Research priorities for the most premature babies born <25 weeks' gestation: results of an international priority setting partnership

Stacey Peart,[1,2,3] Olivia Ray,[4] Laura Galletta,[5] Amber Bates,[6] Rosemarie Anne Boland,[2,3] Peter G Davis,[1,2,3] Chris Gale,[7,8] Samantha Johnson,[9] Suzannah Kinsella,[10] Marian Knight,[11] Louise S Owen,[1,2,3] Louise Pallot,[6] Trisha M Prentice,[12,13] Patricia Santhanadass,[14] Kayleigh Stanbury,[11] David Tingay,[5,12] Clare L Whitehead,[2,3,15] Brett James Manley,[1,2,3] Charles C Roehr,[11,16,17] Pollyanna Hardy,[11] Steering Group

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For numbered affiliations see end of article.

Correspondence to
Dr Stacey Peart;
stacey.peart@thewomens.org.au

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ABSTRACT

Objective The James Lind Alliance (JLA) Most Premature Babies Priority Setting Partnership aimed to identify the most important areas for research for infants born <25 weeks' gestation.

Design Employing standardised JLA methodology, questions for research were sought from stakeholders via an online survey. Summary questions were formed and checked against existing evidence, with unanswered questions compiled into a second shortlisting survey for prioritisation by stakeholders. A stakeholder consensus workshop was held to determine the top 10 research priorities.

Participants People with lived experience of neonatal intensive care, including parents/carers of preterm infants and adults born preterm, and healthcare professionals caring for preterm infants across Australia, New Zealand and the UK.

Main outcome measure The top 10 research priorities for infants born <25 weeks' gestation.

Results From 844 questions received from the initial survey, 81 summary questions were formed, of which 80 were unanswered and included in the second shortlisting survey. The 19 top-ranked questions were taken to the final prioritisation workshop, where the top 10 research priorities were determined by people with lived experience and healthcare professionals. The most important research question identified was 'What can be done in the neonatal intensive care unit to improve long-term health and developmental outcomes?'. Other important areas for research included antenatal interventions and neonatal care at birth, preventing intraventricular haemorrhages, managing pain, postnatal corticosteroid treatment and supporting families.

Conclusions This study identified the most important areas of research for infants born <25 weeks' gestation, as determined jointly by stakeholders. These findings should be used to guide future research and funding aimed at improving meaningful outcomes for these infants and their families.

WHAT IS ALREADY KNOWN ON THIS TOPIC

- ⇒ The highest risk extremely preterm infants born <25 weeks' gestation have not been well studied. The highest risk extremely preterm infants born <25 weeks' gestation have not been well studied.
- ⇒ To improve outcomes, it is critical to establish the research priorities of people with lived experience and the healthcare professionals who care for them.

WHAT THIS STUDY ADDS

- ⇒ The identification of the most important areas of research for infants born <25 weeks' gestation as determined by key stakeholders.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

- ⇒ These research priorities should be used by researchers, funders and other stakeholders as the basis to guide impactful research to improve meaningful outcomes for infants born <25 weeks' gestation and their families.

INTRODUCTION

Preterm birth (<37 weeks' gestation) accounts for 10% of live births globally and is the leading cause of death in children under 5 years of age.[1] Preterm birth rates are increasing.[2] With advances in neonatal care, survival rates for infants born extremely preterm (<28 weeks' gestation) have improved considerably.[3 4] Consequently, intensive care is offered at earlier gestations.[5–8] However, infants born extremely preterm have worse survival and long-term outcomes compared with more mature infants[9–13]; the incidence being inversely proportional to gestational age at birth.

While there is much research addressing the care of extremely preterm infants, evidence is lacking for the highest risk subset of infants born <25 weeks' gestation. A systematic review of studies published from 2010 to 2020 by Pavlek *et al* demonstrated that only 1% of preterm infants enrolled in randomised controlled trials were <24 weeks' gestation.[14] This poor representation is multifactorial. Infants born <25 weeks' gestation are a relatively small but growing population,

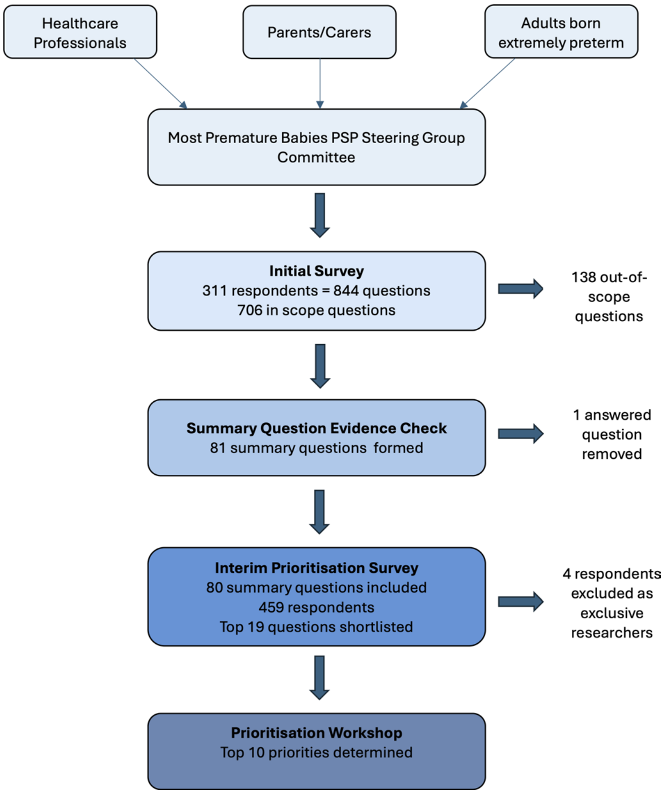

Figure 1 Priority setting partnership flow chart. PSP, Priority Setting Partnership

accounting for 1.5% of preterm births.[15] Poor inclusion rates for this cohort may be due to their exclusion based on the perception that infants born <25 weeks' gestation have in the past been considered non-viable and too high risk. Given the unpredictable and stressful nature of extremely preterm birth, particularly at gestations bordering viability, obtaining prospective consent can be challenging, further limiting the ability to recruit a representative sample of infants <25 weeks' gestation. As fetal development is a continuum, this cohort of infants is embryonically and physiologically different from infants born 25–27 weeks' gestation.[16] Effective and safe treatments in more mature infants cannot be assumed to be equivalently effective at lower gestations. Due to a lack of evidence to guide management in this subset of extremely preterm infants, clinical practices and outcomes vary widely.[5 17–19]

To meaningfully improve the outcomes for infants born <25 weeks' gestation and their families, it is critical to establish the priorities of people with lived experience of extremely preterm birth, as well as healthcare professionals caring for this population. Studies suggest that parents of preterm infants may have different perspectives and priorities regarding neonatal care and outcomes compared with healthcare professionals.[20–24] There is growing recognition of the importance of collaborating with people with lived experience to enable effective translational research.[25–31]

Such joint decision-making underpins the mission of the James Lind Alliance (JLA), a non-profit initiative aiming to bring together patients, carers and healthcare professionals to identify important areas for research.[32 33] The JLA Most Premature Babies Priority Setting Partnership (PSP) is an International Perinatal Research Partnership initiative, a collaboration between the Murdoch Children's Research Institute's Melbourne Children's Trials Centre, Australia, and the University of Oxford's National Perinatal Epidemiological Unit, UK (https://www.npeu.ox.ac.uk/ctu/partnerships).

The aim of the Most Premature Babies JLA PSP was to jointly identify the most important areas for research for infants born <25 weeks' gestation.

METHODS

The Most Premature Babies PSP was conducted in accordance with JLA methodology[34]; two cross-sectional surveys were disseminated, and a consensus workshop held using modified Delphi and nominal group techniques.

Establishing a Steering Committee

A Steering Committee, chaired by a JLA adviser, was formed. Members were identified and invited by consensus of the core project team to collate a representative group of people with varied experience and expertise in preterm birth and research in this area. This comprised 17 members with equal representation from Australia and the UK, including parents/carers of infants born ≤25 weeks' gestation (n=2), adults born ≤25 weeks' gestation (n=2), healthcare professionals (n=9) from medical, nursing and allied health disciplines, a research methodologist, project coordinators (LG, KS) and a project lead/neonatologist (SP).

Scope

Given the broad nature of the topic, the scope of this PSP was limited to the management and care of infants born <25 weeks' gestation during their initial hospital admission, including:

- Antenatal management to improve outcomes.
- Resuscitation practices and provision of intensive care.
- General management during hospital admission.
- Diagnosis and treatment of common conditions affecting this group of infants.
- The impact of inpatient neonatal care on the physical and mental health and well-being of parents/carers of this group of infants.
- The impact of inpatient neonatal care on the health and developmental outcomes of this group of infants.

Topics out of scope included:

- Antenatal treatments to *prevent* preterm birth.
- Causes of premature labour.
- Infants born <22 or ≥25 weeks' gestation.
- The care of infants born <25 weeks' gestation *after* discharge from hospital.
- Infants born <25 weeks' gestation in low- and middle-income countries.

Gathering the uncertainties

An online survey (LimeSurvey, Hamburg, Germany) invited respondents from Australia, New Zealand and the UK to list their top three questions relating to infants born <25 weeks' gestation. Respondents were eligible if they were a parent/carer of a child born preterm, an adult who was born preterm or a healthcare professional caring for preterm infants. Respondents were excluded if they were exclusively involved in research. Respondent demographics were obtained including category of respondent (parent/carer, adult born extremely preterm or healthcare professional), gestational age at birth (as applicable) or healthcare category (as applicable), sex, age bracket, ethnicity, geographic location and highest level of education. The survey ran from 24 October 2022 to 13 March 2023. During this time, it was promoted among people with lived experience and healthcare professionals caring for preterm infants. We

Table 1 Survey 1 respondent demographics

	Overall, n (%)	Healthcare professionals, n (%)	Parents/carers, n (%)	Adults born extremely preterm, n (%)	Other,* n (%)	Unanswered, n (%)
Survey completion	311	210 (67.5)	71 (22.8)	13 (4.2)	14 (4.5)	3 (1)
Sex						
Female	239 (76.8)	147 (70)	67 (94.4)	12 (92.3)	12 (85.7)	2 (66.7)
Male	58 (18.6)	52 (24.8)	3 (4.2)	1 (7.7)	1 (7.1)	0 (0)
Non-binary	1 (0.3)	0 (0)	1 (1.4)	0 (0)	0 (0)	0 (0)
Not answered	13 (4.2)	11 (5.2)	0 (0)	0 (0)	1 (7.1)	1 (33.3)
Location						
Australia/New Zealand	159 (51.1)	103 (49)	42 (59.2)	6 (46.2)	6 (42.9)	2 (66.7)
UK	117 (37.6)	82 (39)	22 (31)	4 (30.8)	8 (57.1)	1 (33.3)
Other	33 (10.6)	23 (11)	7 (10)	3 (23.1)	0 (0)	0 (0)
Not answered	2 (0.6)	2 (1)	0 (0)	0 (0)	0 (0)	0 (0)
Ethnicity						
Caucasian/White British	236 (75.9)	156 (74.3)	57 (80.3)	10 (76.9)	12 (85.7)	1 (33.3)
Indian	18 (5.8)	17 (8.1)	0 (0)	0 (0)	1 (7.1)	0 (0)
Asian	18 (5.8)	16 (7.6)	2 (2.8)	0 (0)	0 (0)	0 (0)
African	6 (1.9)	3 (2.9)	2 (2.8)	0 (0)	0 (0)	1 (33.3)
Aboriginal/Torres Strait Islander	2 (0.6)	0 (0)	2 (2.8)	0 (0)	0 (0)	0 (0)
Māori	2 (0.6)	1 (0.5)	1 (1.4)	0 (0)	0 (0)	0 (0)
Other	14 (4.5)	3 (1.4)	7 (9.9)	3 (23.1)	1 (7.1)	0 (0)
Not answered	15 (4.8)	14 (6.7)	0 (0)	0 (0)	0 (0)	1 (33.3)
Highest education						
Postgraduate degree/higher education	188 (60.5)	159 (75.7)	16 (22.5)	4 (30.8)	8 (57.1)	1 (33.3)
Bachelor’s degree	83 (26.7)	36 (17.1)	34 (47.9)	7 (53.8)	6 (42.9)	1 (33.3)
Graduate or advanced diploma/certificate	29 (9.3)	13 (6.2)	13 (18.3)	2 (15.4)	0 (0)	1 (33.3)
Senior secondary education	5 (1.6)	0 (0)	5 (7)	0 (0)	0 (0)	0 (0)
Did not complete secondary education	2 (0.6)	0 (0)	2 (2.8)	0 (0)	0 (0)	0 (0)
Not answered	4 (1.3)	2 (1)	1 (1.4)	0 (0)	0 (0)	0 (0)

*Includes members or employees of an organisation representing preterm infants and their families.

engaged 15 partner organisations (online supplemental material section 1) supporting preterm infants and their families to assist in disseminating the survey across social media platforms and their networks. It was promoted at neonatal conferences, within neonatal intensive care units and via email among multidisciplinary collegial networks. Promotional material was developed containing a QR code linking to the survey, along with a website (https://www.npeu.ox.ac.uk/most-premature-babies) and social media channels. Representativeness of respondents was monitored and attempts made to increase awareness among under-represented groups. This included increasing engagement with partner organisations and focused social media presence to enhance inclusion of under-represented groups.

Forming summary research questions

All submitted questions were categorised as ‘within scope’ or ‘out of scope’ by the project lead. Within scope, questions with common themes were grouped together to form a summary research question: a clear, easily understood question about a broad topic which accurately reflected the content received from respondents, rather than a specific research question. All summary research questions were reviewed and agreed on by consensus of the Steering Committee. An approach towards keeping summary research questions broad and inclusive was taken in preference to multiple specific research questions.

All summary research questions underwent an evidence check to determine if they had been adequately answered by the current literature. This search was limited to full-text systematic reviews published in English within the last 10 years (online supplemental material section 2). The search was conducted between June and August 2023 across MEDLINE, EMBASE, CINAHL and the Cochrane Database of Systematic Reviews. After screening all titles and abstracts, the remaining relevant full texts were analysed. Questions were considered answered if they were addressed in a systematic review with moderate to high-quality evidence, and where the population of interest was infants born <25 weeks’ gestation. In cases where a summary question was addressed within the literature, but with clearly stated low-quality evidence, the summary question was considered unanswered. Questions were considered partially answered if there was either high-quality evidence in the overall population of extremely preterm infants but not focused on infants born <25 weeks’ gestation or conflicting evidence. If it was unclear whether the summary question was adequately answered, the question and relevant references were reviewed by the Steering Group for consensus. Unanswered and partially answered questions were included in the interim prioritisation survey, with answered questions excluded.

Interim prioritisation

A second online survey (LimeSurvey) invited respondents to select their top 10 priorities from the remaining summary research questions. This involved a two-stage process: (1) selecting all summary research questions that were of interest and (2) selecting from that shortlist their top 10 most important summary research questions. The order of questions presented to

Table 2 Prioritisation survey respondent demographics

	Overall, n (%)	Healthcare professionals, n (%)	Parents/carers, n (%)	People with lived experience, n (%)	Other, n (%)	Unanswered, n (%)
Survey completion	455	304 (66.8)	104 (22.9)	37 (8.1)	10 (2.2)	0
Sex						
Female	383 (84.2)	241 (79.3)	99 (95.2)	34 (91.9)	9 (90.0)	
Male	68 (14.9)	60 (19.7)	5 (4.8)	2 (5.4)	1 (10.0)	
Non-binary	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	
Not answered	4 (0.9)	3 (1)	0 (0)	1 (2.7)	0 (0)	
Location						
Australia/New Zealand	111 (24.4)	77 (25.3)	23 (22.1)	7 (18.9)	4 (40.0)	
UK	265 (58.2)	164 (53.9)	75 (72.1)	21 (56.8)	5 (50.0)	
Other	79 (17.4)	63 (20.7)	6 (5.8)	9 (24.3)	1 (10.0)	
Not answered	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	
Ethnicity						
Caucasian or White British	385 (84.6)	252 (82.9)	93 (89.4)	33 (89.2)	7 (70.0)	
Asian	32 (7.0)	30 (9.9)	0 (0)	1 (2.7)	1 (10.0)	
African or Black British	8 (1.8)	4 (1.3)	4 (3.8)	0 (0)	0 (0)	
Aboriginal/Torres Strait Islander	2 (0.4)	0 (0)	2 (1.9)	0 (0)	0 (0)	
Māori	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	
Other	13 (2.9)	5 (1.6)	4 (3.8)	3 (8.1)	1 (10.0)	
Not answered	15 (3.3)	13 (4.3)	1 (1.0)	0 (0)	1 (10.0)	
Highest education						
Postgraduate degree	253 (55.6)	204 (67.1)	31 (29.8)	12 (32.4)	6 (60.0)	
Bachelor's degree/ higher education	149 (32.7)	83 (27.3)	45 (43.3)	18 (48.6)	3 (30.0)	
Graduate or advanced diploma/certificate	22 (4.8)	13 (4.3)	7 (6.7)	2 (5.4)	0 (0)	
Senior secondary education	26 (5.7)	1 (0.3)	13 (12.5)	3 (8.1)	1 (10.0)	
Did not complete secondary education	0 (0)	0 (0)	8 (7.7)	0 (0)	0 (0)	
Other	2 (0.4)	1 (0.3)	0 (0)	1 (2.7)	0 (0)	
Not answered	3 (0.7)	2 (0.7)	0 (0)	1 (2.7)	0 (0)	

respondents was randomly assigned to reduce bias. This survey was promoted and shared in a similar fashion to the first survey and ran from 18 September 2023 until 10 November 2023.

All summary research questions were ranked overall according to the number of times a question appeared in a respondent's top 10 selection (ie, the question with the highest rank was assigned the highest score). To ensure equal weight was given to all respondent categories and not skewed by the largest represented group, questions were ranked according to separate respondent category. The individual ranks for each question from each respondent category were then added together to obtain an overall rank. Where response counts were equal, questions were given equal rank and score. The top 19 ranked summary research questions were included in the priority setting workshop. This number[19] was decided a priori due to this being the maximum number of questions able to be visualised with ease on a screen.

Priority setting workshop

The priority setting workshop was held, bringing together people with lived experience and healthcare professionals from Australia, New Zealand and the UK in an open forum to determine the final top 10 priorities. Given time zone constraints, this workshop was conducted online in two 3-hour sessions held on 30 November and 1 December 2023. Involvement in the workshop was sought from respondents who indicated interest when participating in the interim prioritisation survey, as well as via social media and word of mouth promotion. The final list of participants was reviewed and agreed on by the Steering Group ensuring balanced representation of all stakeholders. All people with lived experience were financially remunerated for their participation. Participants were provided with the shortlisted questions prior to the workshop and asked to rank their top three and bottom three questions. During the workshop, participants were divided into four breakout groups, with balanced representations from all stakeholders and geographic locations in each group and ensuring that people with lived experience were not grouped with a healthcare professional involved in their clinical care. Groups worked together through three discussion and ranking exercises, interspersed with feedback from the other groups. Discussions were held to determine a top 10 list of priorities for each group, which were then shared among all participants at the end of the first workshop day. The following day, groups were reallocated, again ensuring balanced representation, to promote diversity of perspectives and robust discussion. The process of discussing and agreeing by consensus a top 10 list of priorities was repeated. All groups' results were combined to determine the overall top 10 priorities, with the results shared and agreed among all participants at the end of the workshop.

RESULTS

The results from the initial survey, evidence check and prioritisation survey are summarised in figure 1.

The surveys and summary questions can be found at https://www.jla.nihr.ac.uk/priority-setting-partnerships/the-most-premature-babies.

Table 3 Top 19 weighted ranked questions from the interim prioritisation survey according to respondent categories

Question	Combined rank*	Healthcare professional position	Parent/carer position	Adult born extremely preterm position
What is the best way to manage pain and distress in babies?	1	1 (=)	2 (+1)	Joint 2 (+1)
What can be done in the neonatal intensive care unit to improve long-term health and developmental outcomes?	2	2 (=)	3 (+1)	1 (−1)
How can we improve the care of babies at delivery and in the hours after birth to improve outcomes?	3	4 (+1)	Joint 5 (+2)	Joint 2 (+1)
Should intensive care be offered to babies born at 22 and 23 weeks' gestation?	Joint 4	6 (+2)	7 (+3)	5 (+1)
What is the best way to prevent brain injury, including intraventricular haemorrhages (bleeding in the brain)?	Joint 4	3 (−1)	6 (+2)	Joint 9 (+5)
Does reducing sensory stimulation (lights, noise, touch) in the neonatal intensive care unit improve outcomes, and how can this best be achieved?	5	7 (+2)	10 (+5)	Joint 3 (−2)
What are the long-term outcomes and how are they best predicted?	6	Joint 10 (+4)	Joint 9 (+3)	Joint 3 (−3)
What interventions or treatments during pregnancy and labour can improve outcomes?	7	20 (+13)	1 (−7)	4 (−3)
What is the best way for families to be more involved in the care of their baby during their hospital admission?	8	14 (+6)	Joint 11 (+3)	6 (−2)
Should formal neurological assessments be routinely performed?	9	Joint 15 (+6)	Joint 9 (=)	Joint 8 (−1)
How can antenatal counselling be improved for families facing the possible delivery of a baby before 25 weeks' gestation?	Joint 10	Joint 15 (+5)	8 (−2)	Joint 10 (=)
How can infant mental health be improved?	Joint 10	Joint 11 (+1)	15 (+5)	7 (−3)
How can mothers be best supported in breastfeeding and expressing milk?	11	Joint 18 (+7)	4 (−7)	Joint 12 (+1)
What is the best way to position babies in the first few days after they are born to improve outcomes?	12	5 (−7)	Joint 18 (+6)	Joint 12 (=)
Does family-centred care (working together with families to provide care and make decisions for their baby) improve outcomes?	Joint 13	Joint 15 (+3)	Joint 14 (+1)	Joint 7 (−6)
What are the best ways to communicate and support difficult discussions (including end of life decisions) in the neonatal intensive care unit?	Joint 13	Joint 16 (+3)	Joint 13 (=)	Joint 7 (−6)
What impact does having a baby born less than 25 weeks' gestation have on parental/family mental and physical health?	Joint 14	Joint 25 (+9)	Joint 5 (−10)	Joint 8 (−7)
How can families be best supported during their baby's neonatal intensive care admission?	Joint 14	Joint 16 (+2)	Joint 11 (−3)	Joint 11 (−3)
What is the most effective postnatal steroid treatment regimen to improve outcomes?	15	8 (−7)	Joint 18 (+3)	Joint 13 (−3)

Position remained in the same rank (=). Position increased in rank (+). Position decreased in rank (−).
Rank 1–10 🟩. Rank 11–15 🟨. Rank 16–20 🟥.
*Combined ranked questions weighted equally between the respondent groups.

Initial survey

From the initial survey, there were 311 respondents, yielding 844 questions, of which 706 (83.6%) were within scope. Respondent characteristics are summarised in table 1. Healthcare professionals constituted the majority of respondents (67.5%), with 51.1% of respondents overall from Australia or New Zealand. There was greater representation of females (76.8%), Caucasians (75.9%) and those with tertiary-level education (96.5%) across all respondent categories. Of respondents with lived experience for whom data were available, 69/84 (82%) were born, or had a child born, at ≤25 weeks' gestation.

Evidence check

A total of 81 summary research questions underwent an evidence check, after which 80 remained unanswered and were included in the prioritisation survey (online supplemental material section 3). The summary question regarding survival rates of infants born <25 weeks' gestation met criteria to be considered answered.[5 13 18 35]

Prioritisation survey

The second survey received 459 responses, of which four were excluded as they came from respondents who were exclusively researchers, leaving 455 responses for analysis. Table 2 summarises the respondent demographics. Most (66.8%) respondents were healthcare professionals and from the UK (58.2%). Again, there was greater representation of females (84.2%), Caucasians (84.6%) and those with tertiary-level education (93.1%) across all respondent categories. Of respondents with lived experience for whom data were available, 66/136 (49%) were born, or had a child born, at ≤25 weeks' gestation. The top 19 highest combined ranked questions, weighted equally according to respondent category, are presented in table 3.

Priority setting workshop

The final workshop was attended by 30 participants, including 10 parents/carers, 6 adults born extremely preterm and 14 healthcare professionals with equal representation from Australia/New Zealand and the UK. There were four Steering Group members observing and four JLA facilitators leading the breakout groups. Of the participants with lived experience for whom data were available, 9/14 (64%) were born, or had a child born, at ≤25 weeks' gestation. The final top 10 research priorities are presented in table 4. The most important research question identified was 'what can be done in the neonatal intensive care unit to improve long-term health and developmental outcomes?'. Other important areas for research included antenatal interventions and neonatal care at birth, preventing intraventricular haemorrhages, managing pain, postnatal corticosteroid treatment and supporting families. There were three questions from the prioritisation survey top 10 list which did not make it into the final workshop top 10 (online supplemental material section 4).

Table 4 Final ranking including top 10 questions

Rank	Question	Group 1	Group 2	Group 3	Group 4
1	What can be done in the neonatal intensive care unit to improve long-term health and developmental outcomes?	1	1	1	1
2	How can we improve the care of babies at delivery and in the hours after birth to improve outcomes?	2	2	2	2
3	What is the best way to prevent brain injury, including intraventricular haemorrhages (bleeding in the brain)?	4	3	5	4
4	What interventions or treatments during pregnancy and labour can improve outcomes?	6	5	4	5
5	What are the long-term outcomes and how are they best predicted?	3	4	11	3
6	What is the best way to manage pain and distress in babies?	7	6	6	6
7	What is the most effective postnatal steroid treatment regimen to improve outcomes?	8	10	3	7
8	What is the best way for families to be more involved in the care of their baby during their hospital admission?	12	7	7	8
9	How can antenatal counselling be improved for families facing the possible delivery of a baby before 25 weeks' gestation?	9	8	10	9
10	How can families be best supported during their baby's neonatal intensive care admission?	5	11	13	11
11	What impact does having a baby born less than 25 weeks' gestation have on parental/family mental and physical health?	10	9	12	10
12	How can mothers be best supported in breastfeeding and expressing milk?	11	14	8	15
13	What are the best ways to communicate and support difficult discussions (including end of life decisions) in the neonatal intensive care unit?	16	13	9	12
14	Does reducing sensory stimulation (lights, noise, touch) in the neonatal intensive care unit improve outcomes, and how can this best be achieved?	13	12	14	13
15	Should intensive care be offered to babies born at 22 and 23 weeks' gestation?	14	15	15	14
16	Does family-centred care (working together with families to provide care and make decisions for their baby) improve outcomes?	17	16	16	16
17	What is the best way to position babies in the first few days after they are born to improve outcomes?	15	17	17	17
18	How can infant mental health be improved?	18	18	18	18
19	Should formal neurological assessments be routinely performed?	19	19	19	19

Rank 1–10 🟩. Rank 11–15 🟨. Rank 16–20 🟥.

DISCUSSION

This PSP identified the most important questions for research in infants born <25 weeks' gestation as determined through partnership between people with lived experience and healthcare professionals. To our knowledge, this is the first JLA PSP of its kind to address this high-risk population and is unique in encompassing the parent–infant dyad rather than a specific disease. Given their significant rates of mortality and adverse health and developmental outcomes,[5,13,17,18,35] infants born <25 weeks' gestation are an under-researched group. The limited current evidence for their clinical management is largely either extrapolated from studies in the broader group of extremely preterm infants or based on heterogenous observational studies alone. The observation that 80/81 (98.8%) summary questions were unanswered in the current available evidence supports this.

There was significant overlap of shared priorities between people with lived experience and healthcare professionals, as evidenced by the similarities in the respondent-weighted top 19 ranked questions from the interim prioritisation survey (table 3). In particular, the top seven ranked questions were the same, although in a different order. All respondent groups had a similar number of their top 19 ranked questions included in the final workshop, including their highest ranked priorities. The discriminative values between the top 19 questions were small, often with only one response separating them, suggesting that they are all important priorities to stakeholders. It is necessary to highlight the differences in priorities between people with lived experience and healthcare professionals. The strong theme among healthcare professionals' priorities was clinical management of the infant, while people with lived experience voiced themes of parental and family support. This likely speaks to what is meaningful and impactful at an individual level. The common priority of research relating to long-term outcomes reflected by all respondent categories likely resonates as a common experience both in the overarching aim of clinical management and the daily life of a person with lived experience.

At the workshop, there was a high degree of consensus between groups to determine the top 10 priorities. The final order of priorities may be attributed to the preference given to questions which encompassed a 'big picture' perspective rather than more specific treatments or outcomes. Notably, several questions regarding family impact, support and involvement were ultimately ranked much higher after the final prioritisation workshop. This highlights the crucial importance and validity in involving people with lived experience in research development to ensure that meaningful areas of research are addressed. While the top research priority is broad in nature, it emphasises the necessity of understanding and improving long-term outcomes for this population.

The range of questions included in the top 10 priorities reflects the complex and nuanced needs of infants <25 weeks' gestation and their families. Compared with other PSPs, including the preterm birth PSP[36] in which the top 10 priorities were focused 'PICO' questions addressing specific clinical management topics, the top 10 priorities of this PSP are holistic and varied, encompassing several challenges faced by the most preterm infants and their families during their intensive care journey and beyond. These findings are in keeping with a systematic review by Webbe *et al*[24] demonstrating the differences in priorities between people with lived experience and healthcare professionals in the neonatal setting, with important outcomes extending beyond specific disease states to the global status of the child and family. The nature of these research priorities also emphasises the relevance of alternative research methodologies needed to address these important topics.

The strengths of this study include its standardised, reproducible methodology, the large number of respondents and international collaboration with stakeholders at all stages of the process.

However, the findings are limited in their generalisability as this PSP was limited to high-income settings and targeted to English-speaking respondents from Australia, New Zealand and the UK. Males and people of Indigenous and non-Caucasian ethnicity were under-represented. Given these limitations in diversity, caution should be exercised if implementing research based on these priorities that includes non-represented populations whose values may differ. Future efforts should aim to focus on a more global perspective. Despite best efforts to engage with all key stakeholders, there was greater representation from healthcare professionals; however, this imbalance was mitigated by ranking responses according to respondent category to ensure equal weighting. Within healthcare professionals, there was representation from medical, nursing and allied health professionals.

In conclusion, this PSP determined a top 10 list of research questions aimed at improving the care and outcomes of infants born <25 weeks' gestation, relevant to both people with lived experience and healthcare professionals. These topics should be used as a basis on which to develop focused research questions amenable to study. Researchers are encouraged to use these results as a voice for advocacy when commissioning funding bodies to implement studies that improve relevant and meaningful outcomes for the most premature infants and their families.

Author affiliations
[1]Newborn Research Department, The Royal Women's Hospital, Melbourne, Victoria, Australia
[2]Clinical Sciences, Murdoch Children's Research Institute, Melbourne, Victoria, Australia
[3]Department of Obstetrics, Gynaecology and Newborn Health, The University of Melbourne, Melbourne, Victoria, Australia
[4]Gloucestershire Hospitals NHS Foundation Trust, Cheltenham, UK
[5]Murdoch Children's Research Institute, Melbourne, Victoria, Australia
[6]Consumer Advisory Group, Murdoch Children's Research Institute Centre of Research Excellence in Newborn Medicine, Melbourne, Victoria, Australia
[7]Academic Neonatal Medicine, Imperial College London, London, UK
[8]Centre for Paediatrics and Child Health, Imperial College London, London, UK
[9]Department of Population Health Sciences, University of Leicester, Leicester, UK
[10]James Lind Alliance, National Institute for Health and Care Research, Evaluation Trials and Studies Coordinating Centre, Southampton, UK
[11]National Perinatal Epidemiological Unit, Clinical Trials Unit, Oxford Population Health, University of Oxford, Oxford, UK
[12]Neonatal Services, Royal Children's Hospital, Melbourne, Victoria, Australia
[13]Department of Paediatrics, University of Melbourne, Melbourne, Victoria, Australia
[14]not applicable, Oxford, UK
[15]Department of Maternal Fetal Medicine, The Royal Women's Hospital, Melbourne, Victoria, Australia
[16]Faculty of Health Sciences, University of Bristol, Bristol, UK
[17]Newborn Services, Southmead Hospital, Bristol, UK

X Stacey Peart @Stacey_H_84, Rosemarie Anne Boland @roseandtoby, Chris Gale @DrCGale, Marian Knight @Marianfknight and Brett James Manley @drbretty

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Collaborators Steering Group: 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; Dr Olivia Ray, Gloucestershire Hospitals NHS Trust, UK. Clinical representative/s: Professor Charles Roehr, Neonatologist, Southmead Hospital, Bristol UK; Dr Clare Whitehead, Obstetrician, The Royal Women's Hospital, Melbourne, Australia; Professor Brett Manley, Neonatologist, The Royal Women's Hospital, Melbourne, Australia; Professor David Tingay, Neonatologist, Murdoch Children's Research Institute, Melbourne, Australia; Professor Chris Gale, Neonatologist, Imperial College London, London, UK; Dr Trisha Prentice, Bioethicist and Neonatologist, The Royal Children's Hospital, Melbourne, Australia; Professor Marian Knight, Professor of Maternal and Child Population Health, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford UK; Associate Professor Rosemarie Boland, Neonatal Nurse, Murdoch Children's Research Institute, Melbourne, Australia. Methodology: Associate Professor Pollyanna Hardy, Statistician, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford, UK; Professor Samantha Johnson, Developmental Psychologist, University of Leicester, Leicester, UK. Project coordinators: Ms Laura Galletta, Murdoch Children's Research Institute, Melbourne, Australia; Ms Kayleigh Stanbury, University of Oxford National Perinatal Epidemiology Unit (NPEU) Clinical Trials Unit, Oxford, UK. Project lead: Dr Stacey Peart, Newborn Research, The Royal Women's Hospital, Melbourne, Australia; JLA adviser: Ms Suzannah Kinsella, James Lind Alliance.

Contributors PH, CCR, DT, BJM, LG and SP conceived the study and set up the Steering Group. All Steering Group members contributed to protocol development, survey design and summary question review. SP analysed the data and undertook the evidence checking process. SK facilitated as a James Lind Alliance advisor. PGD and LSO acted as PhD supervisors for SP and were not members of the Steering Committee. First, second and senior authorship was determined by contribution and all other author positions alphabetical as agreed by all authors. SP, DT, BJM, PH, CG, TMP, LG, AB and OR drafted the manuscript. All authors revised the manuscript and approved the final manuscript as submitted. SP is responsible for the overall content as guarantor. CCR and PH are joint last authors

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Author note Person with lived experience representative: OR, AB, LP, PS. Co-second author: OR, LG.

ORCID iDs

Stacey Peart http://orcid.org/0009-0005-2989-7071
Rosemarie Anne Boland http://orcid.org/0000-0002-8127-2133
Peter G Davis http://orcid.org/0000-0001-6742-7314
Chris Gale http://orcid.org/0000-0003-0707-876X
Marian Knight http://orcid.org/0000-0002-1984-4575
Louise S Owen http://orcid.org/0000-0002-9490-6377
Trisha M Prentice http://orcid.org/0000-0003-4271-7950

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