

Research prioritisation exercises related to the care of children and young people with life limiting conditions their parents and all those who care for them: protocol for a systematic scoping review

Protocol agreed 10/03/17: prior to screening studies against inclusion criteria
Minor amendments made: 21/04/17 prior to second screening; 10/05/17 at data extraction piloting (highlighted in yellow)

Organisational affiliation of the review

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This review is being undertaken as part of the Martin House Research Centre programme of work.

Conflicts of interest

None declared.

BACKGROUND

The Martin House Research Centre (MHRC) was launched in January 2017. The aims of MHRC are to: undertake and effectively disseminate world leading research on the care of children and young people with life-limiting conditions and the provision of children's palliative care; build research capacity within children's palliative care; and act as a hub and facilitator for children's palliative care research. Organised around three workstreams; epidemiology, health and care services, and interventions, the Centre's broad remit will also encompass, the care and support needs of children and families, and span clinical, parenting, caring, social, spiritual, financial and practical aspects.

Research involving children and young people has in the past been seen as difficult, for example around the issues of obtaining informed consent, and therefore not undertaken to the same extent as in adults. This has resulted in clinical decisions having to be based on converting the findings of research in adults to apply to children. For example, drugs are licenced for specific indications and patient groups based on the results of clinical trials, usually carried out in the adult population, age 18 to 65. It is common therefore in the

absence of evidence for children to be prescribed medicines in ways that are not included in the license. Children and young people are different biochemically and physiologically from adults, so this is not satisfactory. [1] However, over the last decade or so initiatives such as International Alliance for Better Medicines for Children in 2006 have been set up and it has become commonly agreed that children are not 'little adults'. [2] Ethical considerations have been overcome, so children and young people are encouraged to be involved in investigations into the most effective diagnoses, treatments and delivery of care specifically for them. [3-5] The evidence base generally for all aspects of care for children and young people and understanding of what is important to patients and their families but there are still some significant gaps. [6] This is particularly so for research to benefit of children and young people with life limiting conditions, the area of specific relevance to the work of the MHRC. The needs, experiences and preferences of children, young people and their parents and carers are increasingly driving research agendas and informing the design of studies. This helps ensure the most appropriate questions are asked and answered appropriately.

In order to ensure that finite resources are made best use of and focussed on the most relevant areas, the first action planned is to undertake a research prioritisation exercise. [7, 8] Having undertaken some preparatory internet searches we identified a significant number of published research prioritisation exercises relevant, in varying degrees, to children and young people with life limiting conditions and their parents and carers. [9-12] We found studies published between 2014 and 2017 related to the UK context. As a result, our first step is to undertake a systematic scoping review of existing research prioritisation exercises. The findings of the scoping review will be used to develop a framework of existing priorities, and to inform the design and format of our research consultation exercise.

Systematic scoping review aims and objectives

The aim of this systematic scoping review is to provide an overview of existing research prioritisation exercises relevant to children, young people with life limiting conditions and their families and carers.

Our objectives in undertaking this review are: to identify, as far as possible, all relevant studies; document key information about the prioritisation exercise, including purpose and methods used; provide a map of the conditions and age groups 'covered' in the literature and stakeholders contributing to the consultations; provide an overview of the research priorities identified.

Rationale for undertaking a systematic scoping review

The broad scope of the MHRC remit and need to ensure resources are maximised justified a research prioritisation exercise as a first step. The existence of relevant research prioritisations within our remit, if arrived at through consultation with relevant stakeholders, should provide an evidence based framework for our consultation. We are not aware of any existing reviews or overviews that take in the broad and complex area of research priorities for children and young people with life limiting conditions.

We will use the rigor of systematic review methods to ensure identification, selection and quality assessment of relevant published research prioritisation exercises. The synthesis will provide an overview of the conditions where priorities have been set and present the areas that are most frequently identified as priorities for future research.

METHODS

We will undertake a systematic scoping review of existing research prioritisation exercises focussed on children and young people with life limiting conditions/**life-shortening and life-threatening conditions**, their families and carers. Systematic searches will be undertaken, as will dual independent screening and data extraction of included studies. An assessment of the quality of the included studies will be undertaken. We aim to provide an overview of the studies identified and map the research priorities reached through consensus. Given the nature of the included studies and the aim of the review, a meta-analysis is not planned.

Inclusion criteria

Population

There will be no restrictions on the people or stakeholders undertaking the prioritisation exercise.

Condition

The prioritisation exercise should be aimed at identifying research for the benefit of: neonates (birth to 28 days), children and/or young people (28 days to age 25) with life limiting conditions/**life-shortening and life-threatening conditions**; their family, parents, carers; and/or the professional staff caring for them.

We will use the definitions for life-limiting/life-shortening and life-threatening conditions adopted by Together for Short Lives. [13]

- Life-limiting/life-shortening conditions are those for which there is no reasonable hope of cure and from which children or young people will die. Some of these conditions cause progressive deterioration rendering the child increasingly dependent on parents and carers.
- Life-threatening conditions are those for which curative treatment may be feasible but can fail, such as cancer; are also included. Children in long-term remission or following successful curative treatment are not included in this review.

Where an exercise includes priorities for mixed age groups (children and/or young people and adults) these will be included only if details of the priorities are reported separately for children and young people.

Where an exercise sets priorities for neonates, children and/or young people but not exclusively those with LLCs, these studies will only be included if priorities specifically related to LLCs are reported.

Outcomes

Any form of prioritisation for future research, such as topic areas or specific research questions.

Setting

Any setting related to care of children and young people with life limiting conditions both as the setting for undertaking the exercise and/or for the research priorities set.

While we will search the international literature, to focus generalisability to the UK setting, we will, when screening, exclude exercises exclusively undertaken in non-OECD countries.

Study Design

Studies using any consultation method aimed at achieving a consensus between stakeholders, that is people identified as having a role relevant to the topic for example children and young people with a life limiting condition, their parents/carers and/or healthcare professionals and/or academics. This may be a single group (e.g. PICU nurses) or may be a range of stakeholders (e.g. PICU nurses, parents, and paediatricians). Studies must include at least two rounds of consultation to be included. Consultation rounds may be in any combination of formats or a repeated format (e.g. electronic survey + face to face; two electronic surveys).

Abstracts will be excluded as they are unlikely to include sufficient information, but full papers will be searched for where the abstract appears to report a relevant study.

Research and development analyses and knowledge gap analyses will be excluded as these are different concepts to agreeing research priorities.

Search strategy

The search strategy has been developed by an experienced information specialist (KW) in collaboration with the review team.

Electronic sources

Searches of ASSIA, CINAHL, Embase and MEDLINE will be conducted; the strategy for use in Ovid MEDLINE (Appendix 1) will be modified for the remaining databases. KW will carry out the searches, create an EndNote library and remove duplicate records.

Search terms

Terms describing: children and young people; families; research, priorities, James Lind Alliance, and Delphi methods will be included, using MeSH terms where available. Studies in animals will be excluded.

Other sources

We will check the reference lists of included papers identified from the electronic searches. We will search for grey literature, in particular, the James Lind Alliance Prioritisation Setting

Partnership website will be hand searched for on-going and completed research prioritisation exercises.

Restrictions

An English language only restriction will be used. Healthcare, treatment and service deliver for children and young people with life limiting conditions have changed considerably over the last few decades. In addition, the identification of priorities for future research implies that subsequent research is likely to have been undertaken. For these reasons the searches will be restricted to prioritisation exercises published from 2000 to date.

Study selection and data extraction

Study selection will be performed independently by two researchers. Discrepancies will be resolved by discussion or by recourse to a third researcher. Titles and abstracts will be screened first, then at second screening full papers will be assessed for inclusion or reason for exclusion recorded.

A data extraction form will be designed and piloted independently by two researchers using a selected range of studies. The review team will discuss and amend the data extraction items as necessary; this will be an iterative process and reflect the nature of the studies identified. Once finalised, one researcher will data extract the included studies and a second researcher will check at least 30% of the data. Discrepancies will be resolved by discussion or by recourse to a third researcher. Given time and resource constraints we will not be contacting authors for missing or unclear data.

Items for data extraction may include:

- Reference number, author, year of publication, journal title
- Aims of the exercise (e.g. identify questions; identify topics; scope)
- Study setting and context (e.g. country; health condition(s); health care setting; age group included – if no age restriction, are CYPs interests reported separately?)
- Identification of existing research/ research uncertainties identified (e.g. not done/done/at what stage)
- Perspective(s) included (e.g. nurses on PICU only; nurses and children aged 11-14)
 - are details of participants experience documented (yes/no)
- Methods used to achieve consensus:
 - theoretical model (e.g. Delphi, +/- modifications)
 - undertaken in collaboration with JLA (yes/no)
 - starting point and number of iterations (e.g. base line information provided/sought + consultation rounds)
 - means of communication (e.g. email/post +/- face to face meeting(s))
 - participants included in consensus process (e.g. clinicians, care providers, parents, children) including the numbers and degree of involvement
- Degree of consensus aimed for/ degree achieved
- Priorities identified
- Planned use of priorities (e.g. dissemination; programme of research)*
- Issues or observations reported in the results*

- Issues or observations reported in the conclusions*

**These items in particular will be discussed following piloting of the data extraction form to assess their value and the practicalities of extracting this data.*

If possible and considered appropriate when the included studies have been identified, the researchers will indicate their assessment, in broad terms, of the type of research method(s) likely to be needed to address each of the priorities identified (e.g. quantitative and/or qualitative).

When piloting the data extraction forms: due to the nature and variety of research prioritisation exercises included, it was not found to be appropriate to extract data about issues or observations reported in the results or conclusions. Nor was it felt possible to suggest even in broad terms the type of research method(s) likely to be needed to address each of the priorities identified.

Quality assessment

We are not aware of any validated instrument for quality assessing research prioritisation exercises. However, the WHO developed a quality assessment framework to examine the methods used in prioritisation exercises carried out within the organisation[14]. This will be modified for use in this review.

Strategy for collating, summarising and reporting the data

We will aim to chart the data, and collate, summarise and report the results based, as far as possible, on the enhancements of Levac et al[15] to the scoping review framework suggested by Arksey and O'Malley[16]. However, much will depend on the studies identified and the type of priorities set; for example whether areas for research are specified or specific research questions have been agreed.

A narrative overview of the included studies will be presented. We will provide a description of the methods used to set priorities.

We will produce tables and charts to map the priorities against the life limiting conditions, the ages of the children and young people, the settings, and participants in the exercises. This will demonstrate where priorities have already been identified and where they have not yet been considered.

We will map the priorities identified in the studies. Priorities will be presented pictorially and if possible include indications of the number of studies, and/or the range of stakeholders involved in the consultation process[17]. We will also aim to categorise the priorities by the research study methods likely to be needed to address the issue identified.

DISSEMINATION

A full report, a lay summary report (4 sides of A4) and related presentations will be prepared and used to inform the structure of a consultation event. The mapping will provide a framework for group discussions which will feed into a consensus meeting.

The full report of the scoping review, including the methods used, will be submitted for publication in a peer reviewed journal. This report and the summary report and associated presentations will be made available on the Martin House Research Centre website: www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse.

We will also present the findings of the scoping review at relevant meetings and conferences as appropriate.

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Appendix 1: Literature search strategy for MEDLINE

Database: Ovid MEDLINE(R) Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE(R) Daily and Ovid MEDLINE(R) <1946 to Present>
Records identified 3171

- 1 exp Child/ (1711150)
- 2 exp Infant/ (1033024)
- 3 Adolescent/ (1791294)
- 4 (child* or infant* or newborn* or new-born* or neonat* or neo-nat* or baby* or babies or pediat* or paediat* or schoolchild* or preschool*).ti,ab,kf. (1811535)
- 5 (adolescen* or juvenile* or youth* or teenage* or youngster*).ti,ab,kf. (349423)
- 6 (young people* or young person*).ti,ab,kf. (24096)
- 7 exp Family/ (273430)
- 8 (family* or families or parent* or mother* or father* or maternal or paternal or mom*1 or mum*1 or dad*1 or sister* or brother* or sibling* or son or sons or daughter*).ti,ab,kf. (1533594)
- 9 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 (4805080)
- 10 Health Priorities/ (9858)
- 11 James Lind*.ti,ab. (100)
- 12 psp.ti,ab. (4766)
- 13 (research* adj2 priorit*).ti,ab,kw. (5271)
- 14 (priorit* adj2 set*).ti,ab,kw. (3591)
- 15 (research adj4 Delphi).ti,ab. (178)
- 16 10 or 11 or 12 or 13 or 14 or 15 (22057)
- 17 9 and 16 (4431)
- 18 ("non human" or animal or animals or monkey or monkeys or chimpanzee or chimpanzees or primate or primates or macaque or macaques or hamster or hamsters or rat or rats or mouse or mice or mice or mice or bird or birds or chicken or chickens or goat or goats or cattle or cattles or cow or cows or pig or pigs or dog or dogs or cat or cats or lamb or lambs or bovine or bovines or sheep or sheeps or rabbit or rabbits or horse or horses or equine or equines or camel or camels).ti. (1567441)
- 19 17 not 18 (4373)
- 20 limit 19 to yr="2000 -Current" (3171)