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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.

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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 Institute 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 Institute 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; Harmony 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 (Treble 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:

- 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.
- 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
Comorbidities [‡]	Sepsis	3	Comorbidities [‡]	Hyaline membrane disease	3
	Subgaleal haemorrhage	1		Pulmonary atresia	1
	Seizures	4		Pulmonary hypertension	1
	Organ failure	2		Hypotension	1
	Raised intracranial pressure	1		Sepsis	1
	Sepsis	1		Multi organ failure	1
	Intracranial haemorrhage	1		Patent ductus arteriosus	3
	Hypovolemic shock	1		Metabolic acidosis	1
	Pulmonary hypertension	1		Intravascular coagulation	1
	Hypoglycemia	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			
	For baptism or blessing	3			
EOLC [¶] preferences & priorities	Memory making; photos, bath, cuddles	3	EOLC [¶] preferences & priorities	Not applicable [¶]	2
	Withdrawal of ventilation outside	1		No documentation	2
	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 tetrahydrofolate reductase deficiency (MTHFR)	1
	Ornithine transcarbamylase deficiency	1		Common truncus arteriosus	1
	Severe pulmonary hypertension	1		Congenital fibre type disproportion	1
	Congenital heart disease	3		D-bifunctional protein deficiency	1
Comorbidities [‡]			Comorbidities [‡]	Hypoplastic left heart	
	Necrotising enterocolitis	3		Myotubular myopathy	
	Multiorgan failure	4		Critical aortic & mitral stenosis	
	Mitral valve dysplasia	1		Global delay	1
	Ventricular septal defect	1		Microcephaly	1
	Chronic lung disease	3		Failure to thrive	1
	Oropharyngeal dysplasia	1		Hypotonia	1
	Metabolic encephalopathy	1		Seizures	1
	Hyperammonaemia	1		Obstructed breathing	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 [∞]			
	Deterioration despite max treatment	18	Prenatal diagnosis	5		
	Clinical investigations	2	Postnatal diagnosis	4		
	Dependency on ventilator	11	No curative treatment options	8		
	Multi organ failure	4	Severe disability with poor quality of life	1		
	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				
EOLC preferences & priorities	Admission to ICU	1	EOLC preferences & priorities	Spend quality time together	1	
	Comfort	4		Focus on comfort	4	
	Hold baby before EOL	5		To go home	5	
	Give baby a bath	2		Being together as family	2	
	Spend time together as family	1		Take baby to beach; put feet in water	1	
	Strong Islamic faith	1		Breast feed baby	1	
	Support for sibling	1				
	Parents requested twins to be together	1				
Bereavement follow up contact	No documentation	10	Bereavement follow up	1 [‡]	0	
	1 [‡]	5		>1	5	
	>1	6		Nil	4	
	Nil	10				

[†] 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

[‡] 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

CARE MANAGEMENT TRAJECTORY TYPE 1
EARLY DE-ESCALATION DUE TO CATASTROPHIC EVENT

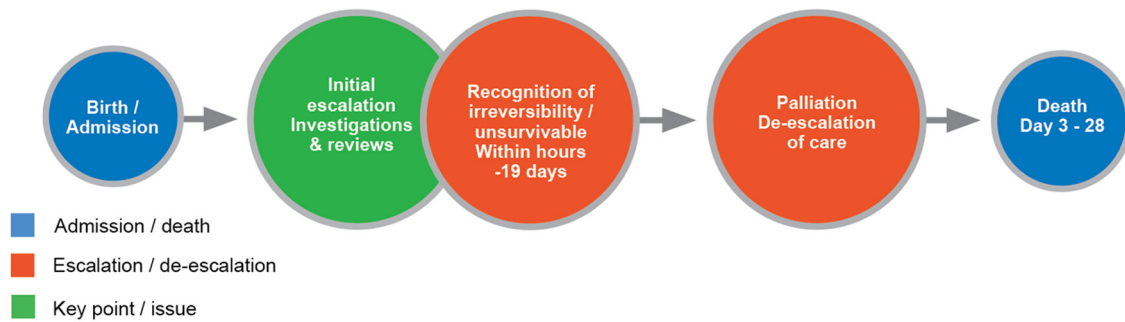


Fig. 1. Care management trajectory type 1.

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 (pre-term 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 Institute 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).

CARE MANAGEMENT TRAJECTORY TYPE 2
TREATMENT WITH CURATIVE INTENT THROUGHOUT

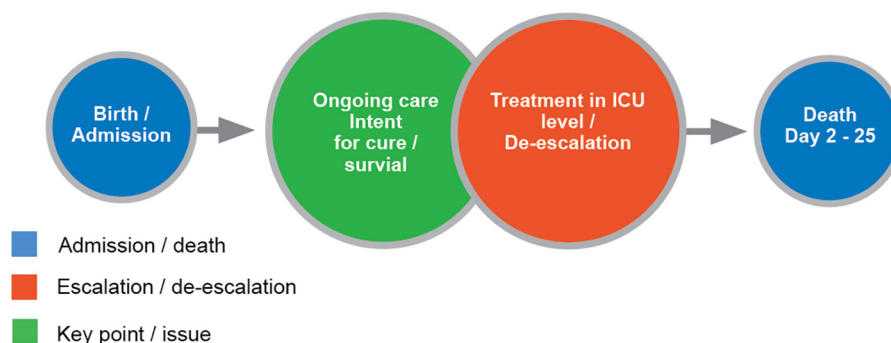


Fig. 2. Care management trajectory type 2.

CARE MANAGEMENT TRAJECTORY TYPE 3
TREATMENT WITH CURATIVE INTENT UNTIL SIGNIFICANT POINT

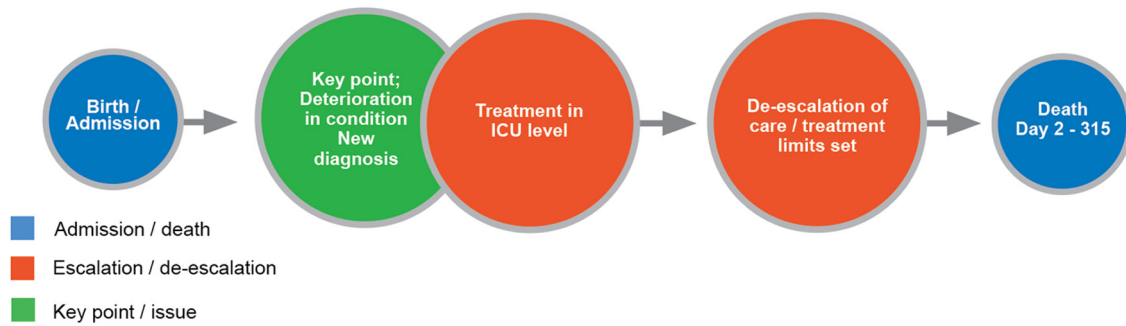


Fig. 3. Care management trajectory type 3.

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; Harmony 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).

CARE MANAGEMENT TRAJECTORY TYPE 4
EARLY TREATMENT LIMITS SET

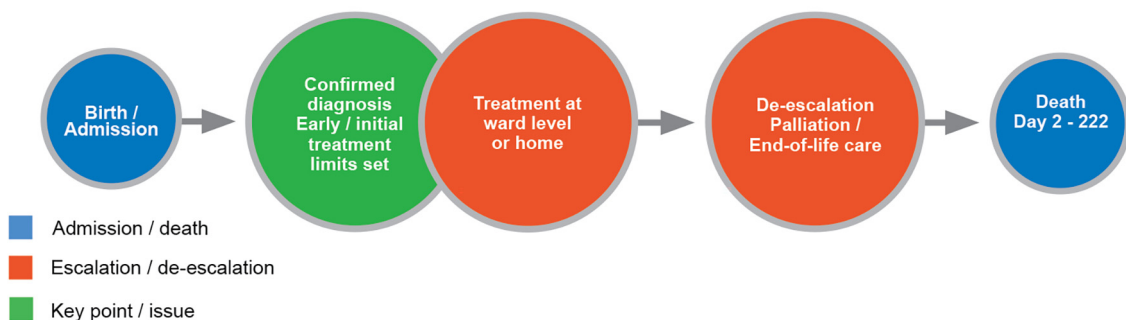


Fig. 4. Care management trajectory type 4.

Bereavement support and coordination for the families of infants after their death was not consistently reported. We found that many families appeared to not 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>.

References

- Assarroudi, A., Heshmati Nabavi, F., Armat, M. R., Ebadi, A., & Vaismoradi, M. (2018). Directed qualitative content analysis: The description and elaboration of its underpinning methods and data analysis process. *Journal of Research in Nursing*, 23(1), 42–55. <https://doi.org/10.1177/1744987117741667>.
- Audigé, M., Gillam, L., & Stark, Z. (2020). Treatment limitation and advance planning: Hospital-wide audit of paediatric death. *Journal of Paediatrics and Child Health*, 56(6), 893–899. <https://doi.org/10.1111/jpc.14771>.
- Australian Commission on Safety and Quality in Health Care (2016). *National Consensus Statement: Essential elements for safe and high-quality paediatric end-of-life care*. Sydney: ACSQHC. <https://www.safetyandquality.gov.au/sites/default/files/migrated/National-Consensus-Statement-Essential-Elements-for-safe-high-quality-end-of-life-care.pdf>.
- Australian Institute of Health and Welfare (2019). *Australia's Children - in brief*. <https://doi.org/10.25816/5e152818d082c>.
- Australian Institute of Health and Welfare (2019). *AIHW National Hospital Morbidity Database*. <https://www.aihw.gov.au/>.
- Australian Institute of Health and Welfare (2020). *National Hospital Morbidity Database*. <https://www.aihw.gov.au/reports-data/myhospitals/hospital/h0746>.
- Benchimol, E., Smeeth, L., Guttman, A., Harron, K., Moher, D., Petersen, I., ... Comittee., R. W. (2015). The REporting of studies conducted using observational Routinely-collected health data (RECORD) statement. *PLoS Medicine*, 12(10). <https://doi.org/10.1371/journal.pmed.1001885>.
- Bolognani, M., Morelli, P. D., Scolari, I., Dolci, C., Fiorito, V., Uez, F., ... Soffiati, M. (2020). Development of a perinatal palliative care model at a level II perinatal center supported by a pediatric palliative care network. *Frontiers in Pediatrics*, 8, Article 574397. <https://doi.org/10.3389/fped.2020.574397>.
- Campling, N., Cummings, A., Myall, M., Lund, S., May, C. R., Pearce, N. W., & Richardson, A. (2018). Escalation-related decision making in acute deterioration: A retrospective

- case note review. *BMJ Open*, 8(8), Article e022021. <https://doi.org/10.1136/bmjopen-2018-022021>.
- Catlin, A., & Carter, B. (2002). Creation of a neonatal end-of-life palliative care protocol. *Journal of Perinatology*, 22(3), 184–195. <https://doi.org/10.1038/sj.jp.7210687>.
- Charlebois, J., & Cyr, C. (2015). Quality indicators for paediatric palliative care. *Paediatrics & Child Health*, 20(3), 145–147. <https://doi.org/10.1093/pch/20.3.145>.
- Commonwealth of Australia (2019). Risk of preterm birth. <https://www.health.gov.au/resources/pregnancy-care-guidelines/part-d-clinical-assessments/risk-of-preterm-birth>.
- Currie, E., Christian, B., Hinds, P., Perna, S., Robinson, C., Day, S., & Meneses, K. (2016). Parent perspectives of neonatal intensive care at the end-of-life. *Journal of Paediatric Nursing*, 31, 478–489. <https://doi.org/10.1016/j.pedn.2016.03.023>.
- Department of Health Western Australia (2015). Perinatal Palliative Care Model of Care. Western Australia. <https://ww2.health.wa.gov.au/~media/Files/Corporate/general%20documents/Health%20Networks/WA%20Cancer%20and%20Palliative%20Care/Palliative%20care/Perinatal-Palliative-Care-Model-of-Care.pdf>.
- Ekberg, S., Bowers, A., Bradford, N., Ekberg, K., Rolfe, M., Elvidge, N., ... Vickery, A. (2021). Enhancing paediatric palliative care: A rapid review to inform continued development of care for children with life-limiting conditions. *Journal of Paediatrics and Child Health*, 58, 232–237. <https://doi.org/10.1111/jpc.15851>.
- Fontana, M. S., Farrell, C., Gauvin, F., Lacroix, J., & Janvier, A. (2013). Modes of death in paediatrics: Differences in the ethical approach in neonatal and pediatric patients. *Journal of Pediatrics*, 162(6), 1107–1111. <https://doi.org/10.1097/PCC.0000000000002259>.
- Fraser, L. K., Fleming, S., & Parslow, R. (2018). Changing place of death in children who died after discharge from paediatric intensive care units: A national, data linkage study. *Palliative Medicine*, 32(2), 337–346. <https://doi.org/10.1177/0269216317709711>.
- Fraser, L. K., Lidstone, V., Miller, M., Aldridge, J., Norman, P., McKinney, P. A., & Parslow, R. C. (2014). Patterns of diagnoses among children and young adults with life-limiting conditions: A secondary analysis of a national dataset. *Palliative Medicine*, 28(6), 513–520. <https://doi.org/10.1177/0269216314528743>.
- Gibelli, M., De Carvalho, W. B., & Krebs, V. L. J. (2021). Limits of therapeutic intervention in a tertiary neonatal intensive care unit in patients with major congenital anomalies in Brazil. *Journal of Paediatrics and Child Health*, 57(12), 1966–1970. <https://doi.org/10.1111/jpc.15630>.
- Gilmour, D., Davies, M. W., & Herbert, A. R. (2017). Adequacy of palliative care in a single tertiary neonatal unit. *Journal of Paediatrics and Child Health*, 53(2), 136–144. <https://doi.org/10.1111/jpc.13353>.
- Government of Western Australia (2021). King Edward Memorial Hospital history and alumni. <https://kemh.health.wa.gov.au/About-us/History>.
- Harmoney, K., Mobley, E. M., Gilbertson-White, S., Brogden, N. K., & Benson, R. J. (2019). Differences in advance care planning and circumstances of death for pediatric patients who do and do not receive palliative care consults: A single-center retrospective review of all pediatric deaths from 2012 to 2016. *Journal of Palliative Medicine*, 22(12), 1506–1514. <https://doi.org/10.1089/jpm.2019.0111>.
- Harris, P. A., Taylor, R., Minor, B. L., Elliott, V., Fernandez, M., O'Neal, L., ... Consortium, R. (2019). The REDCap consortium: Building an international community of software platform partners. *Journal of Biomedical Informatics*, 95, 103208. <https://doi.org/10.1016/j.jbi.2019.103208>.
- Horridge, K. (2015). Advance care planning: Practicalities, legalities, complexities and controversies. *Archives of Disease in Childhood*, 100, 380–385. <https://doi.org/10.1136/archdischild-2014-305945>.
- Hsieh, H. F., & Shannon, S. E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research*, 15(9), 1277–1288. <https://doi.org/10.1177/1049732305276687>.
- Jack, B. A., Mitchell, T. K., O'Brien, M. R., Silverio, S. A., & Knighting, K. (2018). A qualitative study of health care professionals' views and experiences of paediatric advance care planning. *BMC Palliative Care*, 17(1), 93. <https://doi.org/10.1186/s12904-018-0347-8>.
- Kamrath, H. J., Osterholm, E., Stover-Haney, R., George, T., O'Connor-Von, S., & Needle, J. (2019). Lasting legacy: Maternal perspectives of perinatal palliative care. *Journal of Palliative Medicine*, 22(3), 310–315. <https://doi.org/10.1089/jpm.2018.0303>.
- Kenner, C., Press, J., & Ryan, D. (2015). Recommendations for palliative and bereavement care in the NICU: A family-centered integrative approach. *Journal of Perinatology*, 35 (Suppl. 1), S19–S23. <https://doi.org/10.1038/jp.2015.145>.
- Kukora, S., Gollehon, N., & Laventhal, N. (2017). Antenatal palliative care consultation: Implications for decision-making and perinatal outcomes in a single-Centre experience. *Archives of Disease in Childhood. Fetal and Neonatal Edition*, 102(1), F12–F16. <https://doi.org/10.1136/archdischild-2016-311027>.
- Marc-Aurele, K. L., & English, N. K. (2017). Primary palliative care in neonatal intensive care. *Seminars in Perinatology*, 41(2), 133–139. <https://doi.org/10.1053/j.semperi.2016.11.005>.
- Mitchell, S., & Dale, J. (2015). Advance care planning in palliative care: A qualitative investigation into the perspective of paediatric intensive care unit staff. *Palliative Medicine*, 29(4), 371–379. <https://doi.org/10.1177/0269216315573000>.
- Mitchell, S., Morris, A., Bennett, K., Sajid, L., & Dale, J. (2017). Specialist paediatric palliative care services: What are the benefits? *Archives of Disease in Childhood*, 102(10), 923–929. <https://doi.org/10.1136/archdischild-2016-312026>.
- Mitchell, S., Plunkett, A., & Dale, J. (2014). Use of formal advance care planning documents: A national survey of UK Paediatric intensive care units. *Archives of Disease in Childhood*, 99(4), 327–330. <https://doi.org/10.1136/archdischild-2013-304463>.
- Mitchell, S., Spry, J. L., Hill, E., Coad, J., Dale, J., & Plunkett, A. (2019). Parental experiences of end of life care decision-making for children with life-limiting conditions in the paediatric intensive care unit: A qualitative interview study. *BMJ Open*, 9(5), e028548. <https://doi.org/10.1136/archdischild-2016-311027>.
- Moro, T., Kavanaugh, K., Okuno-Jones, S., & Vankleef, J. A. (2006). Neonatal end-of-life care: A review of the research literature. *The Journal of Perinatal & Neonatal Nursing*, 20(3), 262–273. <https://doi.org/10.1097/00005237-200607000-00015>.
- National Centre for Health Statistics (2019). Linked birth and infant death data. <https://www.cdc.gov/nchs/nvss/linked-birth.htm>.
- National Institute for Health and Care Excellence (2016). End of life care for infants, children and young people, with life limiting conditions: Planning and management. <https://www.nice.org.uk/guidance/ng61/resources/end-of-life-care-for-infants-children-and-young-people-with-lifelimiting-conditions-planning-and-management-pdf-1837568722885>.
- O'Brien, B. C., Harris, I. B., Beckman, T. J., Reed, D. A., & Cook, D. A. (2014). Standards for reporting qualitative research: A synthesis of recommendations. *Academic Medicine*, 89(9), 1245–1251. <https://doi.org/10.1097/ACM.0000000000000388>.
- Office for National Statistics (2019). Child and infant mortality in England and Wales: 2019. <https://www.ons.gov.uk/peoplepopulationandcommunity/birthsdeathsandmarriages/deaths/bulletins/childhoodinfantandperinatalmortalityinenglandandwales/2019>.
- Palliative Care Australia (2018). Paediatric Addendum - Palliative Care Service Development Guidelines. Retrieved from Canberra: https://palliativecare.org.au/wp-content/uploads/dlm_uploads/2018/12/PalliativeCare-Paediatricaddendum-2018_web.pdf.
- Quinn, M., & Gephart, S. (2016). Evidence for implementation strategies to provide palliative care in the neonatal intensive care unit. *Advances in Neonatal Care*, 16(6), 430–438. <https://doi.org/10.1097/ANC.0000000000000354>.
- Ramelet, A., Bergstraesser, E., Grandjean, C., Dorsaz, A., Fahrni-Nater, P., Cignacco, E., & Zimmermann, K. (2020). Comparison of end-of-life care practices between children with complex chronic conditions and neonates dying in an ICU versus non-ICUs: A substudy of the pediatric end-of-life Care needs in Switzerland (PELLICAN) project. *Paediatric Critical Care Medicine*, 20(5). <https://doi.org/10.1097/PCC.0000000000002259>.
- Sidgwick, P., Fraser, J., Fortune, P. M., & McCulloch, R. (2019). Parallel planning and the paediatric critical care patient. *Archives of Disease in Childhood*, 104(10), 994–997. <https://doi.org/10.1136/archdischild-2018-315222>.
- Stark, Z., Hynson, J., & Forrester, M. (2008). Discussing withdrawing and withholding of life-sustaining treatment in a tertiary paediatric hospital: A survey of clinician attitudes and practices. *Journal of Paediatrics and Child Health*, 44, 392–398. <https://doi.org/10.1111/j.1440-1754.2008.01351.x>.
- Stutz, M., Kao, R. L., Huard, L., Grotts, J., Sanz, J., & Ross, M. K. (2018). Associations between pediatric palliative care consultation and end-of-life preparation at an academic medical center: A retrospective EHR analysis. *Hospital Pediatrics*, 8(3), 162–167. <https://doi.org/10.1542/hpeds.2017-0016>.
- Tan, J. S., Docherty, S. L., Barfield, R., & Brandon, D. H. (2012). Addressing parental bereavement support needs at the end of life for infants with complex chronic conditions. *Journal of Palliative Medicine*, 15(5), 579–584. <https://doi.org/10.1089/jpm.2011.0357>.
- Together for Short Lives (2017). *A Perinatal Pathway for Babies with Palliative Care Needs* (2nd ed.). <https://www.togetherforshortlives.org.uk/resource/perinatal-pathway-babies-palliative-care-needs/>.
- Trebble, T. M., Hansi, N., Hydes, T., Smith, M. A., & Baker, M. (2010). Process mapping the patient journey: An introduction. *BMJ*, 341, Article c4078. <https://doi.org/10.1136/bmj.c4078>.
- Western Australian Department of Health (2021). Western Australian Paediatric Strategy for End-of-life and Palliative Care 2021–2028. Retrieved from Perth: <https://ww2.health.wa.gov.au>.
- World Health Organisation (1998). WHO Definition of Palliative Care. Retrieved from: <https://www.who.int/cancer/palliative/definition/en/>.
- World Health Organisation. (2012). Preterm birth. <https://www.who.int/news-room/fact-sheets/detail/preterm-birth>.