**Study Protocol**

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Parent and professional experiences of 24/7 paediatric end-of-life care: a mixed methods study

**Short title:**

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# Protocol Version Number and Amendment History

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| 1.1 | Julia Hackett | 12/08/2022 | Data management/data protection policy | **JH** |
| 1.2 | Laura Barrett | 20/10/2022 | 7.3 data collection9.1 Informed consent9.2 participant burden and distress9.4 data management | **JH** |
| 1.3 | Laura BarrettJulia Hackett | 12/12/2022 | Amended study design  | **JH** |
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Contents

[1 Protocol Version Number and Amendment History 2](#_Toc121820347)

[2 Plain English summary 4](#_Toc121820348)

[3 Background 7](#_Toc121820349)

[4 Study aim and research objectives 8](#_Toc121820350)

[5 Study design 10](#_Toc121820357)

[6 Patient and public involvement 11](#_Toc121820358)

[7 Study methods 12](#_Toc121820359)

[7.1 Sample and sampling strategy 12](#_Toc121820360)

[7.2 Recruitment 14](#_Toc121820361)

[7.3 Data collection 15](#_Toc121820362)

[7.4 Data analysis 18](#_Toc121820363)

[7.5 Integration of findings and development of recommendations 20](#_Toc121820365)

[8 Study management and oversight 20](#_Toc121820366)

[8.1 Study team 20](#_Toc121820367)

[8.2 Sponsorship 21](#_Toc121820368)

[8.3 Study management and oversight 21](#_Toc121820369)

[9 Governance and ethics approval 22](#_Toc121820370)

[9.1 Informed consent 22](#_Toc121820371)

[9.2 Participant burden and distress 23](#_Toc121820372)

[9.3 Participant confidentiality 24](#_Toc121820373)

[9.4 Data management / Data protection 25](#_Toc121820374)

[9.5 Researcher safety and distress 26](#_Toc121820375)

[10 Dissemination 27](#_Toc121820376)

[11 Timeline 27](#_Toc121820377)

[12 References 28](#_Toc121820378)

# Plain English summary

Children and young people (CYP) with life-limiting conditions are living longer with increasingly complex symptoms. It is difficult for families to know what to do if something goes wrong out-of-hours (OOH) when their usual health care services are not available. Providing good quality care and support, round the clock, across a range of services, is key to supporting CYP and families’ choice over place of care and symptom control. All CYP and families should be able to access specialist advice and support 24/7 at end-of-life.

Currently, 24/7 children’s palliative care is not provided in the same way across the UK. We do not have a good understanding of how this is provided, what professionals think of current provision, what would help them deliver better care; or of CYP and families’ experiences of accessing care OOH, and what they need.

In order to support the decision making and provision of 24/7 paediatric palliative care in the North East and Yorkshire region, this study will develop a new evidence-based service intervention to help the delivery of, and access to, 24/7 paediatric end-of-life care. The service will be designed to meet the needs of both families receiving 24/7 end of life care and of professionals and services delivering this care.

The thirty-month study will involve speaking to health professionals to find out what they think about the current provision of 24/7 paediatric end-of-life care, and their expectations and needs of a new service. We will also speak to families to find out about their experiences of OOH care. We will look at data that is already collected by the NHS to find out about differences across the region in A&E and emergency hospital admissions in the last 12 months of life and in where children die, and in order to identify possible examples of good practice we will also compare the region to other areas where 24/7 care services have been implemented.

Finally, in partnership with parent advisors, we will develop an intervention and recommendations/guidance for best practice so that the service can be replicated across the UK, and also for parents to inform their care and support of their child; and a plain English summary. Outputs will be disseminated via key stakeholders, managed clinical networks, and parent facing organisations.

**Scientific Abstract**

We define out-of-hours (OOH) as care delivered between the hours of 17:00-09:00 and at weekends.

**Background**

Making end-of-life care available 24/7 is one of the NHS Foundations for the Ambitions of Palliative and End-of-Life Care (PEoLC) and remains the top UK research priority for palliative care. Children and young people and their families should have access to specialist medical advice and skilled nursing support services where, when, and how they need it, as recommended within the government commitment to end-of-life care and current National Institute for Clinical Excellence guidelines. The distress of uncontrolled symptoms cannot wait for ‘opening hours’ and inefficient care planning across the in-hours and OOH interface is a key barrier to achieving timely access to symptom control, choice of place of care at the end of their lives, and good CYP and family experience. Adults at end-of-life are offered community nursing 24/7 when required, but this is not always available to children.

The North East and Yorkshire region is made up of four ICBs: NHS Humber and North Yorkshire, NHS North East and North Cumbria, NHS South Yorkshire, and NHS West Yorkshire. Across these areas there is significant variation in access to, and provision of, paediatric end-of-life care and as highlighted in a recent report by Together for Short Lives, for many families in this region, there is no 24/7 access to children’s nursing care and advice from a paediatric palliative care consultant. This means that the NICE guidance is not being met. Where care is being delivered out of hours, this is largely provided by third sector organisations, such as children’s hospices or by children’s community nursing services whom ‘step-up’ out of goodwill.

In order to support the decision making and provision of 24/7 paediatric palliative care in the North East and Yorkshire region, this study will develop a new service intervention to help the delivery of, and access to, 24/7 paediatric end-of-life care. By conducting this development work, through exploring parent and professional experiences and needs, and patterns of care at end-of-life, this study will develop an intervention grounded in evidence, which will work in everyday practice. The new intervention components will reflect the current service landscape in the region, incorporate evidence from the region and beyond, will be context specific, and ultimately ensure that CYP and their families have access to specialist medical advice and skilled nursing support where, when and how they need it.

**Aim**

The proposed study aims to develop a complex intervention to help the delivery of, and access to, 24/7 paediatric end-of-life care in the North East and Yorkshire region, by assessing parent and professional experiences and needs, and patterns of care at end-of-life and outcomes in other regions where interventions have been developed.

**Methods**

A mixed-methods approach will be used to understand models of practice and access, and to explore parents’ and professionals' experiences in order to develop a service intervention. A logic model has been developed to provide a graphical representation of the programme theory, this will be refined according to findings in order to develop an intervention and demonstrate scalability, recommendations/guidance for best practice will also be produced.

**Workstream 1:** focus groups with specialist and generalist professionals to explore what they think about current provision of paediatric palliative care, their expectations and needs of a new service and the barriers and facilitators of implementation.

**Workstream 2:** semi-structured interviews with parents to explore their experiences of accessing and receiving 24/7 care; and their expectations and needs of a new service.

**Workstream 3:** a retrospective cohort study on the use of A&E and emergency hospital admission and place of death in the last 12 months of life will assess disparities within the region, and quasi experimental methods will evaluate the impact of interventions introduced elsewhere to identify possible good practice.

**Proposed findings**

The findings of this study will support the development of an evidence informed intervention to deliver 24/7 paediatric palliative care, which best support parents, and will mitigate against poor end-of-life and bereavement outcomes. Training and awareness raising will improve professionals’ skills and confidence, positively benefiting staff, children and young people and their families.

A logic model for 24/7 care delivery, refined based upon findings and knowledge exchange activities will be produced. Recommendations/guidance for best practice will be developed and shared so that the service can be replicated across the UK. Recommendations/guidance for parents to inform their care and support of their child, will also be developed in partnership with parent advisors, in addition to a plain English summary.

# Background

Advances in medicine, interventions, and technologies have resulted in children and young people with life-limiting (LLC) conditions living longer with increasingly complex symptoms1 but 4500 children still die in the UK every year. Children with LLC often require numerous hospital admissions and there is often an increased risk of additional, unnecessary hospital admissions, especially towards end-of-life2, 3. This may not be what children and families want and it has a financial cost to the NHS which could be avoided.

Palliative care services for CYP in the UK have developed locally with heavy reliance on individual clinicians and third sector organisations, such as children’s hospices4. That ad hoc provision means delivery of palliative care is often ‘inconsistent and incoherent’5. Currently in England, whilst there are more than 50 children’s hospices, there are tertiary children’s hospitals, without a paediatric palliative care service.

Making children’s palliative care available 24/7 is key to delivering equitable, high quality care and support to CYP with LLCs and families, and remains the top UK research priority for palliative care. The NICE guidelines6 and quality standards of providing care7 include: ‘Infants, children and young people approaching the end-of-life and being cared for at home have 24 hour access to both children’s nursing care and advice from a consultant in paediatric palliative care’. It is also considered the most important element of palliative care for CYP and families5. The distress of uncontrolled symptoms cannot wait for ‘opening hours’8 and inefficient care planning across the in-hours and OOH interface is a key barrier to achieving timely access to symptom control, choice of place of care at end-of-life, and good CYP and family experience. Adults at end-of-life are offered community nursing 24/7 when required, but this is not always available to children. CYP and families should have access to specialist medical advice and skilled nursing support services where, when, and how they need it. A whole systems approach is required9.

Across the UK, 24/7 children’s palliative care is not currently being provided consistently, with a postcode and a diagnosis lottery. Not all families have access to hospice provision, and some do not want this. Children with cancer are more likely to receive good quality 24/7 care, and therefore are more likely to achieve better outcomes, such death at home2, than CYP with non-cancer diagnoses. Current provision does not meet the service specification, NICE guidance7, or the government commitment, with many children not having access to OOH care.

Across the North East and Yorkshire region there are significant variations in access to, and provision of paediatric end-of-life care, and for many families in the region there is no 24/7 access to children’s nursing care and advice from a paediatric palliative care consultant. To support decision making and provision in the region, this study will explore parent and professional experiences and needs of paediatric end-of-life care, examine patterns of care at end-of-life, and then develop a 24/7 paediatric end-of-life care intervention, which is based on evidence. This will ensure that any new services, which are developed and implemented, are designed to meet the needs of both families receiving 24/7 end of life care, and of professionals and services delivering this care.

# Study aim and research objectives

This study will develop a complex intervention to help the delivery of, and access to, 24/7 paediatric end-of-life care in the North East and Yorkshire region, by assessing parent and professional experiences and needs, and patterns of care at end-of-life and outcomes in other regions where interventions have been developed.

We have developed a draft logic model to provide a graphical representation of the initial programme theory (Figure 1). This demonstrates the anticipated core components of the intervention, how they will interact to produce change; anticipated short, medium, and long term outcomes; and how the intervention will have impact13. Throughout the study, the logic model will be iteratively refined.

Funding

Time

Expertise

Partnerships

Communication and information sharing

Coordination of care

Geography and location of staff

Travel

Specialist Paediatric Palliative Care Service

Community nursing care extended / out of hours

Specialist advice line out of hours

Generalist professional training

Care coordination with hospice / community professionals

Clinic delivery in District General Hospitals

Bereavement support

Named professional out-of-hours

Home visits at end of life

Out-of-hours consultant advice

**Short term change in:**

Knowledge

Awareness

Skills

Attitude

Ability to flex up

**Medium term change in:**

Increased early identification of CYP with palliative care needs

Increased awareness of professionals of palliative care service and needs of CYP with LLCs

Support the offer of choice of place of care

Help manage complex cases / difficult decision making

Effective symptom control

**Long term change in:**

Increase in numbers of CYP with ACPs

Better CYP and family experience

Prevent unnecessary hospital admissions

Continuity of care

Choice of place of care achieved

Enable generalist professionals to deliver high quality care to CYP

**Assumptions**

Right level of specialist and advanced skills and knowledge in the teams

Right leadership and support for the specialist teams

Generalist professionals and hospice colleagues share and support the vision

**External factors**

Increasing numbers of CYP with LLCs

Resources

Interprofessional team working

Funding

Prior knowledge and experience of professionals

Figure 1: logic model to describe the delivery of a new 24/7 paediatric end-of-life care service in West Yorkshire and the Humber

This study will explore the following objectives:

1. To explore specialist and generalist paediatric health professionals’ perspectives on the current provision of paediatric palliative care, and their expectations and needs of a new service; and the barriers and facilitators of implementation.
2. To explore CYP and their families’ perspectives on the current provision of 24/7 paediatric palliative care; and their expectations and needs of a new service.
3. To assess regional disparities between use of A&E and emergency hospital admissions in the last 12 months of life, e.g. by condition, ethnic group, deprivation category and geographical locations.
4. To assess regional disparities in the place of death of children.
5. To evaluate changes in A&E and emergency hospital admissions in the last twelve months of life in other areas that have implemented a 24/7 service, to identify possible good practice.
6. To evaluate changes in place of death in other areas that have implemented a 24/7 service to identify possible best practice.
7. To develop an intervention and refine the programme theory according to findings, demonstrate scalability of the intervention, and develop recommendations/guidance for best practice.

# Study design

This is a mixed-methods study, to design a new 24/7 paediatric palliative care service, by developing an evidence based intervention, by exploring professional and parent experiences, and child outcomes.

Normalization Process Theory (NPT) is the main theoretical driver for the study and underpins the development of the intervention. This provides a useful conceptual tool as it provides a robust analytical framework for understanding implementation. We have combined this with logic modelling as they are flexible, adaptable and work together, providing a way to understand collaborative work that needs to be done for a new intervention to become embedded with a given context14. In WS1 and WS2 the core constructs of NPT will be explored, specifically: coherence (sense-making work), cognitive participation (relationship work or commitment/buy-in), collective action (enacting work), and reflexive monitoring (appraisal). These have informed data collection strategies and tools. We will be able to focus on how, and in what ways, an intervention would be initially received, how individually and collectively people practically conceptualise and make sense of it. Specifically, topic guides for both workstreams will be designed with reference to NPT and existing knowledge about 24/7 end-of-life care, focusing mainly on implementation work, barriers, enablers and anticipated achievements15-17.

WS1 and WS2 are designed in parallel and are interactive. Findings from each stage will inform and guide the focus of data collection across both workstreams. For example, emergent findings in parent interviews will influence questioning in professional interviews/focus groups and visa-versa.

# Patient and public involvement

We have worked closely with the Martin House Research Centre (MHRC) Family Advisory Board (FAB), which is an advisory board of parents and other adult family members of children with a life-limiting condition including parents of a child who has died (n~20) on the development of this application.

The FAB (referred to as ‘our parent advisors’ herein) highlighted: the importance of including parents of children with cancer as well as parents of children with other conditions, in order to learn what works well as a comparison to what works less well; the importance of including variability in the setting in which the out-of-hours care took place, as not all out-of-hours issues occur in the community, and developing a sampling strategy that will achieve this; and the need to advertise the study outside of NHS and hospice organisations, e.g. through social media.

We have identified key points during the study that our parent advisors will be asked to provide input:

* Helping to develop the recruitment approach and topics to be covered in interviews:
* Helping to prepare participant information sheets and study adverts and letters.
* Helping to refine and test the interview topic guide.
* Providing feedback on the quality and content of initial interviews to inform any refinements to the topic guide.
* Developing and sense-checking analytical themes during the analysis.
* Interpreting study findings and developing recommendations for policy and practice.
* Help to co-produce the study outputs, particularly the family facing outputs, and provide ideas for meeting the communication objectives for the study, e.g.: identify routes for dissemination, sense-checking public facing outputs (e.g. website content, Plain English summaries).

To ensure effective public involvement during the project, we will establish and work in partnership with a parent advisory panel. This will be established at the start of the project and include 4-5 parents as members, recruited to achieve some diversity in experiences. Members will be recruited through the research team’s extensive networks, e.g.: the MHRC and new members will be recruited if people withdraw their involvement during the study.

They will be active in providing guidance at all research stages. The group will meet ~4 times each year via video-call, depending on the involvement that is required at different stages in the study. Members will also informally liaise with JH for interim updates on progress, and by email, post or telephone when needed between meetings or when reviewing documents, depending on individual parent preferences regarding mode of communication.

A PPI log, guided by the Public Involvement Impact Assessment Framework18, will record planned and unplanned involvement, including details about who is involved and how, and how these activities impact on the study.

Anonymised findings will be fed back to all participants, both professionals and parents, at the end of the project. For professionals this will be via presentations to their service / clinical teams. For parents, this will be via recommendations/guidance and a plain English summary, which will both be co-produced with the project’s parent advisors. In addition, all will be provided with the academic papers.

# Study methods

## Sample and sampling strategy

**Workstream 1**

Specialist and generalist professionals from the 4 ICBs who are involved in the delivery of paediatric palliative care and meet the following criteria:

* Delivered care and support to CYP (0-18 years) with palliative care needs and their families in the region.
* Specialist who is a member of either the specialist paediatric palliative care team or the paediatric oncology outreach team, or hospice care team.
* Generalist who is a community-based paediatrician, is a member of the children’s community nursing team, or a general practitioner.

Professionals from the specialist paediatric palliative care services based in the 3 tertiary centres across the 4 ICBs (may include: consultants, lead nurses, psychologists, pharmacists) (12-24); clinical care teams from each of the 6 hospices (12-18); and paediatric oncology outreach nurses from each of the 3 tertiary centres (18-24) will be eligible to be recruited. Therefore, a total of 6 focus groups, with ~42-66 specialist professionals.

Generalist professionals, including children’s community nurses, community-based paediatricians, and general practitioners, from the 4 ICBs (24-32); paediatric consultants and nurses, from the 3 tertiary centres in the 4 ICBs (18-24), and professionals from District General Hospitals from the 4 ICBs (16-24), will also be eligible to be recruited in order to explore the delivery of care from all perspectives. Therefore, a total of 6 focus groups, with ~58-80 generalist professionals.

**Workstream 2**

The sample for workstream 2 will include parents or legal guardians of a child across the 4 ICBs who meet the following criteria:

* Parent/legal guardian is ≥18 years old.
* Their child has a life-limiting diagnosis or has died: for interviews at baseline child has died ≥3 months ago and <3years ago).
* Their child is <19 years old (or died when <19 years old) (i.e. age range of children included is 0-18).
* Evidence that end-of-life care has been discussed (i.e. child is supported by a palliative care service or has an advance care plan) or their child has died.

Parents will be recruited from all 4 ICBs. Sample size ~24-35 parents

We will not include:

* Parents/legal guardians aged <17 years old.
* Those who lack capacity to participate, guided by the 2005 Mental Capacity Act.

All adults who provide regular parenting for a child with a LLC or a child who has died will be invited to take part (e.g.: mothers, fathers, step-parents, adoptive parents, legal guardians). Referred to as ‘parents’ herein. Exclusion criteria for bereaved parents have been informed by recent literature19-21, our own experiences of carrying out research with this population22, and through discussion with our parent advisors.

A stratified maximum variation purposive sampling strategy23 will be undertaken to ensure the sample includes similar size sub-groups of parents who are currently receiving elements of palliative care for their child with a LLC and parents whose child has died, and reflects the diversity within this population of parents both in terms of their own characteristics and their child’s condition. These purposive sampling criteria include: diagnoses of the child, including cancer, as the existing research suggests they experience better quality 24/7 care, therefore achieve better outcomes2 than CYP with non-cancer diagnoses; age; care received during COVI-19 lockdowns; and ethnic origin, all of which have been identified as potentially affecting access to palliative care24. Variations in provision will also be sought (e.g.: access to children’s hospice; for bereaved parents the setting in which end-of-life care took place, the organisations providing palliative care and where their child died). This will enable diverse perspectives to be captured, and comparative analyses where appropriate25. To capture the diversity of perspectives, we anticipate a sample of ~20-35 parents will be required. We will monitor recruitment to ensure we are achieving a spread across these key characteristics.

Supporting diversity and inclusion is highly important, especially given the higher prevalence of life-limiting conditions in children from deprived areas and ethnic minority groups1. We will ensure the WS2 sample includes those from a range of ethnic origins and socioeconomic statuses.

**Workstream 3**

All children in the region who have died aged <18 years old in the past ten years, with comparison with other regions of England without these services.

## Recruitment

The recruitment strategy will ensure the work of all actors make sense of the intervention (coherence), the means by which they participate in it (cognitive participation), and the forms of appraisal they apply (reflexive monitoring). By working in partnership at the outset, throughout, and at the conclusion, particularly in developing and refining the logic model, this will ensure their voices are heard throughout all study phases.

**Workstream 1**

Within the North East and Yorkshire region there are 6 children’s hospices, 33 NHS Trusts with numerous DGHs, 3 tertiary centres and community nursing teams, which provide care to CYP at end-of-life. Participants will be recruited from organisations in which these services are located/cover.

Participants will be invited to participate via an email, including the participant information sheet, distributed from their professional organisation on behalf of the research team. Interested participants will be asked to contact the research team directly. Participants will provide electronic consent in advance of taking part in an interview.

**Workstream 2**

Participants will be recruited from organisations in the North East and Yorkshire region which provide paediatric palliative care to CYP at end-of-life (NHS Trusts, children’s hospices, community nursing teams), and via parent facing organisations, e.g. Together for Short Lives, and via social media sources e.g. Twitter and Facebook. Parent advisors are keen that we use these sources to avoid healthcare practitioner gate-keeping.

We will work with parent advisors and local organisations (Bradford community groups and Born in Bradford team) to help with respect to wording/content of recruitment materials, the conduct of interviews, and questions/approaches to use to explore cultural/religious perspectives. We will use a range of methods (postal and internet) to advertise and communicate with parents about the study to ensure we reach a wide range of parents and do not rely on households having the internet. Parents will also have a choice over mode of contact with the research team.

## Data collection

**Workstream 1**

We will conduct separate in-depth focus groups, via video-call (Zoom or Teams) with specialist professionals and generalist professionals. Focus groups will be conducted at one time point. They will explore professionals’ views about and experiences of current provision of 24/7 paediatric palliative care; the impacts of the pandemic on service delivery and practice; delivering care; supporting generalist staff to care for CYP 24/7; training needs; continuity; and expectations and needs of a new service.

**Workstream 2**

We will conduct face-to-face, telephone, or video-call (Zoom or Microsoft Teams), in-depth semi-structured interviews, depending on individual participants’ preference. Although face-to-face interviews are most commonly used in qualitative research, there are a growing number of studies that successfully use telephone interviews26-28, including our own with parents of children with a LLC29, 30. Interviews will be conducted at one time point.

Interviews will explore parents’ accounts of any OOH palliative care their child and the wider family have received, in particular focusing on how and why they contacted OOH, what happened, and what happened afterwards; their experiences of OOH care provided during the end-of-life period, and if applicable when their child died; any discussions or planning around preferred place of death; the impacts of COVID-19 on the OOH care they were able to access; and their expectations and needs of a new service.

Interviews will explore parents’ accounts of any OOH palliative care their child and the wider family received, in particular focusing on how and why they contacted OOH, what happened, and what happened afterwards; their experiences of OOH care provided during the end-of-life period, and if applicable when their child died; any discussions or planning around preferred place of death; the impacts of COVID-19 on the OOH care they were able to access; and their expectations and needs of a new service.

**Workstream 3**

Linked inpatient hospital HES-ONS data for all children aged ≤18 years old at the time of death between 2012/13-2021/22 (or later data if available at time of delivery - e.g. 2022/23) in England (n~50,000). A request will be sent to NHS Digital for all inpatient (NHS Digital Admitted Patient Care Dataset - 'APC') and A&E records (NHS Digital Emergency Care Data Set - 'ECDS') and death records (NHS Digital HES-ONS linked mortality data - 'death data') for children with a death record in the HES-ONS linked mortality data with age at death ≤18 years (Figure 2).

Only pseudonymised data (data where the identifiers are cryptographically generated by NHS Digital and can only be recovered to re-identify individuals by NHS Digital, but can be used to link records from e.g. inpatient and A&E datasets belonging to the same individual) will be provided by NHS Digital to the research team.

Request to NHS Digital for APC, ECDS and death record data from 1 April 2012 to 31 March 2022 for all children who died aged ≤18 years

May be multiple rounds of discussion to finalise data

NHS Digital assessment of application, including Independent Group Advising on Release of Data (IGARD)

Provision of pseudonymised data to the research team

NHS Digital ECDS data

(A&E records)

NHS Digital HES-ONS mortality data

(Death records)

(inpatient records)

Intermediate dataset\* with any exact addresses of death stripped out (replaced by classification as home, hospice, hospital)

(Death records)

(inpatient records)

Analysis dataset

(Combines demographics, numbers of A&E visits and emergency inpatient admissions in up to 18 months before death)

NHS Digital APC data

(inpatient records)

Figure 2: Anticipated data flows. Only pseudonymised data are released to the research team. Dashed line indicates data that will be held/data processing by the research team

\*In the event that more sensitive data are required in the death records (i.e. exact address of death) to classify place of death, this classification will be performed and sensitive data removed before linkage of the requested datasets.

## Data analysis

**Workstream 1 & 2**

The implementation of this complex intervention will owe as much to the work of parents as it does to service providers and professionals. The experiences of parents will therefore explicitly be used to explore experiences of care.

All interviews will be analysed using Normalization Process Theory (NPT)17, which consists of four core constructs representing how interventions are embedded and 'normalized’ within routine care. These are: coherence, cognitive participation, collective action, and reflexive monitoring; that are singly and together envisaged as interacting with the healthcare context16.

Interviews will be transcribed and read by members of the research team.

First, data will be analysed using thematic Framework Analysis31. Two analysts will work together through the analytical steps. This will help to ensure rigour, authenticity and dependability of findings. A combination of inductive and deductive coding will be utilised. Interviews will be inductively coded, with themes agreed, refined and developed through discussion with the wider team, including the parent advisors. Second, a theory-driven analyses will be conducted, whereby the initial inductive themes will be mapped against the four NPT constructs. This two-stage approach will help to avoid forcing the data into predetermined conceptual categories and ensure our interpretation remains data driven32. Findings from parent data will be used to examine/verify findings from healthcare professionals and viva-versa. Discussions within the team will contribute to the final overarching analysis. NVivo version 12 will be used for managing, coding and exploring data.

**Workstream 3**

The outputs from this workstream will be in two main categories:

* 1. a description of emergency secondary care use in the last year of life and place of death in the region, with assessment of associations with demographics and condition categories.
	2. an evaluation of the impact of any 24/7 care interventions introduced within regions on emergency secondary care use in the last year of life and place of death.

This analysis will utilise routinely collected hospital and death certificate data.

For (i) emergency secondary care events will be summarised for the region over the period, disaggregated by type of event (A&E visit, emergency inpatient admission) and by demographics (e.g. age at death, sex, ethnic groups, deprivation category of home address), geographical location and category of condition. Place of death (home, hospital, hospice/other) will be summarised in the same way. Poisson family models will be used to explore associations between counts of emergency secondary care use, demographics, geographical location and category of condition. Place of death will be summarised and disaggregated as above. Logistic regression or similar models will be used to assess associations between place of death and demographics, location, category of condition.

For (ii) an assessment will be made of whether any identified previously implemented 24/7 care interventions in other regions have changed any of the key outcomes – place of death and use of A&E and emergency inpatient admission towards end-of-life. Within these analyses we can account for changes in other key variables which are known to influence these outcomes e.g. child’s diagnoses, distance from hospital, demographic information (age, ethnicity, levels of deprivation). These analyses will be used to identify types of interventions that have been shown to have the biggest impact on the key outcome measures, as a guide towards possible best practice. Depending on whether the parallel trends assumption stands, a difference in difference analysis (with comparable regions without interventions) or interrupted time series analyses (within region) will assess whether any introduced 24/7 services have impacted the key outcomes (e.g.: % deaths at home or number of A&E visits or hospital admissions in last 3 or 12 months of life).

Data cleaning and quality/missingness assessment determines any required multiple imputation to minimise the effects of potential biases, e.g. from complete case analyses. From past experience, we expect few missing data for the key variables of interest.

For each individual in the data we assign:

* Sociodemographic variables using all available data for individuals e.g. ethnic group assigned as the most common, non-missing value across all hospital data.
* Main diagnoses and co-morbidities using available diagnostic codes in the hospital and death registration data.

In order to explore possible impacts of the introduced 24/7 services on the costs of the usage of A&E and emergency hospital admissions in the last 12 months of life we will also run cost estimations of each unscheduled service using standard UK price weighting methodology.

## Integration of findings and development of recommendations

Intervention development and feasibility testing through drafting, reviewing and revising through key stakeholder workshop as per MRC guidance on developing complex interventions.

Findings from the 3 workstreams will be used to refine the logic model and intervention *iteratively throughout* in order to demonstrate scalability of the intervention and develop recommendations/guidance for best practice. The findings will also identify target populations that may be particularly in need/suited to pilot studies.

**Knowledge exchange workshop**

Two knowledge exchange workshops will be held in months 27 and 29 of the project. These events have been developed in line with the Knowledge Exchange Framework33 and will be held virtually. Parent advisors will provide input into the design of the KEEs.

In month 27 the workshop will be with regional key stakeholders, which will provide a forum for getting feedback from stakeholders to identify problems, implementing possible solutions and assessing intervention acceptability, feasibility and engagement, with the overall objective of refining it.

The workshop in week 29 and has two key purposes: to raise awareness of the research; and to work in partnership with key stakeholders to develop recommendations regarding 24/7 paediatric end-of-life care provision. It will be attended by senior representatives of key professional organisations and charities, including chairs of the managed clinical networks for PPC across the UK.

Parent advisors will provide input into the design of the KEEs. We will aim to identify “key principles” regarding provision for commissioners, managed clinical networks, service leads and practitioners, which are both flexible and adaptable to each local area.

After the event, the research team will produce final drafts of study outputs, confirm they remain aligned to research findings, circulate for final comments and agreement.

# Study management and oversight

## Study team

Dr Julia Hackett (Study lead)

Professor Lorna Fraser

Associate Professor Lucy Ziegler

Dr Stuart Jarvis

Mrs Laura Barratt

Ms Emma McLorie

## Sponsorship

University of York

## Study management and oversight

Julia Hackett (JH), an experienced qualitative researcher with significant experience of conducting research in paediatric palliative care, will lead the study, supported by Lorna Fraser (LF), Director of the Martin House Research Centre (MHRC) ([www.york.ac.uk/mhrc](http://www.york.ac.uk/mhrc)) and Professor Lucy Ziegler (LZ), who has over twenty years of experience of conducting palliative care research and a strong portfolio of mixed methods research. Professors Fraser and Ziegler will support and mentor Dr Hackett in her role as PI, they are highly experienced, with strong track records in this field.

Laura Barrett (LB) and Emma McLorie (EM), both experienced qualitative research associates, have been appointed to work on the study under the supervision of JH, to conduct the qualitative data collection and undertake the analysis. JH will be responsible for the study’s timely completion, the quality of the work, adherence to ethical standards, and effective dissemination and impact. JH will meet with LB and EM weekly throughout the study, and LF and LZ will join these meetings monthly.

A Study Steering Committee will be established with an independent chair and representation from paediatricians, palliative care specialists, commissioners, NHS England, NICE, parents/carers (to represent the Parent Advisory Panel members), appropriate national charities (Together for Short Lives), and professional bodies (Royal College of Paediatrics and Child Health, Association for Paediatric Palliative Medicine). This panel will meet every six months to assess progress of the study against the defined milestones and deliverables and provide advice and expertise to the Study Management Team.

The MHRC Management Team (who together contribute qualitative, topic and clinical expertise) will review study progress and provide expert input when needed. The MHRC Advisory Board, which brings clinical, service, academic and family expertise from outside MHRC, will also offer guidance to the study team, particularly around interpretation of study findings and development of recommendations for policy and practice.

# Governance and ethics approval

The study will be conducted to protect the human rights and dignity of the participant as reflected in the 1996 version of the Helsinki Declaration. The explicit wishes of the participant will be respected including the right to withdraw from the study at any time, the interest of the participant will prevail over those of science and society, provision will be made for indemnity by the investigator and sponsor and a contact name for further information will be provided.

We will seek ethical approval from the NHS Health Research Authority, addressing issues concerning informed consent, participant burden and distress, participant confidentiality, data management and researcher safety and distress, as summarised below.

## Informed consent

**Workstream 1 – health professionals**

The local Principal Investigator (PI) at each participating NHS site will identify suitable participants and circulate recruitment materials. Interested professionals will be asked to contact the research team directly. As with the parents/guardians, professionals will receive a Participant Information Sheet explaining the study and what will happen to them and their information if they take part. Professionals will provide written or electronic consent in advance of taking part in an interview or focus group.

**Workstream 2 - parents**

The study will use a 3-stage consent process.

1. Participants will be provided with brief information about the study from a recruiting organization and can express an interest in taking part by completing a “consent to be contacted form” to return to the research team, or by emailing or phoning the study team directly. Parents who see information about the study on social media or via recruiting organization websites or newsletters will be directed to contact the study team, who will then send them the information pack either by email or post. If posted, the information pack will contain a consent form and a stamped addressed envelope, marked ‘confidential’ for the participant to use.
2. A researcher from the team will arrange a call with potential participants who are interested in taking part to explain and discuss the study, to provide an opportunity for them to ask questions, and to check that the parent understands what the study is about, what will happen to them if they take part and how we will use their information.
3. If the participant is happy to continue, the formal process of documenting consent will be started and an interview will be arranged. Both the researcher and the participant will sign and date a hard copy or electronic Consent Form to confirm that consent has been obtained prior to the start of an interview. The participant will receive a copy of this document and a copy will be securely filed by the study team at the University of York.

Consent will be monitored throughout the interviews of parents, e.g. looking for disengagement or withdrawal, checking that they are happy to continue; and participants will be reminded that they can choose not to answer questions that are too distressing or that they would prefer not to answer, and that they can stop the interview at any time.

Participants will be informed of their right to withdraw at any time and without giving a reason. This is covered in the participant information sheet and will be reiterated prior to starting the interview.

If a participant chooses to withdraw they will be able to withdraw their information as long as it has not already been used in the study (i.e. for analysis). To ensure fairness and to allow for concurrent data collection and analysis we have specified that participants can withdraw any data they have provided up to 30 days after their interview. No further data will be collected from participants who withdraw and all data able to be withdrawn (from their contribution) will be destroyed unless consent is provided to include data collected up to the point of withdrawal in the study.

## Participant burden and distress

It is possible that parents may feel pressurised to participate, they will be informed that the decision about whether to participate is voluntary and will not affect any services or benefits they or their children receive. Taking part may lead to parents becoming distressed/upset. The fact that this may happen will be explicitly addressed in the participant information sheet and by the researcher at the start of the interview in order to put parents at ease and also impart to them a sense of control.

So that parents can weigh up the risk of becoming distressed during an interview, an outline of areas that the interviews will cover will be provided in the participant information sheet. This will also help to make sure that participants are prepared for the interview.

If a participant becomes distressed during the interview, the interviewer will ask the participant if they want to continue or to take a break, and be guided by the participant, who may wish to continue despite experiencing some distress. In cases where distress is significant or sustained, the interviewer may suggest that the interview is stopped, although the decision will be made with the participant. In these cases, a second interview may be arranged to enable parents to participate fully if they wish. Where needed, assistance will be given to help the participant identify appropriate sources of support, or a recommendation made that they contact their GP or mental health professional. If appropriate, the interviewer may also ask the parent/guardian if they would like to be contacted by the trusted health professional that they were able nominate during the consent process. If this is the case, then the researcher will immediately contact the trusted professional via the telephone number supplied on the consent form.

At the end of the interview, the researcher will ask the parents whether they would like to receive a follow-up call (via the arranged method), two to three days after the interview (timeframe chosen by parent). This will be used: a) to check the parent is happy for their interview to be used for the study; b) as an opportunity for the parent to raise anything that they have reflected upon since the interview; c) and to signpost them to organisations for support, such as Together for Short Lives, if they require any further support. If parents/guardians still need support at the end of this call, the study team member conducting the call will ask the parent/guardian if they would like to be contacted by a trusted health professional that they will be asked to nominate during the consent process. If this is the case, then the researcher will immediately contact the trusted professional via the telephone number supplied on the consent form.

As a research centre we have a safeguarding procedure. If the researcher has serious concerns regarding the safety of a parent, they should follow the research centre’s safeguarding procedure and the identifying centre's safeguarding protocol. They will advise the parent during the call of this process. At all times during an interview, there will be a senior researcher on call. The researcher will first telephone the researcher on call, who they will discuss their concerns with. As a team, a decision will be made as to whether it is necessary to escalate their concerns to either the nominated trusted health professional or the identifying centre. In a case of immediate danger, the chief investigator will contact the police.

## Participant confidentiality

Participants will be informed of their right to confidentiality, and what this means if they disclose information that suggests that they or others are at serious risk of harm. Participants will also be informed that they have the right to withdraw from the study at any time, and to exclude their data from the study if not already analysed as part of the research.

All personal data will be stored in password-protected files, using a participant identifier to link participants’ details to their data (i.e. interview transcript). On entering the study all participants will be given a study identifier code. Codes and the corresponding names will be kept in a separate file location to the study data, in a password-protected file. This information and all data will be stored on the University of York servers and will not be accessed by anyone outside of the research team.

Qualitative data will comprise interview transcripts and researcher field notes. The interview transcripts, which will have all potentially identifiable data removed, will be the primary data source for the study. Audio-recordings, which will be password-protected and stored securely on the University of York server following participants’ interviews, will be kept until the transcripts have been checked and anonymised. They will then be destroyed.

Quotations from participants may be used in research reports and other publications and presentations; however, care will be taken to protect the anonymity of participants so that others are not able to identify them. Any quotations that are used in the final report or any other publications will be anonymised through giving the participant a pseudonym. Additionally, the characteristics of participants and their children will be presented in a way that will ensure anonymity, e.g. condition categories rather than individual diagnoses will be reported.

## Data management / Data protection

In line with the 2018 General Data Protection Act and the Research Governance Framework for Health and Social Care Research, data (anonymized interview transcripts and field notes) will be securely archived by the University of York for a minimum of 10 years. Paper consent-to-contact forms and consent forms will be stored in a locked filing cabinet in a locked office at the University of York. Electronic consent-to-contact and consent forms will be stored on a secure University of York server. These will be kept for 10 years in line with the University of York's requirements.

All information collected during the study will be kept strictly confidential. Information will be held securely in paper and/or electronic formats at the University of York. The University of York complies with all aspects of the 2018 General Data Protection Act and operationally this will include obtaining consent from patients and carers to record personal details including name, postal and email address, and contact telephone numbers; and appropriate storage, restricted access and disposal arrangements for patient and carer personal details.

Consent data from healthcare professionals may be gathered via Qualtrics. All personal information will be held within the UK or European Economic Area in full compliance with data protection legislation.

All participants will be anonymised at the point of consent, by assignment of a study identifier code. Personal data and pseudonymised data will be stored separately in a restricted access folder on a secure university server and access will be password protected.

* All data will be stored in accordance with data protection requirements and will be kept either in a locked filing cabinet in a secure office or in the case of electronic data on a secure sever with a password-protected computer and files.
* Participants’ names and contact details will be stored in a secure place (a locked office) and only accessed by the research team. Electronic data will be stored on password-protected secure computers in the research team members’ locked offices.
* Audio recordings of the interviews will be downloaded onto a password-protected area of the University of York server and deleted from Zoom or the recording device. Audio recordings will only be used during analysis if needed, e.g. to check the meaning of a participant response. These will be destroyed when the transcripts have been analysed.
* No data will be stored on a home computer or laptop.
* All anonymised interview data will be stored for a minimum of 10 years, which will allow time for any academic challenge to be made. All data will be deleted after this time.
* Personal data and consent forms will be stored for up to 3 years after the end of the study. It will then be destroyed.

## Researcher safety and distress

To ensure researcher safety, we will adhere to the University of York policy and procedure on lone working and employ a buddy system to monitor researchers’ whereabouts and safety if visiting participants or conducting virtual interviews from University premises outside of normal office hours. We will also develop risk management protocols that are consistent with those used by the Trusts and hospices we will be working with. All research staff will be GCP (Good Clinical Practice) trained.

We will only use experienced qualitative researchers who have previously worked on sensitive topics to interview participants and analyse data. However, it is possible that the researchers may experience some emotional distress as a result of interviewing parents about their experiences of end of life care for their child or during the process of data analysis. To monitor and manage this, the researchers will meet weekly throughout the study to reflect on data collection and analysis. In preparing for data collection, the researchers will also have an opportunity to conduct one or two pilot interviews with our parent advisors. Additionally, debrief meetings will take place 1-3 days after the first few interviews (between the interviewer and the study's Chief Investigator), and then organised as and when needed after this and throughout the data collection process. These strategies have been used successfully in similar studies.

# Dissemination

The main outputs from this study are:

Output 1: Prototype intervention: A prototype will be developed through synthesis of evidence and stakeholder consensus.

Output 2: Logic model for 24/7 care delivery. This has been drafted at the application stage of the study and will be refined iteratively throughout based on study findings. It will incorporate findings on inputs, outputs, contextual factors, implementation influencers, CYP and family impacts, and intervention mechanisms and outcomes. This will be disseminated in conjunction with the briefing for commissioners, managed clinical network and service leads, and health professionals (Output 2).

Output 3: Research briefing for commissioners, managed clinical network and service leads, and health professionals setting out key findings and implications for practice and training. A short, text-based output, written specifically for commissioners, network and service leads, and health professionals, will highlight key research findings; and implications regarding provision, practice and staff support/training. This will be produced in partnership with key stakeholder groups during the knowledge exchange event in month 29 of the project. These will be designed to ensure that they are relevant, accessible, and feasible to implement within the NHS and hospice sector. These will be provided to commissioners, clinical networks and service leads, and health professionals.

Output 4: Plain English summary and recommendation/guidance for parents. A short, text-based output, developed in partnership with the project’s parent advisory group, will highlight key research findings and implications regarding provision and access. This will be distributed via parent facing organisations, e.g.: Together for Short Lives and Child Bereavement UK.

Outputs 5 and 6: A minimum of two open access articles in a high impact journals, and presentations at key national conferences. These will reach the wider clinical and academic audiences.

Output 7: Final report for Marie Curie. All the study outputs will be available via the study website, social media, and via links from other websites.

# Timeline

The timeline for key phases of the study is provided below and a more detailed Gantt chart is provided in Appendix ?.

# References

1. Fraser LK, Gibson-Smith D, Jarvis S, et al. Estimating the current and future prevalence of life-limiting conditions in children in England. 2021; 35: 1641-1651. DOI: 10.1177/0269216320975308.

2. Gibson-Smith D, Jarviw WS and Fraser LK. Place of death of children and young adults with a life-limiting condition in England: a retrospective cohort study. *Archives of Disease in Childhood* 2020; 1: 1-6.

3. Jarvis S, Livingston J, Childs AM, et al. Outpatient appointment non-attendance and unplanned health care for children and young people with neurological conditions: a retrospective cohort study. . *Developmental Medicine and Child Neurology* 2018; 61: 840-846.

4. Lives. TfS. *A guide to children's palliative care*. 2018. Bristol.

5. Noyes J, Edwards RT, Hastings RP, et al. Evidence-based planning and costing palliative care services for children: novel multi-method epidemiological and economic exemplar. *BMC Palliative Care* 2013; 12: 18. DOI: 10.1186/1472-684X-12-18.

6. Villanueve G, Murphy MS, Vickers D, et al. End of life care for infants, children and young people with life limiting conditions: summary of NICE guidance. *BMJ* 2016; 355.

7. (NICE) NIfHaCE. *End of life care for infants, children and young people; quality standards*. 2017. London.

8. Widdas D, McNamara K and Edwards F. *A core pathway for children with life-limiting and life-threatening conditions*. 2013. Bristol: Together for Short Lives.

9. Bainbridge D, Brazil K, Ploeg J, et al. Measuring healthcare integration: Operationalization of a framework for a systems evaluation of palliative care structures, processes, and outcomes. *Palliative medicine* 2016; 30: 567-579. 2016/03/05. DOI: 10.1177/0269216315619862.

10. Papworth A, Fraser LK and Taylor J. Development of a managed clinical network for children's palliative care - a qualitative evaluation. *BMC Palliative Care* 2021; 20.

11. Medical Research Council. Developing and evaluating complex interventions: new guidance: 2009, <https://mrc.ukri.org/documents/pdf/complex-interventions-guidance/> (2009, accessed 01 November 2021).

12. Moore GF, Audrey S, Barker M, et al. Process evaluation of complex interventions: Medical Research Council guidance. 2015; 350: h1258. DOI: 10.1136/bmj.h1258 %J BMJ : British Medical Journal.

13. Weiss CH. Theory-based evaluation: Past, present, and future. 1997; 1997: 41-55. DOI: <https://doi.org/10.1002/ev.1086>.

14. Finch TLea. Making sense of a cognitive behavioural therapy intevention for fear of falling: qualitative study of intervention development. *BMC Health Services Research* 2014; 14: 436.

15. May CR, Cummings A, Girling M, et al. Using Normalization Process Theory in feasibility studies and process evaluations of complex healthcare interventions: a systematic review. *Implementation Science* 2018; 13: 80. DOI: 10.1186/s13012-018-0758-1.

16. Murray E, Treweek S, Pope C, et al. Normalisation process theory: a framework for developing, evaluating and implementing complex interventions. *BMC Medicine* 2010; 8: 63. DOI: 10.1186/1741-7015-8-63.

17. May C and Finch T. Implementing, Embedding, and Integrating Practices: An Outline of Normalization Process Theory. *Sociology* 2009; 43: 535-554. DOI: 10.1177/0038038509103208.

18. Popay JMCaG. *The Public Involvement Impact Assessment Framework Guidance*. Universities of Lancaster 2014.

19. Butler AE, Hall H and Copnell B. Bereaved parents’ experiences of research participation. *BMC Palliative Care* 2018; 17: 122. DOI: 10.1186/s12904-018-0375-4.

20. Hynson JL, Aroni R, Bauld C, et al. Research with bereaved parents: a question of how not why. *Palliative medicine* 2006; 20: 805-811. 2006/12/07. DOI: 10.1177/0269216306072349.

21. Sque M, Walker W and Long-Sutehall T. Research with bereaved families: a framework for ethical decision-making. *Nursing ethics* 2014; 21: 946-955. 2014/03/13. DOI: 10.1177/0969733014521097.

22. Martin House Research Centre. <https://www.york.ac.uk/healthsciences/research/public-health/projects/martinhouse/research/> (2022).

23. Palinkas LA, Horwitz SM and Green CAea. Purposeful Smapling for Qualitative Data Collection and Analysis in Mixed Method Implementation Research. *Adm Policy Ment Health* 2015; 42: 533-544.

24. Taylor JT, Booth A and Beresford B, et al. Specialist paediatric pallitaive care for children and young people with cancer: A mixed-methods systematic review. *Palliative medicine* 2020; 34: 731-755.

25. Barbour R. *Introducing Qualitative Research*. London: Sage Publications Ltd, 2008.

26. Irvine A, Drew O and Sainsbury R. 'Am I not answering your questions properly?' Calrification, adequacy and responsiveness in semi-structured telephone and face-to-face interviews. *Qualitative Research* 2012; 13: 87-106.

27. Johnson DR, Scheitle CP and Ecklund EH. Beyond the in-person interview? How interview quality varies acorss in-person, telephone, and skype interviews. *Social Science Computer Review* 2019.

28. Heath J, Williamson H and Williams L, et al. "It's just more personal": Using multiple methods of qualitative data collection to facilitate participation in research focusing on sensitive subjects. *Applied Nursing Research* 2018; 43: 30-35.

29. Maddison J, Taylor J and O'Neill M, et al. Outcomes for gastrostomy-fed children and their parents: qualitative findings from the 'YourTube' study. *Developmental Medicine and Child Neurology* 2021.

30. Hackett J, Heavey E and Beresford B. 'It was like an airbag, it cushioned the blow': A multi-site qualitative study of bereaved parents' experiences of using cooling facilities. *Palliative medicine* 2022: 2692163211059345. 2022/01/21. DOI: 10.1177/02692163211059345.

31. Ritchie J, Lewis J and O'Connor. Carrying out qualitative analysis. In: Lewis Ra (ed) *Qualitative Research Practice: A guide for social science students and researchers*. London: Sage, 2003.

32. O’Donnell A, Hanratty B, Schulte B, et al. Patients’ experiences of alcohol screening and advice in primary care: a qualitative study. *BMC Family Practice* 2020; 21: 68. DOI: 10.1186/s12875-020-01142-9.

33. Economic and Social Research Council. How to do effective knowledge exchange, <https://www.ukri.org/councils/esrc/impact-toolkit-for-economic-and-social-sciences/how-to-do-effective-knowledge-exchange/#contents-list> (2017).