



The PRioRiTy (Prioritising Recruitment in Randomised Trials) Study

Setting priorities for trial recruitment research



Final Report

Acknowledgements

The authors would like to sincerely thank all contributing individuals who completed and / or disseminated the surveys and also the people that participated in the final prioritisation workshop in Birmingham. The authors also wish to thank Katherine Cowan, Senior Advisor to the JLA for facilitating the final consensus meeting in Birmingham.

Funding

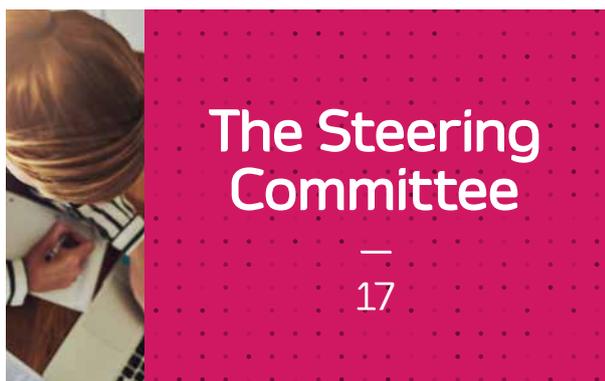
This research was funded under the Health Research Board Knowledge Exchange and Dissemination Scheme Awards 2015, through the Health Research Board -Trials Methodology Research Network (HRB-TMRN) and with the support of the James Lind Alliance throughout this PSP.



The following organisations had representatives on the **PRioRiTy Steering Group**



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Foreword by Professor Declan Devane

We live in a world where there are ever increasing demands for research to be relevant and for care to be firmly evidence based. Randomised trials have long been considered the gold standard for testing the effectiveness of interventions, and they are often used to guide and inform healthcare practices. However, they are often wrought with challenges. One challenge, in particular, is that of slow or suboptimal recruitment.

In the same way that we expect that the resolution of uncertainties about health care requires reliable and robust research, this same approach needs to be taken to uncertainties about how we plan, design, analyse, report and disseminate randomised trials.

The PRioRiT y study was a pioneering adventure between research teams in Ireland and the United Kingdom, that culminated in identifying and prioritising, for the first time, the key methodological questions that need to be addressed in order to improve recruitment to trials.

PRioRiT y was a truly wonderful journey, and we are delighted with how many people and organisations across Ireland and the UK got behind it. I would like to personally thank those that completed the surveys and took part in our workshop in Birmingham and made this list of priorities a reality.

By working with the James Lind Alliance, we gained a greater insight into how to work closely and meaningfully with members of the public in research, but also are more aware as to the incredible benefits they can bring to a project such as this. We sincerely thank the JLA for their guidance and support throughout this journey.

I would like to thank the Health Research Board in Ireland for funding this work and for recognising that 'research on research' can change the way we do randomised trials for the better, reduce waste and increase the value of research.

Finally, I would like to thank the PRioRiT y Study Steering Group, for their help, enthusiasm and for the time, energy and expertise they contributed to this study.

A handwritten signature in black ink, appearing to read 'Declan Devane', written over a white background.

Professor Declan Devane

SCIENTIFIC DIRECTOR OF THE HEALTH RESEARCH BOARD - TRIALS METHODOLOGY RESEARCH NETWORK (HRB-TMRN) AND PRINCIPAL INVESTIGATOR ON THE PRIORITY STUDY.



Foreword by Dr. Derick Mitchell

The importance of evidence from randomised trials is widely recognized in healthcare worldwide. However, recruiting people to these trials is often challenging, and many questions arise when recruitment targets are missed.

It is only through research that we will be able to address the questions about trial recruitment that remain unanswered. Research can both identify the key barriers and inform the development of interventions to reduce difficulties and support those undertaking recruitment.

I was delighted to be one of so many individuals and organisations in Ireland and the UK who collaborated to produce this top 10 list of research priorities for trial recruitment. By consulting widely, the PRioRiTy initiative has enabled patients, carers, relatives, triallists, researchers and healthcare professionals to identify what is most important to those with experience of trial recruitment and how to improve it.

From a personal perspective, including those who have not typically contributed to setting the research agenda was a key element of this initiative. Unlike in the UK, Ireland needs to recognise this systematic, democratic and transparent process with respect to considering topics as part of its wider research prioritisation process.

The top 10 list, as well as the other topics identified, provide researchers and funding agencies worldwide with a rich resource when they are considering future research projects to ensure better targeting of resources and funding.

A handwritten signature in black ink that reads "Derick Mitchell". The signature is written in a cursive, slightly slanted style.

Dr. Derick Mitchell

CHIEF EXECUTIVE OF IRISH PLATFORM FOR PATIENT ORGANISATIONS, SCIENCE & INDUSTRY (IPPOSI) AND PRIORITY STUDY STEERING GROUP MEMBER.



Introduction

Why set Priorities for recruitment research?

Challenges in how randomised trials are designed and conducted are commonly experienced at different stages of the trial process. There remain uncertainties on many aspects of the trial process, from planning and design to conduct, analysis, reporting and dissemination. Methods to boost trial recruitment has been identified as a topic of importance that needs to be addressed.

Participant recruitment is critical to the success of every randomised trial yet optimising recruitment remains a difficult, ongoing challenge for the trial community. Evidence suggests that less than 50% of trials meet their recruitment target or meet their target without an extension [1, 2]. Difficulties in recruitment often result in delays and additional costs in conducting trials and additional costs associated with the need for extensions. Having to commit additional resources for recruitment efforts may also impact negatively on the quality of follow-up for those already recruited, further compromising the trial outcome. Difficulties arise in using the trial results to make informed decisions about clinical care if they have failed to reach the recruitment target necessary for an adequately powered study. From a funder perspective, inadequate recruitment wastes valuable funding and the question the trial was funded to answer remains unanswered leaving treatment decisions uncertain.

For these reasons, it is important that research into how trial recruitment might be improved is conducted so that scarce resources might be directed to areas considered important by key stakeholders and the strength of trials' findings can be increased.

Background

Who carried out the research and why?

The PRioRiTy study was a collaborative study led by the Health Research Board - Trials Methodology Research Network (HRB-TMRN; <https://www.hrb-tmrn.ie>) in Ireland with the support of the James Lind Alliance (JLA) in the UK. The HRB-TMRN is a collaborative network, which seeks to improve the planning, design, conduct, analysis, reporting and dissemination of randomised trials nationally. The HRB-TMRN was established in 2014 and endeavours to improve the understanding of trial methodology through a suite of activities including training and education, online support and primary methodology research. The JLA is a non-profit making initiative which was established in 2004. The JLA is funded by the National Institute for Health Research (NIHR) and is centrally coordinated by NIHR, Evaluation, Trials and Studies Coordinating Centre (NETSCC). It brings patients, carers and clinicians together in priority setting partnership (PSPs) to identify and prioritise the unanswered questions about the effects of treatments that they agree are most important [3]. It is evident that what researchers want to study, can be very different from what patients and clinicians want to study, therefore a collaboration with the JLA was identified as the best means of bridging this gap. While the JLA PSP framework is a tried and tested methodology for identifying and prioritising treatment uncertainties, this is the first PSP concerned with research methodology uncertainties. Therefore, the methods used during the study were developed to accommodate a focus on methodological rather than treatment uncertainties while utilising and respecting the JLA PSP framework.

94%

94% of the consensus meeting participants strongly agreed or agreed that the written information sent in advance of the workshop was helpful.



Objectives

What were we trying to achieve?

The aim of this PSP was to identify unanswered questions around trial recruitment, and then prioritise those that trial participants, trial recruiting clinicians and trial researchers agreed are the most important. Unanswered questions around trial recruitment were defined as the recruitment challenges encountered by recruiters, trial designers and persons being recruited to randomised trials of health care interventions.

This PSP brought together people, across the UK and Ireland who are, or have been, involved directly in designing, conducting and taking part in randomised controlled trials. A randomised trial is a type of research study that compares groups of people receiving different interventions and looks at which of these improves health outcomes the most. An intervention is anything that aims to make a change to someone's health. For example, providing a counselling service, giving a drug, or giving people information and training are all described as interventions. The decision about which group a person joins in a randomised trial is at random, which means that a person is put into one of the intervention groups by chance.

The bringing together of people in this PSP engaged researchers, clinicians, trial experts, JLA staff, the public and/or their representatives in an exercise of discussion, knowledge exchange and consensus, in identifying, agreeing, prioritising and disseminating a list of the most important unanswered questions surrounding recruitment to trials. Recruitment refers to the process of how people join trials.

“

Then we had the amazing task of agreeing where each of the 20 was placed... very good discussion and reasoning.

PATIENT / MEMBER OF THE PUBLIC.

“

Very interesting experience; Different perspectives well represented; Process effective.

NON-RESEARCHER INVOLVED IN RECRUITING PARTICIPANTS



Methodology

What was the process?

The process involved a number of stages as follows: (i) establishing a Steering Group (ii) identifying and engaging with partners (iii) identifying and engaging with stakeholders (iv) Initial survey development and deployment (v) collating the uncertainties into research questions (vi) development of the Interim Survey (vii) Voting and ranking survey items for shortlisting (viii) final prioritisation through a face to face workshop.



i. Establishing the Steering Group

A Steering Group to oversee the PSP was established in accordance with JLA guidance and held its first meeting in May 2016. The Steering Group included individuals and representatives of organisations who could reach and advocate for key stakeholder groups i.e., members of the public invited to join a trial, researchers, healthcare providers who are recruiters and trialists. The primary role of the Steering Group was to discuss and agree the strategic orientation and processes of the PRioRiT y PSP. The protocol was developed in collaboration with the JLA and with reference to the JLA guidebook modified as appropriate for the methodology, rather than treatment, focus of the PSP.

ii. Identifying and inviting potential partners

The next stage in the process was to invite potential partner organisations to engage with the PRioRiT y PSP. The unique subject of the PRioRiT y PSP meant that some changes to the usual exclusion criteria of stakeholder groups were made. The JLA PSP process does not usually invite representatives of the research community (e.g. front line research staff and methodologists) who are not also clinicians, patients or carers to participate in the priority setting process; this stakeholder group was included in this PSP given their various roles in randomised trials.

Potential partner organisations were identified through a process of peer knowledge and consultation, through the Steering Group members' respective networks and through the JLA's existing contacts. The PRioRiT y PSP ensured that a wide range of partners representing the broad stakeholder groups across Ireland and the UK were secured so that the uncertainties gathered would be from as wide a range of potential contributors as possible. Potential partners were contacted and informed of the establishment and aims of the PRioRiT y PSP and invited to participate in the PSP. Partners were expected to help promote the PSP to their members and to encourage participation in the survey used to gather uncertainties. In line with JLA guidance, we did not include representatives of the pharmaceutical industry.



94% strongly agreed or agreed that they felt able to contribute to the workshop and put their views across.



94% strongly agreed or agreed that the process was fair and independent.



94% strongly agreed or agreed that everyone had the opportunity to contribute to the discussion.

iii. Identifying and Engaging the Stakeholders

The stakeholders for this PSP (listed below) were the people to whom the online survey for the initial gathering of information around possible uncertainties was distributed.

- Members of the public who had been invited to participate in a randomised trial or participated in Trial Steering Committees (TSCs). They could be an individual or representing a patient organisation;
- Front line clinical and research staff who were or had been involved in recruitment to randomised trials (e.g., postdoctoral researchers, clinicians, nurses, midwives, allied health professionals);
- People who had established expertise in designing, conducting, analysing and reporting randomised trials (e.g. Principal Investigators / Chief Investigators);
- People who are familiar with the trial methodology research landscape (e.g. funders, programme managers, network coordinators).

iv. Initial survey development and deployment

Initial feedback from stakeholders on the challenges in trial recruitment that they perceived as important and wished to see addressed was sought in an online survey, which was available between July and August 2016 (4 weeks). This survey contained five questions (Table 1) asking people to consider their own experiences of being involved in randomised trials across specific areas (planning and design, conduct, information, trial recruiters and motivation). The specific areas were derived from the ORRCA project (Online Resource for Recruitment research in Clinical triAls) recruitment research domains (4). The ORRCA project has created a web-enabled database (<http://www.orrca.org.uk/>) populated with published and ongoing recruitment research. Respondents were invited to submit comments in an open-ended format to each question. They were also offered a sixth open-ended comment box for any other items not considered in the five thematic questions. The survey also asked demographic questions as well as asking respondents to identify which stakeholder group they belonged to.



Table 1: Survey questions

1	What questions or comments do you have (if any) about improving how trials are planned and designed?
2	What questions or comments do you have (if any) about improving how trials are carried out?
3	What questions or comments do you have (if any) about the information people are given when they join a trial?
4	What questions or comments do you have (if any) about trial recruiters who invite people to take part?
5	What questions or comments do you have (if any) about the motivation of people in joining a trial?
6	Do you have any other questions or comments?

The surveys to collect recruitment uncertainties were constructed in SurveyMonkey® and embedded into the PRioRiTy sub-page of the HRB-TMRN website. The survey link was distributed by email and the survey was also available in paper format if participants preferred this format. Steering Group members and partners were asked to promote the PSP and surveys to stakeholders via email, web sites, relevant meetings, social media and any other opportunities that arose. A social media promotion plan was developed, with all Steering Group members requested to use pre-worded tweets, which included the link to the survey. No incentives were offered for return of the survey.

While no formal target sample size was set for both surveys, the number of surveys being returned by each stakeholder group was monitored on a weekly basis. Where any stakeholder group participation in the survey was <10% of the total responders, efforts were made to ensure targeted promotion of the survey among that group across the relevant partners.



v. Collating initial survey responses and developing questions

The information obtained from the initial survey was assembled and categorised using Microsoft Excel and merged into a single database. The constant comparative method of analysis was used to identify emergent themes from that data. Responses to each survey question were re-written as research questions. Longer responses were broken down into several key excerpts as appropriate and multiple questions were created. Responses judged to be not relevant to trial recruitment were classed as “out of scope” and saved for future analysis. Questions with common themes and issues were merged into broader questions where appropriate and duplicates removed. This process was reviewed regularly by team members for consistency and reliability of emerging themes and issues. Any discrepancies or issues arising from specific responses were discussed by the research team. The recruitment research domains used for the searchable database ORRCA (Online Resource for Recruitment research in Clinical triAls (<http://www.orrca.org.uk/>)), were used as a framework to aid this process [4]. This allowed members of the team to use the same classification of research questions across recruitment themes.

vi. Development of the Interim Survey

A second follow-up interim survey was conducted to prioritise and shortlist the newly developed questions. The interim survey was open during November 2016 (3 weeks) and asked the stakeholders to select up to 10 important questions. Invitations to this survey were not restricted to those participating in the initial survey. The resulting shortlist of questions for research was cross referenced with the identified systematic reviews. Questions were confirmed as unanswered if there was no recent systematic review of research evidence addressing the recruitment question or if a systematic review of research evidence confirmed that the recruitment question remains unanswered.



88% strongly agreed or agreed that the workshop achieved its aim of helping members of the public, health professionals, researchers and those involved in designing and reporting on research to work together to set priorities for future research.

vii. Voting and ranking survey items for shortlisting

The Steering Group used ranked weighted scores to decide which of the interim survey research questions to take forward to the final priority setting workshop. We followed the standard JLA approach as described in detail in the JLA Guidebook (www.jla.nihr.ac.uk/jla-guidebook/). Response counts were allocated for each research question across each stakeholder group. Summed scores from each stakeholder group were calculated separately. For each of the stakeholder groups, the highest ranked question was allocated a maximum score (for example if 30 questions in the list, the #1 ranked question would get a score of 30), the next one down a lower score (i.e. -1) and so on down the list, until the lowest ranked question received the lowest score (i.e. 1). To get the overall ranking, the scores for each question from each of the stakeholder groups were added together. The question with the highest aggregated score was ranked number one overall and the lowest score was ranked lowest overall. This gave the overall interim ranking to the research questions as well as rankings for each of the stakeholder groups, whilst minimising bias owing to numbers of responses from each stakeholder group.

viii. Priority setting workshop

A final prioritisation workshop was held in December 2016 in Birmingham, UK, in order to condense the number of questions generated by the stakeholder surveys to (a minimum of) a “Top 10” list of research questions agreed by consensus. The workshop followed the standard JLA approach as described in detail in the JLA Guidebook (www.jla.nihr.ac.uk/jla-guidebook/) and was facilitated by the JLA’s Senior Advisor. Thirty-one participants representing the stakeholder groups were invited to the face-to-face final priority setting workshop and on the day 26 attended. This was made up of 10 public members from trials or trial steering committees, 7 frontline researchers or non-researchers involved in recruitment, 6 trial methodologists, and 3 researchers or Principal Investigators. Some members of the PRioRiTty Steering Group attended as observers. The 26 participants were divided into 3 groups with a JLA facilitator for each group. Each group was provided with the shortlisted questions in individual question cards with stakeholder group rankings from the voting process and an example quote from the original survey submissions noted on the back of them. The questions had been sent to the participants prior to the meeting so that they could have some time to familiarise themselves with the list and decide on what was important to them. The facilitators then guided the participants through the process of discussing the questions and agreeing, by consensus, a “Top 10” from within them.



Results

What did we find?

The initial survey was completed by 790 respondents. Of those; 9% identified themselves as a person invited to take part in a trial, 10% as a non-researcher (e.g. clinician or health professional) involved in recruiting participants, 12% as a trial methodologist (someone who specialises in the methods of how trials are designed, run, analysed and reported), 16% as a principal / chief investigator, 19% as a researcher involved in aspects of the trial other than frontline recruitment, 21% as a researcher involved in recruiting participants and 13% as other. Respondents to the survey were predominantly from England (74%), followed by the Republic of Ireland (17%), Scotland (4%), Wales (2%) and Northern Ireland (<1%). A total of 1,880 questions for research were formulated from the 790 survey responses. Merging similar themed questions and removing duplicates reduced this to 496 questions. Where appropriate, questions were merged into broader questions following review and discussion with the steering group. Finally, questions asked by more than 15 people and/or at least 6 of the 7 stakeholder groups were selected to progress to the interim survey of stakeholders. These criteria were developed through consultation with the Steering Group after presenting data across stakeholder groups. This resulted in the inclusion of 31 unique questions for ranking in the interim survey.

The interim survey was completed by 815 respondents. Of those, 8% as a non-researcher (e.g. clinician or health professional) involved in recruiting participants, 11% as a trial methodologist (someone who specializes in the methods of how trials are designed, run, analysed and reported), 13% identified as a person invited to take part in a trial, 18% as a researcher involved in recruiting participants, 23% as a principal / chief investigator, 25% as a researcher involved in aspects of the trial other than frontline recruitment, and 2% as other. Respondents to the interim survey were predominantly from England (77%), followed by the Republic of Ireland (9%), Scotland (7%), Wales (5%) and Northern Ireland (2%).

“

Very positive and energising experience
thank you

RESEARCHER

Following the ranking conducted in the interim survey, twenty-five questions were brought forward for discussion and final prioritising at the face-to-face consensus meeting.

The “Top 10” research questions prioritised are:

OVERALL RANKING	UNCERTAINTY AS RESEARCH QUESTION
1	How can randomised trials become part of routine care and best utilise current clinical care pathways?
2	What information should trialists communicate to members of the public who are being invited to take part in a randomised trial in order to improve recruitment to the trial?
3	Does patient/public involvement in planning a randomised trial improve recruitment?
4	What are the best approaches for designing and delivering information to members of the public who are invited to take part in a randomised trial?
5	What are the barriers and enablers for clinicians/healthcare professionals in helping conduct randomised trials?
6	What are the key motivators influencing members of the public's decisions to take part in a randomised trial?
7	What are the best approaches to ensure inclusion and participation of under-represented or vulnerable groups in randomised trials?
8	What are the best ways to predict recruitment rates to a randomised trial and what impact do such predictions have on recruitment?
9	What are the best approaches to optimise the informed consent process when recruiting participants to randomised trials?
10	What are the advantages and disadvantages to using technology during the recruitment process?

Next Steps

The top 10 questions for research can be viewed on a user-friendly, dedicated website www.priorityresearch.ie. Research teams conducting specific research relevant to any of the PRioRiTty research questions are requested to submit basic details of their work to the HRB-TMRN through the PRioRiTty website as a repository and central record platform for the level of research ongoing for each research question. This resource will be maintained by the HRB-TMRN and will be freely searchable and accessible.

The JLA PSP process for methodological uncertainties

The PSP processes developed by the JLA aim to be robust and methodologically defensible. The PRioRiTty study involved adapting the PSP process, to suit the context of this topic area, namely trial methodology. JLA representatives were involved in every step of this study to ensure the original PSP methodology was respected fully. Some limitations in this methodology did exist.

For example, while the surveys aim to attract a representative sample of respondents, this is not always achieved. While every effort is made to remove barriers to participation and to engage participants who are under-represented or seldom-heard, this does not guarantee that everyone who could take part does so. It is hoped that the involvement of healthcare professionals who can represent the interests of a diverse range of patients goes some way to addressing this. Ultimately, however, participants are inevitably self-selecting and may therefore generate a respondent bias. Ways in which the PRioRiTty study addressed this issue are detailed throughout the report.

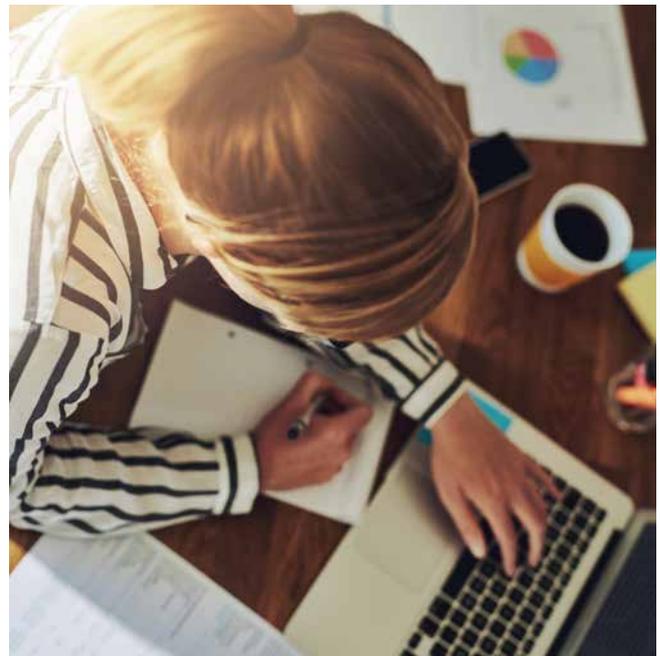
Similarly, the final workshops can only involve a limited number of individuals. Care is taken to achieve a balance of participants, so that no single perspective, personal or professional, dominates the discussion and the decision-making. The JLA requires participants to declare their interests and compiles and distributes participant biographies before each workshop.

Neutral facilitation is intended to help ensure that everyone has their say and that consensus is achieved fairly. Participants are expected to adhere to the principle of partnership working, to respect different opinions and to be pragmatic. By its very nature, consensus decision-making requires compromise.

“

The PRioRiTty study constitutes pioneer work for setting priorities in methodological research. The chosen topic of recruitment in randomized trials is highly relevant across stakeholders of clinical research not only in the UK but internationally.

PEER REVIEWER OF PRIORITY PUBLICATION - TRIALS



Additional information

The attendees at the final consensus meeting agreed and requested that a further prioritisation of questions 11-20 should be carried out. As such questions 11-20 are also available to view on the PRioRiTty website.



88% strongly agreed or agreed that the workshop venue was suitable for their needs.

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The Steering Committee

The Priority Setting Partnership was managed by a Steering Committee. The Steering Committee included the following participants:

The Steering Group

NAME	AFFILIATION	ROLE IN SG
Prof Declan Devane	HRB-TMRN (Scientific Director)	Chair
Dr Valerie Smith	HRB-TMRN/School of Nursing & Midwifery, NUI Galway (Research Fellow) (Now Trinity College Dublin)	Deputy-Chair
Dr Patricia Healy	School of Nursing & Midwifery, NUI Galway (Research Fellow)	Representing frontline research
Prof Paula Williamson	University of Liverpool/MRC Trial Methodology	Trial expertise
Prof Mike Clarke	Queen's University Belfast	Trial expertise
Prof Shaun Treweek	University of Aberdeen/Trial Forge	Trial expertise
Dr Derick Mitchell	Irish Platform for Patients Organisations, Science and Industry (IPPOSI) (Director)	Representing public and patients
Dr Sandra Galvin	HRB-TMRN (Programme Manager)	Trial expertise
Dr Mary Clarke Moloney	Clinical Operations Manager, Health Research Institute, University of Limerick and University Hospital Limerick	Representing frontline research
Dr Amanda Blatch-Jones	National Institute for Health Research (NIHR), Research on Research programme	Priority setting expertise
Ms Eleanor Woodford Guegan	National Institute for Health Research (NIHR), Research on Research programme	Priority setting expertise
Ms Beccy Maeso	James Lind Alliance	Priority setting expertise
Prof Carrol Gamble	University of Liverpool/ ORRCA database	Recruitment expertise
Mr Derek Stewart OBE	NIHR Clinical Research Network Associate Director for Patient and Public Involvement	Representing public and patients
Ms Hannah Reay	NIHR Clinical Research Network: West Midlands Workforce Development Lead	Representing frontline research
Mrs Caroline Whiting	James Lind Alliance	Priority setting expertise/Secretariat support
Dr Abdel Douiri	NIHR Research Design Service/King's College London	Representing frontline research
Mr Chris Bray	UK Trial Managers' Network/Oxford Diabetes Trials Unit	Representing frontline research
Prof Martin O'Donnell	Clinical Research Facility, Galway	Trial expertise/ PI experience
Professor Peter Brocklehurst	Birmingham Clinical Trials Unit (BCTU), University of Birmingham	Trial expertise/ PI experience
Joan Jordan	EUPATI trainee/ IPPOSI	Representing public and patients/ Trial participant
Prof Sue Pavitt	NIHR Clinical Research Network/ University of Leeds	Trial expertise

Publicity

As well as alerting funders, partners and Steering Committee members are encouraged to publish the findings of the PRioRiTy PSP using both internal and external communication mechanisms, to raising awareness of the results among the public and scientific audiences. The JLA can also capture and publicise the results, through descriptive reports of the process itself. The Partnership is asked to keep the JLA informed of activity undertaken to publicise the results of the priority setting exercise.

A paper outlining the methodology used, and also the 'Top 10' list of question is available to read, *Trials* (2018) 19:147. <https://doi.org/10.1186/s13063-018-2544-4>

The full list of questions, including example original survey quotes is available to view on <https://priorityresearch.ie/>

The report was written and prepared by Dr Patricia Healy (NUI Galway) and Dr Sandra Galvin (HRB-TMRN) on behalf of the PRioRiTy Steering Group.

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Room 235, 1st Floor, Áras Moyola,
National University of Ireland Galway
Tel: + 00 353 91 494492
Email: hrb-tmrn@nuigalway.ie
[@hrbtmrn](#) #trialrecruitment

James Lind Alliance
Email: jla@southampton.ac.uk
www.lindalliance.org

Please turn over for full report.



“

“The use of the PSP process for this project provided some very useful learning around the successful engagement of the public and patients in the conversation about trial methodology. We found that the public were well able to contribute to the methodological discussion. Having identified and prioritised the most significant challenges when recruiting people to trials it is now time to move forward to addressing those challenges

DR PATRICIA HEALY - COORDINATOR FOR THE PRIORITY STUDY, RESEARCH FELLOW, NATIONAL UNIVERSITY OF IRELAND GALWAY, IRELAND

METHODOLOGY

Open Access



Identifying trial recruitment uncertainties using a James Lind Alliance Priority Setting Partnership – the PRioRiTty (Prioritising Recruitment in Randomised Trials) study

Patricia Healy^{1,2*} , Sandra Galvin^{1,2}, Paula R. Williamson³, Shaun Treweek⁴, Caroline Whiting⁵, Beccy Maeso⁵, Christopher Bray⁶, Peter Brocklehurst⁷, Mary Clarke Moloney⁸, Abdel Douiri⁹, Carrol Gamble³, Heidi R. Gardner⁴, Derick Mitchell¹⁰, Derek Stewart¹¹, Joan Jordan¹⁰, Martin O'Donnell^{1,12}, Mike Clarke^{1,13}, Sue H. Pavitt¹⁴, Eleanor Woodford Guegan¹⁵, Amanda Blatch-Jones¹⁵, Valerie Smith^{1,16}, Hannah Reay¹⁷ and Declan Devane^{1,2}

Abstract

Background: Despite the problem of inadequate recruitment to randomised trials, there is little evidence to guide researchers on decisions about how people are effectively recruited to take part in trials. The PRioRiTty study aimed to identify and prioritise important unanswered trial recruitment questions for research. The PRioRiTty study - Priority Setting Partnership (PSP) included members of the public approached to take part in a randomised trial or who have represented participants on randomised trial steering committees, health professionals and research staff with experience of recruiting to randomised trials, people who have designed, conducted, analysed or reported on randomised trials and people with experience of randomised trials methodology.

Methods: This partnership was aided by the James Lind Alliance and involved eight stages: (i) identifying a unique, relevant prioritisation area within trial methodology; (ii) establishing a steering group (iii) identifying and engaging with partners and stakeholders; (iv) formulating an initial list of uncertainties; (v) collating the uncertainties into research questions; (vi) confirming that the questions for research are a current recruitment challenge; (vii) shortlisting questions and (viii) final prioritisation through a face-to-face workshop.

Results: A total of 790 survey respondents yielded 1693 open-text answers to 6 questions, from which 1880 potential questions for research were identified. After merging duplicates, the number of questions was reduced to 496. Questions were combined further, and those that were submitted by fewer than 15 people and/or fewer than 6 of the 7 stakeholder groups were excluded from the next round of prioritisation resulting in 31 unique questions for research. All 31 questions were confirmed as being unanswered after checking relevant, up-to-date research evidence. The 10 highest priority questions were ranked at a face-to-face workshop. The number 1 ranked question was "How can randomised trials become part of routine care and best utilise current clinical care pathways?" The top 10 research questions can be viewed at www.priorityresearch.ie.

(Continued on next page)

* Correspondence: Patricia.healy@nuigalway.ie

¹Health Research Board – Trials Methodology Research Network, Galway, Ireland

²School of Nursing and Midwifery, NUI Galway, Galway, Ireland

Full list of author information is available at the end of the article



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Conclusion: The prioritised questions call for a collective focus on normalising trials as part of clinical care, enhancing communication, addressing barriers, enablers and motivators around participation and exploring greater public involvement in the research process.

Keywords: Recruitment challenges, Participation in randomised trials, Survey, Priority setting partnership, James Lind Alliance, Trial methodology,

Background

Challenges in how randomised trials are designed and conducted are commonly experienced at various stages of the trial process. Uncertainties remain on many aspects of the trial process, from planning and design to conduct, analysis, reporting and dissemination.

Participant recruitment is critical to the success of every randomised trial yet optimising recruitment remains a difficult, ongoing challenge for the trial community. Evidence suggests that less than 50% of trials meet their recruitment target with or without an extension [1, 2]. Difficulties in recruitment often result in delays and additional costs in conducting trials and additional costs associated with the need for extensions. Having to commit additional resources for recruitment efforts may also impact negatively on the quality of follow up for those already recruited, further compromising the trial outcome. Difficulties arise in using the trial results to make informed decisions about clinical care if they have failed to reach the recruitment target necessary for an adequately powered study. From a funder perspective, inadequate recruitment wastes available funding and the question the trial was funded to answer remains unanswered, leaving treatment decisions uncertain. Furthermore, a number of systematic reviews, focusing on interventions to improve recruitment, reveal a shortage of high-quality evidence from randomised evaluations of trial recruitment interventions, and what little evidence that is available has a very narrow scope [2–5]. This is reinforced by Tudur-Smith *et al.* (2014) who conducted a Delphi study with Clinical Trials Units (CTUs) in the United Kingdom (UK) to identify topics of importance and to establish consensus for research priorities around trial methodology with “Methods to boost recruitment in trials” being identified as the highest priority [6].

For these reasons, it is important that research into how trial recruitment might be improved is conducted urgently so that scarce resources might be directed to areas considered important by key stakeholders. Research questions that are both important to stakeholders and have not been answered to date, may be identified and prioritised through a priority setting partnership (PSP) [7]. This paper reports on the processes and prioritised questions for research identified by the Prioritising Recruitment in Randomised Trials Priority Setting Partnership (PRioRiT_y PSP). PSPs have previously used the James Lind Alliance (JLA) method

to bring relevant stakeholders together to jointly identify priorities for research concerning treatment for conditions and illnesses [8, 9].

The role of the PSP is to identify questions for research (or “uncertainties”) that are both important to stakeholders and have not been answered to date, and to then prioritise these through engagement across the various stakeholder groups [7]. Prioritised questions are usually broad overarching questions, for which several more specific questions might be identified. The PRioRiT_y PSP was a collaborative project by the Health Research Board Trials Methodology Research Network (HRB-TMRN; <https://www.hrb-tmrn.ie>) in Ireland with the support of the JLA in the UK. The HRB-TMRN is an all-Ireland collaborative network, which seeks to improve the planning, design, conduct, analysis, reporting and dissemination of randomised trials nationally. The HRB-TMRN was established in 2015 and endeavours to improve the understanding of trial methodology nationally through a suite of activities including training and education, online support and primary methodology research. The JLA is a non-profit making initiative that was established in 2004. The JLA is funded by the National Institute for Health Research (NIHR) and is centrally coordinated by NIHR, Evaluation, Trials and Studies Coordinating Centre (NETSCC). It brings patients, carers and clinicians together in PSPs to identify and prioritise the unanswered questions about the effects of treatments that they agree are most important [7]. While the JLA PSP framework is a tried and tested methodology for treatment uncertainties, this is the first PSP concerned with research methodology uncertainties. Therefore, the methods used during the study were developed to accommodate a focus on methodological rather than treatment uncertainties while utilising the JLA PSP framework.

The purpose of the study was to identify unanswered questions around trial recruitment research, and then prioritise these based on agreement from across the relevant trial stakeholder groups.

Methods

The PRioRiT_y PSP was formally initiated in a meeting at the International Clinical Trials Methodology Conference in November 2015, where members (from the HRB-TMRN, Trial Forge (<http://trialforge.org>), NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC; [20](http://</p>
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www.southampton.ac.uk/netscc/index.page) and the MRC-HTMR (<https://www.methodologyhubs.mrc.ac.uk>) agreed the scope and nature of the project. Methodological uncertainties around trial recruitment were defined as the recruitment challenges encountered by recruiters, trial designers and persons being recruited to randomised trials of health care interventions. To collect a representative range of opinions, the PRioRiTty working group brought people together from across the UK and Ireland who were, or had been, involved directly in any aspect of randomised trials.

Establishing the Steering Group

A Steering Group to oversee the PSP was established in accordance with JLA guidance and held its first meeting in May 2016. Potential steering group members were identified by the HRB-TMRN and the JLA through an open and inclusive approach of peer knowledge and consultation with respective networks and existing contacts. Membership included equal representation from researchers, clinicians, trial experts, the public and/or their representatives, as well as JLA staff. The primary role of the Steering Group was to discuss and agree the strategic orientation and processes of the PRioRiTty PSP. The protocol was developed in collaboration with the JLA and with reference to the JLA guidebook modified as appropriate for the methodology, rather than treatment, focus of the PSP.

Identifying and inviting potential partners

The first stage in the process was to invite potential partner organisations to engage with the PRioRiTty PSP. As per the JLA PSP guidance, industry representation was excluded from this PSP, as traditionally the health research agenda has been largely directed by the industry agenda, with the voice of patients and carers rarely included. However, the unique subject of the PRioRiTty PSP meant that some changes to the usual exclusion criteria of stakeholder groups were made. The JLA PSP process does not usually invite representatives of the research community (e.g. front line research staff and methodologists) who are not also clinicians, patients or carers to participate in the priority setting process; this stakeholder group was included in this PSP given their various roles in randomised trials.

Potential partner organisations were identified through a process of peer knowledge and consultation, through the Steering Group members' respective networks and through the JLA's existing contacts. The PRioRiTty PSP ensured that a wide range of partners representing the broad stakeholder groups across Ireland and the UK were secured so that the uncertainties gathered would be from as wide a range of potential contributors as possible. Potential partners were contacted and informed of the establishment and aims of the PRioRiTty PSP and invited to participate in the PSP. Partners were expected to help promote the PSP to their members and to encourage participation in the survey used to gather uncertainties. In line with JLA guidance, we did not include representatives of the pharmaceutical industry.

Identifying and engaging the stakeholders

The stakeholders for this PSP (listed below) were the people to whom the online survey for the initial gathering of information around possible uncertainties was distributed:

- Members of the public who had been invited to participate in a randomised trial or participated in Trial Steering Committees (TSCs). They could be an individual or representing a patient organisation;
- Front line clinical and research staff who were or had been involved in recruitment to randomised trials (e.g. postdoctoral researchers, clinicians, nurses, midwives, allied health professionals);
- People who had established expertise in designing, conducting, analysing and reporting randomised trials (e.g. Principal Investigators/Chief Investigators);
- People who are familiar with the trial methodology research landscape (e.g. funders, programme managers, network coordinators).

Initial survey development and deployment

Initial feedback from stakeholders on the challenges in trial recruitment that they perceived as important and wished to see addressed was sought in an online survey, which was available between July and August 2016 (4 weeks). This survey contained five questions (Table 1) asking people to consider their own experiences of being

Table 1 Initial PRioRiTty survey questions

1	What questions or comments do you have (if any) about improving how trials are planned and designed?
2	What questions or comments do you have (if any) about improving how trials are carried out?
3	What questions or comments do you have (if any) about the information people are given when they join a trial?
4	What questions or comments do you have (if any) about trial recruiters who invite people to take part?
5	What questions or comments do you have (if any) about the motivation of people in joining a trial?
6	Do you have any other questions or comments?

involved in randomised trials across specific areas. The specific areas or domains (planning and design, conduct, information, trial recruiters and motivation) were derived from the Online Resource for Recruitment research in Clinical trials (ORRCA) project recruitment research domains [10]. The ORRCA project has a web-enabled database (<http://www.orrca.org.uk/>) populated with published and ongoing recruitment research. Respondents were invited to submit comments in an open-ended format to a broad question about each of the ORRCA domains. For example; what questions or comments do you have (if any) about improving how trials are planned and designed? They were also offered a sixth open-ended comment box for any other items not considered in the five thematic questions. The survey also asked demographic questions and asked respondents to identify which stakeholder group they belonged to.

Collating and analysing initial survey responses and developing questions

The constant comparative method of analysis was used to identify emergent themes in the survey responses. The constant comparative analysis method is an iterative and inductive process of reducing the data through constant recoding [11]. The information obtained from the initial survey was assembled and categorised using Microsoft Excel and merged into a single database. Responses to each survey question were re-written as research questions by two members of the project team who independently extracted the data. Longer responses were broken down into several key excerpts as appropriate and multiple questions were created. Responses judged to be not relevant to trial recruitment were classed as “out of scope” and saved for future analysis. Questions with common themes and issues were merged into broader questions where appropriate and duplicates removed. The reliability of emerging themes and issues was reviewed regularly by the two project team members swapping a portion of their respective data and comparing findings for consistency. Any discrepancies or issues arising from specific responses were adjudicated by a third team member and discussed by the research team if necessary. The recruitment research domains used for the searchable database ORRCA (<http://www.orrca.org.uk/>), were used as a framework to aid this process [10]. This allowed members of the team to use the same classification of research questions across recruitment themes. A literature review was conducted in parallel with this process to ensure that all of the included questions in the interim survey were questions for which there was insufficient evidence to adequately consider them as being

unanswered. Our search strategy was developed by reviewing and combining search strategies from the Cochrane systematic review on “Strategies to improve recruitment to randomised controlled trials” [4] and from the ORRCA project [10]. The search was run (July 2016) across MEDLINE, EMBASE, Cochrane Database of Systematic Reviews, Social Sciences Citation Index and ERIC. The findings of relevant, up to date (published in the proceeding 3 years as per standard JLA guidance) systematic reviews identified were mapped to the questions and reviewed by at least two members of the Steering Group (see [Appendix](#)).

Development of the interim survey

A second follow-up interim survey was conducted to prioritise and shortlist the newly developed questions. The interim survey was open during November 2016 (3 weeks) and asked the stakeholders to select up to 10 important questions. Invitations to this survey were not restricted to those participating in the initial survey. The resulting shortlist of questions for research was cross referenced with the identified systematic reviews. Questions were confirmed as unanswered if there was no systematic review of research evidence addressing the recruitment question or if a systematic review of research evidence confirmed that the recruitment question remains unanswered. The literature review to establish if the proposed research questions were unanswered confirmed that all 31 questions formulated from the initial survey were included in the interim survey.

Survey dissemination and promotion

While no formal target sample size was set for both surveys, the number of surveys being returned by each stakeholder group was monitored on a weekly basis. Where any stakeholder group participation in the survey was < 10% of the total responders, efforts were made to ensure targeted promotion of the survey among that group across the relevant partners.

Both the initial survey and the interim survey to collect recruitment uncertainties were constructed in SurveyMonkey® and embedded into the PRioRiTty sub-page of the HRB-TMRN website. The survey link was distributed by email and the survey was also available in paper format if participants preferred this format. Steering Group members and partners were asked to promote the PSP and surveys to stakeholders via email, web sites, relevant meetings, social media and any other opportunities that arose. A social media promotion plan was developed, with all Steering Group members requested to use pre-worded tweets, which included the link to the survey. No incentives were offered for return of the survey.

Voting and ranking survey items

The Steering Group used ranked weighted scores to decide which of the interim survey research questions to take forward to the final priority setting workshop. We followed the standard JLA approach as described in detail in the JLA Guidebook (www.jla.nih.ac.uk/jla-guidebook/). Response counts were allocated for each research question across each stakeholder group. Summed scores from each stakeholder group were calculated separately. For each of the stakeholder groups, the highest ranked question was allocated a maximum score (for example if there were 30 questions in the list, the number 1 ranked question would be assigned a score of 30), the next one down a lower score (i.e. minus 1) and so on down the list, until the lowest ranked question received the lowest score (i.e. 1). To obtain the overall ranking, the scores for each question from each of the stakeholder groups were added together. The question with the highest aggregated score was ranked number 1 overall and the lowest score was ranked lowest overall. This gave the overall interim ranking to the research questions and the rankings for each of the stakeholder groups, whilst minimising bias owing to numbers of responses from each stakeholder group.

Priority setting workshop

A final prioritisation workshop was held in December 2016 in Birmingham, UK, in order to condense the number of questions generated by the stakeholder surveys to (a minimum of) a “Top 10” list of research questions agreed by consensus. The workshop followed the standard JLA approach as described in detail in the JLA Guidebook (www.jla.nih.ac.uk/jla-guidebook/) and was facilitated by the JLA’s Senior Advisor. Reimbursement of expenses (all members) and remuneration for people’s time (members of the public only) was guided by the INVOLVE UK guidelines. All expenses were processed centrally by the JLA for all stakeholders. Streamlining of this process through a single line of communication facilitated appropriate procurement, efficient submission of expenses and timely reimbursement.

Results

Initial survey

Completeness of initial survey

The initial survey was completed by 790 respondents with 382 (48%) of those answering at least one of the open ended questions. Only one person requested a hard copy of the initial survey. Completeness of the initial survey across survey sections is outlined in Table 2.

Table 2 Completeness of initial survey

Demographic questions	Number	Completed (%)
Consent to participate (yes)	790	100%
Age (scale)	777	98%
Respondent’s role in trials	717	91%
Where respondent lives	720	91%
Gender	711	90%
Affiliated trial subject	711	90%
Wish to be involved in future research (yes/no)	713	90%
Specific open-ended feedback questions		
“How trials are planned and designed”	382	48%
“How trials are carried out”	350	44%
“Information people are given when they join a trial”	359	45%
“Trial recruiters who invite people to take part”	291	37%
“The motivation of people in joining a trial”	314	40%
“Other questions or comments”	149	19%

Demographic information – initial survey

The proportion of completed surveys from each stakeholder group is presented in Table 3. The stakeholder group with the highest response were researchers involved in recruiting participants (21%, $n = 150$). The number of completed survey responses across stakeholder groups, ranged from 61 to 150 (see Table 3).

Table 3 Initial survey respondent roles

Which one of the following best describes your main role in a randomised trial?	Number	Percentage
Answer options		
A person invited to take part in a trial	83	12
A researcher involved in recruiting participants	154	21
A non-researcher (e.g. clinician or health professional) involved in recruiting participants	77	11
A principal/chief investigator	124	17
A researcher involved in aspects of the trial other than frontline recruitment	183	26
A trial methodologist (someone who specializes in the methods of how trials are designed, run, analysed and reported)	87	12
Other (please specify)	9	1
Total ^a	717	100

^aData were missing in 73 respondents

Information about the clinical subject area of experience was available for 711 respondents. The highest proportion of respondents identified cancer (18%), followed by neurology – neurodegenerative diseases, vascular diseases (12%) (Table 4).

Respondents to the survey were predominantly from England (74%), followed by the Republic of Ireland (17%), Scotland (4%), Wales (2%) and Northern Ireland (<1%).

Initial survey responses, collating themes and merging questions

A total of 1880 questions for research were formulated from the 790 survey responses, which had 1693 open text responses. Merging similar themed questions and removing duplicates reduced this to 496 questions. Where a duplicate question was removed, the total cumulative frequency of the number of times this question was mentioned across stakeholders was noted and each new version of the database was saved to allow

Table 4 Summary of initial survey respondents affiliated trial subject areas

Trial subject area	Number	Percentage
Oncology/haematology – cancer	142	20
Neurology – neurodegenerative diseases, vascular Diseases	93	13
Cardiovascular diseases	44	6
Mental health	42	6
Metabolism – diabetes mellitus	38	5
Orthopaedics/musculoskeletal	34	5
Reproductive Health	34	5
Gastroenterology – hepatology, nephrology	29	4
Paediatrics/neonates	22	3
Respiratory	20	3
Inflammatory conditions (e.g. osteoarthritis, rheumatoid arthritis, fibromyalgia)	15	2
Surgery	15	2
Ophthalmology	14	2
Vaccines – preventive vaccines	13	2
Dementia/ageing	12	2
Infectious diseases	9	1
Dermatology	7	1
Palliative care	4	1
Involved in multiple trials	38	5
Other	79	11
Total ^a	704	100%

^aData were missing or response was “Do not know” in 86 respondents

traceability of concepts and questions from verbatim quotations from individual respondents.

Where appropriate, questions were merged into broader questions following review and discussion with the steering group. Finally, questions asked by more than 15 people and/or at least 6 of the 7 stakeholder groups were selected to progress to the interim survey of stakeholders. These criteria were developed through consultation with the Steering Group after presenting data across stakeholder groups. This resulted in the inclusion of 31 unique questions for research.

Interim survey

Completeness of interim survey

The interim survey was completed by 815 respondents (female 71%, male 29%), of whom 100% selected at least one research question for prioritisation.

Demographic information – interim survey

A total of 802 respondents provided information on their stakeholder group. A full breakdown of affiliated roles is presented in Table 5.

Respondents to the interim survey were predominantly from England (77%, $n = 603$), followed by the Republic of Ireland (9%, $n = 72$), Scotland (7%, $n = 54$), Wales (5%, $n = 38$) and Northern Ireland (2%, $n = 14$).

Information about the clinical subject area of experience was available for 775 respondents (Table 6).

Interim survey ranking

The Steering Group followed the standard JLA approach and used ranked weighted scores across all stakeholder

Table 5 Interim survey respondent roles

Answer options	Number	Percentage
Which one of the following best describes your main role in a randomised trial?		
A person invited to take part in a trial	108	13
A researcher involved in recruiting participants	146	18
A non-researcher (e.g. clinician or health professional) involved in recruiting participants	63	8
A principal / chief investigator	186	23
A researcher involved in aspects of the trial other than frontline recruitment	206	25
A trial methodologist (someone who specialises in the methods of how trials are designed, run, analysed and reported)	90	11
Other	3	< 1
Total	802	98

Table 6 Summary of interim survey respondents affiliated trial subject areas

Trial subject area	Number	Percentage
Oncology/haematology – cancer	161	21
Neurology – neurodegenerative diseases, vascular diseases	77	10
Cardiology – cardiovascular disease	62	8
Orthopaedics/musculoskeletal	56	7
Mental health	51	7
Metabolism – diabetes mellitus	47	6
Vaccines – preventive vaccines	45	6
Reproductive health	33	4
Critical care	24	3
Infectious diseases	21	3
Gastroenterology – hepatology, nephrology	18	2
Surgery	16	2
Respiratory	15	2
Paediatrics/neonates	13	2
Inflammatory conditions (e.g. osteoarthritis, rheumatoid arthritis, fibromyalgia)	11	1
Ophthalmology	9	1
Involved in multiple trials	34	4
Other	82	11
Total ^a	785	96

^aData missing in 30 respondents

groups to decide which of the interim survey research questions to take forward to the final priority setting workshop. For this workshop, based on JLA experience in prioritising questions, 25 questions were brought forward for discussion and final prioritising by the group.

Final prioritization workshop

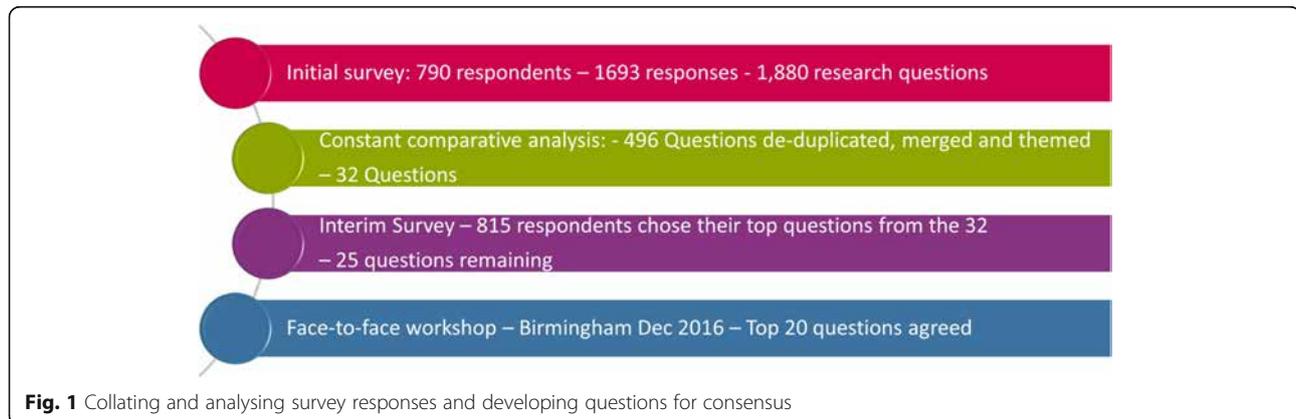
The final prioritisation workshop took place in Birmingham in December 2016 and a final Steering Group meeting was held the following day to review the results of the workshop. There were 31 participants representing the stakeholder groups who were invited to the face-to-face final priority setting workshop and on the day 26 participants attended. This was made up of 10 public members from trials or trial steering committees, 7 frontline researchers or non-researchers involved in recruitment, 6 trial methodologists, and 3 researchers or principal investigators. Some members of the PRioRiTty Steering Group attended as observers. The 26 participants were divided into three groups with a JLA facilitator for each group. Each group was provided with the short-listed questions in individual question cards with stakeholder group rankings from the voting process and an example quote from the original survey submissions noted on the back of them. The questions had been sent to the participants prior to the meeting so that they could have some time to familiarise themselves with the list and decide on what was important to them. The facilitators then guided the participants through the process of discussing the questions and agreeing, by consensus, a “Top 10” from within them.

The group agreed that a ranked “Top 10” list would be created and ranking for an additional ten questions (11–20) would also be carried out. Therefore, the PRioRiTty list of research questions features a list of both the “Top 10” (Table 7) and the research questions ranked 11–20 are available (www.priorityresearch.ie).

The process of managing the data analysis is illustrated in Fig. 1.

Table 7 The “Top 10” research questions prioritised

Overall ranking	Uncertainty as research question
1	How can randomised trials become part of routine care and best utilise current clinical care pathways?
2	What information should trialists communicate to members of the public who are being invited to take part in a randomised trial in order to improve recruitment to the trial?
3	Does patient/public involvement in planning a randomised trial improve recruitment?
4	What are the best approaches for designing and delivering information to members of the public who are invited to take part in a randomised trial?
5	What are the barriers and enablers for clinicians/healthcare professionals in helping conduct randomised trials?
6	What are the key motivators influencing members of the public’s decisions to take part in a randomised trial?
7	What are the best approaches to ensure inclusion and participation of under-represented or vulnerable groups in randomised trials?
8	What are the best ways to predict recruitment rates to a randomised trial and what impact do such predictions have on recruitment?
9	What are the best approaches to optimise the informed consent process when recruiting participants to randomised trials?
10	What are the advantages and disadvantages to using technology during the recruitment process?



Accessibility of the research question list and repository of relevant research

The top 10 questions for research can be viewed on a user-friendly, dedicated website (www.priorityresearch.ie). Research teams conducting specific research relevant to any of the PRioRiTty research questions are requested to submit basic details of their work to the HRB-TMRN through the PRioRiTty website as a repository and central record platform for the level of research ongoing for each research question. This resource will be maintained by the HRB-TMRN and will be freely searchable and accessible.

Discussion

The JLA process of priority setting through partnership and consensus is already well-established around treatment uncertainties. We believe this is the first time the process has been used to identify uncertainties around research methodology.

This Priority PSP identified that stakeholders believe that to improve the process of how people are recruited to randomised trials, research attention needs to focus on normalising trials as part of clinical care, enhancing communication, addressing barriers, enablers and motivators around participation and exploring greater public involvement in the research process.

Challenges encountered

While using a modified JLA process contributed greater efficiency to the study, there were several challenges throughout the process that had to be addressed at each stage. Given that the topic under discussion was research methodology, we were concerned that the voice of the public may have been overshadowed by that of research academics and practitioners. This issue was discussed at all stages, from completing the surveys through to the face-to-face workshop. To minimise this

risk, regular checking of survey responses and dedicated promotion of the survey to the public, in particular, was carried out. Our experience suggests that engaging people to take part in research about how to improve recruitment of people into trials is difficult and this is reflected in a lower response to the initial and interim surveys from members of the public relative to other stakeholder groups. However, our efforts from the beginning of the work, and in particular our Steering Group public representatives, helped ensure that the involvement of the public was meaningful and relevant throughout the project, that the process and language was accessible and that public representatives understood the work and felt that they could contribute on an equal basis.

The face-to-face workshop was not without its challenges. The workshop participants were assigned to small groups initially, each of which had a mixture of stakeholders. The dynamic of each group was slightly different, due to different backgrounds, perspectives, personalities, communication styles, expertise and levels of confidence. The groups were required to cover some complex issues regarding research methodology in discussing the uncertainties presented and reach a consensus around prioritising them. The discussions were robust and lively with each person, as might be expected, taking strong ownership of their own priorities and personal preferences. The presence of an experienced facilitator to moderate those discussions was fundamental in ensuring full, fair, respectful and equal participation. The facilitators took steps to ensure that no one dominated, or was excluded from, the discussion. Pragmatism was required within each group to reach acceptable compromises and revision of opinions in the search for consensus. The small groups then reconvened to one large group to agree the final “top ten”. Good facilitation was again instrumental here to

reach a democratic compromise where no-one felt coerced to let go of the priorities around which they felt strong ownership.

Another significant challenge centred on the language used during all stages of this PSP, for example, how the term “trial participants” was interpreted; some viewed this as patients only, others thought of clinicians and healthcare workers as “trial participants”. Other items centred around classifying “hard-to-reach” or “vulnerable” groups, for which public and patient contributors on the Steering Group were able to provide more appropriate wording such as “seldom-heard” groups. To overcome issues around appropriate language and in a bid to provide distilled language, the “GET IT” glossary (<http://getitglossary.org/> [12]) was used and words were hyperlinked to their specific translation on all materials, from website to initial survey to interim survey.

The submission of a large volume of data in the initial survey raised challenges around data management. The large volume of data needed to be interpreted, categorised and combined into themes whilst at the same time remaining true to the richness of the submission. The two members of the project group independently analysing the data, randomly and regularly checked a sample of each other’s interpretation and categorisation with a third member adjudicating on any disagreements. All data were filed in a manner that facilitates tracking each recruitment question back to the original submission by the stakeholder.

The involvement of a wide stakeholder group in a face-to-face meeting also presented new challenges to the team, for which the JLA have considerable experience. For example, while the research team may be very familiar with using email for communication, this was less so for some of the invited participants, with many not familiar with using email attachments or checking their email infrequently. The team adapted by communicating via phone and text, making sure that every email was accompanied by a phone reminder or text message where needed.

Many of the challenges we encountered have previously been described by others attempting public involvement with research [13–15]. Similar to their experiences, we found that a flexible and responsive approach was needed to successfully address the challenges.

Implications of PRioRiT

We believe that the PSP model was applied successfully to the identification and prioritisation of methodological uncertainties in trial recruitment and that such an approach has merit in identifying uncertainties in

other trial processes e.g. retention, reporting etc. The use of the PSP process for this project also provided some very useful learning around the successful engagement of the public and patients in the conversation about trial methodology. Given that the challenge of recruiting to trials is an international problem, repeating this priority setting exercise in other countries may need to be considered. The PRioRiT study was Ireland and UK centred but having found the PSP process to be effective it could be replicated in other jurisdictions where different trial infrastructural supports are available. International collaborations should now be fostered and the efforts of research groups all over the globe combined to address these prioritised methodological questions.

Conclusion

This bringing together of people, engaged researchers, clinicians and the public in an exercise of discussion, knowledge exchange and consensus, in identifying, agreeing, prioritising and disseminating a list of the most important methodological uncertainties surrounding recruitment to trials. Methodology research, such as the PRioRiT PSP, is an essential adjunct to clinical research. Such so called “research on research” is acknowledged as an important contributor to reducing waste and inefficiencies in research [16]. The critical end point of the PRioRiT PSP is a top ten list of trial recruitment uncertainties, determined by those directly involved in trials, which will inform future research around developing more effective recruitment procedures and processes that encourage participation in randomised trials. The investigation of, and answers to, the research questions identified by PRioRiT will inform future research designs and increase the efficiency of recruitment to trials. This, according to Salman et al. (2014), will minimise avoidable sources of waste and inefficiency in research [17]. Further, ongoing partner engagement in designing future studies around the identified priorities will be encouraged and supported so that trials might be better designed and implemented in the future. Recruitment to trials is an international issue that would be best addressed by engaging in international collaborations for those ongoing partner engagements.

International research groups are encouraged to collaborate and contribute evidence to answer the prioritised recruitment questions. Researchers are encouraged to identify opportunities for building robust proposals to answer these priorities and research funders are encouraged to integrate the priorities into their organisational plans, research strategies and funding calls.

Appendix

Table 8 Evidence checking

Uncertainty as research question	Evidence reviewed (878 screened, 76 full text, 34 extracted)
1. How can randomised trials become part of routine care and best utilise current clinical care pathways?	• No evidence available
2. What information should trialists communicate to members of the public who are being invited to take part in a randomised trial in order to improve recruitment to the trial?	• No evidence available
3. Does patient/public involvement in planning a randomised trial improve recruitment?	• No evidence available
4. What are the best approaches for designing and delivering information to members of the public who are invited to take part in a randomised trial?	<ul style="list-style-type: none"> • Bonevski, B.; Randell, M.; Paul, C.; Chapman, K.; Twyman, L.; Bryant, J.; Brozek, I.; Hughes, C. Reaching the hard-to-reach: a systematic review of strategies for improving health and medical research with socially disadvantaged groups. <i>BMC Medical Research Methodology</i> 2014; 14:42. • Synnot, A.; Ryan, R.; Prictor, M.; Fetherstonhaugh, D.; Parker, B. Audio-visual presentation of information for informed consent for participation in clinical trials. <i>Cochrane Database of Systematic Reviews</i> 2014;9(5):CD003717. • Treweek, S.; Lockhart, P.; Pitkethly, M.; Cook, J. A.; Kjeldstrom, M.; Johansen, M.; Taskila, T. K.; Sullivan, F. M.; Wilson, S.; Jackson, C.; Jones, R.; Mitchell, E. D. Methods to improve recruitment to randomised controlled trials: Cochrane systematic review and meta-analysis. <i>BMJ Open</i> 2013;3(2):
5. What are the barriers and enablers for clinicians/healthcare professionals in helping conduct randomised trials?	<ul style="list-style-type: none"> • Newington, L.; Metcalfe, A. Researchers' and clinicians' perceptions of recruiting participants to clinical research: a thematic meta-synthesis. <i>Journal of Clinical Medicine Research</i> 2014;6(3):162-72
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7. What are the best approaches to ensure inclusion and participation of under-represented or vulnerable groups in randomised trials?	<ul style="list-style-type: none"> • Bellera, C.; Praud, D.; Petit-Moneger, A.; McKelvie-Sebileau, P.; Soubeyran, P.; Mathoulin-Pelissier, S. Barriers to inclusion of older adults in randomised controlled clinical trials on non-Hodgkin's lymphoma: a systematic review. <i>Cancer Treatment Reviews</i> 2013;39(7):812-817 • Cooper, C.; Ketley, D.; Livingston, G. Systematic review and meta-analysis to estimate potential recruitment to dementia intervention studies. <i>International Journal of Geriatric Psychiatry</i> 2014;29(5):515-25 • Nicholson, L. M.; Schwirian, P. M.; Groner, J. A. Recruitment and retention strategies in clinical studies with low-income and minority populations: progress from 2004 to 2014. <i>Contemporary Clinical Trials</i> 2015;45(Pt A):34-40
8. What are the best ways to predict recruitment rates to a randomised trial and what impact do such predictions have on recruitment?	<ul style="list-style-type: none"> • Cooper, C. L.; Hind, D.; Duncan, R.; Walters, S.; Lartey, A.; Lee, E.; Bradburn, M. A rapid review indicated higher recruitment rates in treatment trials than in prevention trials. <i>Journal of Clinical Epidemiology</i> 2015;68(3):347-54
9. What are the best approaches to optimise the informed consent process when recruiting participants to randomised trials?	<ul style="list-style-type: none"> • Farrell, E. H.; Phillips, K.; Morgan, B.; Savage, K.; Lewis, V.; Whistance, R. N.; Kelly, M.; Mann, M.; Blazeby, J. M.; Kinnersley, K.; Edwards, A. G. K. Audio-visual aids for informed consent for invasive healthcare procedures: A systematic review. <i>British Journal of Surgery</i> 2013;100:60 • Gillies, K.; Cotton, S. C.; Brehaut, J. C.; Politi, M. C.; Skea, Z. Decision aids for people considering taking part in clinical trials. <i>Cochrane Database of Systematic Reviews</i> 2015;11:CD009736 • Halkoaho, A.; Pietila, A. M.; Ebbesen, M.; Karki, S.; Kangasniemi, M. Cultural aspects related to informed consent in health research: A systematic review. <i>Nursing Ethics</i> 2015;23(6):698-712 • Eltorki, M.; Uleryk, E.; Freedman, S. B. Waiver of informed consent in pediatric resuscitation research: a systematic review. <i>Academic Emergency Medicine</i> 2013;20(8):822-34

Table 8 Evidence checking (Continued)

Uncertainty as research question	Evidence reviewed (878 screened, 76 full text, 34 extracted)
	<ul style="list-style-type: none"> • Boland, J.; Currow, D. C.; Wilcock, A.; Tieman, J.; Hussain, J. A.; Pitsillides, C.; Abernethy, A. P.; Johnson, M. J. A systematic review of strategies used to increase recruitment of people with cancer or organ failure into clinical trials: implications for palliative care research. <i>Journal of Pain & Symptom Management</i> 2015;49(4):762-772.e5 • Montalvo, W.; Larson, E. Participant comprehension of research for which they volunteer: a systematic review. <i>Journal of Nursing Scholarship</i> 2014;46(6):423-31 • Nishimura, A.; Carey, J.; Erwin, P. J.; Tilburt, J. C.; Murad, M. H.; McCormick, J. B. Improving understanding in the research informed consent process: a systematic review of 54 interventions tested in randomized control trials. <i>BMJ Medical Ethics</i> 2013;14:28 • Treweek, S.; Lockhart, P.; Pitkethly, M.; Cook, J. A.; Kjeldstrom, M.; Johansen, M.; Taskila, T. K.; Sullivan, F. M.; Wilson, S.; Jackson, C.; Jones, R.; Mitchell, E. D. Methods to improve recruitment to randomised controlled trials: Cochrane systematic review and meta-analysis. <i>BMJ Open</i> 2013;3(2): • Synnot, A.; Ryan, R.; Pictor, M.; Fetherstonhaugh, D.; Parker, B. Audio-visual presentation of information for informed consent for participation in clinical trials. <i>Cochrane Database of Systematic Reviews</i> 2014;9(5):CD003717 • Wilman, E.; Megone, C.; Oliver, S.; Duley, L.; Gyte, G.; Wright, J. M. The ethical issues regarding consent to clinical trials with pre-term or sick neonates: a systematic review (framework synthesis) of the empirical research. <i>Trials [Electronic Resource]</i> 2015;16:502
10. What are the advantages and disadvantages to using technology during the recruitment process?	<ul style="list-style-type: none"> • No evidence available

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Availability of data and materials

The datasets used and/or analysed during the current study are held by the PRioRiTy team (Sandra.galvin@nuigalway.ie).

Authors' contributions

DD, VS, SG, PW, ST and ABJ conceived the study. All authors were involved in designing the study. DD led the study and PH oversaw delivery of study components. SG and PH were responsible for survey development, tracking survey responses and data analysis in both surveys. SG was responsible for website development. DD, HG and VS were responsible for evidence checking of the literature. ST, CG and PRW reviewed the systematic reviews to check for unanswered questions. PH and SG wrote the first draft of the manuscript. CW and BM organised the face-to-face prioritisation meeting and facilitated reimbursement and remuneration to attendees. CW was responsible for communication to all study stakeholders. All authors contributed to the conduct of the study, including survey promotion, interpretation of the results, manuscript review and input. All authors read and approved the final manuscript.

Author information

Not applicable.

Ethics approval and consent to participate

Ethical approval was obtained for the PRioRiTy study from Galway University Hospitals Ethics Committee (CA.1496 – PRioRiTy) Participants indicated written consent to participate at the beginning of the survey.

Consent for publication

Not applicable.

Competing interests

Prof Shaun Treweek is a senior Editor for *Trials*.

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Author details

¹Health Research Board – Trials Methodology Research Network, Galway, Ireland. ²School of Nursing and Midwifery, NUI Galway, Galway, Ireland. ³MRC North West Hub for Trials Methodology, University of Liverpool, Liverpool, UK. ⁴Health Services Research Unit, University of Aberdeen, Aberdeen, UK. ⁵James Lind Alliance, Wessex Institute, University of Southampton, Southampton, UK. ⁶Oxford University Hospitals NHS Foundation Trust, Oxford, UK. ⁷Birmingham Clinical Trials Unit, University of Birmingham, Birmingham, UK. ⁸Health Research Institute, University of Limerick, Limerick, Ireland. ⁹Division of Health and Social Care Research, King's College London, London, UK. ¹⁰Irish Platform for Patient Organisations, Science and Industry (IPPOSI), Dublin, Ireland. ¹¹National Institute for Health Research, London, England, UK. ¹²HRB-Clinical Research Facility Galway, NUI Galway, Galway, Ireland. ¹³Centre for Public Health, Queen's University Belfast, Belfast, UK. ¹⁴Dental Translational and Clinical Research Unit (DenTCRU), Leeds NIHR CRF, University of Leeds, Leeds, UK. ¹⁵NIHR Evaluation Trials and Studies Coordinating Centre (NETSCC), University of Southampton, Southampton, UK. ¹⁶School of Nursing & Midwifery, Trinity College Dublin, Dublin, Ireland. ¹⁷National Institute for Health Research-Clinical Research Network (NIHR CRN), Birmingham, England, UK.

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