Identifying research priorities in preterm birth

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For the last six years, the James Lind Alliance (JLA) has been at the forefront of a quiet revolution in the way research priorities are set[1,2]. Rather than allowing the drug industry or pure researchers, who may never see patients, to decide the research agenda, the JLA enables patients/carers and clinicians to identify and prioritise what matters to them. To date it has facilitated partnerships in nine conditions, from asthma to schizophrenia, and there are nine more underway[3], each adhering to a key JLA principle: that priority setting partnerships (PSPs) include equal input from patients/carers and clinicians.

The JLA was set up in the wake of growing interest in patient and public involvement (PPI), notably the commitment made by the Medical Research Council (MRC) in 2003[4], to involve "patients/consumers in all aspects of the clinical trials it funds." The MRC turned this commitment into action, jointly funding the JLA from its inception, alongside the National Institute for Health Research (NIHR).

While its roots and support lie firmly in the mainstream, some traditionalists have been sceptical about the value of the JLA method. As one researcher challenged: "Why should patients have any useful opinions about what directions research should take?" Emma Halls (now Emma Malcolm), CEO of Prostate Action wrote an article for the *BMJ* entitled 'Where are the clinicians when you need them?'[5]. She said "My frustration is growing and the *BMJ* seems a good place to ask doctors: 'Why the lack of communication? Where is your culture of collaboration in my field of prostate cancer? Do you feel you have your own show, and all the answers?"

Despite this rather desperate plea, the prostate cancer PSP completed its work in the average 18 months, and an impressive amount of activity has followed. To date, this comprises no fewer than fourteen funded research projects and the inclusion of a question about GP awareness of prostate cancer built into Prostate Action's existing GP masterclass.

Dr Vincent Gnanapragasam, from the University of Cambridge and Addenbrooke's Hospital, said of this PSP "Being part of both the research and clinical communities made me realise that in many instances what was being researched is far removed from any implementation in patients. Most recent major progress in prostate cancer treatment and survival has come from good clinical studies which have at their centre the patient. Getting the patient view on research priorities therefore seemed a crucial element that had been missing. I think the true test of what the Prostate PSP has achieved will be in seeing if more funding bodies implement the priorities we have produced."

A PSP in preterm birth is now underway as part of a broad programme of research funded by the NIHR, called 'Improving quality of care and outcome at very preterm birth'[6]. Convened by the Social Science Research Unit (SSRU) at London University's Institute of Education (IoE) it is now gathering uncertainties via a ten member steering group, four of whom represent services users and six clinicians.

With 26 participating organisations involved and a target of agreeing a list of uncertainties and prioritising the 'top 10' by November, this PSP has its work cut out. Zoë Chivers, Innovations Manager at Bliss, a charity providing support and care for premature babies and their families and a member of the steering group points out "Bliss is delighted to have the opportunity to be involved with the PSP. Looking at the priorities before birth, whilst their baby is on the unit and post discharge, gives parents the opportunity to identify areas of research and where there are gaps in the service. Parents are often able to see things more objectively and have different motivations from health professionals when identifying areas of research. We are looking forward to working with the JLA to review the priority areas that casting such a wide net might bring."

In the next eight months, the PSP will publicise the survey being used to gather uncertainties (https://www.surveymonkey.com/s/prembabies) to both the patient and professional communities, and then work with information specialists to sift and categorise the questions received and ensure that they are true unknowns, not answerable by current systematic reviews or clinical studies.

The next step in the JLA process is to work with the UK Database of Uncertainties about the Effects of Treatments (UK DUETs; http://www.library.nhs.uk/duets/) where uncertainties are gathered and tagged to indicate the type of research needed to address them before finally being prioritised by the PSP.

Programme leader Lelia Duley, Professor of Clinical Trials Research at the Queen's Medical Centre in Nottingham explains why she feels this approach to be so valuable: "One in every 100 UK babies is born before 32 weeks, often needing help with breathing, feeding and other essentials. Those who survive may spend months in hospital, and suffer childhood ill health or

disability. We are increasingly aware that clinical research often fails to address the questions about treatments that are of greatest importance to patients, their carers, and their clinicians. Resources for research are limited, and so we need to make sure that the research we do answers questions that are important to clinicians who care for babies born preterm, and their families."

Professor Duley's interest in using the JLA method to bring patients and clinicians together to identify and prioritise treatment uncertainties lies in part in its pragmatic stance. As she says "The current routine practice of immediate clamping of the umbilical cord and removal of the baby to a resuscitaire at the side of the room may be more distressing for the mother and her partner than if initial care were provided at the bedside, allowing parents to share the first minutes of their baby's life. The fact is, we simply don't know whether the standard care of immediate cord clamping is best. Indeed, small randomised trials suggest that clamping within 30 seconds may have disadvantages; most specialists think more trials are needed and this chimes well with informal consultation with service users who have identified timing of cord clamping as ripe for research."

This PSP will thus identify and prioritise research gaps for preterm birth, with a broad scope agreed that takes the work much wider than interventional research, to include antenatal and causal issues for preterm birth, long-term follow-up of preterm babies and family support. During the run up to this partnership it has been found that neonatologists are as interested in preventing preterm births as exploring more about treating premature babies. That said, the PSP leaders suspect that alongside studies of the effects of interventions to prevent or follow preterm birth, studies about its cause and people's experiences will emerge as important.

As Sally Crowe, who chairs both this PSP and the JLA Monitoring and Implementation Group, says "It's certainly an unusual PSP, reflected in the range of participating organisations, which we hope will give it a unique strength. This is a different way of establishing research priorities and while we have seen with other PSPs that not everything that emerges is new, we always see things that are, and, vitally, that are shared between those at the sharp end."

While the PSP itself is funded by NIHR, it is thought likely that it will identify research priorities that relate to a broad range of NIHR research programmes and also to medical research charities which are not only increasingly involved in PSPs themselves, but use their outcomes to guide their work. As Sharmila Nebhrajani, Chief Executive of the Association of Medical Research Charities (AMRC) says "Given that charities fund one third of all public medical research in the UK, and that this money is mainly donated by patients, carers and their families, this scale of funding means that the research patients value should be prioritised."

The power of the JLA in turning priorities into research reality is set out by Sandy Oliver, Professor of Public Policy at the IoE, who says "Research funders listen to the James Lind Alliance so this is an opportunity for people with personal or professional experience of preterm birth to ask for research to address what they want to know."

To achieve all these aims, the wide-ranging but simple survey aims to ensure that all ideas, from clinical and family perspectives, are captured. As Crowe says "This can include questions about possible causes of preterm birth or about care before or during preterm birth, or even much later for families at home. The survey is a 'co-production' by the key players who, after considering all the permutations, decided not to narrow down the options, but allow people to contribute all their ideas and especially the ones that don't fit our normal understanding of research questions."

Crowe is right to highlight the potentially unconventional nature of what may emerge from this PSP. Returning to the JLA's roots, one of the pieces of work that led to its establishment was that by Tallon et al[7] who wanted to see whether there was a mismatch between available research evidence and the research preferences of consumers on interventions for the treatment of osteoarthritis of the knee joint. They searched published and unpublished studies to assess the evidence base, and also carried out focus groups and a postal survey of patients. They found a research agenda dominated by studies of pharmaceutical interventions, while 36% of survey respondents ranked knee replacement as the highest priority for research, and 21% chose education and advice as their first choice.

Similarly a recent editorial in *Nature* written by those at the helm of the schizophrenia PSP stated: "Although the purpose of the JLA process is to enable patients and those who treat them to have a say in what gets studied, it can also change clinical practice. For example, sexual dysfunction caused by antipsychotic medication emerged as a key patient priority. This is typically a low priority for clinicians prescribing medication and for companies assessing drug effectiveness. Without the experience of the JLA process, it is unlikely that this issue would have been afforded as much weight as it was[8]."

It is only by consulting those at the sharp end of illness or disability that we can be truly sure what matters most to them. As Lester Firkins, JLA Chair points out "That this is not normal practice is arrogant and irresponsible… those in research who fear the involvement of 'lay' people need to ask themselves why."

Firkins throws down quite a gauntlet. But as he and the rest of the JLA team prepare to transfer the Alliance's work to the NIHR Evaluation, Trials and Studies Coordinating Centre (NETSCC) from April 2013[9], they are encouraged by the way in which a small and potentially risky enterprise has caught the attention and approval of so many, grown so strong, and now looks set to continue its work right at the heart of the UK's health research community.

References

1. **Partridge N., Scadding J.** The James Lind Alliance: patients and clinicians should jointly identify their priorities for clinical trials. *Lancet* 2004;364:1923-24.
2. **Petit-Zeman S., Firkins L., Scadding J.W.** The James Lind Alliance: Tackling research mismatches *Lancet* 2010;376:667-69.
3. http://www.lindalliance.org/Priority%20Setting % 20Members.asp
4. **Medical Research Council.** *Clinical Trials for Tomorrow*. London: MRC. 2003.
5. **Halls E.** Where are the clinicians when you need them? *BMJ* 2010;340:c1845.
6. www.nottingham.ac.uk/pretermbirth
7. **Tallon D., Chard J., Dieppe P.** Relation between agendas of the research community and the research consumer. *Lancet* 2000;355:2037-40.
8. **Lloyd K., White J.** Democratizing clinical research. *Nature* 2011;474:277-78.
9. http://www.netscc.ac.uk/news/item/21032012.asp

To participate in the survey go to: *https://www.surveymonkey.com/s/prembabies* today and help to make a difference. Deadline for responses 15th July 2012.