School of Nursing

Care Management Trajectories of Infants with Life-Limiting Conditions Who Died Before 12 Months of age And Understanding How We Deliver Palliative Care

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This thesis is presented for the Degree of
Master of Philosophy
of
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8 March 2024

DECLARATION

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgement has been made.

This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007).

This study received human research ethics approval from Institutional and Curtin University Human Research Ethics Committee, Approval Number HRE2020-0676.

Signed:

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Rebecca Iten
8 March 2024

ABSTRACT

Introduction: Infants with life-limiting conditions (LLC) are a heterogeneous population, and furthermore they comprise of the highest deaths in childhood. The provision of palliative care for infants is complex and impacted by unique challenges that may reduce the opportunity to integrate palliative care principles and practices into their care. Infants may receive palliative care in a diverse range of health settings and by interdisciplinary primary healthcare teams, which may not involve specialist palliative care service consultation.

Aim: The overall aim of this study was to better understand how palliative care is delivered to infants with life-limiting conditions and examine the involvement of specialist palliative care service consultation in this population.

Methods: The thesis consists of two studies. The first study (Study 1) was a retrospective patient health record review to explore the care management trajectories of infants, palliative and end-of-life care provided to infants with life-limiting conditions who died before 12 months of age. The infants had received care during 2018-2019 at three hospitals in Western Australia (WA). Two methods of data analysis involved directed content analysis and process mapping. Approvals from Institutional and University Human Research Ethics Committees were obtained. A waiver of consent was approved.

The second study (Study 2) was an integrative review to synthesis the literature for how palliative care is delivered for infants aged less than 12 months with life-limiting conditions from a broader context. Six databases; MEDLINE, CINAHL, ProQuest, Cochrane, Joanna Briggs Institute and EMBASE were searched for peer reviewed theoretical and empirical research from diverse methodologies published in English, from 2010 to 2022. Quality appraisal was completed for 26 patient cases series, 9 qualitative, 5 cross sectional and 1 quality improvement study. Data extraction involved grouping the articles according to the research focus, each article was summarised and tabulated. Data analysis involved deductive content analysis and narrative approach to summarise the synthesised results.

Results: In Study 1, 45 infants were eligible for inclusion. Process mapping led to typology of care management encompassing four trajectories; early de-escalation due to catastrophic event (11 patients); treatment with curative intent throughout (4 cases); treatment with curative intent until a significant point (21 patients); and early treatment limits (9 patients). There was specialist palliative care team involvement for 25% of infants and standardised advance care planning processes were used for just over 10% of infants. Bereavement and psychosocial support for the family were infrequently reported.

In Study 2, the search generated 3,156 articles; a reference management software programme and research screening tool organised and screened the articles. A total 1,674 articles were screened at title and abstract level, with 116 articles that met the eligibility criteria and were independently read in full. The final number of articles included after critical appraisal was 37. Studies were conducted from 14 countries and included cases series of newborns up to age 18 years (24 studies), health professionals (8 studies), and bereaved parents (5 studies). The 37 articles identified and examined two models of palliative care delivery and illustrated differences in care received and experiences of families and health professionals. Health professionals reported lack of palliative care education, challenges for delivering palliative care in intensive care settings and barriers to advance care planning including prognostic uncertainty and transitioning to end-of-life care. Families reported positive experiences with specialist palliative care services and challenges engaging in advance care planning discussions.

Conclusion: The two studies presented in this thesis revealed there are complex issues surrounding the provision of palliative care for infants. Study 1 and Study 2 both identified only a small proportion of infants with LLCs received specialist palliative care consultation and the majority died in intensive care settings. Infants and their families may benefit from earlier integration of palliative care principles and practices in parallel to receiving curative or life-prolonging treatment and include specialist palliative care service consultation. Optimal palliative care should encompass a collaborative and coordinated approach between the primary healthcare teams and specialist palliative care services. There is opportunity to enhance the delivery of palliative care for infants and optimise the experience for families through prioritisation of palliative care education for health professionals, implementation of standardised advance care planning processes and better integrate specialist palliative care services across hospitals and health service in WA.

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We acknowledge that Curtin University works across hundreds of traditional lands and custodial groups in Australia, and with First Nations people around the globe. We wish to pay our deepest respects to their ancestors and members of their communities, past, present, and to their emerging leaders. Our passion and commitment to work with all Australians and peoples from across the world, including our First Nations peoples are at the core of the work we do, reflective of our institutions' values and commitment to our role as leaders in the Reconciliation space in Australia.

LIST OF PUBLICATIONS

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ASSOCIATED ORAL AND POSTER PRESENTATIONS

Oral Presentations

Iten, R., O'Connor, M., Gill, F. J. *Learning from our littlest ones – The care management trajectories of infants with life-limiting conditions who died.* Congress on Innovations and Leadership in Nursing and Midwifery, Perth, WA, May 12, 2023.

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STATEMENT OF CONTRIBUTION OF OTHERS

This thesis contains published work, all of which has been co-authored. The details of the work, and an estimated percentage of contribution are listed below:

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Co-author acknowledgement:	**Conception and design**	**Acquisition of Data and Method**	**Analysis and Statistical Method**	**Interpretation and Discussion**	**Provide feedback on manuscript**
Co-author 1 Fenella Gill	✓	✓	✓	✓	✓
I acknowledge that this represents my contribution to the above research output and I have approved the final version. Signed:					
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We confirm permission has been obtained from all authors to include the manuscript in this thesis.

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GLOSSARY OF TERMS

For the context of this study the following terms and abbreviations are defined:

Child or children includes neonates, infant/s, child/ren and adolescent/s up to 18 years of age.

End-of-life care involves care provided in the days or weeks leading up to a person’s death. Involves management of symptoms, and support to the family.

Family includes parents, carers, siblings, and grandparents.

Health professional includes members part of the multidisciplinary team such as nurses, doctors, social worker. In some cases, the term nurses and doctors will be specifically used.

Infant includes the term neonate/s under 12 months of age. The term infant used throughout this thesis relates specifically to infant/s and neonate/s.

Life-limiting condition (LLC) is defined as a condition/s that may cause illness and disability that progresses over a period of time which may result in the possibility of death in childhood, where cure may not be possible or may fail.

Perinatal palliative care can commence during pregnancy, when a life- limiting condition is diagnosed, continues during delivery and newborn period.

Perinatal period is defined as 20 weeks gestation and up to 28 days after birth.

UK refers to the United Kingdom.

US and USA refer to the United States and the United States of America.

WA refers to Western Australia.

CHAPTER 1

1.1 Introduction

The World Health Organization estimates that there are up to 21 million children (neonates, infants and adolescents) who require palliative care each year (World Health Organization, 2018). International reports have shown infants have the highest prevalence of death in childhood, with over 4 million infant deaths reported globally in 2017 (Fraser, Gibson-Smith et al., 2020; National Centre for Health Statistics, 2020; World Health Organization, 2019). It is important to note that not all infants who die require palliative care due to the nature of their death, for example infants who die suddenly or unexpectedly (World Health Organization, 2019).

Palliative care has been recognised as a global health issue and considered a human right. Palliative care is an approach to care that aims to the enhance quality of life for adults and children with life-limiting or life-threatening conditions. The World Health Organization reported that only 14% of the population worldwide who need palliative care will receive it (World Health Organization, 2023). For children palliative care is defined as:

> *…the total care of the child's body, mind, and spirit, and also involves giving support to the family. It begins when illness is diagnosed and continues regardless of whether or not a child is receiving treatment directed at the disease (World Health Organization, 2023, Key facts, para. 1).*

The overarching philosophy of palliative care for children is to provide holistic and family centred care, prioritising quality of life through effective symptom management (Palliative Care Australia, 2018; World Health Organization, 2018). Palliative care is recognised as being different from other medical specialties in that it not only focuses on the physical symptoms of the child but also addresses the psychosocial, emotional and spiritual needs of the child and their family (Palliative Care Australia, 2018; World Health Organization, 2018, 2023).

Palliative care as a specialty dates from the 1960s. The origin acknowledges the pivotal work of English nurse, social worker and later physician Dame Cicely Saunders, for her influence in transforming the care of terminally ill adult patients with a proactive approach for managing pain and a holistic care approach embracing the person’s psychological, spiritual, and social care (Sisk et al., 2020). The term palliative care was used to describe this work, leading to the emergence of adult hospices in the United Kingdom (UK), the United States of America (USA) and Australia (Sisk et al., 2020).

For children, the palliative care movement began in the late 1970s, and in the early 2000s, there was establishment of paediatric and perinatal palliative care specialty (Hynson & Sawyer, 2001; Sisk et al., 2020). During this time, despite the recognition of the need to improve a child’s suffering through proactive symptom management and a total care approach of the child, translation into clinical practice was slow to occur (Sisk et al., 2020). Much of the early literature and practice focused primarily on children with cancer and on managing care of dying children rather than a continuous care approach for a life-limiting condition (LLC) diagnosis (Sisk et al., 2020). Only later in the 1990s, authors began to report how palliative care could benefit children with a range of non-cancer LLCs (Sisk et al., 2020). This expansion reflected recognition of a need for palliative care across a broader range of diagnoses other than cancer, such as cardiac and neurological conditions (Sisk et al., 2020). These early studies also highlighted several key inadequacies and areas for improvement in palliative care provision, such as the need to support families, improve communication between the family and health professionals, reduce gaps in provision of bereavement care and develop health professionals’ confidence in delivering palliative care through education (Contro et al., 2002; Contro et al., 2004; Wolfe, Grier, et al., 2000; Wolfe, Klar, et al., 2000).

Since the 2000s, there was further progression in the specialty of paediatric palliative care through development of guidelines and principles published by the American Academy of Pediatrics (2008), a curriculum for educating health professionals (2004), and a shift in research focus to build the evidence base of palliative care for children (Sisk et al., 2020). Over the last decade, paediatric palliative care has become increasingly recognised by health professionals and health services as an essential element of comprehensive care for children with LLCs (Sisk et al., 2020).

Despite the recognition and higher profile afforded to paediatric palliative care, Sisk et al. (2020) highlighted several ongoing challenges that included lack of palliative care education for health professionals, inadequate funding to support research, inequitable access to palliative care teams, and the need for better integration of specialist palliative care in health services. In practice these challenges may result in suboptimal palliative care including ineffective symptom management, increased hospitalisations, lower quality of life and inadequate psychosocial care for the family (World Health Organization, 2023).

1.1.1 Specialist Paediatric Palliative Care Teams

The key roles of specialist paediatric and perinatal palliative care teams in hospital and community settings have been articulated by international and national palliative care guidelines (National Institute for Health and Care Excellence, 2016; Palliative Care Australia, 2018; Together for Short Lives, 2013). These include the following:

- assisting with communication between the primary healthcare team and the child's family,
- promoting shared decision making,
- facilitating advance care planning discussions,
- supporting the provision of optimal end-of-life care, symptom management, psychosocial wellbeing of the child and family and,
- bereavement support (Palliative Care Australia, 2018).

Specialist paediatric palliative care teams comprise multidisciplinary health professionals including but not limited to physicians, nurses, and allied health professionals who have undertaken advanced training or acquired clinical expertise and experience in palliative care (Palliative Care Australia, 2018). A number of studies have articulated the importance of early consultation with specialist palliative care teams and the benefits for families, which include improved quality of life through effective symptom management, psychosocial support for the whole family, continuity of care and clear communication between health professionals and family, options for place of care and bereavement support (Contro et al., 2004; Ekberg et al., 2021; Fraser, Bluebond-Langner et al., 2020; Jack et al., 2018; Mitchell et al., 2017). Palliative care best practice requires the primary healthcare team and specialist palliative care team to work collaboratively, and to work in partnership with the family (Ekberg et al., 2021; Palliative Care Australia, 2018). Operationally this means the patient, the child, remaining under the care of their primary healthcare team, with the specialist palliative care team providing consultation (Palliative Care Australia, 2018; Western Australian Department of Health, 2021).

1.1.2 Life-Limiting Conditions

There are a broad range of conditions infants and children may experience which are considered life-limiting or life-threatening (British Association of Perinatal Medicine, 2010; Catlin & Carter, 2002; Department of Health Western Australia, 2015; Together for Short Lives, 2023). Together for Short Lives (2023) is a leading palliative care charity in the United Kingdom and presents four diagnostic categories that describe the different trajectories where infants and children may require palliative care and have been widely adopted. See Table 1.

Table 1. Categories of life-limiting and life-threatening conditions for infants and children

Category	**Example**
Life-threatening or life-limiting conditions for which curative treatment may be feasible but can fail	Palliative care may be considered during treatment or when treatment fails. E.g., cancer, organ failure, extreme prematurity, congenital heart disease
Conditions where premature death is inevitable	There may be periods of intensive treatment aimed at prolonging life. E.g., chromosomal abnormality, bilateral renal agenesis
Progressive conditions without curative treatment options	Palliative care can be provided over the whole trajectory. E.g., severe neuromuscular disorders, severe metabolic conditions, anencephaly
Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health complications and deterioration	Child may have complex medical needs with poor quality of life. E.g., severe cerebral palsy, brain or spinal injury

(Together for Short Lives, 2013, 2023)

The categories can assist health professionals in planning an appropriate care pathway and assessment of palliative care needs (Catlin & Carter, 2002; Together for Short Lives, 2013, 2023). Although the categories are useful, it is recommended to individually assess the need for palliative care with frequent re-assessments as the child’s condition may change rapidly (Together for Short Lives, 2013).

The prevalence of children with LLCs has increased over the last two decades (Fraser, Gibson-Smith, et al., 2020). Data from the UK reported the prevalence of children aged 0-19 years with LLCs had increased from 26.7 per 10,000 in 2001 to 66.4 per 10,000 in 2020 (Fraser, Gibson-Smith, et al., 2020). Internationally and in Australia infants comprise of the highest number of deaths in children and account for the highest prevalence of LLCs (Fraser, Gibson-Smith, et al., 2020; National Centre for Health Statistics, 2020; Australian Institute of Health and Welfare, 2022). The leading causes of deaths are attributed to conditions secondary to preterm births, and congenital abnormalities (Australian Institute of Health and Welfare, 2022; National Centre for Health Statistics, 2020; Office for National Statistics, 2020).

Some changes in health care that may account for the increased prevalence include medical and technological advancements that have improved antenatal detection of abnormalities, obstetric care and neonatal care, including the care for premature infants at the margins of viability (British Association of Perinatal Medicine, 2010; Moro et al., 2006). Opportunity to be admitted to intensive care has also increased, attributed to advancements in medical treatment. A UK report found the number of children admitted to intensive care units increased by 15% between 2004 and 2013 (Birchley et al., 2017). Furthermore, Fraser and Parslow (2018) reported children with LLCs accounted for 73% of deaths in a UK paediatric intensive care unit between 2005 and 2015. The majority of deaths in paediatric or neonatal intensive care units are reported to be preceded by withdrawal or withholding of treatment (Agra-Tunas et al., 2020; Birchley et al., 2017; Inwald, 2008; Moore et al., 2008).

Reports of increasing prevalence of children with LLCs have highlighted the increasing demand for palliative care services (Bowers et al., 2022; Fraser, Gibson-Smith, et al., 2020). To support the growing demand, there is increasing priority placed on the generalist palliative care concept as a sustainable model of palliative care (Gardiner et al., 2012; Marc-Aurele & English, 2017; Quill & Abernethy, 2013). This model promotes the capabilities of generalist health professionals such as general practitioners, nurses in hospitals as well as in community settings to deliver palliative care, with the involvement of specialist palliative care teams reserved for complex cases (Gardiner et al., 2012; Quill & Abernethy, 2013). Furthermore, the generalist model recognises that not all health services and countries will have access to specialist palliative care teams, particularly in developing countries and community and regional settings (Quill & Abernethy, 2013). Neonatology specialists Marc-Aurele and English (2017) discussed the Institute of Medicine’s recommendations for neonatal intensive care units in the USA to adopt a mixed model of consultation by palliative care teams and primary palliative care delivered by treating teams. This mixed model involves specialist palliative care team consultations for cases with challenging decision making, refractory symptom management and complex transition beyond the neonatal intensive care units (Marc-Aurele & English, 2017).

However, challenges to the generalist model have been reported including lack of integration of services (Gardiner et al., 2012; Robinson et al., 2022).

1.1.3 Key Principles of Paediatric Palliative Care

Irrespective of who delivers palliative care for children and their families several key principles and standards for delivering quality paediatric palliative care and end-of-life care have been published by leading organisations to inform best practice (Australian Commission on Safety and Quality in Health Care, 2016, 2021; Together for Short Lives, 2013; Western Australian Department of Health, 2021).These principles are summarised in Table 2.

Table 2. Key principles of paediatric palliative care

Child's best interest	Medical treatment should always consider the child's best interest and consider the benefits versus the burdens (Together for Short Lives, 2013).
Holistic care	Encompasses the child's physical, emotional, cultural, spiritual and psychosocial needs (Together for Short Lives, 2013).
Child and family centred care	Promotes health professionals working in partnership with the family and recognises the challenges the whole family; parents, siblings, grandparents may experience (Western Australian Department of Health, 2021).
Communication	Health professionals will seek regular communication with the child and family that is open and honest to establish therapeutic rapport and identify specific family needs. Recognition for child's developmental age and ability to be involved in decision making if desired (Western Australian Department of Health, 2021).
Parallel planning	Involves planning for treatment and care, in parallel to planning for end-of-life care. There are many conditions which may have uncertainty surrounding prognosis, with the possibility of rapid deterioration. This approach allows parents to plan and prioritise their goals (Together for Short Lives, 2013).
Advance care planning	Involves conversations between medical professionals and parents to consider ongoing medical management of a child, taking into consideration their condition, disease trajectory, and quality of life. It allows families time to prepare for and consider their needs, and where they would like to be cared for such as hospital, home or hospice for their child's end-of-life (Together for Short Lives, 2013).
Bereavement support	Bereavement support is a core element of palliative care, it includes supporting the grieving process in the death of a loved one (Breen et al., 2014). Support and interventions may include telephone contact, information and resources to navigate grief, referral to counselling services, age-appropriate support for siblings, and opportunity to meet with the healthcare team to address questions (Breen et al., 2014). An integrated and coordinated approach is recommended that involves assessment of the family needs, and anticipatory support (Kenner, Press, &

	Ryan, 2015). It is reported bereavement support is often missed across adult and paediatric sectors, as a result from lack of bereavement guidelines, not standardised processes, and lack of health professionals' expertise to provide bereavement support (Breen et al., 2014).
Support for health professionals	Access to education, mentorship and psychosocial support for health professionals providing care to children (Western Australian Department of Health, 2021).

Whilst the principles of palliative care can be applied to all individuals across the life span, there are a number of important differences for the paediatric context. The differences primarily relate to the broad range of conditions seen in children, including the rarity of diseases, some of which may be familial; difficulty in diagnosis; decision making about future care; impact on the whole family including financial and emotional burden, and for some young people and their families, additional challenges experienced when transitioning to adult palliative care services (Catlin & Carter, 2002; Hynson & Sawyer, 2001; World Health Organization, 2018; Zhong et al., 2022).

Specific differences in the nature of paediatric LLCs present additional barriers to provision of palliative care. These include that a child's death or end-of-life care can be a rare event for many doctors, nurses and allied health professionals, therefore, individuals may have limited palliative care experience (Hynson & Sawyer, 2001). Consequently, they are potentially reluctant to, and lack confidence in, considering and delivering palliative care (Hynson & Sawyer, 2001). Another reported barrier is the challenge health professionals experience transitioning from cure orientated care to palliative care. For children the time of transitioning is not always clear, in particular when there is uncertainty surrounding prognosis (Mitchell & Dale, 2015). This uncertainty may increase reluctance by health professionals to refer to specialist palliative care services (Hynson & Sawyer, 2001; Twamley et al., 2014).

A key component of palliative care, is advance care planning that involves shared decision making between the parents and health professionals considering the child's best interest to make decisions about future care and preferences for end-of-life care (Birchley et al., 2017). Well documented are challenges discussing advance care planning with families experienced by non-specialist palliative care health professionals (Feudtner et al., 2011; Heckford & Beringer, 2014; Jack et al., 2018; Stark et al., 2008). Some crucial challenges are the emotive nature of the discussions, uncertainty surrounding children's prognosis, lack of medical consensus amongst the healthcare team regarding the child's prognosis, parental disagreement and expectations of medical treatment (Feudtner et al., 2011; Forbes et al., 2008; Heckford & Beringer, 2014; Jack et al., 2018; Stark et al., 2008; Zhong et al., 2022). These findings may explain why advance care planning may not be undertaken at all, or occur very close to end-of-life (Feudtner et al., 2013; Heckford & Beringer, 2014; Stark et al., 2008). Delaying advance care planning discussions can result in continuation of treatment that has become burdensome to the child or care that does not align with family preferences.

Another barrier to providing palliative care for children is the impact of societal beliefs about death and dying, specifically that children should not die before their parents (Scott et al., 2023). The knowledge and attitudes of health professionals and communities have been explored in both paediatric and adult settings, including families and community health providers (De Clercq et al., 2019; Scott et al., 2023). A 2020 systematic review examining health professionals', parents' and communities' knowledge and attitudes towards paediatric palliative care reported a lack of palliative care knowledge, confusion between end-of-life care and palliative care, impact of cultural and religious beliefs, and availability of advanced medical treatments to offer a cure or prolong life (Saad et al., 2022). Further, health professionals felt palliative care would signify to families that they were ”giving up” and foster a lack of hope (Saad et al., 2022). Health professionals' attitudes may contribute to delayed referral to specialist palliative care and lead to suboptimal palliative care for the child and family.

1.2 Statement of Problem

Optimal palliative care provides infants and their families with comprehensive and holistic support, achieved through early integration of palliative care, effective symptom management, timely commencement of advance care planning, and psychosocial support for the whole family (Kenner et al., 2015; Marc-Aurele & English, 2017). However, existing literature has demonstrated there are unique challenges and barriers to the provision of palliative care for the infant population (Catlin & Carter, 2002; Marc-Aurele & English, 2017; Moro et al., 2006). Understanding these challenges is imperative to improving the care they and their families receive.

The purpose of this study is to better understand how palliative care is delivered to infants in Western Australia, explore the challenges which may impact the delivery of palliative care, and provide recommendations to optimise palliative care infants receive.

1.3 Context of the Study

In Australia, specialist paediatric palliative care services are well established in five states. Integral to the role is to work collaboratively with the generalist healthcare professionals who have direct patient contact in the metropolitan and regional hospitals and community settings, providing specialist advice and mentorship (Donovan, Slater, Baggio, McLarty, & Herbert, 2019). Australia is a large geographical landscape and the current population density is 3.4 people per square kilometer (Australian Bureau of Statistics, 2023). The unique contexts mean that children with LLCs and their families may reside in metropolitan, regional or remote areas (Donovan et al., 2019). The largest state in Australia is Western Australia, with a population of approximately 2.8 million spread across 10 health service regions, see Figure 1.

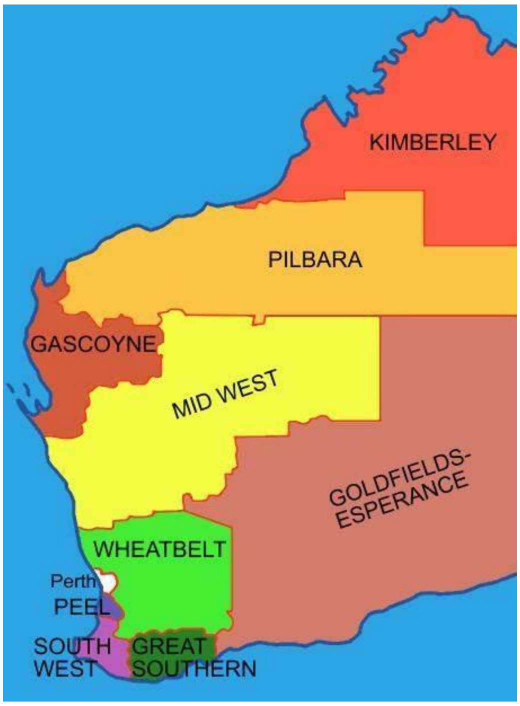

Figure 1. Map of Western Australia regions

(Australian Bureau of Statistics, 2023)

For children and families living in regions beyond Perth metropolitan area, palliative care may be delivered by generalist health professionals, in a variety of health settings, where high rates of health professional turnover are experienced (Western

Australian Department of Health, 2021). This means that the local area knowledge and experience of providing palliative care to children with LLCs is dependent on the individual and may vary substantially (Donovan et al., 2019).

There are two specialist palliative care services providing specialist palliative care provision for WA newborns and children (Department of Health Western Australia, 2015; Western Australian Department of Health, 2021). The paediatric palliative care service for children aged 0 up to adolescence, and the perinatal palliative care service caters for newborns up to the end of the perinatal period (which may commence during pregnancy) (Department of Health Western Australia, 2015). The paediatric palliative care service is a consultative model of care, where referrals are made by the child's primary physician or team (Western Australian Department of Health, 2021). The WA Perinatal Palliative Care Model of Care describes three entry points of referral, these are infants with prenatally diagnosed LLCs, pre-viable preterm births and infants postnatally diagnosed with LLCs (Department of Health Western Australia, 2015). For infants who are exclusively managed as palliative and not expected to survive beyond the hours or days after birth, the parents may choose to receive end-of-life care in the labor and delivery ward or be discharged home with community palliative care support. In other cases an infant will be admitted to the neonatal intensive care unit for ongoing specialist treatment (Department of Health Western Australia, 2015). After admission to the neonatal intensive care unit, or where an infant survives beyond the perinatal period, the paediatric palliative care service may be referred to on a case-by-case basis (Department of Health Western Australia, 2015). A small number of infants discharged home for end-of-life care will receive palliative care support at home through the Silver Chain Hospice service, with support from the perinatal or paediatric palliative care services (Department of Health Western Australia, 2015).

This study was conducted at three tertiary hospitals, catering for the majority of infants with LLCs in WA. The hospitals include a tertiary paediatric hospital (Perth Children's Hospital) where over 30, 000 children are admitted, including 700 neonates each year who are born in other hospitals and require specialist neonatal care (Australian Institute of Health and Welfare, 2020). The second hospital (King Edward Memorial Hospital) is a tertiary women and newborn hospital where approximately 6,500 neonates are born annually (Government of Western Australia, 2021). The third hospital is a tertiary hospital for adults, neonates and children (Fiona Stanley Hospital) where there are approximately 3,300 births and 5,000 children admitted each year (Australian Institute of Health and Welfare, 2020).

1.4 Aim and Objectives

The overall aim of the study was to understand how palliative care is delivered to infants with LLCs, and the involvement of specialist palliative care teams in this population.

There were four objectives to address this aim:

1. To develop a broad understanding of how palliative care is delivered to infants.
2. To identify and characterise the pathways and care management trajectories of infants with LLCs who died within the first year of life and received tertiary care in WA.
3. To map the recorded clinical decision making processes for infants with LLCs, including involvement of parents/family in decision making and other specialist care providers.
4. To make recommendations to optimise palliative care infants receive.

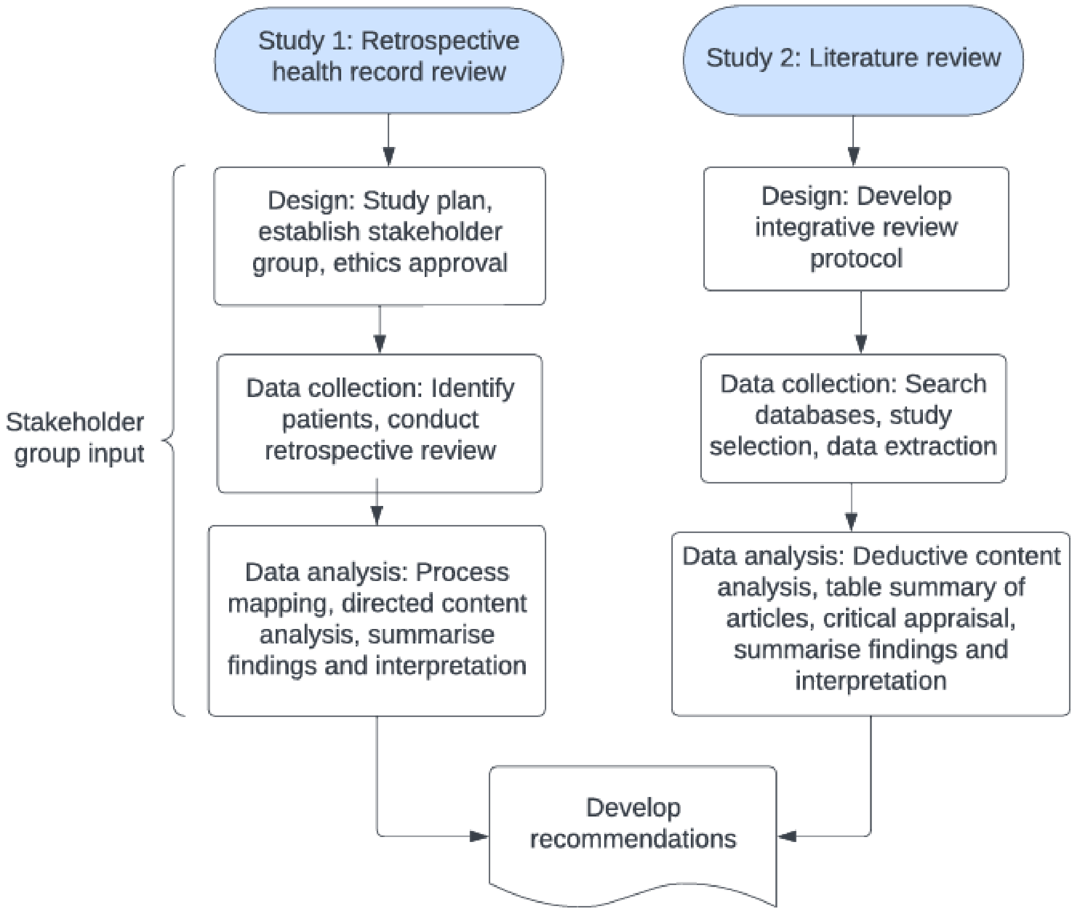

Figure 2. Study 1 and Study 2 approach

The aim of Study 1 was to describe the care management trajectories of infants with LLCs who died before 12 months of age. Study 1 design used a retrospective review of the health records of infants with LLCs who died and received care in three tertiary hospital settings in WA. In Study 2, an integrative review synthesised the literature about palliative care delivery for infants with LLCs.

This study is the first to describe the care management trajectories of infants with LLCs who received care in WA and identify the barriers and facilitators to referral and access to specialist palliative care. The findings will contribute to the current body of evidence to optimise the delivery of palliative care for infants. For health professionals, the research provides an understanding of the heterogeneous infant population and care management trajectories guiding palliative approaches to care and referral to specialist palliative care services. Potential benefits to future infants and their families may include enhanced integration of specialist palliative care services, providing a child and family centred approach, specialist palliative care advice, support with decision making, continuity of care and bereavement support.

1.4.1 Rationale for Research

I am a Registered Nurse and have worked in paediatrics for 11 years across medical and surgical specialties. Over half of my career has been spent in the WA Paediatric Palliative Care Service for six years. In my role as the Palliative Care Nurse Educator, I was responsible for delivering palliative care education to health professionals across WA who are involved in the care of children with LLCs. During this time, I noted and was surprised by how infrequently infants were referred to the service, in particular there were very few infants referred from the neonatal intensive care units. A scoping examination of the morbidity and mortality data at Perth Children's Hospital in 2019 identified more than 80% of infants who received care at the hospital and died did not receive specialist palliative care input (unpublished results). It was also identified that other states in Australia experienced similarly low numbers of referrals of infants to specialist palliative care services (unpublished results). Despite knowing infants comprise the highest number of deaths in childhood and are often diagnosed with LLCs at, or shortly after birth, only a few referrals per year were received. When I explored further, I found similar experiences were anecdotally reported across Australia by other specialist paediatric palliative care teams. Further, in 2020 I initiated and delivered a palliative care education program for nurses working in WA neonatal intensive care units. Feedback from these nurses was that they frequently provided end-of-life care to infants but had not received any specific palliative care education and felt under-prepared to provide palliative care (unpublished results). It was through these experiences that I developed an interest to further explore the area of palliative care for infants for my research degree.

1.5 Outline of the Thesis

This thesis is presented in four chapters, two of these chapters are articles published in peer-reviewed journals.

Chapter 1 – This chapter presents the study context, introduces palliative care for children, describes a background to infants who may require palliative care and some unique challenges of providing palliative care for the infant population, identifies the research problem and study aims and objectives.

Chapter 2 – This chapter consists of a published article titled ‘Care management trajectories of infants with life-limiting conditions – a retrospective patient health record review’. Iten, R., O’Connor, M., Cuddeford, L., & Gill, F. J. (2022) Care management trajectories of infants with life-limiting conditions who died before 12 months of age; a retrospective patient health record review. *Journal of Pediatric Nursing* . https://doi.org/10.1016/j.pedn.2022.11.014.

This publication presents Study 1, the retrospective study methodology and findings.

Chapter 3 – This chapter consists of a published article titled ‘Palliative Care for infants with life-limiting conditions: integrative review’. Iten, R., O’Connor, M., Gill, F. J. (2023) Palliative care for infants with life-limiting conditions: integrative review. *BMJ Supportive & Palliative Care* Published Online First: 09 December 2023. https://doi.ord/10.1136/spcare-2023-004435

This publication, presents Study 2 which synthesised the literature and compared two models of palliative care delivery for infants, explored health professionals’ perspective of barriers and facilitators to providing palliative for care infants and the family experience.

Note: An initial review of the literature was conducted to inform Study 1, then Study 2 was completed second. The reason was a pragmatic decision to enable data collection to be completed before I commenced a period of maternity leave.

Chapter 4 – Presents the discussion which links the two studies and summarises the main findings, details strengths and limitations, presents implications for practice and recommendations for future research.

CHAPTER 2

2.1 Study 1 Retrospective Review

The article “Care management trajectories of infants with life-limiting conditions who died before 12 months of age; a retrospective patient health record review” was published in the Journal of Pediatric Nursing.

Approvals from Institutional and University Human Research Ethics Committees were obtained. A waiver of consent was approved (see Appendix 1 and 2.)

Study 1 design and methodology was based on a study reported by Campling et al. (2018) titled “Escalation-related decision making in acute deterioration: A retrospective case note review’’. Campling’s study described decision making in acute deterioration for adult patients who were hospitalised and died. Data analysis involved process mapping resulting in four care management trajectories which we later adapted for our research. We sought further information from the lead researcher Dr Natasha Campling in November 2021 on the study methodology, specifically to better understand directed content analysis and process mapping. We also sought general advice about the team’s experiences with their stakeholder group and what the care management trajectories have meant for clinical practice. See Appendix 3 for full details.

The collaboration with key health professionals, consumers (bereaved parents), and the research team was a strength of this study. Tapping into the expertise of health professionals from various specialties provided valuable contextual richness to ensure each specialist area was fully understood and facilitated interpretation of the findings. The participation of the bereaved parents was invaluable, adding a depth of understanding and perspectives of their lived experiences. A systematic review identifying the highest ranked principles to optimising consumer involvement were working in partnership, communication, and shared knowledge (Baines & Regan de Bere, 2018). The benefits of consumer participation are strongly recognised to enhance the quality of research, contribute to the translation of research into practice, and can lead to improved patient care (McKenzie & Hanley, 2014; NHMRC, 2016).

2.2 Publication 1

The following contains the article published in November 2022.

Iten, R., O'Connor, M., Cuddeford, L., Gill, F. J. (2022) Care management trajectories of infants with life-limiting conditions who died before 12 months of age; a retrospective patient health record review. *Journal of Pediatric Nursing*. https://doi.org/10.1016/j.pedn.2022.11.014.

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Care management trajectories of infants with life-limiting conditions who died before 12 months of age; a retrospective patient health record review

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ABSTRACT

Purpose: To characterise the care management trajectories of infants with life-limiting conditions, who died before 12 months, including clinical decision-making processes, identification of triggers that led to changes in care management from cure-orientated to palliative care and specialist palliative care team involvement.

Design and methods: Retrospective patient health record review of infants with life-limiting conditions who died before 12 months of age and received care at three hospitals in Western Australia. Two data analysis methods; directed content analysis and process mapping.

Results: A total of 45 patient health records were reviewed. Process mapping led to typology of care management encompassing four trajectories; early de-escalation due to catastrophic event; treatment with curative intent throughout; treatment with curative intent until a significant point; and early treatment limits. Standardised advance care planning processes were used for just over 10% of infants. There was specialist palliative care team involvement for 25% of infants.

Conclusion: Only a proportion of infants received early integration of palliative care principles and practices. Infants and their families may benefit from earlier integration of palliative care, and standardised processes for advance care planning that are done in parallel to treatment.

Practice implications: There is opportunity to further enhance the delivery of palliative care to infants with life-limiting conditions and optimise the experience for families through education for health professionals, implementation of advance care planning and standardisation through policies and clinical practice guidelines.

Background

The highest number of childhood deaths are in infants aged <12 months of age, with three quarters of deaths occurring within the first 28 days of life (Australian Insitute of Health and Welfare, 2019). Furthermore, infants have the highest prevalence of life-limiting conditions (LLC) in childhood (Fraser et al., 2014). The leading causes of infant deaths are congenital malformations and perinatal conditions, including preterm births, low birth weight, birth trauma, and viral infections acquired in utero (Australian Insitute of Health and Welfare, 2019; National Centre for Health Statistics, 2019; Office for National Statistics, 2019).

The care of infants with LLC is complex and impacted by several factors, such as uncertain prognosis, the rarity of diseases, unclear disease trajectories, difficulty in assessment of symptoms and needs, and lack of continuity of care (Kukora et al., 2017; Marc-Aurele & English, 2017; Moro et al., 2006). In addition to the complexity of care, there are increased numbers of infants living with life-limiting and life-threatening conditions, secondary to medical and technological advancements in healthcare which have improved antenatal detection of anomalies, obstetric care, and neonatal care (Moro et al., 2006). Several retrospective studies have reported these infants were often managed in intensive care environments, with a high proportion receiving invasive interventions such as cardiopulmonary resuscitation (CPR) before end-of-life (Bolognani et al., 2020; Fontana et al., 2013; Gibelli et al., 2021). Deaths in the neonatal intensive care unit are commonly preceded by a decision to withdraw or withhold life-sustaining treatment (Audigé et al., 2020; Fontana et al., 2013). The impact on families who

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have an infant with a LLC requiring intensive care is profound. Families are faced with uncertainty about prognosis, lengthy hospital stays, invasive procedures, the prospect of sudden deterioration, ethical issues, and the ever-present possibility of death (Mitchell et al., 2019; Quinn & Gephart, 2016).

Palliative care for children is recognised as a total approach to care which begins at diagnosis of a LLC and can be provided in parallel to treatment, that aims to improve the quality of life of children experiencing a LLC through assessment and treatment of physical symptoms, as well as addressing psychosocial and spiritual needs for the family (World Health Organisation, 1998). Several infant diagnosis categories have been presented for palliative care consideration. In summary these reflect the following; infants with prenatally diagnosed anomalies or LLC; infants born preterm; and infants postnatally diagnosed with a LLC (Catlin & Carter, 2002; Together for Short Lives, 2017). A rapid review of the literature reported early integration of palliative care provided in parallel with treatment that is curative or aimed at prolonging life can improve access to palliative care for these infants in the categories described (Ekberg et al., 2021). This parallel approach aligns with delivering family centred care, prioritises quality of life and comfort and includes the provision of bereavement support (Kenner et al., 2015; Palliative Care Australia, 2018).

Specialist perinatal and paediatric palliative care teams, comprise interdisciplinary health professionals; physicians, nurses, midwives, allied health and chaplaincy professionals working together who all have advanced training and/or clinical experience in paediatric palliative care (Palliative Care Australia, 2018). They provide consultation and work in partnership with primary healthcare teams to support the delivery of palliative care, enhance the quality of life of the infant or child and their family by assisting with communication between teams, facilitating advance care planning (ACP), providing psychosocial and emotional support and coordination of family bereavement support (Palliative Care Australia, 2018). Internationally, there are reports from several countries of infrequent involvement of specialist palliative care for infants with LLC, in particular infants born preterm or managed in neonatal intensive care settings (Bolognani et al., 2020; Harmoney et al., 2019; Ramelet et al., 2020). The reasons for infrequent involvement of specialist palliative care remain unclear, although one study suggested it may be attributed to uncertain prognosis and shorter periods in hospital for infants managed in neonatal intensive care (Stutz et al., 2018).

An essential component of palliative care is supporting families with decision making. Advance care planning is a process that involves discussions between interdisciplinary professionals and families, that provide the opportunity for healthcare teams to guide the family to consider the possibility of acute deterioration, and how best to manage these events that considers the family goals of care, and preferences for end-of-life care (National Institute for Health and Care Excellence, 2016; Palliative Care Australia, 2018; Sidgwick et al., 2019). Triggers for ACP may include diagnosis of a LLC, when death is likely in the short or medium future, significant deterioration during hospital admission, recurrent or unplanned hospital admissions or at parental request (Australian Commission on Safety and Quality in Health Care, 2016). Advance care planning can be commenced in parallel to treatment aimed at cure or prolonging life (Sidgwick et al., 2019). Without these discussions families may not be afforded the opportunity to prepare for these events or consider their preferences and priorities (Horridge, 2015).

In practice, commencing ACP in parallel to treatment presents challenges for health professionals and barriers transitioning from cure-orientated care to palliative care have been reported. These include health professionals' reluctance to begin conversations with families (Catlin & Carter, 2002;Jack et al., 2018; Mitchell & Dale, 2015), the uncertainty of prognosis, moral and ethical dilemmas, disagreement amongst medical teams (Mitchell & Dale, 2015), and fear of causing parental distress (Marc-Aurele & English, 2017). In addition lack of formal training in ACP and associated communication skills, time constraints and competing clinical demands have been reported to contribute to challenges of delivering ACP (Jack et al., 2018; Mitchell & Dale, 2015). These factors may reduce the opportunity to integrate palliative care principles and practices to optimise the care for infants and families (Kukora et al., 2017; Marc-Aurele & English, 2017). Several studies have reported infants and children who received specialist palliative care, were more likely to die at home or outside of intensive care settings, receive less invasive interventions before end-of-life, with documented ACP including preferences for end-of-life care, and increased bereavement support (Audigé et al., 2020; Charlebois & Cyr, 2015; Fraser et al., 2018; Stutz et al., 2018). A systematic review of the literature examined the benefits of specialist palliative care which included bereaved parents' experiences were reported as improved quality of life through the provision of psychosocial and emotional support, guidance with decision making, improved symptom management and support for preferred place of care (Mitchell et al., 2017). Currie et al. (2016) interviewed bereaved parents to identify their perspectives of end-of-life care and palliative care consultation in the neonatal intensive care unit, and they reported specialist palliative care provided an added layer of support and advocacy.

The aim of this study was to explore and characterise the care management trajectories of infants with LLC, who die within their first year of life, to map the recorded clinical decision-making processes, including involvement of families, and identify possible triggers that led to changes in care management from cure-orientated to comfort focused and if or when specialist palliative care teams were involved.

Methods

Design

A retrospective health record review explored and characterised the care management trajectories of infants with LLC who died within their first year of life. The study design and methodologies were adapted from Campling et al. (2018), who reported the complex care management and decision making for adult patients who deteriorated and died during an acute hospital admission. Approvals from Institutional (RGS4033) and University (HRE2020–0676) Human Research Ethics Committees were obtained. A waiver of consent was approved. Reporting guidelines followed were; Reporting of studies Conducted using Observational Routinely-collected health Data (RECORD) (Supplementary File 1) (Benchimol et al., 2015) and the Standards for Reporting Qualitative Research (Supplementary File 2) (O'Brien et al., 2014).

Health professionals and consumers stakeholder group

A stakeholder group, comprised of health professionals and health consumers, guided the study conduct, informed data collection, and contributed to the interpretation and verification of findings. Health professionals included three medical, three nursing, and one midwifery professional in the areas of neonatology, specialist paediatric and perinatal palliative care. Two health consumers were bereaved parents; one with lived experience of specialist palliative care, and one whose child died unexpectedly. Consumer involvement was integral to the study at every stage and was key in supporting the research ethics application for waiver of consent for bereaved families.

Study setting

The study was conducted at three tertiary hospitals where specialist care was provided for neonates and infants in metropolitan Perth, Western Australia (WA). Annually, approximately 6500 neonates are born at site one; the women's and newborns' hospital (Government of Western Australia, 2021), approximately 3300 born at site two; a tertiary hospital for adults, neonates and children (Australian Institute of Health and

Welfare, 2019), and over 700 neonates admitted for specialist neonatal care to site three; the children's hospital (Australian Institute of Health and Welfare, 2020). In WA, specialist palliative care services are available through a perinatal palliative care team and a paediatric palliative care team. Perinatal palliative care caters for prenatally diagnosed anomalies, pre-viable preterm births, and newborns with postnatally diagnosed LLC. Paediatric palliative care may be requested to consult on a case-by-case basis for infants who survive beyond the perinatal period (28 days after birth) (Department of Health Western Australia, 2015). Palliative care and end-of-life care in paediatric and neonatal intensive care units is primarily provided by paediatric intensive care and neonatal intensive care healthcare teams.

Sampling strategy

Patients were identified from the hospitals' death databases following registered service during a one-year period, 2018–2019, at the three hospitals. Inclusion criteria: (a) infant alive for > 24 h, (b) infant died before 12 months of age. Exclusion criteria: (a) sudden infant death syndrome, (b) records or death data not available.

Data collection

A data collection tool was developed with input from clinicians and further reviewed by the stakeholder group to enable extraction of items to map timelines, trajectories of care, key events and interventions (Supplementary File 3). Data were collected from birth through to death and bereavement, these data included clinical and demographic information, diagnosis category, documented specialist palliative care team involvement, treatment and interventions, ACP, end-of-life care, preferred place of care and family bereavement support. Data were managed using REDCap electronic data capture tool (Harris et al., 2019). One researcher was a paediatric nurse with qualifications and experience in specialist paediatric palliative care, the second researcher was a paediatric nurse academic, and the third research member was an experienced palliative care researcher. Two researchers tested the data collection tool for feasibility using a random sample of three patient health records. No changes were made. One researcher extracted data from the patient health records.

At the time of data collection, no standardised ACP forms or proformas were used in WA. Evidence of ACP was assessed through documentation of medical management and family involvement. Family preferences for end-of-life care were assessed through documented spiritual, religious and cultural requirements which included customs and beliefs, religious groups or practices, that may influence care provided (National Institute for Health and Care Excellence, 2016). Timelines were depicted simultaneously to the patient health record review. Field notes captured limitations and data that did not fit within the data collection tool.

Data analysis

Two methods of analysis were applied concurrently.

Directed content analysis

First, extracted data were tabulated and analysed through directed content analysis (Assarroudi et al., 2018; Hsieh & Shannon, 2005). Key concepts to guide analysis were drawn from international and national palliative care standards (National Institute for Health and Care Excellence, 2016; Palliative Care Australia, 2018), and included infant palliative care categories, family centred care, specialist palliative care involvement, ACP and end-of-life care. Patient health records were read multiple times with data directly entered into REDCap using checkboxes, drop-down lists, or transcribed verbatim. Tabulated data and timelines were focused on events that led to infants receiving hospital care after birth; details of treatment and outcomes; events that led to the infant's death; decision making between healthcare teams and families and the care management before end-of-life. Data from field notes were included in the analysis.

Process mapping

Process mapping (Trebble et al., 2010) was achieved through analysis of the depicted timelines and led to grouping patients into one of four care management trajectories, based on Campling et al. (2018)'s four care management trajectories. Together the research team discussed each patient's timeline and allocated a trajectory according to infants' diagnosis category and clinical management. Diagrams were depicted to represent the group experience of the care management trajectories and the sequence of events with a focus on what occurred in each phase.

Health professionals and consumer stakeholder group

The health professionals and consumer stakeholder group verified and contributed to the interpretation of the findings through review and provision of feedback on tabulated data and a case example representing each trajectory. The following questions guided discussion with:

a) The health professionals: Do the care management trajectories accurately represent infants you have been involved in the care of recently; Could you consider how they do or if they don't; Do these demonstrate potential triggers for decision making or referral to specialist palliative care; Could these be applied to clinical practice; Is there anything in the data that you were surprised by or any other comments.
b) The health consumer: Do the care management journeys presented reflect the lived experiences as a parent who has had a child with a life-limiting condition; Can you relate to any of the categories or themes presented from your own lived experience; What are your thoughts or recommendations on the triggers for initiating palliative care based on your experience; Do you have any thoughts on the usefulness of using the triggers identified; Are there any pros or cons.

Seven of the eight health professionals and one health consumer provided feedback (in-person, email, and video meetings) detailed in supplementary file 4. The health professionals feedback confirmed that the patients in this study accurately reflected their encounters in clinical practice. The health consumer provided feedback based on personal experience. There were, however, contrasting views about categorising infants into the four trajectories and application to clinical practice. Two elements robustly discussed were ACP and delivery of palliative care.

Feedback regarding ACP consisted of mixed views. For example, the neonatal physicians identified practical challenges to ACP for the majority of infants in neonatal intensive care. They highlighted that infants' conditions were acute, infants were being managed with curative intent, and their own views were that parents did not want to discuss ACP, especially if there was hope the infant may survive. In contrast, the specialist palliative care team perspective was that ACP and parallel planning supported families to prepare for the death of their infant and documenting these plans can reduce the potential for repetition of distressing conversations and provide a baseline for future conversations. Additionally, neonatal intensive care nurses explained that ACP was inconsistently discussed with families by medical staff, introducing palliative care to families was not spoken about often enough and seemed to be dependent on the individual leading care. One nurse suggested that a lack of a formal policy guiding ACP or referral to specialist palliative care teams may contribute to inconsistent practices. The health consumer advocated the benefits of ACP on the family experience and explained how a documented ACP can reduce repetition of distressing conversations when there is frequent rotation of staff. ACP also provides families the opportunity to plan for the possibility of the infant's death, considering their preferences which can provide families with a sense of control and understanding of what the future may bring.

Differing opinions were also expressed about the timing and referrals to specialist palliative care teams. The neonatal physicians explained that for infants in intensive care environments, where lead clinical teams provide all aspects of care including palliative care, specialist palliative care teams are only required for consultation of patients who have complex care needs. From their perspective, the role of specialist palliative care teams is limited to delivering education for health professionals in intensive care. In contrast, although the palliative care specialist team agreed that specialist palliative care may not be required for all infants with LLC, their perspective was that infants with life-limiting or life-threatening conditions can benefit from receiving care that incorporates palliative care principles and practices early in the disease trajectory. Furthermore, early integration of palliative care principles or referral to specialist palliative care teams prompts a refocus of priorities of care, consideration for what is important to the family and where they want to be cared for at end-of-life. The health consumer identified a potential barrier to integrating palliative care early can be family and health professionals' own misperceptions of palliative care; being that palliative care is a sign of giving up or for when there is nothing else left to do.

Results

Sample characteristics

There were 55 patients identified, of these 10 were excluded due to incomplete data or records not available. A total of 45 patients were included: 17 from site one, two from site two, and 26 from site three. There were 15 females and 30 males, age range at time of death was one-315 days. The length of hospital admission ranged from one-315 days. Patient diagnosis categories were prenatal anomaly diagnosis, nine (20%), born preterm, 17 (38%), and postnatal diagnoses of a LLC, 19 (42%).

Specialist palliative care teams were involved for 11 (24%) patients. For 37 (82%) patients ACP was documented in the patient health records, and for five (11%) ACP was documented in palliative care plans used by specialist palliative care teams. Family preferences and priorities for end-of-life care were documented for 27 (60%) patients, for 14 (31%) there was no documentation, and for three (7%) not applicable due to sudden and unexpected death. Of the 45 infant deaths, 40 (89%) deaths were in hospital; 10 (22%) in a paediatric intensive care unit, 28 (62%) in a neonatal intensive care unit, one (2%) in an inpatient ward, and one (2%) in a regional hospital. Five (11%) infants died at home. Bereavement follow up was inconsistently documented; 24 (53%) families had one or more events of documented bereavement support, and for 21 (46%) there was no documented bereavement support in the patient health record. See Table 1 Key characteristics of the patients within the trajectories.

Typology of care management

Four care management trajectories captured the events and clinical management from birth to death.

Trajectory 1 Early de-escalation due to catastrophic event ($n = 11$)

Trajectory 1 was characterised by patients who experienced a catastrophic event (cardiac arrest, sepsis, brain injury) resulting in admission to paediatric or neonatal intensive care (See Fig. 1 Care management trajectory type 1). The catastrophic event occurred on a median of day two from birth, with five (45%) patients admitted to intensive care on day one.

Initial escalation and treatment included interventions such as cardiopulmonary resuscitation (CPR), intubation, ventilation, advanced life support, and a variety of clinical investigations. The timepoint of recognition of irreversibility of the LLC varied from <24 h to 19 days. The variation in time was attributed to when investigation results became available and to clinical protocols for the acute management of hypoxic-ischemic encephalopathy over the first few days of life. Factors that triggered recognition of irreversibility were investigation results (MRI, CT), patient dependency on mechanical ventilation, and progressive worsening of clinical condition. These factors triggered healthcare team discussions with parents and families to explain the prognosis and likely outcomes for the patient which included the prospect of severe disability and death.

A change in clinical management to palliative care was introduced by the healthcare team for all patients and families. The time taken for families to process information and to reach an agreement with the healthcare team's recommendation for re-direction of care was up to three days. Plans were documented in the patient health records for de-escalation for all patients. Death followed de-escalation; 10 patients died immediately following the withdrawal of ventilation. All deaths were expected, occurring between day three-28. All but one patient died in the intensive care unit; one patient had ventilation support withdrawn in the garden of the hospital at the request of parents and this was the only patient in trajectory type 1 who was referred to the specialist palliative care team.

Trajectory 2 Treatment with curative intent throughout ($n = 4$)

Trajectory 2 was characterised by patients with a prenatally diagnosed LLC (hypoplastic left heart) and three born at the threshold of viability (23 + 2, 24- and 25-weeks gestation), with treatment focus on curative intent throughout (See Fig. 2 Care management trajectory type 2). All patients were admitted to intensive care from day one. The patients in this group were the youngest to die, with median age at death nine days and received the most invasive treatments and interventions before end-of-life.

A key feature in this trajectory was the sudden and unexpected deterioration (apnoeas, bradycardia, low cardiac output related to sepsis) resulting in acute escalation of care including CPR for all patients, who all subsequently died. De-escalation did not occur and following the sudden deterioration, death occurred within minutes to hours. For one patient, there was documentation by the medical team that they were clear with parents the patient would not survive and discussed redirection of care to palliative care. Despite this discussion, the parents wished to continue curative treatment, and the patient subsequently deteriorated, and CPR was attempted, albeit unsuccessfully. For one patient, there was a "not for resuscitation plan" (not for CPR or adrenaline) documented, however, when the patient suddenly deteriorated CPR was attempted. There was no documented rationale to explain the decision to resuscitate the patient. One patient with the hypoplastic left heart condition was being prepared for discharge home when they suddenly deteriorated and suffered a cardiac arrest. All patients died in an intensive care setting on days two-25. There were nil referrals to specialist palliative care team for this group.

Trajectory 3 Treatment with curative intent until a significant point ($n = 21$)

Trajectory 3 was the largest group, characterised by patients with postnatally and prenatally diagnosed LLC (congenital heart disease, cardiomyopathy, and metabolic conditions) and infants born preterm (14 were < 28 weeks, one <32 weeks, and one <34 weeks). Treatment aims were curative intent until a significant point that triggered de-escalation. (See Fig. 3 Care management trajectory type 3). All the patients were admitted to intensive care from day one.

A feature for this group was a long stay in hospital; a median of 11 days, range two-315 days (two patients were hospitalised for >200 days). All patients had multiple health comorbidities, increasing their

Table 1
Key characteristics of the patients within the trajectories.

Trajectory 1 patients (n = 11)			Trajectory 2 patients (n = 4)		
Gestational age[†]	Early preterm	1	Gestational age[†]	Very preterm	1
	Preterm	1		Early preterm	2
	Term	9		Term	1
Age at death (median, IQR)	14 (IQR 7–23) days		Age at death (median, IQR)	9 (IQR 5–17) days,	
Diagnosis	Hypoxic ischemic encephalopathy	5	Diagnosis	Prematurity	3
	Cardiac arrest	1		Hypoplastic left heart	1
	Sepsis	3			
	Subgaleal haemorrhage	1			
Comorbidities[‡]	Seizures	4	Comorbidities[‡]	Hyaline membrane disease	3
	Organ failure	2		Pulmonary atresia	1
	Raised intracranial pressure	1		Pulmonary hypertension	1
	Sepsis	1		Hypotension	1
	Intracranial haemorrhage	1		Sepsis	1
	Hypovolemic shock	1		Multi organ failure	1
	Pulmonary hypertension	1		Patent ductus arteriosus	3
	Hypoglycaemia	1		Metabolic acidosis	1
				Intravascular coagulation	1
Length of admission (median, IQR)	7 (5–9) days		Length of admission (median, IQR)	9 (5–17) days	
Triggers for recognition of irreversibility / un-survivable event	Investigations (MRI)	10	Reason for no de-escalation	Acute care	3
	Severe neurological impairment	11		Palliation not discussed	2
	Dependency on ventilator	10		Parental request	2
Key interventions	Intubation / re-intubation	1	Key interventions	CPR[§]	3
	Advanced life support	1		Intubation / re-intubation	3
	CPR[§]	1		Adrenaline	2
	Withdrawal of ventilation	10			
EOLC[¶] preferences & priorities	For baptism or blessing	3	EOLC[¶] preferences & priorities	Not applicable[◊]	2
	Memory making; photos, bath, cuddles	3		No documentation	2
	Withdrawal of ventilation outside	1			
	For siblings to visit in NICU	1			
	Comfort	4			
	To hold baby before EOL	1			
	For family to be present	1			
	No documentation	2			
Bereavement follow up	1[*]	1	Bereavement follow up	1[*]	0
	>1	4		>1	1
	Nil	4		Nil	3

Trajectory 3 patients (n = 21)			Trajectory 4 patients (n = 9)		
Gestational age[†]	Extreme preterm	14	Gestational age[†]	Term	9
	Very preterm	1			
	Early preterm	1			
	Term	5			
Age at death (median, IQR)	25 (3.5–49.5) days		Age at death (median, IQR)	14 (5.5–142.5) days	
Diagnosis	Extreme prematurity	14	Diagnosis	Trisomy 18	1
	Pulmonary hypertension	1		Nonketotic hyperglycinemia	1
	Cardiomyopathy	1		Severe methylene	1
	Ornithine transcarbamylase deficiency	1		tetrahydrofolate reductase	1
	Severe pulmonary hypertension	1		deficiency (MTHFR)	1
	Congenital heart disease	3		Common truncus arteriosus	1
				Congenital fibre type	1
				disproportion	1
				D-bifunctional protein deficiency	1
				Hypoplastic left heart	
				Myotubular myopathy	
				Critical aortic & mitral stenosis	
Comorbidities[‡]	Necrotising enterocolitis	3	Comorbidities[‡]	Global delay	1
	Multiorgan failure	4		Microcephaly	1
	Mitral valve dysplasia	1		Failure to thrive	1
	Ventricular septal defect	1		Hypotonia	1
	Chronic lung disease	3		Seizures	1
	Oropharyngeal dysplasia	1		Obstructed breathing	1
	Metabolic encephalopathy	1			
	Hyperammonaemia	1			
	Pulmonary hypertension	10			
	Metabolic bone disease	1			
	Hypothyroidism	1			
	Growth failure	1			
	Bronchiolitis	1			
	Sepsis	4			
	Lung hypoplasia	2			
	Heterotaxia	1			
	Asplenia	1			
	Intracranial haemorrhage	2			

(continued on next page)

Table 1 (*continued*)

Trajectory 3 patients (n = 21)			Trajectory 4 patients (n = 9)		
	Systemic hypertension	1			
	Hyaline membrane disease	8			
	Patent ductus arteriosus	1			
	Pulmonary haemorrhage	2			
	Apnoeas	1			
	Gastrointestinal perforation	1			
	Pulmonary stenosis	1			
	Bronchopulmonary dysplasia	1			
	IUGR	1			
Length of admission (median, IQR)	11 (2–35) days		Length of admission (median, IQR)	3.5 (2−10) days	
Significant point triggering de-escalation"	Nil improvement	4	Trigger for early treatment limits"	Prenatal diagnosis	5
	Deterioration despite max treatment	18		Postnatal diagnosis	4
	Clinical investigations	2		No curative treatment options	8
	Dependency on ventilator	11		Severe disability with poor quality of life	1
	Multi organ failure	4			
	Sepsis	4			
	Metabolic acidosis	1			
	Cardiorespiratory arrest	2			
	Poor cardiac function	1			
	Neurological impairment likely	3			
	Futility of treatment	1			
	Recognition we are at the end point	3			
Key interventions	Withdrawal of ventilation	19	Key interventions	Withdrawal of ventilation	2
	Intubation / re-intubation	1			
	Advanced life support	2			
	CPR[§]	2			
	Admission to ICU	1			
EOLC preferences & priorities	Comfort	4	EOLC preferences & priorities	Spend quality time together	1
	Hold baby before EOL	5		Focus on comfort	4
	Give baby a bath	2		To go home	5
	Spend time together as family	1		Being together as family	2
	Strong Islamic faith	1		Take baby to beach; put feet in water	1
	Support for sibling	1		Breast feed baby	1
	Parents requested twins to be together	1			
	No documentation	10			
Bereavement follow up contact	1[±]	5	Bereavement follow up	1[±]	0
	>1	6		>1	5
	Nil	10		Nil	4

[†] Gestational age: Preterm birth is defined as birth before 37 completed weeks of pregnancy; early preterm birth (<34 weeks); very preterm (28 to 32 weeks); extreme preterm (<28 weeks) (Commonwealth of Australia, 2019; World Health Organisation, 2012)

[‡] Some infants had multiple comorbidities

[§] Cardiopulmonary resuscitation

[¶] End-of-life care

[ρ] Preferences and priorities were not applicable due to sudden and unexpected death

[±] Bereavement follow up occurred at least once - within the 1st week following death

["] Some patients had more than one trigger

risk of health complications, involving multiple treating healthcare teams. Interventions and treatments included intubation and ventilation, medications, advanced life support, surgical procedures, and multiple investigations.

Triggers for de-escalation were when patients showed no signs of improvement or continued to deteriorate despite the maximal level of treatment. These included worsening cardiac function, decreasing oxygenation, dependency on mechanical ventilation, metabolic acidosis, sepsis, multi-organ failure and, results from investigations (MRI) confirming the extent of brain injury. These triggers prompted healthcare team discussions with parents for re-direction of care to end-of-life care. Plans were documented in the patient health records for de-escalation for all patients. De-escalation measures included weaning and withdrawal of ventilation, implementing plans for no resuscitation, and medications to manage symptoms such as pain. The median time between the significant point, which triggered de-escalation, and patient death was one day and was preceded by withdrawal of ventilation for 19 (90%) patients. All patients died in an intensive care setting on days two-315. Two patients were referred to specialist palliative care team.

Trajectory 4 Early treatment limits set (n = 9)

Trajectory 4 was characterised by patients with postnatally and prenatally diagnosed LLC (cardiac, genetic, and metabolic conditions), with early treatment limits set (See Fig. 4 Care management trajectory type 4). Three patients were admitted to intensive care on day one, three patients remained on the maternity ward before discharge home, two were admitted to a paediatric ward on day one, and one patient was admitted to hospital at age 14 weeks.

The triggers for early treatment limits were confirmed diagnosis of LLC, lack of curative treatment options, the prospect of severe disability, and anticipated poor quality of life. The median time for implementing early treatment limits was one day from recognition of prognosis. All patients had treatment limits implemented, some of which included not for intubation and ventilation, not for CPR, not for invasive or unnecessary investigations and not for admission to intensive care. The median time from implementing treatment limits and death was 13 days, with 192 days for one patient.

Eight (88%) patients were referred to specialist palliative care teams. Of these, five patients had a confirmed antenatal diagnosis and were referred antenatally to the specialist palliative care team for ACP and

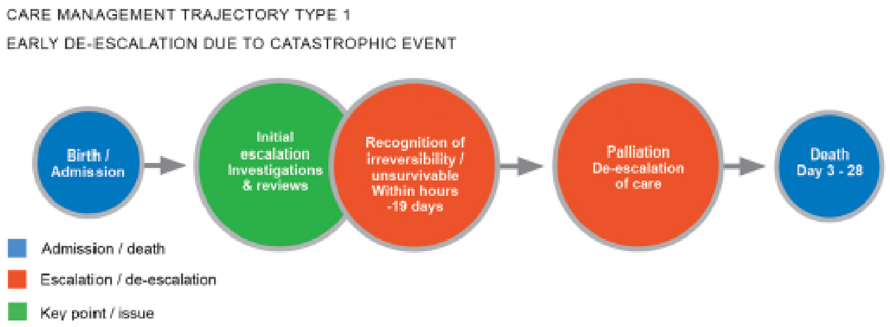

Fig. 1. Care management trajectory type 1.

end-of-life care planning before birth. Plans for once the infant were born included bag and mask ventilatory support, not for admission to intensive care, to offer comfort feeds, patient to remain with mother, medications for comfort, and cuddles with parents. Four patients were diagnosed postnatally and received short periods of acute care and investigations before a confirmed diagnosis. Of the four patients diagnosed antenatally, three were referred to the specialist palliative care team. Reasons for specialist palliative care referral included palliative care assessment, symptom management, ACP, end-of-life care planning, social support, community care planning and bereavement support.

Seven of nine patients had ACP documents that had been used by specialist palliative care teams to document the clinical management for deterioration, family preferences and priorities for end-of-life care, and their cultural, spiritual, and religious wishes. The main primary goals of care were to prioritise comfort and quality of life, management of symptoms, and care to be provided in ward areas or at home. Despite the initiation of treatment limits, patients continued to receive supportive care, which included physiotherapy and or occupational therapy interventions, enteral feeding, seizure medications, antibiotics, and oxygen therapy. Overall, this group received fewer invasive interventions and treatments. A key feature was that five (55%) patients were discharged home for end-of-life care or transferred to hospitals close to home, with the support of specialist palliative care teams and community services.

Discussion

This study illustrated the four care management trajectories for 45 infants with LLC who died. Congenital and perinatal conditions (preterm births, birth trauma) were the most prevalent conditions resulting in infant deaths, reflecting national and international reports of the leading causes of infant deaths (Australian Insitute of Health and Welfare, 2019; National Centre for Health Statistics, 2019; Office for National Statistics, 2019). The key findings were the overriding emphasis on curative intent rather than planning for palliative care in parallel to treatment, identification of triggers and processes for ACP, missed opportunities for integration of palliative care, infrequent involvement of specialist palliative care teams and little documented bereavement support.

The unclear course of diseases, sudden deterioration, significant health comorbidities, and challenges in clinical management featured across all the trajectories. This complex care picture for infants with LLC has been reported by others (Kukora et al., 2017; Marc-Aurele & English, 2017; Mitchell et al., 2019; Moro et al., 2006). Adding to the complex care picture, we also found that different individuals led the infants' care each week in intensive care environments. Mitchell et al. (2019) advocated the importance of continuity of care, and trusted relationships between families and health professionals to support families through decision making.

Despite the ever-present potential for sudden clinical deterioration, many infant deaths in trajectories two and three were not anticipated or planned for until the infants were close to end-of-life. In addition, family preferences for end-of-life care were not consistently documented for the infants in trajectories two and three. The majority of deaths occurred in intensive care environments preceded by a decision for withdrawal of ventilation. This reflects previous studies where high incidences of infants and children who died in intensive care had pre-existing LLC and deaths commonly occurred after withdrawal of ventilation (Bolognani et al., 2020; Fraser et al., 2018; Mitchell et al., 2014).

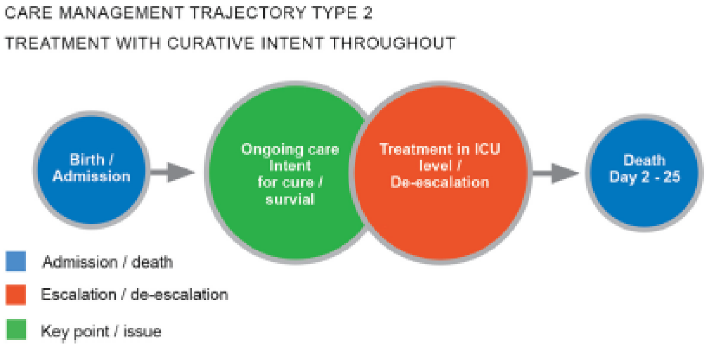

Fig. 2. Care management trajectory type 2.

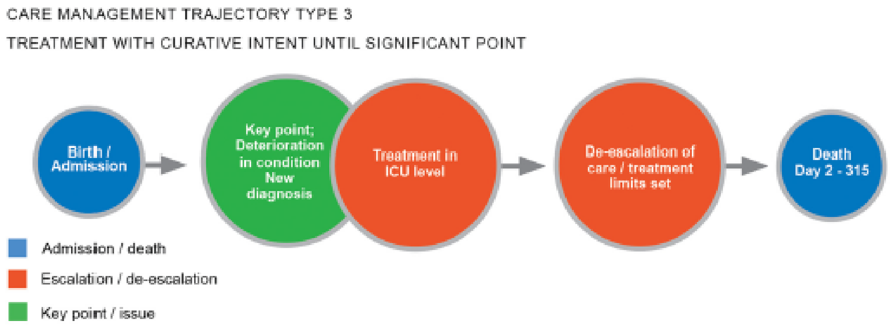

Fig. 3. Care management trajectory type 3.

Furthermore, we noted infants who died in intensive care environments experienced invasive interventions before end-of-life, such as CPR or intubation, in contrast to those who died in wards or at home, this was also supported by other studies (Bolognani et al., 2020; Fontana et al., 2013; Gibelli et al., 2021).

Given the rapidly changing conditions of the infants, commencing ACP discussions in parallel to treatment may be beneficial, and shift the focus towards maximising comfort and quality of life. Stark et al. (2008) reported ACP commonly occurs in the last stages of illness and often when the patient is close to death. Previous research exploring families experiences supports the benefits of ACP in parallel to treatment for families providing opportunity to plan ahead of time, and management of care that prioritises their preferences (Kamrath et al., 2019; Mitchell et al., 2019). Triggers for ACP included confirmed diagnosis of LLC, lack of curative treatment options available, and recognition of irreversibility or deterioration in condition despite maximal treatment. Although these triggers were evident in the small group of infants in trajectory two, de-escalation and ACP were not documented, and the infants died during resuscitation following a sudden deterioration. For trajectories one, three and four, these triggers prompted discussions with families for de-escalation or early treatment limits. Whilst de-escalation occurred in trajectories one and three, these triggers may have highlighted possible missed opportunities for earlier commencement of ACP.

Advance care planning and end-of-life care planning that incorporate the family goals of care, spiritual, religious or cultural values has been recommended (Gilmour et al., 2017; Horridge, 2015), yet these were not consistently documented nor followed a standardised process. In contrast, ACP documents utilised by specialist palliative care teams support health professionals to facilitate discussions, which included family preferences for end-of-life, family goals of care and preferred place of death.

Low referrals to specialist palliative care teams for infants with LLC in neonatal and paediatric intensive care units were consistent with others' reports, in particular for preterm infants (Bolognani et al., 2020; Gilmour et al., 2017; Harmoney et al., 2019; Ramelet et al., 2020). Difficulty predicting prognosis, short periods in hospital and curative focus of care were attributed to reasons for low referrals, as reported by Stutz et al. (2018). In contrast, infants in trajectory four with confirmed LLC and known disease trajectories were referred to specialist palliative care teams, with these infants more likely to be managed at home or in paediatric hospital wards and receive less invasive treatment before end-of-life. These findings are similar to those reported by Bolognani et al. (2020) and were attributed to increased awareness of palliative care and earlier referrals. Specialist palliative care teams worked in partnership with the lead clinical team. They supported the commencement of ACP, parallel planning, accommodation of parents' end-of-life preferences, preferred place of care, and support for managing the dying infant at home. These practices reflect palliative care standards and early integration pathways for infants and children with LLC which include family centred care, ACP, family goals of care and bereavement support (National Institute for Health and Care Excellence, 2016; Together for Short Lives, 2017; Western Australian Department of Health, 2021). Several descriptive studies exploring experiences of parents of infants with LLC who died supported the importance of early integration of palliative care, and involvement of families in decision making and end-of-life care planning (Kamrath et al., 2019; Mitchell et al., 2019; Tan et al., 2012).

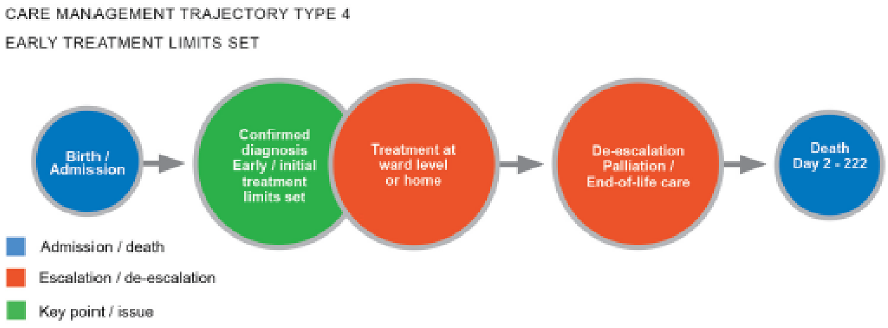

Fig. 4. Care management trajectory type 4.

Bereavement support and coordination for the families of infants after their death was not consistently reported. We found that many families appeared to not to have received follow up bereavement support from the hospital after the first week following the death of their child, and several had no documented follow up. Bereavement support is recommended to comprise support for the whole family, that includes a plan identifying a key person or team who will provide follow up, an opportunity to meet with the healthcare team to address matters related to their infant's care and death, and identifying community support services (Currie et al., 2016; Kenner et al., 2015; Western Australian Department of Health, 2021). Tan et al. (2012) explored the experience of parents whose infants died in acute care settings, highlighting the importance of anticipatory bereavement care to support parents with the death of their child.

From the findings of this study and what is reported in the literature we advocate for health professionals to provide care to infants with LLC that encompasses palliative care principles and practices in parallel to treatment that is aimed at cure or prolonging life. Specialist palliative care teams could be considered for specific cases or to support the family if they wish to be at home.

Practice implications

There is potential to further enhance health professionals' clinical practice in delivering care to infants with LLC that encompasses palliative care in parallel to treatment, and to optimise the support and experience for families. This can be achieved in several ways. At the individual staff member level, the provision for ongoing education can help develop effective communication skills and build confidence in adopting palliative care principles and practices in partnership with specialist palliative care teams. At the organisational level, the implementation of a standardised framework for documenting and facilitating ACP conversations can promote parallel planning and an understanding of the families' goals of care. Bereavement support for families should include the provision of a coordinated and a standardised approach for families following the death of an infant. At a systemic level, a focus on policies and guidelines can promote adoption of palliative care principles and practices including bereavement support.

Limitations

Limitations included the retrospective nature of this study and reliance on documentation by health professionals, including the involvement of families in decision making and end-of-life care planning. Gaps in documented bereavement support were identified; it is possible support and interventions were not documented or they may have been recorded elsewhere (e.g., in the mother's medical record or allied health records). Infants included in this study were a heterogenous group, which may limit the generalisation of the findings to other settings. A further limitation, given there were 10 infants who died in paediatric intensive care, is there was no paediatric intensive care health professional or health consumer perspective on the stakeholder group. We also did not examine differences in care provided between neonatal or paediatric intensive care health professionals. This study did not include the perspective of families which would contribute significantly to further understanding the journeys of infants and experience of their families. Finally, this study did not review the care management of infants with LLC who did not die.

Conclusion

This review highlighted the complex management of infants with LLC, who died in the first year of life. The infants comprised of a heterogenous group, characterised into one of four care management trajectories. Dual competing goals of care for infants with LLC who are susceptible to sudden deterioration were identified. Recognising triggers for de-escalation are integral to providing best practice palliative care. For infants who receive care in intensive care environments, there is potential to optimise the experience of the family through early integration of palliative care principles and practices in parallel to receiving curative or life-prolonging treatment. In addition, ACP can facilitate the goals of the family and formal coordination of bereavement support. There is opportunity to enhance the care and experience of families by adopting a structured approach to providing best practice palliative care through consultation, support, education, policies, and guidelines.

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CRediT authorship contribution statement

Rebecca Iten: Conceptualization, Methodology, Data curation, Investigation, Formal analysis, Writing – original draft, Project administration, Writing – review & editing. **Moira O'connor:** Conceptualization, Methodology, Supervision, Formal analysis, Writing – review & editing. **Lisa Cuddeford:** Writing – review & editing. **Fenella J. Gill:** Conceptualization, Methodology, Formal analysis, Supervision, Writing – review & editing.

Declaration of Competing Interest

The authors have nothing to declare.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.pedn.2022.11.014.

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CHAPTER 3

3.1 Study 2 Integrative Review

Chapter 3 presents the publication *“Palliative care for infants with life-limiting conditions: integrative review”* that syntheses the literature for how palliative care is delivered to infants.

3.2 Publication 2

This article has been accepted for publication in BMJ Supportive and Palliative Care, 2023 following peer review, and the Version of Record can be accessed online at https://doi.org/10.1136/spcare-2023-004435

Iten, R., O'Connor, M., Gill, F. J. (2023) Palliative care for infants with life-limiting conditions: integrative review. *BMJ Supportive & Palliative Care.* Published Online First: 09 December 2023.

Palliative care for infants with life-limiting conditions: integrative review

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ABSTRACT

Background Infants with life-limiting conditions are a heterogeneous population. Palliative care for infants is delivered in a diverse range of healthcare settings and by interdisciplinary primary healthcare teams, which may not involve specialist palliative care service consultation.
Objective To synthesise the literature for how palliative care is delivered for infants aged less than 12 months with life-limiting conditions.
Methods An integrative review design. MEDLINE, CINAHL, ProQuest, Cochrane, Joanna Briggs Institute and EMBASE were searched for research published in English language, from 2010 to 2022, and peer reviewed. Critical appraisal was completed for 26 patient case series, 9 qualitative, 5 cross-sectional and 1 quality improvement study. Data analysis involved deductive content analysis and narrative approach to summarise the synthesised results.
Results 37 articles met the eligibility for inclusion. Two models of palliative care delivery were examined, demonstrating differences in care received and experiences of families and health professionals. Health professionals reported lack of palliative care education, challenges for delivering palliative care in intensive care settings and barriers to advance care planning including prognostic uncertainty and transitioning to end-of-life care. Families reported positive experiences with specialist palliative care services and challenges engaging in advance care planning discussions.
Conclusion There are complex issues surrounding the provision of palliative care for infants. Optimal palliative care should encompass a collaborative and coordinated approach between the primary healthcare teams and specialist palliative care services and prioritisation of palliative care education for nurses and physicians involved in providing palliative care to infants.

WHAT WAS ALREADY KNOWN

⇒ Palliative care for children provides a total approach to care and is recommended to begin at diagnosis of a life-threatening or life-limiting condition.
⇒ The palliative care approach aims to improve quality of life for children with life-limiting conditions and their families.

WHAT ARE THE NEW FINDINGS

⇒ Integration of specialist palliative care services was not a standard practice for infants, this was particularly evident in neonatal intensive care units.Although not all infants with LLCs require input from a specialist palliative care service, a palliative approach to care should be delivered by all health professionals.

WHAT IS THEIR SIGNIFICANCE

⇒ Clinical: Prioritisation of palliative care education for nurses and physicians involved in the care of infants at an individual and organisational level may lead to improved knowledge and confidence to provide palliative care across a range of clinical settings and further integrate specialist services to optimise palliative care delivery to infants and their families.
⇒ Research: Further research to evaluate the impact of palliative care education for nurses and physicians.

BACKGROUND

Palliative care for infants and children provides a total approach to care and is recommended to begin at diagnosis of life-threatening or life-limiting conditions (LLCs).[1–3] It promotes quality of life and can be provided in parallel to curative treatment.[1–3] This approach supports patient-centred and family-centred care to provide treatment that meets the family's preferences and needs.[2 3] A recent rapid review of the literature highlighted key areas to enhance the delivery of palliative care for children were: patient-centred and family-centred care, early integration of palliative care to improve access for families and across a variety of settings, advance care planning with support from specialist palliative care teams, and development of educational resources and skills training for health professionals.[4]

Perinatal and paediatric palliative care services have become established specialties over the past two decades, comprising interdisciplinary health professionals including physicians, nurses, midwives, social workers and chaplains who have advanced training and/or experience in palliative care.[3 5 6] Key features of specialist palliative care services include providing consultation to patients and their families and working in partnership with the patient's healthcare teams to provide management of symptoms, support with decision making, advance care planning, as well as psychosocial, spiritual and bereavement support for the whole family.[1–3] Recent research exploring families' experience of palliative care described what constitutes 'good' palliative care as receiving coordinated care, where palliative care is integrated early with specialist palliative care service professionals, involves families throughout the care of the child and at end-of-life, is individual to the family needs and supports the whole family.[7]

Infants aged less than 12 months comprise the highest number of deaths in childhood.[8 9] The leading causes of infant deaths are congenital malformations and perinatal conditions related to low birth weight, preterm births, birth trauma and viral infections.[9–11] Many infants' deaths occur in intensive care settings.[12–16] Delivery of palliative care for infants has been reported to be impacted by specific barriers such as uncertain prognosis, use of invasive life-saving interventions including mechanical ventilation and cardiopulmonary resuscitation at end-of-life, complexity of symptom management and lack of continuity of care due to rotational staffing in intensive care units.[17–19] A recent study examining the care management trajectories of infants with LLCs who had died found that even for infants born preterm or with congenital conditions, deaths were not anticipated or planned for until close to end-of-life.[16] Additional challenges that have confronted families included prolonged hospital stays, prognostic uncertainty while at the same time facing the risk of sudden deterioration and possibility of death of their infant.[16 20]

Infants with LLCs may be cared for in a variety of hospital and community settings, including intensive care, lower acuity wards and community settings such as hospice and home.[3 16 21] Availability of specialist palliative care services may vary across these settings. This means that palliative care for infants is often delivered by the primary or treating healthcare teams such as intensive care physicians and nurses. An Australian study found less than a quarter of infants (11/45) with LLCs who died received input from a specialist palliative care service.[16] Given the evolving role of specialist palliative care services, the heterogeneous population of infants with LLCs and their unique needs in a diverse range of healthcare settings, it is unknown if palliative care is being optimally provided for these infants and their families. The aim of this integrative review was to synthesise the literature to better understand how palliative care is delivered for infants aged less than 12 months with LCC. The objectives were: (1) to explore how palliative care is delivered for infants with LLCs; (2) to understand the role of specialist palliative care services for infants with LLCs; (3) to identify factors impacting palliative care delivery to infants.

METHODS

The integrative review method was selected to provide a broad understanding of the diverse palliative care literature. The review was guided by the approach used by Whittemore and Knafl[22] and followed five steps: problem identification, literature search, data evaluation, data analysis and presentation of results. A protocol was developed and published before the review was started in the Open Science Framework.[23] The PRISMA Statement reporting checklist for Systematic Reviews was followed.[24]

Search strategy

An electronic search of six databases was conducted from a broad variety of health disciplines: MEDLINE, CINAHL, ProQuest, Cochrane, Joanna Briggs Institute and EMBASE. The search strategy included a combination of key words in the title or abstract relating to (palliative or terminal or 'end of life*') ADJ3 (care or therap* or treatment*) adjacent to (care or therap* or treatment*) and ("life-limiting*" or chronic* or complex*) adjacent to (condition* or disease* or illness*) and ("congenital abnormalit*" or "congenital anomal*" or prematur*) and (infant* or baby or babies or pediatric* or paediatric* or newborn* or neonat*). Search terms were adapted and modified for each database. See box 1 for Medline search strategy. Hand searching of reference lists of retrieved papers was also undertaken.

Box 1 Medline search strategy

Medline search strategy
- ⇒ ('congenital abnormalit*' or 'congenital anomal*' or prematur*).ti,ab. OR
- ⇒ Chronic Disease/OR
- ⇒ (('life-limiting*' or chronic* or complex*) ADJ3 (condition* or disease* or illness*)).ti,ab.

AND
- ⇒ ((palliative or terminal or 'end of life*') ADJ3 (care or therap* or treatment*)).ti,ab. OR
- ⇒ Terminal Care/or Palliative Care/

AND
- ⇒ (infant* or baby or babies or pediatricpaediatric* or paediatric* or newborn* or neonat*).ti,ab. OR
- ⇒ Exp Infant/

Limit to English language and year 2010 to 31 July 2022.

Study selection

Included articles comprised diverse methodologies, theoretical and empirical research about how palliative care is delivered for infants. Specifically, articles focused on palliative care for perinatal, neonatal (the term neonate is defined from birth until 28 days of life)[25] and paediatric populations. Studies were included if there was a substantial proportion that included infants. Studies focused on perinatal care were included only if they also included the neonatal period. The starting year 2010 was chosen for inclusion of articles to capture the period of specialist paediatric palliative care emerging as a medical specialty. Inclusions: English language, published online from 31 July 2010 to 31 July 2022, full text available online and peer reviewed. Exclusions: study results obtained more than 2 years prior to 2010, and where results did not specifically relate to infants.

An electronic research screening programme was used to screen publications by titles and abstracts.[26] A selection of 11 key publications were uploaded to research screener to improve eligibility screening.[26] One researcher (RI) independently screened each article in the research screener programme. All three authors (RI, FG, MOC) screened and cross-checked one-third of the articles. Articles that met the inclusion criteria were read in full by one author (RI). Eligible articles were discussed by all three authors to cross-check and address discrepancies.

Data extraction

Articles were grouped according to the research focus: patient case series and perspectives of either health professionals or families. Data extracted from each article were summarised and tabulated; citation (author), year, country, design and method, sample characteristics, specialist palliative care service established, single or multi-site study, aim of study and key findings.

Data evaluation

Critical appraisal of the articles was completed using one of the four checklists according to study designs: patient case series, qualitative, cross-sectional and quality improvement. For quality purposes, a sample of 25% articles from each checklist was completed together by the authors (RI, FG, MOC). One author (RI) independently completed the remaining appraisals and were cross-checked with FG and MOC. Case series and qualitative articles were appraised using checklists from Joanna Briggs Institute Quality Appraisal Framework,[27,28] both checklists consisted of 10 items. Cross-sectional articles were appraised using a 20-item checklist.[29] The quality improvement study was appraised using a 16-item checklist.[30] The quality of the articles was categorised using an approach by Gill *et al*[31] where the total score from the checklist were converted to a percentage. The following percentages were indicative as: strong (80–100%), good (70–80%), average (50–70%) or poor (less than 50%).

Data analysis

A deductive content analysis[5,32] approach was applied using pre-established themes from key palliative care international guidelines.[5,6] These included population characteristics, specialist palliative care service, advance care planning, characteristics of end-of-life care and bereavement support. Content analysis steps involved categorising, quantifying, contrasting and comparing the data to synthesise the themes and evidence.[22] A narrative approach was used to summarise the synthesised results.

RESULTS

The search generated 3156 articles. Articles were exported to a reference management software programme and, guided by key articles, a research screener system screened and organised each article by title, abstract and relevance.[26] Research screener removed 1486 articles that were duplicates or missing titles and abstracts. A total of 1674 articles were screened at title and abstract level. This resulted in 116 articles that met eligibility criteria and were independently read in full by one author (RI), followed by discussion among the research team to cross-check and address discrepancies. Figure 1 displays the PRISMA flowchart. The final number of articles included prior to critical appraisal was 41, including four articles identified through hand searching.

Critical appraisal

Twenty-six articles reported a patient case series method (retrospective review), of these, 23 articles were rated as strong, 3 rated as average and were excluded. For nine qualitative research studies (interviews and focus groups), five articles were rated as strong, three were good and one rated as poor and was excluded. For five cross-sectional designs (surveys), three rated strong, two were rated as good. One quality improvement study was rated as strong. In total, four articles were excluded due to poor quality and/or a score of less than 50%. See online supplemental file 1 for critical appraisal of articles. Following critical appraisal, the final number included in the review was 37. See online supplemental file 2 summary of articles.

Characteristics of articles

Studies were conducted from the USA 13 (35%), UK 5 (13%), Australia 3 (8%), Brazil 3 (8%), Switzerland 3 (8%), Canada 2 (5%), with the remaining 8 (22%) articles from Belgium, Iran, New Zealand, Malaysia, Italy, China and Turkey. Case series studies included populations of newborns to age 18 years; 13 (35%) articles focused on infants aged less than 12 months and children older than 12 months and 11 (30%) focused on newborns less than 30 days old and infants

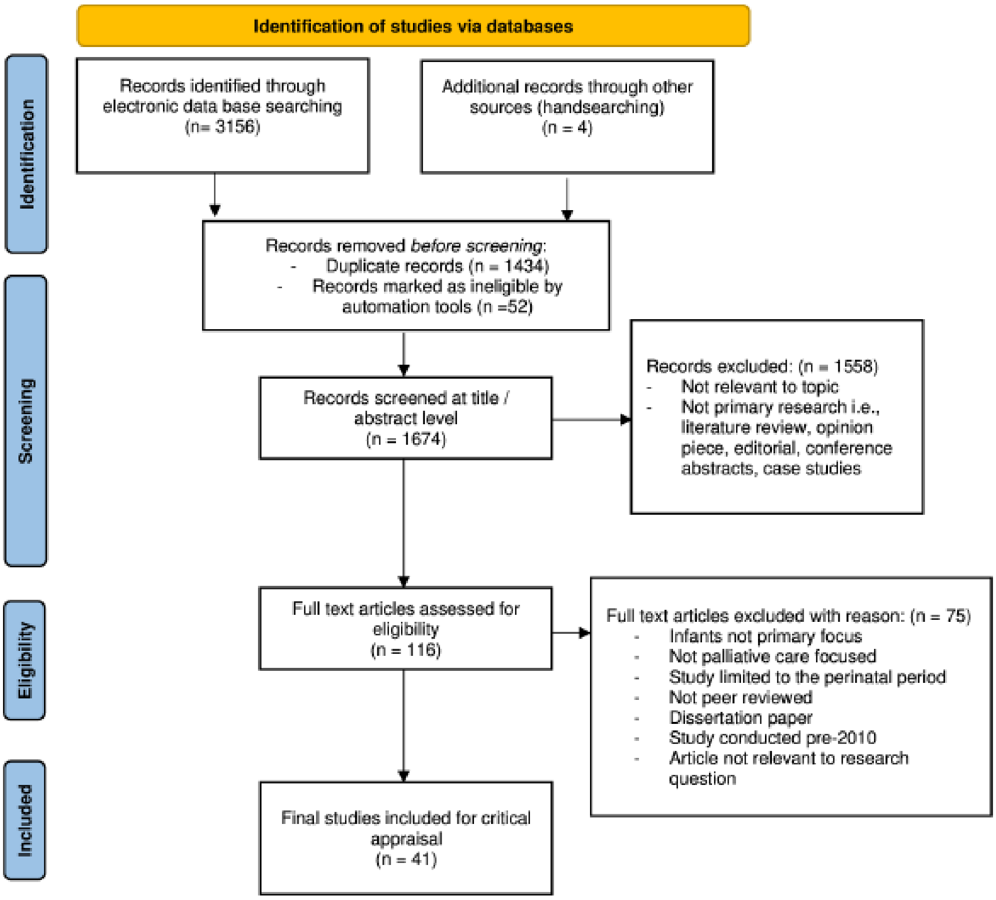

Figure 1 PRISMA flowchart.

aged less than 12 months. Five (13%) articles examined the experience of bereaved parents of infants, and eight (22%) focused on health professionals' experience involving nurses and physicians from areas such as paediatric or neonatal and intensive care units. Most studies 23 (62%) were conducted at a single hospital site and 14 (38%) were multi-site studies. Articles reported on key elements, including the team who delivered palliative care, characteristics of infants who died, advance care planning, barriers and facilitators to provision of palliative care and experience of families. See table 1 for article characteristics.

The availability of specialist paediatric and perinatal palliative care services was reported in 25 (68%) articles. For 10 (27%) articles, it was not reported and two (5%) articles reported that specialist palliative services were not established or limited. In articles where specialist palliative care services were reported, for 16 (43%) it was not routine practice for infants with LLCs and their families to receive specialist palliative care service consultation.[14 15 33–35] Three (8%) articles reported that palliative care was delivered by the primary healthcare team in the neonatal intensive care unit (NICU).[13 33 40] Congenital abnormalities and complications secondary to extreme prematurity were the most reported cause of deaths for infants.[13 15 35 36 38 41–46]

Synthesis of results

This review explored how palliative care was delivered for infants with LCCs and their families, and the role of specialist palliative care services. The 24 case series articles were grouped into two categories of (1) palliative care delivery with specialist palliative care services and (2) palliative care delivery by the primary healthcare team without specialist palliative care consultation. Key topics reported included population characteristics, place of care and death, advance care

Table 1 Article characteristics

Characteristics	n (%)
Study design	
Patient case series	23 (62)
Cross-sectional	5 (13)
Qualitative	8 (22)
Quality improvement	1 (3)
Study participants	
Patient case series (includes n=1 quality improvement article)	24 (65)
Perinatal and infants <12 months	11 (30)
Infants <12 months and children >12 months	13 (35)
Health professional	8 (21)
Neonatal nurses	4 (10)
Neonatal physicians	1 (3)
Mixed neonatal physicians and nurses	1 (3)
Paediatric intensive care physicians and senior nurses	1 (3)
Mixed neonatal/paediatric intensive care physicians	1 (3)
Parent/family (bereaved)	5 (13)
Topics articles reported	
Palliative care delivered by specialist palliative care service or healthcare teams	25 (68)
Characteristics of infants who died (end-of-life care, diagnosis, cause/mode of death, place of death)	28 (76)
Advance care planning	10 (27)
Bereavement support	5 (13)
Barriers and facilitators to palliative care	6 (16)
Health professional experience with palliative care	7 (19)
Parent / family experience with end-of-life care decision making, specialist palliative care service consultation	4 (10)

planning, end-of-life characteristics and bereavement support. The two groups were compared to examine key differences in how palliative care was delivered and how these impacted on care. To explore factors that impacted on palliative care delivery, synthesis of the cross-sectional and qualitative articles included family experience of palliative care and health professional perspective.

Palliative care delivery with specialist palliative care service consultation

The population who received specialist palliative care service consultation were infants and children with a high risk of mortality or medical complexity including receiving invasive life-saving interventions such as mechanical ventilation, older infants (aged more than 12 months),[34 37 43 47] with congenital conditions diagnosed prenatally or following birth trauma,[14 33 42 44 48] and infants with longer NICU stays (more than 30 days).[33] However, this was inconsistently reported. For example, one Canadian article reported for infants diagnosed with congenital heart disease, consultation with a specialist palliative care service was less common (16%), and an article from Ireland reported infants with congenital heart disease comprised 25% of specialist palliative care consultations.[34 48] Reported reasons for consultation with a specialist palliative care service were to guide goals of care/advance care planning discussions, support decision making, optimise symptom management, end-of-life care planning, and care coordination.[33 44 49]

Advance care planning discussions occur between the healthcare team and family and involve consideration of how to provide care to the infant during deterioration and at end-of-life that aligns with the families' preferences and needs.[5] Advance care planning discussions were reported in 10 (27%) articles and included antenatal birth planning, family's preferences for end-of-life care and treatment limitations.[15 37 41–43] Infants with advance care planning and treatment limitations were reported to have received fewer invasive procedures close to end-of-life.[15 35 37 41 50] Antenatal birth plans focused on comfort rather than invasive treatment and resuscitation measures.[42] In contrast, one article from the USA compared infants who received palliative care with those who did not and reported infants in both groups received similar invasive procedures close to end-of-life.[44]

End-of-life care was delivered in a variety of settings including the labour and delivery ward, neonatal and paediatric intensive care, hospice and home.[12 15 42 48 50] A distinguishing feature for families of infants who received specialist palliative care service consultation was that these families were offered options for place of care at end-of-life, resulting in deaths occurring outside of intensive care setting, including inpatient hospital wards, hospice or home.[12 15 21 37 41 43 48 50 51] Two articles captured bereavement support . In both studies, families who had received specialist palliative care service consultation received bereavement support.[15 50]

Palliative care delivery by primary healthcare team without specialist palliative care service consultation

The population who received palliative care delivered by their primary team appeared to be infants in NICUs,[15 35–37 39 43 45 46] in particular infants born preterm or with congenital heart disease.[14 34]

For infants in this group, advance care planning discussions were infrequently reported.[15 35 36 41 43 50] Infants received invasive interventions close to end-of-life, such as mechanical ventilation, cardiopulmonary resuscitation, insertion of central venous access devices, administration of vasoactive medications and antibiotics.[12 35 37 38 45 52] A Brazilian study at a tertiary NICU reported that infants with prognostic uncertainty received invasive treatments more frequently in comparison to infants with confirmed prognosis.[13] One article from the USA compared outcomes for infants between parents who opted for comfort care versus infants who received intensive care treatment and identified no difference in the median age of death.[40] Infant death following withdrawal of life-sustaining treatment was reported in six studies.[12 35 38 41 45 52] A nation-wide Swiss study including children of all ages, that formed part of a larger research programme to understand current end-of-life practices and parental perspectives, reported 84% of infant deaths were preceded by withdrawal of life-sustaining treatment.[38]

A high proportion of infant deaths occurred in intensive care settings.[36,41,45,46,51] Younger infants (aged under 12 months) appeared to be less likely to be transferred or discharged home for end-of-life care.[12,15,21,38,45] Discussions for options for preferred place of death was infrequently reported,[15] and hospice deaths were less common.[43] Bereavement support was not reported in any of the studies.

Family perspective

Five articles reported experiences of infants' bereaved parents using in-depth interviews[20,53,54] or cross-sectional surveys.[39,55] Each article focused on a different aspect of the family experience; consultation from a specialist palliative care service, advance care planning, end-of-life care decision making and parental satisfaction. Families reported that the specialist palliative care team provided support with decision making, facilitated communication for advance care planning, provided emotional support and optimised symptom management.[53,55] Of note, in one study families initially associated the term palliative care with the healthcare team giving up on providing curative treatment and, as a result, they experienced a loss of hope.

One parent stated:

> So when I heard palliative care, I heard hospice, I knew we were teetering on the edge of life. Instead of looking at the positives that it could do, I was just looking at, you know, I'm not ready to transition to that. We're still fighting, I still want aggressive care.[53] p.483

Interestingly, the family later reflected on their experience and stated they found consultation with the specialist palliative care service was beneficial, and they would recommend it for other families.[53]

Three articles reported families' experiences of advance care planning and end-of-life care decision making.[20,53,55] An article from a paediatric intensive care unit in the UK investigated parental experiences of decision making and reported continuity of care and trusted relationships were fundamental to supporting parents with end-of-life care decision making.[20] Advance care planning was reported by parents as being emotionally overwhelming, and they felt they needed to be accepting of the situation before they could engage in these discussions.[20] In addition, not all parents were familiar with advance care planning, some parents felt it would have been helpful and others reported they would not be able to engage in the process to plan for or consider their infant or child's end-of-life.[20] Preferences for end-of-life care (including cultural and spiritual needs) appeared to be unique to each family.[20] Parents described factors that influenced decision making for their infant or child's end-of-life care was their knowledge and experience of the condition, recognition of progressive deterioration, their acceptance of end-of-life, honest and clear communication from health professionals and parents 'goal' for more time with their child.[20,39,53] Another Swiss study, that formed a component of the research programme discussed earlier, involved a survey of 135 bereaved parents from four diagnostic groups (cardiology, neonatology, neurology and oncology) to examine their perceptions of their child's end-of-life care following six quality domains of care; support for the family unit, communication, shared decision making, symptom management, continuity and coordination of care and bereavement support.[39] Parental satisfaction about end-of-life care was reported positively by all diagnostic groups.[39] Interestingly, neonatology parents reported lower satisfaction with shared decision making than the other three diagnostic groups.[39] Furthermore, 80% of neonatology parents reported discussions regarding withdrawal of life-sustaining treatment occurred, however, 65% reported they were actively involved in decisions for withdrawing life-sustaining treatment.[39]

Specific challenges for parents in NICUs were reported to be: not being able to comfort and hold their infant, financial burden of having an infant in hospital, communication conflict with health professionals, lack of continuity of care due to changeover of staff and the open environment of the intensive care unit resulting in lack of privacy.[53] Families who accessed hospice care described the added value of being in a homely environment, availability of continuous support from health professionals, privacy and the opportunity to care for their infant or child and create memories.[54]

Factors that impacted on palliative care delivery

Eight articles reported perspectives of health professionals (physicians and nurses involved in the care of infants) using surveys,[56–58] in-depth interviews[47,59–61] and focus group.[62] Several facilitators and barriers were identified for the provision of palliative care, including referrals/involvement of specialist palliative care service and advance care planning.

Facilitators

A key facilitator for consulting with specialist palliative care service was physicians' and nurses' appreciation of palliative care teams' knowledge and experience navigating uncertainty, expertise in providing emotional and spiritual care for families and assisting with communication,[47] and health professionals' recognition that palliative care shares parallel importance as curative care.[58] Other facilitators to consult with specialist palliative care service were: provision of support for patients with medical complexity and those with lengthy hospital stays.[47] Nurses valued specialist palliative care consultation services' capacity to enable families to have options for preferred place of care and death, provide bereavement support, and care coordination with community teams including hospice care.[59] An important feature reported in six articles was health professionals' recognition that palliative care education and training, including understanding when to refer to specialist palliative care services, is essential for health professionals to deliver optimal palliative care.[47 57–61]

Barriers

A key challenge to offering specialist palliative care service consultation in the NICU appeared to be that a high proportion of infants died within 24 hours of admission.[37] This short time period reduced opportunity for specialist palliative care consultation.[37] Further, health professionals reported how intensive care environments were suboptimal settings for supporting infants at end-of-life and for delivering a palliative approach.[57 58 62] These factors were compounded by time constraints and frequent staff change overs that prohibited opportunity to develop rapport with families.[47 57 58 62] In addition, insufficient knowledge about palliative care and advance care planning due to a lack of formal training and education was reported by nurses and physicians in NICU and paediatric intensive care units.[59–61] Barriers to discussing advance care planning and transitioning to end-of-life care were reported to be due to: prognostic uncertainty, lack of medical consensus about a prognosis, availability of newer technologies and procedures to prolong life, parental desire for and expectations of cure and not 'giving up', and ethical challenges balancing the burdens and benefits of invasive treatments.[56 58 59 62] Neonatology physicians' decision to cease curative treatment was mostly influenced by prognosis—if it was predictable, and their assessment of infants' quality of life.[56]

DISCUSSION

This review identified 37 articles from 14 countries, predominately originating from developed countries. The articles reported a heterogeneous population of infant characteristics, study settings, aims and methods. There was clearly great variation in the care infants received and complex issues surrounding the provision of palliative care and factors impacting consultation with specialist palliative care services. The infant population in the case series articles included a range of disease trajectories, with complex medical needs, and a high proportion of infants was managed in intensive care units. Infants and their families also faced the ever-present possibility of sudden deterioration and death.

While the principles and practice of palliative care for children and adults have many similarities, infants and their families experience unique challenges and considerations.[63] Issues for infants include the rarity of diseases, difficulty to diagnose a condition, prognostic uncertainty, possibility of sudden deterioration, the role of decision makers which may involve extended family members and impact on the whole family.[63] A common theme reported by health professionals and in two of the patient case series studies was prognostic uncertainty, which contributed to the challenges of transitioning from cure-oriented care to palliative care and was a factor that influenced the care infants received. This mirrors a recent study in Australia categorising the trajectories of infants with LLCs who died, which found infants with prognostic uncertainty received curative treatment and were associated with infrequent referral to specialist palliative care services.[16] However, it is important to acknowledge the inherent challenges in the neonatal population where infant prognosis is unknown, and intensive care treatment needs to be initiated before a diagnosis and prognosis can be confirmed. Findings suggest that this is a phenomenon experienced across several continents.

In this review, two distinct models of palliative care were examined: (1) palliative care delivery for the infant and family who received specialist palliative care service consultation and (2) palliative care delivered by the primary healthcare team. Specialist palliative care services were reported as being available in the majority of the case series articles. However, less than a quarter of the articles reported specialist palliative care service involvement in infants' care. For the remaining articles, palliative care was delivered by the primary healthcare team, a feature particularly evident in NICUs. This reflects previous empirical findings and suggests that despite recommendations as best practice, integration of specialist palliative care services is not yet a standard practice for infants with LLCs.[4 19] This finding is not totally surprising given specialist palliative care services are considered relatively new in some countries, so understandably integration and service availability may vary. Specifically, studies conducted in Iran, Brazil, Malaysia and China did not report whether specialist palliative care services were available, perhaps indicating specialist palliative care consultation may not be a uniform approach in all countries and it is unknown the impact cultural beliefs may have on palliative care services.

Neonatal physicians and nurses have a comprehensive role in providing cure-oriented care and expertise in quality end-of-life care to infants. These healthcare teams may not consider or prioritise referral to specialist palliative care services for infants where the care management is focused on curative intent, or in the context of infants' rapid deterioration, and short time periods of care. Interestingly, this was not a factor reported by health professionals and may require further research.

The review highlighted key differences in practice between the two models of palliative care. Infants who received specialist palliative care service consultation were more likely to have had advance care planning discussions and end-of-life planning, including preference for place of death. Without these elements of care, families may not be afforded the opportunity to plan and prepare for their infant's end-of-life care that aligns with their goals and values.[59] Advance care planning discussions were recognised by parents as emotionally overwhelming, however were also acknowledged as important and valuable. Parents' willingness to engage in advance care planning discussions were also variable. This may be influenced by the approach taken by health professionals to frame advance care planning. This involves explaining to parents that advance care planning guides health professionals to deliver care that aligns with the family wishes and can also be achieved alongside infants receiving curative treatment. Using this approach, parents may better appreciate the importance of advance care planning and be able to engage in discussions.

It has long been recognised that parents with a critically ill infant requiring intensive care support experience depression, anxiety and loss of control and hope.[64] At the same time, parents are required to discuss the future care of their infant, a discussion which may occur when the infant's condition is rapidly evolving or has an uncertain prognosis. For some, therapeutic relationships may not yet be established between the family and healthcare team recognised as important factors enabling parents to engage in advance care planning discussions.[20] The challenges experienced by families with advance care planning for infants have also been described in paediatric and adult palliative care settings. For example, studies involving parents of children aged up to 17 years and in the adult patient setting similarly reported family reluctance and difficulty facing reality to have end-of-life discussions.[7 65 66] The review findings support timely referrals to specialist palliative care teams to establish therapeutic relationships, provide continuity of care, assist with decision making and provide psychosocial and emotional support for the whole family.[7 67] Involvement of specialist palliative care teams may play a key role in mitigating some of the challenges experienced by families to support families to engage in advance care planning.

Current guidelines recommend that preferred place of care or death is discussed or considered with the family and can be achieved with support from specialist palliative care services to assist in the transfer of patients for end-of-life care where feasible.[3 5 63] A key finding from this review was that a high proportion of infant deaths occurred in neonatal intensive care settings following withdrawal of life-sustaining treatment . It was not reported what the preferred place of death was for these families. Only a small number of deaths occurred at home or in hospice settings. Those infants who died at home or in hospice care were those who had received specialist palliative care service. While providing choices to families where they wish to be cared for is an important aspect of palliative care, transferring medically complex patients from intensive care units to other settings for end-of-life care may not be practical or feasible.[48] Significant preparation and collaboration of hospital and community providers are required, which can be resource intensive and logistically difficult to arrange.[15 68] Despite these challenges, with timely planning and collaboration, care of the infant at home may provide many benefits, including a homely environment, increased privacy, involving family members such as siblings in care and enhanced ability to meet cultural or spiritual needs.[3] Although a systematic review found inadequate evidence to support home as the preferred place of death for children and young people,[69] the majority of the studies reported perspectives of parents of children with cancer and the results may not be generalisable to other populations, in particular for infants.[69]

Challenges to providing optimal palliative care for infants and their families included primary healthcare professionals' reported lack of knowledge and experience with advance care planning and introducing palliative care resulting in lack of confidence, skills and knowledge to talk with families about palliative care and support decision making. These findings add to existing evidence in both paediatric and adult settings where the importance is recognised of building capacity of health professionals to deliver quality palliative care through education.[70 71] Given the diverse healthcare settings and health professionals involved in providing palliative care to infants, education that addresses specific challenges and complex issues for infants is essential. Targeted paediatric palliative care education through workshops, mentoring and delivering patient-specific education have been found to be useful in developing confidence and improving clinical practice for health professionals.[70 72] Key focus areas for education are recommended to align with current palliative care guidelines, including communication skills, assessment and management of symptoms, and psychosocial care of the patient and family.[3 71 73]

Finally, bereavement contact support was only reported in two of the case series articles, perhaps indicating a potential gap in the provision of palliative care services and bereavement care. Gaps in bereavement support have been widely recognised across adult and paediatric settings, with many services lacking bereavement guidelines.[74] A coordinated approach to bereavement support is recommended, including assessment of individual needs of the family, and may involve providing anticipatory support.[2] Bereavement interventions may include telephone contact, information and resources to navigate grief, referral to counselling services, age-appropriate support for siblings and opportunity to meet with the healthcare team to address questions.[2 74 75]

Strengths and limitations

Strengths of this review were the inclusion of a diversity of palliative care studies from a range of countries, study settings, aims and methods. A rigorous critical appraisal assessing the quality of each of the four study designs was conducted. The qualitative and cross-sectional study designs provided a layer of understanding and insight through exploring family and health professional experiences. Limitations were that only articles published in English and full text available were included. Consequently, there is potential for cultural bias, resulting in stronger perspectives from developed countries and limiting the generalisability of the findings. There is opportunity for researchers to advocate attention to capturing diversity. Additionally, bereavement support was not specifically a search term and was only reported in two of the case series articles. This may have also reflected gaps in documentation in patient records rather than gaps in service.

CONCLUSION

The review highlighted the complexity surrounding provision of palliative care for infants and their families. The infants comprised of a heterogeneous population, with a range of disease trajectories, many receiving intensive care level support. Complexity was compounded by prognostic uncertainty, possibility of sudden deterioration and death. Two models of palliative care delivery were examined, demonstrating differences in both care received and experiences of families and health professionals. For infants who received specialist palliative care service consultation, their families more frequently had advance care planning discussions and end-of-life care planning including preferred place of death. Health professionals reported challenges in transitioning from cure-oriented care to palliative care approaches for infants and their families. Lack of education and training about palliative care was identified by health professionals as a barrier that may impact on their capacity to deliver palliative care and to consult with specialist palliative care services. Optimal palliative care should encompass a collaborative and coordinated approach between the primary healthcare teams and specialist palliative care services. Prioritisation of palliative care education for nurses and physicians involved in the care of infants at an individual and organisation level may lead to improved knowledge and confidence to provide palliative care across a diverse range of clinical settings and further integrate specialist services to optimise palliative care delivery to infants and their families.

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CHAPTER 4

4.1 Discussion and Reflection

The overall study aim was to better understand how palliative care is delivered to infants with LLCs, and examine specialist palliative care teams' involvement. For Study 1 we examined the clinical and palliative care of infants with LLCs who died before 12 months of age at three tertiary hospitals in WA. In Study 2 we systematically examined the literature to understand how palliative care has been delivered to infants over the past decade. This chapter synthesises the research findings. Finally, we discuss implications for practice, scope for future research and make recommendations for optimising the delivery of palliative care to infants.

It was evident in both studies, reflected in local and international contexts, that infants frequently died without the involvement of specialist palliative care service consultation. The majority of infant deaths occur in intensive care settings. The infrequency of advance care planning, and challenges experienced by health professionals transitioning their approach from cure orientated to palliative care are also mirrored in both contexts. These findings are important as they demonstrate there is an opportunity to optimise the delivery of palliative care and for primary healthcare teams and specialist palliative care teams to work in partnership and increase collaboration.

Integration of specialist palliative care services in paediatric health settings has been slow to be adopted, especially in neonatal settings. This is despite the establishment of specialist palliative care services in the majority of jurisdictions, notably in developed countries. Not surprisingly the existence of palliative care services in low-and-middle-income countries remains limited (Fraser, Bluebond-Langner et al., 2020; Sisk et al., 2020; World Health Organization, 2018), and yet the integration of specialist palliative care for infants with LLCs in developed countries is clearly not yet standard practice. There are several possible reasons why integration of palliative care services has yet to occur. First, for low-and-middle-income countries, palliative care may not be a priority for some countries and health systems, including limited access to trained and experienced palliative care clinicians, unavailability of medications to adequately manage symptoms, and the impact of socio-cultural beliefs towards palliative care legislations, health professionals providing care and individuals or family members who require palliative care services (Poudel et al., 2019; World Health Organization, 2023). Second, in looking at the history of palliative care in the paediatric context before the establishment of specialist palliative care services, palliative care and end-of-life care was part of the routine care provided by the primary healthcare team. It seems this traditional practice has continued, with variable involvement of specialist palliative care services across jurisdictions and health settings. In the neonatal context, neonatologists and other intensive care doctors might consider the involvement of specialist palliative care unnecessary given their history of providing palliative care and end-of-life care. Similarly, in the oncology setting a 2020 systematic review examining the factors impacting children with cancer accessing specialist palliative care identified they were less likely to receive input from specialist palliative care teams. This was attributed to the uncertainty of when to refer to palliative care and oncology teams' perception that they were already delivering palliative care to children (Taylor et al., 2020). It is clear the reluctance of health professionals' attitudes and practices towards referring children to palliative care have not changed markedly over the past 20 years. Another finding from Study 2 was that doctors and nurses reported they had not received palliative care education, indicating that another important possible reason is a lack of knowledge about palliative care.

A vital facilitator for delivering safe and quality palliative care is education, and this is strongly recognised and instrumental in changing the attitudes, beliefs and practice of health professionals (Saad et al., 2022). Furthermore, there is an increasing priority for health professional education to support the generalist palliative care model (Gardiner et al., 2012; Robinson et al., 2022). In Australia, a nationally funded education project aimed at increasing health professionals' skills, knowledge and confidence in delivering palliative care for children across all areas of Australia has been established since 2014 (Donovan et al., 2019). Using various education initiatives including mentorship, patient specific education, and resource development. Outcomes from the project reported health professionals found the education to be highly valued, with increased awareness and collaboration of the local and state palliative care services, improved management of care for children and communication skills (Slater et al., 2021). A recent quasi-experimental study, in the Middle East, reported that paediatric nurses who received palliative care education had significantly improved palliative care knowledge and attitudes (Abuhammad & Almasri, 2022). Similarly, a 2014 study in the USA reported health professionals who received palliative care education were more likely to involve palliative care teams earlier in a patient's trajectory (Atwood et al., 2014; Saad et al., 2022). Interestingly, a 2022 review reported a common barrier to health professionals referral of children palliative care teams was concern about the parents' views of palliative care as "giving up" (Saad et al., 2022). This finding demonstrates how health professionals' attitudes towards palliative care and assumptions about what it involves can be a barrier to palliative care.

In both studies it was clear that without involvement of specialist palliative care teams, the core principles of palliative care were not consistently reported or delivered. It is worth noting that it was often the non-medical aspects of palliative care that were often missing. Specifically, advanced care planning; family preferences for end-of-life care, including preferred location of death; psychosocial support for the family; and bereavement support were not consistently reported as having been delivered in Study 1 and 2 publications. These findings demonstrate a clear gap and the potential to improve care in specific domains of palliative care delivery to ensure infants and their families are receiving optimal palliative care that is consistent and aligns with current evidence and standards for best practice. Specifically, the Australian National Safety and Quality Health Service Standards, implemented across Australian health services are mandatory to demonstrate processes are in place to support the provision of safe and quality healthcare for patients (Australian Commission on Safety and Quality in Health Care, 2021).

A component of the Australian national health standards is the Comprehensive Care Standard which addresses end-of-life care to ensure patients receive care that aligns with their individual needs and goals of care, with access to specialist palliative care, advance care planning, support and mentoring for all professionals, and routine assessment of the safety and quality of end-of-life care (Australian Commission on Safety and Quality in Health Care, 2021). Yet, the Comprehensive Care Standard focuses primarily on the end-of-life care dimension, rather than the whole palliative care journey (Australian Commission on Safety and Quality in Health Care, 2021). This results in core elements of palliative care not being specifically addressed, such as ensuring psychosocial aspects of care are provided, and bereavement support is offered. These elements lack a standardised process, and, as a consequence are not routinely documented.

To complement the Australian national health standards, the National Consensus Statement, provides ten recommendations for essential elements for delivering safe and high-quality paediatric end-of-life care (Australian Commission on Safety and

Quality in Health Care, 2016). The national consensus details more in-depth and comprehensive recommendations of care specifically for children and their families, and includes family-centred care, communication and decision making, recognising that a dual approach to care consisting of both palliative care and active treatment can be beneficial to the child and the psychosocial impact to the whole family, including bereavement support (Australian Commission on Safety and Quality in Health Care, 2016). It is clear the Comprehensive Care Standard does not fully reflect the national consensus recommendations. Therefore, there is an opportunity to extend the scope of the national standards to reflect the national consensus recommendations that are more comprehensive and suitable for infants and children. More recently, a Paediatric Palliative Care National Action Plan was published for endorsement (Palliative Care Australia, 2022). Key activities include focus on education for health professionals, referral pathways, guideline development for generalist health professionals, and research development (Palliative Care Australia, 2022). This national approach will aim to improve gaps in paediatric palliative care services and improving resources for health professionals and families.

To further build on delivering quality palliative care, dedicated paediatric hospices can play an integral role in providing comprehensive care to children and their families across the palliative care journey (Tatterton et al., 2023). Internationally, the development of paediatric hospices is more advanced in comparison to Australia, currently in the UK alone there are 54 hospices for children (Sisk et al., 2020; Tatterton et al., 2023). In Australia, there are currently three hospices, established over the past 20 years (Perth Children's Hospital Foundation, 2022). The development of WA’s first paediatric hospice is currently underway, (due to open in 2025) which will include the provision of respite, support for the whole family, and will offer families another option for their preferred location for end-of-life care (Perth Children's Hospital Foundation, 2022). A limitation of the hospice will be the location in Perth and, given the large landscape of WA, it may not be a suitable option for children and their families who live in rural and remote areas that are a long distance from Perth. For infants, however, the majority of infant deaths currently occur in the tertiary intensive care units in Perth, therefore, the hospice will provide a suitable and close by option for families to access. Interestingly, anecdotally, the paediatric hospice in Queensland has a very high proportion of neonates referred for end-of-life care, conversely, in Victoria, a limited number of neonates are referred for end-of-life care. To ensure infants and their families are able to access the WA hospice, it is imperative to develop seamless pathways for referrals to the hospice, and bridge gaps in health professionals' and families' understanding of palliative care (Western Australian Department of Health, 2021).

4.2 Implications for Practice and Future Research

In summary, this research has shown there is opportunity to further optimise palliative care for infants through; improved integration of specialist palliative care services in healthcare settings, and education to further develop health professionals' clinical practice and confidence to provide palliative care.

However, implementing change in practice is not easy or straightforward. Resistance to change is a common response experienced within healthcare services (DuBose & Mayo, 2020). There are several barriers to change in health that need to be addressed to successfully facilitate change (DuBose & Mayo, 2020). Central barriers to change have been reported as health professional fear of change, lack of trust in their leaders, communication barriers and feelings of threat to current practices (DuBose & Mayo, 2020). Specifically in palliative care, factors impacting change reported in a 2016 qualitative study were individual motivation, leadership and culture of change, organisational resources and policy, and ensuring strategies such as education and awareness to successfully implement change (Sommerbakk et al., 2016).

To successfully implement and manage change in practice in a health service environment requires a holistic approach that considers every aspect across the systemic, organisational, and individual levels. Applying Bronfenbrenner (1979) ecological systems model acknowledges the interconnectedness that change implemented at the systemic level will influence the organisational and individual/team levels (Neal & Neal, 2013), as shown in Figure 2.

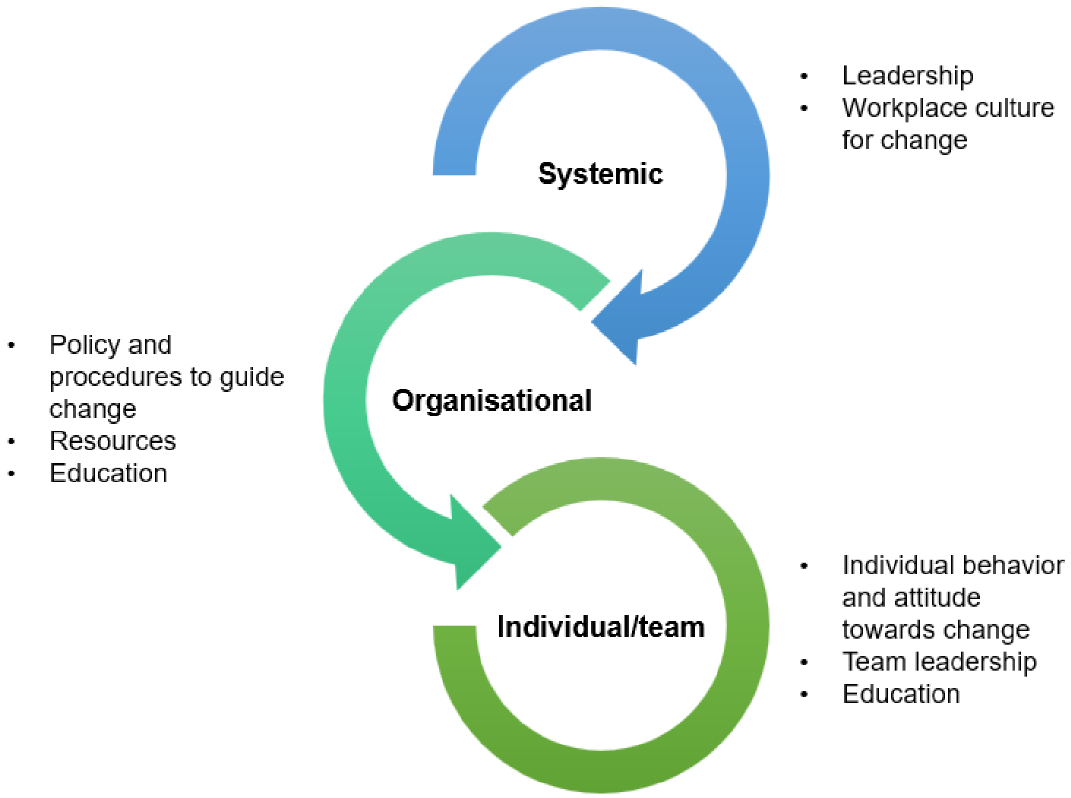

Figure 3. Key features of approach to change across systemic, organisational and individual/team levels

In addition to applying the key features of change across the three levels, targeting the appropriate profession of influence is vital. A 2020 qualitative study reported the influence of hierarchies and power structures as determinants of whether change is successfully implemented (Rogers et al., 2020). Rogers et al. (2020) discussed a conceptual framework for implementing change in health settings includes senior medical staff, hospital management and senior nurses in leadership or management roles. This is particularly relevant in the paediatric context where decisions for referring children to palliative care is largely determined by the medical profession, who are placed high in the hierarchies of decision making in healthcare policies and practices. Therefore, any intervention to affect change in the case of referring children to palliative care will need to target the medical profession.

There are several implications for practice at the systemic, organisational and individual/team level (Neal & Neal, 2013):

At the systemic level, a workplace culture that is open to change is imperative to successfully implement changes (Sommerbakk et al., 2016). Furthermore, a focus on further integrating specialist palliative care across the health service, where palliative care service team consultation is seen as enhancing care and not replacing already existing care. Strategies involve refining existing frameworks and guidelines to better guide clinical practice in palliative care, and inclusion of palliative care education into the curriculum for doctors and nurses. Another key strategy is implementing quality improvement activities to evaluate current palliative care practices and educational initiatives. In Australia, a national Palliative Care Outcomes Collaboration program implemented in health services where palliative care is provided for adults, uses routine collection of a scoring system to identify and measure the quality of palliative care (Eagar et al., 2010). Evaluation of the program showed improved clinical care and patient outcomes (Currow et al., 2015; Eagar et al., 2010). The program is yet to be adopted by paediatric hospitals across Australia, however, it may be an effective initiative to evaluate and improve palliative care delivery to children.

At the organisational level several strategies can be implemented. First, the organisation's commitment to implementing change and adhering to the national health standards to ensure children and their families receive comprehensive quality palliative care. Second, the promotion of the specialist palliative care team service through increasing the presence across health settings by raising awareness about the service. A simple initiative such as having specialist palliative care team presence at patient meetings to identify potential patients who would benefit from their input. Another initiative that has shown to increase the number of palliative care team service consults is a screening tool in a paediatric intensive care unit, where patients are screened weekly to identify who would benefit from a palliative care team consult (Hodge et al., 2023).

Finally, educational initiatives that include developing education programs in collaboration with the specialist palliative care service and specialist teams to meet specific learning needs, increase awareness about advance care planning, provide mentorship programs and opportunity to participate in clinical simulation workshops. Because providing palliative care or end-of-life care can be a rare event for some nurses and doctors, participation in clinical simulation provides opportunities to practice communication skills and end-of-life care using realistic scenarios in a safe learning environment (Smith et al., 2018). A recent report found that after simulation training multidisciplinary professionals from paediatric and neonatal settings experienced increased confidence in palliative care skills such as discussing advance care planning with families (Taylor et al., 2022).

At an individual/team level, strategies include access to professional development opportunities, including attending palliative care conferences to be informed of advancements in palliative care, and targeted palliative care education programs focused on developing individual skills, including self-reflection of strengths and areas to improve. Walter et al., (2019) outlined a conceptual model of barriers and facilitators for primary healthcare teams involving specialist palliative care services and recommendations for individual and team strategies to aid more seamless integration of palliative care. Specifically, for nurses who are at the forefront of delivering patient and family care, professional development opportunities can increase awareness about palliative care principles and practices, develop skills to communicate with families and support doctors with advance care planning discussions, increase ability to provide psychosocial care, bereavement support and identify patients and families who will benefit from specialist palliative care service consultation (DeSanto-Madeya et al., 2020).

4.2.1 Future Research

Further research that can build on the research presented in this thesis includes using qualitative and quantitative methods to explore doctors' and nurses' and other team members' attitudes and knowledge of paediatric palliative care and advance care planning in the WA setting. Studies that further examine the knowledge, barriers and attitudes of health professionals in the Australian setting, similar to the methodology described in Yi-An and Qian (2023) recent study will provide a deeper understanding of the knowledge and attitudes towards palliative care for infants and children across WA settings and identification of barriers specific to the WA context. Specifically, focus should be on exploring doctors' views and perspectives of palliative care, as they are the most influential in referring patients to palliative care, and nurses as they have the most contact with, and opportunity to talk to families.

The existing literature has acknowledged there have been limited studies exploring the family perspective (Saad et al., 2022). Importantly, extending the focus of research beyond the end-of-life phase has been recommended by Saad et al. (2022). Research is needed that explores the family perspective, in particular for families of infants who have had specialist palliative care involvement and their experiences. Increasing the evidence of the family perspective, and where the findings can be generalised will identify further the unmet needs of families, how to support them and lead to a more holistic and family centred approach to palliative care (Saad et al., 2022).

4.3 Strengths and Limitations

Study strengths and limitations were identified in each publication. Strengths of Study 1 were that for the first time it identified and examined the clinical and palliative care of infants with LLCs who died in WA and is one of only two published studies in Australia reviewing palliative care for infants in the past decade. A combined strength of Study 1 and Study 2 is the broad overview of the growing body of evidence in international and Australian context where there is still a gap in optimal palliative care delivery, specifically in neonatal intensive care settings.

There are several limitations for this research. The findings from Study 1 were limited to infants who died and did not capture the care management trajectories of infants with LLCs who survived beyond infancy. The inherent limitations of retrospective studies are dependent on accurate documentation in medical records. This may have resulted in missing or inaccurate data. Study 2 was limited to what was reported in peer reviewed articles, which was predominantly in developed countries, with few articles from developing countries. There is a need to increase the diversity of studies and representation of diverse cultures and to acknowledge the limitations of a review that included articles only published in English where studies may lack diverse representation and may compromise generalisability. A final limitation was that the Study 2 database search was completed in July 2022. Our recent search of the databases (30th December 2023) identified a further ten articles which further support findings and have been discussed in this chapter.

4.4 Recommendations

To optimise how palliative care is delivered to infants and improve integration of specialist palliative care services in health settings, several recommendations are suggested:

Systemic

- Address the key barriers of change within the health setting to further integrate specialist palliative care services.
- For specialist palliative care services and palliative care providers to work in collaboration and informed by the Paediatric Palliative Care National Action Plan.
- Update the Comprehensive Care Standard and other related documents such as the WA Perinatal Model of Care.

Organisational

- To promote greater opportunities for education using diverse approaches to building and developing health professionals' knowledge and confidence to deliver palliative care. Furthermore, to evaluate the efficacy of education initiatives.

Individual/team

- Access professional development opportunities to develop specific skills to provide palliative care.
- Explore the barriers, knowledge and attitudes of health professionals towards palliative care for infants in the West Australian context.
- Explore the family perspective and views of palliative care across their whole journey, not limited to the end-of-life phase.

4.5 Conclusion

This study involved a retrospective review of the care of infants with LLCs who died in WA and an integrative review of the international literature to synthesise how palliative care is delivered to infants with LLCs. The study revealed infants were infrequently referred to specialist palliative care services and that palliative care was predominately delivered in neonatal intensive care units by primary care health professionals. Only a small number of infants received consultation from specialist palliative care service teams. This study found there are gaps and inconsistencies in the integration of specialist palliative care services in health settings, the clinical practice of palliative care delivery to infants and the psychosocial support provided to families across local as well as international contexts. Key recommendations to optimise the quality of palliative care to infants and their families across all aspects of care include addressing the key barriers to integrating specialist palliative care services in infant’s settings, and improving health professionals’ skills and capacity to provide palliative care to infants that is timely and responsive to the individual needs.

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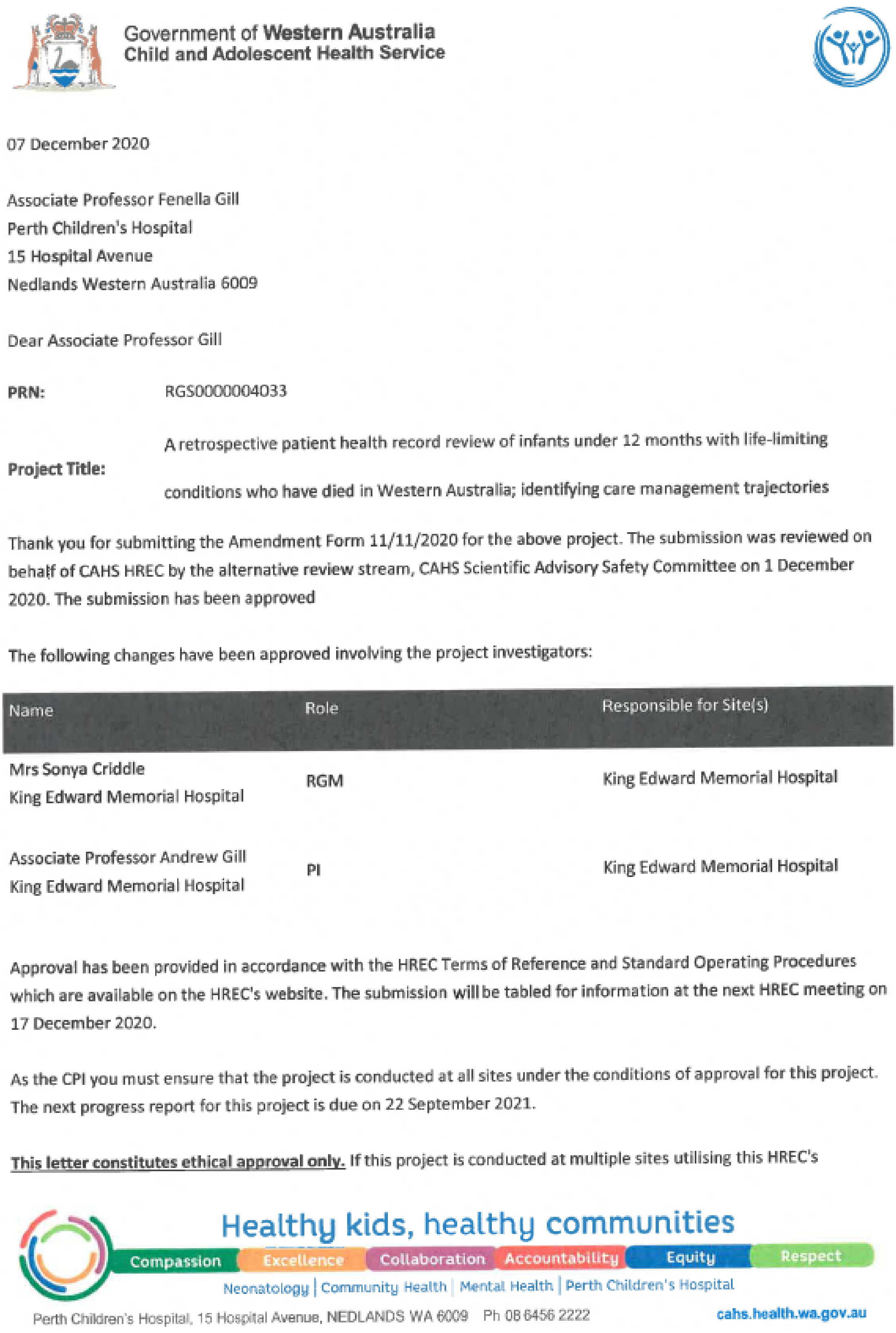
Every reasonable attempt has been made to acknowledge the owners of copyright material. I would be pleased to hear from any copyright owner who has been omitted or incorrectly acknowledged.

APPENDIX 1 CHILD AND ADOLESCENT HEALTH SERVICE HUMAN RESEARCH ETHICS COMMITTEE APPROVAL



Government of **Western Australia**
Child and Adolescent Health Service

07 December 2020

Associate Professor Fenella Gill
Perth Children's Hospital
15 Hospital Avenue
Nedlands Western Australia 6009

Dear Associate Professor Gill

PRN:	RGS0000004033
Project Title:	A retrospective patient health record review of infants under 12 months with life-limiting conditions who have died in Western Australia; identifying care management trajectories

Thank you for submitting the Amendment Form 11/11/2020 for the above project. The submission was reviewed on behalf of CAHS HREC by the alternative review stream, CAHS Scientific Advisory Safety Committee on 1 December 2020. The submission has been approved

The following changes have been approved involving the project investigators:

Name	Role	Responsible for Site(s)
Mrs Sonya Criddle King Edward Memorial Hospital	RGM	King Edward Memorial Hospital
Associate Professor Andrew Gill King Edward Memorial Hospital	PI	King Edward Memorial Hospital

Approval has been provided in accordance with the HREC Terms of Reference and Standard Operating Procedures which are available on the HREC's website. The submission will be tabled for information at the next HREC meeting on 17 December 2020.

As the CPI you must ensure that the project is conducted at all sites under the conditions of approval for this project. The next progress report for this project is due on 22 September 2021.

<u>**This letter constitutes ethical approval only.**</u> If this project is conducted at multiple sites utilising this HREC's

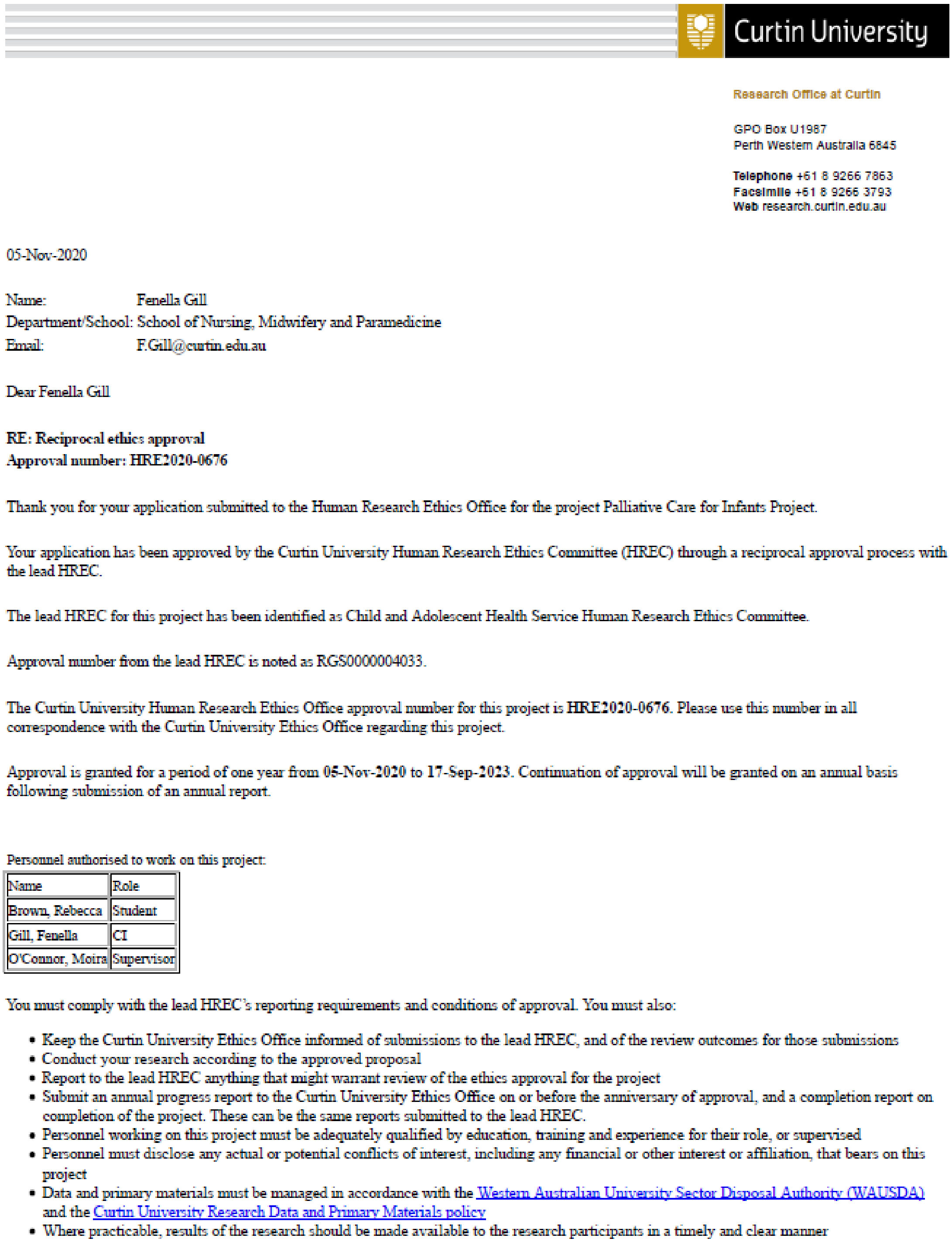
approval, a copy of this letter must be made available to all site PIs to maintain authorisation from their site. If you require further information, please contact the HREC Office on (08) 6456 0516 or CAHS.Ethics@health.wa.gov.au. To find the original letter, click here when logged into RGS.

Yours sincerely

pp

Clinical Professor Catherine Choong
On behalf of the Child and Adolescent Health Service Human Research Ethics Committee

APPENDIX 2 UNIVERSITY HUMAN RESEARCH ETHICS COMMITTEE APPROVAL



Curtin University

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05-Nov-2020

Name: Fenella Gill
Department/School: School of Nursing, Midwifery and Paramedicine
Email: F.Gill@curtin.edu.au

Dear Fenella Gill

RE: Reciprocal ethics approval
Approval number: HRE2020-0676

Thank you for your application submitted to the Human Research Ethics Office for the project Palliative Care for Infants Project.

Your application has been approved by the Curtin University Human Research Ethics Committee (HREC) through a reciprocal approval process with the lead HREC.

The lead HREC for this project has been identified as Child and Adolescent Health Service Human Research Ethics Committee.

Approval number from the lead HREC is noted as RGS0000004033.

The Curtin University Human Research Ethics Office approval number for this project is **HRE2020-0676**. Please use this number in all correspondence with the Curtin University Ethics Office regarding this project.

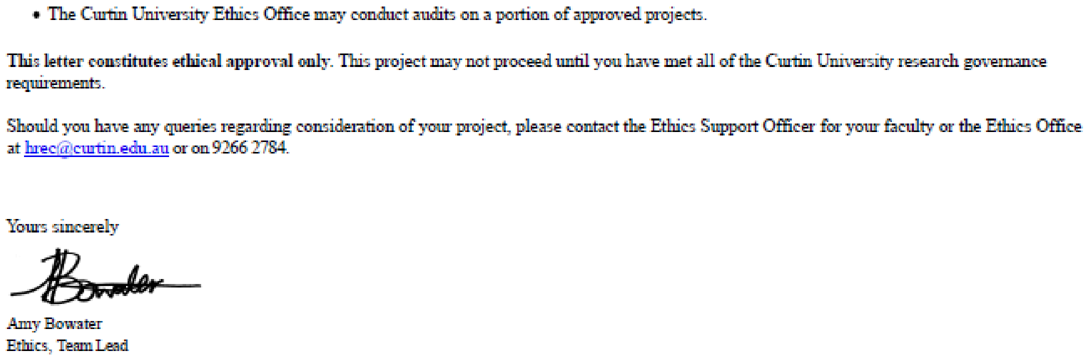
Approval is granted for a period of one year from **05-Nov-2020** to **17-Sep-2023**. Continuation of approval will be granted on an annual basis following submission of an annual report.

Personnel authorised to work on this project:

Name	Role
Brown, Rebecca	Student
Gill, Fenella	CI
O'Connor, Moira	Supervisor

You must comply with the lead HREC's reporting requirements and conditions of approval. You must also:

- Keep the Curtin University Ethics Office informed of submissions to the lead HREC, and of the review outcomes for those submissions
- Conduct your research according to the approved proposal
- Report to the lead HREC anything that might warrant review of the ethics approval for the project
- Submit an annual progress report to the Curtin University Ethics Office on or before the anniversary of approval, and a completion report on completion of the project. These can be the same reports submitted to the lead HREC.
- Personnel working on this project must be adequately qualified by education, training and experience for their role, or supervised
- Personnel must disclose any actual or potential conflicts of interest, including any financial or other interest or affiliation, that bears on this project
- Data and primary materials must be managed in accordance with the Western Australian University Sector Disposal Authority (WAUSDA) and the Curtin University Research Data and Primary Materials policy
- Where practicable, results of the research should be made available to the research participants in a timely and clear manner



- The Curtin University Ethics Office may conduct audits on a portion of approved projects.

This letter constitutes ethical approval only. This project may not proceed until you have met all of the Curtin University research governance requirements.

Should you have any queries regarding consideration of your project, please contact the Ethics Support Officer for your faculty or the Ethics Office at hrec@curtin.edu.au or on 9266 2784.

Yours sincerely

Amy Bowater
Ethics, Team Lead

APPENDIX 3 SUMMARY WITH DR NATASHA CAMPING

Below is a summary of the minutes with Dr Natasha on the 30 November 2021.

Advice for discussion with the stakeholder group	• Ask for their help to interpret the care management trajectories? What does this mean • Not just specialty based
Terminology / phrases	• Consider terminology / phrases that might not be internationally recognised. i.e. comfort care, journeys, pathways
Care management trajectories	Group 2 – smaller number of cases in this group for Bec's research. Why? Because we excluded unexpected deaths or those that didn't have a LLC Group 4 • Higher number of deaths at home • Option for parents • Bereavement support more documented • More palliative care referrals
Referral to palliative care vs palliative approach?	Is a referral to SPC required if everyone can deliver palliative care? Natashas comment > bring it back to what do the parents want? Patient and family centred care.
When describing something not documented	Consistently missed, these are the implications.
Journal advice	Consider avoiding palliative care journal as this could be limiting, you are not trying to sell it to palliative care specialists.