The Most Premature Babies Priority Setting Partnership (PSP) Protocol

Dated: 01 May 2023 - Version 3.0

Lay Summary

Priority Setting Partnerships (PSPs) bring together patients, their carers and clinicians to identify and prioritise unanswered questions (or as they can sometimes be referred to as ‘evidence uncertainties’) for research purposes, in specific health conditions or areas of healthcare.

This PSP is focusing on the Most Premature Babies - those born less than 25 weeks’ gestation (more than 4 months early). We hope to determine the top 10 unanswered questions in this group of high-risk babies. An unanswered question is any uncertainty related to this group of babies that could benefit from research.

We will do this by asking patients (i.e., ex-premature babies who are now adults), parents of premature babies and those healthcare professionals who look after premature babies what they think may be the most important questions that they would like answered. This will help guide future research into the Most Premature Babies and make sure that researchers focus on their most urgent needs.

The key areas of interest of the Most Premature Babies PSP are:

- Pre-birth (antenatal) maternal care to improve outcomes in babies born less than 25 weeks’ gestation
- Care delivered immediately after birth to babies born less than 25 weeks’ gestation
- The clinical management of babies born less than 25 weeks’ gestation
- The diagnosis and treatment of common health conditions affecting babies born less than 25 weeks’ gestation, and
- The impact of neonatal care (within the Neonatal Intensive Care Unit) on the physical and mental health and wellbeing of parents/carers of babies born less than 25 weeks’ gestation

The PSP will *exclude* from its focus questions about the following:

- Pre-birth (antenatal) treatment to prevent premature birth
- Causes of premature labour
- Babies born less than 22 weeks’ gestation and more than 24 weeks’ gestation
- The care of babies born less than 25 weeks’ gestation *after* they have been discharged from hospital
- Babies born less than 25 weeks’ gestation in low- and middle-income countries

This PSP is funded by the Nuffield Department Public Health (NDPH) Pump Priming Scheme, administered by the University of Oxford, for the UK arm of the project. For the Australian arm of the project, funding is provided by the Royal Children’s Hospital Foundation, Melbourne Australia.

1. Purpose of the PSP and background

The purpose of this protocol is to set out the aims, objectives, and commitments of the Most Premature Babies Priority Setting Partnership (PSP) in line with James Lind Alliance (JLA) principles. The Protocol is a JLA requirement and will be published on the PSP's page of the JLA website. The Steering Group will review the Protocol regularly and any updated version will be sent to the JLA.

The JLA is a non-profit making initiative, established in 2004. It brings patients, carers, and clinicians together in PSPs. These PSPs identify and prioritise the evidence uncertainties, or 'unanswered questions', that they agree are the most important for research in their topic area. Traditionally PSPs have focused on uncertainties about the effects of treatments, but some PSPs have chosen to broaden their scope beyond that. The aim of a PSP is to help ensure that those who fund health research are aware of what really matters to patients, carers, and clinicians. The National Institute for Health Research (NIHR – www.nihr.ac.uk) coordinates the infrastructure of the JLA to oversee the processes for PSPs, based at the NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC), University of Southampton.

The International Perinatal Research Partnership (INPRES) is a collaboration between the Murdoch Children's Research Institute's (MCRI) Melbourne Children's Trials Centre (MCTC) Melbourne, Australia, and the University of Oxford's National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford, United Kingdom. Its mission is to develop and facilitate clinical and observational research in a cross-border collaborative approach, which in turn will lead to novel, scientific and practice-changing discoveries in the perinatal, neonatal, and paediatric population. The Partnership's ultimate goal is to improve health outcomes for infants and their families. It is with this goal that the Most Premature Babies PSP was established.

2. Aims, objectives, and scope of the PSP

The aim of the Most Premature Babies PSP is to identify the most important unanswered questions about babies born less than 25 completed weeks' gestation, from patient, parent/carer and clinical perspectives, and to then prioritise those that patients, parents/carers and clinicians agree are the most important for research to address.

The objectives of the PSP are to:

- Work with clinicians, parents/carers of the most premature babies, and adults who were themselves born extremely premature to identify uncertainties about the management and care of the Most Premature Babies (born <25 completed weeks' gestation)
- To agree by consensus a prioritised list of those uncertainties for research, including finalising a list of the highest-priority Population – Intervention – Comparator – Outcome (PICO) questions to be answered by research.
- To publicise the results of the PSP and process
- To take the results to research commissioning bodies to be considered for funding
- To identify research to develop in the short-, medium- and long-term as programmes of work.

The scope of the Most Premature Babies PSP is defined as:

- Antenatal management to improve outcomes in infants born less than 25 weeks of gestation
- Resuscitation practices and provision of intensive care to infants born less than 25 weeks of gestation
- General management of infants born less than 25 weeks of gestation during their hospital stay
- Diagnosis and treatment of common conditions affecting infants born less than 25 weeks of gestation
- The impact of inpatient neonatal care on the physical and mental health and wellbeing of parents/carers of babies born less than 25 weeks of gestation
- The impact of inpatient neonatal care on the health and developmental outcomes of babies born less than 25 weeks of gestation.

The PSP will exclude from its scope questions about:

- Antenatal treatments to prevent premature birth
- Causes of premature labour
- Infants born less than 22 weeks' gestation and more than 24 weeks' gestation
- The care of infants born less than 25 weeks' gestation *after* they have been discharged from hospital
- Infants born less than 25 weeks' gestation in low- and middle-income countries

The Steering Group is responsible for discussing what implications the scope of the PSP will have for the evidence-checking stage of the process. Resources and expertise will be put in place to perform this evidence checking.

3. The Steering Group

The Steering Group membership includes parents/carers of extremely premature infants, adults who were themselves born extremely premature, and clinicians, as individuals or representatives from a relevant group.

The Most Premature Babies PSP will be led and managed by a Steering Group involving the following:

Parents/carers of the most premature babies' representative/s:

- Ms Amber Bates, Centre of Research Excellence in Newborn Medicine Consumer Advisory Group, Australia
- Ms Patricia Shanti Santhanadass, Oxford UK

Adults born extremely prematurely representative/s:

- Ms Louise Pallot, Centre of Research Excellence in Newborn Medicine Consumer Advisory Group, Australia
- Ms Olivia Ray, Buckinghamshire, UK

Clinical Representative/s:

- Prof Charles Roehr, Neonatologist, Southmead Hospital, Bristol UK
- Dr Clare Whitehead, Obstetrician, Royal Women's Hospital, Melbourne Australia
- A/Prof Brett Manley, Neonatologist, Royal Women's Hospital, Melbourne Australia
- Prof David Tingay, Neonatologist, Royal Children's Hospital, Melbourne Australia
- Prof Chris Gale, Neonatologist, Imperial College London, London UK
- Dr Trisha Prentice, Bioethicist and Neonatologist, Royal Children's Hospital, Melbourne Australia
- Prof Marian Knight, Professor of Maternal and Child Population Health, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford UK
- Dr Rosemary Boland, Neonatal Nurse, Murdoch Children's Research Institute, Melbourne Australia
- Prof Samantha Johnson, Developmental Psychologist, University of Leicester, Leicester UK

Methodology

- Ms Pollyanna Hardy, Statistician, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford UK

Project Lead:

- Dr Stacey Peart, Neonatologist, Royal Women's Hospital, Melbourne Australia

Project Coordinator:

- Ms Laura Galletta, Murdoch Children's Research Institute, Melbourne Australia

Project Support:

- Ms Kayleigh Stanbury, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford UK

James Lind Alliance Adviser and Chair of the Steering Group:

- Ms Suzannah Kinsella, JLA (April 2023 onwards)
- Ms Katherine Cowan, JLA (until March 2023)

The Steering Group will agree the resources, including time and expertise that they will be able to contribute to each stage of the process, with input and advice from the JLA.

4. Partners

Organisations and individuals will be invited to be involved with the PSP as partners. Partners are organisations or groups who will commit to supporting the PSP, promoting the process, and encouraging their represented groups or members to participate. Organisations which can reach and advocate for these groups will be invited to become involved in the PSP. Partners represent the following groups:

- People who were born extremely premature
- Parents/carers of people who were born extremely premature
- Health and social care professionals - with experience in delivering neonatal care to extremely premature infants and their families.

Exclusion criteria

Some organisations may be judged by the JLA or the Steering Group to have conflicts of interest. These may be perceived to potentially cause unacceptable bias as a member of the Steering Group. As this is likely to affect the ultimate findings of the PSP, those organisations will not be invited to participate. It is possible, however, that interested parties may participate in a purely observational capacity when the Steering Group considers it may be helpful.

5. The methods the PSP will use

This section describes a schedule of proposed steps through which the PSP aims to meet its objectives. The process is iterative and dependent on the active participation and contribution of different groups. The methods used in any step will be agreed through consultation between the Steering Group members, guided by the PSP's aims and objectives. More details of the method are in the Guidebook section of the JLA website at www.jla.nihr.ac.uk where examples of the work of other JLA PSPs can be seen.

Step 1: Identification and invitation of potential partners

Potential partner organisations will be identified through a process of peer knowledge and consultation, through the Steering Group members' networks. Potential partners will be contacted and informed of the establishment and aims of the Most Premature Babies PSP.

Step 2: Awareness raising

PSP need to raise awareness of their proposed activity among their patient, carer, and clinician communities, in order to secure support and participation. Depending on budget, this may be done by a face-to-face meeting, or there may be other ways in which the process can be launched, e.g., via social media. It may be carried out as part of steps 1 and/or 3. The Steering Group should advise on when to do this.

Awareness raising has several key objectives:

- To present the proposed plan for the PSP
- To generate support for the process

- To encourage participation in the process
- To initiate discussion, answer questions and address concerns.

Step 3: Identifying evidence uncertainties

The Most Premature Babies PSP will carry out a consultation to gather uncertainties from patients, parents/carers and clinicians. A period of 8 months will be given to complete this exercise (a duration which may be revised by the Steering Group if required).

The Most Premature Babies PSP recognises that the following groups may require additional consideration.

- People with cognitive, physical, visual and/or auditory processing problems
- People of non-English speaking backgrounds
- People of low socio-economic status

The Steering Group will use the following methods to reach the target groups

- Online surveys
- Paper-based surveys
- Face-to-face or online invitations conducted opportunistically
- Social media awareness campaigns
- Word of mouth (Steering Group members; hospitals and affiliations involved in the care of extremely premature infants)

Existing sources of evidence uncertainties may also be searched.

- Published randomised controlled trials and other published research studies, i.e. qualitative and observational studies
- Systematic reviews (e.g., Medline, Embase) with the assistance of a librarian where applicable
- PROSPERO for systematic reviews currently underway
- Clinical practice guidelines

Step 4: Refining questions and uncertainties

The consultation process will produce ‘raw’ questions and comments indicating patients’, parents’/carers’ and clinicians’ areas of uncertainty. These raw questions will be categorised and refined by Dr Stacey Peart, into summary questions which are clear, addressable by research, and understandable to all. Similar or duplicate questions will be combined where appropriate. Out-of-scope and ‘answered’ submissions will be compiled separately. The Steering Group will have oversight of this process to ensure that the raw data is being interpreted appropriately and that the summary questions are being worded in a way that is understandable to all audiences. The JLA Adviser will observe to ensure accountability and transparency.

This will result in a long list of in-scope summary questions. These are not research questions and to try and word them as such may make them too technical for a non-research audience. They will be framed as researchable questions that capture the themes and topics that people have suggested.

The summary questions will then be checked against evidence to determine whether they have already been answered by research. This will be done by Dr Stacey Peart with the support and assistance of the Steering Group. The PSP will complete the JLA Question Verification Form, which clearly describes the process used to verify the uncertainty of the questions, before starting prioritisation. The Question Verification Form includes details of the types and sources of evidence used to check uncertainty. The Question Verification Form should be published on the JLA website as soon as it has been agreed to enable researchers and other stakeholders to understand how the PSP has decided that its questions are unanswered, and any limitations of this.

Questions that are not adequately addressed by previous research will be collated and recorded on a standard JLA template by Dr Stacey Peart. This will show the checking undertaken to make sure that the uncertainties have not already been answered. The data should be submitted to the JLA for publication on its website on completion of the priority setting exercise, taking into account any changes made at the final workshop, in order to ensure that PSP results are publicly available.

The Steering Group will also consider how it will deal with submitted questions that have been answered, and questions that are out of scope.

Step 5: Prioritisation – interim and final stages

The aim of the final stage of the priority setting process is to prioritise through consensus the identified uncertainties about the most premature babies. This will involve input from patients, parents/carers, and clinicians. The JLA encourages PSPs to involve as wide a range of people as possible, including those who did and did not contribute to the first consultation.

There are usually two stages of prioritisation:

1. Interim prioritisation is the stage where the long list of questions is reduced to a shorter list that can be taken to the final priority setting workshop. This is aimed at a wide audience and is done using similar methods to the first consultation. With the JLA's guidance, the Steering Group will agree the method and consider how best to reach and engage patients, parents/carers, and clinicians in the process. The most highly ranked questions (around 25) will be taken to a final priority setting workshop. Where the interim prioritisation does not produce a clear ranking or cut off point, the Steering Group will decide which questions are taken forwards to the final prioritisation.

2. The final priority setting stage is generally a one-day workshop facilitated by the JLA. With guidance from the JLA and input from the Steering Group, up to 30 patients, parents/carers and clinicians will be recruited to participate in a day of discussion and ranking, to determine the top 10 questions for research. All participants will declare their interests. The Steering Group will advise on any adaptations needed to ensure that the process is inclusive and accessible.

Step 6: Follow-on stage

A further workshop, involving the Steering Group along with interested patients, parents/carers, clinicians, and researchers, will be held to determine the top 10 PICO style questions. This will involve reviewing the ranked priorities, which may include those outside the top 10 ranked uncertainties/priorities, to determine which unanswered questions can be made amenable to answer within a clinical trial. All participants will declare their interests.

6. Dissemination of results

The Steering Group will identify audiences with which it wants to engage when disseminating the results of the priority setting process, such as researchers, funders and the patient and clinical communities. They will need to determine how best to communicate the results and who will take responsibility for this. Previous PSPs' outputs have included academic papers, lay reports, infographics, conference presentations and videos for social media.

It should be noted that the priorities are not worded as research questions. Through the undertaking of a follow-on stage in order to develop the top 10 PICO style questions, published separately to the top 10 priorities, the Steering Group will be able to better work with researchers and funders to help answer the research questions that will address the issues that people have prioritised. The dissemination of the results of the PSP will be led by Dr Stacey Peart, supported with the assistance of the Steering Group.

The JLA encourages PSPs to report back about any activities that have come about because of the PSP, including funded research. Please send any details to jla@soton.ac.uk.

7. Agreement of the Steering Group

The Most Premature Babies PSP Steering Group agreed the content and direction of this Protocol on the 20th of July 2022.