of JLA facilitators and PSP stages.

Procedure

In addition to the first author, one of the other authors evaluated each article, and individual and comparative appraisals were discussed among the authors until agreement was reached. At least two authors were involved in each of the study selection procedures. A predefined procedure was developed for consulting a third author, or the whole research team, in cases of discrepancies; however, this was never necessary (ie, decisions to accept or reject unclear articles were based on a dyad consensus). The first author and one other author extracted the characteristics and findings of each study.

Quality appraisal

The most recent *JLA Guidebook*⁶ served as the context for investigating the descriptions of the studies' methods. A quality assessment was not included in the remit of this scoping review.⁸

Patient and Public Involvement

No patient was involved.

Collating, summarising and reporting results

Findings related to the scoping review's research questions, based on the JLA approach, were extracted and documented. The information shown in [table 2](#) includes the studies' aims, suggested uncertainties and—depending on the version of the JLA guidelines used—how these uncertainties were determined. We also collected information on the stakeholders (including members of the PSP), whether a JLA advisor/facilitator was used, and the JLA process stages: (1) setting up a PSP, (2) gathering uncertainties, (3) data processing and verifying uncertainties, (4) interim priority setting and (5) final priority setting. The results are presented based on the *JLA Guidebook* steps, which have remained consistent across versions.^{6 9–11}

RESULTS

In total, 37 studies met the inclusion criteria; their characteristics are summarised in [table 2](#).

The publication years ranged from 2010 to 2018. The number of studies using this process has increased annually, with 12 published in 2017. In our sample, 27 of the studies were from the UK,^{1 5 7 12–35} 8 were from Canada,^{36–43} and 1 each was from India⁴⁴ and Spain.⁴⁵

The JLA process participants were patients, carers and clinicians aged ≥18 years. The studies collectively represented patient groups with heterogeneous ages and health conditions/diseases, with later studies generally

more focused on symptoms and function than on diseases ([table 2](#)). Totally, 15 of the studies gave information about ethnicity.^{13 14 16 19 21 23 25–27 32 33 35 36 40 42} One of the studies also gave information about socioeconomic status.²⁶ Another study gave only information about socioeconomic status.⁴⁴

Three of studies described that patient and carers submitted more questions on psychosocial issues, psychosocial stress, depression and anxiety compared with clinicians.^{13 23 40} No studies described disagreement in the prioritisation stages. However, 24 other studies also mentioned psychosocial issues without noting who had done so.^{1 7 14–19 25–27 29 31–39 41–43} Ten studies did not mention psychosocial issues.^{5 12 20–22 24 26 28 44 45} The types of health conditions that were addressed included gastrointestinal,^{26–28} neurological,^{1 5 7 16 21 38} dermatological,^{13 15 17 29 34 45} endocrinal^{14 32 42} and cancer^{22 25 41 43} conditions.

Setting up a PSP

The JLA steering group is made up of key organisations and individuals who can collectively represent all or the majority of issues related to the PSP, either individually or through their networks.⁶

All included studies had a steering group, although they were described differently. Nineteen studies^{1 5 12 14–17 19 20 22 23 25 31 36 37 39–41 45} included patients, carers and clinicians in their steering groups; 16 studies^{7 13 18 24 26–29 32–35 38 42–44} did not include carers in their steering group (ie, only patients and clinicians). In one study,³⁰ the titles of the members on the steering group were not reported; in another,²¹ the steering group did not specifically include patients, carers or clinicians, but rather stated that representation from all stakeholders was ensured.

The number of JLA steps in the PSP process varied across studies from four steps^{1 32 33 37 39 42} to eight steps.^{20 22 44} Five steps, corresponding to *JLA Guidebook* V.4, V.5 and V.6, were most common,^{12 13 15–19 23 24 26–29 31 34 36 38 40 41 43 45} with step 1, initiation; step 2, collection of uncertainties; step 3, collation of uncertainties; step 4, interim priority setting; and step 5, final priority workshop.

Gathering uncertainties

PSPs aimed to gather uncertainties from as wide a range of potential contributors as possible, ensuring that patients were equally confident and empowered compared with clinicians in submitting their perspectives on uncertainties.⁶

With regard to recruitment, various partner organisations, local advertisements, social media, patients, carers and clinicians were PSP information targets. In addition to an online and paper survey, two studies also used face-to-face methods to reach and facilitate involvement by their identified groups.^{5 42}

The questions were usually deliberately open-ended to encourage full responses regarding the experiences of patients, carers and clinicians. One of the 37

Table 2 Characteristics of included studies

Year Author Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
2010 Buckley <i>et al</i> ¹² UK	To identify and prioritise 'clinical uncertainties' relating to treatment of UI.	<p>1. User group*</p> <p>2. <i>JLA Guidebook</i>, year and version</p> <p>3. Age of patient††</p> <p>4. Health condition/disease</p> <p>5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities</p> <p>Organisations were identified, which represented or could advocate for patients, their informal carers and clinicians involved in the treatment or management.</p>	NR	Five steps+NGT 1. Initiation. 2. Consultation. 3. Collation. 4. Prioritisation. 5. Dissemination.
2011 Eleftheriadou <i>et al</i> ¹³ UK	To stimulate and steer future research in the field of vitiligo treatment by identifying the 10 most important research areas for patients and clinicians.	<p>1. Patients, carers, clinicians and researchers.</p> <p>2. <i>JLA Guidebook</i> 2010, V.4.</p> <p>3. NR.</p> <p>4. Vitiligo.</p> <p>5. In total, 660 treatment uncertainties were submitted by 461 participants.</p> <p>Professional bodies and patient support groups; steering group included 12 members with knowledge and interest in vitiligo.</p>	The Vitiligo PSP adopted the methods advocated by the JLA, which were refined to meet the needs of this particular PSP.	Five steps 1. Initiation. 2. Consultation. 3. Collation. 4. Ranking exercise (Interim prioritisation exercise). 5. Final Prioritisation Workshop.
2012 Gadsby <i>et al</i> ¹⁴ UK	To collect uncertainties about the treatment of type 1 diabetes from patients, carers and health professionals, and to collate and prioritise these uncertainties to develop a list of top 10 of research priorities.	<p>1. Patients, carers and clinicians.</p> <p>2. <i>JLA Guidebook</i> 2010, V.4.</p> <p>3. NR.</p> <p>4. Type 1 diabetes.</p> <p>5. In total, 1141 treatment uncertainties were submitted by 583 participants.</p> <p>Members with perspectives in paediatrics and primary care, users of type 1 diabetes services, including patients and carers; a steering group of representatives from these organisations (n=9 plus an independent information specialist) and partner organisations.</p>	JLA, being represented on the steering group	Six steps 1. Setting up the partnership/survey. 2. Collecting uncertainties. 3. Collation activity. 4. Interim priority setting. 5. Final priority setting workshop. 6. Review.
2013 Batchelor <i>et al</i> ¹⁵ UK	To identify the uncertainties in eczema treatment that are important to patients who have eczema, their carers and the healthcare professionals who treat them.	<p>1. Patients, carers and clinicians.</p> <p>2. <i>JLA Guidebook</i> 2010, V.4.</p> <p>3. NR.</p> <p>4. Eczema.</p> <p>5. In total, 1070 treatment uncertainties were submitted by 493 participants.</p> <p>The steering group comprised four patients and carers, including a representative from the National Eczema Society, four clinicians, two dermatologists, a dermatology nurse specialist and a GP and three researchers/administrators at the Centre of Evidence-Based Dermatology.</p>	The PSP was coordinated from the Centre of Evidence-Based Dermatology in Nottingham, with oversight by a representative of JLA, who was the independent chair of the PSP steering group.	Five steps 1. Initiation. 2. Consultation—collection of treatment uncertainties. 3. Collation and treatment uncertainties. 4. Ranking of treatment uncertainties. 5. Workshop to develop research questions.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group†: identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
			<ol style="list-style-type: none"> 1. User group* 2. <i>JLA Guidebook</i>, year and version 3. Age of patient†† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 			
2013	Davila-Seijo et al ⁴⁵	Spain	To describe and prioritise the most important uncertainties about dystrophic epidermolysis bullosa treatment shared by patients, carers and healthcare professionals in order to promote research in those areas.	The steering group comprised eight people, including patients/carers, a representative from the Dystrophic Epidermolysis Bullosa Research Association Spain, a clinician, dermatologists, nurses and researchers; and the Spanish Academy of Dermatology and Venereology.	Workshop advocated by the JLA	Five steps+NGT 1. Initiation. 2. Consultation survey: collection of treatment uncertainties. 3. Ranking exercise. 4. Ranking exercise. 5. Final prioritisation workshop.
2013	Hall et al ⁷	UK	To describe the tinnitus PSP in providing a platform for patients and clinicians to collaborate, to identify and to prioritise uncertainties or 'unanswered questions'.	Membership of the steering group provided a broad representation of people from the field of tinnitus, including professional bodies, charities and advocates for people with tinnitus. The wider working partnership included 56 major UK stakeholders, including individual advocates for people with tinnitus, support groups, hospital centres and commercial organisations.	Independent chairperson representing the JLA	Seven steps 1. Establishing a working partnership. 2. Gathering suggestions for research on the assessment, diagnosis and treatment of tinnitus. 3. Checking and categorising submitted uncertainties. 4. Prioritising the uncertainties. 5. Developing a consensus. 6. Top 10 clinical research questions. 7. Recommendations for future research strategy.
2014	Deane et al ¹⁶	UK	To identify and prioritise the top 10 evidential uncertainties that impact on everyday clinical practice for the management of Parkinson's disease.	The steering group consisted of representatives from Parkinson's UK (n=8) and the Cure Parkinson's Trust (n=1), patients (n=2), carers (n=2), clinical consultants (n=2) and a Parkinson's disease nurse specialist (n=1). Those from Parkinson's UK included representatives with expertise in research development, policy and campaigns (n=5), information and support worker services (n=1), advisory services (n=1) and resources and diversity (n=1).	The JLA provided an independent chair, advised on the methodology, and facilitated the process.	Five steps+NGT 1. Initiation. 2. Consultation. 3. Uncertainties survey. 4. Collation. 5. Prioritization.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
				<p>1. User group*</p> <p>2. JLA Guidebook, year and version</p> <p>3. Age of patient††</p> <p>4. Health condition/disease</p> <p>5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities</p>		
2014	Ingram <i>et al</i> ¹⁷	UK	To generate a top 10 list of hidradenitis suppurativa research priorities, from the perspectives of patients with hidradenitis suppurativa, carers and clinicians, to take to funding bodies.	<p>The steering committee included five patients and carers, including two representatives of the Hidradenitis Suppurativa Trust UK patient organisation; six dermatologists, including two trainees, two dermatology specialist nurses, a plastic surgeon, a general practitioner; the JLA representative and an administrator and stakeholders from various Royal College-related groups.</p>	Three JLA facilitators or four facilitators	Five steps+NTG 1. Identify stakeholders. 2. Invitation to submit uncertainties. 3. Generate 'indicative uncertainties'. 4. Rank uncertainties. 5. Final workshop.
2014	Manns <i>et al</i> ¹⁶	Canada	To improve understanding of kidney function and disease, including those for specific areas, such as dialysis therapies.	<p>The priority setting process was initiated with the formation of an 11-person steering group, which included patients, a caregiver, clinicians, an employee of the Kidney Foundation of Canada and an expert in the JLA approach.</p>	Experienced facilitators	Five steps+NGT 1. Survey. 2. Collation. 3. Combining. 4. Interim prioritisation. 5. Final workshop.
2014	Pollock <i>et al</i> ⁵	UK	To identify the top 10 research priorities relating to life after stroke, as agreed by stroke survivors, carers and clinicians.	<p>A steering group comprising a stroke survivor, carers, a nurse, a physician, allied clinicians, a researcher and representatives from key national stroke charities/patient organisations and from the JLA; the Scottish Government's National Advisory Committee for Stroke. This project was completed in partnership with Chest Heart & Stroke Scotland and the Stroke Association in Scotland.</p>	The facilitators were briefed by members of the JLA on the importance of ensuring equitable participation of all group members	Six steps+NGT 1. Form PSP. 2. Gather treatment uncertainties. 3. Check treatment uncertainties. 4. Interim prioritisation. 5. Final priority setting. 6. Reporting and dissemination.
2014	Rowe <i>et al</i> ¹⁸	UK	To identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians.	<p>The steering committee included patient representatives and eye health professionals. A steering committee and data assessment group comprising the authors of this article oversaw the process and stakeholders from various Royal College-related groups. The Steering Committee also included patient representatives and eye health professionals.</p>	Representative from the JLA convened meetings of the steering committee	Five steps+NGT 1. Establishing the Sight Loss Vision PSP. 2. Survey. 3. Data assessment. 4. Interim prioritisation. 5. Final prioritisation.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
			<ol style="list-style-type: none"> 1. User group* 2. JLA Guidebook, year and version 3. Age of patient†† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 			
2014	Uhm <i>et al</i> ¹⁹	UK	To discover the research questions for preterm birth and to grade them according to their importance for infants and families.	Potential partners were identified through a process of peer knowledge and consultation, steering group members' networks and JLA's existing register of affiliates. Stakeholders from various Royal College-related groups.	Two facilitators from the JLA	Five steps+NGT <ol style="list-style-type: none"> 1. Initiation of the partnership. 2. Identifying treatment uncertainties. 3. Collation: refining questions and uncertainties. 4. Prioritisation – interim and final stages. 5. Publicity and publishing results.
2015	Barnieh <i>et al</i> ¹⁷	Canada	To assess the research priorities of patients on or nearing dialysis within Canada and their carers and clinicians.	The 11-person steering group comprised four patients, one carer, three clinicians, an employee of the Kidney Foundation of Canada (an important funder of kidney research in Canada), an expert in the JLA approach and a researcher. The steering group included individuals from across Canada and different stakeholders.	Facilitators with experience in the JLA methods lead the workshop	Four steps+NGT <ol style="list-style-type: none"> 1. Form PSP 2. Gather research uncertainties. 3. Process and collate submitted research uncertainties. 4. Final priority setting workshop.
2015	Boney <i>et al</i> ²⁰	UK	To identify research priorities for anaesthesia and perioperative medicine.	The steering group comprised representatives of the funding partner organisations, patients and carers, and the JLA. Almost 2000 stakeholders contributed their views regarding anaesthetic and perioperative research priorities. Stakeholders were defined as 'any person or organisation with an interest in anaesthesia and perioperative care'.	Steering group chaired by the JLA adviser	Eight steps <ol style="list-style-type: none"> 1. Enrol partner organisations. 2. Identify research questions. 3. Classify and refine research question. 4. Short-listing. 5. Literature review. 6. Interim prioritisation. 7. Final prioritisation. 8. Publication and dissemination of results.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
				<ol style="list-style-type: none"> 1. User group* 2. <i>JLA Guidebook</i>, year and version 3. Age of patient†† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 		
2015	Kelly <i>et al</i> ²¹	UK	To identify unanswered questions around the prevention, treatment, diagnosis and care of dementia, with the involvement of all stakeholders; to identify a top 10 prioritised list of uncertainties.	Potential partner organisations were identified through the networks of the Alzheimer's Society and the steering group, ensuring representation from all stakeholders. Patients, carers and clinicians were not involved in the steering group.	The Dementia PSP was guided and chaired by an independent JLA representative.	Six steps+NGT 1. Involvement of potential partner organisations. 2. Identifying uncertainties. 3. Question management and analysis. 4. Verifying uncertainties. 5. Interim prioritisation. 6. Final prioritisation workshop.
2015	Stephens <i>et al</i> ²²	UK	To identify the top 10 research priorities relating to mesothelioma (pleural or peritoneal), specifically, to identify those unanswered questions that involved an intervention.	Steering group comprised two patients, one bereaved carer, nine clinicians (including nurses, surgeons, oncologists, chest physicians and palliative care experts) and four representatives of patient and family support groups (one of the representatives was also a bereaved carer); in total, 16 participants.	The steering group was chaired by a JLA facilitator.	Eight steps 1. Establishing a steering group. 2. Initial survey questionnaire. 3. Reviewing the survey responses. 4. Searching. 5. Interim prioritisation. 6. Final priority setting. 7. Identified unanswered questions. 8. An additional PSP.
2016	Knight <i>et al</i> ²³	UK	To identify unanswered research questions in the field of kidney transplantation from end-service users (patients, carers and healthcare professionals).	The steering group included transplant surgeons, nephrologists, transplant recipients, living donors and carers. Additional partner organisations were invited to take part in the process by involving their members in the surveys and helping to promote the process. National patient and professional organisations and charities involved in kidney transplantation were contacted about the project and were invited to contribute to a steering group.	The steering group was chaired by an experienced advisor from the JLA.	Five steps+NGT 1. Organisation and scope. 2. Identification of potential research questions. 3. Refinement of questions and identification of existing literature. 4. Interim prioritisation. 5. Final prioritisation workshop.

Continued

Table 2 Continued

Year Author Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
2016 Rangan <i>et al</i> ²⁴ UK	To run a UK-based JLA PSP for 'surgery for common shoulder problems'	<p>1. Patients, carers and clinicians.</p> <p>2. <i>JLA Guidebook</i> 2013, V.5.</p> <p>3. NR.</p> <p>4. Shoulder surgery</p> <p>5. In total, 652 treatment uncertainties were submitted by 371 participants.</p>	A JLA adviser	<p>Five steps</p> <ol style="list-style-type: none"> 1. Identification and invitation of potential partners. 2. Initial meeting/ awareness raising. 3. Identifying treatment uncertainties. 4. Refining questions and uncertainties. 5. Prioritisation interim and final prioritisation workshop.
2016 van Middendorp <i>et al</i> ⁷ UK	To identify a list of Top 10 priorities for future research into spinal cord injury.	<ol style="list-style-type: none"> 1. Patient, spouse/partner and clinicians. 2. <i>JLA Guidebook</i> 2013, V.5. 3. Ages 18–80 years. 4. Spinal cord injury. 5. In total, 784 treatment uncertainties were submitted by 403 participants. 	Support and guidance were provided by the JLA	<p>Four steps</p> <ol style="list-style-type: none"> 1. Gathering of research questions. 2. Checking of existing research evidence. 3. Interim prioritisation. 4. Final consensus meeting.
2016, Wan <i>et al</i> ²⁵ UK	To establish a consensus regarding the top 10 unanswered research questions in endometrial cancer.	<ol style="list-style-type: none"> 1. Patients, carers and clinicians. 2. <i>JLA Guidebook</i> 2013, V.5. 3. NR. 4. Endometrial cancer. 5. In total, 786 individual submissions from 413 participants. 	An independent advisor from the JLA was chair of the steering group	<p>Six steps+NGT</p> <ol style="list-style-type: none"> 1. Establishing a steering group. 2. Consultative process. 3. Gathering uncertainties. 4. Data analysis and verifying uncertainties. 5. Interim priority setting. 6. Final priority setting.
2017, Britton <i>et al</i> ²⁶ UK	Facilitate balanced input in the priority setting process for Barrett's oesophagus and gastro-oesophageal reflux disease and to reach a consensus on the Top 10 uncertainties in the field	<ol style="list-style-type: none"> 1. Patients, carers and clinicians. 2. <i>JLA Guidebook</i> 2013, V.5. 3. NR. 4. Gastro-oesophageal reflux disease and Barrett's oesophagus. 5. In total, 629 treatment uncertainties were submitted by 170 participants. 	NR.	<p>Five steps+NGT</p> <ol style="list-style-type: none"> 1. Initial survey. 2. Initial response list. 3. Long-list generation and verification. 4. Interim prioritisation survey. 5. Final workshop.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps	Description of stages NGT
				<ol style="list-style-type: none"> 1. User group* 2. <i>JLA Guidebook</i>, year and version 3. Age of patient†† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 			
2017,	Fitzcharles et al ³⁸	Canada	Priorities of uncertainties for the management of fibromyalgia (FM) that could propel future research.	<p>The steering committee was composed of five patients (one patient was a practising pharmacist), five healthcare professionals (one family physician, two rheumatologists, one psychologist and one internist), an internist with previous experience of the JLA process but without specific interest in FM, and a rheumatologist.</p>	Facilitators with experience of the JLA process	Five steps	<ol style="list-style-type: none"> 1. Survey results. 2. In scope uncertainties. 3. Coding uncertainties. 4. Interim prioritisation. 5. Final workshop.
2017,	Hart et al ²⁷	UK	To devise a list of the key research priorities regarding treatment of inflammatory bowel disease, as seen by clinicians, patients and their support groups, using a structure established by the JLA.	<p>A steering committee was established following an initial explanatory meeting and included two patients, two gastroenterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organisation Crohn's and Colitis UK, a representative of the JLA and an administrator.</p>	A JLA facilitator	Five steps	<ol style="list-style-type: none"> 1. Initiation and setting up the committee. 2. Collection of treatment uncertainties. 3. Collation of treatment uncertainties. 4. Ranking of treatment uncertainties. 5. Development of a list of top 10 priorities.
2017,	Hemmelgarn et al ³⁹	Canada	To identify the most important unanswered questions (or uncertainties) about the management of CKD, that is, in terms of diagnosis, prognosis and treatment.	<p>The priority setting process with the formation of a 12-person steering group from across Canada, including patients with non-dialysis CKD, a carer, clinicians (nephrologists), researchers and an employee of the Kidney Foundation of Canada (non-profit organisation for patients with kidney disease).</p>	Jointly organised PSP broadly adhering to the <i>JLA Guidebook</i>	Four steps+NGT	<ol style="list-style-type: none"> 1. Identification and invitation of potential partners. 2. Collection of research uncertainties through a national survey. 3. Refinement and prioritisation. 4. Priority setting workshop.
2017,	Khan et al ⁴⁰	Canada	To identify the 10 most important research priorities of patients, carers and clinicians for hypertension management.	<p>Steering committee of 15 volunteer patients, carers and clinicians from across Canada. Stakeholder not reported in detail.</p>	JLA facilitator from the UK	Five steps	<ol style="list-style-type: none"> 1. Establishing a steering group. 2. Forming PSPs. 3. Collecting potential research questions. 4. Processing, categorising and summarising those research questions. 5. Selecting the top 10 research priorities.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group†, identification and management of partners/stakeholders	JLA	The role of the facilitator/ advisor	PSP	Number of steps	Description of stages
				<ol style="list-style-type: none"> 1. User group* 2. <i>JLA Guidebook</i>, year and version 3. Age of patient†† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 					
2017,	Jones <i>et al</i> ¹¹	Canada	To identify unanswered questions encountered during management of kidney cancer agreement by consensus on a prioritised list of the top 10 shared unanswered questions and to establish corresponding research priorities.	A 15-person steering group was formed with 7 patients/carers and 7 expert clinicians from across Canada. In response, the Kidney Cancer Research Network of Canada, in collaboration with the JLA, Kidney Cancer Canada, the Kidney Foundation of Canada, was formed	The group also included an advisor from the JLA (UK) who provided support and advice throughout the process.		Five steps	<ol style="list-style-type: none"> 1. Formation of steering group. 2. Identifying treatment questions. 3. Collating questions. 4. Interim ranking of questions. 5. Final priority setting workshop. 	
2017,	Lomer <i>et al</i> ²⁸	UK	To provide a comprehensive summary of the research priority findings relating to diet in the treatment of inflammatory bowel disease	Steering committee comprising two patients, two gastroenterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organisation, Crohn's and Colitis UK, a representative of the JLA and an administrator (ie, 13-person steering committee). Stakeholders from various roles, ages and ethnic groups.	A representative of the JLA and an administrator on the steering committee.		Five steps	<ol style="list-style-type: none"> 1. Steering committee. 2. Questionnaire survey. 3. Remaining uncertainties were reviewed. 4. Uncertainties were determined. 5. Final workshop of the steering group. 	
2017,	Macbeth <i>et al</i> ²⁹	UK	To identify uncertainties in alopecia areata management and treatment that are important to both service users, people with hair loss, carers/relatives and clinicians.	Four people representing various patient support groups, four dermatologists and two further individuals to represent the BHNS and the European Hair Research Society; an academic psychologist; a registered trichologist and a GP; and a JLA representative. Two separate steering groups.	A JLA representative provided independent oversight of the PSP and chaired the steering group.		Five steps+NGT	<ol style="list-style-type: none"> 1. Identification and invitation of potential partners. 2. Invitation to submit uncertainties. 3. Collation. 4. Ranking of treatment uncertainties. 5. Final workshop. 	
2017,	Narahari <i>et al</i> ⁴⁴	India	To summarise the process of lymphoedema PSP, discussion during the final prioritisation workshop and recommendation on the top seven priorities for future research in lymphoedema and a brief road map.	The Faculty of Applied Dermatology and the Central University of Kerala participated in the coordinating committee	NR		Eight steps	<ol style="list-style-type: none"> 1. Initiation and setting up a coordinating committee. 2. Literature search. 3. Contacting stakeholders. 4. Listing priorities for research. 5. Random collation of priorities. 6. Ranking exercises. 7. Free lymphoedema medical camp. 8. Final prioritisation workshop. 	

Continued

Table 2 Continued

Year Author Country	Aim of the study	Steering group†: identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
2017 Prior <i>et al</i> UK	To identify and prioritise important research questions for miscarriage.	<ol style="list-style-type: none"> 1. Patients, partners, family members, friends or colleagues and clinicians. 2. <i>JLA Guidebook</i> 2016, V.6. 3. NR. 4. Miscarriage. 5. In total, 3279 questions were submitted by 2122 participants. 	The workshop was chaired by an independent JLA facilitator.	Six steps <ol style="list-style-type: none"> 1. Initiation. 2. Consultation. 3. Identifying uncertainties. 4. Refining uncertainties. 5. Interim prioritisation. 6. Final workshop.
2017 Rees <i>et al</i> ⁴² Canada	Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus	<ol style="list-style-type: none"> 1. Patients, friends and relatives and clinicians. 2. <i>JLA Guidebook</i> 2013, V.5. 3. Ages 18–69 years. 4. Gestational diabetes mellitus. 5. In total, 389 treatment uncertainties were submitted by 75 participants. 	A facilitator familiar with the JLA process	Four steps+NGT <ol style="list-style-type: none"> 1. Survey. 2. Process and collate. 3. Interim ranking. 4. Priority setting workshop.
2017 Smith <i>et al</i> ³⁰ UK	Prioritise research questions in emergency medicine in a consensus process to determine the Top 10 questions	<ol style="list-style-type: none"> 1. Patients, carers and clinicians. 2. <i>JLA Guidebook</i> 2013, V.5. 3. NR. 4. Emergency medicine. 5. In total, 214 initial uncertainties. 	NR.	Six steps <ol style="list-style-type: none"> 1. Online submissions. 2. Working group reviews. 3. Mini systematic reviews. 4. Working group prioritisation exercise. 5. Public prioritisation exercise. 6. Face-to-face final prioritisation.
2018 Fernandez <i>et al</i> ⁵¹ UK	To establish the research priorities for adults with fragility fractures of the lower limb and pelvis that represent the shared interests and priorities.	<ol style="list-style-type: none"> 1. Patients, carers and clinicians. 2. <i>JLA Guidebook</i> 2016, V.6. 3. Age ≥60 years. 4. Fragility fractures of the lower limb and pelvis. 5. In total, 963 treatment uncertainties were submitted by 365 participants. 	A JLA adviser supported and guided the PSP	Five steps <ol style="list-style-type: none"> 1. First survey. 2. Screening. 3. Thematic analysis, original uncertainties turned into overarching indicative questions. 4. Evidence search interim prioritisation. 5. Final workshop.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
			<ol style="list-style-type: none"> 1. User group* 2. <i>JLA Guidebook</i>, year and version 3. Age of patient† 4. Health condition/disease 5. Number of initial uncertainties and participants or returned surveys or uploaded research priorities 			
2018	Finer <i>et al</i> ⁶²	UK	To describe processes and outcomes of a PSP and to identify the top 10 research priorities' in type 2 diabetes.	The steering group comprised five people living with type 2 diabetes (managing their condition in different ways), five clinicians (including a dietician, diabetes specialist nurse, GP and two consultant diabetologists), an information specialist, seven members of the Diabetes UK Research and the senior leadership team, and a JLA senior advisor. The steering group (47% men and 53% women and 26% from black and minority ethnic groups) met 12 times during the PSP process, in person or by teleconference.	The workshop was facilitated by trained JLA advisors.	Four steps+NGT 1. Gathering uncertainties. 2. Organising the uncertainties. 3. Interim priority setting. 4. Final priority setting.
2018	Lecheit <i>et al</i> ⁴³	Canada	To identify the top 10 treatment uncertainties in head and neck cancer from the joint perspective of patients, caregivers, family members and treating clinicians.	The steering committee included five patients with head and neck cancer who were from 3 to 25 years since diagnosis; seven clinicians involved in the treatment and management of head and neck cancer (maxilla-facial prosthodontist, radiation oncologist, speech language pathologist, clinician-researcher, infectious disease specialist, anaplastologist, and two head and neck oncological and reconstructive surgeons). However, a sixth individual (family member) was involved informally throughout the project, despite being unable to commit to regular participation. Alberta Cancer Foundation and the Institute for Reconstructive Sciences in Medicine	The workshop was led by an independent facilitator with extensive experience on JLA PSP projects, supported by two cofacilitators, all of whom were briefed by the JLA senior advisor on recommended JLA protocols.	Five steps+NGT 1. Initial survey development and deployment. 2. Identifying uncertainties through survey data processing. 3. Verifying uncertainties. 4. Interim prioritisation. 5. Final workshop.
2018	Lough <i>et al</i> ⁶³	UK	To identify the shared priorities for future research of women affected by and clinicians involved with pessary use for the management of prolapse.	The steering group comprised three women with pessary experience, three clinicians experienced in managing prolapse with pessaries, two researchers and a pessary company representative, the PSP with guidance from the JLA adviser and project leader. The JLA Pessary PSP was partially funded by a UK Continence Society research grant, two grants from the Pelvic Obstetric and Gynaecological Physiotherapy group of the Chartered Society of Physiotherapy and a funded studentship from Glasgow Caledonian University.	The steering group agreed on the terms of reference and protocol for the JLA adviser and project leader.	Four steps+NGT 1. Gathering questions/uncertainties. 2. Refining the questions and checking the evidence. 3. Prioritising/ranking the questions. 4. Choosing the top 10 priorities by consensus.

Continued

Table 2 Continued

Year	Author	Country	Aim of the study	Steering group† identification and management of partners/stakeholders	JLA The role of the facilitator/ advisor	PSP Number of steps Description of stages NGT
2018	Macbeth <i>et al</i> ³⁴	UK	Identify uncertainties in hair loss management, prevention, diagnosis and treatment that are important to both people with hair loss and clinicians	The steering group comprised four people representing various patient support groups, four dermatologists, a psychologist, a registered trichologist and a GP. A JLA representative ensured key stakeholders were identified through a process of consultation and peer knowledge, building on steering group members' networks and existing JLA affiliates.	The process was facilitated by the JLA to ensure fairness, transparency and accountability.	Five steps+NGT 1. Identification and invitation of potential partners. 2. Invitation to submit uncertainties. 3. Collation. 4. Ranking of treatment uncertainties. 5. Final workshop.
			1. Patients, carers relatives and clinicians. 2. JLA Guidebook 2016, V.6. 3. NR 4. Hair loss (excluding alopecia areata) 5. In total, 2747 treatment uncertainties were submitted by 912 participants.			

*User group means the participants who are involved in the PSP process, not only the survey.

†Age refers to age of patients who are involved in the survey.

‡Steering group, steering committee and coordinating committee are defined as equal concepts.

BHNS, British Hair and Nail Society; OKD, chronic kidney disease; GP, general practitioner; JLA, James Lind Alliance; NGT, nominal group technique; NR, not reported; PSP, priority setting partnership; UI, urinary incontinence.

studies⁴⁴ used an online survey to collect uncertainties; patients and clinicians were invited via email to endorse their priorities based on a table that had been developed from abstracts collected in a literature search. Among the other 36 studies, 12 used open-ended questions^{1 15 18 23 25 32 35 40–43 45} such as 'What questions about the management of hypertension or high blood pressure would you like to see answered by research?' In seven studies, participants (patients, carers and clinicians) were asked to submit three to five research ideas.^{16 17 20 21 27 28 33}

In eight studies, no limit was placed on the types of questions that could be submitted.^{5 13 24 30 31 36 37 39} One study asked about eight open-ended questions requesting a narrative answer.³⁸ Close-ended questions were used in three studies,^{22 29 34} such as 'Do you have questions about the prevention, diagnosis or treatment of hair loss that need to be answered by research?' Five studies did not report their question format.^{7 12 14 19 26}

The number of submitted uncertainties ranged from 8227, submitted by 2587 participants,³² to 323, submitted by 58 participants.⁴⁵ All studies except two^{7 44} reported involving patients, carers and clinicians in the initial survey. Two of the studies addressed verifying uncertainties for example by content experts or librarians.^{40 43} The steering group or researchers were involved in addressing verifying uncertainties in 22 of the studies and^{5 7 14–16 18 20 21 23–27 30 31 33 35 37 39 41 44 45} in 13 of the studies not describing verifying the uncertainties.^{1 12 13 17 19 22 28 29 32 34 36 38 42}

Data processing and verifying uncertainties

Unlike most surveys that are designed to collect answers, JLA PSP surveys are designed to collect questions. The survey responses must then be reviewed, sorted and turned into a list of 'indicative' questions, all of which are unanswered uncertainties.⁶

According to Lechelt *et al*,⁴³ uncertainties are organised through coding, with natural clusters emerging. During this step, duplicates such as similar and related uncertainties are identified. Clinician–patient dyads consolidate and rephrase each cluster of related questions into a single indicative uncertainty, written in lay language using a standard format. Lomer *et al*²⁸ specified that similar uncertainties are combined to create indicative uncertainties. Among our included studies, 20 described refining questions into indicative uncertainties,^{5 13–15 17 19 20 23 24 27–29 31–34 38 39 42 43} while 17 did not describe a concept of indicative uncertainties.^{1 7 12 16 18 21 22 25 26 30 35–37 40 41 44 45}

In total, 16 of the studies described directly ranking and assessing survey-generated uncertainties from a long list ranging from 43 to 226 uncertainties.^{1 5 13 14 19–21 23 24 26 27 30 38 41 43 44}

The wording of the long list of uncertainties was reviewed by the steering group, and, in some cases, wording was altered to make the uncertainties more understandable and to explain complex words not generally well known to the public.¹

Interim priority setting

Interim prioritisation is the stage at which the long list of uncertainties (indicative questions) is reduced to a short list for the final priority setting workshop.⁶

All studies described an interim stage, using the terms: interim priority setting^{14 32}; interim prioritisation^{1 5 42}; and ranking exercise.^{13 44}

Their short lists varied from 22²⁶ to 30 uncertainties.^{12 17–19 22 25 30 36 39} Sixteen of the studies used an interim prioritisation of their top 25 uncertainties that were taken to a final prioritisation workshop, where the participants agreed on their top 10 priorities.^{1 7 13 20 21 23 24 28 29 31 33–35 37 38 40} Three of the studies did not describe the number of short-listed treatment uncertainties.^{15 27 44}

To reduce the number of uncertainties, an interim prioritisation exercise was conducted by email or by post.^{5 18 32} Patients, carers and health professionals were initially invited to examine the long list¹⁸; 14 of the studies used a second online survey,^{1 19 20 23 25 28 29 31–35 38 40} and in one study, the steering group members facilitated an interim ranking exercise.³⁹

Final priority setting

The JLA's final stage is a rank ordering of the uncertainties, with a particular emphasis on the lists of top 10 priorities. For JLA PSPs, a final face-to-face priority setting workshop is conducted with both small group and whole group discussions. The NGT can be used by groups, with voting to ensure that all opinions are considered⁶; 21 of the studies reported using the NGT in the final priority setting workshop.^{5 12 16–19 21 23 25 26 29 32–34 36 37 39 41–43 45}

All of the studies implemented a final priority setting workshop to agree on their top 10 priorities. In most of the studies, these final workshops included patients, carers and clinicians; nine studies mentioned including only patients and clinicians.^{7 23 28 29 33 34 42–44}

DISCUSSION

To our knowledge, this is the first scoping review of published studies using the JLA approach, although the number of steps used by PSPs differed and not all papers describe in detail every aspect of the JLA approach. However, overall they incorporated the same procedural content, which indicates no implications or small implications for our findings. Thus, this scoping review provides unique insight into a broad and varied range of perspectives on PPI using the JLA approach. Interestingly, there were some differences between the questions submitted by patients and carers compared with those submitted by clinicians. The patients focused more on symptoms and function than on disease, while clinicians focused on general treatment. Compared with clinicians, patients submitted more questions about psychosocial issues, psychosocial stress, depression and anxiety.^{13 23 40} There were no studies that described disagreement in the prioritisation steps. The health conditions addressed in these studies were primarily somatic diseases, although

one study was about life after stroke and included mental health.⁵ Thus, the JLA approach is an appropriate and important method for defining research from the perspectives of end users that is, patients and carers.⁴⁶

A key value that informs such partnerships is often described as equality. Equitable partnerships might be defined as a gradation of shared responsibility negotiated in a collaborative and cooperative decision-making environment. Whether such values always align within the JLA process is an open question. Thus, reflecting on and clarifying values about involvement before starting collaborative work might enhance the positive impacts while avoiding the negative impacts of public involvement.⁴⁷

The number of priority setting exercises in health research is increasing,⁴⁸ and our review indicates that the use of the JLA approach is also growing. This approach facilitates broad stakeholder involvement, and it is transparent and easy to replicate. This is consistent with findings by Yoshida,⁴⁸ who argues that there is a clear need for transparent, replicable, systematic and structured approaches to research priority setting to assist policy-makers and research funding agencies in making investments. Increased public involvement can lead to a wider range of identified and prioritised research topics that are more relevant to service users.⁴⁹ A key strength of involving the public and patients, rather than only academics, throughout the partnership process is described in these studies, including having a project led by representatives of a wider range of consumer and clinician organisations.¹ The number of resulting uncertainties reflects this breadth. The studies examined tended to conclude that the JLA principles were welcomed, but consistently emphasised the need for an even broader understanding, better conceptualisation and improved processes to incorporate the results into research. However, few studies focused on how to reach the weakest voices for survey participation. After critically reading these studies, one might ask whether they included the lowest socioeconomic groups and most vulnerable patients. Many respondents, particularly those associated with charity organisations, are likely to be white and middle class and to have high education attainment levels. Yet it is the individuals who are more difficult to reach, such as those in low socioeconomic groups and those who are vulnerable patients, who may have the greatest unmet needs and stand to gain the most from improved treatment.^{25 26 35 42} Given that the JLA is designed to identify shared research priorities, such individuals and their needs may not be reflected in what is typically reported studies. In one case, to better facilitate patient and carer involvement, and to reach those who may not receive and/or respond to email or postal information, a steering group member visited existing support groups and arranged the distribution of information leaflets at local meetings.⁵ Although great efforts were reportedly made²⁵ to include participants from black and minority ethnic groups and care home populations, they were not particularly successful. Lough *et al*³³ reported that the use of an online survey may introduce a bias in

favour of patients who use the internet and social media. It is also likely that those with literacy issues will not participate.¹⁶ Three of the studies^{5 18 42} attempted to facilitate participation among those with language barriers and literacy issues, which implies that efforts need to be made to enable minority groups and learning disabilities to participate in the PSP process. Stephens *et al*²² note another major challenge to involving users in research and patients in the steering group who have incapacitating symptoms and short expected survival durations. Another important issue is that all but two studies^{44 45} were from English-speaking countries and thus represent a relatively limited global population.

According to the *JLA Guidebook*,⁶ PSPs usually report their process and methods, the participants involved, results, reflections on successes, lessons learnt or limitations, and the next steps. It is important that these reports be written in a language understandable to everyone with an interest in the topic, not just to clinicians. Lough *et al*³³ explained that all of the unanswered questions generated by their PSPs would be available on the JLA website and widely disseminated to research commissioners, public health and research funders. However, such reports can be difficult to obtain by those without ready online access or by those with literacy issues. Eleftheriadou *et al*¹³ included implementation of a feasibility study as one of their top 10 priorities; the authors hoped that, following its publication, along with their list of the most important uncertainties, relevant studies would be developed.

Running a PSP and involving the relevant stakeholders in deciding which research should be funded seem to be an effective and sustainable model.²⁴ Without doubt, the essential advantage is integration of this involvement in both research and healthcare. Identifying research priorities is perhaps where the PSP's greatest effect can be achieved.²⁶ Nevertheless, one might ask whether PSPs emphasise basic research less than applied research. Abma *et al*⁵⁰ have argued that the international literature describes corresponding challenges in research agenda setting and follow-up; patient involvement is limited to actual agenda setting, and there is limited understanding of what happens next and how to shape patient involvement activities in follow-up phases. This scoping review process gathered a large number of research priorities from a diverse set of respondents.^{32 34} There has been a clear paradigm shift from a reactive to a more proactive approach described as 'predictive, personalised, preventative and participatory'.²⁵ It is expected that the JLA process will have a clinical impact by driving relevant research studies based on PPI. Crowe *et al*⁵¹ reported that a critical mismatch between the treatments that patients and clinicians want to have evaluated and the treatments actually being evaluated by researchers. This apparent mismatch should be taken into account in future research.

Strengths and limitations

A major strength of this paper is the application of a rigorous and robust scoping review method, including

independent screening and data extraction. The search strategy was carefully performed in conjunction with a research librarian. To strengthen the review's validity, several databases were used, and we have reported them with complete transparency. The studies selected for inclusion were manually searched. Although we searched multiple databases for the period since their inception, we may not have identified all relevant studies. We did not search the grey literature, assuming that empirical research using the JLA approach would be found in indexed databases. As a scoping review, the findings describe the nature of research using JLA's approach and provide direction for future research; hence, this review cannot suggest how to operationalise the JLA process or how to use it in a given context. Another strength is that several of the researchers contributing to this project also work in the clinical areas represented in the studies. In addition, while a quality analysis was beyond the scope of this paper, we have noted varying descriptions within the selected studies (ie, sample sizes, health status and age of groups). Finally, the included studies do not provide information about the impact of involvement, regarding development of consensus, the discussions among all those who took part, the distribution of power and the politics. In future work, it may be important to evaluate how much influence patient/public partners had during the process, besides the impact of the number of participants in the respective groups. Another limitation might involve our inclusion criteria with respect to requirement for peer-reviewed publications, which by definition will use more academic language and may not be readily accessible to the layperson. Lastly, the cost and time involved in a PSP are described in one publication only.²⁴ According to the *JLA Guidebook*, the PSP process will last approximately 12–18 months.⁶

CONCLUSIONS

JLA-based PSP makes a useful contribution to identifying research questions. A range from 327 to 8227 uncertainties were published, with 27 studies from UK. The number of reported steps varied from four to eight. In total, 33 studies mentioned the involvement of a JLA facilitator. Twenty-four included studies that addressed methods for verifying uncertainties, and the use of NGT was reported in 21 studies. Finally, it is important that the results of these studies, including the top 10 priorities, reach those who answered the survey, including the vulnerable groups. Online publishing might contribute to this. Future studies should focus on factors influencing patient and carer involvement in priority setting projects.

Acknowledgements We thank research librarian Malene Wøhlk Gundersen for her helpful and knowledgeable assistance.

Contributors AN, LH, SL, EKG and AB designed the study. AN coordinated the project and is the guarantor. AN, LH, SL, EKG and AB screened articles and performed data extraction. AN conducted the literature search. AN, LH, SL, EKG and AB interpreted the data. AN drafted the manuscript and all authors critically reviewed it. All authors read and approved the manuscript.

Funding This work was supported by the Research Council of Norway (grant number OFFPHD prnr 271870), Lørenskog Municipality and Oslo Metropolitan University. The funders had no role in the study design, data collection and analysis, decision to publish, or preparation of the manuscript.

Competing interests None declared.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement All data relevant to the study are included in the article.

Open access This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>.

REFERENCES

- van Middendorp JJ, Allison HC, Ahuja S, *et al*. Top ten research priorities for spinal cord injury: the methodology and results of a British priority setting partnership. *Spinal Cord* 2016;54:341–6.
- Hanley B, Bradburn J, Barnes M, *et al*. Involving the public in NHS public health, and social care research: Briefing notes for researchers. *UK: Involve* 2004;2:1–61.
- Hoddinott P, Pollock A, O’Cathain A, *et al*. How to incorporate patient and public perspectives into the design and conduct of research. *F1000Res* 2018;7.
- Price A, Albarqouni L, Kirkpatrick J, *et al*. Patient and public involvement in the design of clinical trials: an overview of systematic reviews. *J Eval Clin Pract* 2018;24:240–53.
- Pollock A, St George B, Fenton M, *et al*. Top 10 research priorities relating to life after stroke – consensus from stroke survivors, caregivers, and health professionals. *International Journal of Stroke* 2014;9:313–20.
- National Institute for health research, the James Lind alliance Guidebook: version 7, 2018. Available: <http://www.jla.nihr.ac.uk/jla-guidebook/downloads/Print-JLA-guidebook-version-7-March-2018.pdf>
- Hall DA, Mohamad N, Firkins L, *et al*. Identifying and prioritizing unmet research questions for people with tinnitus: the James Lind alliance tinnitus priority setting partnership. *Clin Invest* 2013;3:21–8.
- Tricco AC, Lillie E, Zarin W, *et al*. A scoping review on the conduct and reporting of scoping reviews. *BMC Med Res Methodol* 2016;16:15.
- National Institute for health research, the James Lind alliance Guidebook: version 6, 2016. Available: <http://jla.nihr.ac.uk/jla-guidebook/downloads/JLA-Guidebook-Version-6-February-2016.pdf>
- Cowan K, Oliver S. The James Lind alliance Guidebook: version 5, 2013. Available: <http://www.jlguidebook.org/pdfguidebook/guidebook.pdf>
- Cowan K, Oliver S. James Lind alliance Guidebook: version 4, 2010. Available: <http://www.bvsde.paho.org/texcom/cd045364/guidebook.pdf>
- Buckley BS, Grant AM, Tincello DG, *et al*. Prioritizing research: patients, carers, and clinicians working together to identify and prioritize important clinical uncertainties in urinary incontinence. *NeuroUrol Urodyn* 2010;29:708–14.
- Eleftheriadou V, Whitton ME, Gawkrödger DJ, *et al*. Future research into the treatment of vitiligo: where should our priorities lie? results of the vitiligo priority setting partnership. *Br J Dermatol* 2011;164:no–6.
- Gadsby R, Snow R, Daly AC, *et al*. Setting research priorities for Type 1 diabetes. *Diabetic Medicine* 2012;29:1321–6.
- Batchelor JM, Ridd MJ, Clarke T, *et al*. The eczema priority setting partnership: a collaboration between patients, carers, clinicians and researchers to identify and prioritize important research questions for the treatment of eczema. *Br J Dermatol* 2013;168:577–82.
- Deane KHO, Flaherty H, Daley DJ, *et al*. Priority setting partnership to identify the top 10 research priorities for the management of Parkinson’s disease. *BMJ Open* 2014;4:e006434.
- Ingram JR, Abbott R, Ghazavi M, *et al*. The hidradenitis suppurativa priority setting partnership. *Br J Dermatol* 2014;171:1422–7.
- Rowe F, Wormald R, Cable R, *et al*. The sight loss and vision priority setting partnership (SLV-PSP): overview and results of the research prioritisation survey process. *BMJ Open* 2014;4:e004905.
- Uhm S, Crowe S, Dowling I, *et al*. The process and outcomes of setting research priorities about preterm birth — a collaborative partnership. *Infant* 2014;10:178–81.
- Boney O, Bell M, Bell N, *et al*. Identifying research priorities in anaesthesia and perioperative care: final report of the joint National Institute of academic Anaesthesia/James Lind alliance research priority setting partnership. *BMJ Open* 2015;5:e010006.
- Kelly S, Lafortune L, Hart N, *et al*. Dementia priority setting partnership with the James Lind alliance: using patient and public involvement and the evidence base to inform the research agenda. *Age Ageing* 2015;44:985–93.
- Stephens RJ, Whiting C, Cowan K, *et al*. Research priorities in mesothelioma: a James Lind alliance priority setting partnership. *Lung Cancer* 2015;89:175–80.
- Knight SR, Metcalfe L, O’Donoghue K, *et al*. Defining priorities for future research: results of the UK kidney transplant priority setting partnership. *PLoS One* 2016;11:e0162136.
- Rangan A, Upadhaya S, Regan S, *et al*. Research priorities for shoulder surgery: results of the 2015 James Lind alliance patient and clinician priority setting partnership. *BMJ Open* 2016;6:e010412.
- Wan YL, Beverley-Stevenson R, Carlisle D, *et al*. Working together to shape the endometrial cancer research agenda: the top ten unanswered research questions. *Gynecol Oncol* 2016;143:287–93.
- Britton J, Gadeke L, Lovat L, *et al*. Research priority setting in Barrett’s oesophagus and gastro-oesophageal reflux disease. *Lancet Gastroenterol Hepatol* 2017;2:824–31.
- Hart AL, Lomer M, Verjee A, *et al*. What are the top 10 research questions in the treatment of inflammatory bowel disease? A priority setting partnership with the James Lind alliance. *ECCOJC* 2017;11:204–11.
- Lomer MC, Hart AL, Verjee A, *et al*. What are the dietary treatment research priorities for inflammatory bowel disease? a short report based on a priority setting partnership with the James Lind alliance. *J Hum Nutr Diet* 2017;30:709–13.
- Macbeth AE, Tomlinson J, Messenger AG, *et al*. Establishing and prioritizing research questions for the treatment of alopecia areata: the alopecia areata priority setting partnership. *Br J Dermatol* 2017;176:1316–20.
- Smith J, Keating L, Flowerdew L, *et al*. An emergency medicine research priority setting partnership to establish the top 10 research priorities in emergency medicine. *Emerg Med J* 2017;34:454–6.
- Fernandez MA, Arnel L, Gould J, *et al*. Research priorities in fragility fractures of the lower limb and pelvis: a UK priority setting partnership with the James Lind alliance. *BMJ Open* 2018;8:e023301.
- Finer S, Robb P, Cowan K, *et al*. Setting the top 10 research priorities to improve the health of people with type 2 diabetes: a diabetes UK-James Lind alliance priority setting partnership. *Diabetic Medicine* 2018;35:862–70.
- Lough K, Hagen S, McClurg D, *et al*. Shared research priorities for pessary use in women with prolapse: results from a James Lind alliance priority setting partnership. *BMJ Open* 2018;8:e021276.
- Macbeth A, Tomlinson J, Messenger A, *et al*. Establishing and prioritizing research questions for the prevention, diagnosis and treatment of hair loss (excluding alopecia areata): the hair loss priority setting partnership. *Br J Dermatol* 2018;178:535–40.
- Prior M, Bagness C, Brewin J, *et al*. Priorities for research in miscarriage: a priority setting partnership between people affected by miscarriage and professionals following the James Lind alliance methodology. *BMJ Open* 2017;7:e016571.
- Manns B, Hemmelgarn B, Lillie E, *et al*. Setting research priorities for patients on or nearing dialysis. *CJASN* 2014;9:1813–21.
- Barnieh L, Jun M, Laupacis A, *et al*. Determining research priorities through partnership with patients: an overview. *Semin Dial* 2015;28:141–6.
- Fitzcharles M-A, Brachaniec M, Cooper L, *et al*. A paradigm change to inform fibromyalgia research priorities by engaging patients and health care professionals. *Canadian Journal of Pain* 2017;1:137–47.
- Hemmelgarn BR, Pannu N, Ahmed SB, *et al*. Determining the research priorities for patients with chronic kidney disease not on dialysis. *Nephrol Dial Transplant* 2017;32:847–54.
- Khan N, Bacon SL, Khan S, *et al*. Hypertension management research priorities from patients, caregivers, and healthcare providers: a report from the hypertension Canada priority setting partnership group. *J Clin Hypertens* 2017;19:1063–9.
- Jones J, Bhatt J, Avery J, *et al*. The kidney cancer research priority-setting partnership: identifying the top 10 research priorities as defined by patients, caregivers, and expert clinicians. *Can Urol Assoc J* 2017;11:379–87.

42. Rees SE, Chadha R, Donovan LE, *et al.* Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus. *Canadian Journal of Diabetes* 2017;41:156–63.
43. Lechelt LA, Rieger JM, Cowan K, *et al.* Top 10 research priorities in head and neck cancer: results of an Alberta priority setting partnership of patients, caregivers, family members, and clinicians. *Head Neck* 2018;40:544–54.
44. Narahari SR, Aggithaya M, Moffatt C, *et al.* Future research priorities for morbidity control of lymphedema. *Indian J Dermatol* 2017;62:33–40.
45. Davila-Seijo P, Hernández-Martín A, Morcillo-Makow E, *et al.* Prioritization of therapy uncertainties in dystrophic epidermolysis bullosa: where should research direct to? an example of priority setting partnership in very rare disorders. *Orphanet J Rare Dis* 2013;8:61.
46. Chalmers I, Ignorance CT. Confronting therapeutic ignorance. *BMJ* 2008;337:a841–7.
47. Gradinger F, Britten N, Wyatt K, *et al.* Values associated with public involvement in health and social care research: a narrative review. *Health Expectations* 2015;18:661–75.
48. Yoshida S. Approaches, tools and methods used for setting priorities in health research in the 21st century. *J Glob Health* 2016;6.
49. Barber R, Boote JD, Parry GD, *et al.* Can the impact of public involvement on research be evaluated? a mixed methods study. *Health Expectations* 2012;15:229–41.
50. Abma TA, Pittens CACM, Visse M, *et al.* Patient involvement in research programming and implementation: a responsive evaluation of the dialogue model for research agenda setting. *Health Expect* 2015;18:2449–64.
51. Crowe S, Fenton M, Hall M, *et al.* Patients', clinicians' and the research communities' priorities for treatment research: there is an important mismatch. *Res Involv Engagem* 2015;2.