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The James Lind Alliance Process approach: A scoping review

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Running head: James Lind Alliance approach

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ABSTRACT

Objective: To summarize studies' descriptions of the James Lind Alliance (JLA) approach to the Priority Setting Partnership (PSP) process and how this process is used to identify treatment uncertainties and develop Top 10 priorities lists.

Design: Scoping review.

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

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

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

Results: Database searches yielded 322 potentially relevant studies, of which 33 met inclusion criteria. Included studies were those published from 2011 to 2018. JLA process participants were patients, carers and clinicians, aged 18 years or older, who had experience with the study-relevant diagnoses. All studies reported having a steering group, though partners and stakeholders were described differently across studies. The number of JLA PSP process steps varied from four to eight. Treatment uncertainties were typically collected via an online survey hosted on, or linked to, the PSP website. The number of submitted treatment uncertainties varied across studies, from 323 submitted by 58 participants to 8,227 submitted by 2,587 participants.

Conclusions: Patient and public involvement makes meaningful contributions to health research. Patient, carer and clinician collaboration on a PSP can be used to identify and

prioritize treatment uncertainties. However, representation of those with different health conditions depends on their having the capacity and resources to participate. No studies reported difficulty with developing their Top 10 priorities.

Article Summary

Strengths and limitations of this study

- This is the first scoping review of published studies used the JLA approach.
- Give possibilities for large involvement of patient, carer and public in setting the research agenda.
- It is hoped that the Top 10 prioritise list will lead to future research that will address issues of importance for the clinical management of the different diseases.
- Very few participants were from minority ethnic populations which could limit the generalisability of these priorities to these populations.
- One inherent limitation to this process is that the weakest voices often lack representation which could limit the generalisability of these priorities to these populations.

Keywords: James Lind Alliance, Priority Setting Partnership, Patient and Public Involvement, patient involvement in research.

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 to actively contribute, through discussion, to decision-making regarding research design, acceptability, relevance, conduct and governance-from study conception to dissemination.[3]

Researchers have noted that involving health care 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 study.[1, 4]

The James Lind Alliance (JLA), a United Kingdom-based non-profit initiative, 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 treatment uncertainties relating to a specific condition.[5] According to Hall et al.,[6] 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,[5] uncertainties and how to prioritize these are key features of the JLA process. The process begins by defining unanswered questions (i.e., 'uncertainties') about the effects of treatment and health care—questions that cannot be adequately answered based on existing research evidence such as reliable, up-to-date systematic reviews—and then prioritizes the uncertainties based on their importance. The most recent JLA Guidebook version 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 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.[5]

The JLA provides independent facilitation and guidance in the identification and prioritization processes. The PSP process uses a systematic and transparent design and approach consisting of several steps, outcomes and ways to secure user involvement. This process forms part of a widening approach to PPI in research. Once a partnership is set up, there is a defined process for collecting uncertainties and interim priority setting, which leads to a list of approximately 20–30 uncertainties used in a final priority setting workshop at which a Top 10 list of priorities is agreed upon.[7] 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.[5] Considering the growing importance of PPI to research and the limited evaluation of this process to date, there is a clear need for aggregated information about how the JLA approach is currently used in health research. To our knowledge, no review has yet been published describing how the JLA approach is used to identify uncertainties in health care. Thus, this scoping review describes the JLA approach to the PSP process and how it is used to identify treatment uncertainties and develop Top 10 priority lists. Specifically, we asked:

- What characterizes published studies' aims, user groups, adherence to the JLA approach, health conditions and number of initial treatment uncertainties?
- How are PSPs organized?
- What processes are used to gather and verify uncertainties?

METHODS

Study design

Our study protocol was developed using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines and registered with the PROSPERO international prospective registry of systematic reviews (registration number CRD#42018093569).

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Identifying relevant studies

A systematic search was conducted through October 2018 using five databases: Embase, Medline (Ovid), PubMed, CINAHL and Cochrane Library. The search strategy in each database was: «james lind*» OR «priorit* setting partnership*». This search identified 638 records and 322 potentially relevant citations. After removing duplicates and screening titles and abstracts based on our inclusion and exclusion criteria, the full text of 91 studies was examined in greater detail. A total of 33 studies met all criteria for review and were subsequently investigated. These numbers were verified by a university librarian. See Flow chart, figure 1.

Selecting relevant studies

A pre-screening process included reviewing the search results and excluding all articles that were not research studies, 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.

Table 1 Criteria for inclusion and exclusion

Inclusion criteria	Exclusion criteria
• All steps from James Lind Alliance • List of Top 10 priorities • Adults (aged 18 years or older)	• Unpublished literature • Articles not written in English • Priority Setting Partnership without James Lind Alliance • James Lind Alliance without Priority Setting Partnership • Protocols • Erratu • Editor • Thesis • Comments review • Guidelines • Randomized controlled trials (RCT)

Charting data

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

Procedure

In addition to the first author evaluating every article, individual and comparative appraisals were discussed among the authors until they reached agreement. A pre-defined procedure was developed for consulting a third author, or the whole research team, for cases of discrepancies; however, this was never necessary (i.e., decisions to accept or reject unclear articles were based on dyad consensus). The first author and one other author extracted the characteristics and findings of each study.

Quality appraisal

The most recent JLA Guidebook [5] 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

Patients and public were not involved in this study.

Collating, summarizing 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—if the JLA guidelines were used—how these uncertainties were determined. We also collected information on the stakeholders (including members of the PSP), whether a JLA advisor or 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; 5) Final priority setting. The results are presented based on the JLA Guidebook steps, which have remained consistent across versions.[5, 9-11]

Table 2 Characteristic of included studies

Year Author Country	Aim of the study	1. User group* 2. James Lind Alliance (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	Steering group*** identification and management of partners/stakeholders	JLA The role of the facilitator/ Advise.	Priority Setting Partnership (PSP) Number of steps Description of stages Nominal Group Technique (NGT)
2011 Eleftheriadou et al [16] United Kingdom (UK)	Stimulate and steer future research in the field of vitiligo treatment, by identifying the 10 most important research areas for patients and clinicians.	1. Patients, carers, clinicians and researchers 2. JLA Guidebook 2010, version 4 3. Not reported (NR) 4. Vitiligo 5. Total 660 treatment uncertainties were submitted by 461 participants	Professional bodies and patient support groups. Steering group, included 12 members with knowledge and interest in Vitiligo.	The vitiligo PSP adopted the methods advocated by the JLA which were refined to meet the needs of this particular PSP.	5 steps 1. Initiation 2. Consultation 3. Collation 4. Ranking exercise (Interim prioritization exercise) 5. Final Prioritisation Workshop
2012 Gadsby et al [7] UK	Collect uncertainties about the treatment of Type 1 diabetes from patients, carers and health professionals, and to collate and prioritize these uncertainties to develop a Top 10 list of research priorities.	1. Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Type I diabetes 5. Total 1141 treatment uncertainties were submitted by 583 participants	Members with perspectives in paediatrics and primary, users of Type 1 diabetes services, including patients and carers. A steering group of representatives from these organizations (n = 9 plus an independent information specialist) and partner organisations.	JLA by being represented on the steering group.	6 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 et al [12] UK	Identify the uncertainties in eczema treatment that are important to patients who have	1. Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. NR	The steering group comprised four patients and carers, including a representative from the National Eczema Society, four clinicians, two dermatologists, a	The PSP was coordinated from the Centre of Evidence-Based Dermatology in Nottingham, with	5 steps 1. Initiation 2. Consultation – collection of treatment

	eczema, their carers and the healthcare professionals who treat them.	4. Eczema 5. Total 1070 treatment uncertainties were submitted by 493 participants	dermatology nurse specialist and a general practitioner and three researchers /administrators at the Centre of Evidence-Based.	oversight by a representative of the JLA, who was the independent chair of the PSP steering group.	uncertainties 3. Collation of treatment uncertainties 4. Ranking of treatment uncertainties 5. Workshop to develop research questions
2013 Davila-Seijo et al [40] Spain	Describe and prioritize the most important uncertainties about Dystrophic Epidermolysis Bullosa treatment shared by patients, carers and health care professionals in order to promote research in those areas.	1.Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. Age 21 – 54 years 4. Dystrophic Epidermolysis Bullosa 5. Total 323 treatment uncertainties were submitted by 58 participants	The steering group comprised eight people with experience in Dystrophic Epidermolysis Bullosa including patients/carers, a representative from the Dystrophic Epidermolysis Bullosa Research Association Spain, clinician; dermatologists and nurses and researchers/ and the Spanish Academy of Dermatology and Venereology.	Workshop advocated by the JLA.	5 steps +NGT 1. Initiation 2. Consultation survey: collection of treatment uncertainties 3. Ranking exercise 4. Ranking exercise 5. Final prioritization workshop
2013 Hall et al [6] UK	Describe the Tinnitus PSP in providing a platform for patients and clinicians to collaborate to identify and prioritize uncertainties or 'unanswered questions'.	1. Patients and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Tinnitus 5. Total 2483 treatment uncertainties were submitted by 825 participants	Membership of the steering group provided a broad representation of people from the field of Tinnitus in the UK, including professional bodies, charities and advocators for people with Tinnitus. The wider working partnership included 56 major UK stakeholders including individual advocators for people with Tinnitus, support groups, hospital centres and commercial organizations.	Independent chairperson, representing JLA.	7 steps 1. Establishing a working partnership 2. Gathering suggestions for research on the assessment, diagnosis & treatment of tinnitus 3. Checking & categorizing submitted uncertainties 4. Prioritizing the uncertainties 5. Developing consensus 6.Top ten clinical research questions

					7.Recommendations for future research strategy
2014 Deane et al [15] UK	Identify and prioritise the Top 10 evidential uncertainties that impact on everyday clinical practice for the management of Parkinson's disease.	1. Patients, carers, family, friends, clinicians 2. JLA guidebook 2013, version 5 3. NR 4. Parkinson's disease 5. Total 4100 treatment uncertainties submitted by 1000 participants	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 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.	5 steps + NGT 1. Initiation 2. Consultation 3. Uncertainties survey 4. Collation 5. Priorisation
2014 Ingram et al [19] UK	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.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Hidradenitis suppurativa 5. Total 1495 treatment uncertainties submitted by 371 participants	The steering committee included five patients with Hidradenitis suppurativa and carers, including two representatives of the Hidradenitis suppurativa Trust UK patient organization, 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.	Three JLA facilitators or four facilitators.	5 steps+ NTG 1. Identify stakeholders 2. Invitation to submit uncertainties 3. Generate "Indicative uncertainties" 4. Rank uncertainties 5. Final workshop
2014 Pollock et al [4] UK	Identify the Top 10 research priorities relating to life after stroke, as agreed by stroke survivors, carers and clinicians.	1. Patients, carers, clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Life after stroke 5. Total 548 treatment uncertainties	A steering group comprising a stroke survivor, carers, nurse, physician, allied clinicians, researcher and representatives from key national stroke charities/patient organizations, and from the JLA. The Scottish Government's National Advisory Committee for Stroke. This project was	The facilitators were briefed by members of the JLA on the importance of ensuring equitable participation of all group members.	6 steps +NGT 1. Form PSP 2. Gather treatment uncertainties 3. Check treatment uncertainties 4. Interim prioritisation 5. Final priority setting

			completed in partnership with Chest Heart and Stroke Scotland and The Stroke Association in Scotland.		6. Reporting & dissemination
2014 Rowe et al [30] UK	Identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians.	1. Patients, carers and clinicians 2.JLA Guidebook 2013, version 5 3. The average age of participants was 65,7 years old 4. Sight loss or an eye condition 5. Total 4461 treatment uncertainties submitted by 2220 participants	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.	Representative from the JLA convened meetings of the steering committee.	5 steps + NGT 1. Establishing the Sight Loss Vision PSP 2. Survey 3. Data assessment 4. Interim prioritisation 5. Final prioritisation
2014 Uhm et al [32] UK	Discover the research questions for preterm birth and to grade them according to their importance for infants and families.	1. Patients, carers and clinicians 2. NR 3. NR 4. Preterm birth 5. Total 593 research questions were raised completed by 386 people	Potential partners were identified through a process of peer knowledge and consultation, steering group members' networks and JLAs existing register of affiliates. Stakeholders from various Royal College-related groups.	Two facilitators from the JLA.	5 steps + NGT 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 et al [33] Canada	Assess the research priorities of patients on or nearing dialysis within Canada and their carers and clinicians.	1. Patients carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. On or nearing dialysis 5. Total 1820 treatment uncertainties and number of participants are not reported	The 11 persons 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	Facilitators with experience in the JLA methods lead the workshop.	4 steps + NGT 1. Form PSP 2. Gather research uncertainties 3. Process and collate submitted research uncertainties 4. Final priority setting workshop

			Canada and different stakeholders.		
2015 Boney et al [13] UK	Identify research priorities for Anaesthesia and Perioperative Medicine.	1. Patient, carer and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Anaesthesia and Perioperative Medicine. 5. Total 1420 treatment uncertainties submitted by 623 participants	Steering group comprising 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.	8 steps 1. Enrol partner organisations 2. Identify research question 3. Classify and refine research question 4. Shortlisting 5. Literature review 6. Interim prioritisation 7. Final prioritisation 8. Publication and dissemination of results
2015 Kelly et al [20] UK	Identify unanswered questions around the prevention, treatment, diagnosis and care of dementia with the involvement of all stakeholders as well as to identify a Top 10 prioritised list of uncertainties.	1. Patients, carers/relatives, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Dementia 5. Total 1563 uploaded surveys	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 are not involved in the steering group.	The Dementia PSP was guided and chaired by an independent representative of the JLA.	6 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 et al [29] UK	Identify the Top 10 research priorities relating to mesothelioma (pleural or peritoneal), and specifically to identify those unanswered questions that involved an intervention.	1. Patients, current and bereaved carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Mesothelioma 5. Total 453 initial surveys	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	The steering group was chaired by a JLA facilitator.	8 steps 1. Establishing a steering group 2. Initial survey questionnaire 3. Reviewing the survey responses 4. Searching 5. Interim

			was also a bereaved carer) = 16 participants.		prioritisation 6. Final priority setting 7. Identified unanswered questions 8. An additional PSP
2016 Knight et al [21] UK	Identify unanswered research questions in the field of kidney transplantation from end service users (patients, carers and healthcare professionals).	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Kidney transplantation 5. Total 497 treatment uncertainties submitted by 183 participants	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 invited to contribute to a steering group.	The steering group was chaired by an experienced advisor from the JLA.	5 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
2016 Rangan et al [27] UK	To run a UK based JLA PSP for 'Surgery for Common Shoulder Problems'.	1. Patients, carers and clinicians, 2. JLA Guidebook 2013, version 5 3. NR 4. Shoulder surgery 5. Total 652 treatment uncertainties submitted by 371 participants	The steering group was made up of the most relevant stakeholders and included patients, physiotherapists, general practitioners, shoulder surgeons, anaesthetists and pain control experts, orthopaedic nurses and an academic clinician National networks and interest organisations.	A JLA adviser .	5 steps 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
2016 Van Middendorp et al [1] UK	Identify a Top 10 list of priorities for future research into spinal cord injury.	1. Patient, spouse/partner and clinicians 2. JLA Guidebook 2013, version 5 3. Age 18-80 4. Spinal cord injury	The steering group comprised representatives from each stakeholder organisation, including an independent information manager. Stakeholders included consumer	Support and guidance was provided by the JLA.	4 steps 1. Gathering of research questions 2. Checking of existing research evidence 3. Interim

		5. Total 784 treatment uncertainties submitted by 403 participants	organisations, clinician societies, carers representatives.		prioritisation 4. Final consensus meeting
2016, Wan et al [31] UK	Establish a consensus regarding the top ten unanswered research questions in Endometrial cancer.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Endometrial cancer 5. Total 786 individual submissions from 413 participants	As part of the JLA process, all organisations that could reach and advocate for patients, carers and clinicians were invited to become involved in a PSP. A steering group composed of representatives from these groups was then formed to ensure the study remained inclusive and fulfilled its aim to deliver and publicise a list of shared research priorities. Group of 23 stakeholders not described in details.	An independent advisor from the JLA was Chair of the steering group.	6 steps + NGT 1. Establishing a Steering Group 2. The consultative process 3. Gathering uncertainties 4. Data analysis and verifying uncertainties 5. Interim priority setting 6. Final priority setting
2017, Britton et al [14] UK	Facilitate balanced input in the priority setting process for Barret's oesophagus and gastro-oesophageal reflux disease and to reach a consensus on the top ten uncertainties in the field.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Gastro-oesophageal reflux disease and Barrett's oesophagus 5. Total 629 treatment uncertainties submitted by 170 participants	Professionals, patients and charity representatives formed a Steering Committee. The steering committee identified the broader. British Society of Gastroenterology, National health service, the university of Manchester, Association of Upper Gastro intestinal surgeons and Primary Society for Gastroenterology.	NR.	5 steps + NGT 1. Initial survey 2. Initial response list 3. Longlist generation and verification 4. Interim prioritisation survey 5. Final workshop
2017, Hart et al [18] UK	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.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Inflammatory bowel disease. 5. Total 1636 treatment uncertainties submitted by 531 participants	A steering committee was established following an initial explanatory meeting and included two patients with Inflammatory bowel disease, 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	A JLA facilitator.	5 steps 1. Initiation and setting up the committee 2. Collection of treatment uncertainties 3. Collation of treatment uncertainties 4. Ranking of treatment

			Colitis UK, a representative of the JLA and an administrator.		uncertainties 5. Development of Top 10
2017, Hemmelgarn et al [34] Canada	Identify the most important unanswered questions (or uncertainties) about the management of Chronic kidney disease (i.e. in terms of diagnosis, prognosis and treatment).	1. Patients, carers, clinicians and policy-makers 2. JLA Guidebook 2013, version 5 3. Age 65 and over 4. Non-dialysis Chronic kidney disease 5. Total 2241 treatment uncertainties submitted by 439 participants	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-for-profit organization for patients with kidney disease.	Jointly organized PSP broadly adhering to the JLA Guidebook.	4 steps + NGT 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 [36] Canada	Identify the 10 most important research priorities of patients, carers and clinicians for hypertension management.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Hypertension 5. Total 673 individual research questions submitted by 386 participants	Steering committee of 15 volunteer patients, carers, and clinicians from across Canada. Stakeholder NR in detail.	JLA facilitator from UK.	5 steps 1. Establishing a Steering Group 2. Forming priority setting partnerships 3. Collecting potential research questions 4. Processing, categorising, and summarising those research questions 5. Selecting the Top 10 research priorities
2017, Jones et al [35] Canada	Identify unanswered questions encountered during management of kidney cancer and agree by consensus on a prioritized list of the Top 10 shared unanswered questions and establish corresponding research priority.	1. Patients, carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patient with kidney cancer 5. Total 2004 treatment questions submitted by 225 participants	A 15 persons 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.	The group also included an advisor from the JLA (UK) who provided support and advice throughout the process.	5 steps 1. Formation of Steering Group 2. Identifying treatment questions 3. Collating questions 4. Interim ranking of questions 5. Final priority setting workshop
2017, Lomer et al [22]	Provide a comprehensive	1. Patients, carers and clinicians	Steering committee	A representative of the JLA and an administrator	5 steps

UK	summary of the research priority findings relating to diet in the treatment of inflammatory bowel disease.	2. JLA Guidebook 2016, version 6 3. NR 4. Dietary treatment of inflammatory bowel disease. 5. Total 1671 treatment uncertainties submitted by 531 participants	comprising of two patients with inflammatory bowel disease. 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 James Lind Alliance; and an administrator (13 involved in the steering committee). Stakeholders from various roles, ages and ethnic groups.	in the steering committee.	1. Steering Committee 2. Questionnaire survey 3. Remaining uncertainties were reviewed 4. Uncertainties determined 5. Final workshop of Steering Group
2017, Mcbeth et al [25] UK	Identify uncertainties in Alopecia Areata management and treatment that are important to both service users, people with hair loss, carers/ relatives and clinicians.	1. Patients, partners/parents/ carers and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Alopecia Areata 5. Total 2747 treatment uncertainties submitted by 912 participants	Four people with hair loss 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 general practitioner and a JLA representative. Two separate steering group.	A JLA representative provided independent oversight of the PSP and chaired the steering group.	5 steps + NGT 1. Identification and invitation of potential partners 2. Invitation to submit uncertainties 3. Collation 4. Ranking of treatment uncertainties 5. Final workshop
2017, Narahari et al [39] India	Summarises the process of PSP lymphedema, discussion during the final prioritisation workshop, and recommendation on the top seven priorities for future research in lymphedema and a brief roadmap.	1. Patients, theraoists and nurses 2. JLA Guidebook 2013, version 5 3. NR 4. Lymphedema 5. Total 137 respondents uploaded research- priorities	The faculty of Applied Dermatology and the Central University of Kerala participated in the coordinating committee.	NR.	8 steps 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 lymphedema medical camp

					8. Final prioritisation workshop
2017 Prior et al [26] UK	Identify and prioritise important research questions for miscarriage.	1. Patients, partners, family members, friends or colleagues and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Miscarriage 5. Total 3279 questions submitted by 2122 participants	The steering group was a balanced composition of women who had experience of miscarriage, charities that represented them and clinicians. Some members representing charities or clinicians also had personal experience of pregnancy loss.	The workshop was chaired by an independent JLA Facilitator.	6 steps 1. Initiation 2. Consultation 3. Identifying uncertainties 4. Refining uncertainties 5. Interim prioritisation 6. Final workshop
2017 Rees et al [38] Canada	Engaging patients and clinicians in establishing research priorities for Gestational Diabetes Mellitus.	1. Patients, friends and relatives and clinicians 2. JLA Guidebook 2013, version 5 3. Age18-69 4. Gestational Diabetes Mellitus 5. Total 389 treatment uncertainties submitted by 75 participants	A steering committee consisting of 3 patients and 3 clinicians (1 family physician who practises intrapartum care, an endocrinologist and a neonatologist); a facilitator familiar with the JLA process and a project manager. The Diabetes Obesity and Nutrition Strategic Clinical Network with Alberta Health Services supported this research. NR as stakeholder.	A facilitator familiar with the JLA process.	4 steps + NGT 1. Survey 2. Process and Collate 3. Interim ranking 4. Priority setting workshop
2017 Smith et al [28] UK	Prioritise research questions in Emergen medicine in a consensus process to determine the Top 10 questions.	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Emergen medicine 5. Total 214 number of initial uncertainties	The steering group members are not reported with titles, but consist of 16 members. Royal College of Emergency Medicine.	NR.	6 steps 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 Finer et al [17] UK	Describe processes and outcomes of a priority setting partnership to identify the 'top 10 research priorities' in	1.Patients, carers, clinicians 2. JLA Guidebook 2016, version 6 3. NR 4.Diabetes 2	The steering group comprised five people living with Type 2 diabetes (managing their condition in different ways), five clinicians (including a dietician, diabetes	The workshop was facilitated by trained James Lind Alliance advisors.	4 steps+ NGT 1. Gathering uncertainties 2. Organising the

	Type 2 diabetes.	5. Total 8227 treatment uncertainties were submitted by 2587 participants	specialist nurse, general practitioner and two consultant dialectologists), an information specialist, seven members of the Diabetes UK research and senior leadership team, and a James Lind Alliance senior advisor. The steering group (47% men and 53% women and 26% from black and minority ethnic groups) met 12 times during the priority setting partnership process, in person or by teleconference Diabetes UK.		uncertainties 3. Interim priority setting 4. Final priority setting
2018 Lechelt et al [37] Canada	Identify the Top 10 treatment uncertainties in head and neck cancer from the joint perspective of patients, caregivers, family members, and treating clinicians.	1. Patients, carers, family members, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patient with head and neck cancer 5. Total 818 treatment uncertainties submitted by 161 participants	The steering committee included 5 patients with head and neck cancer who were from 3 to 25 years since diagnosis; 7 clinicians involved in the treatment and management of head and neck cancer (maxillofacial prosthodontist, radiation oncologist, speech language pathologist clinician-researcher, infectious disease specialist, anaplastologist, and 2 head and neck oncologic 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 2 co-facilitators, all of whom were briefed by the JLA senior advisor on recommended JLA protocols.	5 steps +NGT 1. Initial survey development and deployment 2. Identifying uncertainties through survey data processing 3. Verifying uncertainties 4. Interim prioritization 5. Final workshop
2018 Lough et al [23] UK	Identify the shared priorities for future research of women affected by and clinicians involved with	1. Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. Age 30-89	The steering group comprised three women with pessary experience, three clinicians experienced in managing prolapse with pessaries, two researchers and a pessary company	The steering group agreed the terms of reference and protocol for the JLA adviser and project leader.	4 steps +NGT 1. Gathering questions/uncertainties 2. Refining the questions and checking the evidence

	pessary use for the management of prolapse.	4. Pessary use in women with prolapse 5.Total 669 questions submitted by 210 participants	representative, the PSP with guidance from the JLA adviser and project leader. Funding the JLA Pessary PSP was partially funded by a UK Continence Society (UKCS) research grant, two grants from the Pelvic Obstetric and Gynaecological Physiotherapy group (POGP) of the Chartered Society of Physiotherapy and a funded studentship from Glasgow Caledonian University.		3. Prioritising /ranking the questions 4. Choosing the Top 10 priorities by consensus
2018 Mcbeth et al [24] UK	Identify uncertainties in hair loss management, prevention, diagnosis and treatment that are important to both people with hair loss and clinicians.	1. Patients, carers and relatives and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Hair Loss (excluding Alopecia Areata) 5. Total 2747 treatment uncertainties were submitted by 912 participants	The steering group comprised four people with hair loss representing various patient support groups, four Dermatologists, a Psychologist, a registered Trichologist and a General Practitioner. A JLA representative provided 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.	5 steps + NGT 1. Identification and invitation of potential partners 2. Invitation to submit uncertainties 3. Collation 4. Ranking of treatment uncertainties 5. Final workshop

* 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 equally concept

RESULTS

Thirty-three studies met inclusion criteria; their characteristics are summarized in table 2.

The publication years for the included studies ranged from 2011 to 2018, collectively providing a broad evidence base relating to implementation of the JLA process in clinical health and social practice. The number of studies using this process have increased annually, to a total of 11 published in 2017. Twenty-five of the included studies were from the United Kingdom,[1, 4, 6, 7, 12-32] six from Canada [33-38] and one each from India [39] and Spain.[40]

The JLA process participants were patients, carers and clinicians, aged 18 years or older, with experience in the study-specific diagnoses. The studies collectively represented patient groups with heterogeneous ages and health conditions/disease, with later studies generally more focused on symptoms and function than on diseases (table 2).

Compared with clinicians, patients and carers contributed a greater number of questions regarding psychosocial issues, psychosocial stress, depression and anxiety.[16, 21, 36] The types of health conditions that were addressed included gastrointestinal,[14, 18, 22] neurologic,[1, 4, 6, 15, 20] dermatologic,[12, 16, 19, 24, 25, 40] endocrine [7, 17, 38] and cancer [29, 31, 35, 37] conditions.

Setting up a Priority Setting Partnership

The JLA steering group is made up of key organizations and individuals who can collectively represent all or the majority of issues related to the PSP, either individually or through their networks.[5]

All included studies had a steering group, though they were described differently. Sixteen studies [1, 4, 7, 12, 13, 15, 19, 21, 29, 31-36, 40] included patients, carers and clinicians in

their steering groups. Fifteen studies [6, 14, 16-18, 22-27, 30, 37-39] did not include carers in their steering group (i.e. only patients and clinicians). In one study,[28] the titles of the members on the steering group were not reported; in another [20] 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, 17, 23, 33, 34, 38] to eight steps.[13, 29, 39] Five steps, corresponding to JLA Guidebook versions 4, 5 and 6, were most common,[12, 14-16, 18, 19, 21, 22, 24, 25, 27, 30, 32, 35-37, 40] with Step 1: Initiation; Step 2: Collecting of uncertainties; Step 3: Collation of uncertainties; Step 4: Interim priority setting; Step 5: Final priority workshop.

Gathering uncertainties

PSPs aimed to gather treatment uncertainties from as wide a range of potential contributors as possible, ensuring that patients were equally confident and empowered compared to clinicians in submitting their perspectives on treatment uncertainties.[5]

With regard to recruitment, various partner organizations, 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.[4, 38]

A qualitative survey was most commonly used to generate questions and themes during this step, with questions reflecting the PSP's scope. The questions were usually deliberately open-ended to encourage full responses regarding patients', carers' and clinicians' experiences. One of the 33 studies [39] used an online survey to collect treatment 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 32 studies, 12 used

open-ended questions [1, 12, 17, 21, 26, 30, 31, 35-38, 40] 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.[13, 15, 18-20, 22, 23] In six studies, there were no limits placed on the types of questions that could be submitted.[4, 16, 27, 28, 33, 34] Close-ended questions were used in three studies,[24, 25, 29] such as, 'Do you have questions about prevention, diagnosis or treatment of hair loss that need answered by research?' Four studies did not report their question format.[6, 7, 14, 32]

The number of submitted treatment uncertainties ranged from 8,227 treatment uncertainties submitted by 2,587 participants [17] to 323 treatment uncertainties submitted by 58 participants.[40] Every study except two [6, 39] reported involving patients, carers and clinicians in the initial survey.

Data processing and verifying uncertainties

Unlike most surveys, which 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.[5]

According to Lechelt et al.,[37] uncertainties are organized through coding, with natural clusters emerging. During this step, duplicates such as similar and related treatment 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.,[22] specified that similar uncertainties are combined to create indicative uncertainties. Among these studies, 18 described refining questions into indicative uncertainties,[4, 7, 12, 13, 16-19, 21-25, 27, 32, 34, 37, 38] while 15 did not describe a concept of indicative uncertainties.[1, 6, 14, 15, 20, 26, 28-31, 33, 35, 36, 39, 40]

In total, 15 of the studies described directly ranking and assessing survey-generated uncertainties from a longlist ranging from 50 to 226 uncertainties.[1, 4, 7, 13, 14, 16, 18, 20, 21, 27, 28, 32, 35, 37, 39]

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

Interim priority setting

Interim prioritization is the stage at which the longlist of treatment uncertainties (indicative questions) is reduced to a shortlist for the final priority setting workshop.[5]

All studies described an interim stage, using the terms: interim priority setting;[7, 17] interim prioritization;[1, 4, 38] and ranking exercise.[16, 39]

Their shortlists varied from 22 [14] to 30 treatment uncertainties.[19, 28-32, 34] Fourteen of the studies used an interim prioritization of their Top 25 that were taken to a final prioritization workshop, where the participants agreed on their Top 10 priorities.[1, 6, 13, 16, 20-27, 33, 36] Three of the studies did not describe the number of shortlisted treatment uncertainties.[12, 18, 39]

Different groups conducted the interim step using different approaches. To reduce the number of uncertainties, an interim prioritization exercise was conducted over email or by post.[4, 17, 30] Patients, carers and health professionals were initially invited to examine the longlist;[30] 12 of the studies used a second online survey [1, 13, 17, 21-26, 31, 32, 36] and in one study the steering group members facilitated an interim ranking exercise.[34]

Final priority setting

The JLA's final stage is a rank ordering of the uncertainties, with a particular emphasis on the Top 10 priorities list. 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 considered.[5]

Nineteen of the studies used the NGT in the final priority setting workshop.[4, 14, 15, 17, 19-21, 23-25, 30-35, 37, 38, 40]

All of the studies implemented a final priority setting workshop to agree upon their Top 10 priorities. In most of the studies, these final workshops included patients, carers and clinicians; nine studies mentioned only including patients and clinicians.[6, 21-25, 37-39]

DISCUSSION

To our knowledge, this is the first scoping review of published studies using the JLA approach. We have described these studies' characteristics, PSP configurations, how they gathered and verified treatment uncertainties, and how they agreed upon their Top 10 priorities. Although the number of steps used by PSPs differed, overall their steps incorporated the same procedural content. This scoping review thus provides unique insight into a broad and varied range of perspectives on PPI using the JLA approach. User groups represent patients' health conditions/disease, carers who follow these patients across disease progression and clinicians who treat the patients and follow up with carers. 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.[16, 21, 36] The health conditions addressed in these studies were primarily somatic diseases, though one study was about life after stroke and included mental health.[4] Thus, the

JLA approach is an appropriate and important method for defining research from the end-users' (i.e., patients' and carers') viewpoints.[41]

The authors of these studies argue that many basic research endeavours do not lead to results that are useful to the end-user. When patients, carers and those representing the health care system express what is of greatest importance to them, they collectively provide a broad evidence base related to implementing the JLA principles in health research. Our findings are consistent with those reported by Crowe et al.,[42] that there has been a critical mismatch between the treatments that patients and clinicians want to have evaluated and the treatments actually being evaluated by researchers.

A key value informing such partnerships is often described as equality. Equitable partnerships might be defined as a gradation of shared responsibility negotiated in a collaborative and co-operative decision-making environment. Whether these 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 negative impacts of public involvement.[43]

The number of priority setting exercises in health research is increasing;[44] our review indicates that use of the JLA approach is also increasing. This approach facilitates broad stakeholder involvement, is transparent and easy to replicate. This is consistent with findings by Sachiyo,[44] who stated that there is a clear need for transparent, replicable, systematic and structured approaches to research priority setting in order to assist policymakers and research funding agencies in making investments. Increased public involvement can lead to a wider range of identified and prioritized research topics that are more relevant to service users.[45] If research addresses the questions most relevant to patients and clinicians, decision makers will be better equipped to design and deliver health services that meet those needs. 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 organizations.[1] The number of resulting uncertainties reflects this breadth. The studies tended to conclude that the JLA principles were welcomed, but consistently emphasized the need for an even broader understanding, better conceptualisation and improved processes to incorporate the results into research. To this end, 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 socio-economic groups and most vulnerable patients. Many respondents, particularly those associated with charity organizations, are likely to be white, middle class and have high education attainment levels. At the same time, individuals who are more difficult to reach, such as those in low socio-economic groups and who are vulnerable patients, may have the greatest unmet needs and stand to gain the most from improved treatment.[14, 26, 31, 38] In one study, 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.[4] Although great efforts were reportedly,[31] made to include participants from black and minority ethnic groups and care home populations, they were not particularly successful. According to Lough et al.,[23] use of an online survey may introduce a bias in favour of patients who use the internet and social media. It is also unlikely that those with literacy issues will participate.[15] Three of the studies,[4, 30, 38] attempted to facilitate participation among those with language barriers and literacy issues. Stephens et al.,[29] have pointed out another major challenge to involving users in research: involving patients on the steering group who have incapacitating symptoms and short expected survival durations. Yet another important issue is that all but two studies [39, 40] were from English-speaking countries and thus represent a relatively limited global population.

What motivates patients, carers and clinicians to participant in PSP research? Individualistic motivations include increased confidence and a chance to have an input. Collectivistic motivations include 'helping through being part of something bigger'. Mobilisation, including a desire for change and 'being asked', are also important. In fact, 'being asked' was unanimously agreed upon as the best motivator for participation, while a sense that research results were not communicated to participants was a strong negative factor against future participation.[46] According to the JLA Guidebook,[5] PSPs usually report their process and methods, the participants involved, results, reflections on successes, lessons learned or limitations and next steps. It is important that these reports be written in language understandable to everyone with an interest in the topic, not just to clinicians. Lough et al.,[23] 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, these reports can be difficult to obtain by those without ready online access or those with literacy issues. Eleftheriadou et al.,[16] included implementation of a feasibility study as one of their Top 10 priorities and hoped that, following its publication along with their list of most important treatment uncertainties, relevant studies would be developed.

Running a PSP and involving relevant stakeholders in deciding which research should be funded seem to be an effective and sustainable model.[27] Without doubt, the essential advantage is integration of this involvement in both research and health care. Identifying research priorities is perhaps where the PSP's greatest effect can be achieved.[14] Nevertheless, one might ask whether PSPs emphasize basic research less than applied research. Abma et al.,[47] 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.[17, 24] Based on cumulative data, there has been a clear paradigm shift from a reactive to a more proactive approach described as 'predictive, personalized, preventative and participatory'.[31] As such, it is expected that the JLA process will have a clinical impact by driving relevant research studies based on PPI.

STRENGTH AND LIMITATION

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 operationalize 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. Finally, while a quality analysis was beyond the scope of this paper, we have noted varying descriptions within the selected studies (i.e., sample sizes, health status and age of groups).

CONCLUSION

JLA-based PSP makes a useful contribution to identifying research questions. Through this process, patients, carers and clinicians work together to identify and prioritize unanswered treatment uncertainties. One inherent limitation to this process is that the weakest voices often

lack representation. For the method to best represent those with different health conditions, representatives must have the capacity and resources to participate. Finally, it is important that the results of these studies, including the Top 10 priorities, reach those who answered the survey—including those whose online access may be limited. Future studies should focus on factors influencing patient and carer involvement in priority setting projects.

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Author Contributions

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 and all authors critically reviewed the manuscript. All authors read and approved the manuscript.

Competing interests

None.

Data sharing statement

No additional data are available

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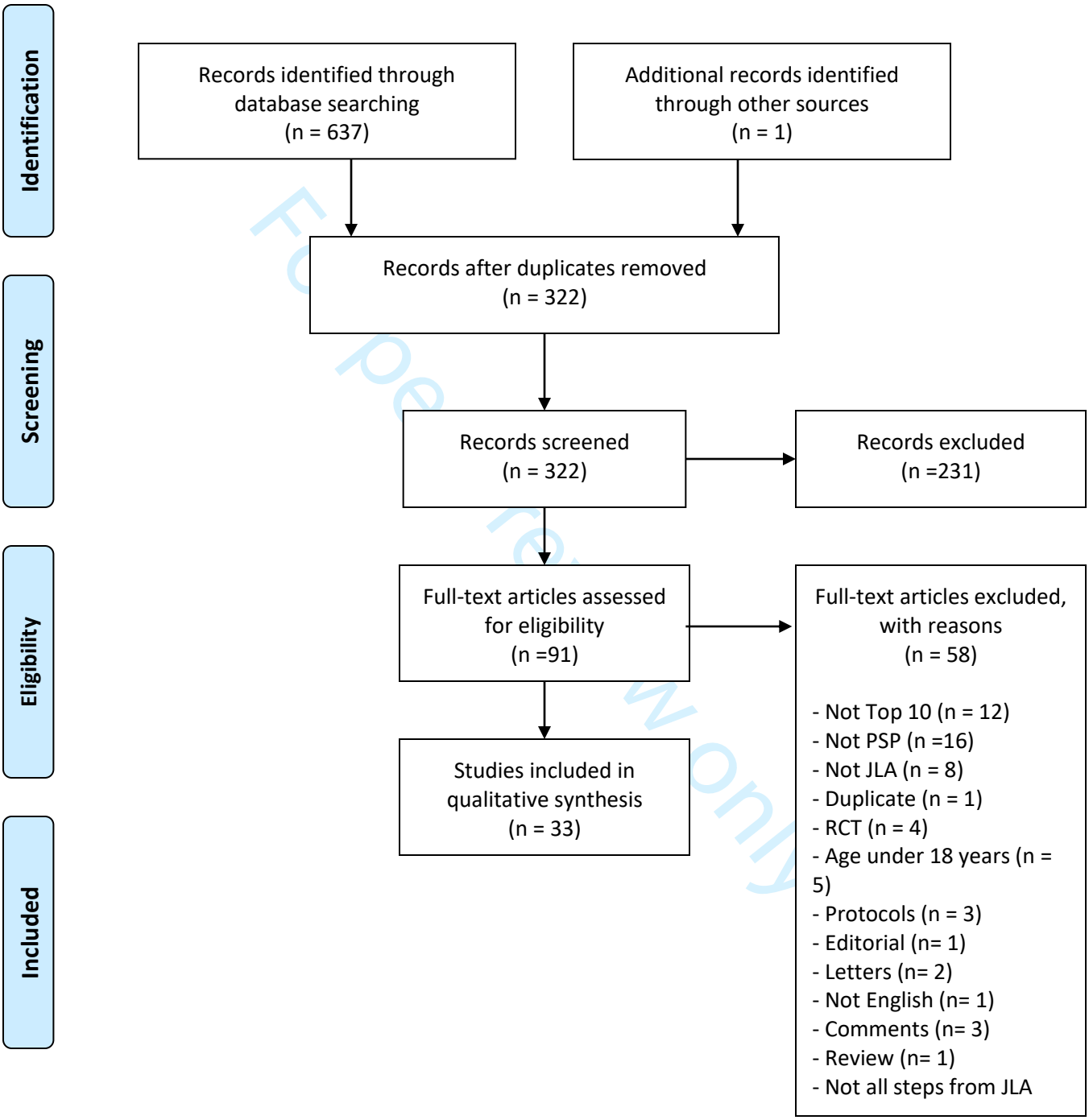
Figure 1 approximately here. FLOW CHART.

PRISMA 2009 Flow Diagram



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Reporting checklist for systematic review and meta-analysis.

Based on the PRISMA guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

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Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement

		Reporting Item	Page Number
	#1	Identify the report as a systematic review, meta-analysis, or both.	1
Structured summary	#2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis	3

		methods; results; limitations; conclusions and implications of key findings; systematic review registration number	
Rationale	#3	Describe the rationale for the review in the context of what is already known.	4-5
Objectives	#4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	5
Protocol and registration	#5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address) and, if available, provide registration information including the registration number.	6
Eligibility criteria	#6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rational	6-7
Information sources	#7	Describe all information sources in the search (e.g., databases with dates of coverage, contact with study authors to identify additional studies) and date last searched.	6
Search	#8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	#9	State the process for selecting studies (i.e., for screening, for determining eligibility, for inclusion in the systematic review, and, if applicable, for inclusion in the meta-analysis).	7

Data collection process	#10	Describe the method of data extraction from reports (e.g., piloted forms, independently by two reviewers) and any processes for obtaining and confirming data from investigators.	6
Data items	#11	List and define all variables for which data were sought (e.g., PICOS, funding sources), and any assumptions and simplifications made.	7-8, 30
Risk of bias in individual studies	#12	Describe methods used for assessing risk of bias in individual studies (including specification of whether this was done at the study or outcome level, or both), and how this information is to be used in any data synthesis.	7, 29
Summary measures	#13	State the principal summary measures (e.g., risk ratio, difference in means).	See note 1
Planned methods of analyis	#14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I2) for each meta-analysis.	See note 2
Risk of bias across studies	#15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	See note 3
Additional analyses	#16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	See note 4

Study selection	#17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6
Study characteristics	#18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citation.	9-20
Risk of bias within studies	#19	Present data on risk of bias of each study and, if available, any outcome-level assessment (see Item 12).	7, 29
Results of individual studies	#20	For all outcomes considered (benefits and harms), present, for each study: (a) simple summary data for each intervention group and (b) effect estimates and confidence intervals, ideally with a forest plot.	21-25
Synthesis of results	#21	Present the main results of the review. If meta-analyses are done, include for each, confidence intervals and measures of consistency.	3
Risk of bias across studies	#22	Present results of any assessment of risk of bias across studies (see Item 15).	N/A see 15
Additional analysis	#23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A see 16
Summary of Evidence	#24	Summarize the main findings, including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., health care providers, users, and policy makers	25-29

Limitations	#25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review level (e.g., incomplete retrieval of identified research, reporting bias).	29
Conclusions	#26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	29-30
Funding	#27	Describe sources of funding or other support (e.g., supply of data) for the systematic review; role of funders for the systematic review.	30

Author notes

1. N/A not relevant in this scoping review

2. N/A not relevant in this scoping review

3. N/A not relevant in this scoping review

4. N/A not relevant in this scoping review

BMJ Open

The James Lind Alliance Process approach: A scoping review

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1

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The James Lind Alliance Process approach: A scoping review

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Running head: James Lind Alliance approach

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ABSTRACT

Objective: To summarize 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 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 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 (e.g., 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 8,227 submitted by 2,587 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 prioritize

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.

Article Summary

Strengths and limitations of this study

- This is the first scoping review of published studies using the JLA approach.
- This approach provides for large-scale involvement of patients, carers and the public in setting the research agenda.
- Top 10 priority list may lead to future research that addresses issues of importance for the clinical management of different diseases.
- Very few participants were from minority ethnic populations, which could limit the generalizability of these priorities to these populations.
- The weakest voices often lack representation, which could limit the generalizability of these priorities to these populations.

We were not in contact with the JLA Coordinating Centre and all relevant literature, such as grey literature and studies which do not described JLA.

Keywords: James Lind Alliance, Priority Setting Partnership, Patient and Public Involvement, patient involvement in research.

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 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 health care 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 United Kingdom-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 prioritize 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] uncertainties and how to prioritize these are key features of the JLA process. The process begins by defining unanswered questions (i.e., 'uncertainties') about the effects of treatment and health care—questions that cannot be adequately answered based on existing research evidence such as reliable, up-to-date systematic reviews—and then prioritizes 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 prioritization 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, categorizing, and summarizing 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]

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, summarize uncertainties and determine the lists of Top 10 list priorities. Thus, the objective of this scoping review is to summarize 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. Specifically, we summarize the following study details: The year of publication, the authors, the country; the aims of the study; user groups, the version of the JLA guidebook used; and the age of patients and their health conditions/diseases. In addition, we examine the processes that are used to gather and verify uncertainties. We investigate the number of initial uncertainties submitted according to the number of participants and the group they represent as well as information about characteristics of the

steering group and whether they reported use of the JLA facilitator. Lastly, we identify the number of steps in the PSP, describe these steps and note whether they referred to use NGT.

METHODS

Insert here: Figure 1 approximately here. FLOW CHART.

Identifying relevant studies

A systematic search was conducted up until 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 Flow chart, figure 1.

Selecting relevant studies

A pre-screening 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.

Table 1 Criteria for inclusion and exclusion

Inclusion criteria	Exclusion criteria
• All steps from James Lind Alliance • List of Top 10 priorities • Adults (aged > 18 years or older)	• Unpublished literature • Articles not written in English • Priority Setting Partnership without James Lind Alliance • James Lind Alliance without Priority Setting Partnership • Protocols • Errata • Editorial • Thesis • Comments • Review • Guidelines • Randomized controlled trials (RCT)

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 pre-defined procedure was developed for consulting a third author, or the whole research team, in cases of discrepancies; however, this was never necessary (i.e., decisions to accept or reject unclear articles were based on 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 involved.

Collating, summarizing 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; (5) final priority setting. The results are presented based on the JLA Guidebook steps, which have remained consistent across versions.[6, 9-11]

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Table 2 Characteristics of included studies

Year Author Country	Aim of the study	1. User group* 2. James Lind Alliance (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	Steering group*** identification and management of partners/stakeholders	JLA The role of the facilitator/ Advisor	Priority Setting Partnership (PSP) Number of steps Description of stages Nominal Group Technique (NGT)
2010 Buckley et al. [12] United Kingdom (UK)	Identify and prioritize ''clinical uncertainties'' relating to treatment of urinary incontinence (UI)	1.Patients, carers, clinicians 2. Not reported (NR) 3. Age ≥40 years 4. UI 5. In total, 494, "raw" treatment uncertainties	Organizations were identified which represented or could advocate for: patients their informal carers and clinicians involved in the treatment or management	Not reported (NR)	5 steps + NGT 1. Initiation 2. Consultation 3. Collation 4. Prioritization 5. Dissemination
2011 Eleftheriadou et al. [13] (UK)	Stimulate and steer future research in the field of vitiligo treatment, by identifying the 10 most important research areas for patients and clinicians	1. Patients, carers, clinicians and researchers 2. JLA Guidebook 2010, version 4 3 NR 4. Vitiligo 5. In total, 660 treatment uncertainties submitted by 461 participants	Professional bodies and patient support groups. Steering group included 12 members with knowledge and interest in Vitiligo	The Vitiligo PSP adopted the methods advocated by the JLA, which were refined to meet the needs of this particular PSP	5 steps 1. Initiation 2. Consultation 3. Collation 4. Ranking exercise (Interim prioritization exercise) 5. Final Prioritisation Workshop
2012 Gadsby et al. [14] UK	Collect uncertainties about the treatment of Type 1 diabetes from patients, carers and health professionals, and to collate and prioritize these uncertainties to develop a list of Top 10 of research priorities.	1. Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Type I diabetes 5. In total, 1,141 treatment uncertainties submitted by 583 participants	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 organizations (n = 9 plus an independent information specialist) and partner organizations	JLA by being represented on the steering group	6 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 et al. [15] UK	Identify the uncertainties in eczema treatment that are important to patients who have eczema, their carers and the health care professionals who treat them	1. Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Eczema 5. In total 1,070 treatment uncertainties submitted by 493 participants	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	The PSP was coordinated from the Centre of Evidence-Based Dermatology in Nottingham, with oversight by a representative of a JLA, who was the independent chair of the PSP steering group	5 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
2013 Davila-Seijo et al. [16] Spain	Describe and prioritize the most important uncertainties about Dystrophic Epidermolysis Bullosa treatment shared by patients, carers and health care professionals in order to promote research in those areas	1.Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. Age 21- 54 years 4. Dystrophic Epidermolysis Bullosa 5. In total 323 treatment uncertainties submitted by 58 participants	The steering group comprised eight people including patients/carers, a representative from the Dystrophic Epidermolysis Bullosa Research Association Spain, a clinician; dermatologists and nurses and researchers/ and the Spanish Academy of Dermatology and Venereology	Workshop advocated by the JLA	5 steps + NGT 1. Initiation 2. Consultation survey: collection of treatment uncertainties 3. Ranking exercise 4. Ranking exercise 5. Final prioritization workshop
2013 Hall et al. [7] UK	Describe the Tinnitus PSP in providing a platform for patients and clinicians to collaborate to identify and prioritize uncertainties or 'unanswered questions'	1. Patients and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Tinnitus 5. In total, 2,483 treatment uncertainties submitted by 825 participants	Membership of the steering group provided a broad representation of people from the field of Tinnitus , including professional bodies, charities and advocators for people with tinnitus. The wider working partnership included 56 major UK stakeholders including individual advocators for people with Tinnitus, support groups, hospital centres and commercial organizations	Independent chairperson, representing the JLA	7 steps 1. Establishing a working partnership 2. Gathering suggestions for research on the assessment, diagnosis and treatment of tinnitus 3. Checking and categorizing submitted uncertainties 4. Prioritizing the uncertainties

					5. Developing consensus 6.Top 10 clinical research questions 7.Recommendations for future research strategy
2014 Deane et al. [17] UK	Identify and prioritize the Top 10 evidential uncertainties that impact on everyday clinical practice for the management of Parkinson's disease	1. Patients, carers, family, friends, clinicians 2. JLA guidebook 2013, version 5 3. NR 4. Parkinson's disease 5. In total, 4,100 treatment uncertainties submitted by 1,000 participants	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	5 steps + NGT 1. Initiation 2. Consultation 3. Uncertainties survey 4. Collation 5. Priorization
2014 Ingram et al. [18] UK	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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Hidradenitis suppurativa 5. In total, 1,495 treatment uncertainties submitted by 371 participants	The steering committee included five patients and carers, including two representatives of the Hidradenitis Suppurativa Trust UK patient organization, 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	Three JLA facilitators or four facilitators	5 steps + NTG 1. Identify stakeholders 2. Invitation to submit uncertainties 3. Generate "indicative uncertainties" 4. Rank uncertainties 5. Final workshop
2014 Manns et al. [19] Canada	Improve understanding of kidney function and disease, including for specific areas, such as dialysis therapies	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. Age 18 to > 80 years 4.Patients on or near dialysis	The priority-setting process was initiated with the formation of an 11-person steering group that included patients, a caregiver, clinicians, an employee of the Kidney Foundation of Canada, and an expert in the	Experienced facilitators	5 steps + NGT 1. Survey 2. Collation 3. Combining 4. Interim prioritization 5. Final workshop

		5. In total, 1,820 treatment uncertainties submitted by 317 respondents	JLA approach		
2014 Pollock et al. [5] UK	Identify the Top 10 research priorities relating to life after stroke, as agreed by stroke survivors, carers and clinicians	1. Patients, carers, clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Life after stroke 5. In total, 548 treatment uncertainties	A steering group comprising a stroke survivor, carers, a nurse, a physician, allied clinicians, a researcher and representatives from key national stroke charities/patient organizations, 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	The facilitators were briefed by members of the JLA on the importance of ensuring equitable participation of all group members	6 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 et al. [20] UK	Identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians	1. Patients, carers and clinicians 2.JLA Guidebook 2013, version 5 3. Average age of participants= 65.7 years 4. Sight loss or an eye condition 5. In total, 4,461 treatment uncertainties submitted by 2,220 participants	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	Representative from the JLA convened meetings of the steering committee	5 steps + NGT 1. Establishing the Sight Loss Vision PSP 2. Survey 3. Data assessment 4. Interim prioritization 5. Final prioritization
2014 Uhm et al. [21] UK	Discover the research questions for preterm birth and grade them according to their importance for infants and families	1. Patients, carers and clinicians 2. NR 3. NR 4. Preterm birth 5. In total, 593 research questions submitted by 386 people	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	5 steps + NGT 1. Initiation of the partnership 2. Identifying treatment uncertainties 3. Collation: refining questions and uncertainties 4. Prioritization – interim and final stages.

					5. Publicity and publishing results
2015 Barnieh et al. [22] Canada	Assess the research priorities of patients on or nearing dialysis within Canada and their carers and clinicians	1. Patients carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. On or nearing dialysis 5. In total, 1,820 treatment uncertainties number of participants not reported	The 11- persons 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	4 steps + NGT 1. Form PSP 2. Gather research uncertainties 3. Process and collate submitted research uncertainties 4. Final priority - setting workshop
2015 Boney et al. [23] UK	Identify research priorities for anaesthesia and perioperative medicine	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Anaesthesia and perioperative medicine. 5. In total, 1,420 treatment uncertainties submitted by 623 participants	The steering group comprised representatives of the funding partner organisations, patients and carers and the JLA Almost 2,000 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	8 steps 1. Enrol partner organizations 2. Identify research questions 3. Classify and refine research question 4. Short-listing 5. Literature review 6. Interim prioritization 7. Final prioritization 8. Publication and dissemination of results
2015 Kelly et al. [24] UK	Identify unanswered questions around the prevention, treatment, diagnosis and care of dementia with the involvement of all stakeholders identify a Top 10prioritized list of uncertainties	1. Patients, carers/relatives, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Dementia 5. In total, 1,563 uploaded surveys	Potential partner organizations 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.	6 steps + NGT 1. Involvement of potential partner organisations 2. Identifying uncertainties 3. Question management and analysis 4. Verifying uncertainties 5. Interim prioritization

					6. Final prioritization workshop
2015 Stephens et al. [25] UK	Identify the Top 10 research priorities relating to mesothelioma (pleural or peritoneal), specifically, identify those unanswered questions that involved an intervention	1. Patients, current and bereaved carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Mesothelioma 5. In total, 453 initial surveys	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.	8 steps 1. Establishing a steering group 2. Initial survey questionnaire 3. Reviewing the survey responses 4. Searching 5. Interim prioritization 6. Final priority setting 7. Identified unanswered questions 8. An additional PSP
2016 Knight et al. [26] UK	Identify unanswered research questions in the field of kidney transplantation from end service users (patients, carers and health care professionals)	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Kidney transplantation 5. In total, 497 treatment uncertainties submitted by 183 participants	The steering group included transplant surgeons, nephrologists, transplant recipients, living donors and carers. Additional partner organizations 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 organizations and charities involved in kidney transplantation were contacted about the project and invited to contribute to a steering group	The steering group was chaired by an experienced advisor from the JLA	5 steps + NGT 1. Organization and scope 2. Identification of potential research questions 3. Refinement of questions and identification of existing literature 4. Interim prioritization 5. Final prioritization workshop
2016 Rangan et al. [27] UK	To run a UK based JLA PSP for 'Surgery for Common Shoulder Problems'	1. Patients, carers and clinicians, 2. JLA Guidebook 2013, version 5 3. NR 4. Shoulder surgery 5. In total, 652 treatment uncertainties submitted by 371 participants	The steering group was made up of the most relevant stakeholders and included patients, physiotherapists, GP, shoulder surgeons, anaesthetists and pain control experts, orthopaedic nurses and an academic clinician	A JLA adviser	5 steps 1. Identification and invitation of potential partners 2. Initial meeting/ awareness raising 3. Identifying treatment uncertainties

15

			National networks and interest organizations		4. Refining questions and uncertainties 5. Prioritization interim and final
2016 Van Middendorp et al. [1] UK	Identify a list of Top 10 priorities for future research into spinal cord injury	1. Patient, spouse/partner and clinicians 2. JLA Guidebook 2013, version 5 3. Age 18-80 years 4. Spinal cord injury 5. In, total, 784 treatment uncertainties submitted by 403 participants	The steering group comprised representatives from each stakeholder organization, including an independent information manager. Stakeholders included consumer organizations, clinician societies and carers representatives	Support and guidance were provided by the JLA	4 steps 1. Gathering of research questions 2. Checking of existing research evidence 3. Interim prioritization 4. Final consensus meeting
2016, Wan et al. [28] UK	Establish a consensus regarding the Top 10 unanswered research questions in endometrial cancer	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Endometrial cancer 5. In total, 786 individual submissions from 413 participants	As part of the JLA process, all organizations that could reach and advocate for patients, carers and clinicians were invited to become involved in a PSP. A steering group composed of representatives from these groups was then formed to ensure the study remained inclusive and fulfilled its aim to deliver and publicize a list of shared research priorities. A group of 23 stakeholders was constituted, but was not described in details	An independent advisor from the JLA was Chair of the steering group	6 steps + NGT 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 et al. [29] 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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Gastro-oesophageal reflux disease and Barrett's oesophagus 5. In total, 629 treatment uncertainties submitted by 170 participants	Professionals, patients and charity representatives formed a steering committee. The steering committee, which identified the broader. Priorities. The British Society of Gastroenterology, National Health Service, the University of Manchester, the Association of Upper Gastrointestinal Surgeons and the Primary Society for Gastroenterology	NR.	5 steps + NGT 1. Initial survey 2. Initial response list 3. Longlist generation and verification 4. Interim prioritization survey 5. Final workshop

2017, Fitzcharles et al. [30] Canada	Priorities of uncertainties for the management of fibromyalgia (FM) that could propel future research	1.Patients, carers and clinicians 2.JLA Guidebook 2013, version 5 3. Age 18 to >70 years 4. Fibromyalgia 5. In total, 4,557 treatment uncertainties submitted by 550 participants	The steering committee was composed of five patients (one patient was a practicing pharmacist), five health care professionals (one family physician, two rheumatologists, one psychologist, one internist), an internist with previous experience of the JLA process but without specific interest in FM, and a rheumatologist	Facilitators with experience of the JLA process	5 steps 1. Survey results 2. In scope uncertainties 3. Coding uncertainties 4. Interim prioritization 5. Final workshop
2017, Hart et al. [31] UK	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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Inflammatory bowel disease. 5. In total, 1,636 treatment uncertainties submitted by 531 participants	A steering committee was established following an initial explanatory meeting and included two patients, two gastro-enterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organization Crohn's and Colitis UK, a representative of the JLA and an administrator	A JLA facilitator	5 steps 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 Top 10 priorities
2017, Hemmelgarn et al. [32] Canada	Identify the most important unanswered questions (or uncertainties) about the management of chronic kidney disease (CKD) i.e. in terms of diagnosis, prognosis and treatment.	1. Patients, carers, clinicians and policy-makers 2. JLA Guidebook 2013, version 5 3. Age 65 ≥ years 4. Non-dialysis CKD 5. In total 2,241 treatment uncertainties submitted by 439 participants	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 organization for patients with kidney disease)	Jointly organized PSP broadly adhering to the JLA Guidebook	4 steps + NGT 1. Identification and invitation of potential partners 2. Collection of research uncertainties through a national survey 3. Refinement and prioritization 4. Priority setting-workshop
2017, Khan et al. [33] Canada	Identify the 10 most important research priorities of patients,	1. Patients, carers and clinicians	Steering committee of 15 volunteer patients, carers, and clinicians from across Canada.	JLA facilitator from the UK	5 steps 1. Establishing a steering group

	carers and clinicians for hypertension management	2. JLA Guidebook 2013, version 5 3. NR 4. Hypertension 5. In total 673 individual research questions submitted by 386 participants	Stakeholder not reported in detail		2. Forming priority setting partnerships 3. Collecting potential research questions 4. Processing, categorizing, and summarizing those research questions 5. Selecting the Top 10 research priorities
2017, Jones et al. [34] Canada	Identify unanswered questions encountered during management of kidney cancer agreement by consensus on a prioritized list of the Top 10 shared unanswered questions and establish corresponding research priorities	1. Patients, carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patients with kidney cancer 5. In total 2,004 treatment questions submitted by 225 participants	A 15 persons steering group was formed with seven patients/carers and seven 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	5 steps 1. Formation of steering group 2. Identifying treatment questions 3. Collating questions 4. Interim ranking of questions 5. Final priority-setting workshop
2017, Lomer et al. [35] UK	Provide a comprehensive summary of the research priority findings relating to diet in the treatment of inflammatory bowel disease	1. Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Dietary treatment of inflammatory bowel disease. 5. In total 1,671 treatment uncertainties submitted by 531 participants	Steering committee comprising of two patients. two gastro-enterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organization, Crohn's and Colitis UK, a representative of the JLA and an administrator (i.e., 13 persons steering committee). Stakeholders from various roles, ages and ethnic groups	A representative of the JLA and an administrator on the steering committee.	5 steps 1. Steering committee 2. Questionnaire survey 3. Remaining uncertainties were reviewed 4. Uncertainties determined 5. Final workshop of the steering group
2017, Macbeth et al. [36] UK	Identify uncertainties in alopecia areata management and treatment that are	1. Patients, partners/parents/ carers and clinicians	Four people representing various patient support groups, four dermatologists and two further individuals to represent the BHNS	A JLA representative provided independent oversight of the	5 steps + NGT 1. Identification and invitation of potential partners

	important to both service users, people with hair loss, carers/ relatives and clinicians	2. JLA Guidebook 2016, version 6 3. NR 4. Alopecia areata 5. In total 2,747 treatment uncertainties submitted by 912 participants	and the European Hair Research Society; an academic psychologist; a registered trichologist and a GP and a JLA representative. Two separate steering groups	PSP and chaired the steering group	2. Invitation to submit uncertainties 3. Collation 4. Ranking of treatment uncertainties 5. Final workshop
2017, Narahari et al. [37] India	Summarizes the process of Lymphedema PSP, discussion during the final prioritization workshop, and recommendation on the Top 7 priorities for future research in lymphedema and a brief road map	1. Patients, theorist and nurses 2. JLA Guidebook 2013, version 5 3. NR 4. Lymphedema 5. In total, 137 respondents uploaded research- priorities	The Faculty of Applied Dermatology and the Central University of Kerala participated in the coordinating committee	NR	8 steps 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 lymphedema medical camp 8. Final prioritization workshop
2017 Prior et al. UK	Identify and prioritize important research questions for miscarriage	1. Patients, partners, family members, friends or colleagues and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Miscarriage 5. In total, 3,279 questions submitted by 2,122 participants	The steering group was a balanced composition of women charities that represented them and clinicians. Some members representing charities or clinicians also had personal experience of pregnancy loss	The workshop was chaired by an independent JLA facilitator	6 steps 1. Initiation 2. Consultation 3. Identifying uncertainties 4. Refining uncertainties 5. Interim prioritization 6. Final workshop
2017 Rees et al. [38] Canada	Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus	1. Patients, friends and relatives and clinicians 2. JLA Guidebook 2013, version 5 3. Age18-69 years 4. Gestational diabetes mellitus	A steering committee consisting of three patients and three clinicians (one family physician who practises intrapartum care, an endocrinologist and a neonatologist); a facilitator familiar with the JLA process and a	A facilitator familiar with the JLA process.	4 steps + NGT 1. Survey 2. Process and collate 3. Interim ranking 4. Priority setting-workshop

		5. In total, 389 treatment uncertainties submitted by 75 participants	project manager. The Diabetes Obesity and Nutrition Strategic Clinical Network with the Alberta Health Services supported this research. Stakeholders not reported.		
2017 Smith et al. [39] UK	Prioritize research questions in emergen medicine in a consensus process to determine the Top 10 questions	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Emergency medicine 5. In, total 214 number of initial uncertainties	The steering group members are not reported with titles but consist of 16 members. The Royal College of Emergency Medicine	NR.	6 steps 1. Online submissions 2. Working group reviews 3. Mini systematic reviews 4. Working group prioritisation exercise 5. Public prioritization exercise 6. Face- to- face final prioritization
2018 Fernandez et al. [40] UK	Establish the research priorities for adults with fragility fractures of the lower limb and pelvis that represent the shared interests and priorities	1.Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. Age ≥ 60 years 4. Fragility fractures of the lower limb and pelvis 5. In total, 963 treatment uncertainties submitted by 365 participants	The steering group consisted of patient representatives, healthcare professionals and carers with established links to relevant partner organizations to ensure that a range of stakeholder groups were represented.	A JLA adviser supported and guided the PSP	5 steps 1. First survey 2. Screening 3. Thematic analysis. original uncertainties turned into over-arching indicative questions 4. Evidence search interim prioritazion 5. Final workshop
2018 Finer et al. [41] UK	Describe processes and outcomes of a PSP to identify the Top 10 research priorities' in Type 2 diabetes	1.Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4.Type 2 diabetes 5. In total, 8,227 treatment uncertainties were submitted by 2,587 participants	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 dialectologists), an information specialist, seven members of the Diabetes UK research and senior leadership team, and a JLA senior advisor. The steering group (47% men and	The workshop was facilitated by trained JLA advisors	4 steps + NGT 1. Gathering uncertainties 2. Organizing the uncertainties 3. Interim priority setting 4. Final priority setting

			53% women and 26% from black and minority ethnic groups) met 12 times during the PSP process, in person or by teleconference Diabetes UK		
2018 Lechelt et al. [42] Canada	Identify the Top 10 treatment uncertainties in head and neck cancer from the joint perspective of patients, caregivers, family members, and treating clinicians	1. Patients, carers, family members, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patient with head and neck cancer 5. In total, 818 treatment uncertainties submitted by 161 participants	The steering committee included five patients with head and neck cancer who were from 3 - 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 oncologic 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 co-facilitators, all of whom were briefed by the JLA senior advisor on recommended JLA protocols	5 steps + NGT 1. Initial survey development and deployment 2. Identifying uncertainties through survey data processing 3. Verifying uncertainties 4. Interim prioritization 5. Final workshop
2018 Lough et al. [43] UK	Identify the shared priorities for future research of women affected by and clinicians involved with pessary use for the management of prolapse	1. Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. Age 30-89 years 4. Pessary use in women with prolapse 5.In total, 669 questions submitted by 210 participants	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 (UKCS) research grant, two grants from the Pelvic Obstetric and Gynaecological Physiotherapy	The steering group agreed the terms of reference and protocol for the JLA adviser and project leader	4 steps + NGT 1. Gathering questions/ uncertainties 2. Refining the questions and checking the evidence 3. Prioritizing /ranking the questions 4. Choosing the Top 10 priorities by consensus

			group (POGP) of the Chartered Society of Physiotherapy and a funded studentship from Glasgow Caledonian University		
2018 Macbeth et al. [44] UK	Identify uncertainties in hair loss management, prevention, diagnosis and treatment that are important to both people with hair loss and clinicians	1. Patients, carers relatives and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Hair loss (excluding alopecia areata) 5. In total, 2,747 treatment uncertainties were submitted by 912 participants	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	5 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 * User group means the participants who are involved in the PSP process, not only the survey.

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

3 *** Steering group, steering committee and co-ordinating committee are defined as equal concepts.

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RESULTS

In total, 37 studies met the inclusion criteria; their characteristics are summarized 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, 12-15, 17, 18, 20, 21, 23-29, 31, 35, 36, 39-41, 43-45] eight from Canada [19, 22, 30, 32-34, 38, 42] and one each from India [37] and Spain.[16]

The JLA process participants were patients, carers and clinicians, aged ≥ 18 years. The studies collectively represented patient groups with heterogeneous ages and health conditions/disease, 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, 17, 19, 21, 24, 26, 28, 29, 31, 33, 38, 41, 43, 45] One of the studies also gave information about socio-economic status. [29] Another study gave only information about socio-economic status. [37]

Compared with clinicians, patients and carers contributed a greater number of questions on psychosocial issues, psychosocial stress, depression and anxiety.[13, 26, 33] However, 24 other studies also mentioned psychosocial issues without noting who had done so. [1, 7, 14, 15, 17-22, 28-32, 34, 36, 38, 40-45] Ten studies did not mention psychosocial issues. [5, 12, 16, 23-25, 27, 29, 35, 37] The types of health conditions that were addressed included gastrointestinal,[29, 31, 35] neurologic,[1, 5, 7, 17, 24, 30] dermatologic,[13, 15, 16, 18, 36, 44] endocrine [14, 38, 41] and cancer [25, 28, 34, 42] conditions.

Setting up a Priority Setting Partnership PSP

The JLA steering group is made up of key organizations 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, 14-19, 21-23, 25, 26, 28, 32-34, 40] included patients, carers and clinicians in their steering groups; 16 studies [7, 13, 20, 27, 29-31, 35-38, 41-45] did not include carers in their steering group (i.e., only patients and clinicians). In one study,[39] the titles of the members on the steering group were not reported; in another, [24] 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, 22, 32, 38, 41, 43] to eight steps.[23, 25, 37] Five steps, corresponding to JLA Guidebook versions 4, 5 and 6, were most common: [12, 13, 15-21, 26, 27, 29-31, 33-36, 40, 42, 44] with Step 1, initiation; Step 2, collecting of uncertainties; Step 3,collation of uncertainties; Step 4,interim priority setting; Step 5final 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 organizations, 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, 38]

The questions were usually deliberately open-ended to encourage full responses regarding the experiences of patients, carers and clinicians. One of the 37 studies [37] 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, 16, 20, 26, 28, 33, 34, 38, 41, 42, 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.[17, 18, 23, 24, 31, 35, 43] In eight studies, no limit was placed on the types of questions that could be submitted.[5, 13, 19, 22, 27, 32, 39, 40] One study asked about eight open-ended questions requesting a narrative answer. [30] Close-ended questions were used in three studies,[25, 36, 44] 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, 21, 29]

The number of submitted uncertainties ranged from 8,227 submitted by 2,587 participants [41] to 323 submitted by 58 participants.[16] All studies except two [7, 37] reported involving patients, carers and clinicians in the initial survey. Two of the studies addressed verifying uncertainties example by content expert or Librarian. [33, 42] The steering group or researchers were involved in addressing verifying uncertainties in 22 of the studies, and [5, 7, 14-17, 20, 22-24, 26-29, 31, 32, 34, 37, 39, 40, 43, 45] in 13 of the studies not describing verifying the uncertainties. [1, 12, 13, 18, 19, 21, 25, 30, 35, 36, 38, 41, 44]

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.,[42] uncertainties are organized 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.,[35] specified that similar uncertainties are combined to create indicative uncertainties. Among our included studies, 20 described refining questions into indicative uncertainties,[5, 13-15, 18, 21, 23, 26, 27, 30-32, 35, 36, 38, 40-44], while 17 did not describe a concept of indicative uncertainties.[1, 7, 12, 16, 17, 19, 20, 22, 24, 25, 28, 29, 33, 34, 37, 39, 45]

In total, 16 of the studies described directly ranking and assessing survey-generated uncertainties from a longlist ranging from 43 to 226 uncertainties.[1, 5, 13, 14, 21, 23, 24, 26, 27, 29-31, 34, 37, 39, 42]

The wording of the longlist 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 prioritization is the stage at which the longlist 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, 41] interim prioritization;[1, 5, 38] and ranking exercise.[13, 37]

Their short lists varied from 22 [29] to 30 uncertainties.[12, 18-21, 25, 28, 32, 39] Sixteen of the studies used an interim prioritization of their Top 25 uncertainties that were taken to a final prioritization workshop, where the participants agreed on their Top 10 priorities.[1, 7, 13, 22-24, 26, 27, 30, 33, 35, 36, 40, 43-45] Three of the studies did not describe the number of shortlisted treatment uncertainties.[15, 31, 37]

To reduce the number of uncertainties, an interim prioritization exercise was conducted by email or post.[5, 20, 41] Patients, carers and health professionals were initially invited to examine the longlist;[20] 14 of the studies used a second online survey [1, 21, 23, 26, 28, 30, 33, 35, 36, 40, 41, 43-45] and in one study the steering group members facilitated an interim ranking exercise.[32]

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 considered [6] 21 of the studies reported use of the NGT in the final priority- setting workshop.[5, 12, 16-22, 24, 26, 28, 29, 32, 34, 36, 38, 41-44]

All of the studies implemented a final priority- setting workshop to agree upon their Top 10 priorities. In most of the studies, these final workshops included patients, carers and clinicians; nine studies mentioned only including patients and clinicians.[7, 26, 35-38, 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, overall they incorporated the same procedural content. Thus, this scoping review thus 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, 26, 33] 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 co-operative 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 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 Sachiyo,[48] who argues that there is a clear need for transparent, replicable, systematic and structured approaches to research priority- setting to assist policymakers and research funding agencies in making investments. Increased public involvement can lead to a

wider range of identified and prioritized 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 organizations.[1] The number of resulting uncertainties reflects this breadth. The studies examined tended to conclude that the JLA principles were welcomed, but consistently emphasized the need for an even broader understanding, better conceptualization 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 socio-economic groups and most vulnerable patients. Many respondents, particularly those associated with charity organizations, are likely to be white, middle class and have high education attainment levels. Yet it is the, individuals who are more difficult to reach, such as those in low socio-economic groups and who are vulnerable patients - may have the greatest unmet needs and stand to gain the most from improved treatment.[28, 29, 38, 45] 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 made ,[28] to include participants from black and minority ethnic groups and care home populations, they were not particularly successful. Lough et al.,[43] 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.[17] Three of the studies,[5, 20, 38] 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.,[25] note another major challenge to involving users in research, involving patients in the steering group who have incapacitating symptoms and short expected survival durations. Another important issue is that all but two studies [16, 37] 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 language understandable to everyone with an interest in the topic, not just to clinicians. Lough et al.,[43] 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.[27] Without doubt, the essential advantage is integration of this involvement in both research and health care. Identifying research priorities is perhaps where the PSP's greatest effect can be achieved.[29] Nevertheless, one might ask whether PSPs emphasize 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.[41, 44] There

has been a clear paradigm shift from a reactive to a more proactive approach described as 'predictive, personalized, preventative and participatory'.[28] 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 operationalize 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 (i.e., 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 amongst 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 on 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 only described in one publication. [27] 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. Through this process, patients, carers and clinicians work together to identify unanswered uncertainties. A range from 327 to 8,227 uncertainties were published from 2010 to 2018, 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 addressed methods for verifying uncertainties and 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 those whose online access may be limited. Future studies should focus on factors influencing patient and carer involvement in priority setting projects.

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Author Contributions

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.

Competing interests

None.

Data sharing statement

All data relevant to the study are included in the article or uploaded as supplementary information.

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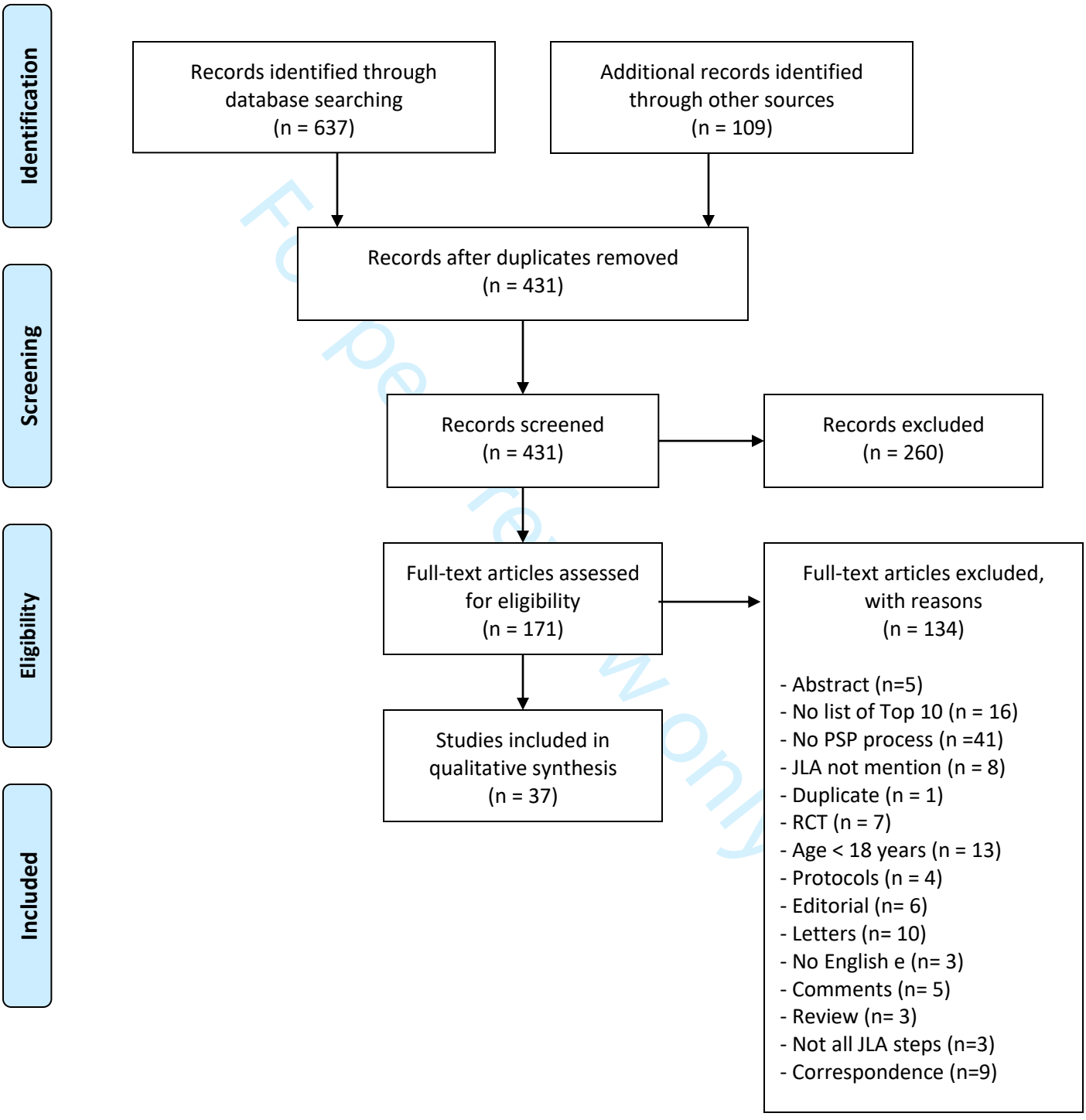
Figure 1 approximately here. FLOW CHART.

PRISMA 2009 Flow Diagram



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PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4-5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	5
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	N/A
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	6-7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	7
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	7-8, 32
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	7, 30
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	See note 1

PRISMA 2009 Checklist

Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	See note 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	See note 3
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	See note 4
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	9-21
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	7, 32
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	22-26
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	3
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/A 15
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A 16
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	27-30
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	30-31
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	31

PRISMA 2009 Checklist

FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	31

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

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BMJ Open

The James Lind Alliance Process approach: A scoping review

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1

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The James Lind Alliance Process approach: A scoping review

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Running head: James Lind Alliance approach

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ABSTRACT

Objective: To summarize 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 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 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 (e.g., 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 8,227 submitted by 2,587 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 prioritize

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.

Article Summary

Strengths and limitations of this study

- This is the first scoping review of published studies using the JLA approach available with involvement of patients, carers and the public in setting the research agenda.
- The weakest voices often lack representation, which could limit the generalizability 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

Keywords: James Lind Alliance, Priority Setting Partnership, Patient and Public Involvement, patient involvement in research.

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 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 health care 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 United Kingdom-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 prioritize 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] uncertainties and how to prioritize these are key features of the JLA process. The process begins by defining unanswered questions (i.e., 'uncertainties') about the effects of treatment and health care—questions that cannot be adequately answered based on existing research evidence such as reliable, up-to-date systematic reviews—and then prioritizes 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 prioritization 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, categorizing, and summarizing 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]

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, summarize uncertainties and determine the lists of Top 10 list priorities. Thus, the objective of this scoping review is to summarize 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 they have operationalized the JLA methods?
- How do the studies describe involvement of different user groups?
- What processes are used to gather and verify uncertainties?

METHODS

Insert here: Figure 1 approximately here. FLOW CHART.

Identifying relevant studies

A systematic search was conducted up until 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 Flow chart, figure 1.

Selecting relevant studies

A pre-screening 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.

Table 1 Criteria for inclusion and exclusion

Inclusion criteria	Exclusion criteria
• All steps from James Lind Alliance • List of Top 10 priorities • Adults (aged > 18 years or older)	• Unpublished literature • Articles not written in English • Priority Setting Partnership without James Lind Alliance • James Lind Alliance without Priority Setting Partnership • Protocols • Errata • Editorial • Thesis • Comments • Review • Guidelines • Randomized controlled trials (RCT)

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 pre-defined procedure was developed for consulting a third author, or the whole research team, in cases of discrepancies; however, this was never necessary (i.e., decisions to accept or reject unclear articles were based on 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 involved.

Collating, summarizing 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; (5) final priority setting. The results are presented based on the JLA Guidebook steps, which have remained consistent across versions.[6, 9-11]

1

Table 2 Characteristics of included studies

Year Author Country	Aim of the study	1. User group* 2. James Lind Alliance (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	Steering group*** identification and management of partners/stakeholders	JLA The role of the facilitator/ Advisor	Priority Setting Partnership (PSP) Number of steps Description of stages Nominal Group Technique (NGT)
2010 Buckley et al. [12] United Kingdom (UK)	Identify and prioritize ''clinical uncertainties'' relating to treatment of urinary incontinence (UI)	1.Patients, carers, clinicians 2. Not reported (NR) 3. Age ≥40 years 4. UI 5. In total, 494, "raw" treatment uncertainties	Organizations were identified which represented or could advocate for: patients their informal carers and clinicians involved in the treatment or management	Not reported (NR)	5 steps + NGT 1. Initiation 2. Consultation 3. Collation 4. Prioritization 5. Dissemination
2011 Eleftheriadou et al. [13] (UK)	Stimulate and steer future research in the field of vitiligo treatment, by identifying the 10 most important research areas for patients and clinicians	1. Patients, carers, clinicians and researchers 2. JLA Guidebook 2010, version 4 3 NR 4. Vitiligo 5. In total, 660 treatment uncertainties submitted by 461 participants	Professional bodies and patient support groups. Steering group included 12 members with knowledge and interest in Vitiligo	The Vitiligo PSP adopted the methods advocated by the JLA, which were refined to meet the needs of this particular PSP	5 steps 1. Initiation 2. Consultation 3. Collation 4. Ranking exercise (Interim prioritization exercise) 5. Final Prioritisation Workshop
2012 Gadsby et al. [14] UK	Collect uncertainties about the treatment of Type 1 diabetes from patients, carers and health professionals, and to collate and prioritize these uncertainties to develop a list of Top 10 of research priorities.	1. Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Type I diabetes 5. In total, 1,141 treatment uncertainties submitted by 583 participants	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 organizations (n = 9 plus an independent information specialist) and partner organizations	JLA by being represented on the steering group	6 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	Identify the uncertainties in	1. Patients, carers and clinicians	The steering group comprised four patients and carers, including a	The PSP was coordinated from the Centre of	5 steps

Batchelor et al. [15] UK	eczema treatment that are important to patients who have eczema, their carers and the health care professionals who treat them	2. JLA Guidebook 2010, version 4 3. NR 4. Eczema 5. In total 1,070 treatment uncertainties submitted by 493 participants	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	Evidence-Based Dermatology in Nottingham, with oversight by a representative of a JLA, who was the independent chair of the PSP steering group	1. Initiation 2. Consultation – collection of treatment uncertainties 3. Collation and treatment uncertainties 4. Ranking of treatment uncertainties 5. Workshop to develop research questions
2013 Davila-Seijo et al. [16] Spain	Describe and prioritize the most important uncertainties about Dystrophic Epidermolysis Bullosa treatment shared by patients, carers and health care professionals in order to promote research in those areas	1.Patients, carers and clinicians 2. JLA Guidebook 2010, version 4 3. Age 21- 54 years 4. Dystrophic Epidermolysis Bullosa 5. In total 323 treatment uncertainties submitted by 58 participants	The steering group comprised eight people including patients/carers, a representative from the Dystrophic Epidermolysis Bullosa Research Association Spain, a clinician; dermatologists and nurses and researchers/ and the Spanish Academy of Dermatology and Venereology	Workshop advocated by the JLA	5 steps + NGT 1. Initiation 2. Consultation survey: collection of treatment uncertainties 3. Ranking exercise 4. Ranking exercise 5. Final prioritization workshop
2013 Hall et al. [7] UK	Describe the Tinnitus PSP in providing a platform for patients and clinicians to collaborate to identify and prioritize uncertainties or 'unanswered questions'	1. Patients and clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Tinnitus 5. In total, 2,483 treatment uncertainties submitted by 825 participants	Membership of the steering group provided a broad representation of people from the field of Tinnitus , including professional bodies, charities and advocators for people with tinnitus. The wider working partnership included 56 major UK stakeholders including individual advocators for people with Tinnitus, support groups, hospital centres and commercial organizations	Independent chairperson, representing the JLA	7 steps 1. Establishing a working partnership 2. Gathering suggestions for research on the assessment, diagnosis and treatment of tinnitus 3. Checking and categorizing submitted uncertainties 4. Prioritizing the uncertainties

					5. Developing consensus 6.Top 10 clinical research questions 7.Recommendations for future research strategy
2014 Deane et al. [17] UK	Identify and prioritize the Top 10 evidential uncertainties that impact on everyday clinical practice for the management of Parkinson's disease	1. Patients, carers, family, friends, clinicians 2. JLA guidebook 2013, version 5 3. NR 4. Parkinson's disease 5. In total, 4,100 treatment uncertainties submitted by 1,000 participants	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	5 steps + NGT 1. Initiation 2. Consultation 3. Uncertainties survey 4. Collation 5. Priorization
2014 Ingram et al. [18] UK	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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Hidradenitis suppurativa 5. In total, 1,495 treatment uncertainties submitted by 371 participants	The steering committee included five patients and carers, including two representatives of the Hidradenitis Suppurativa Trust UK patient organization, 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	Three JLA facilitators or four facilitators	5 steps + NTG 1. Identify stakeholders 2. Invitation to submit uncertainties 3. Generate "indicative uncertainties" 4. Rank uncertainties 5. Final workshop
2014 Manns et al. [19] Canada	Improve understanding of kidney function and disease, including for specific areas, such as dialysis therapies	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. Age 18 to > 80 years 4.Patients on or near dialysis	The priority-setting process was initiated with the formation of an 11-person steering group that included patients, a caregiver, clinicians, an employee of the Kidney Foundation of Canada, and an expert in the	Experienced facilitators	5 steps + NGT 1. Survey 2. Collation 3. Combining 4. Interim prioritization 5. Final workshop

		5. In total, 1,820 treatment uncertainties submitted by 317 respondents	JLA approach		
2014 Pollock et al. [5] UK	Identify the Top 10 research priorities relating to life after stroke, as agreed by stroke survivors, carers and clinicians	1. Patients, carers, clinicians 2. JLA Guidebook 2010, version 4 3. NR 4. Life after stroke 5. In total, 548 treatment uncertainties	A steering group comprising a stroke survivor, carers, a nurse, a physician, allied clinicians, a researcher and representatives from key national stroke charities/patient organizations, 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	The facilitators were briefed by members of the JLA on the importance of ensuring equitable participation of all group members	6 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 et al. [20] UK	Identify research priorities relating to sight loss and vision through consultation with patients, carers and clinicians	1. Patients, carers and clinicians 2.JLA Guidebook 2013, version 5 3. Average age of participants= 65.7 years 4. Sight loss or an eye condition 5. In total, 4,461 treatment uncertainties submitted by 2,220 participants	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	Representative from the JLA convened meetings of the steering committee	5 steps + NGT 1. Establishing the Sight Loss Vision PSP 2. Survey 3. Data assessment 4. Interim prioritization 5. Final prioritization
2014 Uhm et al. [21] UK	Discover the research questions for preterm birth and grade them according to their importance for infants and families	1. Patients, carers and clinicians 2. NR 3. NR 4. Preterm birth 5. In total, 593 research questions submitted by 386 people	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	5 steps + NGT 1. Initiation of the partnership 2. Identifying treatment uncertainties 3. Collation: refining questions and uncertainties 4. Prioritization – interim and final stages.

					5. Publicity and publishing results
2015 Barnieh et al. [22] Canada	Assess the research priorities of patients on or nearing dialysis within Canada and their carers and clinicians	1. Patients carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. On or nearing dialysis 5. In total, 1,820 treatment uncertainties number of participants not reported	The 11- persons 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	4 steps + NGT 1. Form PSP 2. Gather research uncertainties 3. Process and collate submitted research uncertainties 4. Final priority - setting workshop
2015 Boney et al. [23] UK	Identify research priorities for anaesthesia and perioperative medicine	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Anaesthesia and perioperative medicine. 5. In total, 1,420 treatment uncertainties submitted by 623 participants	The steering group comprised representatives of the funding partner organisations, patients and carers and the JLA Almost 2,000 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	8 steps 1. Enrol partner organizations 2. Identify research questions 3. Classify and refine research question 4. Short-listing 5. Literature review 6. Interim prioritization 7. Final prioritization 8. Publication and dissemination of results
2015 Kelly et al. [24] UK	Identify unanswered questions around the prevention, treatment, diagnosis and care of dementia with the involvement of all stakeholders identify a Top 10prioritized list of uncertainties	1. Patients, carers/relatives, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Dementia 5. In total, 1,563 uploaded surveys	Potential partner organizations 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.	6 steps + NGT 1. Involvement of potential partner organisations 2. Identifying uncertainties 3. Question management and analysis 4. Verifying uncertainties 5. Interim prioritization

					6. Final prioritization workshop
2015 Stephens et al. [25] UK	Identify the Top 10 research priorities relating to mesothelioma (pleural or peritoneal), specifically, identify those unanswered questions that involved an intervention	1. Patients, current and bereaved carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Mesothelioma 5. In total, 453 initial surveys	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.	8 steps 1. Establishing a steering group 2. Initial survey questionnaire 3. Reviewing the survey responses 4. Searching 5. Interim prioritization 6. Final priority setting 7. Identified unanswered questions 8. An additional PSP
2016 Knight et al. [26] UK	Identify unanswered research questions in the field of kidney transplantation from end service users (patients, carers and health care professionals)	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Kidney transplantation 5. In total, 497 treatment uncertainties submitted by 183 participants	The steering group included transplant surgeons, nephrologists, transplant recipients, living donors and carers. Additional partner organizations 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 organizations and charities involved in kidney transplantation were contacted about the project and invited to contribute to a steering group	The steering group was chaired by an experienced advisor from the JLA	5 steps + NGT 1. Organization and scope 2. Identification of potential research questions 3. Refinement of questions and identification of existing literature 4. Interim prioritization 5. Final prioritization workshop
2016 Rangan et al. [27] UK	To run a UK based JLA PSP for 'Surgery for Common Shoulder Problems'	1. Patients, carers and clinicians, 2. JLA Guidebook 2013, version 5 3. NR 4. Shoulder surgery 5. In total, 652 treatment uncertainties submitted by 371 participants	The steering group was made up of the most relevant stakeholders and included patients, physiotherapists, GP, shoulder surgeons, anaesthetists and pain control experts, orthopaedic nurses and an academic clinician	A JLA adviser	5 steps 1. Identification and invitation of potential partners 2. Initial meeting/ awareness raising 3. Identifying treatment uncertainties

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			National networks and interest organizations		4. Refining questions and uncertainties 5. Prioritization interim and final
2016 Van Middendorp et al. [1] UK	Identify a list of Top 10 priorities for future research into spinal cord injury	1. Patient, spouse/partner and clinicians 2. JLA Guidebook 2013, version 5 3. Age 18-80 years 4. Spinal cord injury 5. In, total, 784 treatment uncertainties submitted by 403 participants	The steering group comprised representatives from each stakeholder organization, including an independent information manager. Stakeholders included consumer organizations, clinician societies and carers representatives	Support and guidance were provided by the JLA	4 steps 1. Gathering of research questions 2. Checking of existing research evidence 3. Interim prioritization 4. Final consensus meeting
2016, Wan et al. [28] UK	Establish a consensus regarding the Top 10 unanswered research questions in endometrial cancer	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Endometrial cancer 5. In total, 786 individual submissions from 413 participants	As part of the JLA process, all organizations that could reach and advocate for patients, carers and clinicians were invited to become involved in a PSP. A steering group composed of representatives from these groups was then formed to ensure the study remained inclusive and fulfilled its aim to deliver and publicize a list of shared research priorities. A group of 23 stakeholders was constituted, but was not described in details	An independent advisor from the JLA was Chair of the steering group	6 steps + NGT 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 et al. [29] 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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Gastro-oesophageal reflux disease and Barrett's oesophagus 5. In total, 629 treatment uncertainties submitted by 170 participants	Professionals, patients and charity representatives formed a steering committee. The steering committee, which identified the broader. Priorities. The British Society of Gastroenterology, National Health Service, the University of Manchester, the Association of Upper Gastrointestinal Surgeons and the Primary Society for Gastroenterology	NR.	5 steps + NGT 1. Initial survey 2. Initial response list 3. Longlist generation and verification 4. Interim prioritization survey 5. Final workshop

2017, Fitzcharles et al. [30] Canada	Priorities of uncertainties for the management of fibromyalgia (FM) that could propel future research	1.Patients, carers and clinicians 2.JLA Guidebook 2013, version 5 3. Age 18 to >70 years 4. Fibromyalgia 5. In total, 4,557 treatment uncertainties submitted by 550 participants	The steering committee was composed of five patients (one patient was a practicing pharmacist), five health care professionals (one family physician, two rheumatologists, one psychologist, one internist), an internist with previous experience of the JLA process but without specific interest in FM, and a rheumatologist	Facilitators with experience of the JLA process	5 steps 1. Survey results 2. In scope uncertainties 3. Coding uncertainties 4. Interim prioritization 5. Final workshop
2017, Hart et al. [31] UK	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	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Inflammatory bowel disease. 5. In total, 1,636 treatment uncertainties submitted by 531 participants	A steering committee was established following an initial explanatory meeting and included two patients, two gastro-enterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organization Crohn's and Colitis UK, a representative of the JLA and an administrator	A JLA facilitator	5 steps 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 Top 10 priorities
2017, Hemmelgarn et al. [32] Canada	Identify the most important unanswered questions (or uncertainties) about the management of chronic kidney disease (CKD) i.e. in terms of diagnosis, prognosis and treatment.	1. Patients, carers, clinicians and policy-makers 2. JLA Guidebook 2013, version 5 3. Age 65 ≥ years 4. Non-dialysis CKD 5. In total 2,241 treatment uncertainties submitted by 439 participants	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 organization for patients with kidney disease)	Jointly organized PSP broadly adhering to the JLA Guidebook	4 steps + NGT 1. Identification and invitation of potential partners 2. Collection of research uncertainties through a national survey 3. Refinement and prioritization 4. Priority setting-workshop
2017, Khan et al. [33] Canada	Identify the 10 most important research priorities of patients,	1. Patients, carers and clinicians	Steering committee of 15 volunteer patients, carers, and clinicians from across Canada.	JLA facilitator from the UK	5 steps 1. Establishing a steering group

	carers and clinicians for hypertension management	2. JLA Guidebook 2013, version 5 3. NR 4. Hypertension 5. In total 673 individual research questions submitted by 386 participants	Stakeholder not reported in detail		2. Forming priority setting partnerships 3. Collecting potential research questions 4. Processing, categorizing, and summarizing those research questions 5. Selecting the Top 10 research priorities
2017, Jones et al. [34] Canada	Identify unanswered questions encountered during management of kidney cancer agreement by consensus on a prioritized list of the Top 10 shared unanswered questions and establish corresponding research priorities	1. Patients, carers, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patients with kidney cancer 5. In total 2,004 treatment questions submitted by 225 participants	A 15 persons steering group was formed with seven patients/carers and seven 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	5 steps 1. Formation of steering group 2. Identifying treatment questions 3. Collating questions 4. Interim ranking of questions 5. Final priority-setting workshop
2017, Lomer et al. [35] UK	Provide a comprehensive summary of the research priority findings relating to diet in the treatment of inflammatory bowel disease	1. Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Dietary treatment of inflammatory bowel disease. 5. In total 1,671 treatment uncertainties submitted by 531 participants	Steering committee comprising of two patients. two gastro-enterologists, two inflammatory bowel disease specialist nurses, two colorectal surgeons, two dietitians, a representative from the UK inflammatory bowel disease charity organization, Crohn's and Colitis UK, a representative of the JLA and an administrator (i.e., 13 persons steering committee). Stakeholders from various roles, ages and ethnic groups	A representative of the JLA and an administrator on the steering committee.	5 steps 1. Steering committee 2. Questionnaire survey 3. Remaining uncertainties were reviewed 4. Uncertainties determined 5. Final workshop of the steering group
2017, Macbeth et al. [36] UK	Identify uncertainties in alopecia areata management and treatment that are	1. Patients, partners/parents/ carers and clinicians	Four people representing various patient support groups, four dermatologists and two further individuals to represent the BHNS	A JLA representative provided independent oversight of the	5 steps + NGT 1. Identification and invitation of potential partners

	important to both service users, people with hair loss, carers/ relatives and clinicians	2. JLA Guidebook 2016, version 6 3. NR 4. Alopecia areata 5. In total 2,747 treatment uncertainties submitted by 912 participants	and the European Hair Research Society; an academic psychologist; a registered trichologist and a GP and a JLA representative. Two separate steering groups	PSP and chaired the steering group	2. Invitation to submit uncertainties 3. Collation 4. Ranking of treatment uncertainties 5. Final workshop
2017, Narahari et al. [37] India	Summarizes the process of Lymphedema PSP, discussion during the final prioritization workshop, and recommendation on the Top 7 priorities for future research in lymphedema and a brief road map	1. Patients, theorist and nurses 2. JLA Guidebook 2013, version 5 3. NR 4. Lymphedema 5. In total, 137 respondents uploaded research- priorities	The Faculty of Applied Dermatology and the Central University of Kerala participated in the coordinating committee	NR	8 steps 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 lymphedema medical camp 8. Final prioritization workshop
2017 Prior et al. UK	Identify and prioritize important research questions for miscarriage	1. Patients, partners, family members, friends or colleagues and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Miscarriage 5. In total, 3,279 questions submitted by 2,122 participants	The steering group was a balanced composition of women charities that represented them and clinicians. Some members representing charities or clinicians also had personal experience of pregnancy loss	The workshop was chaired by an independent JLA facilitator	6 steps 1. Initiation 2. Consultation 3. Identifying uncertainties 4. Refining uncertainties 5. Interim prioritization 6. Final workshop
2017 Rees et al. [38] Canada	Engaging patients and clinicians in establishing research priorities for gestational diabetes mellitus	1. Patients, friends and relatives and clinicians 2. JLA Guidebook 2013, version 5 3. Age18-69 years 4. Gestational diabetes mellitus	A steering committee consisting of three patients and three clinicians (one family physician who practises intrapartum care, an endocrinologist and a neonatologist); a facilitator familiar with the JLA process and a	A facilitator familiar with the JLA process.	4 steps + NGT 1. Survey 2. Process and collate 3. Interim ranking 4. Priority setting-workshop

		5. In total, 389 treatment uncertainties submitted by 75 participants	project manager. The Diabetes Obesity and Nutrition Strategic Clinical Network with the Alberta Health Services supported this research. Stakeholders not reported.		
2017 Smith et al. [39] UK	Prioritize research questions in emergen medicine in a consensus process to determine the Top 10 questions	1. Patients, carers and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Emergency medicine 5. In, total 214 number of initial uncertainties	The steering group members are not reported with titles but consist of 16 members. The Royal College of Emergency Medicine	NR.	6 steps 1. Online submissions 2. Working group reviews 3. Mini systematic reviews 4. Working group prioritisation exercise 5. Public prioritization exercise 6. Face- to- face final prioritization
2018 Fernandez et al. [40] UK	Establish the research priorities for adults with fragility fractures of the lower limb and pelvis that represent the shared interests and priorities	1.Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. Age ≥ 60 years 4. Fragility fractures of the lower limb and pelvis 5. In total, 963 treatment uncertainties submitted by 365 participants	The steering group consisted of patient representatives, healthcare professionals and carers with established links to relevant partner organizations to ensure that a range of stakeholder groups were represented.	A JLA adviser supported and guided the PSP	5 steps 1. First survey 2. Screening 3. Thematic analysis. original uncertainties turned into over-arching indicative questions 4. Evidence search interim prioritazion 5. Final workshop
2018 Finer et al. [41] UK	Describe processes and outcomes of a PSP to identify the Top 10 research priorities' in Type 2 diabetes	1.Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4.Type 2 diabetes 5. In total, 8,227 treatment uncertainties were submitted by 2,587 participants	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 dialectologists), an information specialist, seven members of the Diabetes UK research and senior leadership team, and a JLA senior advisor. The steering group (47% men and	The workshop was facilitated by trained JLA advisors	4 steps + NGT 1. Gathering uncertainties 2. Organizing the uncertainties 3. Interim priority setting 4. Final priority setting

			53% women and 26% from black and minority ethnic groups) met 12 times during the PSP process, in person or by teleconference Diabetes UK		
2018 Lechelt et al. [42] Canada	Identify the Top 10 treatment uncertainties in head and neck cancer from the joint perspective of patients, caregivers, family members, and treating clinicians	1. Patients, carers, family members, and clinicians 2. JLA Guidebook 2013, version 5 3. NR 4. Patient with head and neck cancer 5. In total, 818 treatment uncertainties submitted by 161 participants	The steering committee included five patients with head and neck cancer who were from 3 - 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 oncologic 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 co-facilitators, all of whom were briefed by the JLA senior advisor on recommended JLA protocols	5 steps + NGT 1. Initial survey development and deployment 2. Identifying uncertainties through survey data processing 3. Verifying uncertainties 4. Interim prioritization 5. Final workshop
2018 Lough et al. [43] UK	Identify the shared priorities for future research of women affected by and clinicians involved with pessary use for the management of prolapse	1. Patients, carers and clinicians 2. JLA Guidebook 2016, version 6 3. Age 30-89 years 4. Pessary use in women with prolapse 5.In total, 669 questions submitted by 210 participants	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 (UKCS) research grant, two grants from the Pelvic Obstetric and Gynaecological Physiotherapy	The steering group agreed the terms of reference and protocol for the JLA adviser and project leader	4 steps + NGT 1. Gathering questions/ uncertainties 2. Refining the questions and checking the evidence 3. Prioritizing /ranking the questions 4. Choosing the Top 10 priorities by consensus

			group (POGP) of the Chartered Society of Physiotherapy and a funded studentship from Glasgow Caledonian University		
2018 Macbeth et al. [44] UK	Identify uncertainties in hair loss management, prevention, diagnosis and treatment that are important to both people with hair loss and clinicians	1. Patients, carers relatives and clinicians 2. JLA Guidebook 2016, version 6 3. NR 4. Hair loss (excluding alopecia areata) 5. In total, 2,747 treatment uncertainties were submitted by 912 participants	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	5 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 * User group means the participants who are involved in the PSP process, not only the survey.

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

3 *** Steering group, steering committee and co-ordinating committee are defined as equal concepts.

RESULTS

In total, 37 studies met the inclusion criteria; their characteristics are summarized 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, 12-15, 17, 18, 20, 21, 23-29, 31, 35, 36, 39-41, 43-45] eight from Canada [19, 22, 30, 32-34, 38, 42] and one each from India [37] and Spain.[16]

The JLA process participants were patients, carers and clinicians, aged ≥ 18 years. The studies collectively represented patient groups with heterogeneous ages and health conditions/disease, 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, 17, 19, 21, 24, 26, 28, 29, 31, 33, 38, 41, 43, 45] One of the studies also gave information about socio-economic status. [29] Another study gave only information about socio-economic status. [37]

Three of studies described that patient and carers submitted more questions on psychosocial issues, psychosocial stress, depression and anxiety compered to clinicians. [13, 26, 33] No studies described disagreement in the prioritization stages. However, 24 other studies also mentioned psychosocial issues without noting who had done so. [1, 7, 14, 15, 17-22, 28-32, 34, 36, 38, 40-45] Ten studies did not mention psychosocial issues. [5, 12, 16, 23-25, 27, 29, 35, 37] The types of health conditions that were addressed included gastrointestinal,[29, 31, 35] neurologic,[1, 5, 7, 17, 24, 30] dermatologic,[13, 15, 16, 18, 36, 44] endocrine [14, 38, 41] and cancer [25, 28, 34, 42] conditions.

Setting up a Priority Setting Partnership PSP

The JLA steering group is made up of key organizations 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, 14-19, 21-23, 25, 26, 28, 32-34, 40] included patients, carers and clinicians in their steering groups; 16 studies [7, 13, 20, 27, 29-31, 35-38, 41-45] did not include carers in their steering group (i.e., only patients and clinicians). In one study,[39] the titles of the members on the steering group were not reported; in another, [24] 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, 22, 32, 38, 41, 43] to eight steps.[23, 25, 37] Five steps, corresponding to JLA Guidebook versions 4, 5 and 6, were most common: [12, 13, 15-21, 26, 27, 29-31, 33-36, 40, 42, 44] with Step 1, initiation; Step 2, collecting of uncertainties; Step 3,collation of uncertainties; Step 4,interim priority setting; Step 5final 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 organizations, 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, 38]

The questions were usually deliberately open-ended to encourage full responses regarding the experiences of patients, carers and clinicians. One of the 37 studies [37] 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, 16, 20, 26, 28, 33, 34, 38, 41, 42, 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.[17, 18, 23, 24, 31, 35, 43] In eight studies, no limit was placed on the types of questions that could be submitted.[5, 13, 19, 22, 27, 32, 39, 40] One study asked about eight open-ended questions requesting a narrative answer. [30] Close-ended questions were used in three studies,[25, 36, 44] 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, 21, 29]

The number of submitted uncertainties ranged from 8,227 submitted by 2,587 participants [41] to 323 submitted by 58 participants.[16] All studies except two [7, 37] reported involving patients, carers and clinicians in the initial survey. Two of the studies addressed verifying uncertainties example by content expert or Librarian. [33, 42] The steering group or researchers were involved in addressing verifying uncertainties in 22 of the studies, and [5, 7, 14-17, 20, 22-24, 26-29, 31, 32, 34, 37, 39, 40, 43, 45] in 13 of the studies not describing verifying the uncertainties. [1, 12, 13, 18, 19, 21, 25, 30, 35, 36, 38, 41, 44]

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.,[42] uncertainties are organized 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.,[35] specified that similar uncertainties are combined to create indicative uncertainties. Among our included studies, 20 described refining questions into indicative uncertainties,[5, 13-15, 18, 21, 23, 26, 27, 30-32, 35, 36, 38, 40-44], while 17 did not describe a concept of indicative uncertainties.[1, 7, 12, 16, 17, 19, 20, 22, 24, 25, 28, 29, 33, 34, 37, 39, 45]

In total, 16 of the studies described directly ranking and assessing survey-generated uncertainties from a longlist ranging from 43 to 226 uncertainties.[1, 5, 13, 14, 21, 23, 24, 26, 27, 29-31, 34, 37, 39, 42]

The wording of the longlist 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 prioritization is the stage at which the longlist 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, 41] interim prioritization;[1, 5, 38] and ranking exercise.[13, 37]

Their short lists varied from 22 [29] to 30 uncertainties.[12, 18-21, 25, 28, 32, 39] Sixteen of the studies used an interim prioritization of their Top 25 uncertainties that were taken to a final prioritization workshop, where the participants agreed on their Top 10 priorities.[1, 7, 13, 22-24, 26, 27, 30, 33, 35, 36, 40, 43-45] Three of the studies did not describe the number of shortlisted treatment uncertainties.[15, 31, 37]

To reduce the number of uncertainties, an interim prioritization exercise was conducted by email or post.[5, 20, 41] Patients, carers and health professionals were initially invited to

examine the longlist;[20] 14 of the studies used a second online survey [1, 21, 23, 26, 28, 30, 33, 35, 36, 40, 41, 43-45] and in one study the steering group members facilitated an interim ranking exercise.[32]

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 considered [6] 21 of the studies reported use of the NGT in the final priority- setting workshop.[5, 12, 16-22, 24, 26, 28, 29, 32, 34, 36, 38, 41-44]

All of the studies implemented a final priority- setting workshop to agree upon their Top 10 priorities. In most of the studies, these final workshops included patients, carers and clinicians; nine studies mentioned only including patients and clinicians.[7, 26, 35-38, 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 indicate no or small implications for our findings. Thus, this scoping review thus 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, 26, 33] There were no studies described disagreement in the prioritization 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 co-operative 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 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 Sachiyo,[48] who argues that there is a clear need for transparent, replicable, systematic and structured approaches to research priority- setting to assist policymakers and research funding agencies in making investments. Increased public involvement can lead to a wider range of identified and prioritized 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 organizations.[1] The number of resulting uncertainties reflects this breadth. The studies examined tended to conclude that the JLA principles were welcomed, but consistently emphasized the need for an even broader understanding, better conceptualization 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 socio-economic groups and most vulnerable patients. Many respondents, particularly those associated with charity organizations, are likely to be white, middle class and have high education attainment levels. Yet it is the, individuals who are more difficult to reach, such as those in low socio-economic groups and who are vulnerable patients - may have the greatest unmet needs and stand to gain the most from improved treatment.[28, 29, 38, 45] 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 made ,[28] to include participants from black and minority ethnic groups and care home populations, they were not particularly successful. Lough et al.,[43] 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.[17] Three of the studies,[5, 20, 38] 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.,[25] note another major challenge to involving users in research, involving patients in the steering group who have incapacitating symptoms and short expected survival durations. Another important issue is that all but two studies [16, 37] 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 language understandable to everyone

with an interest in the topic, not just to clinicians. Lough et al.,[43] 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.[27] Without doubt, the essential advantage is integration of this involvement in both research and health care. Identifying research priorities is perhaps where the PSP's greatest effect can be achieved.[29] Nevertheless, one might ask whether PSPs emphasize 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.[41, 44] There has been a clear paradigm shift from a reactive to a more proactive approach described as 'predictive, personalized, preventative and participatory'.[28] 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 operationalize 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 (i.e., 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 amongst 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 on 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 only described in one publication. [27] 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 8,227 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 addressed methods for verifying uncertainties and 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.

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Author Contributions

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.

Competing interests

None.

Data sharing statement

All data relevant to the study are included in the article or uploaded as supplementary information.

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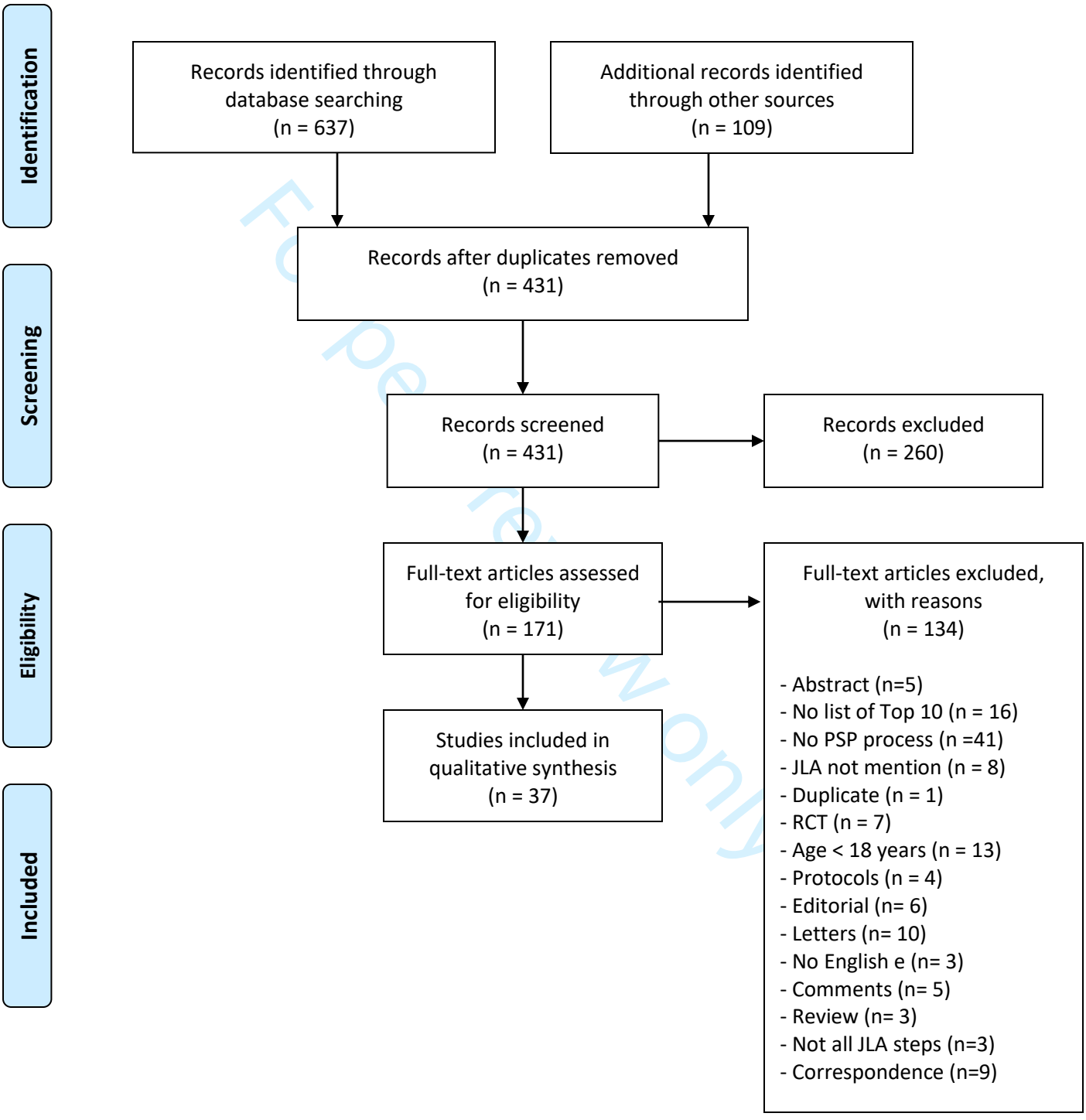
Figure 1. Flow diagram.

PRISMA 2009 Flow Diagram



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PRISMA 2009 Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	4-5
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	5
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	N/A
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	6-7
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	6
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	6
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	7
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	6
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	7-8, 32
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	7, 30
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	See note 1

PRISMA 2009 Checklist

Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	See note 2

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	See note 3
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	See note 4
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	6
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	9-21
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	7, 32
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	22-26
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	3
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	N/A 15
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	N/A 16
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	27-30
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	30-31
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	31

PRISMA 2009 Checklist

FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	31

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