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More than a Top 10:

How James Lind Alliance Priority Setting Partnerships transform research, people and organisations

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About this project

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TwoCan ASSOCIATES

About TwoCan Associates

TwoCan Associates was established in 2004 and specialises in promoting and supporting patient and public involvement in health and social care research. TwoCan works with NHS and voluntary organisations to help clarify the purpose of involvement, to improve policy and practice, and develop meaningful and effective partnerships. The Director, Dr Kristina Staley, was joint lead on this project. Kristina has worked as an Information Specialist on several JLA PSPs and is familiar with the issues that arise in developing researchable questions from the Top 10's. She carried out an early scoping study in 2008 for the JLA, to explore how public sector research funders make use of research priorities and the potential challenges they face. Further information available at: www.twocanassociates.co.uk

About Crowe Associates

Crowe Associates, established 20 years ago has a strong record of facilitation, consultation and project management in health and social care organisations and especially in patient and public involvement. Sally Crowe, the joint lead on this project, has partnered with NIHR, INVOLVE, research networks and many voluntary organisations over the years on aspects of public involvement in research. Sally helped establish the JLA in 2003, was responsible for the methodology and supported many PSPs. She has also published one of the few evaluative papers about the research outputs of PSPs and remains curious and engaged in their influence on the research agenda. Further information is available at: www.crowe-associates.co.uk

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Executive Summary

Introduction

The James Lind Alliance (JLA) Priority Setting Partnerships (PSPs) enable clinicians, patients and carers to work together to identify and prioritise the questions they would like answered by research. PSPs aim to address the mismatch between what researchers choose to research, and what patients, carers and health professionals actually want to know. PSPs thus provide an important and valuable opportunity for the end users of research to help shape the research agenda.

The aim of this project was to identify the most effective ways for JLA PSP research priorities to influence decisions about what research projects get developed and funded, by evaluating different approaches taken by JLA PSPs to date. The objectives were to:

(a) identify examples of success and develop case studies to explore how researchers and funders were positively influenced in each case

(b) identify challenges and tensions in the use of JLA PSP priorities by researchers and funders and how these have been addressed

(c) explore what practical approaches could be taken to maximise the impact of PSPs, both during the process as well as after identifying a Top 10 list of priority topics

We interviewed 20 people who had experience of working on a JLA PSP, or of working with JLA PSP priorities. The interviews took place between April and May 2019. They included 13 PSP Leads, one manager in a funding organisation, three researchers and three patients. The PSP Leads came from a variety of organisations including charities, universities and patient groups, some of which also fund research.

The main audience for this report is past, present and future PSPs. We hope the lessons learnt will help PSPs with the work that happens after the JLA process has concluded, and could usefully inform future evaluations.

Key lessons and conclusions

Even with the small number of PSPs involved in this project, we have revealed a rich and complex picture of the outcomes and impacts of JLA PSPs which go beyond simply funding research, and broaden the definition of what success looks like. Taking part in a JLA PSP can have a dramatic impact on the individuals involved, both professionally and personally. For example, it has enabled patients to expand and enhance their involvement in other parts of the research system, improved the reputations and status of researchers, and changed clinicians' clinical practice. Organisations that lead a JLA PSP report major cultural shifts that result in new partnerships with other organisations, promote greater internal collaboration across departments and/or extend and enhance patient and public involvement in their work. In charities that fund research, the experience has changed relationships between funders and researchers, with less emphasis placed on competition and greater emphasis on working together to achieve a common goal. These 'collateral benefits' come in addition to a shift in research funding towards the issues that matter most to patients, carers and healthcare professionals.

The context for each PSP is hugely significant, for example whether the research area is contested, whether an active research community already exists and whether considerable research on a topic has already been completed. The starting point of the PSP, what it aims to achieve and the specific individuals and organisations involved – these all shape the process and outcomes, making it difficult to draw general conclusions about 'how to succeed'. With this caveat, we have identified some of the key lessons for PSPs, recognising that these will not be universal. We conclude it is important for PSPs to:

- Plan for the end of the PSP at the beginning of the project, in particular to clarify who owns the outputs, who will make decisions about how they are used, who will be accountable for what happens next and how the follow-up work will be resourced
- Develop dissemination plans to reflect the PSP's strategic goals, which may go beyond funding research and reach audiences beyond funders and researchers
- Make greater strategic use of patients, carers, clinicians and researchers in promoting the JLA priorities through their own networks, rather than focusing solely on publications
- Work with funders after the PSP to shape their research agenda, recognising that they may not believe it is their responsibility to respond to the priorities – this influencing work requires people with the right skills and experience, often senior leaders within organisations
- Ensure continued involvement of clinicians, patients and carers in the translation of JLA PSP priorities into themed calls and research projects, so that the spirit of the original questions are maintained
- Carry out foundation work to build researchers' capacity to respond, promoting collective thinking on an issue, as well as identifying barriers to the research and addressing them
- Collect information about research that has been completed in response to JLA PSP priorities and make it publicly available
- Assess the wider impacts of the JLA process and share these amongst all stakeholders to promote a deeper understanding of how PSPs work and the value of JLA PSP priorities

This evaluation has also begun to identify factors within the wider research system that limit the influence of JLA PSP priorities. These include the values held by funders and researchers, and the dominant culture within research organisations. This may mean that funders and researchers use the JLA PSP priorities to endorse and legitimise what they have already planned to do, rather than making significant change. It may be beyond the power of individual PSPs to bring about the wholesale cultural shift required to genuinely change the national research agenda in favour of patients, carers and clinicians' priorities. Addressing deeply embedded beliefs and values is likely to require action from a wide range of stakeholders.

For this reason, many of the lessons contained in this report could be helpfully addressed by others beyond those immediately involved in a JLA PSP, including the JLA Secretariat, JLA Advisers, funders and

researchers. We recommend that the findings form the basis for future conversations, enabling these stakeholders to work together to develop their views on how best to respond. We suggest that some of the questions that could be usefully addressed include:

- **Planning**: If the work of promoting and influencing others at the end of a PSP needs to be properly planned and resourced at the beginning, is there a role for the JLA Secretariat to support this planning? What level of resourcing should be recommended? And where should this resource come from?

- **Disseminating and influencing**: If simply disseminating the priorities is not always sufficient to promote their uptake by researchers and funders, what can be learnt from implementation science about how to encourage others to change their behaviour in light of new evidence?

- **Responding by funding relevant research**: When assessing a research project that aims to address a JLA PSP priority, how can this be judged in a practical and meaningful way? What should funders, grant reviewers and members of funding panels be asking and looking for?

- **Responding in ways other than through research**: How can non-research questions be used for the benefit of patients, carers and clinicians i.e. to meet their information needs and to improve healthcare policy and practice? Which stakeholders need to be involved in this work and how can it be resourced?

- **Translating**: What are the tasks involved in the translation step from JLA PSP priority to research question or themed call? Who needs to be involved? What support and information do they need to do the tasks well?

- **Evaluating**: What are practical and meaningful ways of capturing whether new research addresses a JLA PSP priority? How can the scale and nature of the change to a portfolio be described? How can the change in the nature of individual research projects be captured? How can the wider impacts of the JLA process be captured?

- **Transforming research organisations**: If the goal is to transform the research culture in such a way that it better reflects the needs and interests of the end-users, what are the implications for the way the research organisations currently function, in terms of what they do and how they do it? How do they need to change to be able to respond in a meaningful way to the JLA PSP priorities?

- **Transforming people**: How can individuals' contributions to the process be better recognised and rewarded within the research system? How can the skills and experience that individuals gain through the JLA process be put to better use in the ongoing work and in other contexts?

1. Introduction

The James Lind Alliance (JLA) Priority Setting Partnerships (PSPs) enable clinicians, patients and carers to work together to identify and prioritise the questions they would like answered by research. The shared interests of patients, carers and clinicians have often been overlooked in clinical research. Their questions have been neglected and many areas of potentially important research have not been addressed[1]. Even when researchers do address questions of importance to patients, carers and clinicians, they often fail to provide answers that are useful in practice. JLA PSPs aim to address this mismatch between what researchers choose to research, and the practical information that is needed day-to-day by patients, carers and health professionals. PSPs thus provide an important and valuable opportunity for patients, carers and clinicians to help shape the health research agenda[2].

The JLA process, as described in the JLA Guidebook[3], is now well-established and over 100 PSPs had taken place or were underway in July 2019. The majority of PSPs have been based in the UK, but a substantial number have taken place in other countries such as Canada and the Netherlands, and some PSPs have been international, e.g. Liver Glycogen Storage Disease PSP. This report mainly focuses on PSPs in the UK.

The PSP process itself comes to an end at the point at which a Top 10 list of research priorities has been identified. The partner organisations involved in each PSP then make great efforts to publicise the results amongst UK and sometimes international audiences. They aim to encourage researchers to develop projects that address the top priorities, and to encourage research funders to fund this work.

The aim of this project was to identify the most effective ways for JLA PSP research priorities to influence decisions about what research projects/ programmes get developed and funded, by evaluating some of the different approaches that have been taken to date. This involved carrying out an illuminative evaluation (see Section 3), seeking the views of those with direct experience of conducting PSPs and/ or working with their outputs.

The objectives were to:

(a) identify examples of success and develop case studies to explore how researchers and funders were positively influenced in each case

(b) identify challenges and tensions in the use of JLA research priorities by researchers and funders and how these have been addressed

(c) explore what practical approaches could be taken to maximise the impact of PSPs, both during the process as well as after identifying a Top 10 list of priority topics.

[1] Crowe S, Fenton M, Hall M, Cowan K and Chalmers I. (2015) Patients', clinicians' and the research communities' priorities for treatment research: there is an important mismatch. Research Involvement and Engagement, 1:2.

[2] The James Lind Alliance Priority Setting Partnerships (JLA PSPs) website: About PSPs

[3] The James Lind Alliance Priority Setting Partnerships (JLA PSPs) website: JLA Guidebook

1.1 Who worked on this project and how it was funded

The project was developed and carried out by Sally Crowe from Crowe Associates and Kristina Staley from TwoCan Associates, with support from Joanna Crocker, Caroline Jordan and Polly Kerr from the Nuffield Department of Primary Care Health Sciences, University of Oxford and guidance from an Advisory Group (see Section 3.2).

The work was funded by a grant given by the National Institute for Health Research (NIHR) Oxford Biomedical Research Centre, at the direction of Prof Trish Greenhalgh, Nuffield Department of Primary Care Health Services, University of Oxford.

It was independent of the James Lind Alliance Secretariat (JLA Secretariat) based at the National Institute for Health Research Evaluation, Trials and Studies Coordinating Centre (NETSCC), who did not fund nor direct the project. The extent of their input was to helpfully provide information and contact details for people who had worked on completed PSPs (see Section 3.2). Katherine Cowan, as an independent JLA Adviser and member of the team that helped establish the JLA, was a member of the project Advisory Group and provided advice and guidance throughout.

1.2 Rationale and scope

One of the most commonly asked questions about the JLA PSPs is 'What is the impact of the priorities on research?' To date, this has not been addressed systematically. The JLA Secretariat collect evidence of impact and share this on the JLA website, and individual PSPs (but not all) keep records of research responses and activity relating to their priorities. However, with a large number of PSPs completed, and over 16 years of development and activity, it seemed timely to explore and evaluate the impact of JLA PSPs on research and in other ways.

The Advisory Group for this project helped to determine the scope of the evaluation by identifying a range of outcomes and impacts of JLA PSPs. They concluded that the outcomes (the immediate and observable effects) of a JLA PSP go beyond simply identifying a list of prioritised questions to be answered by research, and include:

- New partnerships and collaborations between the JLA PSP partner organisations
- Other priority issues identified by patients, carers and clinicians – known unknowns, and questions about health policy and practice
- New actions taken by individuals in response to their experience of taking part in a PSP

The main impacts of JLA PSPs (the broad, long-term effects) were considered to be changes in the research culture, i.e. a change to 'what' research gets done and 'how' it gets done, by influencing what research projects are developed and funded. However, the Advisory Group also identified other less tangible impacts including:

- Changes in values within the research community
- Changes within organisations that participated in a PSP
- Changes amongst individuals as a result of taking part in a PSP

The evaluation therefore sought to explore this wide range of outcomes and impacts in relation to a small sample of past PSPs.

The focus of the evaluation was the impact of JLA PSPs from the point of identifying their Top 10 research priorities, to the point at which a priority becomes a funded research project. This meant the following was excluded:

- the JLA process itself and how this influences the outcomes and impact across different PSPs
- the funded research and its success or failure
- the impact of the research findings from JLA PSP priority projects on patients, carers and clinicians.

1.3 Terms used

The terms used in this report are defined as follows:

- **Patient** – someone who is involved in a JLA PSP who has lived experience of a health problem or disease. Some of the non-health related PSPs, e.g. the Adult Social Care PSP, involve stakeholders who are not patients, and sometimes the term service user is used instead.
- **Carer** – a carer of a patient who has a shared, but distinct lived experience from caring for someone with a health problem or disease.
- **Clinician** – a health professional who has clinical experience of a health problem or disease area.
- **PSP Lead** – someone who takes responsibility for the JLA PSP, works closely with the JLA Adviser and the PSP coordinator to champion the PSP and ensure it is successfully promoted, completed and disseminated to funders.
- **PSP Information Specialist** – manages the data generated by the PSP including checking existing research for the evidence for suggested research questions.
- **JLA Adviser** – provides the link between the PSP and the JLA and oversees and chairs meetings, processes and workshops.
- **JLA PSP Priorities** – the outputs of a JLA PSP, which include a Top 10 list of priority topics for research and a shortlist of typically another 10-15 questions that were also voted a high priority, but didn't make the Top 10.

2. Background & Context

2.1 The history of the JLA

The James Lind Alliance (JLA) stemmed from exploratory work by the James Lind Initiative (JLI)[4]. The JLI had its roots in a meeting in 2001 which was convened by the UK Cochrane Centre to 'consider what might be done to increase general knowledge about why treatments need to be tested rigorously, and what rigorous testing of treatments entails'[5]. This discussion evolved into 'how patients, clinicians and policy-makers should respond to uncertainties about the effects of treatments'[4], and a working hypothesis that 'uncertainties reflected in the research choices of people in academia and industry'[4] may not reflect the lived experience of patients and carers, or the clinical experience of health professionals. Discussion moved into exploration of models and methods[6] that could establish which uncertainties mattered to these groups (with the exclusion of industry and non-clinical academia), and also determine the most important of these to stimulate relevant research. This resulted in the Priority Setting Partnerships (PSP) that we know today, with the first PSP addressing asthma[7,8].

The original focus of the JLA created an expectation that prioritised treatment uncertainties would translate into funded clinical trials and systematic reviews of research. The choice to focus on interventional research was partly ideological, given the origins in the UK Cochrane Centre and evidence-based medicine movements, but also pragmatic. It was difficult to imagine a process that could manage collecting and prioritising uncertainties across the whole landscape of a disease process, or indeed address wider health and societal issues. The 'strapline' for the JLA became 'tackling treatment uncertainties together' and the early PSPs were focused on specific medical health conditions (e.g. asthma) and their prevention and treatment.

The evidence-based medicine and healthcare philosophy underpinning the JLA was similarly evident in the approaches used to categorise, structure, and confirm treatment uncertainties gathered through the JLA PSPs. Early PSPs aimed to translate the patient, carer and clinician uncertainties into PICO-formatted research questions (the framework used to structure clinical research questions, which identifies the Problem/Patient/Population, Intervention/Indicator, the Comparison and the Outcome). They also aimed to check all uncertainties with relevant and up to date systematic reviews, and created a Database of Uncertainties in the Effects of Treatments

[4] Chalmers, I (2003) The James Lind Initiative. Journal of the Royal Society of Medicine, 96:575–6.

[5] Chalmers I, Atkinson P, Fenton M, Firkins L, Crowe S and Cowan K (2013) Tackling treatment uncertainties together: the evolution of the James Lind Initiative, 2003-2013. Journal of the Royal Society of Medicine,106:482-91.

[6] Crowe S. (2009) Setting priorities for treatment uncertainties – A review of methods. Oxford: James Lind Alliance.

[7] Partridge N and Scadding J. (2004) The James Lind Alliance: patients and clinicians should jointly identify their priorities for clinical trials. Lancet, 364:1923–24.

[8] Elwyn G, Crowe S and Fenton M. (2010) Identifying and prioritizing uncertainties: patient and clinician engagement in the identification of research questions. Journal of Evaluation in Clinical Practice,16: 627–31.

(DUETs)[9] to capture all uncertainties. Not all of these approaches have survived in the current JLA model.

Recently JLA PSPs have simultaneously broadened in nature for example including topics such as Adult Social Work (2018) and Healthcare Associated Infections (ongoing), as well as narrowing down on smaller clinical areas e.g. Revision Knee Replacement (current), Pessary Use for Prolapse (2017), and specific areas of healthcare e.g. Intensive Care Units (2014) and Physiotherapy (2018). The scope of individual PSPs has also broadened beyond treatment, to include questions about cause, diagnosis, social care and prognosis. This means that the prioritised uncertainties cannot always be answered by clinical research methods, or always by clinical researchers. The approach to categorising the uncertainties and checking whether they have been answered by previous research has therefore been adapted.

While the overall message has slightly changed to emphasise that JLA PSPs now aim to ensure researchers and funders 'are aware of the issues that matter most to the people who need to use the research in their everyday lives', the values underpinning the process have remained the same. It still aims to be a process that is:

- robust and rigorous, particularly in terms of the involvement of a range of patient, carer and clinician perspectives
- transparent - in terms of describing the choices around the methods used and being open about the interests of those involved in the PSP
- accountable, particularly in sharing the outcomes with all stakeholders

One of the challenges for this evaluation is that the different PSPs involved took place at different time points and to some extent with different iterations of the JLA process. Understanding this context is important to explaining some of the differences in views of the interviewees.

2.2 The current JLA process

Currently central support for all past, present and potential JLA PSPs is provided by the JLA Secretariat, which consists of 2.5 whole time equivalent staff who carry out the following:

- recruiting and training JLA Advisers
- managing enquiries and communication with PSPs
- providing advice and guidance to groups considering initiating a PSP
- assessing prospective PSP's readiness to initiate a PSP
- working with JLA Advisers to support and uphold the JLA principles and approach
- maintaining the JLA website and Guidebook

JLA PSPs continue to be self-funding, with a few exceptions. The JLA Secretariat does not directly fund PSPs, but provides valuable infrastructure and networking support. The benefits from this research prioritisation activity are then widely shared.

[9] Fenton M, Brice A and Chalmers I. (2009) Harvesting and publishing patients' unanswered questions about the effects of treatments. In: Littlejohns, P, Rawlins, M (eds). Patients, the Public and Priorities in Healthcare, Abingdon: Radcliffe, pp. 165–80.

The JLA Guidebook, which describes the process of setting up, running and disseminating a JLA PSP in detail, is currently in its eighth edition. Asthma, Urinary Incontinence, Vitiligo and Schizophrenia PSPs took place as the approach was being developed (2007 – 2011), but from approximately 2010, the approach has been mostly consistent and JLA Advisers have ensured that the quality of the process has been maintained. The current process includes the following steps:

- Establishing a Priority Setting Partnership (recruiting partners, obtaining funding, setting up a steering group, agreeing the protocol)
- Gathering uncertainties (via a survey, focus group or other suitable method) to gather the questions that patients, carers and clinicians want research to answer
- Checking uncertainties, agreeing which are in-scope and which are out-of-scope, grouping them and developing summary questions for each group, removing questions that have already been answered by research
- Interim priority setting (usually an online survey), voting on a long-list of questions (typically 50-70 questions) to generate a short-list (typically 20-25 questions)
- Final priority setting at a workshop, agreeing a top-ten list of questions from the short-list
- Publishing the prioritised top-ten uncertainties, all the questions in the long-list and the original uncertainties
- Alerting research funders and researchers to the prioritised topics – usually the Top 10 and the shortlist of questions
- Thinking about and addressing the longer-term impact of priorities (The JLA acknowledges that this can be challenging)

A common misconception, perhaps based on its origins, is that the outputs of the JLA PSPs will be new, neat research questions, ready for funding. However, this is rarely the case since the process has evolved to include areas of research beyond treatments (see above). When patients, carers and clinicians submit their unanswered questions or uncertainties (usually through a survey) their questions can be quite specific, e.g. Would Vitamin D supplements reduce the likelihood of my condition getting worse?, or very broad e.g. 'What is the best diet for people with my condition?'. All related questions are grouped together and summarised by an overarching summary question, which is often broader in nature. Typically, the summary questions that go through to be prioritised, and subsequently form the output of the JLA PSP are therefore best described as questions that encompass a broad *topic or theme*, rather than specific *research questions*.

For funders and researchers, it is more challenging to know how precisely to respond to such broad themes than to a PICO formatted question, as discussed in Sections 4.4 and 4.5. Even the PSPs that have produced PICO formatted outputs have sometimes found these too broad to be immediately researchable. Even their priority questions still required further unpicking. However, since all PSPs typically publish the original uncertainties that underpin each of the summary questions, researchers and funders are able to identify what specific issues patients, carers and clinicians

considered important within each topic, and to understand the nuances between what different groups of people were asking.

2.3 Definitions of success

While all JLA PSPs aim for a similar goal and follow a shared process, the context in which each PSP develops often shapes their specific purpose and intentions. This has a bearing on what are felt to be the important outcomes of each PSP and what each considers to constitute success. For example, some PSPs may simply want to raise the profile of their research area, particularly if it is currently underfunded or inactive, or they may want to identify the research gaps that need addressing within an area that is already popular and very active. Others may want to segment and organise a broad area of research with the input of multiple perspectives, or alternatively, a JLA PSP may be the response to certain research topics being contested, to try to reach consensus amongst stakeholders about the most appropriate direction of travel.

Similarly, the context in which a PSP takes place, and whether it's initiated by a research institution, or by a clinical and health specialist group, or by a health research charity or patient/carer support group, will determine the nature and level of resource and skills available for follow-up work after the JLA process. Success may then need to be defined in terms of what's practically and feasibly possible within a given context, as well as what success means to these different stakeholders.

We also recognise that many of the challenges that JLA PSPs face in trying to change the research culture and influence funding decisions are not unique to them, and it may not always be possible for JLA PSPs to address constraints imposed by the wider research context. An important question to consider when evaluating PSPs is then 'What is within the PSPs' power to change and what external factors set limits to their success?'. Such factors include:

- How much funding is available within research funders' budgets for different types of research and what they are willing to fund
- Fairness in allocating research funding e.g. funding for cancer research versus other conditions or funding for research into rare conditions (as the number of JLA PSPs increase across a broad range of conditions, how different PSPs compete for the same funding pot is beyond the scope of this project)
- Political issues around funding allocation
- The interest, levels of ambition and aspiration for the priorities held by the main stakeholders in the PSP
- Corporate identity and funders' willingness to address different types of research, for example health charity X may identify as funder of biomedical research and be unwilling to fund other types of research
- Lack of capacity/ incentive to do different kinds of research within the existing research community
- Lack of appropriate methods or lack of researcher capability to address the questions raised by patients, carers and clinicians
- Disincentives or unwillingness amongst the research community to address a particular topic

2.4 Implications for this project

For the purposes of this evaluation, we therefore concluded that there isn't a single indicator of success that applies across all PSPs. We are aware that it is not as simple as identifying that a research project that fits with a JLA PSP priority has been funded. Nor is it useful to quantify how much funding has been allocated to a particular priority topic. Some priority topics can be addressed with minimal costs for example through small-scale qualitative research, whilst others might require an expensive clinical trial, and therefore the level of funding bears no relation to the response being meaningful or successful.

We also recognise there are limits to what any JLA PSP can achieve in relation to the wider context, and bringing about research culture change is not their responsibility alone. We have therefore asked PSPs about the context to their PSP and what they did tactically to address their specific issues, within the realms of what was possible for them.

We have drawn out lessons from the different experiences of the PSPs involved in this evaluation, but recognise these will not uniformly apply to all other PSPs. Our aim is to bring attention to important issues in the hope that the various stakeholders in the JLA process can draw out useful learning that will be relevant to their particular context.

Finally, we note that this was a modest project in terms of resources, and was completed within six months. We therefore recognise this was not an extensive evaluation, but hope it has begun to scope the issue of the impacts of JLA PSPs, and has usefully identified areas for future research and evaluation.

3. Methods

3.1 Overall approach

This evaluation took the form of an illuminative evaluation[10], i.e. it aimed to illuminate the outcomes and impacts of JLA PSPs as perceived by the people who took part in the evaluation, rather than judging these against fixed, external criteria. Illuminative evaluation is an established approach in the social sciences that uses naturalistic (qualitative) methods to explore the rationale, development, operations, achievements, and difficulties of an initiative.

The project involved the following stages:

- Initial preparation and consultation
- Planning interviews with key stakeholders
- Conducting interviews and analysing the findings
- Developing the conclusions and recommendations in a final report

These will now be discussed in turn.

3.2 Initial preparation and consultation

Key stakeholders in this project, including JLA Advisers and managers, representatives of funding organisations and representatives from previous JLA PSPs were consulted about the overall project aims and objectives. Their feedback helped to refine the final proposal. An early discussion with researchers and Patient and Public Involvement leads from the Nuffield Department of Primary Care Health Sciences, University of Oxford also helped with initial planning and with obtaining ethical approval from The University of Oxford Central University Research Ethics Committee (Reference number: R61467/ RE001).

Twelve people were invited to form an Advisory Group. They included people who could bring the perspectives of patients, carers, PSP Leads from a charity or university context, clinical researchers, funders of research, experts in public involvement in research and a JLA Adviser. The Advisory Group played an important role in shaping the project plans and developing the final recommendations to ensure these reflected the interests and concerns of all stakeholders in past and future PSPs.

3.3 Planning interviews with key stakeholders

The Advisory Group met at the beginning of the project to review the proposal and consider the overall scope of the evaluation (see Section 1.2). They helped with identifying criteria for the selection of interviewees, as well as the broad topics to be covered in interviews. A discussion of what constitutes 'success' or 'failure' in the context of a PSP as well as the factors that influence outcomes and impacts helped to determine what was appropriate to explore within the limits of this project.

It was agreed that the interviewees should include people involved in PSPs (patients, carers, clinicians, health charity and other organisation representatives) and people who work with the research priorities to develop

[10] Patton M . Utilization-focused evaluation. London: Sage, 1997.

calls for proposals or research projects/ programmes (individual researchers, staff from funding organisations). Overall, the sample of interviewees needed to include:

- PSPs that have reached a stage where they have useful lessons to share from their experience
- PSPs addressing a range of conditions/ topics to avoid bias from the research context
- UK based PSPs for lessons relevant to a UK context
- People with experience of more than one PSP (e.g. PSP Leads or funders)
- Researchers who have used JLA PSP priorities for funding applications (successful and unsuccessful)

The broad topics to cover in the interviews were agreed as:

- Who led the PSP and who owns the outputs and how does this influence what happens next?
- How have priorities been disseminated/ shared with funders/ researchers/ other stakeholders?
- How have funders worked with the priorities? And to what end?
- How have researchers worked with priorities? And to what end?
- How has taking part in a PSP affected the partner organisations, post-PSP?
- How has it affected the individuals who took part in the PSP, post-PSP?
- How have the other outputs of JLA PSPs been used for benefit?

A long-list of potential interviewees was generated based on suggestions from the Advisory Group, the personal experience of the two project leads and advice from the JLA Secretariat. Researchers who had worked with JLA PSP priorities, but had not been involved in a JLA PSP were identified via a call on Twitter. With the help of the Advisory Group, a set of criteria were developed to select interviewees based on their potential to provide useful learning within the broad areas of inquiry as described above (Appendix 2). Applying these criteria generated a short list of 20 people, which included 13 PSP Leads, three patients, three researchers and one manager in a funding organisation. The PSP Leads came from a variety of organisations including charities, universities and patient groups, some of which also fund research. Some interviewees were able to speak about more than one topic, and the final selection was made so that all topics would be covered by the complete sample. Only the two project leads were involved in this selection process to preserve the anonymity of those people interviewed (where requested).

Semi-structured interview schedules were developed for each category of interviewee by focusing on the specific topic areas those people were in a position to discuss. Some questions were general and asked of all interviewees, while some were specific to the individual and their specific areas of expertise and experience.

3.3 Conducting interviews and analysing the findings

All interviewees were invited to participate in the project by email and received an information sheet describing what taking part would involve for them, the steps taken to ensure confidentiality and data protection, and what would happen to their data at the

end of the project. The interviews were all conducted by phone and lasted 30-60 mins. At the start of each interview, the interviewee gave their oral consent to take part, to be recorded and to be re-contacted when a draft report was available for comment. The interviews were digitally recorded and transcribed, and the recordings and pseudo-anonymised transcripts stored on the Oxford University secure IT network (names of individuals, organisations and contact details were removed, but details within the discussion may still make it possible to identify the interviewees).

All the interviews were completed between April and May 2019. Both project leads read all the transcripts and agreed key themes within a broad framework that emerged from the first discussion with the Advisory Group. This framework is based on the stages involved in moving from identifying the Top 10 to developing research questions for funding and the different kind of impacts that the Group members described. It forms the section headings in Section 4 of the report with the findings: Planning, Sharing, Influencing, Responding, Translating, Evaluating and Transforming.

Sub-themes were identified using inductive thematic analysis, i.e. they were generated from the data up, rather than being shaped by existing theory. The two project leads discussed the data and identified these sub-themes. The analysis was shaped by their standpoints as people with considerable experience of either developing or supporting JLA PSPs, and with extensive experience of patient and public involvement in research more generally.

3.4 Developing the conclusions and recommendations

An anonymised draft report was discussed at a final Advisory Group meeting to ask for their views on the findings, and consider how best to share the lessons with different stakeholders in the JLA PSP process. This helped to develop the conclusions and recommendations in Sections 5 and 6. A second draft was then sent to all interviewees for comment, to check they were happy with the way their quotes had been used as well as the overall conclusions and recommendations. This approach ensured that the final report reflected the interests and concerns of all the different stakeholders in the JLA process.

4. Findings

The findings from the interviews have been grouped into the following topics, which will be discussed in turn:

4.1 **Planning**: preparing for the end at the start

4.2 **Sharing**: disseminating the JLA PSP priorities for research

4.3 **Influencing**: persuading others to respond to the JLA PSP priorities

4.4 **Responding:** using JLA PSP priorities to influence strategy and funding decisions

4.5 **Translating**: turning a priority topic into a research project

4.6 **Evaluating**: assessing whether JLA PSPs are making a difference to research

4.7 **Transforming**: changing culture, policy, organisations and people

4.1 Planning: preparing for the end at the start

The current JLA Guidebook advises PSPs to plan to publicise and disseminate the results of their PSP to increase the exposure of the priorities to potential funders and researchers and to promote the JLA process itself. It recommends that Steering Groups consider developing a dissemination plan (see Section 4.2) throughout the PSP process to maximise the chances of success. However, the PSP Leads who were interviewed suggested that PSPs may also need to consider other aspects of the follow-up work in the early planning, to ensure that sufficient resources are allocated to the post-PSP phase. These other aspects relate to:

- Achieving the PSP's strategic goals
- Agreeing ownership of the PSP priorities
- Responding to non-research questions
- Making good use of the skills and experience gained through the PSP

These will be discussed in turn, followed by a section that considers the implications for the JLA Secretariat.

Achieving the PSP's strategic goals

The strategic goals for each PSP may influence decisions about which aspects of their work they wish to disseminate and to which audiences. This needs to be a key element of their dissemination plan, as they may have additional goals that differ from getting the research priorities funded. For example, the PSP Leads on the Mental Health in Children and Young People PSP (2018) were clear that they also wanted the process to raise awareness of (a) the issues around the mental health of young people and (b) the need to disseminate past research findings (see Case Study 1). This is a common aim for a number of PSPs.

Case Study 1: Mental Health in Children and Young People PSP (2018)

The McPin Foundation decided to carry out this PSP as a way of making their contribution to the mental health sector. The PSP complemented other planned programmes of work including the Department of Health and Social Care's Framework for Mental Health Research (2017). Given the high level of interest in Young People's Mental Health at the time, they realised there would be widespread buy-in for the approach.

Vanessa Pinfold, Co-founder and Research Director at McPin viewed the purpose of their PSP as '*beyond research*'. She explained, "*We wanted to use it as a platform for young people's voices to be heard, for the parents to be heard, the teachers to be heard. We wanted all the stakeholders to help us, and to join this growing movement about the importance of kid's mental health. So right from the start we planned a launch event in Parliament… We're a tiny organisation, we don't have lots of resource to sit around policy tables and go to think tanks, so we decided the best use of our time was to try and get a parliamentary launch.*"

The launch proved extremely successful in engaging a wider audience, as Thomas Kabir, Head of Public Involvement at McPin, reported, *"An education specialist in the audience has since been in contact to say, 'What can my school do? We want to be part of the solution.' So, we have real commitment from people who want to address the problem of young people's mental health, of which research is one element. Campaigning, advocacy, policy work, service development are other arms. They are all strings to our bow.*"

The PSP also planned to raise awareness of the need for better dissemination of the findings from past research as Vanessa explained, "*From the very beginning, I was clear that for us to put significant resource into this project, we needed to maximise the impact and influence from it. From my perspective there are two key drivers. The first is obviously the research agenda, but actually, there's a bigger picture here. There are lots of questions people don't know the answers to, even though the research has been done. There's a great big dissemination issue which we also wanted to address.*"

During the initial planning of a PSP, the question of who to invite to join the Steering Group or to partner the PSP depends on its strategic purpose. The general advice is to invite representatives from organisations who could potentially fund the final priorities, but this may not always be what's required, or even possible as was the case in the Lyme Disease PSP (2012):

Lyme Disease is such a contested area, with strongly held views about what the disease is or isn't, that even during the setup of the PSP there were difficulties, and strong feelings that the PSP shouldn't even be happening. We wanted to get clinicians on board who weren't also people with Lyme Disease, to give our PSP greater credibility with the clinical researchers… but that proved extremely difficult. **PSP Lead**

Agreeing ownership of the priorities

If relevant stakeholders are persuaded to join the PSP, it seems important to have an exploratory conversation about who will own the outputs in terms of who will commit to responding to them, who will make decisions about how they are disseminated (see Section 4.2), and who will continue to provide accountability to patients, carers and clinicians for what happens after the PSP. This may mean different organisations reaching an agreement at the start about who will be responsible for these different elements, at least in principle, or it may mean different parts of the same organisation making a joint commitment to respond (see Case Study 2).

> *We had a number of charities there as stakeholder organisations and it certainly gave them a sense of ownership of those priorities. So one of them had a small grant scheme and for the subsequent couple of years they actually required grant applications to that scheme to address those priorities… it has been a very powerful thing for us to be able to say to the charities present 'Look, you signed up to these being the priorities, and here is something that addresses those priorities.'*
>
> **Researcher**

It may be difficult to fully commit when the precise outputs are uncertain, but it still seems important that the stakeholders give consideration to these issues and identify potential future roles and responsibilities:

> *I don't think when we set out to do the partnership that we necessarily were expecting all of the priorities to be owned by [our organisation]. But by the end of the workshop we saw that they were the really important priorities for [our patients] and we wanted to make sure we could do whatever we could to progress them.*
>
> **PSP Lead**

Case Study 2: Type 2 Diabetes PSP

Kamini Shah, Head of Research Funding at Diabetes UK, explained why they engaged different stakeholders before starting their Type 2 Diabetes PSP, *"The first thing we decided was what we might do with the outcomes, how we might influence them being taken forward. We knew we couldn't respond to all the top ten given our funding budget. So we went out to all the other funders of conditions linked to diabetes and its complications, and told them about the PSP. We thought if we wanted to partners for future joint funding calls, then we'd need to get them on board at the beginning."*

Diabetes UK also invested time in preparing all parts of their organisation for delivering the PSP and responding to the outputs, as Kamini described, *"We spent a year getting everyone in the organisation on board… including the people directly involved in the PSP, people in marketing, comms and the finance team. We also trained up our supporter care teams, so they were ready to help people complete the JLA surveys over the phone…. And we got buy-in from the rest of the organisation, so that they were ready to deal with the non-research questions that might require some other kind of response - the campaigning teams and the policy teams. We presented this work as being driven by people with diabetes and by healthcare professionals, and because that's central to our organisation's mission, there was a willingness to support it."*

Responding to the non-research questions

Many interviewees commented that initial planning could also usefully include consideration of how to respond to the issues raised through JLA PSP surveys that do not require a research response. For example, patients, carers and health professionals often raise questions that have already been answered by previous research, but people seem not to be aware of this – the unknown knowns (See Case Study 1). Some PSPs have responded by producing an FAQ (frequently asked questions) document alongside the final report of their PSP. If an organisation has capacity to do this, it may need to allocate resources at the time of planning the PSP and ensure the right people are prepared to lead on it.

Similarly, surveys often identify issues that might usefully inform a campaign rather than a research project, in which case there may be other organisations (or other parts of the same organisation) who may have capacity and interest in taking these up (See Case Study 2). While the idea of taking these kinds of questions elsewhere (e.g. sharing questions about GP (General Practitioner) training and practice with the Royal College of GPs) is often discussed at the time of categorising the responses to the first survey, we were unable to find examples of where this had actually happened in practice.

The JLA Guidebook encourages PSPs to consider these different uses of the out of scope data, but this is always parked until after the prioritisation process is complete, and at that stage it then comes down to individuals' capacity, resource and competing demands. In reality it may not always come to fruition despite good intentions. This is an area for further exploration and evaluation, as the issues raised across multiple PSPs offer a rich resource for useful learning.

Making good use of the skills and experience gained through the PSP

One of the patients who had taken part in a PSP commented that she would have liked to have used her lived experience and knowledge of research to contribute to the next steps of developing research projects, but there wasn't a mechanism that could enable her to do that, *"Coming to that final workshop, there wasn't anywhere further for me to go with it."* As the personal stories confirm (See Section 4.7), patient and carer partners in JLA PSPs gain considerable skills and confidence from the experience and could offer valuable input to other PSPs or research projects with involvement. Planning to make better use of these experts seems an important part of the legacy of a PSP (see Case Study 16).

A role for the JLA Secretariat

Some PSP Leads commented that much of their planning had focused on the JLA process itself, which is undoubtedly important as the process is very time-consuming and requires considerable commitment from all involved. However, as one PSP Lead commented, the work doesn't stop when the priorities have been identified:

> *In many ways this is when the work really starts, because there's no point in identifying your Top 10, if you don't have a plan of what you're going to do with them afterwards. We underestimated the work that's required if you want to really make the best use of the results.* **PSP Lead**

It seems that less consideration is given to what happens at the end of the JLA process, beyond sharing and publicising the prioritised topics. The JLA Secretariat's responsibilities appear to end at the point of identifying the Top 10, but given that planning for the end needs to be included at the start, some suggested that JLA Secretariat, via the JLA Advisers could do more to help PSPs plan for the work that follows, specifically to ensure it is adequately resourced.

We had help with resource planning for the PSP, but then the PSP Lead is left to their own devices at the end… but that's a planning issue as well and that's something the JLA could highlight… maybe it would need at least another six months of a part-time post… to monitor what's going on, or to develop ideas of what to do with the priorities, etc… **PSP Lead**

Key lessons

It is important to plan for the end of the PSP at the beginning of the project in order to:

- Consider all the potential outputs from a PSP and prioritise where there is capacity, enthusiasm and resources for this work
- Consider what role Steering Group members or other participants in the JLA process can contribute to ongoing work and recruit additional people with the required skills and experience
- Ensure commitment amongst funders and other relevant stakeholders (or at least buy-in) to respond to the outputs in a multiple of ways
- Clarify who owns the outputs, who will make decisions about how they are used and who will be accountable for what happens next
- Prepare for responding in ways other than funding research e.g. signposting people to information, informing policy campaigns or health service development
- Maintain the JLA ethos and values beyond the PSP and ensure all future work continues to involve patients, carers and clinicians

4.2 Sharing: disseminating the JLA PSP priorities

Agreeing a communications plan for the end of a PSP is strongly recommended in the JLA Guidebook. Many of the PSPs that were interviewed had developed a comprehensive communications plan to raise awareness of their Top 10 research priorities and often the long list of questions that had been considered at their final workshop. Often all the partner organisations involved in a PSP helped with this dissemination. Large organisations leading PSPs also had the help of their Communications Teams to provide expert advice and resource to do this.

> *Our priorities belong to everybody and they're out there in the public domain… for any funding body who is interested, we had a wide-ranging communication plan to try and make sure that people were aware of these really far and wide.* **PSP Lead**

Common ways to do this included:

- Producing a short summary of the Top 10 or the long-list of 20+ questions – sent to all relevant funders including the Medical Research Council, The Wellcome Trust, the NIHR, NETSCC and Central Commissioning Facility (NIHR CCF), The Economic and Social Research Council (ESRC), soon after the end of the process, embargoed until an official launch
- Holding a launch event e.g. at a scientific conference or in Parliament
- Producing a plain English report for patients and carers, varying in breadth, some just reporting on the process and outcome, some providing context to the prioritised topics
- Writing an academic article for a well-known journal in the relevant field
- Social media campaigns e.g. releasing one priority topic per day over ten days, sending the Top 10 to Linked In professional networks, writing blogs both professional and personal
- Hosting information about the PSP on a PSP-specific webpage and the websites of all partners in the process
- Direct emails to networks and contacts
- Producing press releases for national media
- Giving talks to patient groups, at charity events, scientific conferences and workshops
- In research funding organisations, alerting all current and past grant holders to the priorities
- Using all channels available to PSP partners – magazines and newsletters to publicise the Top 10 and provide links to other reports and articles
- Contacting all the people who had expressed an interest and provided their email address when they took part in one of the surveys as part of the JLA process

The lessons learnt from different PSPs' experiences of these dissemination activities relate to the following issues:

- Producing journal articles and presenting at scientific conferences
- Obtaining news coverage
- Involving people as communicators
- Communicating how PSPs work

- Reflections on which approaches are most effective

These will be discussed in turn.

Producing journal articles and presenting at scientific conferences

Publishing an article in a topic specific journal has proved to be an effective way to reach the researcher audience (see below). This seems to have been straightforward for some of the earlier PSPs, who successfully published in high profile journals, perhaps because of the novelty of the process. However, a more recent PSP reported some difficulty. Their article was rejected because it was not considered to be novel, and in the reviewers' opinion had not identified any new research questions. Another PSP, whose manuscript had been rejected, questioned whether the peer reviewers had sufficient understanding of the JLA process and recognised the significance of the outcome. They took an alternative approach to getting the findings published by writing letters to a journal instead:

> *They refused publication and we thought, well these people here, they're not ready for us yet… But we did manage to get some letters in the BMJ. We sent in a few quick responses to letters and comments on articles in other journals. We had to respond to what had been said, and craft something around that, rather than saying, we've come up with the Top 10 priorities.* **PSP Lead**

Another challenge in producing a journal article is having access to the resource and skills to write it. A clinical member of the PSP Steering Group may do this, but not all PSPs have such willing clinicians involved, for example in the Lyme Disease PSP (2012). Furthermore, clinicians may not consider it worth investing their time in writing an article, as it's not clear they will always gain professionally from the publication. A clinician who led a PSP explained that their work on the PSP would not be valued by their research institution:

> *When you're trying to justify your salary and are asked, 'What are your metrics and what have you published?' - none of this counts. This all goes into the little PPI section… I would say some of this is as important as a good science publication, but it doesn't have that value in the wider system.* **PSP Lead**

Some clinicians and some journals seem to believe that the JLA process is not appropriate for publication in journals that publish scientific research:

> *I don't think any journal's going to be interested in our Top 10. I don't think [the top journal in this field] would give it a second look. It doesn't seem scientific, there's no hypothesis. There isn't really a research question… it's more social science isn't it?* **PSP Lead**

Similarly, some PSP Leads have experienced difficulty in getting an abstract about their PSP accepted at a scientific conference, again suggesting that the values informing decisions about publishing research findings might not fit with the values underpinning the JLA process.

I put in an abstract to [a USA conference] and got completely rejected and then I put in an abstract to [a British conference] and I was gobsmacked that it only went in as an E-poster. First of all, I'm really sorry but this should be receiving oral presentations, because researchers and clinicians need to talk about this… so it's been very poorly received at conferences… it doesn't seem that interesting to them. **PSP Lead**

Such experiences caused some PSP Leads to question whether the JLA process has wider recognition across the national health research infrastructure i.e. whether there is consensus across the system as to the significance of the priorities. They concluded that more could be done to promote the JLA to other parts of the research system, including journal editors.

Obtaining news coverage

One of the PSP Leads reported having made considerable effort to get their priorities covered in the national media, but without success. They concluded that culture of the science media values reporting 'scientific breakthroughs', rather than explaining the research process and how the agenda is set.

> *We really tried hard to get a media centre to launch our report, but they wouldn't, because it wasn't newsworthy. We thought we must have a news story here, because young people's mental health is so topical, but they said, 'No, you haven't got anything new to say.' The feedback was that they're not so interested in questions, they're interested in results… We also sent a press release to lots of places, but we didn't get any stories anywhere. Nothing, not even a tiny mention. People don't seem to really understand the value of asking the right questions, but if you don't ask the right question, the chances are you won't get a good answer either. I don't think that's appreciated.* **PSP Lead**

However, this has not been universally the case. Other PSPs have received mainstream media coverage. A key factor has been making links with reporters who have personal experience or a direct interest in the topic of the PSP.

Involving people as communicators

Many of the PSPs had involved patients, carers and clinicians in their dissemination of the priorities, recognising that their involvement in this phase can have a major impact, giving the messages greater power, credibility and authenticity. It is also in keeping with the ethos and principles of the JLA. One researcher who was interviewed reported having heard about the PSP in their area of research from a patient, long before seeing the related journal article.

'Word of mouth' is always one of the most effective communication tools and this also proved important in spreading the message amongst researcher and patient/ carer communities. When the pool of researchers working on the same topic is small and well-connected, information about the JLA PSP priorities spreads quicker and they seem to be more rapidly adopted (see Case Study 20).

Communicating how PSPs work

Although some PSP Leads reported finding it challenging to explain the concept of PSPs to clinicians and researchers, the feedback from the interviewees was that this background information is very important. Understanding the robustness of the approach and the fact that large numbers of people have been consulted gives more credibility to the outcomes and gains researchers' trust:

> *Sometimes when I talk to people, I don't think they realise that the process has been quite long, very involved, and involved hundreds of people in different ways. Most researchers, when they talk about working with patients it's only with a handful of people, so PSPs may need to do more to highlight this isn't a quick survey, but quite a well thought out process and,*

that those top 10 are quite precious and many people have made huge efforts to get them. **Researcher**

I first heard about the PSP at a conference. It had just been published and they were presenting the findings. And I just went, 'Oh this is brilliant'. And so I went searching for it, and I had a look at the website and had a nose round there just to find out how was it done and then thought, 'OK, this is solid'. **Researcher**

Reflections on which approaches are most effective

A couple of the PSP Leads who had invested considerable time and resource in writing a lengthy plain English report on the findings, questioned whether this effort had been worthwhile, as they were uncertain about the impact. For example, the Palliative and end of life care PSP (2015) chose to produce an extra report that summarised all the questions raised in the first survey, including those that were more philosophical. It was hoped that these additional questions might be addressed by social scientists. However, it is unclear whether such reports have an impact:

We were very lucky, we had co-funders willing to fund an extra researcher for a couple of months to write the second report. Sometimes it is quoted mainly to say that there's one particular theme which wasn't included in the outcomes of the PSP… and we take it with us whenever we go anywhere. But I'm not sure how well it is known actually, and whether we could have done anything else to promote it.
PSP Lead

It's this thing about charities constantly churning out PDF reports and you need to make sure you're asking yourself 'What is the point, right? Like who is reading this?'
PSP Lead

Another PSP Lead made a similar comment about whether writing a long report had been worthwhile, and whether the time and effort would have been better invested in writing a journal article. Other Leads who hadn't written a journal article shared this view, as with hindsight they believed this to be the most effective way to reach the researchers.

I spent a lot of time writing the report and now I think a lot of the content wasn't that relevant. I would still write some form of report, but shorter… I'd have rather invested that time in doing a research paper. It might have added something with a little more velocity to academics. It might have reached a few more people.
PSP Lead

This view was endorsed by a researcher who had developed a research project in response to a PSP priority topic. She reported having found out about the PSP via a journal article and thought this an important communication route. Other interviewees emphasised that a peer-reviewed article was also more likely to give the findings credibility and weight:

People in academia will be trawling through these journals and have alerts coming out from the journals, so new findings probably more easily pop up that way than through some other routes…
Researcher

Because we didn't have a journal article, all we could do was link to an article on our website… a clinician, if they see that the link is to a patient website, they're not going to take it so seriously, but if we had had a paper, that would have been an anchor, more trustworthy from their point of view. ***PSP Lead***

Key lessons

- It is important for PSPs' dissemination plans to reflect their strategic goals, which may go beyond funding research
- Plain English reports are valuable for patient and carer audiences – these can be short
- Producing a journal article seems to be the most effective way to reach researchers, but there may be cultural barriers to publishing the outputs of the JLA PSPs
- Greater strategic use could be made of patients, carers, clinicians and researchers in promoting the JLA PSP priorities – using word of mouth – they can also usefully promote publications
- The JLA process and its significance for the research agenda needs to be promoted amongst the wider clinical research community, to overcome the barriers to publication/ presentation at scientific conferences
- Greater public awareness of the JLA is needed to overcome cultural barriers to reporting the JLA PSP priorities in non-scientific publications and the media

4.3 Influencing: persuading others to respond to the research priorities

PSP Leads often aim to influence three different categories of stakeholder who could potentially respond to JLA PSP priorities, namely:

(a) Funders

(b) Researchers

(c) Fundraisers

The experience of working with each of these and the lessons learned will be discussed in turn.

(a) Funders

Some PSPs are led by organisations that fund research or have authority to decide how to allocate their own research funds, for example NIHR Biomedical Research Centres, (see Section 4.4). PSPs that lack this capacity instead aim to influence other funding bodies to respond. The general assumption seems to be that other funders will want to do this, or should do this if they recognise the significance of the JLA findings, but it has proved to be a significant challenge.

We note there may be parallels here with the use of research evidence to improve healthcare practice. Even when the best quality evidence suggests life-saving and cost-saving benefits, organisations like the NHS find it difficult to make the necessary changes. Implementation science recognises that context, attitudes and resources all influence whether new evidence will lead to change and that additional translational work is needed. In a similar way, expecting large public sector funders of clinical research to respond to this new way of identifying research priorities may need a greater amount of intervention/ support to bring about a different kind of response. As some of the interviewees commented:

> *Communicating the Top 10 might not be enough. I think you could do a PSP and find it's ignored.* **PSP Lead**

> *We just assumed that people would pick them up and run with them, but actually you need to really push it and really do things to make it work.* **PSP Lead**

The lessons that the PSPs learned from aiming to persuade other funders to respond to their priorities included the following:

- The value of engaging funders in the JLA process
- Recognising funders have their own remit and strategy
- Collaborating with other funders to address common priorities
- The skills and resources needed for influencing

The value of engaging funders in the JLA process

In an attempt to create a sense of ownership or a certain amount of buy-in into the JLA process, some PSPs have included funders (except industry funders) as project partners, rather than as members of the Steering Group. These funders have provided resources in-kind (e.g. meeting rooms) and sometimes attended the final workshop. However, the representative of an organisation that is involved may or may not be the person who is ultimately making decisions about research strategy or

portfolios, and maybe unable to influence that level of decision-making, as one PSP Lead commented:

> *We've been having conversations with [a funder] and I was talking to one of their senior staff and when I mentioned the Top 10 they said, 'I've never seen those.' And I have sent them to that person twice. It's not on their radar… I'm not blaming them they are busy, but you can send emails, you can invite people to workshops, you can talk to all their colleagues, but at the end of the day the PSP may be more important to you than anyone else.*
>
> **PSP Lead**

From the perspective of a manager in a funding organisation, being part of the process, for example, by attending the final workshop, may be less useful than working with the PSPs after the Top 10 has been published. At this later stage funders value getting input from the experts on the Steering Group to make sense of the outputs and translate priorities into research questions and projects (see Section 4.5).

> *It's helpful for my staff to go once or twice to see the JLA process and really understand how it works. I'm keen for them to do that because I want them to see how much time and effort, blood, sweat and tears goes into this process and just how important the priorities are. But after that, with our limited resources, our time is better spent in the engagement work with the PSP Leads we partner with, to unpick the priorities and the thinking behind them.*
>
> **Manager in funding organisation**

Recognising funders have their own remit and strategy

Based on their experience to date, some PSP Leads have concluded that large research funding bodies may only respond to JLA PSP priorities, if those priorities fit within their existing research remit or overall strategy:

> *At a [funder] meeting, a senior manager told me that, "We don't have to take any notice of these PSPs. Lots of patients assume that we are going to fund them, but we won't fund them unless we really feel that they meet our priorities and that they're sensible." It was a very important lesson that the funders may not necessarily listen… if they like your PSP and some of the themes that come out of it fit with their current strategy and priorities, you might be lucky and they fund it.* **PSP Lead**

A manager from a funding organisation explained they did not perceive it to be their sole responsibility to respond to JLA PSP priorities, since they see themselves as accountable to a wider range of stakeholders:

> *We don't view the JLA process as specifically there to identify priorities for our research. It is one of a number of sources that we look at, and often a research call that comes out will have evolved through the inclusion of questions from multiple different sources, not just the JLA… and all sorts of information including reviewers' and committee members' comments will shape the final commissioned call. Our process has the best intentions of maximising the benefits for patients and the NHS… so we wouldn't necessarily look at our call and assess how well it sits with the JLA's priorities, because essentially that's not what we're here for.*
>
> **Manager in funding organisation**

Furthermore, from their perspective, not all JLA PSP priorities will fit within their specific remit or their area of expertise. They will be unable to consider any questions that cannot be answered by the type of research their funding organisation supports:

> *Sometimes we might not be able to take something forward because a project would be enormous and hugely expensive, or maybe the topic is not something that is best answered by a controlled trial… recently I spoke to a PSP Lead who very helpfully brought us the priorities with an idea of which ones would be good for us to look at, and which ones would make a perfect PhD project, for example… Sometimes it's a question of who is the best funder of this research… and often that's not us*.
>
> **Manager in funding organisation**

Older PSPs that were carried out when the JLA process focused solely on treatment uncertainties appeared to have more success in persuading a funder, in this case the NIHR, to address their priorities. This is likely due to the fact that the approach at that time was tailored to generate questions that could be answered by a clinical trial or a systematic review:

> *By chance we were one of the PSPs that were ongoing when the NIHR adopted the JLA… and it brought our PSP to their attention...and that was influential in getting the NIHR to engage with us…we'd had contact with those people, so there was a rapport… we were very focused on treatments, and the way we framed the priorities using PICO formatted questions leant them to being instantly fundable.*
> **PSP Lead**

Collaborating with other funders to address common priorities

Even when a PSP does have funders who take ownership of the priorities (See Section 4.4), there may still be topics that are difficult for them to address for a variety of reasons. For example, with condition-specific PSPs that are led by a charity, there may be priorities that aren't relevant to their organisation's purpose and mission, or topics that they don't view as being their concern. This might lead to some issues not being picked up by any of the partners on a PSP. One PSP Lead argued for collaborations across PSPs to form to address such common issues (See Case Study 8).

> *Incontinence was a topic that came up in our PSP, but one of the reasons it hasn't been taken forward is that none of the charities have real ownership of this issue, because it relates to a lot of different types of disease. it can end up that no one feels it is specific enough to their patient community… having some comparison of all the JLAs, doing some analysis to identify common issues would be fascinating and would help to identify topics that fall between the cracks.* **PSP Lead**

Where UK-based charities have been unable to respond to JLA PSP priorities because of limited research funds, they have been able to use their international networks to persuade much larger patient organisations to fund relevant research:

We work internationally and we managed to influence the US charities in our field. They fund much more research than we do, like hundreds of millions of dollars of research, and we worked with them to show that mental health is a clear priority for our members. I think that definitely influenced them… **PSP Lead**

The skills and resources needed for influencing

Many of the PSP Leads concluded that there is a considerable amount of work to do at the end of a PSP to keep the momentum going and to continually raise awareness of the priorities with the aim of influencing others.

It needs at least two years of somebody feeling like it's their baby at the end of the PSP, to be pushing it out, going to conferences and talking about it, going to patient events and talking about it, making sure that the JLA website is up to date… it comes down to the continuity for somebody who has very much bought into it throughout the process… if you don't have that, then it tends to just sit on a pile and everybody forgets about it. **PSP Lead**

One of the PSP Leads commented that the skills required to plan and run a PSP may be different to the skills and experience required to influence other stakeholders at the end. This may mean that a different set of people to those on the Steering Group are needed to take forward the influencing work. This is another issue that may need to be considered and planned for at the start (Section 4.1).

We're like anybody else with their PSP and their priority lists. You're knocking on the door of very, very powerful institutions and you're trying to work out a way through them. I think we are doing quite well but it's early days… we're quite experienced. Happenstance and other forces might also act in our favour, but a whole set of different skills come into play. **PSP Lead**

PSP Leads who work at a senior level in a charity, seem to have the right combination of skills, experience and networks to create positive pressure. They report being able to use their power and influence with researchers to encourage a response to priority topics:

I lobbied a number of people quite hard to say 'Look at this question. There's been nothing in this area for a while and we've got this funding around this, I'd like to see some projects looking at this question.'
PSP Lead

However, currently it seems that the follow-up work after a JLA PSP relies on individuals having sufficient motivation and enthusiasm, rather than there being a strategic plan with allocated roles and responsibilities. Time and resources are an issue for every PSP, simply to complete the process and disseminate the outputs. A common experience is that it *'all took a lot longer than expected'* and Steering Groups may feel exhausted by the time they *'get their PSP over the finishing line'*. This may be an argument for including the ongoing work as part of the initial PSP budget, and for involving organisations in the JLA process who have capacity to absorb this additional influencing role at a later stage (wherever they exist).

After the final workshop, it lost a lot of momentum because I wasn't there to keep everybody together and keep driving it on… I have to proactively keep on top of everything for it to continue, to keep searching for new impacts and to promote

the priorities as well… it could easily fall off the agenda for a lot of people… **PSP Lead**

The assumptions that people were going to carry on doing things after the PSP were just unrealistic really. Unless it was being done by an organisation that had the resources to do that, like a specific charity, and they used it to drive forward their own strategy, whereby the staff are all funded to do this as their day job. But in the context that we did ours, we flew by the seat of our pants, proverbially speaking.
PSP Lead

Being clear about who is responsible for this influencing work becomes particularly important if there is no clear owner of the PSP outcomes (See Case Study 3).

Case Study 3: Cellulitis PSP (2017)

Cellulitis is experienced by people with a range of conditions. There is no patient charity specifically for cellulitis, and therefore no sense of anyone particularly owning the disease. This is why there has been little research on this topic, which was one of the motivations for doing the PSP in the first place. However, this also made it challenging to know how and where to disseminate the priorities. As Kim Thomas, Professor of Applied Dermatology Research, University of Nottingham explains, *"It highlights the importance of having someone or an organisation with a sense of responsibility to influence and persuade. A whole catalogue of things meant that our Cellulitis priorities didn't get pushed out as quite as actively as they could have done. It makes me wonder how proactive I need to be, and concerned that I need to do more with the funders. The Cellulitis PSP has not been as successful as some of the others I have worked on, because it didn't have a critical mass of people willing to keep it up in the air."*

(b) Researchers

The lessons learnt around persuading researchers to undertake research to address JLA PSP priorities relate to the following issues:

- Avoiding conflicts of interest when research institutions lead a PSP
- How the priorities influence researchers
- Which researchers are most responsive

These will now be discussed in turn.

Avoiding conflicts of interest when research institutions lead a PSP

When an NIHR-funded research centre leads on a PSP, they may have an opportunity to use the priorities to decide how to allocate their existing research funds, in much the same way as that a charity may choose to allocate its research budget (Section 4.4). If the researchers in that institution need to apply for funding, they can make use of the JLA PSP priorities to support their

applications. Some interviewees suggested this needed careful handling, to avoid any conflict of interest or a perception that researchers might be using the PSP to further their own interests. To this end, an academic PSP Lead was clear that their institution did not claim exclusive ownership of the priorities, stating that:

> *Our feeling has always been that this is something that we do as a service for everybody, so they're not owned by anyone…* **PSP Lead**

However, this raises the question as to who then has responsibility for promoting the priorities and doing the work of influencing others. Another PSP Lead from a charity believed this is where partnerships between different kinds of organisation prove extremely successful:

> *In terms of lobbying others to respond to the priorities, it does help if there's an organisation that doesn't have any skin in the game to do some of the pulling the strings behind the scenes… there's some real advantage in having the researchers leading it, because then you know they're going to make an application to do some of the research on the back of it… Our partnership with a University worked really well, because they were able to get what they needed out of it and we were able to do the lobbying side of it with our networks*. **PSP Lead**

How the priorities influence researchers

The researchers who were interviewed explained that the JLA PSP priorities had mostly given them confidence to apply for funding for research projects they were already planning to do. The process had not suggested new topics for researchers to consider. Nor was there evidence that researchers had been persuaded to stop pursuing a particular area of interest because it wasn't a JLA PSP priority.

> *X Charity have signposted this topic as its number one interest. I was working on that anyway, but what it's done is reinforced me to do further work on that, in partnership with the Charity… it's meant that they have confidence that we're in tune with them and the community… it gave us absolute authority, rationale and reinforcement that this was the area to research.* **Researcher**

> *I read it [the journal article about the priorities] at a time when I was putting together some fellowship applications. And I was very pleased to see it, because it supported what I was wanting to do already… It was great for including in these sections of the grant and fellowship applications about what is the impact of your research going to be, and what's the need for this research.* **Researcher**

> *There have been some commissioned calls from funders and I was particularly pleased when I saw that priority number two had been funded… It was like the system worked… Albeit it might have just suited those researchers, because that was the research they were going to do anyway, but here was a way to sell it even harder*. **PSP Lead**

Importantly, one researcher explained how knowing that she was responding to a JLA PSP priority gave her the motivation and determination to go through the funding application process:

When you're writing a grant and putting that it's the number one priority, it helps. It just drove me on a little bit, because this is important and this area doesn't get funded particularly well… I actually always had the Top 10 printed out beside me as I was writing the grant, just going, "No this is worth the effort, it's worth the time. This is worth it." **Researcher**

Which researchers are most responsive?

Based on their experience to date, some PSP Leads recommended targeting junior researchers applying for fellowships or PhD funding, as this group seemed most open to the principle of researching what patients, carers and clinicians believe important. Others reported success in influencing the international research community. This proved particularly valuable for the Lyme Disease PSP (2012), where there weren't any UK researchers working on their topic (see Section 4.4).

No UK researchers have indicated an interest which is understandable because no one has ever identified these uncertainties before, so how could the researchers engage?... We've talked quite extensively to people in Norway and in The Netherlands. One researcher said, "Well we don't need to do this now, you've done it so we'll take this as the Top 10 priorities." They have since initiated some research projects, in fact only the beginning of this year they asked me to join an EU funded project. **PSP Lead**

(c) Fundraisers

Some of the PSPs led by charities reported that the JLA process had helped with fundraising for their research amongst their supporters. It had helped to make the case for the value of specific research and has been used to convince their funders that the research is worthy of their support:

I would credit the JLA process and the strategic approach we've taken with helping us with our ten year fund raising programme… one of the things I did shortly after the PSP was to work with our fundraising team to write a case for support, which now has the JLA PSP priorities embedded throughout. So we've got research plans against our fundraising targets for each of the main JLA PSP priority areas. And this helped to build the internal case for raising more money for research as well. **PSP Lead**

If you can show with clarity that you're representing the priorities of your people, and you're authentic about that, that is enormously powerful and it gives you enormous confidence in terms of going out there and speaking about what we do, especially with donors… the JLA is massively impactful in that way. **PSP Lead**

One of the patients who had been involved in a PSP suggested the priorities could provide a focus for fundraising by the patient and carer community, which could prove very effective based on lessons learnt from other contexts:

If you ask Tate Museum members to fundraise generally for the Tate, people tend to do it… but when you ask people to fundraise for a particular painting or a particular educational initiative, you tend to get a better response. Even better, if you ask people what they want funded and then give them an opportunity to contribute to that, then they are even more engaged… There's really interesting and exciting opportunities to do more of that with patients and JLA PSP priorities… if the patient community could do more to fundraise… then there would be a bigger grant available… because it's about patients shaping the direction of research and patients saying, 'This is what needs to be answered and we're going to help fund that.' **Patient**

Key Lessons

- Involving funders in the JLA process may help build relationships but the greatest influence comes from working with funders after the PSP to define research questions
- Not all funders believe working with the JLA PSP priorities is their responsibility – PSPs therefore need to be prepared to 'make the case' for them – knowing how funders choose their own priorities may help
- The work of influencing requires specific skills and experience and needs to be properly planned and resourced – senior leaders in organisations are well-placed to do this
- Partnerships between charities and research institutions seem to provide an effective combination of skills and networks for influencing others
- Researchers are motivated by the JLA PSP priorities, but it is not clear if this is shifting the direction of their research – researchers may need to expand on how they have been influenced by the JLA PSP priorities, rather than simply stating this is the case
- Supporters of research are persuaded to fundraise for JLA PSP priorities – patient and carer communities could do more of this if they feel attached to the priorities they have identified – this potential for fundraising needs further exploration

4.4 Responding: using JLA PSP priorities to influence strategy and funding decisions

When PSPs are led by organisations that fund research or have control over how research funds are spent, they can choose to respond to the priorities in the following ways:

(a) Integrating JLA PSP priorities into an organisational research strategy

(b) Directly funding research projects/ programmes that address JLA PSP priorities

(c) Putting out themed calls for proposals addressing JLA PSP priorities

(d) Integrating JLA PSP priorities into open calls for funding applications

(e) Judging whether research proposals genuinely reflect a JLA PSP priority

The lessons learnt in relation to each of these issues will be discussed in turn.

(a) Integrating JLA PSP priorities into an organisational research strategy

Where a PSP has been led by a charity, a number have been able to incorporate the JLA PSP priorities into their research strategy. The MS Society was one of the organisations that took this approach and found that it helped them to decide what to let go of, as much as clarifying where to focus their efforts:

> *We've made our research strategy focus on the top few priorities where we think we can make the most difference - the fastest. It's given us justification to prioritise and rationalise with the research community because people with MS have told us what they want us to do. What was also important was making sure our Board of Trustees knew about those ten priorities, because now we've got a Board that understands and wants us to report against them*. **PSP Lead**

(b) Directly funding research projects/ programmes that address JLA PSP priorities

PSPs led by organisations that fund research have simply decided to fund projects that address a JLA PSP priority. A number have reported doing this with partner organisations, both in the UK and internationally, to promote efficiency and avoid duplication. The JLA process itself has often been a catalyst for such partnerships, by helping to foster trusting relationships between organisations that have continued well beyond the prioritisation exercise (See Case Study 4). Pooling resources with other organisations through such partnerships has been essential to fund research that otherwise wouldn't go ahead (See Case Study 5).

Case Study 4: Sight Loss and Vision PSP (2013)

The Sight Loss and Vision PSP produced twelve top-ten lists of research priorities for a range of eye diseases and conditions. One of the lists related to Age-related Macular Degeneration (AMD), the most common cause of sight loss in the UK. The top priority in the AMD list was finding an early stage intervention to stop people losing their sight.

Delivering the broad PSP led to over 40 charities and other organisations collaborating for the very first time about researching sight loss. This collaboration throughout the project was one of the important factors that helped to establish a new partnership of four charities, with the sole purpose of addressing the number one AMD research priority. Michele Acton, who was CEO of Fight for Sight at the time, explained, *"The PSP was incredibly useful not just for Fight for Sight, but also as a great coalescing piece of evidence. Identifying the AMD priority for patients gave us a shared goal we could all get behind, based on a strong sense that we could achieve so much more for this group of patients if we all worked together. We are four charities with very different overall missions, but if you draw the Venn diagram, the overlap in the middle is around that early stage intervention for AMD."* The collaboration has since funded a small team, based in Fight for Sight's offices, which is taking forward the work on this priority.

Case Study 5: Multiple Sclerosis (MS) PSP (2013)

The MS Society shared their JLA PSP priorities worldwide with the aim of persuading other MS Charities internationally to address the top issues. This has led to the Society funding, in collaboration with the NIHR and the National MS Society in the US, a phase three clinical trial of high dose simvastatin for people with progressive MS. Susan Kohlhaas, Executive Director of Research at The MS Society reported *"It's costing around five million pounds in total and we've put in just over a million. We worked really hard to broker that partnership between the NIHR, the National MS Society and us, because we knew it was a topic that sat squarely in our top priority list. Justifying that amount of investment would have been really difficult had it not been a top priority. So, that's one success that the JLA process has achieved for us."*

Charities have the additional option of directly funding development projects, rather than academic research (Case Study 6). Such projects may be undertaken by other parts of their organisation, rather than their research departments.

Case Study 6: Autism PSP (2016)

The number eight topic in the Autism PSP Top 10 priorities was a question about employment. James Cusack, Director of Science at Autistica, explained how the organisation decided to respond to this priority, "*We had already set up an initiative around employment where we worked with corporate partners on internships for autistic people. We started with Deutsche Bank, who bravely allowed us to research an internship scheme we ran with them with another autism charity. Since the JLA PSP, we've been rolling that out to other companies and collecting more data on the outcomes, because there's a trend towards people doing these internship schemes as a bit of a PR exercise. So we're using this project as a way to work with corporate partners and to ensure that we're addressing this priority topic as well.*"

(c) Putting out themed calls for proposals addressing JLA PSP priorities

Another option for funders who lead a PSP is to put out a themed call for proposals. This asks researchers to put forward any proposal that fits within a broad topic. For example, after the Palliative and end of life care PSP (2015) had finished, the charity Marie Curie undertook a grant mapping exercise which helped them to prioritise amongst the many research themes that emerged from their process (see Case Study 7). This shaped subsequent themed calls for research proposals to address the topics that were found to have had the least funding.

Case Study 7: Palliative and end of life care PSP (2015)

After their PSP, Marie Curie undertook an in-depth mapping exercise to find out what research was ongoing in the UK that related to any of the 83 questions that had been identified through their JLA process. Sabine Best, Head of Research at Marie Curie, described their approach, *"It was a big piece of work, which involved searching through the UK Clinical Research Collaboration's Health Research Analysis database of research grants (2014). This contains information from all the big funders and the Association of Medical Research Charities, to include three billion pounds' worth of health related research activity in the UK that year. We identified keywords relating to each of the questions and then searched the database to identify projects that were underway."*

A member of staff worked part-time for approximately six months to deliver this project, representing a considerable investment for the charity. Sabine reported that it did help them to decide which topics to pursue, *"We had some key findings from that exercise. For example, we identified 15 questions that had no or very, very little funding, such as four questions about bereavement that had no funding at all. We also looked at the Top 10 in more detail and found that our top priority, how to provide palliative care outside of working hours, was the second lowest funded of the Top 10. Training for carers and families was the least funded of the Top 10, and the one that was most addressed was one around non-cancer diseases. From that we identified priority areas for our themed call to include the topics from the Top 10 that had not been very well addressed."*

The exercise had wider impacts as it was referenced a few years later when the NIHR also put out a themed call for research on end of life care.

Marie Curie (2014) Does current palliative and end of life care research match the priorities of patients, carers and clinicians? A grant mapping analysis of the UK Clinical Research Collaboration's Health Research Classification System dataset 2014.

Some funders reported advantages to working together to put out joint themed calls, either because there is considerable overlap in their JLA PSP priorities (see Case Study 8), or because their research budget is not sufficient to cover certain topics and requires support from other partners. For example, a number of the PSP Leads interviewed, were exploring the possibility of putting out a joint call with the NIHR.

Case Study 8: A collaboration across several PSPs to address the topic of incontinence

A group of PSP Leads met informally in 2015-2016 to share experiences and ideas for follow-up work after a PSP, led by staff from the Parkinson's UK PSP (2014). They considered what their PSPs had in common and realised that many of their patients had similar symptoms, including incontinence and depression. As a Depression PSP (2016) had just been carried out, the group decided to explore the topic of incontinence, focusing on what they could do collaboratively. They concluded it would be helpful to hold a workshop with researchers, patients, carers and clinicians to scope out the topic in more depth, as Sabine Best from Marie Curie reported, "*We wanted to get more of a handle on what was behind those questions and to identify which topics needed more work.*"

They held a one-day workshop with patients and carers, recruited via the various charities involved, as well as researchers and clinicians with expertise in incontinence. This helped to identify where there were mismatches between what was being researched and what questions patients and carers wanted research to answer. The charities involved all agreed to include the prioritised incontinence research questions in their subsequent calls for proposals.

While this project helped to ensure the topic did get research funding, the process was not without its challenges. As Sabine explained, "*The main issue was lack of time and resource*." The charity staff who worked on this project did so as an extra in their already full-time jobs, and none had dedicated resources for it. Sabine concluded this would be helpful for such an approach to maximise its impact in future.

"*My bladder and bowel own my life." A collaborative workshop addressing the need for continence research* (2018).

Developing research capacity where it doesn't yet exist

The biggest challenge of taking an approach based on a themed call is that the skills, capacity or willingness to address to the topic may not yet exist in the research community. The applications received in response to a call may then be limited in number and/ or of poor quality. Where this has been a perceived risk, some PSPs have taken additional steps to bring researchers together to carry out foundation work to develop new areas of research (see Case Study 9).

Case Study 9: Multiple Sclerosis PSP (2013)

To address one of their Top 10 priorities, a question about the most effective early treatment for people with MS, the MS Society first set up a working group of researchers, as Susan Kohlhaas, Executive Director of Research at The MS Society, explained, "*We had one researcher who was interested in the topic but he wanted help from other people, with a broader range of expertise.*" The working group suggested carrying out an initial review of the literature which the Society agreed to fund. This study suggested differences in outcomes for people with MS receiving different types of early treatment. The working group then concluded that a clinical trial would be the best way to find out which treatment was best, and worked together to design one. Their subsequent funding application in the UK was unsuccessful, but they were able to adapt it and use it as the basis for a proposal for an international trial, working with researchers in the US. This time they were successful and the American funding organisation PCORI (The Patient-Centered Outcomes Research Institute), finally agreed to fund the trial at a cost of around ten million dollars.

Where capacity to respond to a research topic does not yet exist, others charities have decided to explore what is stopping the research going forward, and then taken whatever steps are possible to address whatever they perceive to be the shortfall.

> *A clear example is the area of prevention and risk, because the science is not at a place where we really understand what's going on. So, we've done a few small things. We held an international workshop on prevention to ask researchers to identify what we could do that would be affordable and practical for us to do, to move the field forward… They came up with four recommendations which we've started to implement.* **PSP Lead**

> *Continuity of care and how to provide it, it's actually too big a topic… we had some resource in one of our research centres, and a researcher there did a rapid review on the topic. In our next themed call, we plan to provide some detail on this question to help the research community know how to address it.* **PSP Lead**

(d) Integrating JLA PSP priorities into open calls for funding applications

In addition to themed calls, most research funders put out open calls for proposals that allow researchers to put forward research projects on any topic they choose, also known as response-mode funding. Some of the charities that fund research and have led a PSP, have decided to highlight their priorities in these open calls, either by indicating they are interested in a particular topic, or to ask the researchers to state in their application whether their project addresses any of the JLA PSP priorities:

> *One of the areas that came out as really important was around hormones... It's a neglected area, the impact of menopause is never talked about, and we thought we can't have a separate funding call on just that, but what we can do is encourage applications on this topic through a highlight notice which says we are particularly interested in applications on this issue, and that's what we're doing now.* **PSP Lead**

What we did from day one was reference the priorities in our grant application form. Anybody who applies for a grant has to be able to indicate which priority their research was aiming to address. So, any researchers across our community who applies to us for a grant, now knows about the PSP and explain how their research will deliver what patients want... even if it's basic science and treatment is still million miles away, at least they can explain why they believe that their research is the necessary building block to get further down the line. **PSP Lead**

The questions raised by taking this approach and debated by the interviewees included:

- How much weight is given to the JLA PSP priorities when reviewing funding applications?
- How do researchers respond to such requests?
- What are the implications for further patient and public involvement in the research proposals?

These will be discussed in turn.

How much weight is given to the JLA PSP priorities when reviewing funding applications?

None of the charities interviewed for this project had made the decision to *only* fund JLA PSP priorities, as they did not want to risk losing other potentially valuable research:

We don't want an amazing project not to be funded just because it's not one of the Top 10… We're having that discussion between the research committee and our lay members to find which ones should be funded, who can then decide among all the applications they receive, which ones should be a priority for funding. **PSP Lead**

One of the researchers who led on a PSP thought addressing a JLA PSP priority needed to become an explicit criterion for judging the quality of proposals and awarding funding:

I would go so far as to say, this goes to the policy decision… if you've got two grants and one of them with a somewhat higher score didn't fit in the Top 10, and the other one with the lower score did, I think the one that sits in the Top 10 should be funded… I feel as a society we have a responsibility to use the money wisely and therefore fund the people with the highest scores in their grant applications, but I think we need to be prioritising the research that is answering the questions that patients have… otherwise I feel we're tinkering round the edges really… if we all agree that PSPs produce representative data, then there should be more of a requirement for them to be considered in the funding application. **PSP Lead**

However, others were wary of giving too much emphasis to JLA PSP priorities as they were concerned that the process could miss potentially important topics because patients, carers and clinicians may be unaware of what's possible or available:

Our Top 10 was heavily influenced by questions on hearing aids. Hearing aids are the most common management option for hearing loss, so it's obvious that patients and clinicians focus on what they know… As researchers, we're closer to the ground in terms of emerging innovations and we know there are alternatives… but if there are treatments in development that patients aren't aware of, they don't put

those questions forward. So I think the JLA is only one tool in our tool box to make sure that research better reflects what patients and clinicians want and need. **PSP Lead**

Where does basic science fit in with this and how valid is it for basic science questions? Because there will be some basic science questions that were never going to be brought up in a PSP which could be key to scientific and medical progress… So I don't think the PSPs are everything. **PSP Lead**

None of the funders who were interviewed were clear about how much weight the reviewers reviewing grant applications (which often include a mix of patients, carers and researchers), give to projects that address a JLA PSP priority. The relevance of a research project is often part of the scoring system that reviewers use to evaluate proposals, but how much the JLA PSP priorities influence this scoring has not been explored (see below). Therefore several important questions still need to be answered including: does relevance to a JLA PSP priority influence funding decisions, and if so how?, and since the JLA process has been introduced, has this created an observable change over time in terms of the numbers of projects addressing JLA PSP priorities that are being funded? This project was unable to answer these questions.

How do researchers respond to such requests?

The researchers who were interviewed had often referenced the fact that their project was addressing a JLA PSP priority even when this wasn't an explicit requirement in the funding application. They viewed this as an important validation of their choice of topic (see Section 4.3). The researchers assumed their recognition of JLA PSP priorities would be significant in the final funding decision, but had not received any feedback to know for certain this is the case.

When we are writing grant applications we are now repeatedly saying, "We've asked the patients and this is what they want us to do"… Of course, we don't know if we would've had the grants had we not done this, but one would hope that this is something that helps the grant application…When the funders have been charities, we're assuming they're very receptive to that, because obviously they're very patient focussed. **PSP Lead**

What are the implications for further patient and public involvement in the research proposals?

Some of the researchers commented that if they were responding to a priority topic that had been identified via a JLA process, that meant some of the groundwork had been done, lessening the need for them to involve patient and carers at the prioritisation stage. This is helpful as there often isn't funding available for involvement when applying for a grant. Importantly, they were clear that subsequent stages of their research would still need involvement.

It's really hard to get people talking about a potential project and whether it's a good idea or not, when you may never follow through on it. I hate the idea of not following through when someone else has dedicated a time to it… that's where these priority settings have been really, really helpful because it means that you're not wasting someone's time or getting someone's expectations up too much…

obviously when it then comes to designing the research project, after I have worked up the ideas, I work with a local group to get their feedback on the details…

Researcher

One patient with considerable experience of reviewing grants was concerned that not all researchers understood this and that some researchers conclude that responding to a JLA PSP priority was all the involvement they needed to do. She was also concerned that researchers could use JLA PSP priorities to make themselves look good, so they could be publicly seen to be acting morally, which might sometimes lead to tokenism.

How do we get the message across to everyone that saying you have responded to a JLA PSP does not mean patient involvement, job done. Like a lot in patient involvement, people use it to virtue signal.

Patient

(e) Judging whether research proposals genuinely reflect a JLA PSP priority

Whether a researcher responds to an open or themed call, if they put forward a proposal that aims to address a JLA PSP priority, it raises a question about how to judge whether the project genuinely reflects the original priorities set by the patients, carers and clinicians who took part in the PSP. Some interviewees questioned whether this is always a concern in every context. They suggested that if a topic is under-researched, then patients, carers and clinicians will be glad to see any research go forward in that area, no matter how closely it relates to their original questions.

In a breast cancer PSP for example, the specific formulation of a research question would probably much more important because there is so much research already out there. Whereas, in our field, there isn't very much research at all...so any research will be of benefit. **PSP Lead**

Others raised concerns that researchers could seek to 'game the system', and therefore concluded that this question of how well a project reflects a priority does merit more attention. They believe this becomes more significant when the priority encompasses a broad area of research (see Section 4.5), as it then becomes much easier for a researcher to think, 'How can I fit my ideas under this umbrella topic?', rather than 'What research can I do to address this JLA PSP priority?'

Questions about the process of judging how well a researcher's proposal reflects a JLA PSP priority include:

- Is it sufficient for a researcher to simply state that their research addresses a JLA PSP priority? What supporting information is required?
- Who makes the judgement as to whether the proposed project genuinely reflects the priority issue in the way the patients, carers and clinicians who took part in the JLA process described it?
- When projects are scored as part of the application process, how does addressing a JLA PSP priority impact get scored and how does this influence decisions based on overall scores?
- What are the implications for projects that don't address a priority topic?

While many of the interviewees were aware of these questions, none had yet fully developed policies and processes to address them. In the case of charity funders, the

involvement of patients and carers in the review process was often assumed to provide an adequate check on researchers' claims of relevance to patient/ carer priorities. However, it is unclear what those patient/ carer reviewers know about the JLA PSP priorities and the original questions raised in the process. Therefore questions about whether and how this judgement becomes an explicit part of the review process remain unanswered.

> *Researchers have to answer questions in the application form about whether their applications fit into any of the Top 10 priorities... And we state that if any applications are on the borderline at committee meetings, we will ask our lay members to consider the Top 10 priorities when the funding decision is made… But judging whether a researcher's proposal reflects a priority is difficult, we haven't managed that yet. I don't think there's a clear answer for it…* **PSP Lead**

The challenges of making such an assessment led some interviewees to conclude that the emphasis should be on the stage before, when the researcher is translating a JLA PSP priority into a specific research question. They suggested that high quality patient involvement at this stage would help to ensure the question genuinely reflected the patients' interests and concerns, as opposed to those of the researchers (see Section 4.5).

> *I don't think it's acceptable to basically just ask for proposals and see what the academics come up with… here's our JLA PSP priorities and by all means try and fit your research into them… the JLA is the beginning of an involvement process… So identifying something as a priority is the first step, and it's actually a whole series of steps, and researchers need to involve patients throughout.* **PSP Lead**

Key Lessons

- Integrating JLA PSP priorities into a research strategy helps to decide what research to stop funding as much as where to prioritise funding
- Collaboration with other funders or other PSPs helps with picking up topics that might otherwise fall through the gaps and pooling resources for high-cost research
- Themed calls may need prior foundation work for example to (a) build researchers' capacity to respond, (b) promote collective thinking on an issue, and (c) identify barriers to the research and ways of addressing them
- Asking researchers to respond to JLA PSP priorities may have limited impact on the research agenda without also finding a way to assess whether a priority has been addressed in a way that reflects patient/carer and clinicians' interests and concerns – a process that properly assesses this still needs to be developed
- Some researchers may still need to learn about how the JLA process works and how the extent of the patient and carer involvement gives it weight, at the same time as understanding that simply responding to a JLA PSP priority is not sufficient involvement in research

4.5 Translating: turning a priority topic into a research project

Once a priority topic has been identified, there are three additional steps to develop a research project:

(a) mapping which aspects of the topic remain unanswered by research

(b) identifying and shaping the research question(s) that still need to be answered

(c) designing a project to address the research question

These will be discussed in turn, describing how the PSPs and researchers involved in this evaluation have undertaken these steps.

(a) mapping which aspects of the topic remain unanswered

An essential stage in the JLA process is to review the published literature to ensure that any uncertainty identified by patients, carers and clinicians through an initial survey has not already been answered by research. Any questions that have already been answered are not included in the later stages of prioritisation, as there is no need for further research on the topic (See Section 2.2). However, what this literature review often reveals is that some research has taken place but it is not thought to be of high quality, or that the question may have been partially answered. Furthermore, the PSPs rarely have the skills and resource to complete a systematic review of the literature across all the topics that emerge from the first survey, so will not have identified all the evidence that may exist.

Once it has been decided to work on a particular topic, it is therefore important to scope the research landscape around that topic to be certain about what is known, what is currently being investigated and what still needs to be researched. This is of course true for the development of any research study to avoid duplication and wasted resource. In the case of the broad themes that get prioritised via a JLA PSP, this is also an important step to narrow down the area of potential interest.

This step has been undertaken by staff in a research team (see Case Studies 7, 10 and 11), or by individual researchers developing funding applications. We did not come across examples of where patients and carers had been involved in the review of the evidence or the evaluation of how well their uncertainties had been answered (see Section 7).

Case Study 10: Type 2 Diabetes PSP (2017)

One of the top priority topics in the Type 2 Diabetes PSP was mental wellbeing and diabetes. This topic had been raised as a concern many times through different streams of work in Diabetes UK, including via the Diabetes Clinical Studies Groups. Therefore, as Kamini Shah Head of Research Funding, explained, "*It felt like a natural step that we should do something in this area. So we had a research workshop where we brought together the research experts in the field of diabetes and mental wellbeing from across the world together with people who work outside of diabetes, but work in mental wellbeing. The purpose was to find out what research had already been done and to identify what needs to be done next. A report will be produced and an Advisory Group will look at that to develop a themed call for research proposals*."

(b) Identifying and shaping the research question

The questions that interviewees raised and debated on this issue included:

- Who should be involved in identifying and shaping the research question?
- How could patient and public involvement add value at this stage?
- How could patients and the public be practically involved at this stage?

Who should be involved in identifying and shaping the research question?

The process of developing a research question tends to be understood as a highly technical task that is best undertaken by researchers and others with this expertise. The researchers who took part in this evaluation had worked alone to do this. One of the researchers reported they had plans to involve patients and the public in the practical planning and delivery of their work, but had not had direct involvement in deciding and shaping the research question:

> *I've planned for four patient representatives listed as collaborators on this work and they'll be important at different stages… But I think the main coming up with the idea came from me and more the clinical collaborators… Although I have had quite a lot of conversations with patients over the years, which have influenced it.* **Researcher**

For this reason, it is sometimes assumed that the clinicians involved in a PSP are the people who should take on this task of defining the research questions that come out of the JLA PSP priorities. One of the funders described the value of working with such technical experts from a PSP to do this:

> *Clinicians and anyone with a research background from the PSP, they already have an idea in their heads from their discussions, what research questions are underpinning a particular priority. And rather than us trying to guess what they are and then engage with the community to find out if we're along the right lines, it's very helpful if those questions can be provided to us… those are the kind of discussions that we try to facilitate when we work with steering a members after the PSP, which we frequently do.*
>
> **Manager in funding organisation**

Another PSP Lead described a situation in which a funder hadn't worked with anyone from the PSP, and had developed a poor quality themed call as a result:

> *The funder did end up putting out a call on one of the questions, but they didn't speak to anyone beforehand… and the call was so badly worded that no one could put a research proposal together to answer it. It just went unfunded basically, so it's about making sure some of the key people involved in the PSP speak to the right people in the funding organisations… but only if there is someone who doesn't have a conflict of interest in getting involved in the design of the research*. **PSP Lead**

A concern about only involving the technical experts from a PSP in subsequent conversations with funders, is the risk they will bias decisions in favour of their own interests and concerns, either intentionally or inadvertently. Ensuring that multiple perspectives are included through continued patient and public involvement seems important at this stage, as with all other stages of a PSP, to ensure the priority issues for patients, carers and clinicians continue to have an influence.

How could patient and public involvement add value at this stage?

One of the ways involvement at this stage adds value is by challenging clinicians'/ researchers' assumptions about what is included under the umbrella of broad research topics which are frequently prioritised through the JLA process. Research experts often assume they understand the nature of the questions being raised, based on the way research topics in their field have been framed in the past. For example, one PSP Lead reported that experts kept repeatedly misinterpreting one of their priorities, thinking it to refer to an existing body of work, rather than describing a distinct area of research. Involvement can help to challenge such misunderstandings when they occur (see Section 5).

> *Question eight on our list is about how parenting styles affect treatment outcomes for young people with mental health problems. And it seemed when we were talking to funders that they got the wrong end of the stick about this… A lot of the research that is out there is about how parents who have a mental health problem affect their children. That's not what this question is getting at all… so then you need to have a long conversation to try and explain what it really means.*
>
> **PSP Lead**

With the broad priority topics, it is also possible to interpret them in a way that loses the sense of what the patients, carers and clinicians actually meant when they submitted their uncertainties through the JLA process. Some interviewees were concerned that researchers might accidentally or purposefully misinterpret the questions to reflect their pre-existing ideas, perhaps then minimising the impact of a PSP (see Section 5).

> *The challenge we had was because the questions are so broad, it's quite hard to do any research in our areas, and it not meet one of those Top 10…* **PSP Lead**

> *If you put out a broad call to researchers, you will get back what they are already doing.* **PSP Lead**

Again patient and public involvement in the process of developing the research questions would ensure the questions developed remain true to what was originally asked.

> *We're trying to push the priorities forward and keep them real… with their roots in what concerns young people, parents, teachers all the other stakeholder groups… there's this issue of keeping it authentic to what people were actually asking, which isn't a neat research question…* **PSP Lead**

> *I see researchers referencing the priorities in their grant applications so it seems like they are taking notice of them, but if they haven't followed through with additional conversations with people that are affected by the topic, the researchers could be wander away from the spirit of the discussion at the workshop that actually prioritised the issue.* **Patient**

Reflecting on the nature of the questions asked across a wide range of PSPs, it is apparent that patients and carers tend to frame their questions in terms of how to achieve a particular health outcome, whereas clinical researchers and clinicians tend to frame questions in terms of asking whether an intervention works (see Section 5). This means that clinical researchers working alone to develop research questions from priority topics are likely to frame questions in a different way to patients. This has implications for the type of research that may subsequently be developed. Involvement can ensure that the research questions are framed in such a way that planned project actually delivers outputs that are useful for patients, carers and clinicians (see Section 5).

> *A big difference as a result of the JLA process, is that we're not prioritising the science within the priority areas, but we're prioritising what outcomes people with MS want.* **PSP Lead**

> *It's not just about answering the right question, it's about answering it in the right way.* **PSP Lead**

How could patients and the public be practically involved at this stage?

In some sense, patients and carers will have already had extensive involvement in this stage via the JLA process itself. Each priority question is a summary of a whole host of subtly different questions, asked in different ways by patients, carers and clinicians in the initial JLA survey (See Section 2.2). One way to ensure the research questions that subsequently get developed stay true to what was originally asked is to include an analysis of the original survey responses as part of the development of a research question. This provides an in-depth understanding of the issues people were raising and the nuances in their responses:

> *Often you'll go back to that original survey data and analyse them a little bit more and try to tease things out… within in each of your Top 10 priorities, there's potentially another ten questions. You almost need to do another PSP on those…* **PSP Lead**

However, most interviewees reported that researchers and funders were working with the JLA PSP priority topics at face-value i.e. only looking at the summary questions. One PSP that had offered to make their original survey responses available to researchers for exactly this purpose, had not received any requests for the information.

Another approach, taken by the charities who have led on PSPs, is to support patients, carers and researchers to work together to

interpret the JLA PSP priorities areas and define the research questions. In practical terms this involves running joint workshops (see Case Study 11) or helping researchers find patients and carers to work with:

> *We've been written into a couple of bids recently where we said to the researchers that if they got the funding then we would work with them, so that there's resource for us to do so… and that they can work with our panel of patients and carers to work with on going back to the original questions…because obviously, there's more than one question that fits in to some of these themes.* **PSP Lead**

> *We're working with [a funder] to plan a workshop type event where patients and carers will sit on tables alongside researchers and funders, so that they can have their say… and the idea is to bring people together to encourage them to collaborate and work together to decide what research they want to see on, maybe three of the priority topics…and our patients/ carers are going to help design the workshop.. And then hopefully, the research will be funded on the back of it.* **PSP Lead**

Case Study 11: Multiple Sclerosis PSP (2013)

The Research team staff at the MS Society carried out background research to map out what was going on in each of the priority areas, and drew conclusions about what the next steps were likely to be and where to target future investment. This report was sent out to the MS research community with the aim of engaging researchers in the discussion about how to respond, as Susan Kohlhaas, Executive Director of Research at The MS Society explained, "*Because it's easy to let a report sit on a shelf, to put the priorities in your research strategy and then sit back and wait for the research community respond, but we really wanted to start bringing people together around some of those areas we thought were most important."* The report was shared with the MS Society's Clinical Trials Network, who chose to work on four of the priorities.

The MS Society then brought researchers and people affected by MS together to begin to shape research questions and projects, for example addressing the topic of Self-Management. The priority question and the original submissions from the survey were circulated ahead of a 'round table' discussion, which brought together people with MS, health professional specialists and researchers with an interest in self-management. The aim of the discussion was to answer some general questions, including: What does self-management mean for people with MS? What are the essential components of successful self-management interventions? What outcome measures are appropriate? The answers to these questions were used to develop new research proposals.

(c) designing a project to address the research question

Involving patients and carers in developing research projects is a long-established and proven way of ensuring the research reflects patients' and carers' interests in the conceptual, ethical and practical aspects of its design. One of the PSP Leads described the difference made by involving patients and carers at this stage of responding to a JLA PSP priority, illustrating the importance of continuing the ethos and principles underpinning the JLA in all subsequent activity (See Case Study 12).

Case Study 12: Childhood Disability PSP (2014)

One of the priorities of the Childhood Disability PSP was about how to improve continence in disabled children and what interventions (training programmes, medicines and/ or surgery) could speed up the process. The NIHR put out a commissioned call for some initial scoping research to identify what interventions were currently being used in clinical practice. The PSP Lead, Chris Morris, Associate Professor at the University of Exeter, is also a researcher in this field and decided to work with a group of parents of disabled children to develop a response to the call. He described the impact of their involvement as follows, *"In the commissioning call, they'd asked for a survey of practice in the NHS, and a systematic review of evidence around what was known to be effective. And through the involvement, it became evident that we were going to be missing a trick if the survey didn't include families, which I don't think was in the call. It was more what are the health professionals doing in the NHS. So, the public involvement effectively expanded the plans for the survey to include families as well as professionals."*

Their application was successful and their work was funded. Two of the parents who were involved, have become co-investigators in the study, in addition to another group of parents that advises on how the project is carried out.

Key lessons

- Developing research questions has largely been the responsibility of clinicians and researchers working with JLA PSP priorities
- Although the potential added-value of involving patients and carers in the development of the research question is recognised, this has rarely been done
- While some PSPs have involved patients, carers and researchers in workshops to scope a topic, as yet there is not a clear process for the whole of the translation stage

4.6 Evaluating: assessing whether JLA PSPs are making a difference to research

The issues raised by the interviewees in relation to evaluating JLA PSPs included:

- How to assess the impact of a PSP
- How to disseminate the impacts of a PSP

These will be discussed in turn.

How to assess the impact of a PSP

Most of the PSP Leads in research funding organisations have thus far attempted to assess the impact of their PSP by monitoring how many of the research grants they have awarded have been allocated to projects addressing the JLA PSP priorities. Many of the PSPs interviewed were able to list funded research projects that had addressed their priority topics. For example, the Mild to Moderate Hearing Loss PSP had identified several different research projects addressing their priorities from 2015, which included NIHR and Department of Health and Social Care reports, a PhD studentship, an NIHR fellowship, systematic reviews and a feasibility study for a clinical trial. The charities that had led PSPs were also able to identify other kinds of projects that had addressed their priorities, for example evaluating new employment initiatives for autistic people following the Autism PSP (see Case Study 6).

The task of monitoring which priority topics are addressed by research is comparatively easy to do within charities that fund research, as they can include a question about this within their own funding applications:

> *We're using the question about priorities in the application form as way of monitoring which priorities are being addressed, so that in a couple of years' time we can say 'this number of applications have addressed our Top 10'.* **PSP Lead**

Some interviewees suggested that other funders adopt a similar approach, for example, including a tick-box or drop-down menu in NIHR funding application forms, for researchers to state whether their research addresses a JLA PSP priority topic. Without such notification it becomes very difficult to monitor what JLA-related research is being funded. One way to do this is to employ someone to track the mentions of PSPs in reports of completed research projects. This has been tried by one University, utilising the time of an Impact and Communications Officer, as part of their wider role to assess the impact of the School of Medicine's work as a whole.

> *She does spend an awful lot of time just googling and following up what she finds... If you just want to be able to say, we did this PSP ten years ago and now look at all of these different topics that have been researched, then really you can be googling around the actual questions themselves… Her role is quite unique… talking across virtually every other department in the school, they haven't got a role like this. It's remiss really.*
>
> **PSP Lead**

Although the JLA Secretariat does request they be kept informed of all successfully funded research addressing the priorities, it is not clear to the various stakeholders as to who should do this – funders or researchers? The researchers who were interviewed did not consider this to be their responsibility, assuming that the charities funding their work would do so.

Disseminating the impact of JLA PSPs

Some of the PSPs who finished some time ago had shared progress on researching the priorities with their respective patient, carer and researcher communities. For example the Tinnitus PSP (2012) produced a report five years later, detailing work that had addressed their Top 10 priorities, while Diabetes UK was in discussion with a journal producing a special issue on research relating to the priorities from their Type 1 and Type 2 Diabetes PSPs.

The patients who were interviewed said they wished there was a way they could keep track of what work was going on to address their Top 10 priorities:

> *One of my worries is that it's been slightly forgotten… what do we know about what has been achieved, if there was a place where you could find out easily, where you could follow what has been done, that would be just brilliant.* **Patient**

One of the researchers suggested finding a way to inform all of the patients and carers who took part in the original PSP of the results of research addressing priorities in the same way that participants are informed of the finding:

> *When we do clinical trials we spend a lot of time and effort making sure that we've produced a lay summary of what the trial means, and make sure it's gets sent to all of the trial participants, but by then you often are talking five, six years down the line from the PSP… what we don't do enough of, is making sure we send that information to everybody who took part in the PSP as well.* **PSP Lead**

Key Lessons

- Most PSPs have aimed to assess impact by monitoring how many projects are funded that address priority topics – this is resource-intensive and difficult to do across all funders
- Some PSPs have produced reports of the research that has been completed in response to JLA PSP priorities, but it is not clear whose role it is to collect this information and how it could be done systematically
- Patients and carers involved in JLA PSPs would like this information to be more comprehensive and publicly available in one place

4.7 Transforming: changing culture, policy, organisations and people

The experience of going through the JLA process appears to have a profound impact on the people who take part, with changes in their values, thinking, plans and actions. This can lead to a change in the culture of the organisations they work in, and/ or the communities they work with, as well as influencing wider policy and practice. This was described by one of the interviewees like *'the ripples in a pond'.*

These broad impacts relate to:

(a) Changing individuals – patients and clinicians

(b) Changing organisations that took part in a JLA PSP

(c) Changing the research culture

(d) Changing health services policy and practice

These will be discussed in turn.

(a) Changing individuals

Patients

The patients who were interviewed found that the experience of taking part in a PSP had:

- given them enormous satisfaction from having been involved in a meaningful exercise
- improved their confidence
- changed their involvement practice
- enabled them to network with other patients

These impacts are described in more detail in the patients' own words in their personal stories (see Stories 1 and 2). Other people involved in PSPs had also witnessed such changes in the patients they had worked with:

> *With one of the patients I worked with, when I spoke with her dad at the workshop he had tears in his eyes saying how proud he was of her and what she's achieved in the project and that it had made her so much more confident in expressing her opinions. I know she has now taken some steps towards addressing the issues that were holding her back, and it's absolutely wonderful. I can't say it's because she was doing the JLA PSP that she is now able to tackle those issues, but I'd be surprised if it's not…* **PSP Lead**

1. Lynn Laidlaw's story

How did you get involved in the JLA?

I'd submitted questions to the Oral and Dental Health JLA PSP (2018) and I think everyone that submitted questions got invited to apply to take part in the final workshop. So I did because I've got Bechet's syndrome (a rare disease), and one of the manifestations is oral. I've also participated in the Healthcare Associated Infections PSP (current). I'm interested in that is because I am immunosuppressed.

What has been your experience of being involved in JLA PSPs?

The first thing that struck me was it felt very equal and I have reflected on that. I do a lot of research involvement activity and I'm not sure I go to any other forum where the conversation is that equal. It made me realise the importance of excellent facilitation that brings out the best in everyone and has such an impact on what they are doing. It seems to be crucial, if you get all the right people in the room, then having the right conditions and facilitation for conversations to grow.

What has been the personal impact of being involved in a JLA PSP?

My first JLA PSP was a thunderbolt moment. It made such a big impact on me. I kind of floated away from the meeting and I was on a high for days because I just felt like I'd been involved at something that was really meaningful. And I don't feel like that every time that I get involved in research. Research is hope, and research is the difference between life and death and having quality of life. No-one wants researchers to succeed more than patients!

How has being involved in a JLA PSP changed what you do when you're involved in research?

I think it's improved my research grant review. It's given me an extra dimension. When people write in applications about the James Lind Alliance, I can be more critical because I understand the process. Before I might have accepted it at face value. I'm also trying to put a bit of the JLA ethos into my other involvement activity. For example, I was in a conversation (on Twitter) about a Delphi (survey) exercise and I asked 'Oh, are you going to involve patients?' That led to a conversation with the clinician organising it, and I talked about the JLA, and they said they couldn't afford one of those. So, I offered to help them to encourage questions from patients and members of the public in their survey. We wrote some text (in Plain English) encouraging people to put forward a research question, or if that was too hard then to share what's important to them as a patient. If I hadn't have gone through the JLA I don't think I would have been able to contribute to that activity in the way that I did. Finally being part of James Lind Alliance workshops is also a chance for me, as a patient, to network as well, which I think is really important.

2. Irenie Ekkeshis's story

How did you get involved in a JLA PSP?

It came about because I had contacted Fight for Sight, (a sight loss charity) and asked them to amend their information on the disease that I had contracted (Acanthamoeba keratitis - AK) back in 2011. AK had caused total vision loss in one eye, and this had a dramatic impact on my life. After various conversations with staff at Fight for Sight, they sent me an invitation saying 'We're running this PSP and we're looking for patients to participate and we thought that the cornea one would be really relevant to you.' So I agreed to attend the final workshop.

How has your experience in a JLA PSP affected you?

It's changed the course of my life and had a massive impact on me, and what I do now. It was the first time that I realised that patients should and could have a say in the whole healthcare ecosystem. They weren't just victims or the 'done to' but that they could positively impact what's going on.

But I have to say I'd never heard of it before, and it had never occurred to me that that didn't happen already, because why wouldn't funders say, 'Well we've got a limited pot of funds, and we have to be really careful because resources are tight, so what we're going to do is concentrate our efforts in these areas where we feel that there's the most need, or where there's the potential to have the most impact.' And it felt like a no brainer that everybody involved in that situation would be invited to come together and jointly agree that stuff.

What in changed for you personally?

Before the workshop I thought my opinion wasn't as valid as others who are more educated in the topic (doctors, researchers etc). I couldn't really understand how I, with the limited experience of my own situation, could have an equal voice in a conversation with a professor. Then I realised that this JLA process was going to be different, not where you have a patient in the room and then everyone smiles, nods and tilts their head to one side and goes, 'Thank you for telling your really dramatic story and now we're going to get down to our professional business and you can leave the room!' Instead it was a very skilfully managed group, and this helped us to respect, reflect and understand and build on each other's points of view. It's quite a deliberative process and I thought it was amazing, I really did. I walked out the room at the end and I felt three feet taller.

How has it affected you since?

I felt quite excited about what I had done, and the impact it might have and that spurred me on to do more, to start campaigning, to think about the patient voice and how it's used, and where participation's really meaningful. This was great for me as I had lost my job during the course of my illness, and I was feeling like I'd lost a sense of purpose and direction in my life since becoming ill.

What else have you done since the PSP?

I basically started a campaign to raise awareness of the danger of water and contact lenses mixing, which has had knock-on effects all over the optical sector. I've achieved changes in lens packaging and more prominent warnings of the risk factors for the disease. My work has been supported by industry-led initiatives like Love Your Lenses Week www.loveyourlenses.com/why-love-your-lenses/ which is all about contact lens health and information about how to wear and take care and use contact lenses more safely.

I also set up a consultancy called the New Citizenship Project, where we work with organisations to explore what it means to really invite people to meaningfully participate. We apply lots of what I learnt during that PSP to our work, and it remains an inspiration to me of what can be achieved when you give people an equal voice and agency. We work with all kinds of sectors, including cultural heritage, health, the third and even the commercial sector. I could honestly say (and I know this is a very grand extrapolation from my PSP participation) it showed me what meaningful participation could be like and inspired me to encourage more of it in the world.

What do you think the wider impacts have been?

Research funding for my eye disease is really limited, and funding for eye disease in general is poor. Since the PSP, the profile of my eye disease has really skyrocketed, no one had really heard about it before and now it's taken very seriously and the impacts of that are really far reaching.

Clinicians

The clinicians who were involved in PSPs reported that the experience had:

- Changed their clinical practice
- Made them more aware of patients' lives and their concerns
- Made them more aware of the aspects of healthcare that still need improving
- Enhanced their visibility, status and credibility amongst their peers
- Increased their willingness and confidence to involve patients and carers in their work
- Expanded their networks to support future work

These impacts are described in more detail in the clinicians' own words in their personal stories (see Stories 3 and 4) and below:

People often ask me 'Why should I do a PSP?' I say it's the issue of visibility, to be seen to be raising the profile of research in our area, that's good for everybody involved. As an academic, JLA PSPs can support the Research Excellence Framework, but for me personally, it's all about if I'm going to invest the next five to ten years of my life researching something, I want to feel that I'm researching something that somebody wants to know the answer to… It gives me massive confidence that, yes, enough people were thinking this is an important topic, so I'm prepared to focus in this way. And it's a great way of building networks and contacts. After you've done a PSP, suddenly you've got hundreds of people who can help you on the journey moving forward. **PSP Lead**

3. Noémi Roy's story

How did you get involved in the JLA?

I'm a clinician (haematologist) working with people with lifelong blood conditions, and I wanted to do a JLA PSP in this area, focussing on six rare inherited anaemias. I became the PSP Lead and we completed the process in Summer 2018.

How has taking part in a JLA PSP affected your research?

One of the things I'm interested in is quality of life and not a lot of money goes into research on this in these rare conditions. Now, when we are writing grant applications, we can reference the priority on quality of life, which gives us more credibility than simply saying, 'We've asked the patients and they support us.' It validates some of the work we are already doing. I'm also using the PSP questions to drive research on fatigue, which came up as a very strong factor that patients want research on, but few people are focusing on that. I that think the real advantage is focussing on quality of life, non-medical interventions, non-drug-based interventions - the kinds of things that wouldn't necessarily be thought about if it wasn't for the PSP.

What difference has being involved in a JLA PSP it made to you professionally?

I would say it's had a huge impact professionally with the contacts that I made, and it has increased my visibility and credibility with colleagues. I'm part of the European Haematology Association, some guideline groups, some rare anaemia groups and EuroBloodNet which is a European Reference Network. In my interactions with these groups, I always bring in the JLA approach, asking whether what we're doing is going to make any difference to patients. It's become part of what I talk about and consider when looking at different courses of action.

The PSP also brought me into contact with a patient charity and they've invited me as their research ambassador to a conference. I spent the weekend with the patients and their families, which was a real 'eyeopener'. I heard stories that I would never have heard otherwise, about how doctors relate to patients, and how the choice of words used are important such as giving people fear, hope or being realistic. If it wasn't for the JLA PSP I wouldn't have had that interaction with them.

Some of the personal relationships I have made with the patients I've met have had a deep impact on me, particularly to see the way that taking part in a PSP has been of tremendous benefit to them personally, in how they view themselves and their contribution to society. I wonder how we can capture that. It's an enormously important outcome of the process, but doesn't figure in any measures.

Has it made a difference to your clinical practice?

It has made a difference to how I practice medicine. It's made my patients seem more human, that might sound weird, but it's helped me see my patients in a wider context, from a different point of view - because when you're dealing with patients through their medical care, the focus is the condition that they have. When you're dealing with patients through the PSP, you see much more of their life, their aspirations, their fears, their reservations and really what's important for them, their values. Now, all of these things *can* come out of a clinical consultation but often they won't, as the focus is more limited.

So it has given me more perspective and when I'm talking to patients about their treatment, I now do that in a more collaborative way.

Finally, going through all of the submitted questions from the survey, we came across some really sad stories. You could see the struggle families had experienced in getting anyone to listen to them, either taking them seriously when they were saying they thought something was wrong with their child, or even with logistics, like nobody ever picking up the phone when they tried to get in contact with their medical team. I think it's made me more aware of how the little things really make a big difference. So, now I think I try harder to get the details right for my patients much more often.

4. Alexander Heazell's story

What has been your involvement in the JLA?

It was part of my NIHR Clinician Scientist Fellowship programme in Maternal and Fetal Health research and I initiated the Stillbirth PSP (2015). I was challenged whilst I was submitting the proposal for what I was going to do. Someone asked, 'How do you know your research questions are important? You should do a JLA PSP priority Setting Partnership', and so we did.

As a researcher what has been the professional impact for you from doing a PSP?

For a very long time we struggled to get research in Stillbirth onto research funders' radar, anyone's radar, except the pregnancy charities who already bought into its importance. There is an element of 'shoulder shrugging' about stillbirth so to be able to say this was of sufficient importance that we could pull together a priority-setting process, and develop these questions, that means that these questions merit answering. So, I think actually the whole process validated the topic, rather than just this is a relatively small number of people who have come together, otherwise it's just me writing grants saying stillbirth is important. Actually, it was of sufficient importance that over 1,100 people completed a questionnaire to help and to tell us what the important questions were, I think it gives it more weight.

The process of undertaking the PSP enlarged my network with stakeholder organisations and other potential funders, and also a more local network of people interested in the JLA approach and local processes have developed out of that.

It also improved our relations with the hospital trust library staff, they saved the day as we were looking for people to do the literature searches. It also gave me a better understanding of the breadth of literature in stillbirth, which fitted in with the systematic review work that I was doing as well.

What about your experience of working with public contributors to research?

It gave me a really good insight into patient and public involvement (PPI), I think there's a lot of people that subscribe to it, but actually don't really do it. Maybe they feel they have to do it because there's a box on the form requiring them to write something and they won't get funded if it's blank. At a fairly key point in my career the JLA PSP experience made me realise how important PPI can be and you can actually gain a huge amount from it. I would say subsequent to that, my fellowship and my research has had quite a strong PPI component and we're just at the moment writing up how we think the PPI has changed what we did. At the end of this month my PPI group are going to do some of the analysis of interview transcripts from women who participated in a clinical trial. I don't think I would have got to that point without the PSP. It has been a very affirmative process.

(b) Changing the organisations that took part in a JLA PSP

Taking part in a PSP changed organisations in a number of ways including:

- Enhancing their status and credibility
- Changing their culture and values, leading to different ways of working
- Creating new infrastructure, policies and projects

These will be discussed in turn.

Enhanced status and credibility

Having been through the JLA process, which has a strong reputation for being a robust and high quality process, a number of interviewees reported that their organisations had gained status, visibility and credibility in their respective fields:

> *When I contacted the Royal College of GPs and said, "Could we write an online course for your website?", they agreed. I think it was having been through the PSP that helped, because they could see that we were sensible… almost everything we've done comes from doing that PSP. It truly did give us gravitas. It was immensely important.* **PSP Lead**

> *It's helped us to establish ourselves. We're now the UK's X charity now… if you were to ask influential people, they would see us as a central voice… the JLA is one of several things that have happened which has led to that... We've started talking with confidence as well, which comes from having the authority, knowing that you're speaking to people's priorities. If someone criticises us, I don't think any of the team feels too bothered about it, because we've followed the process and we feel like we're doing the right thing.* **PSP Lead**

> *It demonstrated leadership in a disparate sector, and it demonstrated that we could deliver what was quite a big project for us at that time… It has enabled us to move into a level of operation and engagement with external organisations that we simply would not have done beforehand… It was hugely positive in saying to the external world that here is an organisation that is taking a leadership role in X research.*
>
> **PSP Lead**

> *It's put us on the map.* **PSP Lead**

Changed culture and values, leading to different ways of working

Some of the organisations that carried out a PSP chose to do so because they knew the JLA process to be in strong alignment with their existing culture and values, and therefore saw it as a means of achieving the goal of becoming patient-centred in their work:

> *Right from day one of setting up our Centre, was that patients were at the heart of everything that we did. That's why we were early adopters of JLA because it just sang to our existing values... So, it wasn't like we had to make this big sea change.*
>
> **PSP Lead**

For others, the JLA represented the start of a culture change, particularly around becoming more collaborative, both with external partners (see Case Study 13) and internally amongst different departments (see Case Study 14). For organisations that fund research it has changed the nature of their relationship with the researchers they fund

(see Case Study 15). Working towards a shared goal of patient-centred research appears to help all these kinds of partnerships to first establish and then subsequently sustain their motivation.

Case Study 13: Sight Loss and Vision PSP (2013)

The Sight Loss and Vision PSP brought over 40 organisations on board, initially to help with dissemination of the survey. Michele Acton, who was CEO of Fight for Sight at the time, believes the experience had a huge impact on her organisation by promoting a culture of working in partnership with other related charities, *"Last year Fight for Sight worked with 23 partner organisations, whereas before the PSP we partnered with two. The PSP helped to give all those organisations a shared agenda, an output which the whole sector could use and it helped build trust between the organisations."*

Working in partnership is never easy as Michele describes, *"It can be painful at times and difficult and sometimes you think 'Can we actually deal with this?', but you have to constantly think about what is in the best interests of patients and how to make it work."*

Uniting around the shared value of putting patients first is what helps to cement these partnerships and maintain efforts, *"It's part of the shift from saying 'We fund research that researchers want funded' to saying 'We fund research that patients want.' And that's a massive cultural shift that we're all contributing to."*

Case Study 14: Type 2 Diabetes PSP (2017)

A shared goal to be patient-centred helps different parts of an organisation to collaborate, as opposed to working in silos. Kamini Shah Head of Research Funding at Diabetes UK, described such a change in her organisation, *"We are a patient charity, so we're driven by what people with diabetes want. We should be doing that as an organisation together, but there are definitely challenges in that. Because we started having those conversations in the build up to the PSP to get everyone on board, that was what helped us the most and has helped us to start working more closely together."*

Case Study 15: Multiple Sclerosis PSP (2013)

As a result of the taking part in the PSP, the MS Society has changed the way it works with researchers, promoting greater collaboration amongst academics, as well as between the charity and the researchers they fund, as Susan Kohlhaas, Executive Director of Research at The MS Society explained, *"Originally, we had a traditional funder/researcher relationship - we give you a grant, you report against these milestones and we'll be in touch if anything goes wrong. The JLA made us think more about how we're going to work alongside other funders, academics and different institutions, to try and get things done."*

The MS Society is part of a Stop MS fundraising appeal that is raising money for their research programme. They realised they needed to ask a range of researchers to work on the feasibility of such a trial and therefore, *"We decided to put out an invite to different kinds of researchers and had around thirty applications. The instinct as a funder is to go through a competitive process and pick the best. But we decided not to do that, because we felt that creating divisions at an early stage would just lead to problems down the road. So, we accepted everybody onto a consortium, which has actually worked really well. We're having to work closely with them as a team, setting objectives and time frames, but how they get to those objectives is up to them. We've done all the feasibility work in just over a year, which has been helped by being much more flexible, and encouraging the community to be less competitive and more collaborative."*

Observing and learning from the experience of conducting a PSP can also encourage others to decide to carry out a PSP themselves or adopt a similar process in another context, transferring the change in ways of working across departments and different institutions:

> *About a year after we did the JLA, the PSP Lead moved roles from research into strategy. And he developed our organisational strategy using a very similar process… It became a cross-organisational priority setting exercise that helped us to work out how to prioritise our activities beyond research*. **PSP Lead**

> *Because we're quite high profile within our local area, others have been watching what we've been doing and then converting. You grow as an organisation, and we all watch and learn from each other*. **PSP Lead**

New infrastructure and resources

Some of the PSPs created new structures to support patient and carer involvement in their organisations, as an integral part of the JLA process (Case Study 16). Others have decided to do this as a consequence of the PSP (Case Study 17). Part of the legacy of a JLA process is therefore to create the skills, resources and capacity to support increasing involvement in subsequent ongoing research.

Case Study 16: Mental Health in Children and Young People PSP (2018)

The Mental Health in Children and Young People PSP set up a Young People's Advisory Panel and a Young People's Network to support the process, which they have continued to fund. The plan is for these young people to continue to work with the McPin Foundation on the rest of their journey responding to the priorities. The priorities are on the agenda at every Panel meeting. Thomas Kabir, Head of Public Involvement at McPin reported, *"Now with this new resource, which includes the staff who have skills and experience of working with young people, we're getting researchers coming to us. They want our help with young people's involvement in their bids. We can draw this pool of young people to come to meetings or take part in other activities, when these kinds of request come in."*

Case Study 17: Autism PSP (2016)

Conducting the Autism PSP led Autistica to set up and launch its Discover Network, which brings autistic people, their carers and researchers together, with the goal of making research more authentic and impactful. James Cusack, Director of Science has observed, *"The Network has brought the community closer together and ensured autistic people are not just involved in setting priorities, but also in the development of research ideas and the full research cycle. That wouldn't have happened if we hadn't done the JLA first. We have since been partners on research applications, which is something that hadn't happened in the past. It also ensures we follow a process for funding research which is most likely to be useful to people, because we're focusing on what people want. For us that's massive."*

Some PSPs have also prompted organisations to develop new resources to meet the needs of patients and carers that have become apparent through the JLA process (See Case Study 18).

Case Study 18: Sight Loss and Vision PSP (2013)

One of the issues that emerged from the Sight Loss and Vision PSP is that patients want to know which new treatments are on the horizon. In response to such questions, the NIHR Horizon Scanning Research & Intelligence Centre conducted two horizon scanning reviews in collaboration with Fight for Sight, looking for new and emerging treatments for corneal conditions and for inherited retinal diseases. As Michele Acton, who was formerly the CEO for Fight for Sight reported, "*It is very difficult for people to get the facts in a clear and concise manner about what potential treatments might be on the horizon. Every time there's something in the newspaper about a cure for blindness, people have to rely on good journalism, or make their own assessment of whether that might be a treatment that could help them. The two horizon scanning reports helped give patients and clinicians the necessary facts about the clinical trials in progress.*"

(c) Changing the research culture

Many of the PSPs could identify ways in which their JLA process had brought about a shift in the research culture within their specific research communities. The exact nature of the shift depended on the context, and the dominant concerns in their fields at the time their process started. Sometimes it resulted in greater willingness to engage with the topic (Case Study 19), or a greater recognition of the need for research (Case Study 20) or greater consensus around whether research is required (Case Study 21). However, the PSP Leads emphasised that the JLA process cannot not be expected to resolve all of the conflicts and challenges, but does add value by helping to move the debates forward.

Case Study 19: Lyme disease PSP (2012)

A point of contention in the Lyme disease community is around complex cases, where patients report symptoms of Lyme disease or continuing symptoms after treatment, but standard blood tests prove negative. The patient community believe the diagnostic tests have in-built limitations, while the clinical/ research communities attribute the symptoms to another cause, including psychological problems. The patient group that led the Lyme Disease PSP set out with the intention of getting researchers to engage with this area of research as Stella Huyshe-Shires from Lyme Disease Action described, *"What we wanted was to have an official voice saying 'There are uncertainties'. We wanted to engage clinicians in the process, because they would only believe these uncertainties if they participated."*

The patient group had experienced years of being dismissed because of their assumed 'lack of scientific training', (they were all scientists), and hoped the process would give them some legitimacy as Stella explained, "*International researchers had told us that they had nearly given up on Lyme disease research because there are easier diseases to work on, and they feel 'battered' by some groups of patients. So we were working in this political maelstrom really, and we hoped that by doing the JLA, we would prove that we are science based and we know what we're talking about. We were very careful."*

The PSP struggled to get any clinicians involved in the process other than health professionals who were also Lyme disease patients. However, Stella believes the project still achieved their goal for legitimacy in that "*The PSP at least forced some people to look at us and think, 'This is sensible, it's got a good background, whatever it comes up with will be right."* She has found it has opened doors for them to engage with the Department of Health and its agencies, as well as some professionals and researchers, although so far only those working in primary care, rather than secondary care.

The shift has been slow. The priorities themselves got very little traction at the time they were launched in 2012. They were presented for the first time at a UK professional meeting five years later in 2017, although Lyme Disease Action was not invited. To date, none appear to have been addressed through research.

Case Study 20: Stillbirth PSP (2015)

Researchers working on stillbirth report being part of a small, close-knit community, which has helped to create ownership and acceptance of the priorities across the field. Alex Heazell, Professor of Obstetrics at the University of Manchester, reported, "*The journal article about our priorities was published in 2015 and has been cited 32 times since then. And then other researchers in the fields of miscarriage and sudden infant death syndrome have used our PSP as a basis for theirs.*"

The combined impact of these three PSPs across these aspects of pregnancy and early childhood has successfully challenged a wider cultural acceptance that 'these things just happen', as opposed to being events that can and should be prevented. Alex explained, "*Because the PSPs identified all of those questions, it challenged that sort of shoulder shrugging about whether anything more could be done*."

Case Study 21: Autism PSP (2016)

The autistic community is deeply divided over which types of research on autism are ethically acceptable and should go ahead. Some people are concerned that research in this field is aiming to 'cure' autism or eliminate the condition through eugenics. Autistic people also vary greatly in terms of how they are affected, as James Cusack, Director of Science at Autistica, explained, *"Autistic people can on one hand have high support needs, can be minimally verbal and have issues with aggression. So they are obviously having an extremely difficult time. But then there are people who are outstanding people in society, professors in universities who are doing very well. We know some autistic people out there who, if they were given the choice tomorrow, would take a drug that would alleviate some of the core issues, whereas other people would find that ethically, just completely unacceptable. It may be overambitious to think you can address the needs of all of these different groups within one PSP."*

The aim of the autism PSP was to try to build some consensus across the community about which research on autism should go ahead. James believes it has partly achieved this goal, "*It did help us to build consensus on where to focus our efforts and helped to justify what we are doing. That generated a lot of buy-in from the community, but ultimately we still ran into problems. There are still concerns and a distrust of research. We have to embrace the fact that different views exist and there is no one right view. The JLA won't solve all of it. It gets you closer, but there's still work to be done."*

(d) Changing policy and practice

Many of the JLA PSPs across many different conditions raise questions about health service delivery as well as research. Some organisations, either as part of the PSP or beyond, have responded to these kinds of priorities directly either by changing the service they provide (personal communication), by developing new practice guidelines (See Case Study 22). One of the international PSPs has succeeded in changing national health policy (See Case Study 23) and has been included to illustrate the potential for national influence in the UK.

Case Study 22: Tinnitus PSP (2012)

One of the questions that was prioritised in the Tinnitus PSP was about how to manage tinnitus in children. David Stockdale, Chief Executive of the British Tinnitus Association, explained that they decided to address this issue immediately through developing paediatric guidelines, based on the evidence already available "*We were able to get a group together to develop new guidelines which were then published through the British Society of Audiology, which is responsible for guidelines in our area of work, so it has credibility. That was done basically at zero cost through the contributions of some excellent people who could see the need, and were willing to put in the time to make sure that document was produced.*"

David's influence seems to have been essential to drive this work forward which he described as, *"Just soft lobbying, it was knowing who'd be good people to write it and just saying, 'Look we've got this question, you all agree it needs to happen, when are you going to write it?' The JLA was instrumental in creating the motivation as most of the people writing it had been involved in the PSP in some way."*

Case Study 23: Canadian Dementia PSP (2017)

The Canadian Governments new strategy for Dementia "*A Dementia Strategy for Canada: Together We Aspire*" made explicit mention of the Canadian Dementia PSP as part of its recommendation to establish and review strategic dementia research priorities for Canada. The strategy states:

"Another example of a stakeholder-engaged priority setting approach comes from a collaborative effort among the Alzheimer Society of Canada, the Toronto Rehabilitation Institute, and the Canadian Consortium on Neurodegeneration in Aging. The Canadian Dementia Priority Setting Partnership initiative was undertaken to better understand research priorities of those living with dementia, caregivers, families, health and social care providers and the general public. The Partnership questions focused on living with dementia, dementia prevention, treatment and diagnosis. Questions on cure and the biological mechanisms of dementia were not included in this prioritization exercise. The outcome of this priority setting process differs from those dementia research priorities outlined by the World Health Organization in 2016, which was informed by researchers, clinicians, and health and care workers. For example, stigma, and early treatment were prioritized by the Partnership while the WHO priorities included a strong focus on prevention, diagnosis and therapies. These differences demonstrate the importance of dialogue with multiple stakeholders when setting and reviewing research priorities."

Key lessons

- The outcomes and impacts of JLA PSPs extend much further than the direct impact on the research
- Future evaluations of JLA PSPs will need to find ways to capture and report the impacts on the people, the organisations, policies and practice, and the wider research culture, as well as how these evolve over time

5. Conclusion

The aim of this evaluation was to find out how JLA PSP priorities have successfully influenced decisions about which research projects to develop and fund, and to draw out lessons for past, present and future PSPs. Even with the small numbers of PSPs involved in this project, we have revealed a rich and complex picture of the outcomes and impacts of JLA PSPs which go beyond simply funding research. The context for each PSP is hugely significant. The starting point of the PSP, what it aims to achieve, the individuals involved, the organisations that lead it – all shape the JLA process and outcomes, making it difficult to draw out general conclusions about 'how to succeed' and broadening the definition of what success looks like.

In this section, we draw on our experience and our insightful discussions with Advisory Group members to reflect on the findings. We note that our interpretation is influenced by many years' experience of patient and public involvement in research (SC and KS), of developing the JLA approach (SC) and from working in different capacities across a number of PSPs (SC and KS). We recognise that other stakeholders with different experiences may have different interpretations and that understanding their views will be important for deciding how best to respond to the findings (see Recommendations).

Based on the experiences of the PSPs and other stakeholders we interviewed, it is possible to identify some of the actions that future PSPs could take to maximise their influence. The most important of these seems to be proper planning and resourcing of the work that takes place *after* a PSP concludes, to continue to raise awareness of the priorities, to seek to influence individual researchers and funding organisations, and to monitor and capture the wide range of possible impacts. We note that health charities and patient organisations often have the skills, resource and networks required for this work, and have frequently proved to be effective leads and/ or partners for PSPs.

Key lessons include the importance of:

- Planning for the end of the PSP at the beginning of the project, in particular to clarify who owns the outputs, who will make decisions about how they are used, who will be accountable for what happens next and how the follow-up work will be resourced
- Developing dissemination plans to reflect the PSP's strategic goals, which may go beyond funding research and reach audiences other than funders and researchers
- Making greater strategic use of patients, carers, clinicians and researchers in promoting the JLA priorities through their own networks
- Working with funders after the PSP to shape their research agenda, recognising that they may not believe it is their responsibility to respond to the priorities – this influencing work requires people with the right skills and experience, often senior leaders within organisations
- Carrying out foundation work to build researchers' capacity to respond, promoting collective thinking on an issue, as well as identifying barriers to any research and addressing them

- Ensuring continued involvement of clinicians, patients and carers in the translation of JLA PSP priorities into research questions and projects, so that the spirit of the original questions are maintained
- Collecting information about research that has been completed in response to JLA PSP priorities and making it publicly available

However, this evaluation has also begun to identify factors within the wider research system that can limit the influence of JLA PSP priorities. These include the values held by funders and researchers, and the dominant culture within research organisations. This may mean that funders and researchers use the JLA PSP priorities to endorse and legitimise what they have already planned to do, rather than making significant changes. It may be beyond the power of individual PSPs to bring about the wholesale cultural shift required to genuinely change the national research agenda in favour of patients, carers and clinicians' priorities. Addressing deeply embedded beliefs and values is likely to require action from a wide range of stakeholders.

The dominant culture within many research organisations is that of evidence-based medicine. For this reason, funders and researchers may see their responsibility as generating the best quality evidence to inform healthcare policy and practice, and may therefore prioritise methods and research questions that align with the established hierarchy of quality evidence.
Typically, systematic reviews and meta-analyses rank most highly, randomized controlled trials (RCTs) rank above observational studies, while expert opinion and anecdotal experience are ranked at the bottom[11]. These values may be in in conflict when addressing the questions which patients, carers and clinicians want answered by research, as their questions may be better answered by methods other than clinical trials (see below). As yet this tension seems unresolved. Nor has there been sufficient debate as to whether it is public funders' responsibility to respond to priorities as defined by patients, carers and clinicians. We note that health charities, whose culture and values already align with meeting the needs of patients and carers, may experience fewer challenges in changing their research agendas.

Overall, there seems to be a general lack of understanding of the JLA process and its goals in the wider research community, beyond those individuals immediately involved in a PSP. This seems to create unmet expectations. One of the common responses to the JLA PSP priorities is, '*This is not telling us anything new*.' The list of priority topics can be familiar to clinicians and researchers, rather than identifying completely new research topics. We suggest this is because patients, carers and clinicians are identifying the questions that remain unanswered, precisely because they are difficult to research, low-tech or 'unexciting', and therefore these are questions that researchers may have previously decided are 'un-researchable'. More importantly, while the topics may not be new, the way that patients, carers and clinicians frame their questions may be different to researchers, requiring a novel methodological response, rather than

[11] Greenhalgh T (July 1997). "How to read a paper. Getting your bearings (deciding what the paper is about)". *BMJ*. **315** (7102): 243–6.

opening up a completely different research area (see below).

Another common response to the JLA PSP priorities is a concern that '*These questions are too broad for us to be able to work with them*.' Historically, when the JLA focused solely on treatments, the priorities often took the form of PICO formatted questions, which were often easier to adopt straight into the existing system. The JLA process has since evolved to cover a broad spectrum of health related issues and is now generating a much wider range of priorities, but it seems that researchers and funders are still expecting to work with the outputs at face value*, as if these are the research questions*. We suggest that the JLA PSP priorities are better understood as broad topic areas and that multiple research questions may emerge from each one. This creates the need for an additional step of working with the priorities to translate them into research questions and prioritise amongst possible research projects. Many of the health charities that led the PSPs in this evaluation have included such a step, but thus far they have taken different approaches towards slightly different ends. There is not yet consensus on what the task of translation involves and who needs to be included in the process.

Based on our experience of working on PSPs, we suggest that patients/ carers and clinicians/ researchers tend to frame the questions they ask in different ways. Patients/ carers tend to focus on *outcomes* asking questions such as "How can I stop my cancer coming back? How can I reduce the curve in my spine? How do I reduce the risk of another psychotic episode?' Patients and carers want to know what *combinations* of medical treatment, self-management, lifestyle change and alternative therapy are likely to be effective. They are often keen to hear about the positive experience of their peers asking questions such as 'What can we learn from the people who are managing their condition well?' In contrast, clinicians/ researchers, often as the people who deliver or who are the intervention, tend to frame questions in terms of 'Does X *intervention* work?' These clinician/ researcher questions are more amenable to answering with an RCT, and align more closely with the dominant research culture (see above). We note that researchers/ funders seem to approach patient/ carer questions by in effect asking, 'What RCTs might address this question?', rather than 'How can I answer the question that patients/ carers are asking?' These tensions highlight the need for continued patient, carer and clinician involvement throughout the translation from JLA PSP priority to research project, to ensure that the priorities are not lost or distorted in the process.

We note that much of the current activity that follows a JLA PSP often highlights the need for continued *patient* and *carer* involvement, while the clinician's perspective is assumed to be included when clinical researchers start to work with the priorities. We suggest there can be a difference between frontline clinicians who are the end-users of research, and the, often more senior, clinical researchers who may come with their own beliefs about research priorities that can dominate both the JLA process and any response to the outputs. Such potential conflicts of interest need to be surfaced and checked in subsequent processes, and the continued involvement of frontline or 'jobbing' clinicians assured. Similarly, we note that research organisations that have their own patient and carer

involvement processes, often assume these can do the job of interpreting the JLA PSP priorities. However, this can mean that one or two patients/ carers on a panel are given the responsibility of assessing whether a particular research project is an adequate response to a broad JLA PSP priority, *without* those patients/carers having knowledge of the JLA process or access to the original responses of the hundreds of people whose views helped shape the question. This raises the question of how JLA PSP priorities can be meaningfully integrated into current grant review processes.

We conclude that challenges arise at the end of a PSP when the priorities are handed over to funding systems and researchers (NIHR and others) that are not connected to the JLA process, that operate within the boundaries of an evidence-based culture, and that may have their own processes of involving patients, carers and clinicians who are again disconnected from the original JLA PSPS. We therefore suggest that further work is required to clarify what additional steps need to be taken to translate the JLA PSP priorities into research questions, in a way that maintains the spirit of the JLA process all the way through. Furthermore, the organisations that respond to the PSP outputs as yet have limited oversight, or any sense of responsibility for what happens to JLA PSP priorities. This fundamental question of whose responsibility is it to respond to the priorities still needs to be widely debated.

In summary, while the JLA process itself can no doubt still be improved, and the JLA PSPs supported and encouraged to do more with their outputs, there is a vital need for parallel commitment from other parts of the wider research system to change and adapt their response. A much deeper and broader cultural change is required to ensure that the overarching goal of delivering research that is more relevant and useful to the end-users is genuinely achieved.

6. Recommendations

We have chosen not to develop recommendations for follow-up action on behalf of others, and instead suggest that the different stakeholder groups (the JLA Secretariat, JLA Advisers, researchers, funders, patients, carers and clinicians) are engaged in a conversation about the findings, and work together to develop their views on how best to respond. Based on the key lessons from this evaluation, we suggest that some of the questions that could be usefully addressed include:

- **Planning**: If the work of promoting and influencing others at the end of a PSP needs to be properly planned and resourced at the beginning, is there a role for the JLA Secretariat to support this planning? What level of resourcing should be recommended? And where should this resource come from?
- **Disseminating and influencing**: If simply disseminating the priorities is not always sufficient to promote their uptake by researchers and funders, what can be learnt from implementation science about how to encourage others to change their behaviour in light of new evidence?
- **Responding by funding relevant research**: When assessing a research project that aims to address a JLA PSP priority, how can this be judged in a practical and meaningful way? What should funders, grant reviewers and members of funding panels be asking and looking for?
- **Responding in ways other than through research**: How can non-research questions be used for the benefit of patients, carers and clinicians i.e. to meet their information needs and to improve healthcare policy and practice? Which stakeholders need to be involved in this work and how can it be resourced?
- **Translating**: What are the tasks involved in the translation step from JLA PSP priority to research question or themed call? Who needs to be involved? What support and information do they need to do these tasks well?
- **Evaluating**: What are practical and meaningful ways of capturing whether new research addresses a JLA PSP priority? How can the scale and nature of the change to a portfolio be described? How can the change in the nature of individual research projects be captured? How can the wider impacts of a JLA PSP be captured?
- **Transforming research organisations**: If the goal is to transform the research culture in such a way that it better reflects the needs and interests of the end-users, what are the implications for the way the research organisations currently function, in terms of what they do and how they do it? How do they need to change to be able to respond in a meaningful way to the JLA PSP priorities?
- **Transforming people**: How can individuals' contributions to the process be better recognised and rewarded within the research system? How can the skills and experience that individuals gain through the JLA process be put to better use in the ongoing work and in other contexts?

Appendix 1: Abbreviations

AMD Age-related Macular Degeneration

BRC Biomedical Research Centre

CEO Chief Executive Officer

DUETs Database of Uncertainties in the Effects of Treatments

ESRC Economic and Social Research Council

EU European Union

FAQ Frequently Asked Question

GP General Practitioner

JLA James Lind Alliance

JLI James Lind Initiative

MS Multiple Sclerosis

NETSCC NIHR Evaluation, Trials and Studies Coordinating Centre

NHS National Health Service

NIHR National Institute for Health Research

NIHR CCF NIHR Central Commissioning Facility

PCORI Patient-Centered Outcomes Research Institute

PhD Doctor of Philosophy

PICO Problem/Patient/Population, Intervention/Indicator, Comparison and Outcome

PPI Patient and Public Involvement

PSP Priority Setting Partnership

UK United Kingdom

Appendix 2: The criteria used to select interviewees for this project

Topic of enquiry / variables to consider to select interviewees
1. Who led the PSP and who owns the outputs and how does this influence what happens next?
• Charity that funds research – how have priorities have influenced internal research strategy or funding decisions
• Charity that doesn't fund research – how have they influenced funders
• NIHR organisation with funding for research e.g. Biomedical Research Centre – how have priorities have influenced internal research strategy or funding decisions – vested interest?
• Umbrella organisation e.g. National Cancer Research Institute – how have they influenced funders
• Clinician/ clinician organisation – how have they influenced funders
• Co funding models whereby several organisations have contributed funds
2. How have priorities been disseminated/ shared with funders/ researchers/other stakeholders?
• Dissemination strategy details can be asked of all interviewees – how they disseminated their Top 10s or how they heard about the Top 10 – aim to identify innovative and effective approaches
• PSPs that have engaged funders as part of the PSP process e.g. funders on Steering Group (trade-off with scope of the PSP?)
• Barriers to dissemination – where publication of Top 10 has been blocked
• Examples of guerrilla dissemination – grassroots activity leading to raised awareness of JLA PSP priorities
3. How have funders worked with the priorities? & To what end?
• Charity or NIHR organisation that has allocated funding to priority topic – how can/ does mentioning JLA influence funding decisions, research strategy – what to do and not to do
• Charity or NIHR organisation that developed call for proposals based on a priority
• Clinical study groups or mixed working groups set up to develop projects from priorities
• Facilitating funders and researchers to come together and discuss priorities – NCRI conference
• PSPs that have collaborated on common priority areas to identify highest priority issues affected broad range of conditions e.g. incontinence
4. How have researchers worked with priorities? & To what end?
• Examples of researchers/ research groups that have developed a project based on a priority topic
• Examples of PSPs where no researchers working on the priorities – lack of researcher capacity (within our scope if we can draw on useful learning to help others?)
• Ask lead expert in a field whether JLA has had influenced the research landscape
5. How has taking part in a PSP affected the partner organisations post-PSP?
• Example where impact on organisational culture
• Example where impact on collaborative working with other organisations
6. How has it affected the individuals who took part in the PSP?
• Example of where an individual has been changed by the experience and taken new courses of action as a result
7. How have the other outputs of JLA PSPs been used for benefit?
• Example of where known unknowns have been used
• Example of where out of scope questions have been used