OPEN LETTER

Parental involvement in a multidisciplinary PhD programme in neonatal brain injury [version 1; peer review: 2 approved with reservations]

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Abstract
Parental and patient and public involvement (PPI) involvement is a core element of the Neonatal Brain Consortium Ireland (NBCI) since its inception. PPI representatives were critical to the development of the Consortium and the animations for parent information as well as the NEPTUNE Neonatal Encephalopathy PhD programme in which they are core members involved in PhD supervision, publications, study days and educational outreach. Key outputs have also included national clinical guidelines and parent information.

Keywords
neonatal encephalopathy, PPI, newborn

This article is included in the Public and Patient Involvement collection.
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Competing interests: No competing interests were disclosed.

Grant information: Health Research Board Ireland [CDA-2018-008] The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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How to cite this article: Molloy EJ, Daly M, Ryan P et al. Parental involvement in a multidisciplinary PhD programme in neonatal brain injury [version 1; peer review: 2 approved with reservations] HRB Open Research 2020, 3:40 https://doi.org/10.12688/hrbopenres.13009.1

First published: 25 Jun 2020, 3:40 https://doi.org/10.12688/hrbopenres.13009.1
Rationale, aim, scope of project

Neonatal brain injury is a common cause of mortality and disability. Neonatal encephalopathy (NE) is one of the commonest causes of neonatal brain injury in full term infants. For every baby that dies from NE, another will survive with significant lifelong disability. A recent systematic review estimated that in 2010, 1.15 million babies developed neonatal encephalopathy directly related to intrapartum asphyxia with 287,000 deaths, 233,000 infants surviving with moderate/severe disability and 181,000 living with mild impairment. This represents a massive global burden of disease, as these children develop their injury at the very beginning of life. It is estimated that worldwide, NE leads to 50.2 million DALYS each year. A recent Health Research Board Ireland (HRB) Collaborative Doctoral Award to researchers in our Neonatal and Children’s Brain Consortium Ireland (NBCI) has shown that even infants with mild encephalopathy can have cognitive impairments at 5 years of age. There is an urgent need to study this population in greater detail and to establish the cause of brain injury so that ultimately, it can be prevented. In this project we aimed to create a unique national collaborative multidisciplinary research group including parents to optimise the investigation and management of neonatal brain injury. All babies with brain injury routinely get an magnetic resonance imaging (MRI) scan and brain monitoring with an electroencephalogram (EEG) as well as detailed developmental progress. We planned to develop unique expertise in these techniques as well as newer advanced methods. The joint supervision of the PhD students allows a new generation of expertise to be developed in Ireland and join international groups to allow integrated care and further research progress in the future. Researchers in this consortium have internationally recognized multidisciplinary expertise in neonatology, paediatrics, neurodevelopment, family-centred care, clinical trials and methodology, pharmacology, epidemiology, biostatistics, translational research and neuroimaging in neonatal brain injury. PhD students experience the holistic overview of research in this area involving the entire translational paradigm from basic science research, translational clinical research, clinical trials to epidemiology and population health while getting in depth expertise in their chosen area.

Aims

Neonatal brain injury has a multifactorial aetiology and causes significant neurological morbidity such as cerebral palsy. The only treatment available is therapeutic hypothermia (TH) for term infants with encephalopathy but morbidity and mortality rates remain high. There is an urgent need for adjunctive therapies to improve neurodevelopmental outcomes. This project aims to combine the international expertise of a multidisciplinary group to improve the outcomes of infants with Neonatal Brain injury and strengthen collaborative links including neonatal neurointensive care and followup.

Implementation

The resulting HRB Neonatal Encephalopathy PhD Training Network (NEPTuNE) comprises five PhD projects. The PPI group were involved as follows: student interview process, active members of the Steering Committee, regular participants in Study Days and advocates for the project at other meetings. Each PPI member is connected to one of the five PhD projects as a member of the supervisory team. This involvement has had a significant impact on the direction of our research, from the initial input to the funding application, selecting motivated students, further development of projects during collaborative Study Days and strategic focus for NEPTuNE and the NBCI. PPI members have also been involved in creating two animations for parents on therapeutic hypothermia and also on the Neonatal Intensive care unit. These meetings were helpful to allow the entire research team to understand the concerns of parents. We discussed each project as well as the motivation for all participant’s involvement and planned outputs from discovery to clinical guideline implementation and family information.

Evaluation/impact

We are particularly proud of the amazing involvement of all team members and the generosity of the PPI group in attending the clinical workshops in MRI, EEG, Clinical evaluation, core outcome data sets and also in interviewing the PhD candidates. This has resulted in an open discussion on all aspects of the projects relating to families and enhances plans for dissemination. In particular we are revising some of the consent and information forms for routine MRI in the NICU. We found having a group who communicate well with us and each other is ideal and it means the same parents are not obliged to contribute constantly. This also increases the sustainability of the group.
We have held an annual national Neonatology meeting for 10 years which has a morning session hosted by the National Parent Organisation (INHA) and an afternoon scientific session targeting medical health care professionals with international guest speakers to both events. We also ensure that at every workshop and meeting a PPI representative will give a presentation and is involved in the programme development.

Lessons learnt
Initially we were concerned that PPI involvement would inadvertently have a coercive element and place an extra burden on families. However by communicating by email, phone and with occasional face to face meetings after the first few introductions we have managed to involve sites all over Ireland with less impact on all collaborator time. In addition the project co-ordinator organises the teaching and communication between the group. Increased funding for meetings and opportunities to financially compensate the PPIs (all of whom work on a voluntary basis for the INHA) would enhance integration of families.

Future directions
We plan a series of papers on family reflections for the journal Pediatric Research written by the PPI group as well as ongoing guideline development and parent information with the Royal College of Physicians of Ireland. We are also co-writing a multidisciplinary paper with core PPI involvement related to the importance of sleep and circadian rhythms in the NICU as they are also experienced in publishing. We will start a PPI forum every 6 months to the executive committee to get valuable feedback and updates on appropriate involvement on the project. We have strong interdisciplinary links in Europe with the European Society for Paediatric Research (ESPR) and the European Foundation for the Care of Newborn infants (ECFNI) which is the first pan-european organisation and network to represent the interests of preterm and newborn infants and their families. We also hope to develop Family centred care in Neonatology and also advance neonatal neurointensive care in collaboration with our international partners in Johns Hopkins University, USA. This project has potential to be extended to preterm infants. Preterm infants are also at high risk of brain injury particularly cerebral palsy and neurodevelopmental delay.

Key messages
- Personal contacts and previous collaborations are really helpful
- Core involvement as co-applicants and access to the grant as it is written
- Continued involvement in project development, programme governance and co-writing manuscripts

Data availability
Underlying data
No data is associated with this article

Acknowledgements
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11. Ms Siobhan Horkan, Administrator and organiser of National Therapeutic Hypothermia Group, Ireland

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14. Dr Jean Quigley, School of Psychology, Trinity College Dublin, Dublin, Ireland

15. James Meaney, Director of CAMI and Professor of Radiology, Trinity College Dublin, Dublin, Ireland

16. Dr Joanne Balfe, Consultant Paediatrician, Tallaght Hospital and Laura Lynn Hospice, Dublin, Ireland

17. Dr David Webb, Consultant Paediatrician Neurologist, Our Lady’s Children’s Hospital & Tallaght Hospital, Dublin, Ireland

18. Gunter Shumann, Professor and Chair in Biological Psychiatry; Director, Centre for Population Neuroscience and Stratified Medicine, King’s College London, London, UK

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Open Peer Review

Current Peer Review Status: ?  ?

Version 1

Reviewer Report 16 November 2020

https://doi.org/10.21956/hrbopenres.14101.r28214

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Brigitte Vollmer
Clinical and Experimental Sciences, Faculty of Medicine, University of Southampton, Southampton, UK

This interesting and timely open letter, of interest to a large audience of clinicians and researchers in the field.

I have a few comments and I feel these should be addressed prior to indexing:

1. Careful review for grammar, typos, and errors (e.g. in list of associate partners) is required.

2. It is important to clearly define neonatal hypoxic-ischaemic encephalopathy (HIE) as a disorder in the group of neonatal encephalopathies and not imply that neonatal encephalopathy is the same as neonatal HIE. There are a number of other aetiologies of neonatal encephalopathy than perinatal asphyxia (PA) which leads to neonatal HIE. By using terms interchangeably it is not clear whether this project is focussed on HIE as a consequence of PA only or encompasses all aetiologies of NE.

3. "All babies with brain injury get an MRI" - should this read all babies with the clinical presentation of NE undergo MRI to identify brain injury?

4. Future directions: this currently reads a little like a "shopping list" - it would be helpful for the reader to revise this section to improve flow.

5. Would it be helpful to include a table that lists PPI involvement point by point?

Is the rationale for the Open Letter provided in sufficient detail?
Partly

Does the article adequately reference differing views and opinions?
Partly
Are all factual statements correct, and are statements and arguments made adequately supported by citations?  
Yes

Is the Open Letter written in accessible language?  
Partly

Where applicable, are recommendations and next steps explained clearly for others to follow?  
Partly

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Neonatal and Paediatric Neurology; Developmental Neurology; Brain development after early acquired brain injury; advanced MRI.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 29 June 2020

https://doi.org/10.21956/hrbopenres.14101.r27586

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David Sweet  
Regional Neonatal Unit, Royal Maternity Hospital, Belfast, UK

This paper sets out to describe the benefits of parental involvement at all stages of a major research project, from conceptualisation and grant application, through interviewing of prospective PhD students and supervision of the projects.

The paper is clearly written and easy to understand, however my one criticism would be that the initial "Rationale" section focuses too much on neonatal encephalopathy and not on what this paper is about... that is involving parents in design and execution of a major research programme.

In my view the first two paragraphs should lead the reader in to the subject of the importance of patients and parents views on what is appropriate in research.... only then mention the fact the neonatal encephalopathy studies were what were being used in this instance to develop the model of greater parental involvement.

Apart from that only one small correction... the word "follow-up" in the "Aims" section should be hyphenated.
In The Key Messages section make it clearer that you are talking about Patient and Public Involvement, perhaps by stating this in the message.

1. For patient involvement personal contacts and previous collaborations are helpful.

2. Patients can be involved as co-applicants with access to the grant as it is written.
   etc. etc.

**Is the rationale for the Open Letter provided in sufficient detail?**
Partly

**Does the article adequately reference differing views and opinions?**
Yes

**Are all factual statements correct, and are statements and arguments made adequately supported by citations?**
Yes

**Is the Open Letter written in accessible language?**
Yes

**Where applicable, are recommendations and next steps explained clearly for others to follow?**
Partly

**Competing Interests:** No competing interests were disclosed.

**Reviewer Expertise:** Neonatal clinical trials, respiratory distress syndrome.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.