



Shared decision-making experiences in child long-term ventilation: a systematic review

George Peat ^{1,2}, Sally-Ann Delaney³, Faith Gibson ^{4,5}, Lorna K. Fraser^{2,6} and Joe Brierley^{4,7}

¹Department of Health Sciences, University of York, York, UK. ²Collaborative Paediatric Palliative Care Research Network. ³North Middlesex University Hospital NHS Trust, Middlesex, UK. ⁴NIHR Great Ormond Street Hospital Biomedical Research Centre, London, UK. ⁵School of Health Sciences, University of Surrey, Guildford, UK. ⁶Cicely Saunders Institute of Palliative Care, Policy & Rehabilitation, Kings College, London, UK. ⁷Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK.

Corresponding author: George Peat (george.peat@york.ac.uk)



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Implementing shared decision-making practice in child long-term ventilation is challenging. Ensuring caregivers feel informed, acknowledging their unique position and using honest, clear and timely dialogue can support shared decision-making. <https://bit.ly/3r9EpeP>

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Abstract

Introduction: Recent decades have seen an increase in children receiving long-term ventilation. To ensure that long-term ventilation decisions incorporate the perspectives of stakeholders, it is vital that empirical evidence is gathered to substantiate frameworks and guidance on shared decision-making for long-term ventilation. This systematic review and qualitative evidence synthesis aimed to clarify what shared decision-making constitutes in relation to long-term ventilation initiation for children and young people (<21 years).

Methods: A systematic review of qualitative research was undertaken. Searches were conducted in MEDLINE, Embase, CINAHL, PsycINFO and Web of Science.

Results: Findings from 13 studies were included representative of 363 caregivers and 143 healthcare professional experiences. Components that support shared decision-making included acknowledging the unique positionality of caregivers and ensuring caregivers were informed about the implications of long-term ventilation. Beneficial qualities of engagement between stakeholders included honest, clear and timely dialogue using lay, tactful and sensitive language.

Conclusion: Our findings clarify components and approaches supportive of shared decision-making in discussions about long-term ventilation. This review therefore provides a valuable resource to implement shared decision-making practices in the context of long-term ventilation decisions for children and young people.

Introduction

Advances in medical science have enabled many children and young people to survive with the aid of medical technologies. One key technology is long-term ventilation (LTV), defined as ventilatory support lasting at least 3 months and includes invasive and noninvasive options [1]. LTV can be intended as a bridge therapy to a state where the technology is no longer required, such as to support premature babies with respiratory distress syndrome. Alternatively, LTV can be regarded as lifelong treatment to prevent respiratory insufficiency for people with conditions such as Duchenne muscular dystrophy [2]. Regardless of the pathway, whether to initiate LTV is a complex, individual and dynamic decision often made on a case-by-case basis reflective of the heterogeneous nature of these populations [3, 4].

Shared decision-making (SDM) exists as a principle in the UK National Health Service (NHS) constitution that states, “Patients, with their families and carers, where appropriate, will be involved and consulted on all decisions about their care and treatment” [5]. Enacting SDM in practice relies firstly on patients and their families receiving the necessary information about the risks and benefits associated with the available treatment options. More so, patients and their families should be given the opportunity to express their



preferences and, where appropriate, deliberate with healthcare professionals to determine the best treatment plan [6].

The factors that surround a decision to initiate LTV are complex and particularly warrant the existence of SDM principles. For example, there may be considerable uncertainty about the future prognosis of the child. Likewise, the clinical circumstances of children who require LTV means caregivers often must advocate for their child in decisions made about their care. Caregivers must do this while likely contending with their own anxieties and concerns, in environments that are often highly stressful and emotionally draining [7–9]. As such, ensuring caregivers are appropriately informed and supported to be a participant in decisions made is critical to reaching shared agreement on the most appropriate treatment plan for their child.

The value of SDM principles in decisions about LTV for children is formerly recognised in practice guidelines [10]. These guidelines allude to the importance of practices such as consistent, open and empathetic dialogue between all parties. Also highlighted is the continued assessment of patient and family understanding of illness and treatment options. Likewise, service specifications for paediatric LTV published by NHS England state that the child and family must be provided the opportunity to discuss LTV with specialists and be given “condition-specific” information [11].

Despite the publication of such guidelines, there is minimal empirical evidence or understanding of the application of SDM in the practice of LTV initiation. This is significant because the number of children and young people (0–24 years) on LTV is increasing rapidly, with an estimated 2382 in 2019 in the UK [12]. Consequently, there is a growing need to substantiate existing opinion-based guidance with empirical evidence and understanding. To address this gap, in this systematic review and qualitative synthesis, we aim to clarify what SDM constitutes in relation to LTV initiation by exploring how children, young people, caregivers and healthcare professionals practically experience SDM in LTV initiation.

Methods

Study design

This review is reported according to the Enhancing Transparency in Reporting the Synthesis of Qualitative Research framework [13]. The review protocol was not registered.

Eligibility criteria

Studies were included using the following criteria:

- They used qualitative research methods to explore experiences of decision-making concerning the initiation of LTV or tracheostomy leading to LTV (defined as >3 months [1]) in patients <21 years, from any combination of child, caregiver and healthcare professional perspectives.
- In cases where the study focused on tracheostomy initiation, a sample included patients who went on to receive LTV.
- A mixed-methods approach was used, with qualitative data reported separately so it could be extracted.
- They were published in the English language.

Studies were excluded if they included the experiences of decision-making about LTV initiation in patients >21 years.

Search strategy

The search strategy was developed using terms for child, ventilation and decision-making and a qualitative filter was applied (see supplementary material). The SPIDER tool was used to define terms for each concept and advice from an information specialist was sought to support initial development. Exemplar articles were identified before the search and used to check the accuracy of searches. Searches were run in electronic databases MEDLINE, Embase, CINAHL, PsycINFO and Web of Science from inception to April 2022, using a combination of medical subject headings, keyword and free-text terms.

To supplement electronic searches, citation searching, searching of referencing lists and a Google Scholar advanced search were also undertaken. Searches were imported to EndNote (Clarivate Analytics, Philadelphia, PA, USA), duplicates removed and results uploaded to Covidence (Covidence systematic review software; Veritas Health Innovation, Melbourne, Australia; www.covidence.org) for screening.

Title and abstract screening were undertaken by G.P. and S.D., with ongoing discussion throughout. Any disagreements were resolved in consultation with a third reviewer (L.F.). Full-text screening was undertaken with reference to the inclusion criteria by G.P. and S.D. and any disagreements resolved as above.

Data extraction

Key properties of each study, including title, year of publication, year of data collection, country, setting, aims, methodology, methods and sample characteristics, were extracted. All data labelled as “findings”, “results” or pertaining to such, including results reported in the discussion, were extracted to NVivo (version 12 (2018); Lumivero; www.lumivero.com). This included authors’ interpretations as well as quotes from participants.

Quality appraisal

A modified version of the Critical Appraisal Skills Programme [14] was used to undertake quality appraisal with the selected studies. The modified version was used because it provides greater clarity to assess the philosophical underpinning of qualitative studies. Included studies were ranked “high”, “medium” or “low” quality, although studies were not excluded on account of their quality, as doing so is not linked to improved review quality [15].

Synthesis

A thematic analysis [16] of qualitative data was undertaken. A thematic approach was appropriate in line with the aim of the review to explore lived experiences of SDM practice. A case-by-case approach to synthesis was first undertaken, whereby text from each article was coded line-by-line. This process generated a bank of codes which were then iteratively revised as codes from individual papers were compared and renamed where appropriate. Descriptive themes were then developed, grouping codes together to present a summative account of the data. A final layer of synthesis followed with the aim of developing analytical themes that went beyond description to offer a linkage between the review aims and analysed data. Coding was principally carried out by one reviewer (G.P.) with input from the review team, particularly as descriptive and analytical themes were developed.

Results

Identification and selection of studies

The electronic database searches identified 2143 unique results. A total of 2063 studies were excluded during title and abstract screening leaving 80 papers that were assessed against the eligibility criteria during full-text screening. 63 papers were excluded during full-text screening, leading to the inclusion of 17 papers from 13 studies (figure 1).

Characteristics of included studies

Studies were published between 2008 and 2022 with data collection taking place between 2008 and 2019 when specified. Most originated from the USA (11), then Canada (two), with one each from England, Indonesia, Germany and Norway. All had been peer-reviewed before publication.

Studies predominately used semi-structured interviewing for data collection; other approaches included survey/questionnaire design with open-ended questions and secondary analysis of qualitative data. The majority explicitly employed qualitative methodology with specified approaches, including phenomenology, ethnography and a narrative approach; most undertook thematic or content analysis.

Characteristics of participants

The included studies represented the experiences of 363 caregivers. We define caregivers as biological parents, foster parents, adoptive parents and grandparents. Where reported, conditions of children were static encephalopathy, congenital/chromosomal conditions, medical complexity, chronic respiratory disease, central hypoventilation, central nervous system progressive and ventilatory muscle weakness related to neuromuscular disease. Both invasive and noninvasive ventilation were reported. Only one study reported the experiences of a young person using invasive LTV and one person using noninvasive LTV [17]. The experiences of 143 healthcare professionals were also reported; their roles included paediatric pulmonologists, paediatric intensivists, neonatologists, nurses across specialities, social care workers, care coordinators and respiratory, speech and physical therapists.

Quality appraisal

Most studies were of medium to high quality. The aims and objectives of the studies were largely well articulated and supported by a clearly defined and appropriate qualitative methodology. Most studies offered findings that were considered in relation to existing research and discussed in terms of their implications for practice and policy. However, few studies described the theoretical underpinnings of their methodology. Likewise, few studies reflexively considered the relationship between the researcher(s) and study design and delivery.

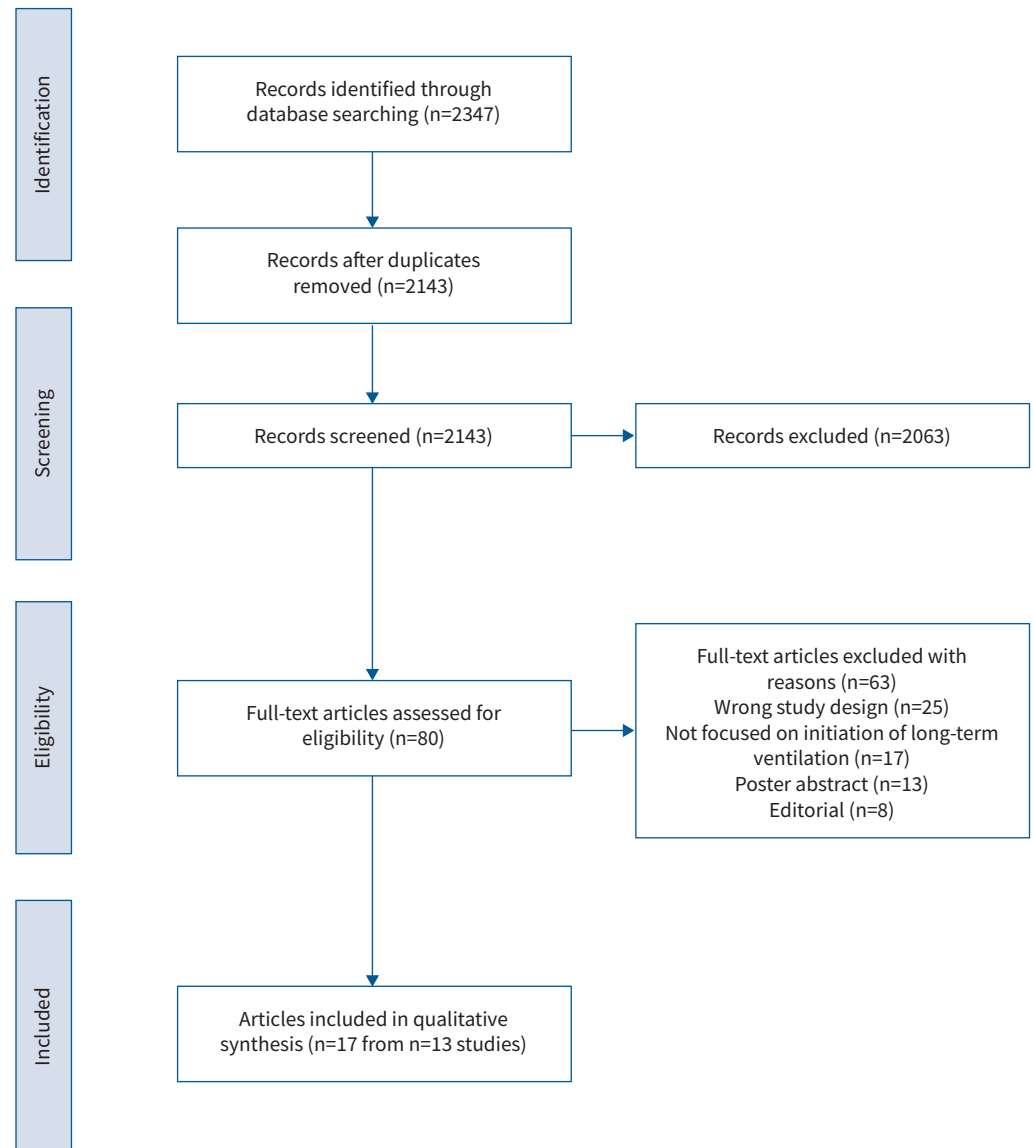


FIGURE 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart showing the inclusion of 17 articles (13 studies) from 2347 identified.

Results of thematic synthesis

Line-by-line coding resulted in 54 codes (examples in table 1). Codes were then grouped, offering eight descriptive themes reflective of the data. Further synthesis resulted in three analytical themes (table 1, figure 2) described below and illustrated with extracts from included studies.

Analytical theme 1: multiple voices; managing caregiver and healthcare professional roles in decision-making

Illustrated here is the variation in roles and voices that decide whether to pursue LTV for a child. Descriptive theme 1 portrays the potential value of multidisciplinary teams. Descriptive theme 2 describes the inherent challenges involved in managing the various perspectives of those involved in the decision and the potential for conflict or disagreement to arise from this engagement.

Value of multidisciplinary teams

Clinical teams supporting children requiring LTV are multidisciplinary and reflective of the medically complex care needs of the child. Managing multidisciplinary teams to coordinate input and information to caregivers relies on effective management of the clinical team. For example, collaborating with

TABLE 1 Relationship between codes, descriptive themes and analytical themes

Analytical themes	Descriptive themes	Codes
Multiple voices; managing caregiver and healthcare professional roles in decision-making	Value of multidisciplinary teams Variation in roles in decision-making	Effective coordination Palliative care involvement Value of nursing staff Balancing caregiver autonomy with clinical paternalism Effective coordination Joint decision All voices heard
Components facilitative of shared decision-making	Types of information caregivers would have valued Engagement with others with similar lived experiences Helpful qualities of clinician/caregiver interaction	“It’s that everyday living that wasn’t explained” Presented with all the facts Home nursing Long-term ventilation as lived Peer support and friendship Patience, honesty and understanding Using lay language and assessing understanding Continuity of clinician and dialogue
Caregiver positionality in the decision-making process	Primitive drives and faith Intense emotions and their impact on information processing Weight of the decision	Getting home Preserving life Religion and spirituality Shock and feeling overwhelmed Grief Loss of hope Making sense of information Is this the right thing to do? Best interests of the child Guilt Impact on family “Can I do this?”

sub-specialists including respiratory specialists, palliative care, and critical care to ensure caregivers received a range of perspectives to inform their decision. Likewise, caregivers benefited from care teams that actively promoted continuity of care [18, 19]. Caregivers particularly valued the inclusion of palliative care teams, regardless of whether LTV was pursued [19–21]. Palliative care is related to hospital teams as well as children’s hospice care. In relation to the former, palliative care offered caregivers and healthcare professionals a “different way of thinking” about their needs and care priorities [20]. Likewise, hospice care provided caregivers and their families a physical space and network of support when deciding whether to initiate LTV for their child:

“I got in touch with the children’s hospice and yes, that has always been like a life-saving oasis for us. And they also tremendously ... Well, they tremendously supported us in the madness.” (Quote from a caregiver participant [21])

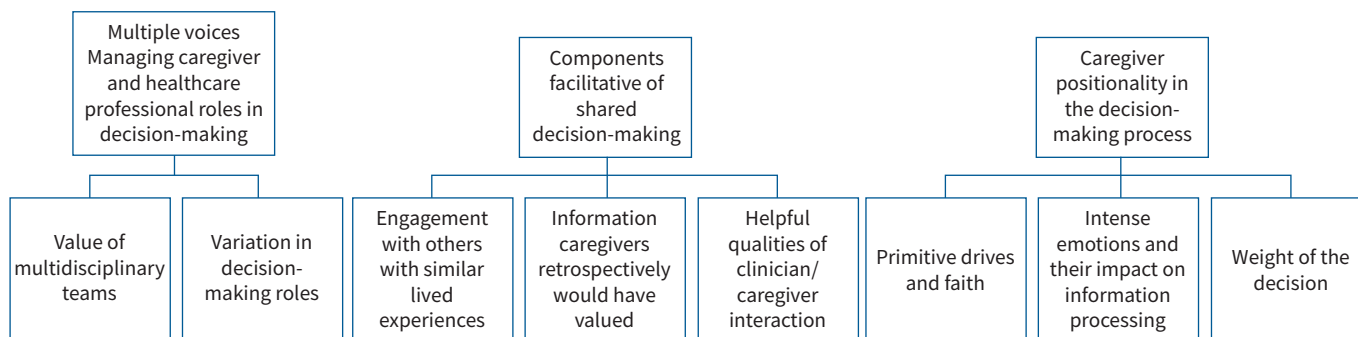


FIGURE 2 Thematic map.

Across specialisms, nursing roles were core to any multidisciplinary LTV team [7, 22]. The proximity of nurses to caregivers can result in a rich understanding of caregiver and child needs, allowing nurses to advocate for and support caregivers in making decisions [7]. Likewise, nurses can act as gatekeepers between the caregiver and other professionals, helping effective communication between both parties:

“... they [nurses] seem to be so invested in these families, and they get so close to them ... we have had very, very difficult decisions made by parents that were facilitated by the nurses. And again, it’s because the nurses just relate to them. They show the families that they’re in their corner...” (Healthcare professional participant [7])

In summary, the medical complexity of children requiring LTV means that the medical teams delivering their care must be multidisciplinary. If well managed, caregivers can benefit from the diverse expertise and perspectives of staff. Nurses were identified as being essential to the makeup of any multidisciplinary team.

Variation in decision-making roles

Most caregivers felt it essential to be active in LTV decision-making for their child, which for some meant leading decisions. These caregivers valued the expertise and guidance of healthcare professionals but ultimately felt they “knew their child best” and, therefore, “it was their decision to make” [7, 18, 20, 21, 23, 24]. Conversely, others wanted healthcare professionals to lead on decisions or to base their decision on the opinion of the healthcare professional(s) [24]. In some instances, caregivers described their role in decision-making as being suppressed or diminished because of the obstinate position of some healthcare professionals:

“The doctors had already made the decision to place the trach. All discussions and answers to questions were designed to convince me to agree with their decision.” (Caregiver participant [8])

Conflict or disagreement related to LTV decision-making between both caregivers and healthcare professionals and between caregivers and their own families exists [7, 17–20, 25]. Disagreements arose when parties involved in the decision disagreed on the pathway that was in the best interests of the child. Disagreements between caregivers and healthcare professionals could be exacerbated when caregivers felt healthcare professionals went beyond describing their child’s health and predicted outcomes to “telling us what you think we should do with that information” [18]. Conversely, examples of SDM were identified [17, 18, 20, 23]. For instance, in one study, over 50% of caregivers recounted a collective and shared decision [20]. SDM appeared to be based on components including relevant information sharing [8, 9, 17, 20], honest, clear and timely communication between parties [7, 17, 20, 23, 26], assessing caregiver understanding [24], and healthcare professionals guiding decisions made [18].

This theme illustrates the varied perspectives of individuals involved in LTV decisions. Where caregivers and healthcare professionals believe the decision is primarily theirs to make, there is the potential for conflict or disagreement to arise. Creating an environment where decisions are shared appears reliant on several factors, alluded to further in analytical theme 2.

Analytical theme 2: components facilitative of shared decision-making

Described here are the components facilitative of shared decision-making. Descriptive themes 1 and 2 highlight information and interaction beneficial to caregivers to support their decision-making. Descriptive theme 3 describes the qualities of caregiver-professional engagement that support caregivers in making a decision that is right for their child and themselves.

Information caregivers retrospectively would have valued

Caregivers wanted information about day-to-day life with a child on LTV “It’s that everyday living that wasn’t explained” [20]. Specifically, caregivers would have benefited from understanding the anticipated or potential impact on daily living such as sleep, supporting a functioning household, family life, travel, and finances [8, 9, 17, 20, 24]. Relatedly, caregivers would have also valued advice on home nursing and financial support [8, 17, 20, 27].

Regarding the delivery of information about LTV, some caregivers preferred to be presented with as much information as possible, regardless of whether the information was upsetting [17, 23]. However, where information was provided in a condensed fashion, such as over one or two conversations, caregivers reported feeling overwhelmed [7, 20]. Therefore, the provision of enough information for caregivers to feel

informed without overloading or overwhelming them can be a challenge, as conveyed by one healthcare professional:

“It’s really a Catch-22. If you don’t give them all the information then there’s a problem with that. ‘We weren’t informed’. If you do give them the information, then they feel pressure, like overwhelmed.” (Health and social care professional [7])

Engagement with others with similar lived experiences

Engaging with other caregivers who had made decisions about LTV was helpful, offering valuable insight into daily life with a child on LTV, including “tips and pointers” to support everyday living [7, 9, 19–22, 28]. Social media platforms were described as spaces to establish connections with others “who know what you are going through” [9, 28]. The benefits of supporting caregiver-to-caregiver engagement during decision-making were equally valued and recognised by healthcare professionals:

“...the most invaluable thing is actually letting the family talk to another family who already live with a child with similar technology and maybe has a similar medical condition, because I find they get sort of that real-life perspective from another family that we can’t provide.” (Healthcare professional participant [22])

Helpful qualities of clinician/caregiver interaction

Caregivers value honest, clear and timely dialogue with healthcare professionals about LTV. This means delivering information using understandable terms, tactful and sensitive language and tailoring communication to suit individual needs [17, 20, 21, 23, 26]. In addition, dialogue about LTV should be delivered in a timely manner, with ongoing opportunities to discuss questions and concerns with a healthcare professional acting on behalf of the team supporting the child, young person, caregiver and family throughout the process:

“It was really good to have a continuity doctor to go through it all. Every step of the way, she was with us. It made talking about it, it made accepting things, easier.” (Caregiver participant [20])

In summary, caregivers can be supported to make decisions about LTV through engagement with others who have faced similar circumstances. Healthcare professionals should engage with caregivers in an honest, clear and timely fashion, maintaining focus on the individual’s needs and the preferences of the child and their caregiver(s).

Analytical theme 3: caregiver positionality in the decision-making process

Described here are the unique positions of the caregivers of children requiring LTV. Three descriptive themes relay the instincts, emotions and pressures that caregivers can face when deciding whether to initiate LTV for their child.

Primitive instincts and faith

Decisions made by caregivers about LTV were understandably influenced by fundamental instincts related to bringing their child home and preserving life [7, 9, 17, 20, 23, 29]. The wish to remove their child from a clinical setting to a home environment was described as dominating decisions and therefore clouding other factors such as the reality of homelife with a child on LTV:

“I was very excited at the idea of just getting home. I just wanted to do whatever it took to get home.” (Caregiver participant [20])

The primal instinct of preserving their child’s life often dominated discourse and decisions on LTV [7, 9, 17]. To this degree, LTV was sometimes pursued as the only option to preserve life “It’s my baby girl, and I’m going to do whatever to keep her alive” (caregiver participant [17]). Some caregivers recounted never considering the possibility of not proceeding with LTV, regardless of future prognosis or the impact on their own quality of life [7, 9, 17, 20].

Religion, spirituality and faith also played significant roles in decisions [8, 17, 30, 31]. For some caregivers, not pursuing LTV for their child was akin to an act of “sin”, with a fear of “going to hell” [30, 31].

Likewise, caregivers described it as being “God’s choice how long (child) is going to live” [17]; therefore, they pursued any medical intervention that would prolong their child’s life. Religion and faith also offered some caregivers a sense of hope that extended to a belief in a miracle occurring, even when their child was in a severe or terminal condition [30]. However, there were few examples of healthcare professionals engaging with caregivers about their spiritual or religious beliefs. Reasons included a sense that healthcare professionals did not appear spiritual to caregivers, therefore deterring engagement on the topic or that discussing religion/spirituality was not the role of healthcare professionals [30].

Intense emotions and their impact on making sense of information

Caregivers experienced intense emotions regarding deciding about LTV for their child. On being confronted with the possibility of LTV, caregivers described a sense of shock and fear, particularly when breathing problems were encountered at birth [8, 17, 20, 28]. Being introduced to LTV also signalled a loss of hope related to the realisation that their child’s respiratory condition would be long-term [20, 21]. In turn, caregivers also experienced grief at the prospect of LTV:

“Do you know when somebody goes through the seven stages when someone dies? I probably went through all of those with the decision to get a trach.” (Caregiver participant [20]).

Caregivers described heightened feelings of fear and confusion when healthcare professionals used over-prescriptive and intimidating language [7, 18, 19]. Unhelpful dialogue can hinder caregivers’ ability to process the information provided [17, 19, 20]. Healthcare professionals alluded to some caregivers being unwilling to accept “bad news”, presenting a barrier to supporting informed and cooperative decisions about LTV [7, 17, 20]. Despite some caregivers’ wish for more emotional support [20, 21], peer-to-peer support was the only evidence of this.

Weight of the decision

The clinical circumstances of children requiring LTV meant caregivers often had to be advocates for their child, playing a significant role in deciding whether initiating LTV was in the child’s best interests. At the same time, some caregivers described the importance of being “rational” advocates [7, 17]. However, in reality, most caregivers had to grapple with their own needs and anxieties while attempting to promote their child’s best interests when making the decision [7–9, 17, 20, 31, 32].

“Can I do this? Can I do this as his full-time caregiver? It’s a medically fragile infant. Will I be able to handle everything and keep him from getting sick, keep him healthy, keep everything in line?” (Caregiver participant [9])

Prognostic uncertainty confounded the weight of the decision on caregivers. Evidence of caregivers being informed about long-term prognosis was limited [24]. Caregivers described receiving limited information about long-term prognosis and the experience of LTV [20]. Where these conversations did take place, healthcare professionals expressed considerable uncertainty about a child’s future with LTV [7, 18, 23]. Consequently, caregivers often had to make decisions with little certainty of the impact it would have, both for their child and themselves. Creating an environment free of judgement and unwanted opinion can help alleviate a degree of the burden felt by caregivers:

“I felt no judgment passed. [The healthcare professionals] were like, ‘There’s no right answer. It’s a toughest place to be. There’s no right answer. There’s no wrong answer. It’s what feels right to each family’.” (Caregiver participant [19])

Being the caregiver of a child requiring LTV is a unique and challenging position to fulfil. Caregivers must balance their own needs alongside advocating for their child often based on an uncertain prognosis. Religion and faith can be salient in decisions made.

Discussion

This systematic review presents a synthesis of qualitative evidence to clarify what SDM constitutes in relation to LTV initiation for children and young people (<21 years). Our findings highlight the potential challenges and considerations in attempting to implement SDM in discussions about LTV. These include acknowledging the unique positionality of caregivers and managing the multiple voices that require input into decisions made. This review has identified components facilitative of shared decision-making, including ensuring caregivers and their families feel informed about both the short- and long-term

implications of LTV and supporting engagement with others with similar lived experiences where appropriate. In addition, helpful qualities of healthcare professional and caregiver engagement have been identified and include honest, clear and timely dialogue, with information delivered using lay, tactful and sensitive language. The findings of this review are discussed in relation to existing evidence and their implications for practice and policy.

A decision to offer a child LTV is rarely straightforward. While in well-characterised populations (*e.g.* chronic lung disease of prematurity), there is substantial evidence to support LTV, in other conditions, such as neurodegenerative disease, the ability to predict future prognosis can be challenging and complex [3]. This review identified caregiver perspectives of children with a range of conditions, including static encephalopathy and congenital/chromosomal disorders, highlighting the prognostic uncertainty in which some LTV decisions must be made [7, 18, 23]. Dealing with uncertainty regarding the prognosis of children on LTV is well recognised in decision-making frameworks and guidance [33]. Therefore, it is recommended that decision-making is viewed as a process as opposed to a one-off event, with continued open and coordinated dialogue between all parties [10, 33]. The findings of this review support the recommendation, with evidence that caregivers strongly value honest, straightforward and timely dialogue delivered using lay terminology and tactful and sensitive language [17, 19–21, 26].

Supporting patients and their caregivers to feel informed about treatment risks, benefits and possible consequences is core to an SDM approach [34]. In the context of decisions about LTV, patients and their caregivers must be informed, as best as possible, about treatment risks, benefits and consequences. In particular, information about the long-term impact of LTV, both on the patient and family, is of clear value in supporting informed decisions. It is recognised that presenting a complete picture of the future is challenging; nevertheless, quantitative data suggests that caregivers are not always aware of the impact of LTV on their lives [1]. Qualitative findings from this review support this evidence, with caregivers reporting a paucity of information about the consequences of LTV for everyday living including supporting a functioning household, family life, travel and finances [8, 9, 17, 20, 24]. Therefore, including a social worker in multidisciplinary teams is likely to be of value in supporting families to consider and be informed about the potential long-term impacts of LTV.

Regarding the delivery of information about LTV to patients, caregivers and their families, this review identified the benefit of a recognised healthcare professional who acts as a gatekeeper between caregivers and the multidisciplinary team involved in the child's care [8]. Specifically, caregivers benefit from the input and expertise of healthcare professionals across various specialities, but in a coordinated and not overwhelming manner. Nurses were identified as particularly vital in supporting caregivers to process and make sense of information while also acting as advocates for the needs of the child and family [7, 22]. The review further identified the importance of drawing on expertise from outside clinical teams to support caregivers in their decision-making. For example, palliative care provided by children's hospices can provide valuable sources of support and refuge. Likewise, engaging with others with direct lived experience of similar decisions was found to be beneficial. As such, the review findings support good practice recommendations regarding the provision of opportunities for interaction with families with children already on LTV, which can offer caregivers a detailed account of what life with a child on LTV may be like [3, 10]. Nevertheless, it is essential to recognise the uniqueness of individual children, as such, engagement with caregivers of children with different comorbidities may prove counterproductive [3].

The complexity, gravity and emotional weight attributed to decisions about LTV means some degree of disagreement is not to be unexpected [1]. This review found evidence of disagreement both between caregivers and healthcare professionals and caregivers and their families [7, 17–20, 25]. Disagreements between caregivers highlight the emotional impact such decisions can have on families; therefore, caregivers must be provided with the necessary emotional and social support when making such decisions. Quantitative studies have identified healthcare professional–caregiver disagreement about the appropriateness of LTV [1]. Methods of resolution recommended include seeking a second opinion and the involvement of local hospital ethics advisory groups or committees [1, 3]; although little evidence of the effectiveness of such methods was found in the identified papers. Identifying approaches and strategies to support conflict resolution is essential, particularly in light of recent high-profile cases in the UK whereby disputes have needed to be resolved by the courts [35]. Future studies should focus on this area of dispute resolution as it is clearly in the best interests of all parties that such disputes are appropriately resolved.

In the past decade, there has been a 2.5-fold increase in the number of children receiving LTV in the UK [12]. As both the prevalence and survival of children with life-limiting conditions increase [12], decisions about whether to initiate LTV will more frequently need to be made by patients, their caregivers and healthcare

professionals. While the principles of SDM should be applied in decisions related to treatment for children with life-limiting conditions more broadly. Advancing understanding of SDM specifically related to LTV initiation is particularly significant because of the weight of these decisions on all involved parties and the prognostic uncertainty often surrounding such decisions. High-quality empirical evidence is, therefore, vital to support the continued development of practical recommendations and policies to support parties in reaching decisions that are in the best interests of the child. Notably, of the 17 papers identified, only one was from the UK [28]. Addressing this gap in UK-specific evidence is essential, particularly considering the unique national legal framework (see [33]).

Strengths and limitations

To the best knowledge of the authors, this is the first systematic review and qualitative evidence synthesis to explore experiences of decision-making from the perspectives of healthcare professionals and caregivers. In addition, this review includes the experiences of 363 caregivers of children with a diverse range of conditions and 143 healthcare professionals ranging from ear, nose and throat specialists to paediatric pulmonologists. Furthermore, most of the data reported in the included studies was collected in the last 10 years (see supplementary material). We hope this evidence synthesis offers a valuable contribution to practice, supporting recent recommendations and frameworks for ethical decision-making in LTV [1, 3, 10, 33].

A limitation of the included studies was the lack of representation of children and young people in the study sample. For example, only one study reported the experiences of a young person using invasive LTV and one person using noninvasive LTV [17]. This situation is likely due to the characteristics of the population; nevertheless, future studies may wish to specifically explore the directly reported experiences of children and young people where feasible. Further, regarding the samples of included studies, there was an over-representation of caregivers and healthcare professionals from the US. The lack of representation from other countries must be acknowledged in drawing conclusions from the review findings. Nevertheless, it is important to highlight the synergy between the review findings and the principles of SDM advocated by UK expert-informed guidelines [10]. Nevertheless, there is clear gap in evidence from other continents regarding LTV decision-making practices that needs to be addressed.

Inconsistent indexing across databases led to complexities in locating all relevant studies. To mitigate against this, manual searching of referencing lists and searches on Google Scholar allowed for the identification of 17 papers from 13 studies.

Points for clinical practice

Implementing SDM practice in child LTV is challenging. Ensuring caregivers feel informed, acknowledging their unique position and using honest, clear and timely dialogue can support SDM.

Conclusion

It is vital that empirical evidence is gathered to substantiate and support the development of SDM frameworks and guidance for LTV in children (*e.g.* [1, 10, 33]). This systematic review offers a synthesis of current practice, understanding and lived experience of LTV decision-making based on empirical qualitative evidence. Our ambition is that it might help ensure that LTV decisions incorporate and acknowledge the perspectives of all relevant stakeholders. Using the existing literature, we have clarified the components that can support SDM, such as the crucial gatekeeper role and factors, such as the positionality of caregivers, that need to be considered to achieve it in practice. As such, we hope this review offers a valuable resource for practitioners and policymakers to consider when implementing SDM practices in the context of LTV decisions for children and young people.

Provenance: Submitted article, peer reviewed.

Author contributions: All authors conceived the study and G. Peat developed the protocol with regular input from S.-A. Delaney, F. Gibson, J. Brierley and L.K. Fraser. G. Peat and S.-A. Delaney undertook the electronic searches, screening, data extraction and quality appraisal. G. Peat conducted the thematic synthesis with regular input from S.-A. Delaney. G. Peat drafted the manuscript with regular input from S.-A. Delaney, F. Gibson, J. Brierley and L.K. Fraser.

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References

- 1 National Confidential Enquiry into Patient Outcome and Death (NCEPOD). Balancing the pressures. London, NCEPOD, 2020. www.ncepod.org.uk/2020ltv/LTV_Full_Report.pdf
- 2 Robinson RO. Ventilator dependency in the United Kingdom. *Arch Dis Child* 1990; 65: 1235–1236.
- 3 Brookes I. Long-term ventilation in children. *Paediatr Child Health* 2019; 29: 167–171.
- 4 Fine-Goulden MR, Ray S, Brierley J. Decision making in long-term ventilation for children. *Lancet Respir Med* 2015; 3: 745–746.
- 5 Department of Health and Social Care. The NHS Constitution for England. Date last updated: 1 January 2021. Date last accessed: 5 December 2022. www.gov.uk/government/publications/the-nhs-constitution-for-england/the-nhs-constitution-for-england#principles-that-guide-the-nhs
- 6 Légaré F, Adekpedjou R, Stacey D, et al. Interventions for increasing the use of shared decision making by healthcare professionals. *Cochrane Database Syst Rev* 2018; 7: CD006732.
- 7 Gower WA, Golden SL, King NMP, et al. Decision-making about tracheostomy for children with medical complexity: caregiver and health care provider perspectives. *Acad Pediatr* 2020; 20: 1094–1100.
- 8 October TW, Jones AH, Greenlick Michals H, et al. Parental conflict, regret, and short-term impact on quality of life in tracheostomy decision-making. *Pediatr Crit Care Med* 2020; 21: 136–142.
- 9 Henderson CM, Raisanen JC, Shipman KJ, et al. Life with pediatric home ventilation: expectations versus experience. *Pediatr Pulmonol* 2021; 56: 3366–3373.
- 10 Ray S, Brierley J, Bush A, et al. Towards developing an ethical framework for decision making in long-term ventilation in children. *Arch Dis Child* 2018; 103: 1080.
- 11 NHS England. Paediatric long term ventilation reporting specification. Date last updated: 1 May 2019. www.england.nhs.uk/publication/paediatric-long-term-ventilation-reporting-specification-technical-detail-specific-data-requirements/
- 12 Barker N, Sinha A, Jesson C, et al. Changes in UK paediatric long-term ventilation practice over 10 years. *Arch Dis Child* 2022; 108: 218–224.
- 13 Tong A, Flemming K, McInnes E, et al. Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Med Res Methodol* 2012; 12: 181.
- 14 Long HA, French DP, Brooks JM. Optimising the value of the critical appraisal skills programme (CASP) tool for quality appraisal in qualitative evidence synthesis. *Res Methods Med Health Sci* 2020; 1: 31–42.
- 15 Dixon-Woods M, Sutton A, Shaw R, et al. Appraising qualitative research for inclusion in systematic reviews: a quantitative and qualitative comparison of three methods. *J Health Serv Res Policy* 2007; 12: 42–47.
- 16 Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Med Res Methodol* 2008; 8: 45.
- 17 Edwards JD, Panitch HB, Nelson JE, et al. Decisions for long-term ventilation for children. Perspectives of family members. *Ann Am Thorac Soc* 2020; 17: 72–80.
- 18 Bogetz JF, Munjapara V, Henderson CM, et al. Home mechanical ventilation for children with severe neurological impairment: parents' perspectives on clinician counselling. *Dev Med Child Neurol* 2022; 64: 840–846.
- 19 Jabre NA, Raisanen JC, Shipman KJ, et al. Parent perspectives on facilitating decision-making around pediatric home ventilation. *Pediatr Pulmonol* 2022; 57: 567–575.
- 20 Boss RD, Henderson CM, Raisanen JC, et al. Family experiences deciding for and against pediatric home ventilation. *J Pediatr* 2021; 229: 223–231.
- 21 Pechmann A, Langer T, Kirschner J. Parents' perspectives on diagnosis and decision-making regarding ventilator support in children with SMA Type 1. *Neuropediatrics* 2022; 53: 122–128.
- 22 Edwards JD, Morris MC, Nelson JE, et al. Decisions around long-term ventilation for children. Perspectives of directors of pediatric home ventilation programs. *Ann Am Thorac Soc* 2017; 14: 1539–1547.
- 23 Mah JK, Thannhauser JE, McNeil DA, et al. Being the lifeline: the parent experience of caring for a child with neuromuscular disease on home mechanical ventilation. *Neuromuscul Disord* 2008; 18: 983–988.
- 24 Smith MA, Clayman ML, Frader J, et al. A descriptive study of decision-making conversations during pediatric intensive care unit family conferences. *J Palliat Med* 2018; 21: 1290–1299.

- 25 23rd Annual Conference of the International Society for Quality of Life Research. *Qual Life Res* 2016; 25: Suppl. 1, 1–196.
- 26 Ferguson E, Wright M, Carter T, *et al.* Communication regarding breathing support options for youth with Duchenne muscular dystrophy. *Paediatr Child Health* 2011; 16: 395–398.
- 27 Henderson CM, Wilfond BS, Boss RD. Bringing social context into the conversation about pediatric long-term ventilation. *Hospital Pediatr* 2018; 8: 102–108.
- 28 Flynn A, Whittaker K, Donne AJ, *et al.* Holding their own and being resilient: narratives of parents over the first 12 months of their child having tracheostomy. *Compr Child Adolesc Nurs* 2021; 44: 255–265.
- 29 Nageswaran S, Golden SL, Gower WA, *et al.* Caregiver perceptions about their decision to pursue tracheostomy for children with medical complexity. *J Pediatr* 2018; 203: 354–360.e1.
- 30 Nageswaran S, Banks Q, Golden SL, *et al.* The role of religion and spirituality in caregiver decision-making about tracheostomy for children with medical complexity. *J Health Care Chaplain* 2022; 28: 95–107.
- 31 Nurnaningsih N, Setiyarini S, Al Mirzanah S, *et al.* Forgoing life sustaining treatment decision-making in critically ill children: parental views and factor's influence. *Clin Ethics* 2021; 16: 246–251.
- 32 Dybwik K, Nielsen EW, Brinchmann BS. Ethical challenges in home mechanical ventilation: a secondary analysis. *Nurs Ethics* 2012; 19: 233–244.
- 33 Larcher V, Craig F, Bhogal K, *et al.* Making decisions to limit treatment in life-limiting and life-threatening conditions in children: a framework for practice. *Arch Dis Child* 2015; 100: s3–s23.
- 34 National Institute for Health and Care Excellence. Shared decision making. Date last updated 17 June 2021. Date last accessed: 5 December 2022. www.nice.org.uk/guidance/ng197
- 35 Turner EJ. The growing complexities with long term ventilated children: a personal perspective. *Med Leg J* 2023; 91: 3–6.