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

4

5 **ABSTRACT**

6 **Background:** In planning high quality research in any aspect of care for children and young
7 people with life-limiting conditions it is important to prioritise resources in the most
8 appropriate areas.

9 **Aim:** To map research priorities identified from existing research prioritisation exercises
10 relevant to infants, children, and young people with life-limiting conditions, in order to
11 inform future research.

12 **Design:** We undertook a systematic scoping review to identify existing research
13 prioritisation exercises; the protocol is publicly available on the project website.

14 **Data sources:** The bibliographic databases ASSIA, CINAHL, MEDLINE/MEDLINE In Process
15 and Embase were searched from 2000. Relevant reference lists and websites were hand
16 searched. Included were any consultations aimed at identifying research for the benefit of
17 neonates, infants, children and/or young people (birth to age 25 years) with life-limiting, -
18 threatening or -shortening conditions; their family, parents, carers; and/or the professional
19 staff caring for them.

20 **Results:** Twenty four research prioritisation exercises met the inclusion criteria, from which
21 279 research questions or priority areas for health research were identified. The priorities
22 were iteratively mapped onto an evolving framework, informed by WHO classifications. This
23 resulted in identification of 16 topic areas, 55 sub-topics and 12 sub-sub-topics.

24 **Conclusions:** There are numerous similar and overlapping research prioritisation exercises
25 related to children and young people with life-limiting conditions. By mapping existing
26 research priorities in the context in which they were set, we highlight areas to focus
27 research efforts on. Further priority setting is not required at this time unless devoted to
28 ascertaining families' perspectives.

29

30 **Keywords:** Delphi Technique; Consensus; infant; infant, newborn; child; Adolescent;
31 Palliative Care; Humans.

32

33

34 **What is already known about the topic?**

- 35
- 36 • There is limited high quality research in many aspects of care for infants, children
37 and young people with life-limiting conditions.
 - It is important to minimise waste in research and maximise use of limited resources.

- 1 • A range of research prioritisation exercises for a variety of aspects of care or
2 conditions exist within the broad scope of this population.

3 **What this paper adds**

- 4 • This paper provides a unique overview of where and by whom a wide range of
5 research priorities for infants, children and young people with life-limiting conditions
6 have been agreed.
7 • The research priorities identified are mapped in the context in which they were
8 agreed, while common topics and themes are highlighted.

9 **Implications for practice, theory or policy**

- 10 • This paper presents an overview of consensus derived research priorities for infants,
11 children and young people with life-limiting conditions, providing the opportunity for
12 a coherent approach to improving the evidence base for this area of practice.
13 • This study highlights the need for broader consideration of stakeholder perspectives
14 when undertaking research prioritisation exercises.
15 • However further identification of research priorities cannot be justified at this time
16 unless ascertaining the perspectives of children and young people and their families.

17

18 **BACKGROUND**

19 The number of children with life-limiting or life-threatening conditions has been rising with
20 latest figures estimating 49,000 children and young people with a life-limiting condition in
21 the UK and approximately 21 million worldwide.^{1, 2} These include conditions for which there
22 is no reasonable hope of cure and from which children or young people will die, as well as
23 conditions for which curative treatment may be feasible but can fail, such as cancer or heart
24 failure. In children and young people, more than 300 diagnoses are life-limiting or life-
25 threatening,³ including Duchene muscular dystrophy, severe cerebral palsy,
26 neurodegenerative conditions, and severe congenital anomalies. Although many of the
27 individual diagnoses are rare, as a group children and young people with a life-limiting
28 condition are a larger patient population than many other long term conditions in children
29 and young people, such as diabetes mellitus.⁴ Many of these children are living longer due
30 to the use of medical technologies, e.g. ventilation and gastrostomy feeding, and more
31 aggressive treatment of complications and they are often high users of healthcare services.^{5,}
32 ⁶ The recent UK National Institute for Health and Care Excellence guidance on End-of-life
33 care for children and young people, although focussed on end of life care, highlighted the
34 lack of evidence base on which the care of these children and young people was based.⁷ The
35 recent Lancet Commission on Palliative Care and Pain Relief states that globally, nearly 2.5
36 million children die in need of palliative care and pain relief, and over 90% of paediatric
37 deaths associated with serious health-related suffering are avoidable.⁸ The American
38 Academy of Pediatrics guidelines and recommendations on paediatric hospice and palliative
39 care are based on available evidence and consensus expert opinion, while acknowledging
40 the need for further clinical and health service research.⁹

1 In terms of medical research involving children and young people with life-limiting
2 conditions has in the past been seen as difficult, for example around issues of access, clinical
3 considerations and obtaining informed consent.¹⁰ This has resulted in clinical decisions
4 having to be based on converting the findings of research in adults to apply to children. For
5 example, drugs are licenced for specific indications and patient groups based on the results
6 of clinical trials, usually carried out in the adult population, age 18 to 65. It is common
7 therefore in the absence of evidence for children to be prescribed medicines in ways that
8 are not included in the license. Children and young people are different biochemically and
9 physiologically from adults, so this is not a satisfactory solution.¹¹ However, over the last
10 decade or so initiatives such as the International Alliance for Better Medicines for Children
11 in 2006 have been set up and it has become commonly agreed that children are not 'little
12 adults'.¹² Ethical considerations have been overcome, so children and young people are
13 encouraged to be involved in investigations into the most effective diagnoses, treatments
14 and delivery of care specifically for them.¹³⁻¹⁵

15 Involving children, including those with life-limiting conditions, directly in research about
16 their lives and the services they use is, however, more established.¹⁶⁻¹⁹ So the evidence base
17 generally for all aspects of care for children and young people and understanding of what is
18 important to patients and their families has grown. But there are still some significant gaps
19 in terms of evidence with respect to particular populations and/or the scope, or
20 comprehensiveness of that evidence.^{7, 10, 20}

21 The identification of priority areas for research using consensus methods is recognised as a
22 good way of ensuring that finite research resources are used to maximum effect.^{21, 22} Prior
23 knowledge and preparatory internet searches identified a significant number of published
24 research prioritisation exercises relevant, in varying degrees, to neonates, infants, children
25 and young people with life-limiting conditions and their parents and carers in the UK.²³⁻²⁶
26 We therefore planned to examine the focus, context and questions identified by existing
27 prioritisation exercises in this area by undertaking a scoping review to:

- 28 • Systematically identify existing research prioritisation exercises relevant to infants,
29 children, and young people with life-limiting conditions and their families and carers
- 30 • Formulate the findings into a framework, mapping existing priorities.

31 **METHODS**

32 The exploratory nature of the review, the broad scope and anticipated volume of literature
33 was suited to scoping review methods. In addition, the absence of any validated quality
34 appraisal tools for consensus exercises precluded a systematic review. The scoping review
35 was undertaken using systematic methods and is reported in line with the PRISMA
36 statement.²⁷ The review protocol was agreed and made publicly available on the Martin
37 House Research Centre website²⁸ prior to screening studies against inclusion criteria;
38 scoping review protocols are not accepted for registration on PROSPERO. Amendments
39 made to the protocol were highlighted and dated.

40 **Eligibility criteria**

41 Studies using any consensus consultation method were included. Studies had to include at
42 least two rounds of consultation.

1 Excluded were research and development analyses and knowledge gap analyses as these
2 are different concepts to agreeing research priorities.

3 There were no restrictions on the people or stakeholders undertaking the prioritisation
4 exercise. Research prioritisation exercises aimed at identifying research for the benefit of:
5 neonates (birth to <28 days), children and/or young people (28 days to age 25) with life-
6 limiting, life-shortening and/or life-threatening conditions; their family, parents, carers;
7 and/or the professional staff caring for them. We used the definitions for life-limiting, life-
8 shortening and life-threatening conditions (hereafter encompassed in the term life-limiting)
9 adopted by Together for Short Lives: ²⁹

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

17 Exercises seeking to identify research priorities for mixed age groups (children and/or young
18 people and adults) were included if details of the priorities were reported separately for
19 children and young people. Likewise, exercises setting priorities for our target age groups
20 but not exclusively those with life-limiting conditions, were included if priorities specifically
21 related to life-limiting conditions were reported.

22 The outcomes of interest were the top ten priorities for future research, such as topic areas
23 or specific research questions presented as the main result of the consultation. Secondary
24 lists, for example priorities for a sub-group of participants, were excluded.

25 **Search sources and strategy**

26 To identify studies for inclusion we searched ASSIA, CINAHL, MEDLINE/MEDLINE In Process
27 and Embase. The search strategy was developed by an experienced information specialist in
28 collaboration with the rest of the review team. The strategy consisted of thesaurus and free
29 text terms for "research prioritisation" combined (using AND) with terms for children and
30 young people and their families. The search strategies are presented in Supplementary File
31 1.

32 The reference lists of included papers and relevant websites, such as the James Lind Alliance
33 (JLA) Prioritisation Setting Partnership (PSP) website, were hand-searched for on-going and
34 completed research prioritisation exercises.

35 There were no restrictions on the setting in which the exercise was undertaken or for the
36 research priorities set. However, to aid generalisability to settings with similar healthcare
37 provision, we restricted exercises to those in the English language and undertaken in
38 Organisation for Economic Co-operation and Development countries. Healthcare, treatment
39 and service delivery for children and young people with life-limiting conditions have
40 changed considerably over the last few decades. In addition, the identification of priorities

1 for future research implies that subsequent research is likely to have been undertaken. For
2 these reasons the searches were restricted to exercises published from 2000 to date.

3 **Study selection and data collection**

4 Study selection was performed independently by two researchers with discrepancies
5 resolved through discussion or by recourse to a third researcher. Titles and abstracts were
6 screened first, then at second screening full papers were assessed for inclusion.

7 A data extraction form was designed and piloted independently by two researchers. Once
8 finalised, one researcher data extracted the included studies and a second researcher
9 checked 30% of the records. Discrepancies were resolved by discussion or by recourse to a
10 third researcher.

11 Items data extracted were: publication details; funding; aims and objectives; study
12 country/ies; setting; health condition/s; age group; methods used to achieve consensus;
13 degree of consensus; priorities identified; and planned use of priorities.

14 **Strategy for collating, summarising and reporting the data**

15 We charted the data, and collated, summarised and reported the results based on the
16 enhancements of Levac et al³⁰ to the scoping review framework suggested by Arksey and
17 O'Malley³¹. This included the research team collectively developing the data extraction
18 tables and discussing sequential iterations and piloting until an appropriate data set had
19 been agreed. Simple narrative and descriptive statistics were used for reporting the
20 included study characteristics and methods.

21 For the synthesis we used the WHO established classifications of functioning, disability and
22 health as a starting point for developing a framework.³² Three researchers independently
23 categorised the identified priorities into the framework. Many of the authors had grouped
24 their questions or issues into topic areas which were also used to inform the process.
25 Repeated discussions and iterations took place, including consideration of the factors that
26 informed prioritisation decision-making.³³

27 **RESULTS**

28 The electronic searches, run in February 2017, identified a total of 7447 records which were
29 loaded into EndNote v7 bibliographic software and de-duplicated leaving a total of 4970
30 records for screening. The study flow chart is presented in Figure 1.

31 Initial screening identified 189 records for full text assessment, from which 24 studies were
32 found to meet the inclusion criteria. A table of characteristics of the included studies is
33 provided in Table 1. A list of excluded studies and the full data extraction tables are
34 available on request from the authors.

35 **Theoretical consensus models used**

36 The consensus method most frequently used in the included consultations was the Delphi
37 Consensus method. The majority of consultations used questionnaires to obtain responses,
38 mainly distributed electronically, but a few were distributed as hard copies.

39 **Settings**

1 Most of the consultations were undertaken in single countries. Three consultations were
2 Europe wide, and a fourth attempted global reach. The majority of consultations were
3 undertaken in, and for use by, centres providing specialist care such as paediatric palliative
4 care (including hospices), neo-natal/paediatric intensive care or oncology units.

5 **Participants in the consultations**

6 Details of participants' professions/roles were generally not clearly reported but the
7 majority were health professionals, including academic health professionals. Other
8 participants included academics, social workers, volunteers, carers, charity staff, teachers,
9 policy makers, and priests.

10 Only eight studies reported including children and young people and/or parents and family
11 members (Table 1). None of the included studies only consulted with these groups. There
12 were some reports on the difficulties of recruiting children, young people and parents, but
13 the majority of papers did not mention any attempts to include family members or explain
14 why they had not.

15 The number of individuals taking part in a consultation exercise overall, and within
16 individual rounds, varied considerably and not all study reports included details of numbers
17 at every stage.

18 **Focus for research priorities**

19 The priority setting exercises either focussed on conditions, such as cancer or cystic fibrosis,
20 an overarching 'any life-limiting condition', or a specified care need, such as requiring
21 paediatric palliative/end of life care, or other therapies.

22 The majority of papers reported age descriptively, for example "children and young adults"
23 or "paediatric", rather than specifying an age range.

24 **Planned use of priorities**

25 The majority of research prioritisation exercises were undertaken with the intension of the
26 results being used by the wider health professional and research communities.^{23, 26, 34-46}
27 Others were undertaken to provide priorities for the benefit of funding and commissioning
28 agencies; raising the profile of paediatric palliative care³⁸; ensuring the perspective of the
29 family was considered in neonatal research.⁴⁴ Eight studies were undertaken specifically to
30 inform the research agenda of those undertaking the exercise for their particular setting.⁴⁷⁻
31 ⁵⁴

32 Seventeen studies referred to searching for existing prioritisation exercises in the
33 background to their papers, either not finding any relevant to their area of interest or
34 justifying a repeat for differences in service delivery between countries. Fourteen studies
35 discussed the priorities identified in the context of existing research; two of these reported
36 modifying the priorities in light of finding relevant research.

37 **Research priorities**

38 From the 24 studies meeting the inclusion criteria, we identified 279 research questions or
39 priority areas for research. The majority of studies presented up to 10 top priorities, 11
40 listed between 12 and 33 and one listed 82 items. The range was 4 to 82, the average 17

1 and the median 11. Eight of the studies reported more than one final list of priorities: these
2 were differentiated by topics, categories, or by groups of participants in six cases, and in two
3 cases, as well as ranking questions, the authors ranked research domains.

4 The priorities were variously called research priorities, items, themes, questions,
5 statements, or issues, and were framed in different ways. We use the collective term
6 'research priorities' from here on. This ranged from detailed specific questions including
7 population, intervention, comparator and outcome (PICO),³⁸ to less explicit, more
8 exploratory questions such as what is best practice for a specific stage of care.^{40, 41} Some
9 studies simply listed, or included, one or two word topic areas.^{25, 34, 45}

10 **Synthesis of research priorities**

11 As the majority of studies sought to identify 10 or less research priorities, and given our
12 objective to identify issues/topic areas considered most pressing or causing greatest
13 concern, we limited inclusion in our synthesis of the data to a maximum of 10 of the overall
14 top priorities reported.

15 This yielded a total of 279 research priorities from the included studies. Some studies
16 presented lists of priorities under more than one category, and some had less than 10
17 priorities (hence the odd number). The complete list of synthesised research priorities
18 identified by the included studies is very large and therefore presented by topic, in
19 Supplementary File 2.

20 Some studies concerned specific types of service or setting (palliative, hospice and end-of
21 life care; tertiary referral centres; oncology; and PICU and NICU). In terms of the scope of
22 the studies with respect to diagnostic categories, they were grouped as either life limiting
23 conditions or cancer.

24 An iterative analysis of the research priorities generated 16 topic areas across which were
25 subsumed 55 sub-topics and 12 sub-sub-topics. These are presented in Table 2 and a high
26 level summary of the key issues provided here.

- 27 • *Epidemiology / population (including access to services)*: the need to and feasibility of
28 collecting data; barriers and facilitators to accessing services; the needs of CYP and their
29 parents; and strategies for health promotion.
- 30 • *Measurement and assessment*: understanding what quality of life means; assessment of
31 symptoms; levels of sedation in babies; assessment of antibiotic levels; psychosocial
32 issues for children with cancer.
- 33 • *Service delivery and models of care*: best models for organisation and delivery of care;
34 differences in quality and cost; how to maintain good governance; use of tele-care;
35 barriers and facilitators to shared care; inter- and multi-agency collaboration; effective
36 transfer between services; palliative care outcomes of importance to CYP and families;
37 effective ways to measure, prevent, record aspects of care.
- 38 • *Health interventions: pharmacological and/or invasive*: avoiding necrotising
39 enterocolitis; preparing children for cancer treatment; best practice in ventilation;
40 meeting nutritional needs; long term effects of cancer treatments.

- 1 • *Symptom management and control*: effective management of symptoms e.g. pain
2 control, in different settings and different stages; non-pharmacological interventions;
3 drug trials.
- 4 • *Other interventions: physical health and functioning*: optimum timing and delivery of
5 interventions; long-term safety and effectiveness of techniques to manage spasticity;
6 achieving, or improving, continence; effective communication with CYP with
7 neurodisability; strategies to manage sleep disturbance in CYP with neurodisability;
8 facilitating engagement in physical activity.
- 9 • *Intervention adherence*: barriers and facilitators.
- 10 • *Emotional and psychological issues*: factors to protect or risk factors when adjusting to
11 living with a LLC; coping mechanisms; anxiety, breathlessness and sleeplessness as
12 clusters; emotional and psychological challenges and experience for CYP; needs of
13 parents, siblings and wider family; support systems and practices; promotion of well-
14 being.
- 15 • *Participation and inclusion*: promoting positive attitudes; supporting independence in
16 terms of participation and mobility.
- 17 • *Communication and decision-making*: recording care; communications within multi-
18 disciplinary teams; communications between staff and CYP and their families; shared
19 decision making.
- 20 • *Other family needs and support*: impact on the family; support for family; support for
21 CYP to self-manage.
- 22 • *Practices related to palliative and end-of-life care*: understanding and supporting parent
23 expectations; understanding what dying well means.
- 24 • *Bereavement*: CYP experience of and coping with bereavement; care and support needs
25 of families.
- 26 • *Ethics*: parental role in ethical decision making; ethical dilemmas for staff.
- 27 • *Workforce*: recruitment, retention and support for staff; training to equip staff to deliver
28 high quality evidence based care; strategies to help staff communicate with CYP and
29 their families.
- 30 • *Funding*: who and how research questions should be set; where funding should be
31 targeted.

32

33 **DISCUSSION**

34 **Main findings/results of the study**

35 In this scoping review we aimed to map research priorities identified from existing research
36 prioritisation exercises relevant to infants, children, and young people with life-limiting
37 conditions. Extensive searches of bibliographic databases and hand searching identified 24
38 research prioritisation exercises that met the inclusion criteria. From these 279 research
39 questions or priority areas for health research were identified. The content of these
40 research priorities was subject to a thematic analysis which generated 16 topic areas with
41 numerous sub-topics across these areas. This demonstrates the wide range of research
42 priorities identified by existing studies, with major themes including medical treatments,
43 condition and symptom management; non-medical aspects of care and support; and topics
44 of shared interest and expertise. No single research prioritisation exercise captured the total

1 range. This demonstrates the value of our prudent use of resources having taken the
2 approach of a scoping review.

3 Given the number and diversity of life limiting conditions and the range of services involved
4 in their care, the large number of published exercises and identified priorities was not
5 unexpected. Whilst these studies generated a large volume of research priorities, it was
6 possible to organise these into a relatively small number of overarching and meaningful
7 topic areas.

8
9 The research prioritisation exercises included in this review were wide-ranging in terms of
10 purpose, inclusivity of stakeholders and experts, topic areas and scope. However, a key
11 limitation of many studies was the lack of involvement of children and young people and
12 parents in the research prioritisation exercise or, where sought, only minimal involvement
13 was secured.^{23, 25, 26, 34, 38, 39, 48, 50} It was not possible to map the priorities by condition or by
14 age group as we thought might be possible based on the systematic approach to
15 undertaking research prioritisation taken by the JLA PSP.⁵⁵ The rigorous methods used by
16 the JLA set a standard to aspire to, but which are challenging even for the JLA to meet. For
17 example we included the JLA PSP on neurodisability, where the authors report the problems
18 of including children and young people in the consensus process.²⁶ An issue echoed in other
19 studies where attempts were made. While including children and young people, parents and
20 other family members is always likely to be challenging given the nature of the conditions
21 concerned, it is still disappointing that so few studies reported even considering
22 perspectives other than those of the health professionals. Even within the involvement of
23 health professionals, this almost exclusively included doctors and nurses, with very little
24 involvement of other relevant professions such as physiotherapists, speech and language
25 therapists, occupational therapists, clinical psychology, and dieticians. This is concerning
26 given the evidence from various fields that what is important to patients can be different to
27 what is important to clinicians.⁵⁶⁻⁵⁹ Also, other professional groups outside of the healthcare
28 sectors such as social workers, priests/chaplains, and teachers may bring new insights and
29 perspectives. A useful and efficient way forward to address this inequity and imbalance in
30 the stakeholder groups involved in identifying research priorities to date would be to use
31 the findings from this review as a basis for consultation with families and particular
32 professional groups. We recently carried out such an exercise to inform and guide the work
33 of our newly established research centre.²⁸

34 The number of exercises identified may illustrate a wide spread awareness of the lack of
35 evidence in this area, and the interest in providing the evidence in a way that maximises
36 value.²² We limited the search period from post-2000 to February 2017. It may be that some
37 progress has been made in producing research evidence. We are aware that the JLA PSP on
38 neurodisability has informed the commissioning of research by the UK's National Institute
39 for Health Research. However, within the studies included in this review, only a few
40 reported that research priorities were limited to those where there was a known degree of
41 uncertainty. It was beyond the remit of this review to evaluate the current evidence
42 available against each of the topic and sub-topics identified by our synthesis.

43 Undertaking thorough literature searches to justify carrying out any study is good research
44 practice. So, while this review is a valuable resource, it does not negate the need for

1 evidence reviews to inform future research and decisions regarding the commissioning of
2 research.

3 The drivers behind the production of the prioritisation exercises varied. For many the reason
4 for doing the exercise was specific to a profession or service at a single site. This may have
5 been in the belief that priorities would vary between sites and professions. However, again
6 the overlap of priorities in this scoping review demonstrates the commonality of issues that
7 need to be addressed. When more research has been carried out and it becomes
8 appropriate to take a fresh look at priorities, we believe a more comprehensive approach
9 would be justified.

10 **Strengths and weaknesses/limitations of the study**

11 A key strength of our review is in the systematic methods used to ensure only those
12 consultations that met our pre-specified criteria were included. Our focus on research
13 priorities for children and young people meant we had to exclude studies that included all
14 ages but did not present the results in a way in which we could identify those relevant to
15 children and young people. This meant included studies where the top ten priorities were
16 clearly not relevant to children and young people but also some where they may have been
17 relevant but not necessarily arrived at with this age group specifically in mind. For example,
18 we had to exclude a well conducted JLA PSP which set priorities for clinical research in
19 primary brain and spinal cord tumours related to any age and which included paediatric
20 representatives on the panel.⁶⁰ We also excluded studies that focussed on the Emergency
21 Department setting as generally the population and conditions presenting are very mixed
22 and priorities therefore not focussed on children and young people with life limiting
23 conditions.^{61, 62} This demonstrates the rigor of our selection process but also underlines
24 further the extensive number of priority setting exercises closely related to the 24 studies
25 that met our inclusion criteria.

26 As this is a scoping review we have not included an appraisal of the quality of the included
27 studies. Given the potential for consensus priorities to influence the future direction of
28 research bids and funding, consideration should be given to the way in which the consensus
29 has been carried out. There is extensive literature on consensus methods,^{33, 63-66} and now on
30 the reporting of Delphi studies in palliative care.⁶⁷ In synthesising the included priorities, we
31 used the existing recognised terminology related to children and young people with life-
32 limiting conditions as a starting point for the framework. Although three researchers
33 independently allocated priorities to topics in an iterative analytical process, we have to
34 acknowledge a level of subjectivity in the synthesis. At all times we were conscious of the
35 need to acknowledge the drivers behind individual studies and the factors that informed the
36 decision-making processes. For example, it was important to know who set the questions, in
37 what context and with what degree of consensus.³³

38 We placed no restrictions on the setting in which priorities were set. However, the
39 epidemiology of life-limiting conditions in children, their management, the availability of
40 services and treatments and outcomes are very different in the high resource and
41 low/middle resource countries therefore the research priorities here will be very different.
42 So, to aid generalisability to settings with similar healthcare provision, we restricted

1 inclusion to those in the English language and undertaken in Organisation for Economic Co-
2 operation and Development countries.

3 **What this study adds**

4 We believe this scoping review presents a unique overview of research priorities, arrived at
5 through consensus processes, with respect to infants, children and young people with life-
6 limiting, life-threatening and life-shortening conditions. By detailing the individual
7 consultations and presenting our synthesis of the priorities identified, we provide evidence
8 of the range and focus of areas for improving the evidence base for care of this population
9 and their families. As such it provides a useful resource for researchers, professionals,
10 funders and commissioners of research and other stakeholders involved in supporting
11 evidence informed practice. The findings provide an opportunity at this time to maximise
12 the use of limited research resources by focussing on filling priority evidence gaps.

13 **CONCLUSION**

14 By taking systematic scoping approach to identification of existing research prioritisation
15 exercises and providing transparency in our methods we believe we present a reliable
16 overview of the priorities already set in this area. Anyone wishing to develop a programme
17 of research or planning a specific project could use this review as a starting point, and
18 justification, for choosing topics or questions. Users will of course need first to check if
19 research has been done or is in progress, on priorities before proceeding

20 There are a significant number of research prioritisation exercises related to children and
21 young people with life-limiting conditions. By mapping the priorities while being sensitive to
22 the context in which they were set, we have achieved our aim to provide an overview of
23 existing research priorities.

24 **DECLARATIONS**

25 **Authorship.** AB conceived the approach, all authors contributed to the design of the review
26 and approved the protocol. KW developed the search strategy and ran the searches. AB and
27 JM undertook the screening and data extraction with BB and LF providing arbitration; AB,
28 JM, BB and LF synthesised the priorities identified. AB drafted the paper and JM, KW, BB and
29 LF provided critical comments. The final version was approved by all authors.

30 **Funding.** This review was undertaken as part of the Martin House Research Centre
31 programme of work.

32 **Declaration of conflicts of interest.** The Authors declare that there is no conflict of interest.

33 **Research ethics and patient consent.** Research ethics and consent were not required for
34 this review of published studies.

35 **Data management and sharing.** The data extraction tables are available from the authors
36 on request.

37 **Acknowledgements.** The team would like to thank Jenny Parry for her work obtaining
38 copies of papers for the review.

1 REFERENCES

- 2
- 3
- 4 1. Connor SR, Downing J and Marston J. Estimating the Global Need for Palliative Care
5 for Children: A Cross-sectional Analysis. *Journal of pain and symptom management* 2017;
6 53: 171-177. 2016/10/22. DOI: 10.1016/j.jpainsymman.2016.08.020.
- 7 2. Fraser LK, Miller M, Hain R, et al. Rising national prevalence of life-limiting conditions
8 in children in England. *Pediatrics* 2012; 129: e923-929. 2012/03/14. DOI:
9 10.1542/peds.2011-2846.
- 10 3. Hain R, Devins M, Hastings R, et al. Paediatric palliative care: development and pilot
11 study of a 'Directory' of life-limiting conditions. *BMC Palliat Care* 2013; 12: 43. 2013/12/18.
12 DOI: 10.1186/1472-684x-12-43.
- 13 4. Royal College of Paediatrics and Child Health. National Paediatric Diabetes Audit.
14 2014.
- 15 5. Feudtner C, DiGiuseppe DL and Neff JM. Hospital care for children and young adults
16 in the last year of life: a population-based study. *BMC Medicine* 2003; 1: 3-3. DOI:
17 10.1186/1741-7015-1-3.
- 18 6. Ananth P, Melvin P, Feudtner C, et al. Hospital Use in the Last Year of Life for
19 Children With Life-Threatening Complex Chronic Conditions. *Pediatrics* 2015; 136: 938-946.
20 DOI: 10.1542/peds.2015-0260.
- 21 7. National Institute for Health and Care Excellence (NICE). End of life care for infants,
22 children and young people with life-limiting conditions: planning and management. 2016.
- 23 8. Knaul FM, Bhadelia A, Rodriguez NM, et al. The Lancet Commission on Palliative Care
24 and Pain Relief - findings, recommendations, and future directions. *The Lancet Global Health*
25 2018; 6: S5-S6. DOI: 10.1016/S2214-109X(18)30082-2.
- 26 9. American Academy of Pediatrics Policy Statement. Pediatric Palliative Care and
27 Hospice Care Commitments, Guidelines, and Recommendations. Section on hospice and
28 palliative medicine and committee on hospital care. *Pediatrics* 2013; 132: 966.
29 10.1542/peds.2013-2731.
- 30 10. Hudson BF, Oostendorp LJ, Candy B, et al. The under reporting of recruitment
31 strategies in research with children with life-threatening illnesses: A systematic review.
32 *Palliat Med* 2017; 31: 419-436. 2016/09/10. DOI: 10.1177/0269216316663856.
- 33 11. Bourns IM. Unlicensed medicines in the UK – legal frameworks, risks, and their
34 management. *@Medicine Access@Point of Care* 2017; 1: 0. 10.5301/maapoc.0000006. DOI:
35 10.5301/maapoc.0000006.
- 36 12. Gazarian M. Delivering better medicines to children: need for better integration
37 between the science, the policy, and the practice. *Paediatr Drugs* 2009; 11: 41-44.
- 38 13. Blyth CC, Chen SC, Slavin MA, et al. Not just little adults: candidemia epidemiology,
39 molecular characterization, and antifungal susceptibility in neonatal and pediatric patients.
40 *Pediatrics* 2009; 123: 1360-1368. DOI: 10.1542/peds.2008-2055.
- 41 14. Einarsdóttir J. Research with children: methodological and ethical challenges.
42 *European Early Childhood Education Research Journal* 2007; 15: 197-211. DOI:
43 10.1080/13502930701321477.
- 44 15. World Health Organisation. Children are not little adults In: Children's environmental
45 health training modules. June 2016. Available from:
46 www.who.int/ceh/capacity/training_modules/en/

- 1 16. Coyne I, Sheehan A, Heery E, et al. Improving transition to adult healthcare for young
2 people with cystic fibrosis: A systematic review. *Journal of Child Health Care* 2017; 21: 312-
3 330. DOI: 10.1177/1367493517712479.
- 4 17. Ambresin AE, Bennett K, Patton GC, et al. Assessment of youth-friendly health care: a
5 systematic review of indicators drawn from young people's perspectives. *The Journal of*
6 *adolescent health : official publication of the Society for Adolescent Medicine* 2013; 52: 670-
7 681. 2013/05/25. DOI: 10.1016/j.jadohealth.2012.12.014.
- 8 18. Beresford B and Stuttard L. Young adults as users of adult healthcare: experiences of
9 young adults with complex or life-limiting conditions. *Clinical Medicine* 2014; 14: 404-408.
10 DOI: 10.7861/clinmedicine.14-4-404.
- 11 19. Gibson F, Aldiss S, Horstman M, et al. Children and young people's experiences of
12 cancer care: a qualitative research study using participatory methods. *International journal*
13 *of nursing studies* 2010; 47: 1397-1407. 2010/05/01. DOI: 10.1016/j.ijnurstu.2010.03.019.
- 14 20. Simpson LA, Peterson L, Lannon CM, et al. Special challenges in comparative
15 effectiveness research on children's and adolescents' health. *Health affairs (Project Hope)*
16 2010; 29: 1849-1856. 2010/10/06. DOI: 10.1377/hlthaff.2010.0594.
- 17 21. Cuello-García CA. Setting research priorities to avoid waste—the time is right. *The*
18 *Journal of Pediatrics* 2015; 167: 219-221. DOI: <https://doi.org/10.1016/j.jpeds.2015.06.018>.
- 19 22. Chalmers I, Bracken MB, Djulbegovic B, et al. How to increase value and reduce
20 waste when research priorities are set. *The Lancet* 2014; 383: 156-165. DOI: 10.1016/S0140-
21 6736(13)62229-1.
- 22 23. Baker JN, Levine DR, Hinds PS, et al. Research Priorities in Pediatric Palliative Care. *J*
23 *Pediatr* 2015; 167: 467-470.e463. Research Support, Non-U.S. Gov't. DOI:
24 [10.1016/j.jpeds.2015.05.002](https://doi.org/10.1016/j.jpeds.2015.05.002).
- 25 24. Browne LR, Shah MI, Studnek JR, et al. 2015 Pediatric Research Priorities in
26 Prehospital Care. *Prehospital emergency care : official journal of the National Association of*
27 *EMS Physicians and the National Association of State EMS Directors* 2016; 20: 311-316.
28 2016/01/26. DOI: 10.3109/10903127.2015.1102997.
- 29 25. Clinton-McHarg T, Paul C, Sanson-Fisher R, et al. Determining research priorities for
30 young people with haematological cancer: a value-weighting approach. *Eur J Cancer* 2010;
31 46: 3263-3270. Research Support, Non-U.S. Gov't. DOI:
32 <https://dx.doi.org/10.1016/j.ejca.2010.06.013>.
- 33 26. Morris C, Simkiss D, Busk M, et al. Setting research priorities to improve the health of
34 children and young people with neurodisability: a British Academy of Childhood Disability-
35 James Lind Alliance Research Priority Setting Partnership. *BMJ Open* 2015; 5: e006233.
36 Research Support, Non-U.S. Gov't. DOI: <https://dx.doi.org/10.1136/bmjopen-2014-006233>.
- 37 27. Moher D, Liberati A, Tetzlaff J, et al. Preferred Reporting Items for Systematic
38 Reviews and Meta-Analyses: The PRISMA Statement. *PLOS Medicine* 2009; 6: e1000097.
39 DOI: 10.1371/journal.pmed.1000097.
- 40 28. Martin House Research Centre. Research Prioritisation Exercise - Martin House
41 Research Centre, , [https://www.york.ac.uk/healthsciences/research/public-](https://www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse/projects/prior-exercise/)
42 [health/projects/martinhouse/projects/prior-exercise/](https://www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse/projects/prior-exercise/) (2017, accessed Dec 19 2017).
- 43 29. Together for Short Lives. Definitions for terms and phrases used in children's
44 palliative care,
45 [http://www.togetherforshortlives.org.uk/professionals/childrens_palliative_care_essentials](http://www.togetherforshortlives.org.uk/professionals/childrens_palliative_care_essentials/definitions)
46 [/definitions](http://www.togetherforshortlives.org.uk/professionals/childrens_palliative_care_essentials/definitions) (2017, accessed 06 March 2017).

- 1 30. Levac D, Colquhoun H and O'Brien KK. Scoping studies: advancing the methodology.
2 *Implementation Science* 2010; 5: 69. DOI: 10.1186/1748-5908-5-69.
- 3 31. Arksey H and O'Malley L. Scoping studies: Towards a Methodological Framework. *Int*
4 *J Soc Res Methodol* 2005; 8. DOI: 10.1080/1364557032000119616.
- 5 32. World Health Organisation. International Classification of Functioning, Disability and
6 Health (ICF), <http://www.who.int/classifications/icf/en/> (accessed 20 Dec 2017).
- 7 33. Lomas J, Fulop N, Gagnon D, et al. On being a good listener: setting priorities for
8 applied health services research. *The Milbank quarterly* 2003; 81: 363-388. 2003/08/28.
- 9 34. Bradley JM, Madge S, Morton AM, et al. Cystic fibrosis research in allied health and
10 nursing professions. *J Cyst Fibros* 2012; 11: 387-392. Congresses Research Support, Non-U.S.
11 Gov't. DOI: <https://dx.doi.org/10.1016/j.jcf.2012.03.004>.
- 12 35. Brenner M, Hilliard C, Regan G, et al. Research priorities for children's nursing in
13 Ireland: a Delphi study. *J Pediatr Nurs* 2014; 29: 301-308. Comparative Study. DOI:
14 <https://dx.doi.org/10.1016/j.pedn.2014.01.003>.
- 15 36. Downing J, Knapp C, Muckaden MA, et al. Priorities for global research into children's
16 palliative care: results of an International Delphi Study. *BMC Palliat Care* 2015; 14: 36.
17 Research Support, Non-U.S. Gov't. DOI: <https://dx.doi.org/10.1186/s12904-015-0031-1>.
- 18 37. Fletcher-Johnston M, Marshall SK and Straatman L. Healthcare transitions for
19 adolescents with chronic life-threatening conditions using a Delphi method to identify
20 research priorities for clinicians and academics in Canada. *Child Care Health Dev* 2011; 37:
21 875-882. Research Support, Non-U.S. Gov't. DOI: [https://dx.doi.org/10.1111/j.1365-](https://dx.doi.org/10.1111/j.1365-2214.2011.01318.x)
22 [2214.2011.01318.x](https://dx.doi.org/10.1111/j.1365-2214.2011.01318.x).
- 23 38. Lioffi C, Anderson A-K and Howard RF. Development of research priorities in
24 paediatric pain and palliative care. *British Journal of Pain* 2016; 11: 9-15. DOI:
25 10.1177/2049463716668906.
- 26 39. Malcolm C, Knighting K, Forbat L, et al. Prioritization of future research topics for
27 children's hospice care by its key stakeholders: a Delphi study. *Palliat Med* 2009; 23: 398-
28 405. Research Support, Non-U.S. Gov't. DOI:
29 <https://dx.doi.org/10.1177/0269216309104061>.
- 30 40. Quinn C, McCarthy S, Devins M, et al. Prioritisation of future research topics in
31 paediatric palliative care in Ireland: a Delphi study. *International Journal of Palliative Nursing*
32 2017; 23: 88-97. DOI: 10.12968/ijpn.2017.23.2.88.
- 33 41. Ramelet AS, Gill F and ACCCN Paediatric Intensive Care Special Interest Group. A
34 Delphi study on National PICU nursing research priorities in Australia and New Zealand. *Aust*
35 *Crit Care* 2012; 25: 41-57. Multicenter Study Research Support, Non-U.S. Gov't. DOI:
36 <https://dx.doi.org/10.1016/j.aucc.2011.08.003>.
- 37 42. Steele R, Bosma H, Johnston MF, et al. Research priorities in pediatric palliative care:
38 a Delphi study. *J Palliat Care* 2008; 24: 229-239. Research Support, Non-U.S. Gov't.
- 39 43. Tume LN, van den Hoogen A, Wielenga JM, et al. An electronic delphi study to
40 establish pediatric intensive care nursing research priorities in twenty European countries.
41 *Pediatr Crit Care Med* 2014; 15: e206-213. Research Support, Non-U.S. Gov't. DOI:
42 <https://dx.doi.org/10.1097/PCC.000000000000109>.
- 43 44. Uhm S, Crowe S, Dowling I, et al. The process and outcomes of setting research
44 priorities about preterm birth — a collaborative partnership. *Infant* 2014; 10: 178-181.
- 45 45. Wielenga JM, Tume LN, Latour JM, et al. European neonatal intensive care nursing
46 research priorities: an e-Delphi study. *Arch Dis Child Fetal Neonatal Ed* 2015; 100: F66-71.

- 1 Research Support, Non-U.S. Gov't. DOI: [https://dx.doi.org/10.1136/archdischild-2014-](https://dx.doi.org/10.1136/archdischild-2014-306858)
2 [306858](https://dx.doi.org/10.1136/archdischild-2014-306858).
- 3 46. Zeigler VL and Decker-Walters B. Determining psychosocial research priorities for
4 adolescents with implantable cardioverter defibrillators using Delphi methodology. *J*
5 *Cardiovasc Nurs* 2010; 25: 398-404. Research Support, Non-U.S. Gov't. DOI:
6 <https://dx.doi.org/10.1097/JCN.0b013e3181d817b0>.
- 7 47. Aylott M. Research priorities: a Delphi survey. *Paediatric Nursing* 2000; 12: 16-20.
- 8 48. de Vries K, Walton J, Nelson K, et al. An examination of the research priorities for a
9 hospice service in New Zealand: A Delphi study. *Palliat Support Care* 2016; 14: 232-240. DOI:
10 <https://dx.doi.org/10.1017/S1478951515000838>.
- 11 49. Fochtman D and Hinds PS. Identifying nursing research priorities in a pediatric clinical
12 trials cooperative group: the Pediatric Oncology Group experience. *J Pediatr Oncol Nurs*
13 2000; 17: 83-87. Research Support, U.S. Gov't, P.H.S.
- 14 50. Medlow S and Patterson P. Determining research priorities for adolescent and young
15 adult cancer in Australia. *Eur J Cancer Care (Engl)* 2015; 24: 590-599. Research Support, Non-
16 U.S. Gov't. DOI: <https://dx.doi.org/10.1111/ecc.12291>.
- 17 51. Monterosso L, Dadd G, Ranson K, et al. Priorities for paediatric cancer nursing
18 research in Western Australia: a Delphi study.[Erratum appears in *Contemp Nurse* 2002
19 Apr;12(2):135]. *Contemp Nurse* 2001; 11: 142-152.
- 20 52. Soanes L, Gibson F, Bayliss J, et al. Establishing nursing research priorities on a
21 paediatric haematology, oncology, immunology and infectious diseases unit: a Delphi
22 survey. *Eur J Oncol Nurs* 2000; 4: 108-117. DOI: <https://dx.doi.org/10.1054/ejon.2000.0083>.
- 23 53. Williams A, Sell D, Oulton K, et al. Identifying research priorities with nurses at a
24 tertiary children's hospital in the United Kingdom. *Child Care Health Dev* 2016; 04: 04. DOI:
25 <https://dx.doi.org/10.1111/cch.12340>.
- 26 54. Wilson S, Ramelet AS and Zuiderduyn S. Research priorities for nursing care of
27 infants, children and adolescents: a West Australian Delphi study. *J Clin Nurs* 2010; 19: 1919-
28 1928. DOI: <https://dx.doi.org/10.1111/j.1365-2702.2009.03025.x>.
- 29 55. James Lind Alliance Priority Setting Partnerships NETSCC Team and Cowan K. The
30 James Lind Alliance Guidebook. Feb 2016; Version 6.
- 31 56. Haywood KL, Collin SM and Crawley E. Assessing severity of illness and outcomes of
32 treatment in children with Chronic Fatigue Syndrome/Myalgic Encephalomyelitis (CFS/ME):
33 a systematic review of patient-reported outcome measures (PROMs). *Child Care Health Dev*
34 2014; 40: 806-824. 2014/03/26. DOI: 10.1111/cch.12135.
- 35 57. Michener LA. Patient- and clinician-rated outcome measures for clinical decision
36 making in rehabilitation. *Journal of sport rehabilitation* 2011; 20: 37-45. 2011/03/18.
- 37 58. Kirwan JR, Newman S, Tugwell PS, et al. Progress on incorporating the patient
38 perspective in outcome assessment in rheumatology and the emergence of life impact
39 measures at OMERACT 9. *The Journal of rheumatology* 2009; 36: 2071-2076. 2009/09/10.
40 DOI: 10.3899/jrheum.090360.
- 41 59. Serrano-Aguilar P, Trujillo-Martin MM, Ramos-Goni JM, et al. Patient involvement in
42 health research: a contribution to a systematic review on the effectiveness of treatments for
43 degenerative ataxias. *Social science & medicine (1982)* 2009; 69: 920-925. 2009/08/04. DOI:
44 10.1016/j.socscimed.2009.07.005.
- 45 60. MacDonald L and on behalf of the Neuro-Oncology Group. *Top 10 priorities for*
46 *clinical research in primary brain and spinal cord*. June 2015. Final report of the James Lind
47 Alliance Priority Setting Partnership in Neuro-Oncology.

- 1 61. Hartshorn S, O'Sullivan R, Maconochie IK, et al. Establishing the research priorities of
2 paediatric emergency medicine clinicians in the UK and Ireland. *Emerg Med J* 2015; 32: 864-
3 868. Research Support, Non-U.S. Gov't. DOI: [https://dx.doi.org/10.1136/emered-2014-](https://dx.doi.org/10.1136/emered-2014-204484)
4 [204484](https://dx.doi.org/10.1136/emered-2014-204484).
- 5 62. Quest TE, Asplin BR, Cairns CB, et al. Research priorities for palliative and end-of-life
6 care in the emergency setting. *Acad Emerg Med* 2011; 18: e70-76. Consensus Development
7 Conference Research Support, U.S. Gov't, P.H.S. DOI: [https://dx.doi.org/10.1111/j.1553-](https://dx.doi.org/10.1111/j.1553-2712.2011.01088.x)
8 [2712.2011.01088.x](https://dx.doi.org/10.1111/j.1553-2712.2011.01088.x).
- 9 63. Hsu CC and Sandford BA. The Delphi Technique: Making Sense of Consensus.
10 *Practical Assessment Research & Evaluation* 2007; 12(10).
- 11 64. Murphy MK, Black NA, Lamping DL, et al. Consensus development methods, and
12 their use in clinical guideline development. *Health technology assessment (Winchester,*
13 *England)* 1998; 2: i-iv, 1-88. 1998/04/30.
- 14 65. Sinha IP, Smyth RL and Williamson PR. Using the Delphi Technique to Determine
15 Which Outcomes to Measure in Clinical Trials: Recommendations for the Future Based on a
16 Systematic Review of Existing Studies. *PLOS Medicine* 2011; 8: e1000393. DOI:
17 [10.1371/journal.pmed.1000393](https://doi.org/10.1371/journal.pmed.1000393).
- 18 66. Viergever RF, Olifson S, Ghaffar A, et al. A checklist for health research priority
19 setting: nine common themes of good practice. *Health Research Policy and Systems* 2010; 8:
20 36. journal article. DOI: [10.1186/1478-4505-8-36](https://doi.org/10.1186/1478-4505-8-36).
- 21 67. Junger S, Payne SA, Brine J, et al. Guidance on Conducting and REporting DElphi
22 Studies (CREDES) in palliative care: Recommendations based on a methodological
23 systematic review. *Palliat Med* 2017; 31: 684-706. 2017/02/14. DOI:
24 [10.1177/0269216317690685](https://doi.org/10.1177/0269216317690685).