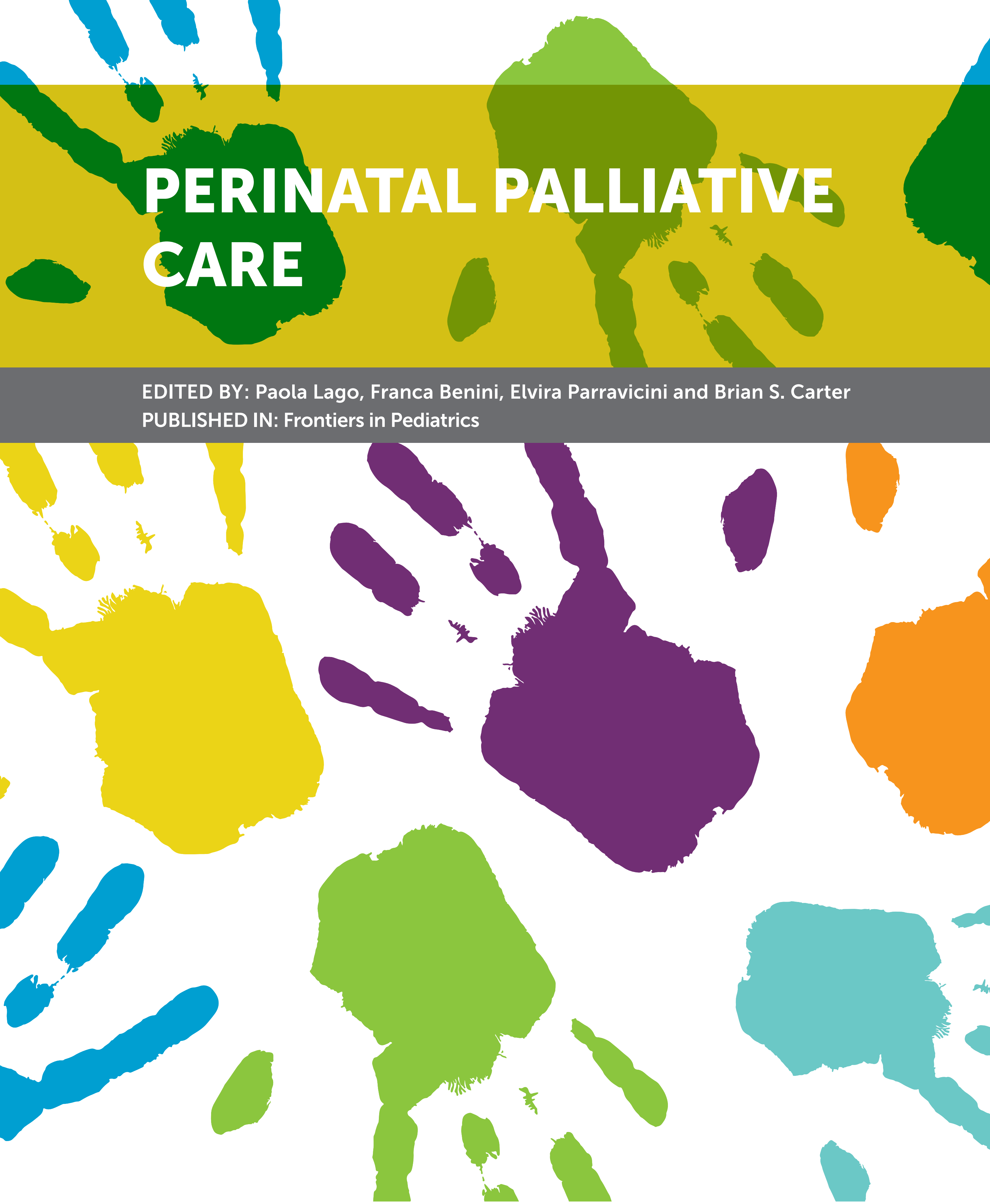

PERINATAL PALLIATIVE CARE

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PERINATAL PALLIATIVE CARE

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EDITORIAL
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Editorial: Perinatal Palliative Care Comes of Age

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Editorial on the Research Topic

Perinatal Palliative Care Comes of Age

Advances in medical technology have allowed for the prenatal detection of anatomical congenital anomalies leading to well-recognized patterns of concern that may involve any organ system. They may be due to chromosomal or other genetic differences. Or they may result of altered maternal-placental-fetal metabolism, immune function, or environmental exposure to teratogens. The affected fetus, and newborn, is said to have a life-limiting condition and postnatal life may be brief. Even with resuscitation and admission to a neonatal ICU (NICU), hospitalization may be prolonged only to lead to technology dependence, and adverse prognoses.

Perinatal palliative care (PnPC) has grown out of the field of hospice and palliative medicine (HPM) and increasingly available services of pediatric HPM. The goal of PnPC is to address and support the family living with a concerning, life-limiting, or life-threatening fetal diagnosis. Their pregnancy narrative has been disrupted—their story, in a way, has been broken. PnPC offers a comprehensive interdisciplinary approach for the care of the parents during their continued pregnancy, to help them redirect their attention to deal with the news that they have received and consider the story that they can best live with moving forward for their newborn and themselves. For each family, and unique newborn, care is directed toward achieving a state of comfort by satisfaction of the newborn's basic needs, pain and symptom management, and supportive psychosocial and spiritual care for family members across generations in a culturally sensitive framework. PnPC also provides support for professionals.

While PnPC addresses the perinatal journey that starts prenatally with a concerning fetal diagnosis, it continues at birth and may accommodate resuscitative measures to smooth neonatal transition and optimize time with a family while the baby is alive—even when invasive interventions and a lengthy NICU stay may not be the elected treatment paradigm. PnPC provides accompaniment for the baby and his/her family as an inpatient and may continue into the post-discharge outpatient timeframe. It also may be an option that offers a plan of care directed at improving the baby's quality of life when cure-oriented and life-prolonging treatment is no longer the goal of care for a baby who has been treated in the NICU.

The interdisciplinary approach that PnPC brings is one of collaboration across professional disciplines, including high-risk obstetricians, neonatologists, and other specialists, in accordance with each specific diagnosis. Services to help provide comfort to the baby (lactation consultant, feeding therapists, speech pathologist, OT/PT) and emotional, psychological, and spiritual support to families (Social Work, Psychology, Child Life, and Chaplaincy or engaging the family's faith community) are summoned to help. Yet, while there are over 300 programs of PnPC reported internationally, there is no current standard of PnPC team composition or service provision for the fragile population it addresses.

Here we provide 13 peer-reviewed manuscripts aimed at addressing much of what PnPC is, offers, and considers, its interdisciplinary team nature, and the need for ongoing study and development.

Beginning with a concerning fetal diagnosis, Marc-Aurele describes for us in her article, the Decisions Parents Make When Faced with Potentially Life-Limiting Fetal Diagnoses and the Importance of Perinatal Palliative Care how families consider gaining additional diagnostic information and the value of additional pediatric specialty consultation to inform parents of what to expect as they continue a pregnancy. She introduces birth planning, which is further elaborated upon by Cortezzo et al. who expound on the conversations that precede and follow the development of any birth plan, comparing it to other forms of Advance Directives. In Perinatal Palliative Care Birth Planning as Advance Care Planning Cortezzo relates how birth planning allows families to express their fears, values, hopes, and wishes.

Symptom management follows as Cortezzo and Meyer review of caring for newborns and their end-of-life symptoms in Neonatal End-of-Life Symptom Management. This subject is also addressed in a separate chapter, End-of-Life Care for Neonates: Assessing and Addressing Pain and Distressing Symptoms by Haug et al. Importantly, the paucity of rigorous evidence-based research on therapeutics is noted, and the PnPC clinician may be at his best when he adopts practices from other populations while remaining considerate of the constraints and context of the neonate's physiology. Kain RN addresses the importance of various cultural and spiritual considerations that caregivers should understand in their approach to patients and families of varied faith traditions. Perinatal Palliative Care: Cultural, Spiritual, and Religious Considerations for Parents—What Clinicians Need to Know is a useful summation of many, though understandably not all, such traditions.

An excellent model of caring for fragile newborns with anticipated short lives follows in the perspective piece by Wool and Parravicini. In The Neonatal Comfort Care Program: Origin and Growth Over 10 Years they relate the development of the program at Columbia University in New York and how it has grown to serve countless newborns and families in sharing meaningful, humane, and comfortable time together—even when life for the newborn is brief. Another model of PnPC is reported in a perspective piece by Locatelli et al. from Bologna, Italy. "Percoso Giacomo": An Italian Service of Perinatal Palliative Care. Named for a palliative pathway taken by the family of little Giacomo, this chapter provides an excellent review of key pieces in making PnPC functional.

For many around the world, it will be important to understand how to develop a perinatal palliative care program. While this is understandably determined in some manner by local practices in obstetrics and neonatal care, as well as resources, a nice review of essential mindsets, materials and models should be welcome by all. Lago et al. present such material in Summary of the Key Concepts on How to Develop a Perinatal Palliative Program. This theme is further given attention with the presentation of a specific model of development by Bolognani et al. from Italy in their brief report, Development of a Perinatal Palliative Care Model at a Level II Perinatal Center Supported by a Pediatric Palliative Care Network.

The potential development of moral distress in professional staff is very real within the neonatal ICU caregivers. Mills and Cortezzo provide a superlative paper that any physician or nurse would benefit from reading entitled, Moral Distress in the Neonatal Intensive Care Unit: What Is It, Why It Happens, and How We Can Address It. Additional address of these and broader measures of caring for the caregiver are found in an excellent scoping review by advance practice nurse Grauerholz et al. Fostering Vicarious Resilience for Perinatal Palliative Care Professionals.

No address of PnPC would be complete without addressing barriers to palliative care. Benini et al. provide key insights to such barriers in the perinatal context in their opinion paper, Barriers to Perinatal Palliative Care.

As PnPC programs develop, work through, and overcome barriers, and experience success, it is important to be able to evaluate their impact and, in many ways, "prove" their value to healthcare organizations, hospitals, and health care professionals. Jennifer et al. present an evaluation of the impact of a 3-day training course for interdisciplinary clinicians. Assessment of Healthcare Professionals' Self-Perceived Competence in Perinatal/Neonatal Palliative Care After a 3-Day Training Course. It is important that any future training events be similarly evaluated to assess participant confidence, competence, and best practices.

We sincerely hope that this collection of works from around the world will help give confidence to those needing it, direction for those on the journey of developing PnPC programs and understanding to all.

AUTHOR CONTRIBUTIONS

BC, EP, FB, and PL conceived the paper. BC created the first draft. All authors contributed to edits and accepted the final copy of the manuscript.

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REVIEW
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Perinatal Palliative Care Birth Planning as Advance Care Planning

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Purpose of Review: A significant number of pregnancies are complicated by a fetus with a life-limiting diagnosis. As diagnoses are made earlier in the pregnancy, families experience anticipatory grief and are faced with navigating goals of care for a baby that has yet to be born. With the support of the care team, families can begin to grieve, plan, and make meaningful memories during the duration of the pregnancy, the birth of their baby, and life of the child. Creating a palliative care birth plan, which expands beyond the traditional concept for delivery planning to include prenatal, perinatal, and neonatal care has become an important method for parents to process the diagnosis, for parents to document their wishes, and for members of the care team to communicate with the goal of supporting and enhancing the experience of the family. This articles reviews recent and relevant literature on the importance of birth planning and the role of perinatal palliative care when a life-limiting fetal diagnosis is made.

Recent Findings: The process of birth planning is an important component of perinatal palliative care. Through this process, families can express their fears, values, hopes, and wishes. It also offers an opportunity for providers to communicate these wishes for the remainder of the pregnancy, the delivery, birth, and time afterwards. This has been demonstrated to decrease maternal stress and promote family centered care.

Summary: Perinatal birth planning is an important component of perinatal palliative care when a fetus has a life-limiting diagnosis. The process of birth planning can be supportive and therapeutic as well as an important communication tool. With multiple practices and designs of perinatal palliative care programs, there are no standard tools even though important components have been identified. Ultimately, the strategies outlined here can be used as advance care planning tools.

Keywords: perinatal palliative care, birth plan, advance care planning, neonatal, life-limiting diagnosis

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INTRODUCTION

Advances in prenatal care and diagnostic technologies have allowed for earlier and more frequent identification of life-limiting fetal diagnoses, leading to increased numbers of families seeking support in decision-making surrounding the pregnancy, delivery, and neonatal care of medically complex newborns (1–3). Perinatal palliative care (PnPC) consultation as a part of family-centered fetal care offers opportunities for exploration of goals of care and planning for value-driven medical care for the remainder of the pregnancy, during delivery, and in the neonatal period (4–8). While practices vary and some PnPC teams investigate a spectrum of options that includes interruption of pregnancy, most PnPC consultation occur once a family decides to continue a pregnancy with a potentially life-limiting fetal diagnosis. PnPC teams ensure that families demonstrate sound medical understanding and prognostic awareness. They aid in exploration of unique value-systems from which families may choose various pathways including expectant pregnancy management, close monitoring of the pregnancy, interventions during labor and delivery, comfort-focused newborn care, or subspecialty neonatal evaluation and medical interventions. PnPC providers advocate for clear communication and support each family in vocalizing goals of care and wishes for their child's life (9).

Creation of a palliative care birth plan is an important method of exploring and documenting requests for the remainder of the pregnancy, labor and delivery, and the subsequent care of a newborn with a life-limiting diagnosis (10–12). The process of palliative care birth planning is both functional and therapeutic for families navigating goals of care discussions. Ultimately a care plan, the culmination of the important birth planning process, is developed to document and communicate the family's wishes with the multidisciplinary medical team. In this way, the palliative care birth plan expands beyond the traditional concept of a birth plan, which communicates preferences for the logistics surrounding delivery, to include prenatal, perinatal, and neonatal care. The PnPC birth plan can give families a sense of control in a challenging time and an opportunity to honor the life of their child. This article reviews recent literature on PnPC birth planning in the setting of a life-limiting fetal diagnosis and provides recommendations on developing documents that can be used as effective communication tools in advance care planning.

POTENTIALLY LIFE-LIMITING FETAL DIAGNOSES

In the United States alone, there are over 1 million fetal deaths and over 15,000 neonatal deaths each year (13). The majority of these, with up to 3% of pregnancies complicated by a potentially life-limiting fetal diagnosis, are the result of congenital malformations or chromosomal anomalies (14–16). Expanded early genetic screening or diagnostic options and advances in imaging technology have resulted in earlier and more frequent identification of life-limiting fetal diagnoses. A life-limiting condition is one having a low likelihood of long-term survival without severe morbidity that could impact quality of life. Multidisciplinary fetal care centers have expanded to include perinatal palliative care specialists who assist families facing complicated pregnancies in exploring options for care of the fetus and newborn (1, 3, 17). Some fetuses with life-limiting diagnoses will die *in utero*. Others may die during labor and delivery or in the immediate neonatal period. Others still may survive with varying degrees of interventions for hours, days, or more (1, 10, 18). Within these diagnoses are a wide range of pathology with a high lethal potential where individuals are expected to have varying degrees of survival and life spans. **Table 1** shows a list of common potentially life-limiting fetal diagnoses that perinatal palliative care teams are commonly consulted for.

TABLE 1 | Examples of common potentially life-limiting fetal diagnoses.

Genetic/Metabolic
- Trisomy 13
- Trisomy 18
- Osteogenesis imperfect (severe phenotypes)
- Triploidy
- CPT2 deficiency

Cardiac
- Acardia
- Inoperable lesions
- Lesions with poor prognosis despite palliative surgery options

Central nervous system
- Anencephaly
- Alobar holoprosencephaly
- Pontocerebellar hypoplasia
- Hydrancephaly
- Giant encephaloceles
- Spinal muscular atrophy

Severe pulmonary hypoplasia
- Bilateral renal agenesis with anhydramnios (Potter's sequence)
- Thanatophoric dysplasia and other severe skeletal dysplasias
- Short rib polydactyly syndrome
- Congential diaphragmatic hernia with low O:E lung volumes
- Severe hydrops fetalis

Other
- Severe pentalogy of cantrell
- Limb-body wall complex
- Severe forms of conjoined twins

With opportunities for multidisciplinary evaluation and increased access to perinatal palliative care, more women, between 20 and 40%, are choosing to continue pregnancies with a fetal life-limiting diagnosis (11, 19). Of those choosing to continue the pregnancy, reports of live births range from 60 to 82% (11, 20–23). When a child survives into the neonatal period, families face many decisions surrounding treatment and approaches to care. Some of these options may alter the disease trajectory but are unable to correct the underlying physiology or significantly alter the prognosis of the condition for the child. Earlier and enhanced fetal diagnosis gives families time to receive multidisciplinary fetal care with expert explanation of the suspected or confirmed diagnosis, support in understanding the prognosis and accepting levels of uncertainty, and value-driven shared decision-making in determining appropriate care or treatment options. With this information and support, families can utilize the ongoing prenatal period to create and share hopes, requests, and goals for the remainder of the pregnancy and their child's care after delivery, enhancing opportunities to parent in the face of a life-limiting diagnosis.

Abbreviations: PnPC: Perinatal Palliative Care.

The response to a life-limiting fetal diagnosis is unique to each family and their pregnancy journey. Some families seek comfort-focused medical care for the remainder of the pregnancy and for their child while others seek support in exploring or requesting varying degrees of life-sustaining interventions during the pregnancy and after birth. In a recent evaluation of parental experiences in the face of a life-limiting prenatal diagnosis, 52% of families reported creating birth plans that focused on comfort-care for the remainder of the pregnancy, delivery, and neonatal care, 27% requested some degree of intervention but placed limitations on more extreme levels of life support after delivery, and 25% requested full interventions and monitoring during the remainder of the pregnancy and life-sustaining therapies after delivery (24). Understanding the value system behind parental-decision making, building a supportive longitudinal care relationship, and guiding goals of care discussions are critical roles of the perinatal palliative care team in creating a comprehensive palliative care plan of care encompassing the remainder of prenatal care, perinatal care including preferences about delivery, and post-natal/neonatal care. The authors use the terms palliative care birth planning throughout the article to describe this processes and palliative care birth plan to describe the document created to communicate the wishes to the care team.

PARENTAL DECISION-MAKING EXPERIENCE WITH A PRENATALLY IDENTIFIED LIFE-LIMITING DIAGNOSIS

When a life-limiting diagnosis is made, many variables impact decisions regarding pregnancy continuation and subsequent choices for further care. These factors include maternal age and medical history, religious beliefs, cultural values, severity of anomalies, expected prognosis, gestation at diagnosis, and socioeconomic status (25–28). Additionally, many families consider the worth of their baby, potential quality of life, and personal ethics in the context of the known information (29–31). Several studies have shown the impact of religion on decisions for pregnancy continuation and decisions regarding care after delivery (32, 33). There are times this can lead to feelings of hopefulness, peace, or acceptance. Alternatively parents may experience fatalistic beliefs or fears related to decisions that may be viewed as deviating from their religious beliefs (28). Knowing these factors can help the care team better understand families' decision-making processes as well as help support them through this journey. There are times when personal values and cultural beliefs conflict and families can become lost, experiencing great emotions and turmoil at times leading to ambivalence in the difficult decisions they are asked to make (34).

A qualitative study that described if and how parents decide to proceed with a pregnancy when a diagnosis is made highlighted a three phase process families experience that includes information gathering, assessing implications of multiple uncertainties, and ultimately decision making (35). The manner in which information is presented, as well as parental perception of provider's views, affect the parents' experience and potentially informed decision making (17). During the information gathering phase, providers must be conscious of framing bias and the possibility for this to influence parental choices, as evidenced by the finding that families are more likely to seek interventions if outcomes are portrayed in terms of survival (32). Poor choice architecture and framing bias can be contradictory to the effort to provide non-directive counseling prenatally. Additionally terms such as "lethal," when used for life-limiting conditions, can convey a sense of finality to families, can leave them feeling hopeless, and may alter the decisions made for the remainder of the pregnancy and afterwards (36). Families have shared experiences of hearing insensitive or rushed remarks that cause lasting trauma (8, 37–39). For example, phrases such as "incompatible with life" were found to be insensitive and unhelpful by parents (40). Families take information in the context of their pre-existing views of disability, parenthood, certainty of the information received, their journey to pregnancy, and their support system when making subsequent decisions (41).

Studies of parental experiences show the importance of clear, concise communication at the time of diagnosis. Simultaneously, acknowledging uncertainty and reviewing further plans for testing or follow up in a timely manner is important (42). Families desire support, empathy, and acknowledgment of the difficult situation they are in and the complexity of decision making (43). Parents want information and support as they continue through their pregnancy journey (44). Often, multidisciplinary support and subspecialty care helps families feel that their questions are being addressed and they are receiving comprehensive care that ultimately aids in coping with the diagnosis (43, 45–47).

Unfortunately, many families feel that this information is poorly communicated. Reports indicate families feel discussions are framed based on providers' view of quality of life which may be vastly different from theirs. Families appreciate providers who offer comprehensive care and planning as well as encouragement to embrace the moments and milestones of their pregnancy (40). Providers must utilize reflexive practices so that members of the treatment team are aware of their own views and how information is portrayed during this vulnerable time (17). Families desire continued support while taking time to process the information and do not wish to feel pressured in to making delicate and important decisions. Parents also want providers to learn about them and their baby as individuals which creates the feeling of significance and that their child matters to the medical team (30). Families also appreciate continuity of care and consistency. Pregnant women and their support people are most comfortable when there is a dedicated provider or care team who takes the time to know them and is willing to explore the fears, questions, and uncertainty that lies ahead with them. Families appreciate learning about all support and treatment options from providers as opposed to seeking resources and information on their own (40, 48).

When the decision is made to continue the pregnancy, the parental experience evolves. Recent literature describes parents' experiences and emotional processing with continuing a pregnancy with a lethal fetal diagnosis (44, 49–53). While variation exists in how individuals cope and process information, an overarching theme is not wanting to have regrets for the remainder of the pregnancy and the decisions made. Parents experience a variety of emotions ranging from shock to existential crisis and ultimately life remodeling as they learn about the diagnosis, live with it, experience the birth and at times the death of their child. Across all of these phases, parents have a variety of coping strategies. As they begin to comprehend the implications of the diagnosis, parents seek information, and use that to revise goals for the pregnancy. Then parents may begin to visualize concrete plans for the remainder of the pregnancy, labor and delivery, the birth, and possibly the death of their child. While parents are preparing, they feel as if they are advocating for their baby, one of the most important responsibilities of parenting. During this time, in addition to the support from family, community, and social networks, families rely on the support of the medical providers (44, 50). Psychosocial support enhances their ability to process the information and be active in truly shared decision-making (51–53). As every individual processes information differently and requires different types of support, it is imperative that providers learn about their patient and listen to them so that the medical team can tailor the care to the unique needs of the family. It is likewise important that providers understand the stages and phases parents go through while processing the diagnosis so that they can recognize where their patient is along the journey and as a result provide the appropriate support and anticipate which conversations families are ready or prepared to engage in.

When a woman learns her fetus has a life-limiting diagnosis, she begins to grieve the loss of a baby she is already attached to (54, 55). Some have noted that receiving the diagnosis meets the criteria of a traumatic experience (17, 56). It is important to recognize that this is true for not just the pregnant woman but her partner, if she has one, and family as well. As a result support should be provided to all parties (57). Healthcare providers need to be equipped to provide psychosocial and bereavement support shortly after the diagnosis is made, through the remainder of the pregnancy and afterwards (58, 59). The support and experience the woman and her family receive has impact far beyond the current pregnancy (60). Initial reactions often include a complex dynamic of grief, guilt, hopelessness, and anger (17, 28, 34, 43, 61). At the same time, parents may maintain a feeling of joy and that their baby is a blessing (62). Families are facing having to make difficult and imperfect decisions that impact this current pregnancy and their relationships afterwards (17, 29, 62).

ROLE OF PERINATAL PALLIATIVE CARE-SUPPORTED ADVANCE CARE PLANNING IN ENHANCING OPPORTUNITIES TO PARENT

The importance of quality, collaborative perinatal palliative care is well-recognized (63–65). When parents choose to continue a pregnancy with a life-limiting diagnosis, they have the unique experience of simultaneously grieving the future for which they have hoped, embracing the milestones of the pregnancy, and planning for how they wish to spend the time they will have with their baby. Many will experience a loss of identity and a sense of isolation (39). Parents often seek the support of their medical providers who have familiarity with what is initially foreign and often overwhelming to them (66). Parents also seek opportunities to feel or regain a sense of control. By being able to make decisions about the remainder of the pregnancy and care of their baby, they are given an opportunity to actively parent and may feel a sense of empowerment (50, 66–68). As the pregnancy progresses, parents continue to bond with their baby. They seek respect for the child's life and shared celebration of the pregnancy milestones and mementos such as maternity pictures, heartbeat recordings, detailed ultrasound images, pregnancy journals, or intentional family activities or rituals. By providing guidance, exploring value systems and goals of care, and creating a space for questions and fears, PnPC teams help families feel supported as parents plan for the life and possibly death of their baby (66, 69, 70).

While discussing the birth and expected death of their child can be difficult for families, the process of creating a palliative care birth plan is also therapeutic and grants families a sense of control and confidence in sound communication of their needs, requests, and hopes (67). Exploration of goals of care, assistance in framing limitations of intervention, and advocacy for sound symptom management are best supported by palliative care trained providers. In the process of advance care planning, PnPC team members balance supportive listening and exploration of family value-systems with facilitation of discussion of goals and the translation of those goals in to medical care plans (10–12, 19, 71–73). The therapeutic relationship that develops between the family and providers leads to increased satisfaction and valued experiences during a difficult time. PnPC providers utilize the process of birth planning as a tool to explore meaningful support from the medical team and ideals the family visions for the remainder of the pregnancy and time with their child. When a family knows that their wishes are heard, it validates their parental role (67, 74). Families find joy in the opportunities to love, parent, and make memories with their baby even before the delivery (75). These experiences are chances to celebrate their child's life and to begin to form an enduring memory and legacy.

The experience of parenting and legacy making are included in the concept of the palliative care birth planning and can occur during the prenatal, perinatal and neonatal care phases. PnPC providers should facilitate exploration of how families seek opportunities to parent, make memories, and create a legacy as the PnPC team is well-equipped to anticipate or suggest what opportunities, choices, and options are available and may be important to the family (40). Photography, recordings, sibling involvement, creation of tangible keepsakes, bathing, and family-centered or faith-based rituals are commonly valued memory making opportunities during the remainder of the pregnancy and afterwards (76). Some families also seek to find meaning beyond their interaction with their babies and hope to create a lasting legacy for their child through organ/tissue donation, contributions to research or education, or breast milk donation. As advocates for family well-being, PnPC providers often balance planning for these valuable memory-making and life-honoring opportunities with careful watch for grief and complicated bereavement. This is especially true when carefully planned time with a baby after delivery or the opportunity to donate organs after death does not come to fruition as hoped.

EXPLORING GOALS OF CARE DURING PERINATAL PALLIATIVE CARE CONSULTATION

The perinatal palliative care birth planning process is ideally longitudinal and interdisciplinary. Families benefit from multiple points of contact with care teams. Many PnPC providers advance conversations incrementally through several visits, focusing initially on rapport building and ensuring appropriate medical understanding and prognostic awareness before transitioning to exploration of goals of care and birth planning (10). This allows for manageable amounts of information to be received by families and gives space for reflection and formation of questions. PnPC is interdisciplinary, utilizing care team members with specific training in psychosocial assessment and spirituality as well as communication and medical expertise to optimize a climate of shared decision-making (77). Parents appreciate receiving balanced information about the diagnosis and all treatment options before making decisions. They appreciate being part of the care discussions, having their voices heard, acknowledgment of their difficult situation, and ongoing support (30, 71). They rely on providers to encourage them to express their hopes, fears, and goals (78). Providers and parents collaborate to make decisions and plans, balancing autonomy and parental authority with medical recommendations and prognosis (79).

Discussing goals of care is a key communication skill when caring for patients with a complex or life-limiting illness (69). These conversations generally begin with exploration of parental values, goals, and fears leading to creation of a care plan that honors these views and wishes. When exploring goals of care with the family, it is pertinent providers assess the family's understanding of the diagnosis, prognosis, and range of treatment options. If providers can be present and supportive during these emotional conversations, there is an opportunity to learn more about the family, their views of quality of life, and how they make difficult decisions. This allows the provider to better understand and align with the family and ultimately helps to develop a care plan that is consistent with the family's goals and values. Early discussion of goals of care allows for time to reflect and plan compassionately and thoughtfully (80). For example, wishes for fetal monitoring during labor or newborn medical intervention should be established before families present for the birth of their child (7, 81–83). It is important to acknowledge that goals of care may evolve throughout the course of a pregnancy or after delivery as information available or the family's views and experience may change. For example, if an infant is unable to ventilate despite artificial respiratory support, this could be an opportunity to pause and reevaluate goals of care in the context of the evolving clinical situation. Ongoing perinatal palliative care support provides families with a safe place to explore the uncertainty of life-limiting fetal diagnoses and a team committed to careful communication of goals of care and palliative care birth plans (5, 67). **Table 2** uses a modified REMAP framework to outline key components of exploring goals of care in the setting of life-limiting fetal diagnosis (79).

TABLE 2 | Exploring goals of care for families facing life-limiting fetal diagnoses [modified REMAP framework (80)].

Reframe medical understanding and prognostic awareness
- Help family explore "What does all of this mean?"
- Acknowledge uncertainly and balance of "hope and reality"

Expect emotion
- Ask open ended questions (i.e., what are you most worried about?)
- Validate and respond to emotion. Be okay with silence

Map out value system family uses to makes decisions
- Inquire about faith, spirituality, religion
- Help to put those values in context of child's care (i.e., what is most important to you when thinking about how you want the team to care for your child?)

Align with values and hopes
- Reflect and summarize what we are hearing
- Seek clarification and verification

Propose a plan
- Suggest how identified goals may be achievable
- "I am hearing you ask that we create a plan that focuses on _______ for Baby's care after delivery" (time, comfort, better understanding diagnosis, etc.)

THE PALLIATIVE CARE BIRTH PLAN AS AN ADVANCE CARE PLAN

While the process of palliative care birth planning goes far beyond the tangible specifics of care surrounding delivery, the palliative care birth plan itself is important for families and providers. The palliative care birth plan functions as a communication and advocacy tool among care team providers. Providers caring for the mother often have different perceptions of family's understanding than providers caring for the baby (67). In order to fully support the family and provide seamless care, there must be open communication and shared understanding between all team members. The care team for a family delivering an infant with potentially life-limiting diagnosis will be comprised of both obstetrical and neonatal providers, including physicians, advanced care nurse practitioners, nurses, respiratory therapists, and more. The birth plan reflects the exploration of goals of care, summarizes the pregnancy journey, and shares the voice of the family while simultaneously documenting requests to all potential medical care providers for care during labor, delivery and the neonatal period for both mother and baby.

In order to meet the needs of both the families and providers, the palliative care birth plan should be viewed as an advance care planning document. Advance care planning includes deciding what medical care is desired in the setting of a life-limiting illness, exploring and sharing personal values, and documenting and communicating wishes (84). Each palliative care birth plan is unique to the family that creates it and serves as a tool to share their wishes and medical decisions regarding care of the mother and baby for the remainder of the pregnancy, around the time of delivery, and the subsequent neonatal care. As palliative care birth plans typically reflect discussions with medical care teams, the documents are generally created with the direct support of perinatal palliative care providers.

Palliative care birth plans include a breadth of information pertinent to providers involved in the care of the mother and baby. While many templates exist, key information should be included that will aid in seamless care during the delivery process and afterwards. General information such as the parent's names, the baby's given name (as many EMR's utilize automatic naming conventions based on the mother's name), and the parents' understanding of the diagnosis is important. Likewise, contact information for support persons should be readily available. This allows the care team to delegate tasks such as contacting family to others so that the parents can focus on the labor and delivery process while still having access to the support they desire. The document can include some specifics about goals during the pregnancy including celebrating milestones, memory-making such as pregnancy photography and preferences for follow up and delivery location. Wishes for maternal care during labor and delivery should be addressed ahead of time in the palliative care birth plan. This includes if the mother desires fetal monitoring, if she would want a cesarean delivery, analgesia, who should be present during the time of labor, and any special requests. Some women request music, aromatherapy, dim lights, or time alone with their partner during labor as a way to relax during this potentially stressful, exciting, and anxiety-provoking time. If they baby is live born, the medical care of the baby including specific requests for resuscitation or interventions should be clearly documented. This is an opportunity to express to the care team the wishes based on detailed discussions that have occurred throughout the pregnancy. Common discussions addressed include options for comfort measures only, non-invasive medical interventions, and invasive medical interventions. Beyond those

TABLE 3 | Key components of a palliative care birth plan.

Important information for the care team
- The parents' names
- The baby's name (if known)
- A diagnosis for the baby (if known) and a summary of what the family has been told to expect around/after the time of delivery. Any pertinent medical information should be included
- Names and numbers for important members of the care team (obstetrician, pediatrician, geneticist, other subspecialists)
- Names and numbers for important support persons (family, clergy, friends who should be involved)

Wishes for labor and delivery
- Vaginal or cesarean birth
  - ○ Indications for when they would want a cesarean delivery
- Fetal heart monitoring during labor
- Analgesia for mother
- Special requests during labor
- Who should be present
- Who should cut the umbilical cord
- Preferences for if the baby is stillborn

Wishes for medical care of the baby
- Invasive interventions vs. comfort measures only
  - ○ Any specific interventions they would or would not want
- Wishes for holding the baby
- Wishes for delaying routine procedures or providing them while the baby is in the parent's arms
- Wishes for feeding the baby
- Wishes for medications
- Wishes for additional testing

Wishes for memory-making and support
- Who should be in the room after the baby is born
- If there are siblings/family members, who will talk with them and how do they wish to be involved
- Photographs and videos
- Keepsakes: footprints, handprints, locks of hair, crib card, ID bands, blankets, clothing, heart beat recording
- Wishes for bathing the baby and special outfits
- Spiritual rituals/wishes

Plans for if the baby survives the first day
- If the family would like to take their baby home vs. receive end of life care in the hospital
- If they wish to be discharged home
  - ○ Code status at discharge
  - ○ The name of the hospice/home care that will support them
  - ○ Anticipated care needs at home
  - ○ Information about whom to notify if baby dies at home

Plans for if the baby dies before discharge
- Plans to ensure the baby is comfortable during the dying process
- Wishes to keep the body in their room
- Wishes for organ/tissue donation if eligible
- Wishes for autopsy or further testing
- Funeral home information
- Special wishes about transportation of the body

Any other additional requests the family may have

TABLE 4 | Important components of provider notes on birth planning.

Perinatal palliative care meeting note

Team members present:

Timing and reason for the visit

Family members present at the meeting

The name of the baby

Maternal history

Pertinent OB labs

Results of fetal testing and imaging

Name of the subspecialists involved in the medical care

Social History

> Share with others the important insight you have gained. Talk about the family structure, siblings, jobs, support structures, and faith. It is also helpful to talk about additional stressors and how the individuals involved in the shared decision-making are processing and coping with the diagnosis

Impressions, Counseling, and Plan

- Description of anomalies
- Counseling on prognosis and potential complications
  - Who was present and a detailed description of what the family has been told to expect as far as morbidity, mortality, and anticipated medical challenges (including the ranges and both short and long term complications). Discuss what treatment options, care paths, and interventions have been offered and how that is expected to change the disease trajectory.
- Goals of Care
  - Discuss the wishes of the family that they have expresses (comfort measures, non-invasive interventions, invasive interventions). Explain the various options and interventions discussed in detail (this is a guide for the providers). Also expand on any uncertainty and things that are important to the family if their goals are conflicting or cannot be met. Discuss labor and delivery wishes in the delivery room, code status, site of newborn care, anticipated procedures, or medications (help the providers who haven't met the family prepare for what to anticipate/expect), memory-making, religious ceremonies, plans for if the baby dies before discharge, and plans for if the baby survives to discharge

goals, parents may have specific requests for feeds, testing, and symptom management. Equally important, if applicable, are wishes for end-of-life care including location of death, symptom management, organ donation, autopsy, and funeral arrangements. One of the most important components of the birth plan to parents is the opportunity for memory-making. Who is included and what keepsakes or rituals are important is unique to each family. **Table 3** outlines details important to a birth plan.

Families identify palliative care birth planning as extremely important in preparing for the medical management of their child (67). It is not uncommon for families to request supportive care, varying degrees of resuscitation/delivery room interventions, and even life-sustaining or life-extending therapies for their child with a life-limiting fetal diagnosis. In creating requests for care, parents balance desires with fears. Common hopes and desires include meeting the child alive, enjoying time together as a family, and even providing a life and home for the child. These are balanced and contrasted with fears of pain, suffering, and extreme medical complexity (24, 79). The PnPC providers consistently utilize goals of care discussions and multidisciplinary birth planning to support families and address this duality resulting in requests for perinatal and post-natal care that is consistent with family values and rooted in medical understanding and prognostic awareness (85).

As much as the palliative care birth plan is a voice for families and communicates their advance care plan to the team, maternal and neonatal medical teams often seek expanded details on the medical care to be provided (67). It is important that all providers understand conversations that having taken place about the diagnosis, prognosis, and treatment options available. This often requires thoughtful and detailed documentation beyond the palliative care birth plan in the medical record. Delivery hospital teams should be directed toward details of collecting post-natal genetic samples or providing further neonatal assessments. Furthermore, documenting a family's decision-making structure and unique needs helps providers to partner with them during labor and delivery and provide optimal family-centered support while minimizing distress and miscommunication. An outline of pertinent information to include in provider notes around advance care planning in the form of birth planning can be found in **Table 4**.

Multidisciplinary fetal care teams have varying formats for discussion and planning upcoming deliveries of complex neonates and babies with life-limiting diagnoses. Discussions including key stakeholders (OB, neonatology, palliative care, etc.) ensure that providers are in agreement and all questions are addressed. Including families, psychosocial and spiritual support staff, and nursing teams may offer the opportunity to ensure interdisciplinary communication, allowing insight in to what is most important to the family and how to collaborate in family-centered-care after delivery. Additionally, palliative care birth plans should be readily available to providers and ideally documented in the electronic medical record before the time of delivery.

DISCUSSION

When a pregnancy is complicated by a life-limiting fetal diagnosis, the remainder of the pregnancy is vastly different from what was hoped for and expected. The grieving process may start well before the family even meets the baby. Often, families seek opportunities to celebrate and honor the life of their child while simultaneously planning for the ongoing pregnancy and navigating decisions surrounding care of the newborn with a life-limiting condition.

Exploration of goals of care, assistance in framing limitations of intervention, and advocacy for sound symptom management are well-supported by palliative care-trained providers. Multidisciplinary fetal care centers are growing to include perinatal palliative care specialists, who support families in processes of exploring value-systems, establishing goals and requests for medical care, and communicate those care plans with the broader multidisciplinary delivery team. Ultimately, perinatal palliative care birth planning and the creation of a palliative care birth plan with the guidance of an interdisciplinary perinatal palliative care team functions as an advance care planning tool for both families and providers. Further research dedicated to perinatal palliative care practices surrounding birth planning and multidisciplinary meetings as well as family experiences related to advance care planning during complicated pregnancies will lead to improved supportive care of families experiencing life-limiting fetal diagnoses.

AUTHOR CONTRIBUTIONS

DC, KE, and AS conceptualized and designed the report, drafted the initial manuscript, and approved the final manuscript as submitted.

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REVIEW
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Moral Distress in the Neonatal Intensive Care Unit: What Is It, Why It Happens, and How We Can Address It

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Moral distress is prevalent in the neonatal intensive care unit (NICU), where decisions regarding end-of-life care, periviable resuscitation, and medical futility are common. Due to its origins in the nursing literature, moral distress has primarily been reported among bedside nurses in relation to the hierarchy of the medical team. However, it is increasingly recognized that moral distress may exist in different forms than initially described and that healthcare professions outside of nursing experience it. Advances in medical technology have allowed the smallest, sickest neonates to survive. The treatment for critically ill infants is no longer simply limited by the capability of medical technology but also by moral and ethical boundaries of what is right for a given child and family. Shared decision-making and the zone of parental discretion can inform and challenge the medical team to balance the complexities of patient autonomy against harm and suffering. Limited ability to prognosticate and uncertainty in outcomes add to the challenges faced with ethical dilemmas. While this does not necessarily equate to moral distress, subjective views of quality of life and personal values in these situations can lead to moral distress if the plans of care and the validity of each path are not fully explored. Differences in opinions and approaches between members of the medical team can strain relationships and affect each individual differently. It is unclear how the various types of moral distress uniquely impact each profession and their role in the distinctively challenging decisions made in the NICU environment. The purpose of this review is to describe moral distress and the situations that give rise to it in the NICU, ways in which various members of the medical team experience it, how it impacts care delivery, and approaches to address it.

Keywords: moral distress, decision-making, neonatal intensive care, ethical confrontation, periviability, end-of-life care, medical futility

INTRODUCTION

American philosopher Martha Nussbaum writes "in all situations of choice, we face a question that I call 'the obvious question': what shall we do? But sometimes we also face, or should face, a different question, which I call 'the tragic question:' is any of the alternatives open to us free from serious moral wrongdoing?"(1) In Nussbaum's "tragic question" lies the crux of moral distress. This concept in relation to the field of medicine was first described and defined by Jameton in 1984 as "the psychological distress of being in a situation in which one is constrained from acting on what one knows to be right"(2). Though its first applications were primarily limited to nursing practice, the concept has since been broadened to include other groups of healthcare professionals.

The concept of constraint is central to the essence of the original definition moral distress and is a predominant theme in nursing literature. Moral dilemma or conflict, on the other hand, is more often reported as being experienced by physicians (3). In circumstances where there is moral conflict, values systems or duties relating to multiple treatment options are incompatible with one another and lead to psychological distress (4). The provider feels he must act in a way or provide care that is contrary to what he believes is the appropriate care plan (5). This discrepancy inherently challenges one's ethical principles. As a result, the term ethical confrontation has also been used to describe the associated distress (6). In an effort to provide the most inclusive review possible, moral constraint, moral conflict, and ethical confrontation will be considered in the discussion of moral distress.

While it is necessary to expand the definition of moral distress beyond the idea of moral constraint as providers are rarely faced with one definitive action on which to make a moral or ethical judgement, there must be a clear distinction between separate albeit related concepts of uncertainty, ethical dilemmas, and moral distress. In the neonatal intensive care unit (NICU), there is seldom absolute assurance in the outcome for a baby with the choices or treatment options available. There is always an element of uncertainty in outcomes that can lend itself to varying degrees of distress when discussing treatment options and decision-making with families. In addition, the NICU is wrought with ethically ambiguous clinical circumstances and complex decisions for a vulnerable population. This innately leads to feelings of internal discord, powerlessness, and uncertainty in physicians, nurses, and other healthcare professionals. This type of uncertainty, while undoubtedly causing stress and discomfort, should not be confused with moral distress. While there are times when these situations lead to distress if values are challenged, the presence of uncertainty or ethical dilemmas do not automatically equate to moral distress.

Frequently, though, decisions regarding end-of-life care and life-sustaining measures provoke moral distress, requiring a closer look at how this ultimately influences the care that is provided. Providers may feel constrained by the uncertainty in these decisions and having to counsel parents with limited information about the possible outcomes. Regardless of the etiology, moral distress can impact patient care and provider longevity (5, 7–9). If addressed, though, providers can minimize the negative affects while supporting each other and fostering personal growth (10, 11). The purpose of this review is to describe the role of moral distress in the NICU, reasons it occurs, how it presents unique challenges to different healthcare professions, and how providers can address it.

THE SCOPE OF THE PROBLEM

While it is accepted that moral distress is ubiquitous for those who work in healthcare, the true prevalence in the NICU is not well-established (12). Some historical challenges in assessing the prevalence include a focus in the literature on parental moral distress and adult patients, multiple definitions of moral distress, and an emphasis on the nursing profession (13, 14). While moral distress can affect any member of the healthcare team, it is most prevalent in those who provide direct care, such as physicians and nurses. This is likely due to the close relationships formed with patients and their families and the sense of responsibility in providing care (15). As such, it is increasingly recognized that moral distress can affect any member of the medical team regardless of profession.

For nurses, common causes of moral distress can be patient-focused or nursing-focused (16). Patient factors largely focus on quality of life, advocacy for the patient, and communication challenges with the families and care team. Nursing factors may include not having a voice within the care team, unclear roles, personal or team conflict, and feeling their integrity is being compromised. Historically, it was thought that the factors leading to moral distress were rooted in paternalistic approaches to medicine where nurses were instructed by physicians to provide care they did not always feel was appropriate (5).

With newer multidisciplinary care models, a recent focus on understanding healthcare provider moral distress has given more insight to the scope of this issue. In a recent qualitative analysis of attitudes around moral distress of NICU physicians and nurses, it was reported that up to 72% of providers experience moral distress at least once a month (10). Other studies show similar findings with up to 58% of nurses and physicians reporting specific work related moral distress (8). While it presents differently for each profession, all members of the medical team are vulnerable to this phenomenon.

Despite the prevalence, there is significant variability around the degree and frequency at which providers experience moral distress or ethical confrontation. This is likely due in part to the characteristics of the individuals included in studies. One study evaluating the impact of experience and knowledge of providers on the frequency of ethical confrontation reported that 35% of experienced NICU nurses and 19% of pediatric or obstetric residents experienced this challenge frequently at work. Overall, in the presence of ethical confrontation, greater experience and a higher level of understanding were associated with lower rates of moral distress. Perhaps with experience providers are able to process and cope with ethically challenging situations in a

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Abbreviations: NICU, Neonatal Intensive Care Unit.

manner that they do not feel moral constraint. On the other hand, increased knowledge about particular medical situations can open the possibility of experiencing moral distress. Individuals at medical centers that reported lower rates of moral distress around periviable resuscitation were less knowledgeable about outcomes for extremely preterm infants and were less likely to offer resuscitation at the lowest gestational ages. Those with more experience in the consequences of extreme prematurity, especially nursing staff, tend to overestimate poor outcomes and have higher rates of moral distress around offering resuscitation. Overall, the culture of a given center had the greatest impact on levels of moral distress, with the lowest frequencies of moral distress reported at centers that were the most homogenous with respect to language spoken and religious affiliation (6). This is in contrast to the longstanding notion that moral distress primarily originates from medical team hierarchy and suggests that heterogeneous values and background may contribute more to moral distress than was originally thought. Results of this and similar studies varied considerably with respect to the frequency and intensity of moral distress experienced by varying members of the medical team (6, 8, 12, 17).

Interestingly despite the challenges associated with moral distress, up to 76% of healthcare professionals feel it is a necessary part of caring for critically ill neonates (10). They view this feeling as a byproduct of being a caring, compassionate provider who is invested in the well-being of their patient and providing the best possible care. It is also viewed as a sign of innovation and progress in patient care (11). The internal struggle created by a difficult situation can, at times, be necessary. It challenges providers to acknowledge their own distress and biases in caring for patients and talking with families. This is important because the information communicated to families and the manner in which it is communicated can impact decisions made and ultimately lead to transference of distress (5). Simply because the medical team and/or parents agree upon a specific treatment course does not guarantee the accuracy or benefit of the treatment. Without the challenge of differing opinions, the ethical, moral, and medical appropriateness of such a decision goes unscrutinized (11). Discussions that emerge from moral distress promote exploration of multiple courses of action and encourage the most informed decision possible.

While providers feel that moral distress is an innate part of caring for NICU patients, it is accompanied by some degree of burden. Physician trainees in the NICU reported, through longitudinal narrative writing, that they experienced conflict in multiple situations that caused them distress and led them to question their own morals (18). The unique position of trainees in the medical team hierarchy places them at risk for constraint distress. They may feel obligated to provide care they do not agree with and are unable to voice these disagreements due to lack of confidence, fear of unfavorable evaluation, or concern for retaliation. As a result, they may feel numb and cope through detachment and desensitization, which can lead to compassion fatigue and burnout over time (18, 19). The emotional toll, which transcends beyond trainees, can lead to disengagement and ultimately effect patient care with avoidance behaviors amongst staff, increase length of patients' hospital stays, adverse patient outcomes, and increased pain levels in patients (12, 20). In addition, moral distress can contribute to increased burnout, decreased job retention by the healthcare provider, decreased staff retention by institutions, career transitions, threatened moral integrity, and a sense of failure to perform a professional and moral duty (5, 7–9). In order to provide optimal patient care and a safe environment for providers to thrive, when moral distress is prevalent providers are faced with a balancing act of minimizing the negative impact and leveraging the potential for progress (9).

WHY IS MORAL DISTRESS SO PREVALENT IN THE NICU?

Many factors can contribute to moral distress. Intensive care settings, staffing shortages, the need for timeliness/efficiency, and situations involving perceived futile care or different perspectives on end-of-life decisions place individuals at higher risk for moral distress (12). Structural aspects of the NICU, including lack of a consistent care team, poor communication, and understaffing lead to moral distress due to feelings of providing substandard care (21). Beyond the structure itself, variation in providers' own views and values within an institution can lead to reports of greater moral distress (6). Perhaps there are factors innate to the providers themselves and the culture of the institution that play a role in the presence of moral distress. When providers on the same care team have different values and moral compasses, there is a higher likelihood of some team members feeling moral constraint and subsequently moral distress based on the care provided.

The vulnerability of the NICU population, combined with the need for decision-making by proxy further compounds this risk. In the face of advancing medical knowledge and increasing reliance on technology and life sustaining measures, parents and healthcare providers struggle to find balance and determine what is truly in the best interest of the child (22). The frequency of end-of-life situations and the complex nature of NICU care lends itself to moral distress. One study found that a higher number of deaths and issues surrounding end-of-life care and resuscitations are associated with the highest levels of moral distress (8, 17). Interactions between team members and with parents and family can ease or amplify these feelings (17). Similarly, the degree to which medical team members agree with one another and with the parents' decisions can influence perceptions of constraint or control. It is far more common for healthcare providers to feel they are "doing too much" rather than too little. While this can be seen as an ethical dilemma, the resulting concern for inflicting undue harm and suffering results in a greater degree of moral distress (23). Interestingly, while physicians were more likely to disagree with the level of care a patient was receiving than nurses were, their reported levels of moral distress were lower than those of nurses. This may relate to the notion that increased education and experience with these challenging clinical scenarios allows physicians to better delineate ethical questions from moral distress. It also may relate to the fact that nurses are the individuals actively carry out the care plan.

While many factors contribute to how decisions are made for a vulnerable patient who cannot convey their own wishes, as with a neonate, there are external constraints beyond provider judgement that can impact and alter the care plan. Some factors that influence decisions include limitations put forth by laws and regulations, institutional policy, parental wishes and views of quality of life, hierarchy of the healthcare system, or ethics committee rulings. If these factors are in opposition to provider beliefs, it can lead to significant moral distress (25). Similarly, if there is a perception that medical interventions are causing unnecessary suffering, it can lead to feelings of helplessness (26).

Just as important to consider are factors internal and innate to the provider. If there is uncertainty or doubt about the diagnosis, prognosis, or most effective treatment course, it can lead to feelings of powerlessness, fear, lack of knowledge, or hesitation to advocate for their patient (8). These internal constraints are commonly described in the literature but less frequently recognized as sources of moral distress (9, 13, 27, 28). While some have advocated to expand the definition of moral distress to encompass situations beyond where there is moral constraint, these circumstances of uncertainty, even with a broader definition of moral distress are more appropriately described as moral sensitivity or ambiguity rather than moral distress (3, 4).

While moral distress can arise from any complex situation, most common scenarios that give rise to moral distress in the NICU involve end-of-life care, medical utility and futility, periviability, and disagreements about care plans (24). Each of these challenging clinical scenarios, which are described in greater detail below, involve medically complex decision-making.

End-Of-Life-Care and Palliative Care

There are over 15,000 neonatal deaths each year in the United States and the majority of these occur in the NICU (29). As a result, end-of-life care is routinely tasked to providers who take care of babies in an intensive care setting. Transitions to end-of-life care are never easy and are fraught with grief and uncertainties at a time of heightened emotions for all involved. More than 80% of the time, deaths in the NICU are preceded by a decision to limit, withdraw, or withhold life-sustaining treatments (30). This requires difficult conversations about the prognosis and multiple treatment options and approaches to care available. Once a decision to redirect care is made or it is evident that a patient is dying despite invasive medical interventions, subsequent care can greatly influence parental coping and medical team attitudes surrounding the death.

How providers define or personally view aggressive care, redirection of care, and the reasons for pursuing these care paths can contribute to feelings of moral distress. Singh et al. claim that when an actively dying patient is extubated so the mother may hold the infant as he or she passes, it is ethically different than compassionate withdrawal of care in the setting of a severe neurologic insult with likely poor long-term outcomes. The study also noted that timing of death did not significantly differ for infants who received full support vs. those for whom care was withdrawn (31). Both resulted in the infant's passing, but perhaps, the authors suggest, the former situation offers a clearer assessment of futility and allows moral judgement of providers to align with the actions taken.

The timing of when options such comfort measures, redirection of care, or pursuing invasive medical interventions are discussed is important as it can also impact feelings of moral distress. It may feel different to parents or healthcare providers to withhold care and refrain from initiating a particular care path than to withdraw or redirect care after invasive interventions are initiated. With increasing medical capabilities to care for the tiniest, sickest infants, trials of therapy are not uncommon. Parents may need time to process the information and prognosis, especially with unanticipated events or diagnoses and providers must balance the need to minimize suffering to the patient and utilize resources appropriately. Parents and providers utilize a shared decision-making approach to determine the boundaries of care plans, based not only on prognostic factors, but parental values and goals of care (32). Concern for prolonging suffering while awaiting parental decisions may intensify the medical team's feelings of failure to prevent undue suffering or harm to the patient. Likewise, due to provider discomfort with having these difficult conversations or their own perceptions about the appropriateness of such care plans, discussions often occur late in the hospital course. This can lead to distress and constraint of other providers or the families (32). Moral distress may result from the choice to provide invasive medical interventions, withhold therapies, or withdraw invasive medical interventions especially in circumstances in which there is little data to drive the decision that is in opposition to what the provider believes to be morally right.

Consider a former premature child corrected past term with bronchopulmonary dysplasia who thrives on continuous positive airway pressure and awaits parental decision regarding a tracheostomy to aid in development and facilitate transition to home. Despite declining redirection of care when the baby was critically ill, parents have consistently expressed the desire for an acceptable quality of life over quantity of life. In their eyes, dependence on any medical technology long-term is not an acceptable quality of life. They are increasingly distressed and often in disagreement with the medical team over the amount of invasive care the child is receiving. They express they would not want their child to be dependent on a tracheostomy or gastrointestinal tube feeds for an extended period of time. Ultimately, they elect to redirect care and remove the continuous positive airway pressure mask. The baby dies over the course of a several days. Some of the medical team expresses anguish over withdrawing non-invasive support on a baby with a favorable neurodevelopmental prognosis. They are faced with the constraint of parental desires and request to redirect care despite their own beliefs and views. In the zone of parental discretion, one may choose to honor such a request with appropriate counseling and risk assessment. While there is a level of moral judgement that this treatment course exposes the child to risk and ultimately death, the decision of the parents does not constitute medical neglect as there are also significant risks and burdens associated with the alternate care paths. A child with a tracheostomy who is ventilator dependent requires around-the-clock care and vigilance. They may have significant medical complications including death related to the tracheostomy and long-term ventilator support. In this challenging scenario, the involvement of palliative care and ethics is important. Through their involvement, the care team can better understand the complex factors that went in to the difficult decision made by the parents. This also allows for an avenue for staff to express their concerns, better understand the ethical questions at hand, and process their personal views in relation to the case.

Medical Utility and Futility

Consideration of medical futility when pursuing life-sustaining measures is important but difficult to apply to a clinical setting. Given the present degree of uncertainty in neonatal outcomes and continual advances in therapies and medical technology available, it is challenging to determine if interventions being provided have a reasonable likelihood of success and what defines success for any particular baby/family. Futility can be defined as a treatment that fails to provide discernable benefit (33). It innately requires delineation of goals as it is a term relative to the desired outcome. This is a challenging concept as many interventions provide some benefit and invasive interventions may allow an individual to live for a period of time. Due to the increasing use of technology and medical innovations, providers may feel that care is at times overly aggressive, without clear or definitive benefit. This can lead to distress and concern that they are causing undue suffering to their patient. It is reported that over half of attending neonatologists and over one-third of nurses have provided care they view as "overly burdensome." Similarly, 80% of physicians and 69% of nurses felt they had saved a child who "should not be saved," despite personal and moral objections to doing so (34). The decision of whether or not to continue aggressive care in the setting of a poor prognosis can be morally distressing. In particular, neurodevelopmental outcomes and the impact on long-term perceived quality of life is of great concern when weighing benefit against risk. Simply enabling survival is no longer the goal. A delicate balance between preserved quality of life, incorporating family goals, and minimizing suffering complicates the decision of the most appropriate treatment plan.

Quality of life is inherently personal as what is considered acceptable for one person may not be for another. While physicians overwhelmingly support parental involvement in decisions regarding end-of-life and palliative care, there is a tendency to anchor in the statistical majority with respect to outcomes (35). This can greatly influence how healthcare providers counsel a family and the degree of moral distress following a parental decision that does not align with their own views. In these situations, providers are asking themselves if a life with profound disabilities is a life worth living and if providing life-sustaining measures is worth the cost (36). While weighing these costs, they view what that life could potentially look like. Physicians tend to place a much higher value on cognitive function and independence than families do (37). This is likely, in part, due to the limited context and surroundings in which they interact with children with severe impairments. It can cause moral distress when a provider is challenged to accept different values and provide balanced counseling and at times provide interventions with goals in opposition to their personal values.

Regardless of the underlying diagnosis, when the medical team is asked to provide care that will extend a child's life, they often do so with the best interest of the baby in mind. However, when this is in the context of a child who will have a profound cognitive disability, the best interest standard often comes in to question (38). This is largely based on weighing the burdens and benefits of various treatment pathways for the child in the context of their family. While providers may have personal views, they cannot presume to know if the child would prefer death to a life with profound impairments especially when the child would have no other comparison (39). Often times the treatments available will extend the child's life and potentially facilitate leaving a hospital setting. The interventions themselves do not place the child at risk for imminent, preventable harm. As a result, it is difficult for providers to use ethical principles of best interest and non-maleficence as justification for advocating against such interventions. While there will likely be burdens for the child and family with the pursuit of invasive interventions, some feel these burdens are miniscule in comparison to the life the child is able to have and the potential relationship the baby is able to develop with their family and surroundings (40). With this notion, some advocate for providers to focus on the moral value of that relationship and the capacity for a caring relationship as opposed to neurodevelopmental outcomes when determining if invasive interventions are appropriate. The relational potential for which some advocate is morally meaningful regardless of others ability to see the reciprocity of the relationship (40). It is only with the help of the providers that these potentials can be realized. By acknowledging the significance of this relationship, providers can support their patients' families and potentially feel more comfortable with the interventions they are being asked to provide.

Often, providers find themselves in the zone of parental discretion when the risk of an option does not clearly outweigh the benefit. In such circumstances, parents are assumed to have their child's best interest at heart and know what is best for their family. As a result they are encouraged to be the decision-makers for their child unless the decision will cause express harm (41). When parental decisions ultimately differ from what providers believe is the right or most appropriate treatment course, either professionally or personally, the uncertainty of the outcome coupled with constraint of parental preference may produce profound moral distress for providers (42). This can be exacerbated by significantly differing beliefs and views on which these decisions are based. If a physician or nurse experiences moral distress as a result of disagreement with parental wishes, communication may become filtered through their own views and values, such that it offers more or less hope for achievement of parental goals than truly exists. This has been described as "ethically indefensible" and amounting to "deception" (43).

Consider a child with a long and arduous clinical course fraught with multiple bouts of severe clinical instability. He has undergone several invasive surgical procedures with refractory hypotension and hypoxia leading to irreversible end-organ damage. There has been no progress for months. Despite frequent counseling on the poor prognosis and lack of ability to wean ventilator support, parents continue to advocate for interventions with the goal of extending his life. They request that the medical team minimize discussions around redirection of care as they have expressed their wishes for their child. Physicians rounding on the child find themselves at a loss for contributions to his care plan that will lead to improvement in his clinical status and bedside nurses feel increasingly distressed by the invasive care they must provide. For months, he remains on the ventilator with no evidence of interacting with his surroundings in a meaningful or purposeful manner. When parents are available, they participate in his care. Otherwise, the nursing staff are his primary care-takers. He develops an infection and acutely decompensates. Despite interventions to treat the infection, he ultimately dies. Staff struggled with continuing to provide invasive medical care. Initially there was great distress over the perception that he was suffering. After discussions with the ethics committee and a better understanding of the degree of his neurological injury and appropriate administration of medications, most providers believed inventions were prolonging a short life in the hospital but no longer felt he was suffering. Ultimately, some reported less moral distress after understanding that he was not suffering, that parents needed more time with him, and that his parents needed to see him die under those circumstances to feel that they advocated for him and gave him every chance to survive.

Periviability

Periviable deliveries and resuscitation remain a controversial topic. Different institutions have various thresholds for resuscitation based on patient-specific factors, local data and outcomes, physician attitudes and maternal characteristics (44). Population-based estimation of outcomes lacks the precision necessary to make decisions on an individual level. The extreme uncertainty and lack of clear evidence or definitive clinical guidelines on periviable resuscitation can make counseling parents and caregivers challenging. In addition, knowledge and experience of the medical team members are recognized as factors that influence providers' personal views and greatly impact thresholds for resuscitation, estimation of mortality risk, and assessment of long-term outcomes they convey to parents (6).

Discussions around resuscitation often occur at a time with high emotions and stress where outcomes are uncertain and decisions are often made quickly. Providers are tasked with guiding parents through the recognition of their own values while providing available evidence to aid in a shared decision-making approach to care. However, the decisions that are ultimately made can remain a moral and ethical challenge for providers when their personal views and values are in opposition to the care plan they are developing (45). The vast majority of the time, neonatologists and parents are able to come to a mutually agreed upon course of action through discussion and goal sharing. In fact, in a cross-sectional study surveying neonatologists about their preferences surrounding periviable resuscitation, physicians chose options that aligned with parental wishes 66% percent of the time, as compared with options supported by institutional guidelines just 34% of the time (46). The agreeable nature of acting in accordance with parental wishes supports previous findings that understanding parental values and aiding in developing a care plan based on those values results in a lower frequency of moral distress. Further, defaulting to parental wishes and realizing there is no clear right or wrong option despite variation in personal views could partially remove the burden of decision-making from the neonatologist and reduce the moral dilemma he or she would otherwise face. There may also be an element of constraint from institutional guidelines that parental preference helps to override. Rather than strict criteria, guidelines may incorporate parental discretion and offer options that can be tailored to a particular patient. In this way, uncertainty is acknowledged, and shared decision-making can proceed with less impact from medical team bias or distress. A better understanding of the complexities involved in prognostication and decision-making can foster discussions and address discomfort or questions individuals may have (45).

Conflict and Disagreement

While differing views and opinions can result in discussions that encourage progress and novel ideas, it can also serve as a nidus for moral distress and residue that degrades the relationship between members of the care team and with the family (13, 47). Differences in the application of the zone of parental discretion may lend itself to variability in the amount of parental latitude given in determining interventions and treatment goals. Physicians may be viewed as overindulgent to seemingly unreasonable requests or too inflexible in incorporating parental perspectives. Physicians may agree to parents' wishes if those wishes do not constitute medical neglect while nurses feel distressed at having to carry out interventions that they feel cause more harm than good (48). Consultants may struggle to find helpful recommendations for a case they feel is futile. Different teams caring for the same patient may have opposing recommendations that can erode confidence and trust of the family and care team (13, 47, 49). In these situations, achievement of consensus despite initial disagreement may ultimately allow parents and caregivers to justify their decisions internally and serve to assuage discomfort associated with such ethical dilemmas.

ADDRESSING MORAL DISTRESS

As moral distress can have a significant impact on providers, patients, their families, and the care management plans that are derived, it is imperative that providers go beyond recognizing moral distress and develop effective ways to address it (50). This can lead to improved moral resilience, the capacity to tolerate moral distress and effectively function while mitigating the negative effects (49). While improving moral resilience is greatly dependent upon an individual's internal resources and ability to navigate ethically challenging situations, mindful and intentional reflection of the situation is pertinent to help providers process their emotions. There needs to be training around emotional support and coping for healthcare providers (18).

One of the most important initial steps is recognizing moral subjectivity. This can promote open discussions about the different views and perspectives of others that are driving their opinions and lead to a feeling of comradery. As a result, a shared sense of burden and responsibility for the decisions that are made can develop (5). Specific to end-of-life settings, studies suggest that higher levels of emotional intelligence may temper the negative effects of moral distress (20). By being able to recognize and reflect on emotions and incorporate them with cognitive reasoning, one can more productively manage and cope with morally challenging situations rather than having them lead to moral distress. Ultimately, this leads to improved interactions with patients and decreased levels of stress and anxiety (20). An important factor in this practice is improved and effective communication skills amongst all team members.

There have been many suggestions to support recognizing and addressing moral distress. Included in these suggestions are workshops, debriefings, ethics training, and practices such as reflective writing (20, 51, 52). These methods all have a uniting theme of bringing to the forefront the emotional impact of various situations and events. Writing, specifically, tasks an individual with critically examining the experience as a way of processing and coping. Narrative medicine can aid with coping and enhance physician empathy (18). While studies in other avenues have shown a positive effect in this type of interventions, a small study for NICU nurses in Iran failed to show a difference in moral distress intensity or frequency after 8 weeks of narrative writing (53). This may suggest that there are more complex factors than the act of writing about traumatic events that must occur in order for the practice to be effective. Beyond these measures of personal reflections, the way in which an individual approachs conversations and arrives at or navigates through challenging decisions can have a positive impact on moral distress.

Counseling and Decision-Making

Since the early 2000's, decision-making in neonatology has seen a shift in practice from providing information to parents and recommending a course of treatment based on evidence alone to a shared decision-making process. In this collaborative approach, factors such as parental values, emotions, trust in the care being provided, life experiences, goals of care, and other personal considerations are taken in to account when developing a care plan (54). The medical team and family must balance the often competing principles of autonomy, beneficence, and non-maleficence. This model of care allows for various stakeholders to express their personal views and values that help them determine a care plan. Values that predominate among healthcare professionals are intertwined with their own personal values. They often emphasize concepts such as patient dignity, quality of care, integrity, a duty to "do no harm," and alleviate suffering. There are times when their personal views of quality of life vastly differ from that of the parents. As a result, while a shared decision-making approach ensures that all members invested in the care of the baby have input, there are still situations where providers enact a treatment plan they do not feel is optimal for their patient (5). The highest level of distress reported by nurses occurs when following family's wishes when the medical team perceived they were doing "too much" (55). Regardless of the providers' personal views, through the processes of shared decisions-making, he can better understand the views and values of the family. Through recognizing important factors for them in determining quality of life or an appropriate care plan, the provider can make recommendations that align with those views rather than their own personal views. This understanding and approach to navigating goals of care or treatment plans can lead to confidence that the care plan established is a collaborative approach to care that is most appropriate for the individual baby and family regardless of the providers' personal views. As a result, this approach to counseling and decision-making can lead to decreased moral distress.

Continuity of Care

Inherent in successful shared decision-making is the cultivation of a relationship between the neonatologist, other members of the medical team, the patient, and their parents or other surrogate decision makers. Due to the nature of prematurity and critical illness in newborns, it is not uncommon for a child and their family to encounter multiple care teams throughout their NICU stay, with physician teams rotating approximately every 1–4 weeks. While nursing care teams are typically more consistent, they also involve some degree of variability. Though the same team members may come and go throughout the hospitalization, they participate in different parts of a given patient's care. This can make it difficult for parents to develop consistent relationships that foster trust and an understanding of values.

Despite efforts at standardization and adherence to evidence-based guidelines, there are often differences in the care provided due to the relative paucity of definitive data and algorithms for treatment in neonates. Constraint distress may be experienced due to a plan set in place by another physician or care team and in turn, bedside nurses or other staff members may be faced with the dilemma of aligning with the changing recommendations of the medical team while advocating for frustrated parents and original care plans (5, 16). Their personal views on the best course of action may also make counseling families or communicating with other staff more difficult, thus impacting overall the team's decision-making ability.

Watching a patient suffer due to poor communication and lack of continuity of care leads to high levels of self-reported moral distress (14, 23, 49). It is important with the multiple hand-offs and transitions in care that there are detailed conversations between care providers about the conversations that have taken place and that the unit has a collaborative approach to care. This will allow the providers to better understand how families make decisions, what information has been conveyed to them, and what the expectations for the care plan and treatment options are. Standardizing this approach can lead to seamless care despite changing providers. All providers should be engaged in methods to promote teamwork and to work collaboratively. It is important to ensure all members of the multidisciplinary team have an opportunity to share their thoughts and information to truly promote an environment of shared understanding and community (8). There should be clear methods in place to address conflict or disagreements that may arise among team members (12). It will also allow the family to feel more confident that the providers know their child and decreases the chances of abrupt changes in the care plan.

Education and Communication

An integrative review examining moral distress in NICU nurses around palliative care found that experiences of moral distress were variable (56). Factors associated with a higher level of moral distress included conflicts among care providers, lack of continuity of care, perceived futile care, false hope, and fluctuation in patients' clinical status. In response to recognizing moral distress, many centers have developed formal and informal educational opportunities to attempt to address causes of distress (18). While the etiology of moral distress appear multifactorial, certain interventions appear to alleviate moral distress. Education around end-of-life care, formation of a care team with the focus of establishing goals of care, and a protocol with a clear plan to address the dying process tend to decrease moral distress.

One center developed comprehensive educational interventions with modules to address nursing moral distress around end-of-life care (57). Overall comfort level with dying patients increased after participating in the educational program. After the educational sessions, nurses reported less compassion fatigue and noted that learning about self-care was important. They also benefited significantly from educations around legal and ethical issues surrounding neonatal end-of-life care. The importance of communication and the many emotions surrounding these situations was also highlighted. Through educational sessions, the importance of the desire for support was recognized. Regardless of the format, having avenues to discuss situations and allow staff to feel supported and heard is critical. Providing support through shared experience is beneficial and a way to foster meaningful relationships (18).

Specific training around communication and environments conducive to meetings and support for staff beyond traditional debriefings is crucial (12). It is known that that those who are less informed about outcomes may make a moral judgement based on misinformation or lack of information and experience moral distress as a result of treatment decisions that challenge their judgment (6). Clear and consistent communication with all members of the team regarding rationale, expected outcomes and options provided to the parents may help to alleviate distress that occurs as a result of simply "not knowing." To facilitate communications in an educational environment, some centers have introduced rounds or designated sessions for multidisciplinary teams to discuss challenging patients. One center described their approach to a multidisciplinary conference with the goal of promoting communication and consensus building (58). In a structured setting, all members of the NICU staff were invited to attend a discussion about a patient where there was distress or concerns related to the patient care. Input is sought from all team members as well as invited ethicists with the hope of navigating through the complex issues with the goal of improving communication and collaboration to reach a consensus about the case at hand. It appeared that participants felt these conferences took place later than they should in the course of care. Comfort with expressing distress directly correlated to the perceived support of the institution. While communication was cited as a cause of distress, the sessions were attended predominately by those who already felt comfortable recognizing and addressing distress. As a result it was suggested that with education around communication there may be an improved comfort level with crucial conversations that would lead to improved comfort in attending such sessions with the intent of improving patient care and decreasing moral distress.

Role of Ethics Consultation

With the frequency of ethically challenging situations in the NICU, the utilization of ethics consultations is common. These consultations can serve as forum to promote discussions on moral subjectivity and clarifying ethical challenges that arise (5, 59). This can aid in supporting moral resilience and alleviating compassion fatigue. Recommendations may vary from one institution to the other and laws vary from state to state. Lack of futility clauses and vague language regarding definitions of benefit and harm further complicate many complex clinical situations. In most cases, physicians and nurses feel the ethics committee role is to give advice and help promote a better understanding of the situation. In cases where parents and physicians disagree despite open communication an education, team members feel the ethics committee should make the decision on the most appropriate care plan. A small minority of providers would allow courts and the legal system to make the final decision in the case of clear disagreement (60).

Without clear disagreement, despite moral dilemma physicians and nurses would rather be involved with making the decisions. An ethics committee can help facilitate thoughtful discussions and open communication to better understand the issues that are leading to distress. Through these discussions, the important distinction between an ethical dilemma and moral distress can be made. There can be multiple treatment options that are acceptable while personal views on the most appropriate path may differ. By allowing thoughtful discussions about these options and why they are ethically acceptable, providers may have less moral distress as they have a better understanding in the complexity behind such decisions and also have an opportunity to express their views and concerns (49). Beyond that, ethics committees can help the team navigate communication with parents while understanding that the parents have ultimate responsibility of acting as moral agents for their baby (61). It is only with the recognition of the moral and ethical dilemmas unique to the NICU that organizational support for establishing ethical framework to support complex decision-making can truly come to fruition (62). With this support and structure in place, there can be improved quality of care through less conflict and distress amongst team members.

DISCUSSION

Moral distress is far more prevalent than even the current literature describes. It transcends healthcare professions and affects the entire healthcare team, the family, and most importantly the patient. While historically described as a negative emotion resulting from poor communication, discrepant values, and paternalistic hierarchy, it can be a source of growth and progress if leveraged correctly. However, if left unaddressed, it can contribute to burnout, job dissatisfaction and, ultimately, a lower quality of care provided.

The multitude of circumstances that can lead to moral distress require a thoughtful and tailored approach to patient care. Ethics rounds, debriefing after deaths, codes, or other challenging situations, and exceptional communication among team members and with caregivers are the cornerstones of minimizing the negative impact of moral distress while leveraging its role in progress. Understanding that different team members will feel moral distress to varying degrees is imperative. Offering options, such as the choice to abstain from care with which they disagree, may be one way to combat constraint in those who are most at risk to feel powerless or "without a voice." Simulation or case-based discussion can provoke thought and acknowledgment of one's own feelings in various situations so that providers feel confident in their approach to care. In addition, not delaying difficult conversations with parents can minimize the trauma associated with aggressive medical interventions or end-of-life care and will encourage open, deliberate communication. This will allow providers and families to feel that they made the most informed decision.

Most importantly, perhaps, is the recognition of moral distress as an entity, its impact on care provided, and helping staff identify its presence. By acknowledging the influence and implications of moral distress, providers are better equipped to minimize the negative effects and provide safer, more resilient care. Utilization of behavioral medicine resources, team discussions, mentorship and buddy systems for emotional and moral support can encourage self-awareness and a focus on addressing moral distress. Fostering a culture of openness, ethical sincerity and support for those who are struggling can reduce consequences such as burnout and job dissatisfaction. Communication with team members can offer insight in to other perspectives and guide future actions. Guidance from seasoned mentors and reassurance from colleagues regarding the normalcy of moral distress and advice for managing it provides the tools for personal and professional growth.

The impact of moral distress on decisions made in the NICU is under-recognized and represents a potential area for improvement in communication among staff members and with parents. Future areas of investigation should focus on the zone of parental discretion and its boundaries. In addition, given that physicians are most often involved in the medical decision-making for a child, further research on optimal interventions for moral distress tailored to address the specific challenges of different professions would aid in providing additional tools to combat moral distress.

AUTHOR CONTRIBUTIONS

MM conceptualized and drafted the initial manuscript. DC aided in conceptualizing, designing, and editing the manuscript. All authors approved the final manuscript as submitted.

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REVIEW
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Neonatal End-of-Life Symptom Management

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Purpose of review: Despite advances in technology and treatment options, over 15,000 neonates die each year in the United States. The majority of the deaths, with some estimates as high as 80%, are the result of a planned redirection of care or comfort measures only approach to care. When curative or life-prolonging interventions are not available or have been exhausted, parents focus on preserving quality of life and eliminating needless suffering. Parents hope their child will have a peaceful death and will not feel pain. A significant component of end-of-life care is high quality symptom evaluation and management. It is important that neonatal providers are knowledgeable in symptom management to address common sources of suffering and distress for babies and their families at the end-of-life (EOL).

Recent findings: Medically complex neonates with life-threatening conditions are a unique patient population and there is little research on end-of-life symptom assessment and management. While there are tools available to assess symptoms for adolescents and adults, there is not a recognized set of tools for the neonatal population. Nonetheless, it is widely accepted that neonates experience significant symptoms at end-of-life. Most commonly acknowledged manifestations are pain, dyspnea, agitation, and secretions. In the absence of data and established guidelines, there is variability in their clinical management. This contributes to provider discomfort and inadequate symptom control.

Summary: End-of-life symptom assessment and management is an important component of neonatal end-of-life care. While there remains a paucity of studies and data, it is prudent that providers adequately manage symptoms. Likewise, it is important that providers are educated so that they can effectively guide families through the dying process by discussing disease progression, physical changes, and providing empathetic support. In this review, the authors make recommendations for non-pharmacological and pharmacological management of end-of-life symptoms in neonates.

Keywords: Perinatal palliative care, neonatal, end-of-life, symptom management, pharmacological management

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INTRODUCTION

Neonatal end-of-life care is pivotal for the patient, parents, and providers. Whether the need for end-of-life care is the result of a comfort measures only approach with a known life-limiting diagnosis, the result of redirection of care after a trial of therapy, or the end result despite maximal invasive medical interventions, it is an incredibly difficult time filled with emotions and uncertainty. At a time typically filled with joy and hopes for the future they will have with their child, parents of neonates at EOL are grieving the loss of their baby even before death occurs (1). Parents note that when goals have transitioned to end-of-life care, they worry that their child will experience pain and physical suffering. They hope for a peaceful death with minimal discomfort for their child (2–8). One of the focuses of palliative care is the prevention and alleviation of symptoms including physical, emotional, and perceived suffering of the patient (9). In addition to providing support, it is paramount that providers focus on pain and symptom management (10, 11). At the same time, there can be many unknowns. For clinicians as well as families this is a time of uncertainty. It is often difficult to predict the timing of death, whether it be hours, minutes, days, or more (8). Effective management requires anticipation of the disease manifestations as well as the severity of symptoms for each patient. This can vary greatly and is often related in part to the underlying diagnoses and conditions leading to death. For any patient population, anticipating the symptom burden can be challenging even with a strong understanding of the pathophysiology. There is an added layer of complexity when caring for neonates as they cannot communicate their symptoms or express how they are feeling. For older children, adolescents, and adults, the gold standard of symptom assessment is self-report. However, there is no such standard that exists for preverbal patients. In neonates, there are few reliable tools to accurately assess symptoms and observational measures can be blunted depending on the gestational age and underlying disease process. This makes it difficult for providers to know if a neonate is experiencing symptoms at the EOL and if interventions are alleviating the symptoms to a satisfactory and acceptable level. In spite of all of these challenges, it is pertinent that providers are able to assess and treat symptoms a neonate may have at EOL so that the baby, the parents, and family have a peaceful experience.

SCOPE OF NEONATAL END-OF-LIFE CARE AND SYMPTOM MANAGEMENT

In the United States, there are over 15,000 neonatal deaths each year (12). The majority of these deaths are the result of congenital malformations or chromosomal anomalies, with up to 3% of pregnancies being complicated by a fetal life-limiting diagnosis (13–15). While some families opt for a comfort measures approach to care at home, the majority of neonatal deaths occur in the neonatal intensive care unit (NICU). Regardless of the location of care, most deaths, with some estimates as high as 80%, are the result of a comfort measures only approach to care or a redirection of care after a trial of therapy (16). As a result, there is a unique opportunity to thoughtfully and methodically plan to treat symptoms based on the anticipated needs unique to the pathophysiology effecting each neonate (8, 17–20).

Likely in part due to the paucity of data, there is significant variability in neonatal EOL comfort practices. A recent survey of members of the American Academy of Pediatrics Section on Neonatal-Perinatal Medicine inquired about neonatal end-of-life practices across America (21). Of the respondents, nearly half did not have institutional guidelines for comfort care. Of those that did, 80.9% addressed pain management and over 75% discussed non-pharmacological management. Other symptoms, such as secretions and anxiety, were addressed less than half the time.

END-OF LIFE SYMPTOMS

Symptoms experienced during EOL care that can be applicable to the neonatal population include agitation, dyspnea, neuroirritability, pain, and increased secretions (20, 22). Agitation and neuroirritability, generally speaking, are unpleasant states of arousal. They can be the result of the underlying disease process or from interventions and medications. Common manifestations include autonomic signs, increased motor activity, restlessness, and disturbed or disrupted sleep. Dyspnea is discomfort with breathing and is largely a subjective experience or sensation. Though subjective, a majority of verbal patients report dyspnea at the end-of-life and family members frequently perceive the patient is having dyspnea (23–26). As a result, it is reasonable to associate physical signs of increased work of breathing and tachypnea with the subjective experience of dyspnea at EOL. Pain, defined as an unpleasant sensation that results in discomfort and distress, is commonly experienced by pediatric patients in the last days of life (19, 22, 23, 26). The etiology and degree of pain experienced can vary greatly. It is important to know that there are different types of pain. Nociceptive pain is the result of tissue damage or inflammation and can be somatic, localized to a specific region, or visceral, affecting the internal organs. Neuropathic pain is the result of damage or irritation to the nerves. Identifying the type of pain that is occurring and understanding the etiology and underlying mechanism can help direct more effective approaches to therapy. Much of the early work of Anand was focused on understanding the neuroanatomy, neurophysiology, and neurochemistry of pain in fetuses and neonates (27). At 20–22 weeks gestation fetuses have developed their peripheral pain sensors and the neuronal migration of the ascending pathway to connect with the thalamus and sensory cortex has occurred. The peripheral nervous system and spinal cord are well-developed by 30 weeks gestation. The descending pathways develops later. This would indicate that neonates as young as 22 weeks gestation have the same number of pain receptors as an adult on a much smaller surface area and without the complete development of the inhibitory pathways. In addition, substance

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Abbreviations: NICU, Neonatal Intensive Care Unit; EOL, end-of-life; PPS, Proportionate Palliative Sedation; PST, Sedation Therapy; PSU, Palliative Sedation to Unconsciousness.

P and other neurotransmitters can also cause excitation of uninvolved neurons leading to lower pain thresholds and more robust, increased and longer pain responses in even the youngest neonates. These findings were accompanied by recognition in the clinical setting of pain responses and led to the acceptance the neonates are capable of experiencing pain and a change in practice around assessing and treating pain in neonates. With the understanding and acceptance that neonates experience pain, it is appropriate to assume that neonates should be properly monitored for signs of pain and treated accordingly (27–30). Excessive secretions are not as commonly seen but can be experienced if the neonate is unable to effectively swallow. This can be the result of the underlying disease process or from overall decreased tone, alertness, and activity during the dying process.

ASSESSMENT OF SYMPTOMS

While there are approved and validated methods to evaluate EOL symptoms for adults and older children, there are limited data and methods for neonates (20, 22, 31). Considering the clinical circumstances surrounding death, neonates may not be able to mount the normal physiologic response to pain that is seen in older populations. Similarly, neonates may not display other symptoms in the same manner other individuals do, making it difficult for providers to assess and effectively manage them. As a result, the methods and tools utilized in adolescents and adults cannot be translated to this population. The evaluation is largely reliant on clinician and family perception of symptoms. A recent qualitative exploratory study interviewed NICU nurses about the EOL care their patients received (32). While small, this study displayed themes of uncertainty, discomfort, and chaos. Nurses were not always certain, based on the physical signs they were seeing and on the clinical situation, if their patient was experiencing pain or any other symptoms. Lack of education, resources, and consistency in the approach to EOL care contributed to overall provider discomfort. In addition, providers may be focused on other tasks such as supporting the family which lend to subtle physical signs going unnoticed (8).

These challenges, along with historically limited documentation, makes it difficult to draw conclusions or to come to a consensus about standardized approaches to symptom assessment in neonates. One study of 20 neonates with planned redirections of care described the documentation of symptoms exhibited and the treatment of those symptoms during the dying process (8). The authors noted that all patient documentation had missing data related to EOL care and that there was a varying amount of detail with regard to the infant's symptoms and use of medications. Specifically, no pain scores were documented after invasive medical interventions were withdrawn or care was redirected. With gaps in documentation, providers may not be able to accurately assess the baby. Furthermore, this leads to significant limitations in research. Without detailed documentation regarding symptoms and interventions, there are challenges in drawing conclusions about current assessment and management of neonatal EOL care, as well as challenges in developing innovative or consistent guidelines and approaches to that care.

Pain, one of the most common symptoms discussed at EOL in older populations, has been historically challenging to assess in neonates. Additionally, there is concern that pain has been underappreciated and undertreated at baseline in this population. Research shows that fetuses and neonates have well-developed peripheral nervous systems and pain-related neurotransmitters are present as early as 22 weeks gestation; it has been accepted that neonates, even when born premature, can experience pain (27, 33, 34). In recent years, there have been improvements in assessment and management of neonatal pain, especially post-operatively (28, 29, 34–37). While not specific to EOL care, it is pertinent that providers utilize a neonatal specific pain assessment tool (9, 20). Carter describes at length the various pain assessment tools available for neonates and concludes that the evidence does not allow for recommendation of one specific tool but that using a validated tool is pertinent (20). It is pertinent that providers have been trained to use the tool and are experienced with it to increase the reliability of the reported results. An ideal tool is one that is validated in both premature and term neonates for subacute and chronic pain. Given the challenges of assessing pain in neonates, strong tools combine both behavioral and physiological data in the assessment. Common pain assessment tools suitable for this population are COMFORTneo, CRIES (Cry, Requirement for more oxygen, Increased vital signs, Expressions, Sleeplessness), N-PASS (Neonatal Pain, Agitation, and Sedation Scale), NIPS (Neonatal Infant Pain Scale), and PIPP (Premature Infant Pain Profile) (20, 38). While there is a paucity of data regarding pain and comfort in neonates at the end-of-life, one study showed that significantly fewer neonates had their pain evaluated with a pain assessment scale routinely used in other pediatrics patients receiving end-of-life care (20, 39, 40).

Other symptoms, such as agitation, dyspnea, and secretions are evaluated based on the clinical circumstance and physical assessment. Agitation or neuroirritability are suspected when there is self-reported or perceived increased motor activity, restlessness, and disturbed sleep. Similarly, assessment of dyspnea is reliant on a subjective report either by the patient or a visual assessment by another individual based on markers such as respiratory rate, the presence of hypoxia, and work of breathing.

To date, there remains little research on symptom assessment, symptom management, and the evaluation of interventions used in EOL care of neonates (41). Although there is a lack of standardized assessment tools or standards for interventions for neonatal EOL care, some providers advocate for treating common distressing conditions at the EOL in neonates whether or not they appear to be in distress. In these conditions, older, verbal populations commonly experience a high symptom burden from the same sources of suffering and their self-report and observations scores correspond to the severity of their burden. Providers suggest that neonates experience the same symptom burden and ought to be treat similarly (17, 42). For example, in verbal pediatric patients there is a high likelihood of dyspnea occurring after ventilator withdrawal, and there is also a high likelihood of dyspnea causing additional secondary symptoms that can be alleviated with pain medications (43). The same can be assumed with neonatal populations experiencing similar distressing symptoms after going through similar clinical experiences.

Parent Perception

Parental perception of suffering is extremely important as parents know their baby better than anyone and are sensitive to signs of pain or distress others may not be aware of. Parents may perceive their baby is distressed by physical changes they observe during the dying process. They often equate these changes with pain or discomfort (32, 44, 45). As symptom assessment has a component of subjectivity, there can be differing opinions from those interacting with the baby. In fact, one small study of EOL experiences for neonates revealed that 85% of providers felt symptoms were well-managed while only 57% of parents felt their baby was comfortable (44). Another study asked the bereaved parents of 40 neonates about their observations of symptoms during the last week of life (46). Mothers and fathers were given a list of 22 symptoms and on average the mothers reported 6.63 symptoms in their baby's last week of life while the fathers reported and average of 5.67 symptoms. Mothers most often noted respiratory distress, agitation, and pain while fathers most often noted respiratory distress, agitation, and lethargy. Both reported respiratory distress as the worst symptom in severity and the most difficult symptom to watch as they perceived it as the most uncomfortable symptom. On average parents reported their baby experienced a moderate amount of suffering. Only 6 of the symptoms inquired about where routinely documented in the medical record and of those there was no significant difference between parenteral or provider report. While parents and providers may not agree on the extent of suffering, provider initiated discussion around EOL care with parents allows for improved care of the neonate and reduction in anxiety of parents and possible tension between parents and the care team.

MANAGEMENT OF END-OF-LIFE SYMPTOMS

While there is a body of evidence and guidance for the treatment of EOL symptoms in adults and adolescents, there is limited data for neonates (20). As a result, there is a great deal of variability in pharmacological and non-pharmacological end-of-life symptom management for neonates (32, 47, 48). While there is variability, it is important that symptoms including pain are addressed by both pharmacological and non-pharmacological means (49). Other symptoms seen that may need to be addressed include dyspnea, agitation, neuroirritability, and secretions (20, 22).

Non-pharmacological Management

Preparing the family for what to expect during the dying process helps to alleviate parental anxiety. As a result, effective communication with families that supports the parental role and conveys empathy is required. Providers must prepare families for the dying process with clear and honest conversations, realizing that families will want varying degrees of information about the physiologic changes (9, 49). By understanding the changes that may occur, some families are less distressed by symptoms such as gasping and color changes (50, 51). They are able to appreciate that the physical changes occurring do not necessarily mean their child is experiencing dyspnea. They feel more prepared for the death, have less grief, and are more satisfied with the care their child received (2, 52–54). One study of 131 bereaved caregivers of children who died of cancer revealed that they did not feel prepared for the medical needs of their child as they approached death (55). Parent who felt unprepared for the changes at the time of death perceived their child suffered more. They expressed that they desire clear, honest communication about the medical issues around the time of death. Without an understanding of these changes, they may feel powerless or unsure of what their child is experiencing. By fostering open communication, providers can set expectations, address misconceptions, and encourage the family to convey when they are concerned that their baby is having symptoms so that they are addressed in a timely fashion. This is important not only for the comfort of the baby and the parent's experience with their child, but also for how the parents process the death of their baby. Parent perception of suffering has a lasting impact that can prolong and complicate their grief. Parents who are uncertain about what to expect at the EOL have more severe grief afterwards (56, 57).

Care should be taken to reduce uncomfortable procedures or invasive touching/stimulation (58). When symptoms occur, interventions such as decreasing stimulation, massage, skin care, mouth care, elevating the head or repositioning for dyspnea, fluid restriction, and gentle suctioning may be beneficial (9, 42, 49). Gentle suction and repositioning, for example, are more effective than pharmacological therapy the majority of the time for secretion management and noisy breathing. Swaddling, skin-to-skin contact, and non-nutritive sucking can be beneficial for agitation and pain. These measures help to meet the basic needs of neonates and promote positive bonding experiences (59). Families appreciate the opportunity to bond with and parent their baby (60). Eighteen nurses in Scandinavia were interviewed about their practices around skin-to-skin contact for parents and neonates during EOL care (61). Skin-to-skin interaction is known to decrease pain, improve labored breathing, and have positive psychosocial effects. Providers believe facilitating this care promotes comfort and bonding for both the mother and baby. This, in conjunction with promoting emotional and physical intimacy for families, can be very effective in alleviating dyspnea and agitation as hypoxia and hypercapnia that can occur during this time also have natural sedative effects (39). While there is expressed knowledge of the benefit of non-pharmacological interventions, documentation of their use in the medical chart remains limited (8). This makes it difficult to know the true extent to which these techniques are utilized and are effective.

Pharmacological Management

Non-pharmacological interventions will often alleviate some but not all symptoms neonates experience at the EOL. If anticipated and discussed ahead of time, parental apprehension and resistance to using medications to treat symptoms can be alleviated. Historically, providers have been hesitant to administer medications, specifically opioids and benzodiazepines, out of concern that they will hasten death. This concern of providing medications that have respiratory depression as a side effect in a medically fragile neonate has led to inadequate treatment of symptoms (39). However, there is a commonly accepted ethical notion known as the principle of double-effect which states that an action is justifiable if the nature of the act is good, the good effects are the intended effects as opposed to the bad effects, and that the good effects outweigh the bad given the circumstances (20, 47). Along these lines, it is considered good medical practice and standard of care to alleviate symptoms and pain during the dying process. In addition, multiple studies have shown that administering opioids and sedatives in appropriate doses to treat EOL symptoms does not hasten death, carries little risk of respiratory depression, and increases perceived comfort during the dying process (20, 62, 63).

While there remains variability in practice around administering medications for EOL symptoms in neonates, many providers give some type of medication. Typical classes of medications utilized are opioids, benzodiazepines, antipyretics, anticholinergics, diuretics, hypnotics, and anticonvulsants (9, 42, 64). The majority of studies in neonates are limited to intravenous administration in a hospital setting. A study of 20 neonates with planned redirections of care noted that most babies received pharmacological therapy. Eighty-five percent received opioids or benzodiazepines prior to withdrawal of invasive medical interventions and 60% received them afterwards. The majority received intravenous medications with one receiving oral medications, and five receiving aerosolized forms (8). One of the earliest studies looking at practices around neonatal EOL care reviewed deaths over a 3 year period in the NICU secondary to withdrawal of or withholding invasive medical interventions (18). Eight-four percent of neonates were given opioid analgesia. When care was redirected in the setting of perceived suffering or with major surgical or congenital anomalies, infants were more likely to receive opioids, most commonly morphine or fentanyl. If deaths outside of the delivery room were excluded, birth weight had no impact on the likelihood of receiving analgesia. Doses of opioids given were consistent with typical the pharmacological range when taking in to account previous opioid exposure. There was no significant difference in the time from withdrawal of invasive medical interventions to death for those who received no opioids or <0.2 mg/kg of morphine compared to those who received >0.2 mg/kg of morphine. In the institution at that time, the nurses would routinely ask physicians for opioid analgesia if they felt the neonate was in pain based on their assessment. The use of benzodiazepines was not typical and as a result there was no use of them documented in the study.

Other studies similarly found that there was no difference in birth weight or gestational age in whether or not infants received medications for symptoms as part of EOL care and that the vast majority of patients who died in the delivery room did not receive opioids (39). A study of 171 neonates at a level III NICU showed that 27.5% did not receive opioids or benzodiazepines for EOL care (17). Those in the group weighing <800 g were less likely to receive these medications. However, those weighing >1,500 g did not receive pain medications as often as those weighing 800–1,500 g. It was thought that potentially those in the 800–1,500 g group more often had diagnoses associated with pain. The fact that any died without comfort medications, though, was concerning to the authors.

One study retrospectively reviewed practices around medication administration for neonates at four NICUs in the United States, Canada, and the Netherlands (47). Of note, similar to previous studies, none of the babies who died in the delivery room received medications. Outside of the delivery room setting, most neonates received some type of medication for comfort, an opioid and/or benzodiazepine, in the 48 hours prior to redirection of care and after the redirection of care. Benzodiazepines were utilized more frequently in the NICUs in the United States than in the other countries. However, only the NICUs outside of the United States increased the dose of comfort medications prior to redirection of care. If a neuromuscular blockade was already being used, it was discontinued at two of the centers but continued at the other two. Only the NICU in the Netherlands administered a neuromuscular blockade after redirection of care and that was only to relieve suffering at the request of family.

While it is well-known that there is significant variability in the assessment and approach to treatment of pain in neonates in other countries, there is little evidence with regards to variations in practices around EOL care for neonates. The largest difference in the literature around EOL practices in other countries is related to euthanasia. While a rare occurrence, there are instances where the Dutch and Belgium consider euthanasia of neonates to be acceptable. The prognosis must be certain, there must be evidence of undue suffering, and parents must give consent. While this topic is sensitive, complex, and wrought with ethical debate, it is an acceptable practice for the Dutch and Belgium under rare circumstances that are tightly regulated (65–67).

Classes of Commonly Used Medications

Alpha 2 adrenergic agonists, clonidine, and dexmedetomidine, have analgesic and sedative properties. They been used in the neonatal population, particular after cardiac surgery, for some time (36). These options are appealing as they are usually well-tolerated and have minimal effect on respiratory drive.

Anticholinergics block acetylcholine either centrally or peripherally. As a result, they can cause fever, tachycardia, and other physiologic symptoms. In proper doses these side effects can be minimized and the effect of anhidrosis can be beneficial. Specific to EOL care this can reduce the amount of secretions. It is important to note, though, that there is little evidence that pharmacological therapy is effective for secretion management or the death rattle during the dying process (20).

Anticonvulsants have properties that lend themselves to EOL care in certain circumstances. Seizures themselves are unlikely during the dying process unless there is a preexisting condition. However, some of these medications can be effective at treating agitation or neuroirritability. Gabapentin, which works on the voltage dependent calcium channel, can be beneficial for neonates with difficult to control agitation or neuroirritability (68–71).

Benzodiazepines work by activating the gamma aminobutyric acid A receptors. They are commonly used in neonates for sedative purposes and have no analgesic properties. Side effects can be myoclonic jerks, hypotension, and, at higher doses, respiratory depression, or excessive sedation. In end-of-life care, they can be used to treat agitation and dyspnea.

Cyclooxygenase-2 enzyme inhibitors, acetaminophen and non-steroidal anti-inflammatory drugs, have been well-studied in the neonatal population and as a result have been used to treat fevers and mild pain, or as an adjuvant for pain treatment plans. They could have similar roles in end-of-life symptom management. Non-steroidal anti-inflammatory medications convert arachidonic acid to prostaglandin and are known to have anti-inflammatory, analgesic, and antipyretic properties. They can inhibit platelet function and lead to renal impairment. If already part of an analgesic treatment plan, they would likely be beneficial to continue for EOL care.

Dissociative anesthetics such as ketamine, have amnetic, analgesic, and sedative properties. Ketamineis is not known to cause hypotension or respiratory depression in neonates. However, there have been concerns at higher doses in animal models of apoptosis which has led to limited use until recently in the neonatal population (72, 73). In human models and in the presence of painful stimuli, it has been shown to have a possible protective effect (74). Given the limited data, its use had been limited to invasive procedures. In opioid exposed patients, it could be beneficial to treat pain, dyspnea, and agitation at the EOL.

Opioids are a class of medications that act on the opioid receptors in the body. Depending on the specific medication, they work on the mu, kappa, and delta portion of the receptors with varying degrees of affinity. They have effective analgesic and sedative properties and are readily used in neonates (29, 36). They are most commonly used for and most effective in treating pain, dyspnea, and agitation during the dying process.

Routes of Administration

Historically, in the hospital setting, IV access has been maintained though EOL care as a stable way to provide comfort medications either as continuous or intermittent infusions (64). Both methods can lead to adequate symptom control but some providers feel intermittent infusions, if possible, may make it easier for families to hold their babies and may be more conducive to memory-making. As for subcutaneous drug administration in neonates, there is very little data (75). Recently, there has been a focus on alternative and less invasive routes to provide medications for EOL care. The reasoning is largely to remove invasive equipment such an intravenous needles, aid in memory-making, allow EOL care to occur outside to the NICU, and to allow the option for EOL care in the home setting while still effectively managing symptoms. The route of administration with the most data and experience in these circumstance is oral administration. There are well-established dosage guidelines of oral medications in neonates. Alternately, while oral transmucosal (buccal or sublingual) administration provides several benefits, there are very few medications concentrated or designed for this route of administration making its utility limited (76). One center utilized the limited data available to develop a protocol for neonates receiving EOL care (76). Medications used included morphine, midazolam, lorazepam, scopolamine, and glycopyrrolate with doses equivalent to intravenous dosing. There were clear criteria on when to administer the medications and which dosages to use. While compliance with utilizing the pain assessment tool was low, by report, all survey respondents felt symptoms were controlled with oral transmucosal administration and no adverse side effects were noted. The vast majority said they would recommend this protocol. Similarly, a small retrospective study looking for non-invasive methods of controlling symptoms examined intranasal fentanyl in neonates (77). This medication and mode was evaluated because of the rapid onset of action, ease of administration, and because opioids are utilized to treat two of the most common symptoms-pain and dyspnea. There is more data available on the intranasal route of administration in children as opposed to the sublingual or buccal route. Doses were similar to intravenous doses while accounting for incomplete bioavailability. In the 11 neonates who received intranasal fentanyl for treatment of EOL pain or dyspnea, they received an average of 4.5 doses and had symptom control without the need for intravenous medications. There were no noted side effects and some were able to be cared for in settings outside of the NICU. Another route that is at times utilized in the neonatal population is transdermal. While there are transdermal forms of opioids and alpha 2 agonists available, there are is limited data of their use in neonates to make clear recommendations. The dosages in which these medications are available may be one limitation to the use in the neonatal population.

PALLIATIVE SEDATION

While it is accepted and expected that symptoms should be appropriately managed during EOL care, there is a clear distinction between appropriately managing symptoms and providing the medication with the intent of ending life or hastening death (78–80). This is particularly true when there is a high symptom burden refractory to typical medications and doses. One article looking at Dutch practices found that the intent is not always obvious by reviewing the type and dose of medications used (78). In this nationwide study, the authors utilized retrospective chart review and physician interviews to describe the type, dose, and reason for medication administration surrounding EOL care in the Dutch NICUs. Comparable to other studies, roughly 14% did not receive medications. They found that doses of opioids and sedatives were generally increased following EOL discussions and that documentation of the reasons for this were present <50% of the time despite the physicians explaining the reasoning in interviews. Per interviews, medications were usually titrated to mitigate or prevent symptoms such as pain, agitation, and gasping. In 10% of cases the intent was to hasten the dying process. Opioids were administered above what they considered normal dosages in 5% of patients before EOL decisions were made and in 17% afterwards. Benzodiazepines dosages were above the considered normal dosage in 12% of patients after EOL decisions were made. Roughly 16% of neonates received neuromuscular blockades as part of the EOL care. In interviews reasons included to prevent gasping, per parental request, and to limit suffering. While there has been debate over the years around the use of neuromuscular blockade at the EOL, it is generally accepted that they should not be utilized after redirection of care as the principle of double effect typically cannot be justified and it would bring the patient to his death (9, 81). In cases where a paralytic has recently been given, if time is not allowed for the paralytic to wear off, there should be discussions with the family, the intent should be to relieve suffering, and it should be clear that the burden of waiting an extended period of time for the effects of the paralytic to completely wear off would significantly outweigh the benefits (9, 20, 47). Another study from Europe examining the use of medications with the intent of hastening death in neonates concluded that the medications used are highly effective, the infants were moribund at the time they received the medications, and there was a discrepancy in documentation of the reported intent and effect (79). Furthermore, they felt that it is not easy to distinguish between intent to end life and provide adequate symptom control during the dying process.

When sedation is utilized at the EOL as a means to decrease and control symptom burden, it has been referred to as terminal sedation, palliative sedation therapy, controlled sedation for intractable patients, EOL sedation, and continuous sedation for the dying (82, 83). Palliative sedation can occur across a continuum from anxiolysis to deep sedation with loss of consciousness. As pediatric palliative care has evolved, the field is able to articulate the terminology, objectives and methods to provide effective and ethical management of severe symptoms at the EOL. Providing an effective level of symptom control to match the symptom burden without hastening death through respiratory depression is the objective. The provision of palliative sedation has been supported by numerous professional organizations including the American Medical Association, National Hospice and Palliative Care Organization, American Academy of Hospice and Palliative Medicine, and American Academy of Pediatrics.

Many pediatric palliative care providers apply criteria for patients under consideration for palliative sedation prior to initiation of sedation including limited prognosis of hours to weeks, confirmation that the target symptom is refractory, parental consent, and a goal of comfort care and foregoing resuscitative efforts. Accurately determining prognosis can be challenging for providers and discussion with specialists and colleagues can produce an accurate perspective. While not often utilized in the NICU, palliative sedation may have a role for patients who have refractory symptoms and/or a lengthy history of exposure to pharmacological therapies typically used

TABLE 1 | Neonatal end-of-life symptom management: suggested pharmacologic medications.

Medication	Class	Symptom	Starting dose (per kg) with route and frequency	Comments
Acetaminophen	COX2 inhibitor	Fever Mild pain	15 mg PO/PR q6 6–8 mg IV q8	As an adjuvant for pain
Atropine	Anticholinergic	Secretions	0.01–0.02 mg PO q2	No strong evidence
Dexmedetomidine	Selective alpha 2 agonist	Agitation Pain	0.5–1 mcg IV/IN q2 0.5–1 mcg/kg/hr IV continuous	
Fentanyl	Opioid	Pain Dyspnea	0.5–2 mcg IN/IV q2 1–4 mcg/kg/hr IV continuous	Quicker onset of action
Gabapentin	anticonvulsant	Agitation Neuroirritability	5–15 mg PO q8	
Glycopyrrolate	Anticholinergic	Secretions	0.01–0.02 mg IV q4 0.04–0.1 mg PO q4	No strong evidence
Ketamine	Dissociative anesthetic	Agitation Pain	0.5–1 mg PO/IV q2-4	
Lorazepam	Benzodiazepine	Agitation Dyspnea	0.05–0.1 mg PO/IV q4-6	
Methadone	Opioid	Pain	0.05–0.2 mg IV/PO q12-24 (initially q4 for 3 doses)	
Midazolam	Benzodiazepine	Agitation Dyspnea	0.05–0.1 mg IV q2-4 0.2–0.3 mg Sublingual q2-4 0.25 mg IN q2-3 0.05 mg/kg/hr IV continuous	Short acting
Morphine	Opioid	Pain Dyspnea Agitation	0.05–0.2 mg IV/IM q2-4 0.15–0.5 mg PO/Sublingual q2-4 0.01–0.05 mg/kg/hr IV continuous	

COX-2 inhibitor, Cyclooxygenase-2 enzyme inhibitor; IN, intranasal; IM, intramuscular; IV, intravenous; PO, per oral; PR, per rectum; q2, every 2 h; q2-4, every 2–4 h; q4-6, every 4–6 h; q6, every 6 h; q8, every q hours; q12-24, every 12–24 h.

It may not be appropriate to continue feedings. They can be difficult to digest and cause fluid overload.

to treat EOL symptoms. The latter often results in tolerance and resistance to conventional symptom management. Beyond light sedation, palliative sedation can be classified as proportionate palliative sedation and palliative sedation to unconsciousness.

Proportionate Palliative Sedation

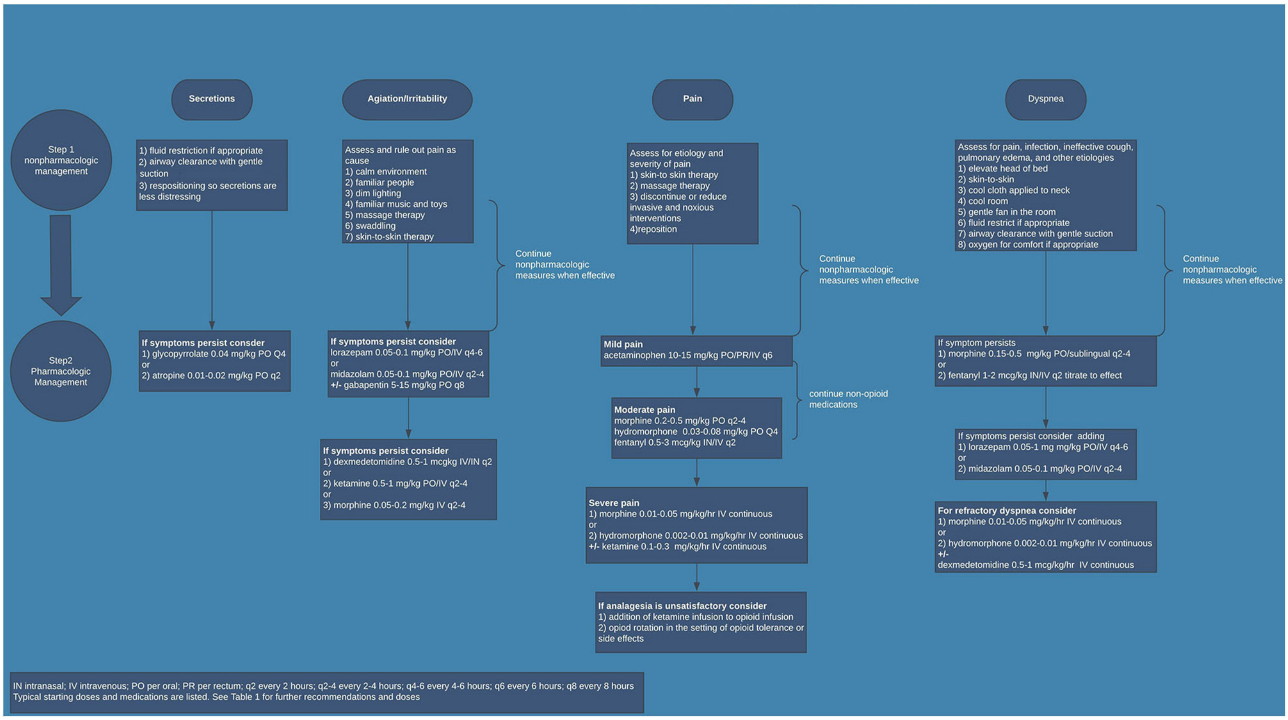
Proportionate Palliative Sedation (PPS) is symptom management that directly targets a specific symptom with a medication's identified mechanism of action while accepting that the side effects of the medication will cause sedation. Pain treated with analgesia is a common scenario in which the symptom is managed with a class of medications that directly targets the symptom. Opioids, for example, provide analgesia and can induce sedation at higher doses. Opioid analgesics directly reduce the suffering caused by pain while the use of hypnotics, such as midazolam, would mask the manifestations of the pain. Alternatively, seizure management, a less common threat to comfort at the EOL, can be complicated by antiepileptic therapy that can induce sedation at levels needed to optimally treat refractory seizures. In PPS the antiepileptic therapy that targets the source of suffering is appropriate and the end-point is relief of suffering while causing the minimum amount of sedation necessary.

The Level of Sedation and Titration

Sedation may vary from intermittent to continuous as the patient may waken spontaneously or may not waken once sedation is initiated. The depth of sedation can vary from light in which the patient readily wakens to voice and light touch to deep. In the latter, the patient is unable to be awake at all. In deep sedation, respiratory depression may be marked by decreased respiratory rate, tidal volumes, and obstructive breathing. In all scenarios, the objective is to provide relief of symptoms. The process for achieving adequate dosing is based upon medication titration proportional to the severity of the symptoms. When a symptom breaks through a previously achieved level of control, dosing increases can be based on the severity. Many providers use the following guide to make medication increases that are proportional to the severity: mild breakthrough −10 to 20% increase, moderate breakthrough −20 to 30% increase, and severe −30 to 50% increase. Any increase in dosing, whether intermittent dosing or continuous infusion, is followed by a period of reassessment to ensure the symptom has been addressed. The term Palliative Sedation Therapy (PST) has been advocated for to describe the active management that is required to adequately treat evolving symptoms in that frequent assessment of symptoms coupled with titration of medications to achieve the balance is needed. This active management process involves parents and family by including them in the symptom assessment.

Palliative Sedation to Unconsciousness (PSU)

There are clinical circumstances in which proportional palliative sedation is inadequate and suffering persists despite adequate titration and dosing. If the symptom is refractory, obtaining comfort can be achieved by using agents that alter the level



IN intranasal; IV intravenous; PO per oral; PR per rectum; q2 every 2 hours; q2-4 every 2-4 hours; q4-6 every 4-6 hours; q6 every 6 hours; q8 every 8 hours
Typical starting doses and medications are listed. See Table 1 for further recommendations and doses

FIGURE 1 | Neonatal end-of-life symptom management.

of consciousness but do not directly reduce the symptom itself. PSU is provided using classes of sedative and hypnotic medications in conjunction with the medication targeted to treat the specific symptom. Distinct from PPS, the intent is to induce loss of consciousness to eliminate suffering. Agents commonly used included propofol, phenobarbital, ketamine, and dexmedetomidine. The addition of a hypnotic is often done only after all measures to treat symptoms have been exhausted and is administered in an addition to the current regimen.

DISCUSSION

Appropriate EOL symptom management, while challenging at times, is important for neonates. Every individual deserves to have their symptoms addressed so they can have as peaceful of a dying process as possible. Not only does it improve comfort for the neonate, it also improves the EOL experience for the family. This allows them to focus on spending time with their baby and make meaningful memories which can help lead to less complicated grief and a positive experience for the limited time they have with their baby. While there are challenges associated with identifying and quantifying some of the symptoms that neonates experience during the dying process, many providers and families report common symptoms including respiratory distress, pain, and agitation. These are the same EOL symptoms that are often experienced in adolescents and adults.

It is important, especially when there is time such as with a planned redirection of care, that the healthcare team clearly communicates with the family about the dying process. Clear communication can lead to an open dialogue to address some of the concerns and fears of the family. By understanding the changes they will see in their baby, they can better prepare themselves and begin to understand the difference between physiologic changes and signs of distress or discomfort. When they perceive less suffering surrounding the death of their child, they have less guilt and less complicated grief.

It is pertinent that symptoms be regularly assessed during the dying process. If EOL care takes place in the NICU, routine assessment by staff should occur. If EOL care occurs outside of the NICU, family and staff should be educated as to the common signs of agitation, dyspnea, neuroirritability, pain, and increased secretions. For agitation and neuroirritability, signs of restlessness, abnormal movements, and disturbed sleep should be monitored closely. With dyspnea, common associations in a non-verbal patient are tachypnea and increased work of breathing. For secretions, increased saliva coming out of the mouth, gagging, and the "death-rattle" are common signs. Pain is the symptom that has the most validated assessment tools in the neonatal population. While not specific to EOL, institutions should have an agreed upon tool routinely used for preterm and term neonates that evaluates chronic and subacute pain. If EOL care is occurring outside of the hospital, family should be educated about ways in which their baby manifests signs of pain or discomfort. Despite the challenges associated with adequately assessing symptoms in neonates, when symptoms appear they should be adequately treated in a multidisciplinary approach. Many of the therapies that are beneficial for treating these symptoms in adults have been utilized in neonates with perceived benefit. Environmental and non-pharmacological factors are incredibly important. Promoting bonding, holding, and skin-to-skin contact is therapeutic for both the neonate and family. Implementing interventions such as decreasing painful procedures, decreasing stimulation, ensuring normothermia, and repositioning can be quite effective. Often times, though, non-pharmacological interventions are not adequate. This is particularly true outside of the delivery room or in situations where a neonate is being compassionately extubated. Identifying if there is a treatable cause of the symptom is important in developing an effective treatment plan. For example, dyspnea that is secondary to a pneumonia may be treated differently than dyspnea secondary to congestive heart failure. Based on current literature, institutional expertise, and clinical experience of the authors, suggestions for common medications are included (see **Table 1**). **Figure 1** is an algorithm for a practical application of and approach to EOL symptom management in the NICU population. Medication choices and doses used may need to be adjusted based on prior exposures. For example, if the neonate is already on an opioid drip the dose may need to be increased. At times, higher doses are needed to achieve adequate symptom management or alternative medications need to be utilized. In rare circumstances, where there is a high symptom burden and traditional treatment options have not achieved appropriate symptom control, palliative sedation may be appropriate. In either of these situations, if the provider is not comfortable or familiar with alternatives such as ketamine, dexmedetomidine, or gabapentin, or if the provider is not experienced with providing palliative sedation, expert input should be sought ahead of time.

Despite the recent focus on neonatal palliative care, there remains a scarcity of data in the literature about EOL symptom assessment and management in neonates. Some providers are still concerned about the possibility of medications causing respiratory depression and hastening the dying process. Studies have not shown this to be the case and it is accepted that it is ethically appropriate and medically necessary to treat EOL symptoms. While this is true, there are disagreements about the classes of or doses of medications to use and providers are left with little guidance on pharmacological management. This contributes largely to the wide variations in practices. Future research is needed to develop standardized tools to assess EOL symptoms in neonates. This would help providers and families have an objective way to evaluate symptoms. Beyond that, further research on the pharmacokinetics and pharmacodynamics and efficacy of the medications used is needed to better guide dosing recommendations and treatment options for these patients. A better understanding could lead to better symptom control and alternative medications, as well as alternative routes for administration. This could potentially lead to more choices for the location of EOL care for certain neonates.

With further research, institutions could develop more comprehensive guidelines for the assessment and management of EOL symptoms in neonates. In order to accomplish this, clinicians must take care in documenting their assessment of symptoms surrounding EOL as well as the interventions used. This includes noting the indication for the intervention and how effective it was at alleviating the symptom. This will lead to overall better control of symptoms, more consistency in the care provided, and less discomfort among staff and parents. One approach would be to develop an EOL symptom flowsheet in the electronic medical record where the various symptoms are listed along with the established mode of assessment. In this flowsheet, staff can easily document if symptoms are present, their severity, interventions used and a reassessment of the symptoms after interventions. This will provide valuable insight for patient care and for research. Even now in the absence of clear data for assessment and management of EOL symptoms in neonates, institutions should have guidelines in place. As part of that process, there should be education for staff around the dying process, EOL symptoms, and symptom management. With a better understanding, provider distress can be decreased. It also serves as an avenue to increase comfort and empower providers to have discussions with families around EOL care. While these discussions can be difficult, they promote a unified understanding of what to expect as well as shared decision-making that can ultimately translate to a better EOL experience for the neonate and the family.

AUTHOR CONTRIBUTIONS

DC conceptualized and designed the report, drafted the initial manuscript, and approved the final manuscript as submitted. MM aided in designing the manuscript, contributed to the content, and approved the final manuscript as submitted. All authors contributed to the article and approved the submitted version.

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OPINION
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Barriers to Perinatal Palliative Care Consultation

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Keywords: perinatal palliative care, barriers, newborn, need, neonatal intensive care

INTRODUCTION

Despite that infant mortality rates in western countries have improved over the past decade, 5.8 infant deaths per 1,000 live births are still reported, mainly due to prematurity and congenital anomalies (1, 2). According to recent data collected in the UK, the survival rate of pre-term infants born at 23+0 to 23+6 weeks of gestation and who receive active treatment is 38% (3). According to a registry study of the National Institute of Child Health and Human Development Neonatal Research Network, 20% of infants born at 22–24 weeks of gestation survives without neurodevelopmental impairment (4). Therefore, unfortunately, still a relevant number of newborns admitted to the neonatal intensive care unit (NICU) could have an unfavorable diagnosis and a poor prognosis. This leads to the notion of perinatal palliative care (PnPC) as an emerging field of palliative care. It involves different healthcare providers, including neonatologists, gynecologists, midwives, and nurses, and in a broader view all professionals who provide support strategies to the mother or the family and the fetus/newborn.

PnPC provides supportive care before and after birth and to families when a child is affected by congenital anomalies inconsistent with long-term survival, or when the extreme prematurity or post-natal irreversible disease of the child drastically reduces the possibility of survival (5, 6). Remarkably, in case of life-limiting fetal anomalies, palliative care could start before delivery (7) and up to 85% of parents continue pregnancies with a poor prenatal diagnosis worldwide (8). Therefore, the American College of Obstetricians and Gynecologists and the American Academy of Pediatrics recommend parental informed consent and discussion of all available care options, including PnPC (9).

Despite the importance of PnPC, several barriers to its implementation and utilization exist (10–12). At present, training programs in the field are proposed, the discipline is described as "very important" by hospital leadership but allocation of personnel, space, money, curriculum time remains poor (12).

In this paper, we comment on the barriers that make the access, planning, and development of PnPC services difficult.

IDENTIFICATION OF BARRIERS AND CRITICAL ISSUES

As a first step, we conducted an evaluation of literature with the aim to identify the most relevant barriers to PnPC and better focus our comment on this topic. In particular, we conducted a literature search on PubMed, TripDatabase, and Google Trend, using different combinations of keywords (e.g., MeSH terms: Perinatal palliative care, Barriers, Newborn, Need, Neonatal Intensive Care, life-limiting illness, end of life, ethical decision; free terms: Gestational age, culture, family, education, model organization, parents, newborn management, information, education,

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and others). No limitations in the search were applied. The reference lists of the retrieved papers were reviewed to find other papers worth consideration. Authors' own personal collections of literature were also browsed. The identified papers were then selected for inclusion according to their relevance for the topic, as judged by the Authors, in line with the non-systematic and narrative nature of this manuscript.

Based on this literature search and our clinical and organizational experience, we have identified four macro-areas of barriers and critical issues for the development of PnPC, namely: (i) Socio-cultural setting; (ii) Patients and their specific conditions; (iii) Training of healthcare providers (HCP); (iv) Regulatory and political issues.

SOCIO-CULTURAL SETTING

Misperception of the Role of Medicine and the Concept of "Health"

The management of PnPC could be influenced by families', nurses', and physicians' perceptions (8, 13, 14). Indeed, socio-cultural and economic context, and religious and moral values of future parents are taped among the main factors that influence the management of a fetus or an infant eligible to PnPC (13). There are widespread cultural barriers to consider death as an event occurring also in the neonatal/pediatric setting. The child is perceived as a human being at the beginning of his/her life journey and therefore the possible—or certain—fatal event is often neglected by parents and even healthcare providers. Medicine itself is often perceived as having the only aim of "cure," and the "Healthy" status is only associated with lack of any disease. Conversely, the WHO proposes a wider definition of "Health," as a state of psychological, physical, and social well-being; Medicine should aim at ensuring this state of health (15). In this perspective, it is important to clarify the notion of the inherent limits of Medicine: in some cases, the adoption of a palliative active care should be preferred over "doing whatever is possible," i.e., overtreatment (16–18).

The very same definition of "Palliative Medicine" is often misinterpreted as "end-of-life care" by parents of children eligible to PnPC and even by healthcare providers (19). This misinterpretation denies patients the opportunity of appropriate treatment.

Difficulties in Considering the Baby as a Person Who Can Feel Pain, Distress, Fear, and Suffering

It is widely accepted that, from the second trimester of gestation onwards, the nervous system of the fetus can perceive pain and distress, and these events have a major impact on fetal and post-natal life (20, 21). Therefore, even extremely premature newborns can "feel" the burden of disease, its symptoms, any invasive diagnostic and therapeutic procedure, as well as the distress due to the disease itself and the separation from mother. Unfortunately, the newborn is considered as unable to feel the above situations and therefore the burden associated with disease is neglected or underestimated. Furthermore, by negating newborn's ability to perceive pain and distress, the decision-making process of the clinical strategy becomes impaired. As a consequence, caring for such children requires a multidisciplinary approach that provides information regarding the risk, benefit, and burden for both mother and fetus.

Poor Information

Information on PnPC and the possibility to "cure" life-limiting or life-threatening diseases is scant, at best, and this lack of information limits the decision-making process by parents of children eligible to PnPC (19).

Moreover, the information that media provide is often only sensationalized news, which can be highly misleading and lacks an honest clinical, ethical and deontological evaluation. This poor information is important both for the patients, their families and for healthcare providers, also since the fear of social judgment limits the possibility of a correct assessment of the patient's status. Furthermore, lack of information and knowledge on PnPC limits the establishment of appropriate interventions by the healthcare system, with the recognition of dedicated units for PnPC.

PATIENTS AND THEIR SPECIFIC CONDITIONS

Heterogeneity of Clinical Situations and Types of Patients

The PnPC-eligible patients could be divided into three main clinical groups (22): (i) extremely premature at the limits of viability (<23 gestational week); (ii) prenatal/postnatal diagnosis of life-limiting/life-threatening diseases with poor prognosis and/or incompatibility with life; (iii) severe/critical clinical conditions without possibility of improvement, with high complexity of care and needing intensive support for vital functions' maintenance. In the second and third group, a range variety of conditions could be identified: neurological, cardio/pulmonary, and nephrological disorders or other genetic/chromosomal anomalies (23–25). Current medical evidence, experience of the healthcare providers, ethical, and regulatory bodies propose different strategies for the treatment of PnPC-eligible patients, according to their status and the type of life-limiting condition (26). Indeed, the diagnosis of a life-limiting condition can be difficult and can lead to doubts and different hypotheses.

Communication With Parents and Families

The parents of a child eligible to PnPC, as his/her legal guardians, have the responsibility of taking decisions. However, the inherent level of complexity of PnPC can lead to different views and perceptions between parents and healthcare providers, also due to the substantial variation in prognostic information given by obstetricians and neonatologists (27). This gap is due, at least in part, to the lack of unambiguous sources, tools, and training and it could lead to uncertain diagnostic and prognostic information (28). Several other factors should be taken into account, including the psychological dimension of the issue, the urgency (actual or perceived), the different role, perceptions, and emotions of healthcare providers and parents, the very same definitions of terms, such as "cure," "disease," "health," and "quality of life." The plethora of different feelings and emotions can be associated with reactions, such as negation of the issue, unrealistic expectations toward medicine, or even delegation of any decision to others and/or impersonal acceptance of the situation. This discrepancy can lead to communication issues between the parents and the healthcare providers, which, in turn, can delay or even deny the possibility of initiating palliative care.

The PnPC team should share the diagnosis, the anticipated prognosis, and potential interventions if any, with compassionate communication that respects patient cultural beliefs and values (29, 30) All the possibilities of care should be presented within a coordinated treatment strategy, focusing on maximizing quality of life, and comfort for the newborn. During counseling, team members should collect the appropriate information in order to give a tailored guidance to the family, free from any personal view and supporting parents in their decision-making process (28, 31). Therefore, team members should ask appropriate questions to explore the family's values, their understanding of the newborn's meaning and quality of life, in order also to establish a relationship based on trust and caring. An advance care plan is shared between parents and PnPC members, a birth plan, and/or a document of family wishes on delivery and resuscitation could be drafted (28).

All care options should be discussed, including pregnancy termination (abortion, within legal limitations), prenatal and postnatal intervention intended to promote survival, and palliative comfort care, which may include interventions to promote comfort and improve quality of life without intending to promote survival (29).

The lack of well-conducted communication can negatively affect the relationship between families of patients and PnPC healthcare providers, and between team members (32).

TRAINING OF HEALTHCARE PROVIDERS (HCP)

Dedicated training programs for perinatal palliative care are fundamental to improving the knowledge of healthcare professionals and the overall standard of care delivered to neonates.

In the past years, mounting interest has been given to palliative care, but this has not been paralleled by a growing in training programs; only scant data are available regarding the implementation of the PnPC educational programs.

In 2000, the American Academy of Pediatrics recommended an adequate training in pediatric palliative care and its subspecialties for all pediatricians to enhance the best possible quality of life for children living with a life-threating or terminal condition, using an integrated interdisciplinary approach (33). From 1975 to 2015, 99% of US medical schools offered education in palliative care, but only about 12–15 h of their entire medical school curricula were dedicated to palliative care (34). Similar results were reported for the European setting (35, 36). More data are available regarding training in PnPC for nurses and midwives (37–39), while only scant information regarding educational programs for neonatologist is available (40). According to the above, a structured curriculum dedicated to neonatal–perinatal medicine fellows to develop the complex set of skills required in this setting is eagerly warranted (40).

In a recent review investigating educational programs and events in the field of PnPC (41), the effectiveness of teaching healthcare professionals, methods of evaluating the teaching, and the teaching strategies used were analyzed. In total, 14 studies were included in the analysis, all published between 2002 and 2017. All studies referred as perinatal bereavement education as effective in terms of improvements in health workers of knowledge and communication skills, comfort in providing end-of-life care, and increased perceptions of the emotional needs of bereaved families. However, different evaluation tools were used and hence and it was not possible to determine which program was most effective.

Training programs in PnPC with specific content need to be developed and made available to all professionals who deal with perinatal death and the bereaved families (38, 40). Therefore, a dedicated program in PnPC should be offered to nurses, midwives, and the neonatologist in order to provide the best care for the fetus or infant and his family (42). Many studies reported that the main barriers to the development of pediatric palliative care are the inadequate training in communication and it is likely that this feature is also present in the PnPC field.

REGULATORY AND POLITICAL ISSUES

Organizational Issues

PnPC services represent a new, dynamic, and complex setting of care, which required dedicated time and resources; however, in daily practice, these are not always readily available.

The organization of a PnPC service poses serious challenges and requires the involvement of both local hospitals and the healthcare system. In PnPC, interdisciplinary competent teams are needed, and they should potentially involve different specialists (such as gynecologists, obstetrical specialists, nurses, neonatologists, geneticists, pediatric palliative care specialists, pediatricians, and bioethicists).

Few reports of experiences of well-structured PnPC networking models are available, and only a limited number of these investigated all difficulties and needs. This complex networking of different experiences needs the definition of shared protocol, dedicated organizational models, and adequate training. Meetings should begin in the antenatal phase and continue later, in order to identify and pursue a mutually agreed and appropriate care program. Dedicated time and appropriate places are needed for multidisciplinary meetings, and they may also help ensure a good communication between families of patients and team members, during the entire course of the patient's disease.

A quiet place should be available for parents during the infant dying process (6). Hospice concepts should be applied to neonatal care, providing a private family room where other family members can gather, and religious support can be given (43).

Resources Issues

From their moment of establishment, neonatology services have been characterized by continuous medical innovations, in a fast-growing field. However, only scant resources are available for this setting, requiring proper allocation (44). Moreover, literature offers complex networking models for PnPC services (45), making it difficult to estimate the costs for the public healthcare system, as well as the different social costs.

(Lack of) Regulations

The lack of dedicated regulations, which clearly define the right of children to PnPC, makes them, in many counties, an optional opportunity and not a duty of the healthcare system. Therefore, access to PnPC is largely dependent on single institutions, which further limits the diffusion of PnPC.

CONCLUSIONS

In this brief analysis, we identified and commented on many of the barriers currently slowing the development of PnPC programs. This issue is challenging, also due to the high heterogeneity of PnPC; however, we can propose some actions that may help in establishing proper PnPC programs.

- Increase the recognition of the actual lack of PnPC programs, in order to increase awareness of this setting.
- Recognize the inherent complexity of PnPC, making it necessary to establish specific training and services and collecting dedicated resources. Improvisation is not feasible in the setting of PnPC.
- Increase social awareness on PnPC, which aims at overcoming prejudices and providing the tools for proper reflections and decision-making.
- Provide proper training on PnPC, in terms of clinical, ethical, organizational, and communicational skills in order to make them able to pursue the best interest of the newborn with life-limiting/life-threatening disease.
- Promote research in PnPC and the collection of shared data, in order to find new tools applicable in different centers and settings.

AUTHOR CONTRIBUTIONS

FB: study design, data analysis, data interpretation, and writing. SC: literature search, data analysis, data collection, and writing. FR: literature search, data collection, and writing. MC: literature search, data interpretation, and writing. PL: literature search, study design, data interpretation, and writing. All authors: approved the final version of the draft.

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Assessment of Healthcare Professionals' Self-Perceived Competence in Perinatal/Neonatal Palliative Care After a 3-Day Training Course

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Background: Perinatal/neonatal palliative care (PNPC) offers a plan of care for improving the quality of life of infants when the prolongation of life is no longer the goal of care. The number of PNPC programs has increased in recent years, but training for clinicians has not kept pace. Therefore, an interdisciplinary team developed a 3-day intensive PNPC training course for physicians, nurses, and other healthcare professionals at Columbia University Irving Medical Center (CUIMC).

Objective: The aim of this study was to assess the efficacy of a PNPC training course in improving the self-reported competence of participants.

Study Design: A cross-sectional survey design was used to obtain data from 88 healthcare professionals who attended the PNPC training course. Data was collected using a validated questionnaire. The questionnaire included 32 items that queried participants about their self-assessed competence using a forced 1–4 Likert scale. The 32 items, which served as the outcome variables, were clustered into the eight domains of palliative care. The survey was administered through a web-based tool at the beginning and the conclusion of the course.

Results: Results from two-sample *t*-tests comparing pre-test and post-test self-assessed competence were statistically significant for each item across disciplines. Additional analysis revealed that after participation in the training course, the statistically significant differences between physicians' and nurses' pre-course self-reported competence disappeared.

Conclusion: The development of an evidence-based curriculum improved the self-reported competence of participants across disciplines, filled a specific gap in nurses' self-reported competence and addressed a global training need.

Keywords: perinatal, neonatal, palliative care, competence, training course

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INTRODUCTION

Perinatal/neonatal palliative care (PNPC) offers a plan of care for improving the quality of life of infants when the prolongation of life is no longer the goal of care or the complexity of the medical condition is associated with an uncertain prognosis. Palliative care in the perinatal period is a coordinated, interdisciplinary approach to obstetric and newborn care that includes a focus on maximizing the quality of life and comfort of newborns with life-limiting conditions (LLC) (1). This focus extends to supportive, coordinated care for parents and family members by providing anticipatory guidance, honoring parents' values, providing targeted consultations, formulating birth and treatment plans for the infant, and offering bereavement counseling.

The number of perinatal palliative care programs has increased in recent years (2, 3), but training and education for clinicians has not kept pace. There remains a gap in knowledge and competence regarding the management of parents expecting an infant with a LLC during pregnancy and birth and after delivery (4, 5). Furthermore, healthcare professionals from different backgrounds and disciplines have varying experiences, training, and confidence with regards to the delivery of PNPC (6–8). While suggestions for education and training have been proposed (9), formal professional coursework for PNPC is still very limited (10).

A policy statement from the American Academy of Pediatrics (11) on pediatric palliative and hospice care recommends that palliative care should be a part of all pediatric education, training curricula, and quality improvement. In addition, the National Consensus Project (NCP) Clinical Practice Guidelines for Quality Palliative Care states that all clinicians in direct care of seriously ill patients should have the necessary training and experience to complete palliative assessments and address common sources of suffering (12).

In response to clinicians' educational needs and recommendations from national organizations, an interdisciplinary team developed a 3-day intensive training course at Columbia University Irving Medical Center (CUIMC).

TABLE 1 | Items assessing self-reported competence grouped by palliative care domain (12).

Domain	Survey Items
Domain 1: Structure and Process	1. Offering the parent(s) the option of palliative care when they are making decisions about a fetus with a life-limiting condition 2. Talking to parent(s) about the range of options including palliative care after their fetus/infant has been diagnosed with a serious medical condition and potential adverse outcome 3. Talking to parent(s) about the range of possible outcomes after their fetus/infant has been diagnosed with a life-limiting condition 4. Engaging in shared decision-making after a life-limiting diagnosis is confirmed 5. Including parent(s) to create a plan of care for the pregnancy and birth 6. Including parent(s) to create a plan of care for the infant after birth 7. Providing prenatal care in a consistent way 8. Communicating with families about priorities of care 9. Understanding the role of palliative care and its integration into intensive care in the NICU 10. Accessing palliative care expertise and support when needed
Domain 2: Physical	11. Attending the delivery and providing palliative care to an infant with a life-limiting condition 12. Assessing the basic needs of infants with life-limiting conditions 13. Making a comprehensive care plan to address an infant's basic needs 14. Developing a nutrition plan for an infant with a life-limiting condition who is breathing and stable on room air 15. Providing interventions to achieve an infant's comfort 16. Assessing pain/discomfort in infants 17. Managing pain/discomfort in infants 18. Assessing other symptoms in infants (respiratory distress, irritability) 19. Managing other symptoms in infants (respiratory, irritability) 20. Following through with documentation of goals of care for an infant with a serious condition and potential adverse outcomes
Domain 3: Psychological and Psychiatric	21. Addressing the unborn child by name 22. Giving parent(s) a sense of normalcy during pregnancy and birth 23. Providing emotional support to parent(s) expecting an infant with a life-limiting condition 24. Exploring with families how they talk with their other children about the infant who is not expected to live
Domain 4: Social	25. Creating a comfortable, caring environment for parent(s) 26. Accessing resources related to perinatal palliative care (clinical resource materials, continuing education) 27. Knowing the community resources available for palliative care and how to access these resources
Domain 5: Religious, Spiritual, Existential	28. Sensitivity to spiritual needs
Domain 6: Cultural	29. Sensitivity to cultural values
Domain 7: Care of the Imminently Dying	30. Having conversations with families about the possibility of their infant dying 31. Recognizing signs of impending death in an infant
Domain 8: Legal and Ethical	32. Identifying and understanding ethical issues related to end of life care for infants

This study had two specific aims. First, the study aimed to evaluate the efficacy of the PNPC training course in improving the self-reported competence of course participants in perinatal and neonatal palliative care. Second, the study aimed to examine differences between physicians' and nurses' self-reported competence.

MATERIALS AND METHODS

Training Course

A 3-day course entitled *Next-Level Perinatal/Neonatal Comfort Care Training Course: Developing a Medical and Interdisciplinary Plan for Each Baby and Support for Their Family* was offered at CUIMC in June 2019. The program was designed by a team of medical experts to cover all aspects of PNPC (13). The target audience included the entire perinatal team: physicians, nurses, nurse practitioners, and other allied health professionals practicing in the perinatal arena (i.e., obstetrics, neonatology) who were interested in improving the practice of PNPC both nationally and internationally. Participants were provided with educational resources including a syllabus with objectives and literature references for each session. The course included presentations from an interdisciplinary team of clinicians including physicians, nurses, and researchers from neonatology and obstetrics as well as other professionals (a social worker, psychologist, speech-language pathologist, child life professional, and chaplain) involved in perinatal care and parent interviews. Faculty presented evidence-based rationale and strategies, offered interactive learning opportunities, modeled elements of effective communication, and practiced hands-on activities with participants. These educational strategies were administered to demonstrate and teach family-centric support and the attainment of a state of comfort for newborns with LLC. The eight domains of palliative care published by the NCP (12) were woven into the course curriculum, which included the following topics:

(1) The current state of the science on perinatal palliative care
(2) Strategies to assist families during decision-making processes
(3) Definition and roles of the members of the interdisciplinary team
(4) Essential elements of the perinatal palliative consult
(5) Advance care planning
(6) Evidence-based interventions in the prenatal, intrapartum, and postnatal periods that promote the health of the mother and family
(7) Essential elements of a neonatal palliative care plan
(8) Identification of potential barriers to institutional uptake of perinatal palliative care.

Study Survey

A survey was developed to assess participants' self-reported competence before and after the course and included 50 items with three sections. Section one included demographic data. Section two contained the outcome variables, which were 32 items addressing self-reported competence (**Table 1**). Participants were asked to rate their competence on a forced Likert scale of one (not competent), two (somewhat competent), three (competent), and four (highly competent). Participants could select "not applicable" if they did not feel as though they could assess their competence for a specific item. Competence was defined as the "habitual and judicious use of communication, knowledge, technical skills, clinical reasoning, emotions, values and reflection in daily practice for the benefits of the individual and the community being served" (14). The 32 items addressing competence were derived using a two-pronged approach. First, the literature was reviewed and a validated questionnaire by Stekenes and colleagues (15) was modified with permission to create the 32 outcome variables in the survey. The items were examined by two neonatologists and a researcher, reworded where needed, and agreement was reached regarding the inclusion of the final items. Next, the NCP's eight domains of quality palliative care were used to organize items and ensure that each domain was represented with at least one item. Section three queried participants' previous education in palliative care.

Study Design

The study was approved by the Institutional Review Board at CUIMC (AAAS4060) and informed consent was obtained from

TABLE 2 | Demographic characteristics of participants.

Demographic Characteristics	Number of Participants (%)
Profession	
Nurse	34/76 (45)
Midwife	2/76 (3)
Physician	31/76 (41)
Other	9/76 (11)
Area of Practice	
Neonatology	49/76 (65)
Obstetrics	9/76 (12)
Palliative Care	8/76 (10)
Pediatrics	3/76 (4)
Other	7/76 (9)
Highest Level of Education	
High School Diploma	2/76 (3)
Undergraduate Degree	24/76 (31)
Master's Degree	21/76 (28)
Doctorate	29/76 (38)
Age	
<30 years	13/76 (17)
31–40 years	32/76 (42)
41–50 years	11/76 (14.5)
51–60 years	11/76 (14.5)
61–70 years	9/76 (12)
Gender	
Male	16/76 (22)
Female	60/76 (78)
Country of Origin	
USA	62/76 (82)
Other	14/76 (18)

TABLE 3 | Pre-course and post-course self-assessed competency for all participants.

Item number	*N* (Pre-test)	*N* (Post-test)	Pre-test M (sd)	Post-test M (sd)	t-test (df); sig.
1	63	44	2.86 (0.99)	3.50 (0.35)	−4.17 (103); $p < 0.001$
2	63	46	2.95 (1.01)	3.48 (0.34)	−3.43 (103); $p < 0.001$
3	64	43	2.81 (0.95)	3.44 (0.40)	−4.06 (105); $p < 0.001$
4	64	44	3.00 (0.86)	3.55 (0.35)	−3.74 (105); $p < 0.001$
5	57	44	2.49 (1.22)	3.57 (0.48)	−5.99 (96); $p < 0.001$
6	61	43	2.77 (1.15)	3.70 (0.26)	−5.88 (92); $p < 0.001$
7	53	45	2.57 (1.10)	3.40 (0.65)	−4.44 (95); $p < 0.001$
8	68	42	2.96 (0.88)	3.57 (0.40)	−4.11 (107); $p < 0.001$
9	72	42	2.97 (0.90)	3.76 (0.23)	−5.87 (110); $p < 0.001$
10	68	40	2.88 (0.97)	3.63 (0.29)	−5.05 (106); $p < 0.001$
11	64	41	2.95 (0.93)	3.61 (0.44)	−4.11 (102); $p < 0.001$
12	70	41	2.97 (0.98)	3.56 (0.40)	−3.81 (108); $p < 0.001$
13	69	38	2.86 (0.86)	3.61 (0.30)	−5.26 (104); $p < 0.001$
14	66	40	2.38 (1.07)	3.43 (0.51)	−6.16 (102); $p < 0.001$
15	69	39	2.91 (1.08)	3.64 (0.24)	−4.94 (103); $p < 0.001$
16	67	39	3.09 (0.96)	3.56 (0.30)	−3.19 (104); $p = 0.001$
17	66	37	3.05 (0.97)	3.54 (0.26)	−3.37 (100); $p = 0.001$
18	67	42	3.19 (0.92)	3.67 (0.28)	−3.32 (105); $p = 0.001$
19	63	39	3.16 (0.88)	3.59 (0.30)	−2.93 (100); $p = 0.004$
20	70	43	2.77 (0.93)	3.58 (0.30)	−5.70 (110); $p < 0.001$
21	53	38	3.32 (0.99)	3.84 (0.30)	−3.20 (84); $p = 0.001$
22	57	42	2.92 (1.00)	3.60 (0.39)	−4.06 (95); $p < 0.001$
23	56	43	2.91 (0.88)	3.56 (0.44)	−4.01 (96); $p = 0.001$
24	72	46	2.33 (0.87)	3.46 (0.48)	−7.49 (113); $p < 0.001$
25	65	41	3.31 (0.75)	3.73 (0.25)	−3.19 (103); $p = 0.001$
26	71	47	2.69 (0.87)	3.60 (0.33)	−6.50 (116); $p < 0.001$
27	73	48	2.55 (0.81)	3.29 (0.51)	−5.05 (115); $p < 0.001$
28	65	43	3.12 (0.58)	3.67 (0.22)	−4.64 (106); $p < 0.01$
29	67	44	3.09 (0.60)	3.68 (0.22)	−5.01 (108); $p < 0.01$
30	68	43	3.01 (0.91)	3.53 (0.45)	−3.38 (108); $p < 0.001$
31	66	41	3.02 (0.94)	3.63 (0.33)	−4.13 (105); $p < 0.001$
32	72	46	2.96 (0.69)	3.68 (0.22)	−5.01 (108); $p < 0.01$

participants at the start of data collection. In order to assess the course outcomes, a pre-test, post-test study was developed. The pre-test, post-test design was cross-sectional and data were collected at two discrete points in time: before the training course commenced and after the course was completed. At the start of the course, a link to the pre-test survey was sent to each participant's electronic inbox using the web-based tool Qualtrics. An introduction to the survey and informed consent were provided on the first page. The training course was considered the intervention and lasted a total of 3 days. At the close of the training course, a link to the survey was emailed to participants. Follow-up emails to solicit completion of the post-test survey were sent 1 and 3 weeks after the conclusion of the course.

Statistical Analysis

Prior to performing the analyses, "not applicable" responses for each item were removed from the dataset and therefore not included in the final analyses. In order to examine differences between pre-test and post-test self-reported competence, each of the thirty-two items were compared using a two-sample t-test assuming unequal variance. Next, the items were clustered into their respective palliative care domains. The means for each item in each clustered domain were compared using a two-sample t-test assuming unequal variance. In addition, an independent sample t-test was completed to examine the difference in the self-reported competence between physicians and nurses at the start of and end of the training course.

RESULTS

A total of 88 professionals from nineteen US states and six countries (Australia, Burundi, Canada, England, Italy, and Russia) attended the course; however, only 76 attended the complete 3-day course and qualified for participation in the study. The pre-test survey response rate was 100% and the post-test survey response rate was 67%. Most participants were physicians or nurses and mainly represented the fields of neonatology and obstetrics. One third of participants had limited clinical palliative care experience (cared for

TABLE 4 | Comparison of physician and nurse differences in self-reported competency in pre-test and post-test items.

	Nurses			Physicians			
	Mean	**SD**	***N***	**Mean**	**SD**	***N***	***t*-test (df); sig.**
Pre-test Items							
Offering parents options	2.53	1.00	30	3.07	0.92	29	−2.12 (57), $p = 0.03$
Talking about options	2.61	1.05	31	3.18	0.94	28	−2.16 (57), $p = 0.03$
Talk about outcomes	2.39	0.98	31	3.21	0.77	29	−3.55 (58), $p = 0.001$
Engaging in shared decision-making	2.65	1.01	31	3.28	0.79	29	−2.65 (58), $p = 0.01$
Consistent prenatal care	2.17	1.07	23	2.85	0.96	26	−2.30 (47), $p = 0.02$
Access palliative expertise/support	2.50	0.99	34	3.18	0.90	28	−2.78 (60), $p = 0.007$
Nutrition plan for infant	2.09	1.05	32	2.70	0.95	31	−2.36 (60), $p = 0.02$
Follow through with documentation	2.51	1.01	35	3.00	0.78	30	−2.17 (62), $p = 0.03$
Conversations about dying	2.68	1.03	34	3.28	0.79	29	−2.59 (60), $p = 0.01$
Post-test Items							
Offering parents options	3.45	0.67	22	3.53	0.61	19	−0.35 (39), $p = 0.72$
Talking about options	3.45	0.67	22	3.48	0.60	21	−0.11 (41), $p = 0.91$
Talk about outcomes	3.40	0.68	20	3.52	0.60	21	−0.61 (39), $p = 0.54$
Engaging in shared decision-making	3.40	0.68	20	3.67	0.58	21	−1.35 (39), $p = 0.18$
Consistent prenatal care	3.30	0.80	20	3.55	0.80	22	−0.99 (40), $p = 0.32$
Access palliative expertise/support	3.47	0.69	19	3.71	0.47	17	−1.18 (32), $p = 0.24$
Nutrition plan for infant	3.42	0.69	19	3.32	0.82	19	−0.42 (36), $p = 0.67$
Follow through with documentation	3.47	0.69	19	3.65	0.48	20	−0.91 (32), $p = 0.36$
Conversations about dying	3.33	0.84	18	3.71	0.56	21	−1.63 (29), $p = 0.11$

fewer than five dying infants in their career), while 47% of participants reported having some previous exposure to palliative care education. Demographic data are presented in **Table 2**.

Analysis of the 32 items assessing self-reported competency showed that pre-course and post-course self-reported competence scores were statistically different for all professionals for each of the 32 items with higher scores on the post-course survey (**Table 3**). Comparisons remained statistically significant when the items were clustered into their respective palliative care domains.

Self-reported competency was then analyzed by discipline. The post-course self-reported competence scores for nurses were statistically higher than their pre-course scores for each of the 32 items. Physicians' post-course self-reported competence scores were higher than their pre-course scores and reached statistical significance for 20 of the 32 items including items 1 ($p = 0.03$), 5 ($p = 0.03$), 6 ($p = 0.01$), 8 ($p = 0.02$), 9 ($p < 0.001$), 10 ($p = 0.005$), 11 ($p = 0.04$), 12 ($p = 0.03$), 13 ($p < 0.001$), 14 ($p = 0.001$), 15 ($p < 0.001$), 16 ($p = 0.004$), 20 ($p = 0.004$), 24 ($p < 0.001$), 26 ($p < 0.001$), 27 ($p = 0.008$), 28 ($p = 0.002$), 29 ($p = 0.001$), 30 ($p < 0.001$), 32 ($p = 0.002$).

Scores for nurses and physicians were also directly compared. Prior to the start of the course physicians' mean scores for self-reported competence were higher than the nurses' mean scores for nine items. In post-course analyses, the nine self-reported competence items were again compared between nurses and physicians and were no longer statistically significant (**Table 4**).

DISCUSSION

This study found that the development and implementation of an evidence-based PNPC training course improves the self-reported competence of its participants across disciplines and in each domain of palliative care. Moreover, participation in the course filled a specific gap in nurses' self-reported competence.

The improvement in the self-reported competence in each item and in each domain of palliative care demonstrates the breadth of the material presented in the training course. The comprehensive training program addressed not only the medical aspects of perinatal palliative care but also the psychological, spiritual, cultural, and ethical aspects of care. The pre-course mean self-reported competence scores were particularly low for participants across disciplines on several items related to the psychological and social aspects of care. Participants reported the lowest self-reported competency scores in the areas of developing a nutrition plan and communication with the siblings of infants with a life-limiting condition. The creation of a nutrition plan is rarely included in PNPC education because most infants with a life-limiting condition have a short life expectancy. However, some infants may be stable at birth and continue to live for days, weeks, or months. Therefore, hunger and thirst must be addressed (16). Furthermore, colostrum care and non-nutritive sucking are recommended by the American Academy of Pediatrics as non-pharmacological interventions to alleviate pain and discomfort in neonates, including at the end of life (11, 17). Although PNPC emphasizes the care and support of the entire family, training is often lacking in this specific area, which was highlighted by the scores in the "somewhat competent" range for communication with siblings. Participation in this PNPC training course adequately addressed these topics as demonstrated by the marked improvement in the post-course self-reported competence scores.

Additional analyses revealed that nurses improved their self-reported competency scores on each item, while physicians improved their scores in 20 of the 32 items. The domains of palliative care most impacted by course participation were related to the integration of palliative care into NICU practices, addressing infants' basic needs, providing interventions to achieve infants' comfort, accessing resources for PNPC and initiating conversations about dying. Future training programs should continue to incorporate these findings to address the specific educational needs of physicians.

Prior to the start of the course, a statistically significant difference between physicians and nurses self-reported competence was calculated for nine items. These items were in three of the eight domains including the structure and process of care, physical aspects of care, and care of the imminently dying and reflect areas of PNPC that physicians often have more experience with. These findings were not surprising since previous work has suggested that physicians are often more confident than nurses in the practice of perinatal and neonatal palliative care (18, 19). After participation in the training course, these differences disappeared. This suggests that an intensive training course can fill a gap in nurses' self-reported competence in the practice of PNPC.

The training course included participants from different disciplines including neonatology, obstetrics, and palliative care. This is particularly important because PNPC practices require an interdisciplinary team of practitioners. Neonatologists are often at the forefront of neonatal palliative care initiatives and obstetricians' interest in acquiring competence in this field is growing (1). This growth is important since obstetricians are often on the front line of delivering news of a fetal diagnosis of a LLC to parents and providing recommendations.

The implementation of this training course addressed an international need. A recent study found that nearly half of the neonatal intensive care units surveyed did not have neonatal comfort care guidelines and 91% of respondents stated that their institution would benefit from PNPC education (20). Previous work suggests that appropriate PNPC education can increase knowledge, self-perceived confidence, and clinical skills (21). Insufficient knowledge about PNPC is a known barrier to its uptake (6). Furthermore, comprehensive training may alleviate some of the distress, helplessness, and discomfort clinicians have reported in previous studies (8). Finally, numerous nationally recognized organizations including the NCP, ACOG and the Worldwide Palliative Care Alliance have recommendations encouraging clinicians and institutions to develop PNPC programs, which can only be done well when there is ample training for clinicians (1, 12, 22). The CUIMC course attracted individuals from several countries. Importantly, despite the vast and diverse demographic origins of the participants, the course content universally improved professionals' self-reported competence in PNPC management.

The present study has several strengths. First, participants represented several disciplines and specialties which contrasts with previous work. Knighting and colleagues evaluated the impact of a training course on self-perceived knowledge and clinical practice in 73 professionals. However, 82% of participants had a nursing background and only three participants were from the field of obstetrics (21). A second strength of the study includes the timing of the administration of the post-course survey. Since participants completed the post-test survey within 1–3 weeks of course completion, it is unlikely that the improvement in self-reported competence can be explained by additional education or experiences outside of the training course.

There are several limitations of the study. In contrast to the study by Knighting and colleagues, participants' pre-course and post-course responses were not matched and fewer participants completed the post-course survey. In addition, the study focused on measures of self-perceived confidence but did not directly assess knowledge and skills. Finally, since this is a cross-sectional study, it does not measure participants' experiences after return to their healthcare systems. It is unknown if the course materials provided professionals with the ability to develop or improve PNPC programs. Therefore, longitudinal research is recommended to measure outcomes in clinical practice environments.

The development and implementation of an evidence-based PNPC course improved the self-reported competence of participants across disciplines and addressed specific self-reported competence gaps of nurses. This study provides a framework for the establishment of a PNPC curriculum, which addresses a global need.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by Columbia University Irving Medical Center Institutional Review Board. The patients/participants provided their written informed consent to participate in this study.

AUTHOR CONTRIBUTIONS

JH, CW, and EP developed the survey, analyzed the data, and wrote the manuscript. All authors contributed to the article and approved the submitted version.

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MINI REVIEW
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End-of-Life Care for Neonates: Assessing and Addressing Pain and Distressing Symptoms

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One of the most essential components of end-of-life (EOL) care for neonates is assessing and addressing distressing symptoms. There is limited evidence to guide neonatal EOL symptom management and therefore significant variety in treatment (1–4). EOL neonatal palliative care should include identifying and relieving distressing symptoms. Symptoms to manage at neonatal EOL may include pain using both non-pharmacologic and pharmacologic comfort measures, respiratory distress, secretions, agitation and neurologic symptoms, nutrition and gastrointestinal distress, and skin care. Also of equal importance is communication surrounding familial existential distress and psychosocial care (1, 5–7). Institutions should implement a guideline for neonatal EOL care as guidelines have been shown to decrease variability of interventions and increase use of pharmacologic symptom management (4). Providers should consult with palliative care teams if available for added multidisciplinary support for family and staff, which has been shown to enhance EOL care in neonates (8, 9).

Keywords: neonatal, end-of-life care, neonatal hospice, neonatal palliative care, neonatal pain

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INTRODUCTION

Neonatal end-of-life (EOL) care includes addressing pain and other distressing symptoms (1, 7, 10, 11). Neonatal intensive care units (NICUs) are generally competent in EOL care of neonates; however, evidence shows there are wide variations in methods for evaluating and addressing symptoms (1, 12). This variation likely stems from limited evidence regarding neonatal EOL symptom management given the lack of on-label neonatal pharmacologic treatments (5, 10). A 2016 survey showed only 55% of NICUs in the United States have a neonatal EOL guideline, and 45% do not have access to a palliative care team, showing there is much room for improvement regarding neonatal EOL care (1). This review aims to incorporate scientific research evidence with clinical expertise to aid in assessing and addressing neonatal EOL symptoms.

Vignette

A 25-week gestation 5-day-old neonate with severe grade 4 bilateral intraventricular hemorrhage and acute pulmonary hemorrhage is currently intubated on high frequency oscillator ventilation. Parents have decided to transition to EOL comfort care. The bedside nurse is concerned regarding increased facial grimacing of the neonate and asks which pain scale to use.

ASSESSMENT OF PAIN IN NEONATES

The most common symptom related to pediatric EOL care is pain, which is often underestimated in neonates (13–15). In comparison to adults, neonates experience pain that is more severe, diffuse, and prolonged (16). Several different pain assessment tools are validated for neonates; however, one has not been found superior over the others (5, 16, 17). These assessment tools do not evaluate for distress from other etiologies such as hunger, and interpretation must be completed in proper clinical context (5).

Pain should be evaluated every 15 min upon initial discovery. Once controlled, we recommend evaluation of pain at least every 3 h. The scale chosen should be part of your institutional guidelines. Your chosen assessment scale should be used consistently, as scenarios allow, and chosen with input from your nursing staff. Assessment scales to consider include but are not limited to the following.

Acute neonatal pain scales to consider:

- Behavioral Indicators of Infant Pain (BIIP) evaluates pain using behavioral state, facial expressions, and hand movements. Validated since 2007 for acute pain in preterm infants 23- to 32-weeks gestation (18).
- Premature Infant Pain Profile (PIPP) uses gestational age, behavior, heart rate, oxygen saturation, brow bulge, eye squeeze, and nasolabial furrow. Validated since 1996 for acute and postoperative neonatal pain (19).

Prolonged neonatal pain scales to consider:

- COMFORTneo Scale uses alertness, calmness, respiratory response, crying, body movement, facial tension, and body muscle tone. It was validated in 2009 for prolonged neonatal pain (20).
- EDIN Scale (Echelle Douler Inconfort Nouveau Né) evaluates facial activity, body movements, quality of sleep, quality of contact with nurses, and consolability. Validated in 2001 for prolonged pain specifically in preterm neonates (21).
- NPASS (Neonatal Pain, Agitation, and Sedation Scale) looks at crying, behavior, facial expression, tone, and vital signs. Validated in 2008 for prolonged postoperative pain as well as pain with mechanical ventilation. It is validated for use down to 23-weeks gestation (22).

Vignette Discussion

Does your unit have a consistent acute pain scale used at EOL? We recommend BIIP, PIPP, or NPASS scale here given the relatively acute nature, prematurity, and use of mechanical ventilation. No one scale has been found superior, though certain scales are validated for particular scenarios such as prolonged pain or gestation.

NON-PHARMACOLOGICAL COMFORT MEASURES

Critically ill neonates frequently undergo painful interventions (16, 23). Care should be given to decrease noxious stimuli. Regularly scheduled procedures, such as bath time, can often be held or deferred; orders for obtaining vitals less often can decrease noxious stimuli (24).

Non-pharmacological neonatal comfort measures include swaddling, facilitated tucking, kangaroo care/skin-to-skin care, non-nutritive sucking with or without oral sucrose, and breastfeeding (16, 25, 26). Each of these methods has been shown to increase comfort or decrease oxidative stress markers of neonates in situations of acute pain. In fact, combining measures enhances comfort over using a single method alone (5, 27).

PHARMACOLOGICAL MANAGEMENT

Non-opioid mediations are recommended for mild pain, while opioid medications with or without adjuvant therapies are used for moderate to severe pain (16, 24). Efficacious administration routes include enteral, intravenous (IV), mucosal (buccal, lingual, or rectal), intranasal, transdermal, and subcutaneous depending on the agent. Agents may need to be combined for optimal comfort (5, 16). See **Table 1** for initial dosing guidelines and **Figure 1** for a general guide to stepwise approach for providers.

The principle of "Double Effect" is an important consideration for clinicians concerned about the use of pharmacologic agents for EOL care. The principle asserts it is morally permissible to unintentionally cause harm as a side effect if the original overarching intent is to bring about a good effect. Extrapolating this principle to EOL care explains the use of medications that might potentially cause respiratory depression given the intent is to relieve pain and suffering (28, 29).

Non-opioid Medications

Sucrose is effective in reduction of pain behavior during acute procedures and noted to have increased efficacy when used in combination with nonnutritive sucking (5, 16, 30).

Although acetaminophen does not show analgesic effectiveness for reducing effects of painful procedures in neonates, it does show an opioid sparing effect (31, 32). Acetaminophen should only be used to treat mild pain or as an opioid sparing agent. Side effects include liver toxicity, although this is less common in neonates than older children (33).

Literature examining nonsteroidal anti-inflammatory agents such as ibuprofen, ketorolac, or indomethacin for neonatal EOL care is lacking and therefore not generally recommended.

Opioids

Opioids should be first-line therapy in management of neonates with moderate to severe pain. Route of administration should take into consideration onset of action, duration of action, and most efficacious route for desired opioid.

Morphine has been shown to be less effectively absorbed when used mucosally due to less lipophilicity than other opioids like fentanyl (34, 35). Side effects include urinary retention, decreased gastrointestinal motility, nausea, vomiting, hypotension, and respiratory depression. Respiratory depression is always preceded by sedation (16, 36).

Benefits of fentanyl include fast onset of action and efficacy of intranasal administration due to high lipophilicity (16, 34).

TABLE 1 | Neonatal pharmacologic agents for end-of-life palliative care with initial doses*.

	Enteral or Mucosal	**Intravenous**	**Other**
NON-OPIOIDS			
Sucrose	Buccal/Lingual: 0.1–1 mL/dose every 2 min		
Acetaminophen	PO: 10–15 mg/kg every 6–8 h PR: 15–20 mg/kg every 6–8 h	7.5–15 mg/kg every 6–8 h	
OPIOIDS			
Morphine	PO/PR/Buccal: 0.1–0.2 mg/kg every 3–6 h	Bolus: 0.05–0.1mg/kg every 2–4 h Continuous: 0.01–0.1 mg/kg/h	SC: 0.1–0.2 mg/kg every 3–6 h
Fentanyl	Buccal: 1–2 mcg/kg every 1–4 h	Bolus: 1–3 mcg/kg every 5–15 min Continuous: 0.5–5 mcg/kg/h	Intranasal: 1–2 mcg/kg every 1–4 h
ADJUVANTS			
Midazolam	PO/Lingual/PR: 0.2 mg/kg every 2–4 h	Bolus: 0.05–0.1 mg/kg every 2–4 h Continuous: 0.03–0.06 mg/kg/h Maximum Continuous: 0.3–0.5 mg/kg/h**	Intranasal: 0.1–0.3 mg/kg every 2–4 h
Lorazepam	PO/PR/Buccal: 0.05–0.1 mg/kg every 6–8 h	Continuous: 0.05–0.1 mg kg every 6–8 h	
Dexmedetomidine		Bolus: 0.5–1 mcg/kg/h Continuous: 0.3–1 mcg/kg/h	
Clonidine	PO: 2–4 mcg/kg every 4–6 h	***	
Phenobarbital	PO/PR: 5 mg/kg every 24 h	Bolus: 5 mg/kg every 24 h	

IV, intravenous; PO, by mouth; PR, rectal; SC, subcutaneous.

**Table lists suggested doses, recommend titrating as required for symptom management.*

***Maximum sedation EOL dosing, also with anticonvulsant activity at this level which may allow other anticonvulsants to be discontinued.*

****No IV formulation available, see dexmedetomidine IV.*

Fentanyl also does not cause histamine release; therefore, it has decreased vasomotor center activity causing less hypotension than morphine. It also has a shorter duration of action than morphine. A notable adverse effect is chest wall rigidity, especially with higher doses and rapid IV administration (16).

Benzodiazepines

Benzodiazepines potentiate inhibitory GABA (gamma-aminobutryric acid) neuronal activity of the central nervous system (37). They are potent sedative and anxiolytic medications without analgesic effect, which can decrease need for opioids (16). Midazolam has rapid onset as well as short duration of action, enabling ease of titration (5). Lorazepam has an intermediate onset of action but with longer duration of action facilitating baseline sedation needs (37). Side effects include myoclonic jerking and hypotension (16).

Alpha 2-adrenoreceptor Agonists

Dexmedetomidine and clonidine are alpha2-adrenoreceptor agonists that have sympatholytic, sedative, and analgesic effects. In contrast to opioids, they do not have significant ventilatory effects. The most notable side effects include bradycardia and hypotension (5, 16).

Alternative Sedation Medications

Phenobarbital is a barbiturate that can be an adjunct medication for palliative sedation (38–40). It has sedative and anxiolytic effects without analgesia via its inhibition of the central nervous system through augmenting the GABA system. The preferred route in this context is oral due to slow absorption causing less severe side effects (5). Dosage when used as an adjunct sedative is much less than for seizure control in neonates (see **Table 1**). Clinicians should use caution if combining with opioids or benzodiazepines as this may increase the risk of respiratory depression. Pentobarbital is another barbiturate with shorter half-life than phenobarbital but is not well studied in end-of-life care for neonates (16).

NMDA-Receptor Antagonist

Ketamine is an N-methyl-D-aspartate (NMDA)-receptor antagonist used to decrease central sensitization to painful stimuli and perception of pain. Ketamine has proven effective in neonates for peri-procedural sedation and analgesia with a short duration of action. It should only be used in combination with a sedative medication due to hallucination risk. Adjunct dose to consider is 0.5–2 mg/kg/dose intravenous although use at end of life is not well-studied (16, 41).

Vignette Discussion

*What comfort medications you consider depend on distressing symptomatology for infant and family. Is the neonate already on an opioid drip for painful chest tubes, ventilator, or recent surgical procedure? At times you may only need titration for improved effect. In other cases, your main distressing symptom may be respiratory distress or secretion management without any pain issues. See **Figure 1**.*

RESPIRATORY SYMPTOM MANAGEMENT

Respiratory Distress

EOL respiratory distress may manifest as retractions, tachypnea, grunting, nasal flaring, or gasping. Non-pharmacologic treatment interventions may include positioning modifications such as elevating the head of the bed and positioning side-lying or prone. Use of a fan with air movement toward the patient's face has been shown to relieve dyspnea symptoms in adult hospice

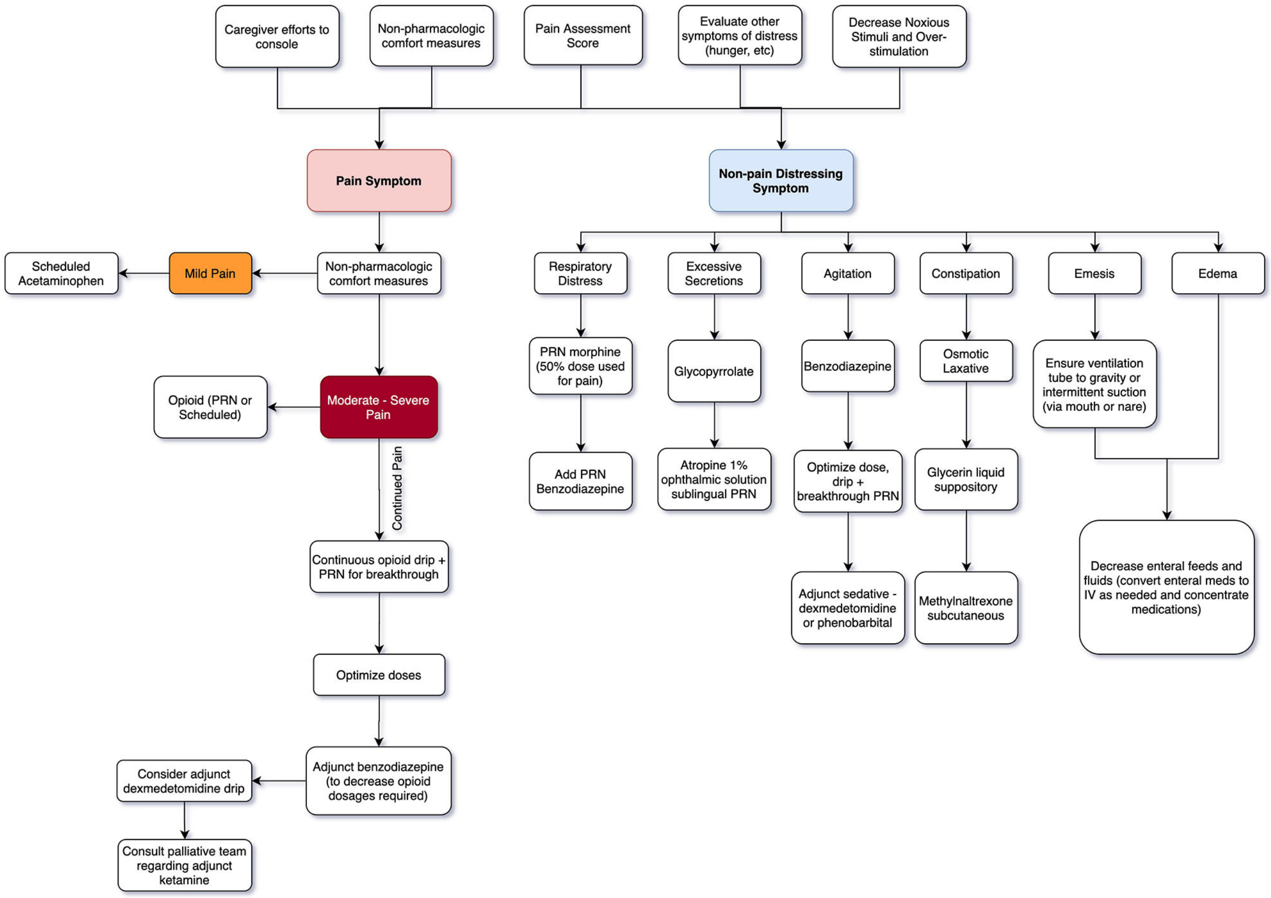

FIGURE 1 | Neonatal EOL Symptom Algorithm.

patients (42). Use of oxygen is not beneficial for respiratory distress for most patients at EOL (43, 44).

Pharmacologic management options include opioids, which can reduce central sensitivity to hypoxia and hypercapnia (45). Morphine is commonly used in neonates and is usually effective at 50% of the dose used for pain (see **Table 1**). Use of an opioid with a benzodiazepine has been shown to significantly reduce respiratory EOL symptomatology (16, 45).

Excessive Secretions

Inability to swallow saliva may lead to pooling in the posterior pharynx and noisy breathing. Anticipatory guidance with the family should be based around treating only if symptoms becomes distressing to the neonate. Least invasive non-pharmacologic treatment would first include decreasing or discontinuing hydrating fluids in an effort to decrease secretion production, positioning the infant to allow gravity to drain secretions (i.e., side-lying or prone), and gentle shallow oral suctioning with soft catheter as needed (46).

Atropine 1% ophthalmic solution may be given sublingually 1–2 drops every 1–6 h as needed (5, 16, 45, 47). Atropine inhibits local salivation via anticholinergic activity. Atropine sublingual drops have shown reasonable effect in reduction of terminal respiratory secretions while not showing cardiac or central nervous system symptoms, though most evidence is adult based at this time (48).

Glycopyrrolate is an anticholinergic that reduces gastric, pharyngeal, tracheal, and bronchial secretions. Dosing PO is 20–100 mcg/kg/dose every 4–8 h as needed. Dosing IV or subcutaneous is 2–10 mcg/kg/dose every 4–8 h as needed. If pharmacologic intervention is needed, glycopyrrolate is generally the preferred pediatric anticholinergic option as it does not cross the blood-brain barrier, so it rarely has central side effects (49). Side effects include mucous plugging due to thickened secretions.

Scopolamine transdermal patches show no increased effectiveness in secretion management and are usually avoided in neonates due to risk of central side effects from crossing the blood brain barrier (50).

Compassionate Extubation

Clinicians must be cautious in the setting of compassionate extubation or discontinuation of respiratory support in neonates. Experts in neonatal EOL care recommend initiating opioids with or without synergistic benzodiazepine and then a slow wean of ventilator settings while monitoring for development of respiratory distress. This allows for titration of medication for symptoms prior to extubation. Parents should be involved with the non-pharmacologic comfort role if able, including holding, swaddling, rocking, and positioning for secretions. Anticipatory guidance should be given to parents in monitoring for pain and respiratory symptoms. Anticipation and prevention of pain, agitation, and dyspnea symptoms is paramount to decrease familial and neonatal discomfort (5, 16).

AGITATION AND NEUROLOGIC SYMPTOM MANAGEMENT

There are multiple etiologies for agitation during EOL care for neonates, which may include brain injury, malformations, seizures, or iatrogenic neonatal abstinence syndrome due to long-term drug exposure from hospitalization. Non-pharmacologic interventions should not be employed first in neurologic scenarios such as seizures.

Pharmacologic interventions for agitation include benzodiazepines as well as opioids as they have a synergistic sedation effect. Barbiturates may also be considered (29). See **Table 1** for agitation medication recommendations. It is important to balance over-sedation and lethargy with desired control of symptoms. Seizure control should include phenobarbital 20 mg/kg/dose up to 40 mg/kg/load IV. Medical consultation with your pediatric neurologist can guide further anticonvulsant recommendations.

NUTRITION AND GASTROINTESTINAL DISTRESS SYMPTOM MANAGEMENT

Nutrition

An important component of neonatal end-of-life care is addressing artificial hydration and nutrition as well as natural hydration/nutrition. There is no reason to preclude breastfeeding or bottle feeding as part of comfort during EOL care if the neonate orally feeds and doing so does not exacerbate distress.

However, discussions around withdrawal of artificial hydration and nutrition present a challenge in neonatal EOL care. Culturally, many parents attach meaning and symbolism of caring for their neonate with nutrition. Using clear language during discussions is of utmost importance (16). Receiving artificial hydration and nutrition may not be in the best interest at EOL as it may worsen symptomatology through fluid overload, respiratory distress, increased abdominal distention, and discomfort or nausea and vomiting (5). Addressing subsequent side effects of dehydration is also important, for example, oral hygiene (see Mouth Care section below) (16).

Constipation

Opioid induced constipation is multifactorial including decreased gastrointestinal motility, inhibition of mucosal transport of electrolytes and fluids, as well as interference with the defecation reflex (16). The risk of opioid-induced constipation increases with duration of opioid therapy. Laxative treatment such as polyethylene glycol is considered first-line with neonatal doses of 0.2–0.8 g/kg/day (51–53). Liquid glycerin suppository 0.2 mL per rectum may be given in addition to scheduled osmotic laxative.

A last resort treatment consideration is methylnaltrexone for reversal of some opioid side effects without precipitating withdrawal. Methylnaltrexone is a peripheral acting mu-opioid receptor antagonist that has emerging data in neonates. Neonatal dosing used is 0.15 mg/kg subcutaneous once daily until bowel movement occurs (54, 55).

SKIN, EYE, AND MOUTH CARE

Physiological changes during EOL may compromise the skin and soft tissues manifesting as changes in skin (color, elasticity, or integrity) or as subjective symptoms such as pain or itching (56). Areas of decreased cutaneous perfusion may exhibit as dusky erythema, mottled discoloration, or areas of localized cooling. Care should include skin assessments and prevention of excessive pressure, friction, moisture, and immobilization during EOL care. Clinicians should monitor for incomplete eyelid closure as this increases risk of exposure keratopathy, which might cause ophthalmic pain and sensitivity (57). Care of mouth xerostomia should also be part of EOL assessment care.

Evidence based treatments for neonatal EOL skin care are lacking; however, pediatric studies show topical emollients may aid in reduction of cutaneous dryness (58). If incomplete eyelid closure is noted, artificial tear ointments have shown to be helpful in multiple intensive care settings (59). Moist sterile water swabs work well for neonatal xerostomia care (60, 61). Topical petroleum jelly to dry lips may be helpful as well (60). Parental involvement in symptom assessment has been shown to be a predictor of quality EOL symptom treatment (62). If counseled appropriately, these skin, eye, and mouth changes can easily be assessed by parents and treated as needed (63).

FAMILIAL EXISTENTIAL DISTRESS AND PSYCHOSOCIAL CARE

Providers should be mindful of the propensity for family members to feel enormous guilt and question their decisions. Many families will be in existential crisis and openly question why this is happening. This part of the grief process should be normalized. Some parents may need reassurance that they are in fact a good advocate for their baby while also being present for their neonate's EOL (11).

Some families have moral distress in believing that withdrawing technological support is not permissible in their cultural or religious customs. Reframing discussions in a way that the family does not have to decide anything may be helpful. For example, the physician may say something like "Additional technological support is inappropriate and will increase the infant's suffering. I recommend we increase the palliative care services and work intensively now to support comfort for your infant" (11).

Helpful discussions for family may include openly asking about religious rituals such as baptism that may be important for closure. Consultations to pastoral care or clergy that align with their religious preference may be of comfort during EOL transition. Consider involvement of social work, child life, and palliative medicine if available at your hospital and circumstances allow. Developing a unit checklist for bedside nurses may aid families in obtaining memories with mementos such as a lock of hair, hospital name bands, crib cards, and photographs (10, 11). Families should also be given bereavement and counselor contact information for the area to aid the grief and healing process after perinatal loss (10, 64, 65).

Anticipatory guidance is an important part of the bereavement process for women who are lactating. Consider consulting your lactation specialist. Helpful methods of care may include the use of medications such as estrogen containing birth control pills, a process of decreasing frequency of milk expression without completely emptying the breast, or milk donation for a time (66, 67).

INSTITUTIONAL NEONATAL EOL CARE GUIDELINES

Most institutions across the United States do not formally address neonatal EOL care in a guideline format (1). However, implementation of clinically practical guidelines increases consistency in patient care as well as education and comfort of staff/trainees (4, 38, 68, 69). Neonatal EOL guidelines should be individualized by institution and case to case as necessary.

DISCUSSION

One of the most difficult parts of neonatal EOL care includes discussion with family. Acknowledgment should be a tool we use with our families, staff, and ourselves. Acknowledge that EOL is sometimes unfair and difficult, and unimaginable even. Acknowledge it is natural to want to lessen the blow of information you have to discuss as the provider, despite research showing this does not help (70). Acknowledge the difficulty of family needing to digest what you discuss. Share with the family how much you wish the situation were different. Speak information as clearly and succinctly as possible. Answer questions, attempt to anticipate questions, and normalize their concerns (71).

EOL care is a multifaceted approach including identifying and treating EOL symptoms while also addressing familial existential distress and psychosocial care (1, 5–7). Implementation of a guideline for neonatal EOL care at your institution can aid in improved EOL processes (4). Consulting with neonatal palliative care teams, if available, can add important multidisciplinary support that enhances neonatal EOL care (8, 9). Increasing neonatal EOL care palliative education with trainees, nursing staff, and colleagues at your institution can increase comfort and competence with EOL care (72). In all, EOL care is a sacred and deeply meaningful part of life around the world. By assessing and addressing symptoms as well as promoting a culture of respect surrounding familial or spiritual traditions, we can help families find closure with a dignified and peaceful neonatal EOL process (73).

AUTHOR CONTRIBUTIONS

SH collected and analyzed data as well as primarily wrote the manuscript. AD and SD critically reviewed and contributed to the final draft of the manuscript. All authors contributed to the article and approved the submitted version.

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REVIEW
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Fostering Vicarious Resilience for Perinatal Palliative Care Professionals

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Background: The demands on healthcare professionals caring for families grappling with a life-limiting condition in an unborn or newly born child can be overwhelming. Clinicians working in emergency/trauma, hospice, and pediatric settings are already at high risk for burnout and compassion fatigue, which can leave healthcare institutions increasingly vulnerable to poor retention, absenteeism, and waning quality of care. The provision of exemplary palliative care requires a cohesive interdisciplinary team of seasoned professionals resilient to daily challenges. In September 2019, the American College of Gynecology, in a committee opinion, published standard of care guidelines for perinatal palliative care. This has created an impetus for exceptional caregiving and a greater demand for both physician and interdisciplinary healthcare provider education, training, and ongoing support that promotes truly beneficent care for pregnant patients confronted with life-limiting fetal conditions.

Methods: A scoping review of the research literature was conducted in order to distinguish the barriers and facilitators of professional resiliency in perinatal palliative care. PubMed, Medline, CINAHL, and EBSCO Psychology & Behavioral Sciences Collections were systematically reviewed. Because of the paucity of studies specific to perinatal palliative care, several interviews of nurses and physicians in that field were conducted and analyzed for content distinctly pertaining to personal practices or workplace factors that support or hinder professional resiliency.

Results: The research indicated that medical professionals often cite a lack of knowledge, inexperience using effective communication skills related to perinatal palliative care and bereavement, challenges with interdisciplinary collaboration, misconceptions about the role and function of palliative care in the perinatal or neonatal settings, moral distress, and workload challenges as encumbrances to professional satisfaction. Strategic implementation of facility-wide bereavement care training, effective communication modalities, and evidenced-based practical applications are critical components for a thriving perinatal palliative care team. Authentic formal and informal debriefing, peer mentoring, adequate caseloads, robust provider self-care practices, exceptional relational efficacy, and cultural and spiritual humility can foster personal growth and even vicarious resilience for perinatal palliative care professionals.

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Conclusions: Support should be strategic and multifaceted. The onus to implement salient measures to cultivate resilience in the perinatal palliative caregiver should not be only upon the individuals themselves but also upon prevailing regulatory governing bodies and healthcare institutions.

Keywords: perinatal, palliative, compassion, burnout, ethical, self-care, grief, resiliency

INTRODUCTION

Perinatal palliative care professionals have an integral role in the decision making and care delivery for many families encountering uncertain perinatal outcomes, some of which may have a reverberating and lasting impact. Palliative care champion Byock (1), in his 1997 book *Dying Well*, described how the time preceding a death can be a time of personal and spiritual growth for many. Researchers Côté-Arsenault et al. [(2), p. 12] said that "contrary to common societal reaction where focus is on what is wrong with the baby, [the study participants] focused on everything positive about their babies." The parents also said that their participation in the research study was a positive experience and added meaning to their children's short lives (2). Nurses in a study that explored their reactions to perinatal death revealed that when adequately supported, "growth and transformation" emerged from "the anguish and grief" (3). This article will systematically explore ways that professional caregivers can glean a fecundate resilience and vicarious personal growth sojourning with their patients who are enduring a life-limiting fetal diagnosis.

The loss of a child in infancy can be a profoundly traumatic experience. Psychologists Jaffe and Diamond (4) described in their 2011 book, *Reproductive Trauma: Psychotherapy with Infertility and Pregnancy Loss Clients,* the hopes and dreams that individuals envision for their future family long before conception. They also portrayed the trauma that can occur when those hopes and dreams go awry. The psychological ramifications when one's fundamental beliefs about themselves and their offspring are disrupted by a life-limiting antenatal diagnosis can be devastating. Lang et al. (5) demonstrated in their research that the death and dying of unborn or newly born children, though culturally disenfranchised, is as emotionally painful as the loss of an older child or adult family member. The researchers also concluded that because of the lack of social support, intrinsic to disenfranchised losses, parents' emotional reactions to perinatal losses tend to be particularly traumatic compared to more traditional types of losses (5). Doka [(6), p. 5] described disenfranchised grief as an experience that "is not openly acknowledged, socially validated, or publicly observed." The birth and/or untimely death of an infant and the associated grief reaction is outside of typical grieving norms, and parents can find themselves on an unworn path of bereavement.

The emotional impact on families confronted with a life-limiting fetal illness is often complex. At the time of diagnosis, parents may experience a variety of emotions including fear, shock, anger, anxiety, sadness, guilt, and even jealousy of other parents with healthy babies (7). The risk for severe psychological distress including depression, anxiety, and obsessive-compulsive behaviors was shown to be as much as five times higher in parents with a very low birth weight infant in comparison to parents with normal birth weight babies (8). In addition, some families have to cope with financial strain, time away from other family members, long commutes, and travel expenses (8). Furthermore, a couple planning a delivery or termination of a fetus with a life limiting condition is likely to encounter disagreements about their decision from healthcare professionals, family members, and friends. In the absence of multidisciplinary planning and support, the antenatal and postpartum bereavement has the potential to be fraught with ambivalence, shame, secrecy, and isolation. Those involved in the care provision may also find that, in the absence of personal resiliency practices, the persistent emotional strain is unsustainable (9, 10).

Palliative care teams, comprised of multidisciplinary professionals serving inpatient and home environments, are a relatively new concept in medical practice, particularly in pediatrics, and perinatal care. The team usually consists of two or more interdisciplinary professionals with complementary skills who collaborate and move toward common focused goals in the delivery of care (11). Team members often depend on and hold each other accountable for different complex and mutable aspects of palliative care. Perinatal teams are structured like adult and pediatric palliative care teams, which usually include medical care providers, social workers, chaplains, respiratory therapists, and lactation consultants. It is recommended that nurses and physicians in these teams be trained in obstetrics and neonatology (12). Because of improved technology and the earlier diagnoses of genetic anomalies and lethal malformations over the past four decades, expectant couples are now being counseled about the health of their unborn child earlier in the pregnancy. Since the onset of antenatal prognostication, the option of pregnancy termination was initially purported as the only measure to avoid suffering, and comfort-focused alternatives were not developed nor presented as a modality of care (13). Now, couples are being offered a broader spectrum of care that includes palliative and hospice provisions for their unborn (14). Thus, frequent acquiescence of the palliative professional's approach to varied patient care situations is essential, and enduring occupational resiliency is indispensable.

METHODS

A scoping review according to the methods described by (15) was used to explore and amplify the key factors necessary for promoting professional stability in the perinatal palliative setting (**Figure 1**). The scope of practice for perinatal palliative care is still evolving, but for the purposes of this study the field of reference was that outlined by the American College of Gynecology (14). There is a body of research that explores various aspects of self-care and supportive measures to prevent compassion fatigue and burnout in healthcare providers in pediatric and palliative settings, but there is a paucity of studies specific to perinatal palliative care. The search included systematic reviews, qualitative case studies, and quantitative survey studies that had been published in the past 11 years (January 2009–January 2020) (**Figure 1**). Searches were made in PubMed, Medline, CINAHL, and an EBSCO Psychology & Behavioral Sciences Collection. Search words included *perinatal*, *neonatal*, and/or *palliative* combined with the words *burnout*, *compassion fatigue*, or *resilience*. Relevant studies were identified from screening summary and abstracts. Full articles were analyzed for components associated with professional caregiver burnout, compassion fatigue, resilience, and satisfaction (**Table 1**). In addition, semi-structured interviews were conducted (**Table 2**). The participants included four registered nurses and two physicians involved in various perinatal palliative and hospice care programs in the following US cities: San Diego, CA; New York, NY; Denver, CO; and Flagstaff, AZ. The interview questions were emailed to the perinatal palliative professionals who had agreed to participate. They either composed and returned written responses or answered the questions in a phone call conducted by the primary author, which were then transcribed. The responses were aggregated and then the content was analyzed for facilitators and barriers to professional resilience by two coders who have graduate level education in nursing or psychology (**Table 3**). There was no specific funding utilized for the conduction of this study.

RESULTS

Database searches revealed 4,247 studies. Initial review and removal of duplicates, unavailable full-text article, marginal relevance to perinatal palliative care, and language barriers reduced to the search to 123 articles. Another 102 were removed because they did not report primary research. Therefore, 21 articles were selected. The literature review process and content analysis of the interviews yielded the following factors that contribute to distress in neonatal and palliative care professionals: challenges associated with both multi- and interdisciplinary collaboration, gaps in training, moral and ethical issues, misconceptions about the role and function of palliative care in the acute setting, and unfeasible workloads. Alternatively, components that supported resilience and career satisfaction included more comprehensive and interdisciplinary bereavement care training, formal and informal debriefing, supportive measures to strengthen teams and care delivery models, individualized self-care measures, and a humble approach to care provision that included both cultural and spiritual humility.

DISCUSSION

Palliative Care Collaboration

Professionals agree that initiating palliative care and education to the family as soon as possible after the devastating diagnosis procures more salient comfort measures, which, in turn, enhances the significance of the care received (16). Care demonstrated to be effective and relevant can improve caregiver satisfaction and resilience. A registered nurse and clinical coordinator of the Neonatal Comfort Care program at New York's Presbyterian Morgan Stanley Children's hospital, Fran McCarthy, recalled a recent case involving a mother who had just received the initial introduction with the comfort care program and staff when she delivered prematurely. A birth plan and end-of-life options and procedures had not yet been reviewed, nor had there been time to develop a trusting alliance. Consequently, the attending medical resident and nurses were distressed about the despondent mother's demands for continued and prolonged resuscitation measures that were not typically indicated for an infant born in the presented condition. The difficult timing issues resulted in a knowledge gap that, in turn, caused parental and staff distress. In contrast, McCarthy recalled that some of the cases she considered to be most optimal had been those in which the staff and the families had adequate time to prepare and nurture a reciprocal relationship prior to the birth of the child. Karen Christie, Registered Nurse Pediatric/Perinatal Case Manager at The Elizabeth Hospice in San Diego, also recalled that more affirming bereavement experiences occurred for families who had received maximum in-depth training and antepartum palliative care planning. In regard to one memorable encounter, Christie said, "I appreciated having continuity and the experience of being part of a spiritually uplifting situation observing the parents' joy in the life, albeit short, of their son."

Bereavement care initiated soon after an ominous prenatal diagnosis can address the anticipatory grief that parents often experience and help them cope with a loss that is typically multifaceted (17, 18). Perinatal palliative care teams aim to mitigate the trauma of disenfranchised grief through supportive modalities, ongoing education, and active care for patients and their families throughout the perinatal and bereavement periods (12, 19). In several studies (2, 20–22), Côté-Arsenault demonstrated how professionals providing effective family-centered palliative support can potentially optimize the parents' adjustment to the painful event. Many parents also reported feeling fulfilled and even joyful to have had the short time to parent and love their severely ill children (2, 21).

Moral, Ethical Issues, and Misconceptions

Palliative, neonatal, and obstetric care teams have had to become increasingly flexible to varying patient values and desires for the trajectory of their pregnancy. The increasing scope in the range of care delivery options has not occurred in the absence of moral distress experienced by those providing the care (23–25). When interviewed about the barriers and challenges to keeping a cohesive and effective palliative care team together, the medical director of the Perinatal/Pediatric Hospice Program at The Elizabeth Hospice and a family practice in San Diego,

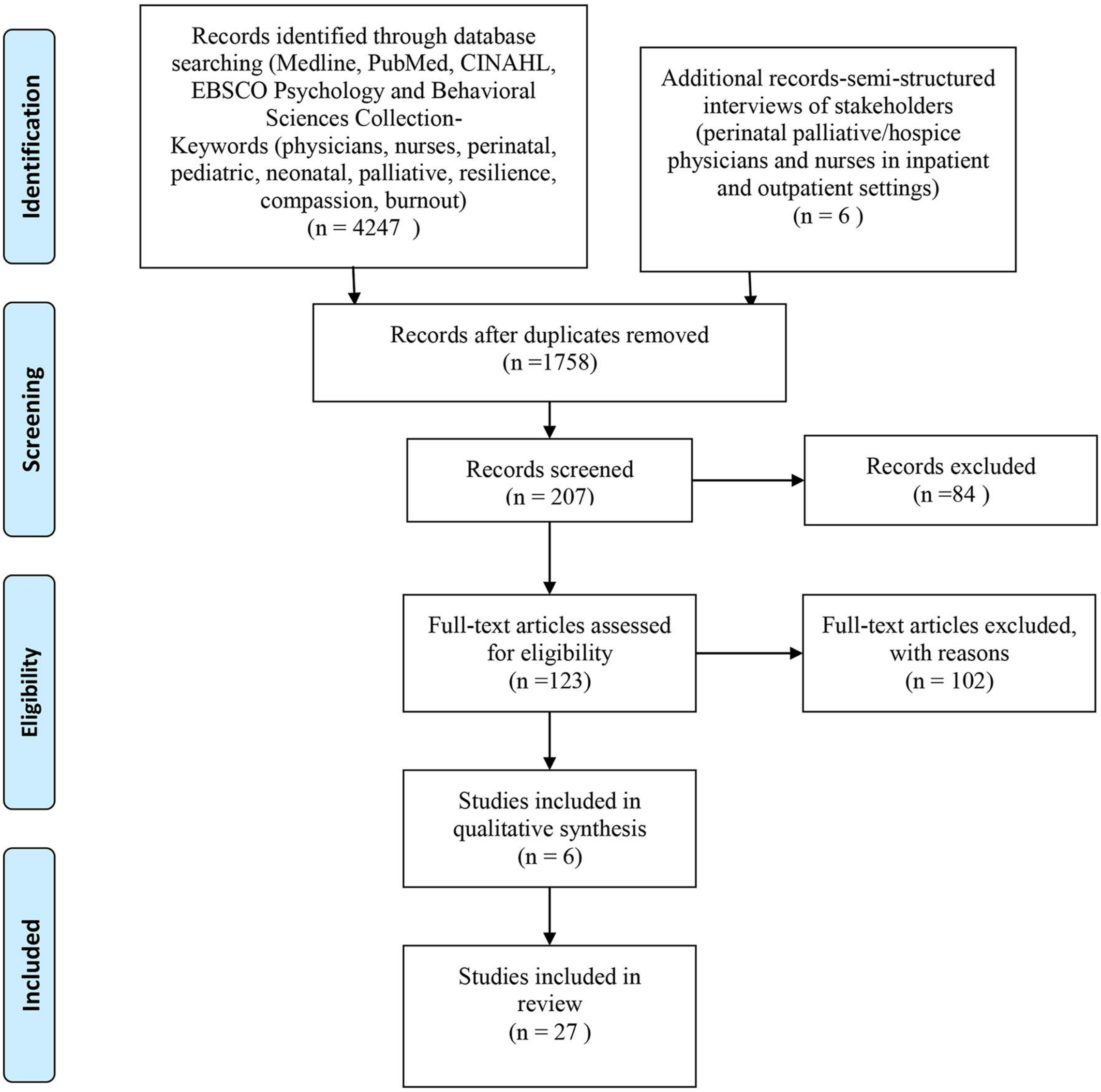

FIGURE 1 | Flow diagram of article section process.

Dr. George Delgado, said the "challenges include emotional stress, compassion fatigue, ethical dilemmas, and cost-cutting across healthcare." From his experience most of the conflicts are ethical, extending from contrasting societal philosophical axioms influencing healthcare, which can leave professionals in an ideological betwixt and between in the delivery of care. Ethical dilemmas not only surround the termination of a live fetus but also accompany the initiation and continuation of life-sustaining treatments that can be perceived as a prolongation of suffering and an exercise in futility for an infant bound to die (24, 26, 27). Both options are indicated as palliative options on the spectrum of services for those diagnosed with life-limiting fetal malformations by the American College of Obstetrics and Gynecology (14). Emerging perinatal palliative care programs are involved in the provision of comfort care to the neonate that neither aims to hasten death nor aggressively prolong life.

Additionally, moral distress can occur when professionals experience reluctance advocating the interests of their patients for fear of discord with the parental prerogative or institutional constraints (28, 29). Practitioners delivering direct patient care in acute situations are more vulnerable to moral distress, which can lead to burnout and staff retention issues (30, 31). Strategies for addressing and alleviating moral distress include frequent communication with the family, clarifying patient understanding of diagnosis/prognosis, engaging in ongoing goal setting, discussing the ethical issues, increasing team collaborative efforts, and using a skilled ethics facilitator for high levels of distress (23, 32).

Misconceptions surrounding the goals and spectrum of palliative care are often the most challenging aspects of its provision and collaboration amid the dominant curative culture of medicine. Caregivers may also deduce that their service

TABLE 1 | Study descriptions and details.

Author(s)	Year of publication	Aim of Study	Methodology	Findings summary
Back et al.	2016	Examine the approaches to building resilience in palliative care physicians—clinician resilience skills & workplace factors	Literature review and stakeholder interviews	Introduced a structural model of individual skills and system construction that support professional resilience
Cortezzo et al.	2019	Study of birth planning for life-limiting fetal diagnosis from both the patient and parents' perspectives	Mixed methods, quantitative-descriptive & univariate analysis; qualitative-thematic analysis	Proposed eliciting parental preferences for greater participation and decision making. Physician comfort levels were adequate, but found that time limitations made communication aspects with birth planning was most unfeasible aspect
Cortezzo et al.	2015	Explore the practices and perceptions of end-of-life care in NICU—examined patient and provider experiences	Survey study- explorative, description cross-sectional evaluation	Found debriefing/closure conferences, bereavement support, and education provision was variable and suboptimal at times. Value of formal palliative care teams rather than individual consultants ascertained.
Forster and Hafiz	2015	Explore the coping strategies and perceptions of support available to professionals encountering pediatric death	Qualitative: multidisciplinary professional interviews	Personal coping strategies, peer support, family support, spiritual beliefs, relational efficacy when caring for the dying child and family all improved coping
Hamric	2014	Illustrate ethical issues and moral distress associated with end-of-life issues that occur in the NICU which impact perinatal palliative care provision.	Case study	Expounded ethical climate building with debriefing, improving communication, multidisciplinary care conferences, and resilience factors
Hutti et al.	2016	Evaluate nurses reactions to caregiving for patient experiencing fetal demise	Recorded group discussions structured by Swanson's Theory of Caring was analyzed	Risks for compassion fatigue were revealed. Coping strategies and interventions based on latest research was presented which included debriefing, mentoring, more perinatal bereavement education, and self-care practices were implicated.
Jonas-Simpson et al.	2016	Explore thematic patterns of obstetrical or neonatal nurse's grief experiences to perinatal death	Qualitative study: interpretive phenomenology	Thematic patterns included: Growth and transformation emerging with anguish and grief, collegial supports, education, aspects of reciprocal mentorship, the intrinsic value of compassionate caregiving
Kamal et al.	2016	Examine the prevalence and predicting factors associated with burnout rates in hospital and palliative care clinicians in the U.S.	Quantitative: Survey study and statistical analysis	Higher rates of burnout correlated with emotional exhaustion and depersonalization, working at smaller organizations, extended work hours, age < 50, and working weekends. Activities related to interpersonal relationships and vacation use were associated with less burnout rates.
Klein	2009	Study issues of ethical dilemma and moral distress in pediatric palliative care—infants born with severe life-limiting anomalies	Case studies	Prognostication challenges, cultural, and social factors can cause moral distress in parents and palliative staff. Beliefs and aspects regarding the suffering of the infant can create emotional dilemma. Ethics and palliative care collaboration can be supportive to staff and caregivers.
Kilcullen and Ireland	2017	Present facilitators and barriers to palliative care provision in an Australian NICU	Qualitative: Thematic analysis of semi-structured interviews	Facilitators of the provision of palliative care included: cultural awareness, experienced and proficient mentors and supportive staff, clinical skills and knowledge, personal knowledge of one's own values, morals, & beliefs, emotional & communication skills, and knowledge of bereavement practices. Barriers to the provision of palliative care included: lack of education/inservices, workspace environment, parents' inaccessibility to infant, nurses' own grief, and included input into development of policies and procedures.
McCloskey and Taggart	2010	Probe the occupational stressors of pediatric palliative care nurses	Qualitative focus group discussions: thematic content analysis	Workload demands included: emotional, ethical, constraints to care delivery, lack of resources, documentation, community misconceptions, maintaining relationships with patient & family, and cultivating autonomy within the profession
Mehta et al.	2016	Study aspects of palliative care teams-pilot study of model for team resilience	Quantitative- Resiliency model testing with physicians	Team building measured a feasible reduction in perceived stress, and improved the perspective taking of the others in the team

(Continued)

TABLE 1 | Continued

Author(s)	Year of publication	Aim of Study	Methodology	Findings summary
Mills et al.	2017	Evaluate perceptions, education, and practices related to self-care in palliative nurses and medical professionals in Australia	Survey studies	Self-care was regarded as important by most. Many had not practiced self-care and also denied having any education on self-care. A large portion of those reported that self-care plans would be considered if training was provided.
Mills et al.	2018	Explore the meaning and practice of self-care in palliative nurses and physicians	Qualitative content analysis of semi-structured interviews	A practice of self-care contributed to a better capacity to care for others. Self-care practices in both occupational and personal settings is necessary. Facilitators of self-care practices included supportive environments, self-assessment, prioritization, and an ongoing planning process. Barriers included overwhelming workload & excessive busyness, stigma against self-care at the workplace, low self-worth or self-criticism, and lack of planning.
Perez et al.	2015	Examine the barriers and facilitators of resiliency in palliative care clinicians	Qualitative: content analysis of semi-structured interviews	Identified stressors, coping strategies, and training needs. Stressors included limited resources & cost-cutting, conflicting interdisciplinary expectations, increased caseloads, intensity of the cases, boundary setting, personal & professional limitations, competing demands from administrative & documentation activities, patients, & staff. Coping strategies included personal self-care, emotional & physical distancing from work off the clock, and social & emotional supports. Training needs included mind-body skills, stress education, cognitive stress management skills, and brief coping strategies to implement in real time.
Profit et al.	2014	Explore aspects of burnout and resilience in NICU	Cross-sectional surveys	NICUs with higher levels of burnout were correlated with a reduced teamwork climate, safety climate, job satisfaction, perceptions of management, and working conditions. A resilient culture was show to enhance safety and quality of care.
Sanso et al.	2015	Determine the relationship between self-care & awareness and development of burnout, compassion fatigue, and coping with death in palliative care professionals	Quantitative- survey study	Self-care and self-awareness improved coping with death and compassion satisfaction, less burnout and compassion fatigue. The data collection after the spiritual training exercises did not have those desired effects.
Dev et al.	2018	Evaluate the relationship between barriers to compassion and burnout in nurses.	Quantitative study: survey	Barriers to compassion, contributors to burnout included workload, clinical barriers, practice of self-compassion, encounters with patient and families
Weitraub et al.	2016	Contrast physician burnout, compassion fatigue, and compassion satisfaction	Quantitative study: linear regression analysis	Overlapping of burnout and compassion fatigue found. Impacts emotional well-being and professional performance of neonatologists. Self-identifying as being Hispanic debriefing and utilization of pediatric palliative care services resulted in compassion satisfaction.
Wright et al.	2011	Examine the facilitators and barriers to the provision of end-of-life care in the NICU settings from the perspectives of neonatal nurses.	Quantitative: Questionnaire survey study	Five barriers were identified as: inability to express values and beliefs regarding palliative care, environmental constraints, engagement in technology, demands from parents, and lack of education. Eight facilitators identified as: staff support, involvement of parents in decision making, support from medical team when palliative care was implemented, parents informed of options, staffing, time allotment with dying baby, policies/guidelines that supported palliative care, and available counseling.
Zwack and Schweitzer	2013	Analyze stakeholder interviews regarding aspects of burnout and resilience	Qualitative study: Concept analysis	Both attitudes and strategies (social, mental, and physical) assisted physicians' success to augment decision making, coping, and promoting resilience in others.

TABLE 2 | Interview questions based on initial research review.

Please avoid including any patient identification information that would violate HIPAA regulations.
1. Tell me about your experience as a healthcare professional involved in caring for pregnant patients confronted with life-limiting fetal conditions?(Certification, length of time, positions)
2. Are there any particularly memorable experiences when you felt the care received by the patient and the child was at its best?
3. Can you recount an experience when you felt that the care and/or prognostication process were less than ideal for the situation?
4. How often does it seem like there is one or more members on the palliative/hospice care team that are in disagreement with the plan of care? (An estimate of average amount of time)
5. Have there been times when you disagreed with the plan of care and felt morally conflicted by the care given? Do these situations occur more often with particular diagnoses rather than others?
6. What types of interdisciplinary issues seem to cause the most conflict in creating and carrying out a plan of care?
7. Does it ever seem like residue of past distress over patient care experiences effects the quality of care that you provide now?
8. Does it seem difficult to keep a cohesive and effective palliative care team together? What do you think are the barriers or challenges?
9. In what way have you changed as a provider than you were when you began providing perinatal palliative care/hospice?
10. Is there someone who was a mentor to you when beginning this type of work? Now?
11. What did the mentor do that was helpful to your professional development?
12. Do you have particular self-care practices that are helpful in managing work related tension or distress? How long have you been practicing these?
13. Does your employer provide any supports to well-being at work or for off-work hours?
14. Do you have any ideas about methods of self-care that could be provided in the work place or by your employer that would be beneficial to employee well-being?

efforts are marginalized when the modality of palliation is disparaged within the healthcare system. Despite decades of research, misunderstandings and erroneous beliefs surrounding the value of palliative care, particularly in the inpatient setting, have been pervasive. Grunauer and Mikesell [(33), p. 3] listed the more prevalent misconceptions regarding the implementation of palliative care in intensive care units as:

> [It] is (1) ineffective and unimportant for most ICU patients, (2) synonymous with hospice and hopeless on the part of the family, patients, and/or clinician, (3) equivalent to the "soft skills" that health-care professionals already innately have, and (4) wasteful in that it absorbs valuable time and resources from intensivists.

However, they went on to state that none of these misconceptions were supported by research. In fact, multiple studies show that initiation of palliative care in a timely manner can improve symptom control, professional communication, the patient's perception of their quality of life, and even lengthen survival times (33). Dispelling the myths about palliative care and holistic care for neonates and infants with life limiting conditions and their families can be accomplished with ongoing education provided to the entire caregiving staff within each unit (18, 33, 34). More research and widespread education can enhance the appreciation of palliative care, which can foster a benevolent environment. Dr. Elvira Parravicini, M.D., neonatologist from Columbia University Irving Medical Center, said that "most obstetricians have come along to supporting us after seeing the benefits and appreciation from the patient of what we have done for them."

Professional Strength

Professional caregivers experiencing distress and high levels of stress have been associated with the development of cynicism, depersonalization of patients, diminished professional satisfaction, impaired decision making, medical errors, increased rates of adverse events, and higher mental and physical morbidities (9, 10, 35). Compassion fatigue or secondary traumatic stress disorder can also occur when empathetic caregivers experience, through a phenomenon known as countertransference, the trauma of their patients. Figley (36) coined the term *compassion fatigue* when he examined his own reaction to caring for military veterans debriefing from their traumatic combat experiences. He (36) said this regarding the personal anguish he encountered: "We feel the feelings of our clients. We experience their fears. We dream their dreams. Eventually, we lose a certain spark of optimism, humor and hope. We tire. We aren't sick, but we are not ourselves."

Palliative and hospice professionals have higher rates of burnout and compassion fatigue than most other specialists with rates averaging as much as 61% in some studies (9, 37). Increased levels of distress and burnout can lead to absenteeism and negatively impact the palliative care team (38). Efforts to mitigate burnout and compassion fatigue are not without significant benefits for both the patient and the caregiver (39). Long-term job satisfaction can also flourish from relational efficacy (40).

Fostering Resilience

Hernandez et al. (41) revealed that vicarious resilience occurs when professional caregivers experience their own growth through the provision of care and witness of patients enduring and recovering from traumatic experiences. Author van Dernoot Lipsky [(42), p. 11] wrote, "Those who support trauma stewardship believe that joy and pain are realities of life, and that suffering can be transformed into meaningful growth and healing when a quality of presence is cultivated and maintained even in the face of great suffering." Therefore, a self-assessment to determine one's level of compassion fatigue or professional satisfaction is essential. The Professional Quality of Life Measure (ProQol) is a tool found to be useful for this appraisal (43). Self-care has been found to be a key aspect in developing resilience and maintaining an instrumental presence in the midst of suffering and loss (10, 35, 44, 45).

Resilience is an individual's ability to cope with adversity and maintain a healthy response to stressors (46). Effective coping and resilience can be nurtured by participation in a variety of

TABLE 3 | Pertinent, categorized quotations from palliative professionals' interviews.

Palliative Care Collaboration

"The family and staff became well acquainted with the [palliative] goals of care and the relationship with the palliative established prior to the delivery."

"A patient with a complex relationship with [her] partner was recently handled well with multi-disciplinary support we had available for her."

"The social worker continued contact and visits with family, providing psychosocial support, birth plan guidance and memory making tools."

"The baby was stillborn, and parents' needs were met, however I felt they would have had more support if they had let the team be more involved."

"We have a great deal of respect for each other. We have extreme trust in each other. We all are in agreement about the mission and vision of our program."

"Most professionals in other disciplines (speech pathology, lactation consultants, chaplains, and child-life therapist) admire the work of our program, moreover there are some who are very comfortable with palliative care and help us a lot facilitating comfort for babies and support for parents. I believe that no professional in our institution has been in conflict with our program. Most obstetricians have come along to supporting us after seeing the benefits and appreciation from the patients of what we have done for them."

Moral, Ethical Issues, and Misconceptions

"The mother was a nurse and asked for oxygen and pain meds to prevent her baby from suffering. Although the healthcare team knew this to be unnecessary nor beneficial, we accommodated the mother's request to empower her during this time. By doing so, she was able to have the motherly control to help her during the grieving process."

"More disagreements occur with other specialties like maternal-fetal medicine physicians or when staff members are not understanding role of perinatal palliative care."

"[Conflict] often occurs because medical professionals feel compelled to suggest abortion whenever there is an anomaly."

"Sometimes the parents choose not to feed the baby, even if feeding might be tolerated."

"Sometimes other professionals have questioned our plan of care because they have a different mindset and are not familiar with a palliative care approach. However, when they see the results, the comfort of the baby and parental satisfaction, they come along and they want to learn more about it."

"Mostly the conflicts are ethical. Many team members are utilitarian in their ethical approaches, not out of malice but due to the prevailing utilitarian ethic in medicine."

"There was one situation where the baby was born severely malformed and the parents had the school age children take pictures with baby who did not have severe brain malformation covered. My concern was the internal difficulties the children may have had with viewing the malformation, although externally they presented as accepting."

"In many institutions, babies with life-limiting conditions are not fed at all, we prefer to offer food for comfort, unless the baby is in end of life stage. The experience of feeding their baby is very important for parents, we had babies who lived for only a couple of days successfully breast-fed. We also strongly believe that is comfortable to end life with a 'full stomach'."

"Sometimes bedside caregivers are morally conflicted and concerned about the futility of care and suffering."

Professional Strength

"I think each patient's experience is different. Their needs are catered differently. Providing that compassion for the families actually allows me to use my vocation as a nurse to my utmost ability. I think that's when my quality of care is the strongest."

"Occasional vacancies are difficult in the neonatal intensive care unit and palliative programs."

"I have become more open minded and attentive. I look at each single patient and look to them to tell me through objective signs and cues as to what to do for them. It is a tremendous gift to learn from my little patients and from their parents."

Educational Strategies

"Field staff are offered presentations and webinars including those on compassion fatigue and burnout."

"Bedside staff [report] not feeling comfortable with nature of palliative care. [It is because of] a lack of training and experience. [They are] used to curative goals."

"The perinatal palliative program was difficult when it was located on the postpartum unit, because the nurses had a hard time transitioning to a palliative approach."

"Babies die differently than other children and the outpatient hospice nurses not always educated to the differences. For instance a baby with trisomy 18 will have a different breathing patterns than typical counterparts."

"My previous experience taking care of hospice patients or elderly as they passed was quite different than taking care of families that lost their babies. The grieving seems to have different, complex elements to it."

Formal and Informal Debriefing

"Our team works very well together and we have discussions, planning, and debriefing as a team. We are friends, a tight, knit group, and are not afraid to express concerns to each other."

"Debriefing is a huge benefit to me. I find that debriefing immediately after a case before going home helps me to relieve that pressure in my heart and mind. It helps me learn from mistakes and set goals for myself for improvement. I also like to call a friend that understands my line of work to debrief with."

"Personally, I think a standardized debriefing practice after unexpected events or difficult situations can promote/be connected to self-care. It is possible that until a situation is debriefed and talked through do healthcare workers really realize how they are feeling about a situation."

"The hospital provides more formal team debriefing on caseloads and challenging cases- increasing and improving these processes."

"In addition, debriefing is essential. It can be a formal debrief or decompressing with a coworker about what happened, how I feel about it, etc. The process of 'talking things through' not only helps me digest the situation, learn from it and grown as a nurse."

Care Delivery Models

"In the world of [maternity care], we are able to provide the time and compassion to help families during these difficult times. Fortunately, fetal demise or palliative care is staffed as a one-to-one care."

"The family expressed their gratefulness afterward that they were able to spend as much time with their baby prior to her death at home, and that her last days were not spent in the hospital. Parents were especially grateful for having avoided the surgery because clearly her death was associated with brain stem anomalies."

Self-Care Initiatives

"This realization can point to the need of self-care and additional supportive interventions."

"They have Reiki, chair massages, hand massages. However, these work best for the people in the office."

"Yes, they provide mental health and debriefing sessions upon request. Also, our clinical manager is very supportive in terms of allowing time to grieve or leave of absence if needed."

"My previous employer and management team were always very supportive and had definitely cared about the overall well-being of the staff. It helps that nurse managers and directors know what it's like as a floor nurse and the difficult situations surrounding [perinatal care] so they know from experience the importance of support, self-care and promoting overall well-being of their staff."

(Continued)

TABLE 3 | Continued

"[My employer] offers plenty of opportunities for employees such as arranged lunches for nurses and physicians, even nicely arranged coffee breaks. They also offer different exercise programs, yoga, and counseling services. I haven't participated in any of these, I believe because I have so much help through my team and my friends."

"[Our routine team] meetings have time for bereavement and support. Additionally, we have an employee assistance program."

"[I] exercise, garden, and do a lot of home maintenance. I am also very involved with family, grandchildren that live nearby."

"Mostly, I enjoy the friendship of many people. My friends care for my psychological needs. I also sing in a choir."

"I have been practicing self-care all of my career. However, am much more organized now in active planning of self-care. Spiritual discussions and personal involvement with an excellent small group of very close friends (most of them work in hospice) and family; owning and interacting with dogs; daily walks and stretching exercises; massage and chiropractic care; keeping up with medical and dental routine care and issues (years ago I delayed these until they became urgent). My belief is that one can only do any type of hospice and palliative care with a wide variety, and regular, self-care activities."

Cultural and Spiritual Humility

"The family was also able to sing praise and worship songs to their lovely daughter and love on her."

"I appreciated having continuity and the experience of being a part of a spiritually uplifting situation observing the parents' joy in the life, albeit short, of their son."

"We also rely on the parents to tell us what they think their child needs. They are more intuitive about their own child's needs."

"Honestly, I pray to Mother Mary a lot during my times of distress. If anyone understands what these patients are going through, it's [Mary]. I pray that she helps and guides me as well."

"[An] anencephalic baby delivered at hospital before I arrived. I baptized the baby, [as the parents were] fearing that a priest or deacon would not arrive in time. We provided comfort to the baby including time on breast."

"I keep in mind that 'I am not God'. I cannot solve everything. Our time is finite, just a drop in the ocean."

"Taking into account their cultural background also is important. Sometimes, the conflicts occur within the partnership of the parents and their cultural differences."

self-care practices such as physical activity, mindful focusing, meditation, breathing exercises, and positive interactions with colleagues and friends (47). Hutti [(48), p. 25] found that "nurses reported strength to cope primarily from three sources: faith, relationships with fellow health care providers, and their own families." Active involvement in a range of personal and employer organized supportive care strategies can augment coping, effective decision-making, and ultimately caregiver resilience in the midst of stressful workplace conditions (40). Dr. Parravinici said in regards to her comfort care team professionals, "We are friends, a tight knit group, and are not afraid to express concerns to each other. My friends care for my psychological needs. For instance, my nurse coordinator tells me things like 'slow down,' 'go home,' or 'be realistic' when I need to hear it."

A cohesive team is essential to the delivery of effective and beneficial palliative care in the perinatal/neonatal setting. Teams that have a well-defined program mission, vision, and goals that are fully embraced by each of the constituents are in an advantageous position to provide the necessary support for both individual and team resilience (11, 49). When the role of each team member is clearly delineated and in agreement with shared aspects of palliative care, the team as a whole benefits (11). Other aspects that can define the efficacy of a team include clear workload or productivity expectations, established lines of accountability, and a constructive evaluation process (49). Fostering respect and appreciation of the individual team members and the team as a whole requires evolving trust, open communication, prompt conflict resolution, and a shared movement toward common goals. Tempering factors that impair team function such as absenteeism, ongoing conflict, poor team structure, insufficient training, fear of conflict, and lack of commitment and accountability can help mitigate team erosion (49).

Studies have shown that caseloads perceived to be manageable by care providers often coincide with professional satisfaction and resilience. Keeton and colleagues [(50), p. 954] said that the "strongest single predictor of emotional resilience and personal accomplishment was control over schedule and hours worked." Burnout and emotional exhaustion have been attributed to an increased workload (31, 51, 52). Specifically, palliative care providers reported more difficulty managing increasing caseloads because of the challenges in estimating the length of time needed for the provision of individual care plans, locating the family members involved in the care, and collaborating with the other team members (47, 53). In order to address issues related to absenteeism, employee retention, and burnout, healthcare institutions and legislative entities should carefully examine staffing polices and reimbursement guidelines (9).

Educational Strategies

Lack of education and training has frequently been cited as a barrier to the provision of consistent and comprehensive palliative and bereavement support in the neonatal setting (7, 54–57). Haug et al. (55) demonstrated, in a nationwide study, that fewer than a third of institutions offer any formal training for physicians or staff on perinatal palliative or comfort care. However, research has shown that professionals exhibit better emotional coping when they have been provided with education related to bereavement and end-of-life issues (56, 58–60). For example, a strong majority (91.8%) of neonatologists involved in that study indicated that increased education in perinatal palliative and end-of-life care would be beneficial (55). Elements of self-care activity, as well as the symptoms and risk factors for both burnout and compassion fatigue, should be included in the education for perinatal palliative care professionals (7, 61). An enhanced knowledge base can increase caregiver confidence when serving patients.

A commitment to continuous learning is key to providing the best possible care for patients (62). "Staff education should be provided during orientation and then periodically throughout the course of every year" [(7), p. S30]. Palliative and bereavement care training with a multifaceted approach improves caregiver communication and confidence. Allyson McCullough, Registered Nurse and Bereavement Care Coordinator at

Bella Natural Women's Care & Family Medicine and former Bereavement Care Coordinator on an inpatient maternity ward, said, "As a new nurse, I was uncomfortable … and felt that I needed to fill the silence with saying the right thing." She went on to explain that education and experience have improved her confidence in communicating with and comforting patients.

Wilkinson and Roberts (63) demonstrated that it is most beneficial to use a range of teaching methods to nurture effective communication techniques. Utilizing experts for on-site training to facilitate group analyses of case studies, role-playing exercises, and demonstrations is an effective means of teaching professional caregivers techniques that will improve their comfort level in delivering bad news and confidence in supporting the family. According to Sweeney et al. [(64), p. 14], role-play is particularly impactful, stating:

> [It] led to increased understanding of and changes in attitudes toward key palliative care principles, interdisciplinary teamwork, and communication of bad news. There was evidence of increased self-awareness. Findings suggest that the interdisciplinary breaking bad news role-play was a [particularly] rich integrative learning experience valued by students.

Professional caregivers who communicate effectively with parents facing a life-threatening illness in a neonate have been shown to have more professional confidence and better patient rapport. Increasing self-awareness, practicing specific responses via role-play activities, improving eye contact and body language, and learning to mirror a patient's own language are key components of providing quality patient care. Boles [(65), p. 307] asserts that empathy, sensitivity, active listening, a calming presence, and advocacy are essential skills "all nurses and psychosocial staff are called to provide at various points in the patient and family's illness journey." McCullough also said, "[it] is often not what you say, but [in] the things not said—the non-verbal communication [and] body language, sitting with the family, holding their hand, crying with them, sitting in silence, and meeting them where they are." Caregivers can provide an environment conducive to emotional healing by acknowledging the individual's feelings, withholding judgmental comments, reassuring them that they are not alone, and essentially giving them permission to grieve.

Patients and families grappling with the life-limiting diagnosis of an unborn or newly born child often report feeling powerless and marginalized in their involvement of care for their child (18). The provision of practical applications to assist the family bonding with and memorializing their severely ill or deceased neonate can empower both the bereaved and the caregivers. Providing personalized care that is directed in part by the patient or parent also fosters relational efficacy and provider resilience (40). Côté-Arsenault (2) said, "Parents in [her] study chose to focus their love and attention on what they could do at the present moment and on memory making," which included memorialization, photography, and comfort measures. Examples of patient-focused care include recognizing physical cues from the baby and asking the parent to assist in determining their child's needs such as holding, skin-to-skin time, breastfeeding, and assessing for comfort, hunger, thirst, and cold (19).

Formal and Informal Debriefing

Research indicates a beneficial advantage to members of a palliative team when they all participate as a group in scheduled discussions about the plan of care, appropriateness of the interventions, and the need for ongoing intensive support (39, 66, 67). Opportunities for formal debriefing should occur after every death and can be arranged to allow for reflection, quality improvement, shared narrative from the staff, and improved interdisciplinary cohesion (67). Many care teams have found that the utilization of a facilitator at these meetings can enhance the debriefing experience. For employers, it is important to note that stress management modalities and pre-incident education improve effective coping after encountering traumatic events (68).

Informal debriefing to a trusted colleague or mentor immediately or soon after a death or traumatic experience can also assist in mitigating the impact and facilitate personal emotional recovery (50, 69, 70). Also, small groups following a standardized debriefing process can help to ensure that all aspects of the incident were appropriately handled and allow for review from multiple perspectives (68). A nurse educator for the maternal ward at Flagstaff Medical Center reported, "I find that debriefing immediately after a case before going home helps me to relieve that pressure in my heart and in my mind." She also reported that her employer had provided ample formal debriefing and mental health assistance, which she thought was indeed helpful for those caregivers needing more intense or prolonged assistance.

Care Delivery Models

An institution should consider the manner in which perinatal palliative care is integrated with each unit. Currently there are two palliative care models integrated into the healthcare system. One model comprises a core group of professionals who provide consultation and care for families meeting the criteria for palliative care. Another model of care educates and trains the entire staff to provide palliative care (71). Casarett et al. [(71), p. 653] found that "families reported better outcomes when the patient received care in a palliative unit than they did when the patient received a palliative care consultation." A pattern of favorable outcomes can fuel compassion satisfaction and foster resilience in the care provider.

In 2002, Catlin and Carter (28) deciphered that those staff members willing to provide perinatal palliative care should be the ones primarily involved in its provision rather than it being required of all in a given unit. However, Mancini (67) countered that all staff members should receive the training to care for any patient they are presented with including those needing end-of-life care. The physicians and nurses I interviewed reported that they had colleagues who had expressed their discomfort with the provision of palliative and end-of-life support and avoided assignments requiring it. A neonatal nurse from Flagstaff, Arizona, said in an interview, "having dedicated members is a challenge because that's usually not the reason why nurses went into this specialty [maternal/child nursing] in the first place … [and] it takes a toll [on the nurse] emotionally, physically, and psychologically." Her statements underscore the need for increased training in palliative and end-of-life care early on in each professional education track. Ongoing education provided to the entire staff about palliative and end-of-life care should train and equip all staff with basic communication modalities to be compassionate and comforting in their interactions with patients. In 2019, Wool and Catlin [(34), p. S28] reported that "an integrated system of care increases quality and safety and contributes to patient satisfaction. The goal for respectful caregiving throughout the entire hospital system is achievable." Professionals should feel free to disclose moral distress and receive the debriefing, spiritual, and emotional support necessary for a manageable resolution and promotion of ongoing occupational resilience (67).

In 2018, U.S. reproductive grief care experts Michaelene Fredenburg and Katie Geppert provided training for all staff members of the Ukrainian L'Viv Children's Hospital Perinatal Unit. The nurses who organized the event indicated that a high incidence of neonatal death and the staff's lack of knowledge about reproductive grief were the galvanizing motivations. The entire unit participated in the training including physicians, nurses, medical assistants, and anesthesiologists as well as social workers and a hospital chaplain. Nurses attending to the NICU patients also listened to the presentation on headsets.

Participants were introduced to research and concepts related to the unique nature of reproductive loss (5), disenfranchised grief (6), and the tasks of grieving (72). Evidence-based grief care best practices were shared as well as basic self-care ideas. Frequent staff debriefings were also recommended. After an uncomfortable amount of silence during the time allotted for questions, one of the nurses ventured to admit, "Please don't interpret our silence as disagreement, it's just that we culturally haven't been allowed to discuss this topic. Thank you for giving us the words and for giving us permission to talk to each other and to the parents of our patients." Concertedly affirmed by the rest of the staff with applause and inquiries, the medical director pledged to adopt the recommended reproductive grief care and self-care policies. Months later, the nurses who had organized the in-service confirmed enhanced patient care and improved staff morale after the training.

Self-Care Initiatives

Self-care is an imperative component to abating compassion fatigue (47, 73). One study reported that only 11% of the participating nurses and doctors practiced self-care [(74), p. 628]. Only 6% of that study's participants had an active self-care plan, and of those respondents "100% reported that they found the use of a self-care plan to be an effective self-care strategy" [(74), p. 627]. Koch and Jones [(30), p. 10] reported, "self-care is not only preventative for practitioners but can serve as a model for family caregivers about the critical importance of self-compassion and care." Earnest intention to care for oneself can, in turn, result in an enhanced capacity for empathy and the service of others. Progressive resilience can be cultivated through a concerted and intentional effort to develop, implement, and fulfill a self-care plan.

The care provider should make an inventory of aspects of their personal and work practices that contribute to both distress and wellness (40, 46). The dimensions of holistic-wellness plans are multifactorial requiring attention to the mind, body, spirituality, and emotions (47, 75–77). Initially, a plan that includes pertinent goals and methods of accountability should be strategically developed. The anticipation of potential barriers can help the individual avoid being derailed from their pursuit (78). Diligent attention to potential subversions can foster timely attainment of the desired outcomes (79). Enlisting the assistance of a supportive person(s) to partner in the interventions and process of evaluating goals can improve accountability and overall beneficence (80). Care providers find that the creation of a written plan or contract for themselves is a motivating factor for their intended endeavor.

A cadre of wellness programs are often available at larger institutions. Kase et al. [(76), p. 3] found that only 60% of pediatric physicians had participated in those programs and their reasons for not partaking in them included "inconvenience of scheduling" (45.3%), time constraints (27.2%), the preference of participants to "handle things on my own" (34.1%), and the sentiment that partaking in the activity would not be helpful (21.9%). Studies have found that "one-size-fits-all" wellness programs offered by an institution might not effectively address specific needs of their employees and professional affiliates (76, 78, 79). Parravicini concurred, stating, "What people find to be helpful for self-care is a highly individual matter. Some prefer tennis, some skiing, some a walk in the park, others getting a beer." Individualized discussions regarding the implementation of self-care strategies that include goals to be accomplished outside the work environment and continuing education can be integrated into occupational debriefing and evaluation conversations. Use of a self-administered assessment tool such as the Professional Quality of Life Assessment (43) or the Self-Care Assessment Worksheet (80) in planned intervals can assist in evaluating favorable progression toward the desired benefits (81).

A self-care plan would be ill equipped if it did not involve caring for personal losses. Caregivers cannot adequately assist others when they ignore their own bereavement, and they should not impose nor inject their personal grief experience into the care of others (82). James and Gilliand (82) observed that the palliative care provider can experience bereavement challenges when they encounter multiple meaningful losses in their domestic and occupational environments. They can find themselves overloaded emotionally and need more time to invest in adequate bereavement (82, 83). Obstacles to adequate grieving can occur when the nature of the loss is ambiguous. The absence of physical reminders, shared memories with others, and memorialization of the loss contribute to ambiguity and carrying out of the tasks associated with a typical grieving trajectory (5, 6, 72). When the grieving individual assimilates a healthier pattern of mourning losses through bereavement education and assistance "the knowledge and perspective gained from one's own growth following grief should serve as a quiet reservoir of strength" [(82), p. 461].

Cultural and Spiritual Humility

A humble approach to assess the particular relational, emotional, cultural, and spiritual needs of the patient and family receiving palliative care is essential. In the midst of the suffering inherent to neonatal end-of-life care, healthcare professionals can have a human but irrational tendency to deduce that their clients' anguish could have been avoided with more expert care (11). A reverent but curious approach with not only patients and their families but also with other consulting or attending providers can enhance open, nonjudgmental communication and collaboration (84). Physician and inspirational author Coulehan [(85), p. 206] wrote:

> [An] operational definition of medical humility includes four distinct but closely related personal characteristics that are central to good doctoring: unpretentious openness, honest self-disclosure, avoidance of arrogance, and modulation of self-interest. Humility, like other virtues, is best taught by means of narrative and role modeling. We may rightly be proud of contemporary medical advances, while at the same time experiencing gratitude and humility as healers.

In addition, Sasagawa and Amieux [(86), p. 925], researchers of multidisciplinary care collaboration, found that "both quantitative and qualitative analyses revealed that humility was viewed as an important character trait for successful inter-professional collaboration." Dr. Parravicini exemplified an unassuming approach to patient assessment when she said, "I have to look at each single patient, look to them to tell me through objective signs and cues what to do for them. It is a tremendous gift to learn from my little patients and from their parents."

Culturally sensitive care is necessary for families enduring the loss of an infant. Caregivers need to be alert and respectful to their clients' cultural, racial, ethnic, spiritual, linguistic, educational, and geographical differences. Cultural competency is a process of gathering knowledge about particular cultural groups; cultural humility is a process of inquiry and interpersonal curiosity approached with openness toward another (87). The pervasive nature of multiculturalism in the United States has made basing cultural competency on knowledge alone increasingly challenging (88). Yancu and Farmer [(89), p. e1–e2] said:

> In practice, cultural diversity is manifested in a broad range of ever-changing behaviors, beliefs, rituals, restrictions, traditions, norms, institutions, and relationships that form the basis of cultural knowledge. This makes cultural mastery something akin to trying to grab onto a cloud.

In response to the change, cultural humility is quickly becoming the preferred style to assessing and addressing the cultural needs of patients (90).

Cultural and spiritual humility are conducive to superlative interpersonal communication and relational efficacy contributing to provider compassion satisfaction (88). Nurse researcher Kalu (91) concluded that religious and spiritual beliefs can support and promote better coping for individuals experiencing a reproductive loss. In her study done in Africa, one woman said, "The midwife sat and listened to me. She was very knowledgeable, spiritual and caring. She asked me if I wanted to see the baby. I said yes and spent time with [her]. I have a good memory of [my baby]. That helped me to find ways to adjust and move on knowing that the baby will always be part of my family" [(91), p. 6]. The inference from this patient exemplifies how a provider's reserved but open communication style can support a patient's spiritual beliefs without imposing any religious maxims.

Awareness of one's personal cultural and spiritual values enhances the caregiver's ability to accommodate multicultural and multiracial differences in the patients they serve (90, 92). Appreciation and practice of one's spirituality can promote professional resilience (40, 47, 93, 94). Belcher and Griffiths [(95), p. 271] said that:

> Personal spirituality and a knowledge base to support spiritual caregiving were significant factors in hospice nursing staff members' competence and confidence in providing this intimate level of care. Respondents related a commitment to, and the ability to achieve, a level of spiritual care that was highly consistent with the spiritual needs of patients: the need for meaning and purpose in life, the need to give love, the need to receive love, and the need for hope and creativity.

Exploration of one's own cultural and spiritual background, particularly in aspects of family, childbearing, and grieving, can be a journey that enhances receptivity and caregiving satisfaction.

Research and Practice Implications

Larger scale qualitative studies would make a valuable contribution to explicit needs of perinatal palliative care providers particularly in the aspects of occupational misconceptions, moral distress, aspects of humility, and professional resiliency. Quantitative studies on the effectiveness of more widespread bereavement education on patient and professional satisfaction indicators throughout a hospital system could potentially help healthcare systems to mitigate caregiver burnout and turnover. Qualitative and quantitative studies utilized to explore the efficacy of team enhancing interventions and structured debriefing processes may also yield benefits for institutions.

Perinatal palliative care is greatly valued in the lives of those grappling with life-limiting neonatal conditions. Support for those gifted and compassionate professionals who provide the caregiving needs to be strategic and multifaceted. The onus to implement salient measures to cultivate resilience in the perinatal palliative caregiver should not be only upon the individuals themselves, but also upon prevailing regulatory governing bodies and healthcare institutions as well. The implementation of efficacious staffing, debriefing protocols, and ongoing bereavement and self-care education are also vital components to resiliency.

There is a societal trend to minimize the impact of death for those that occur within the perinatal period. It is often erroneously believed that suffering and grieving is obsolete because the unborn or newly born child has not lived long enough to establish themselves. The assumption and commentary that one can "just move on and try to have another one" is incredibly hurtful to those mourning the loss of their child. Perinatal palliative care providers support and become that benevolent community necessary for families to endure and mourn. Parravicini said (96) in an interview, "I'm most proud of the babies because they are so abandoned to our care. In fact, what is unique in our program is that we always put ourselves in the babies' shoes." She incites a professional humility that can assist perinatal palliative care providers to flourish within the setting of illness and death. "In its relational aspect, humility includes reverence or awe for the grace and strength of patients and their care-givers, a sense that the care-provider is not self-sufficient but needs the care-receiver" [(97), p. 291].

Byock [(98), p. 10] was posed the question from a woman with a terminal diagnosis, "Do I need to die well?" He went on to clarify that by promoting palliative care he had "hoped to dispel the notion that life ends the moment you receive a lousy diagnosis" [(98), p. 11]. He also profoundly concluded that, "it is possible to feel well within oneself and right with the world, even as one dies" and "that therein lies the hope for us all." [(98), p. 13] The profession of caregiving has traditionally been a trajectory of hope for the future that involves a relationship between the patient and provider. However, in palliative care the aim of care provision is not focused on curing the ailment but on bettering the course of the illness. Professional and personal strengths rise to accommodate the vulnerabilities inherent to perinatal palliative care, but that relational efficacy requires concerted fortification.

AUTHOR'S NOTE

Professionals working in perinatal palliative care are at risk for burnout and compassion fatigue because of the emotional, collaborative, and ethical demands associated with the birth and death of infants with life-limiting illnesses. Consequently, healthcare institutions with perinatal palliative programs may be more vulnerable to poor retention, absenteeism, and waning quality of care. Medical professionals often report a lack of knowledge, inexperience in using effective communication skills (i.e., fear of saying the wrong thing), and emotional anguish concurrent with compassion fatigue as reasons for their emotionally avoidant or even irreverent behaviors in bereavement care delivery. Existential suffering and moral distress are common challenges inherent to the provision of care for life limiting neonatal conditions. There is an increased need for physician and interdisciplinary healthcare provider education, training, and ongoing support that promotes beneficial palliative care in the perinatal setting. Support should be to be strategic and multifaceted. The implementation of bereavement care training, effective communication modalities, and evidenced based practical applications are critical components for a thriving perinatal palliative care team. Authentic formal and informal debriefing, peer mentoring, adequate caseloads, robust provider self-care practices, cultural and spiritual humility can foster personal growth and vicarious resilience for perinatal palliative care professionals.

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All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

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MINI REVIEW
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Decisions Parents Make When Faced With Potentially Life-Limiting Fetal Diagnoses and the Importance of Perinatal Palliative Care

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When parents face a potentially life-limiting fetal diagnosis in pregnancy, they then have a series of decisions to make. These include confirmatory testing, termination, and additional choices if they choose to continue the pregnancy. A perinatal palliative team provides a safe, compassionate, and caring space for parents to process their emotions and discuss their values. In a shared decision-making model, the team explores how a family's faith, experiences, values, and perspectives shape the goals for care. For some families, terminating a pregnancy for any reason conflicts with their faith or values and pursuing life prolonging treatments in order to give their baby the best chances for survival is the most important. For others, having a postnatal confirmatory diagnosis of a life limiting or serious medical condition gives them the assurance they need to allow their child a natural death. Others want care to be comfort-focused in order to maximize the time they have to be together as a family. Through this journey, a perinatal palliative team can provide the support and encouragement for families to express their goals and wishes, as well as find meaning and hope.

Keywords: perinatal palliative care (PPC), decision-making, pregnancy, life-limiting fetal diagnoses, decisions

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INTRODUCTION

During pregnancy, ultrasounds and blood test results in the first two trimesters provide information about the fetus' health. When these results suggest a potentially life-limiting fetal diagnosis, parents are faced with a series of decisions. The first choices involve whether to obtain more diagnostic information via invasive or non-invasive tests during the pregnancy. Invasive testing includes chorionic villus sampling or amniocentesis in order to check for genetic mutations or aneuploidy. Parents need to weigh the risks of invasive testing against their desire for more information and greater certainty to help them with time-sensitive decisions. Although rates of pregnancy loss have been difficult to estimate, a recent meta-analysis suggests no significant risk of miscarriage with chorionic villus sampling and a miscarriage risk of 1:300 with amniocentesis (1). There are other risks associated with invasive testing, however, including bleeding, Rh sensitization, rupture of membranes, and infection (2). The recommended time frame to perform these invasive tests, between 10 and 20 weeks' gestation, is exactly when concerns about the fetus are raised and therefore a relatively narrow window of time in which to make decisions about testing (3). Diagnostic results may take up to 1–2 weeks to return. Although prenatal genetic testing has been expanded to include exome sequencing, it unfortunately cannot identify all possible abnormalities. Despite these limitations, the results may determine whether some families continue the pregnancy.

As an alternative, parents may first choose to decline invasive testing and opt for non-invasive testing which includes cell-free DNA screening and additional imaging. Cell-free DNA screening has a high sensitivity and specificity for Trisomy 21 and Trisomy 18, but lower performance for Trisomy 13, other chromosomal anomalies, and microdeletions (4). If non-invasive cell-free DNA screening is suggestive of a genetic abnormality, invasive testing is still needed to confirm the diagnosis before termination. Additional imaging includes fetal echocardiogram, three-dimensional fetal ultrasound, or fetal magnetic resonance imaging. For advice and opinion on the suspected diagnosis and outcomes, parents may also want to consult with pediatric subspecialists. What adds to the complexity of the situation is that maternal fetal medicine specialists have two patients: the mother and the fetus. Their duty is to separate information about the risks and benefits of various choices for both patients, recognizing that decisions made will have linked outcomes (5). As such, the maternal fetal medicine specialist will also review for the mother potential pregnancy complications associated with fetal anomalies: preeclampsia, preterm labor, polyhydramnios, and mirror syndrome (6–10).

After gathering as much data as possible, parents will consider whether or not to terminate the pregnancy. This decision often must be made within days or a couple of weeks after learning about a fetal abnormality. While many technologies offer to improve diagnostic certainty or prognostication, parents need to know about limitations in predicting survival and outcome variability. Each test, imaging modality, and consultation is an option for parents to pursue or decline. For some families, terminating a pregnancy is not consistent with their faith or values so undergoing invasive or non-invasive testing to inform a decision to terminate does not make sense. It is possible that a life-limiting fetal diagnosis may not be discovered until after termination is no longer an option, but families may still want information to prepare themselves for possible outcomes. Other families may feel the degree of uncertainty associated with some information acquired during pregnancy is not worth the additional stress. And some families want more time to process the news before obtaining more data.

PERINATAL PALLIATIVE CONSULTATION

"Perinatal palliative care refers to a coordinated care strategy that comprises options for obstetric and newborn care that include a focus on maximizing quality of life and comfort for newborns with a variety of conditions considered to be life-limiting in early infancy. With a dual focus on ameliorating suffering and honoring patient values, perinatal palliative care can be provided concurrently with life-prolonging treatment" (11). There is a misconception that palliative care applies only to cases where the goals are to allow a natural death. In fact, perinatal palliative teams advocate primarily for respect of parental wishes, supporting a spectrum of goals from comfort-focused to life-prolonging care. A perinatal palliative team is typically multidisciplinary including a physician, nurse, social worker, and spiritual counselor (12). Some palliative teams are imbedded within fetal diagnostic centers, enabling simultaneous palliative consultation (13). For centers without integrated palliative teams, however, there are barriers to early referral and palliative consultation might not occur for weeks (14). Because decisions to terminate often need to occur relatively quickly, families who chose termination are likely to miss the opportunity for perinatal palliative consultation. In practice, consultation with a perinatal palliative team most frequently occurs after parents have declined termination with plans instead to continue the pregnancy (15). No matter which decision parents make (i.e., to terminate or continue the pregnancy), however, all parents experience losses, find themselves planning for a future they did not hope for or expect, and can benefit from the additional support for decision-making that perinatal palliative teams can provide (16).

Providing support to parents as they make decisions is central to the perinatal palliative consultation. Amidst the variable and sometimes overwhelming emotions that parents can experience, the perinatal palliative team allows parents to explore their goals in order to promote shared decision-making with regard to obstetric management as well as postnatal care for the baby. Mixed, changing, and sometimes conflicting feelings of shock, concern, disbelief, denial, anger, love, shame, hope, and guilt are normal for parents to experience when hearing serious news about their baby (17). The perinatal palliative team fosters a safe, compassionate, and supportive environment for parents to process their emotions and discuss their values (18). Without the ability to address their feelings or spiritual distress, parents may not be ready to explore their hopes for their baby's care, let alone make changes to the plan of care. For example, a spiritual counselor on the perinatal palliative team is especially important if a parent is worried about the "right" thing to do with respect to her faith. Addressing the meaning of the situation in the context of her faith is necessary before approaching decision-making about treatment options for the baby. By validating and reflecting what parents express and in turn building trust, the perinatal palliative team can support a parent's voice in decision-making and facilitate communication between parents and other medical teams (19).

BIRTH PLANS

One way in which the perinatal palliative team supports a parent's voice and decisions is to collaborate on a birth plan. A birth plan is similar to an advanced directive for the pregnancy, delivery, and neonatal care. By sharing their birth plan with the medical team, families create additional opportunities for informed and shared decision-making. A birth plan documents parental hopes, wishes, and goals of care, serving as a communication tool for the entire medical team. Personalized birth plans vary in content but generally address the following components: pain control for the mother, preference for fetal monitoring, mode of delivery, who to be present in the delivery room, resuscitation measures desired at birth, medical management for the baby, as well as wishes for memory making, and ceremonies (20). **Table 1** presents suggested topics for a birth plan. Writing a birth plan allows parents to identify and articulate goals ahead of time before a potentially overwhelming emotional moment. Parents have reported that creating and using a birth plan gave

TABLE 1 | Suggested topics for a birth plan.

Plan of care for mother
- Maternal information including medical record number and date of birth
- Preference for fetal monitoring and mode of delivery
- Requested persons to be present in the delivery room
- Music choices for labor and delivery
- Requests for any limitations in medical personnel
- Pain control preferences
- Request for information regarding maintenance and suppression of lactation

Plan of care for baby
- Planned name
- Designated person to cut the umbilical cord
- Resuscitation measures desired at birth
- Goals for medical management for the baby
    - Code status
    - Anticipated location for care
    - Expectations for respiratory and nutrition support
    - Desired diagnostic testing, imaging, consultations
    - Symptom management
    - Any limitations for routine newborn care (i.e. screening and prophylaxis)
- Selected persons to be present with the baby
- Memory making including caregiving activities, photography, and creation of mementos
- Desired spiritual care or ceremonies

End-of-Life Plan
    - Plans for home hospice care
    - Organ donation
    - Autopsy
    - Preference regarding use of a cooling bassinet
    - Final Arrangements
    - Requests for bereavement counseling resources

them a sense of control, was therapeutic, and helped them feel prepared (21).

The most challenging sections of the birth plan to complete usually address fetal monitoring, mode of delivery, and goals of care for baby's management. The 2019 ACOG Committee Opinion on Perinatal Palliative Care states that, "Decisions regarding the appropriateness of intrapartum fetal monitoring in cases like this should be individualized" (11). Some families are willing to accept the possibility of their baby's death during unmonitored labor in order to avoid the operative risks of cesarean section. Other families would rather have fetal monitoring during labor and if necessary, cesarean delivery for fetal distress, to increase the chances for their baby to be born alive. For example, some may want the opportunity for the entire family to meet the baby or to perform a ceremony at birth. For some families, having a postnatal confirmatory diagnosis of a life-limiting or serious medical condition gives them the assurance they need to allow their child to have a natural death. And other families may want to pursue life-prolonging treatments in order to give their baby the best chances for survival.

The uncertainty of whether the baby will survive the pregnancy and birth can make planning for the neonatal period challenging. Some families are ready to consider in detail the choices for baby's care after birth while other families want to defer those decisions until after the baby is born. The perinatal palliative team can help to explain how care for the newborn can be adapted to align with the goals of care. For example, families may want a time-limited trial of respiratory and nutrition support in order to see if the baby can breathe or eat on her or his own. If the baby is unable to breathe independently or feed by mouth safely, then families will need to consider whether providing long-term ventilation or artificial nutrition aligns with their goals for care. As it was during pregnancy, each test, imaging modality, and consultation for the baby is an option for parents to pursue or decline.

The birth plan will vary based on a family's faith, values, and experiences. For example, some families will want specific spiritual care or ceremonies after birth to honor their faith. But there are also some universal themes (22). Janvier et al. found that among families with children who lived with Trisomy 13 or 18, there were "common hopes to bring their child home, give their child a good life, and be together/a family" (23). Parents often have an intrinsic need to feel that they have "done the best that they could" for their child and want to avoid having regrets in the future (24). Maintaining hope is an important source of strength for parents when facing difficult situations (25). A parent's hope can change with time and new information, from hoping for a normal pregnancy, to hoping the baby will be healthier than predicted, to hoping that the baby will be alive at birth to meet the family. During goals of care planning, the perinatal palliative team can encourage and help families to maintain hope as circumstances change. Nearing the day of delivery, parents may worry about future regrets in choosing comfort-focused care, and want time-limited trials of therapy until the baby's diagnosis is confirmed. It is important to validate that concerns about regret can inform the goals of care and to acknowledge that allowing time to pass might reduce parental decision regret (26). There is also the possibility that at birth, the baby's presentation may not match the prenatal diagnosis thereby suggesting a different prognosis. Health care providers need to be flexible and remember that the birth plan serves as a guide for both mother and baby care providers, but it can be modified at any point before and after delivery. Ultimately, what is most important is that parents are supported, ideally by perinatal palliative teams alongside other medical teams, to discuss and express the rationale behind their decisions.

MEMORY MAKING

Whether the goals of care are focused on life-prolongation or comfort, most families whose baby may have a short life want to plan for memory making. The perinatal palliative team offers expertise and experience to support parents in the many ways they want to make memories. Memory making includes "any intervention or experience that encourages contact or interaction between the parents and newborn and any intervention that results in the creation or collection of mementos" (27). Mementos include molds of the baby's hands and feet, foot and handprints, blankets, hats, and clothing that the baby touched, as well as crib cards, hospital bands, and other personal items that were associated with the baby in the hospital. Now I Lay Me Down to Sleep is a non-profit international organization which offers free professional retouched photography to families who have a baby with a life-limiting condition (28). Families may want to take their own photographs or hire a photographer instead. Although parents may not want to see photos of their child right away, the portrait sessions give them the opportunity to have documentation of their child's life available for when they are ready. After the death of an infant, interviewed families emphasize the importance of having as many parenting experiences as possible to reflect upon in bereavement (29). Opportunities for caregiving can include bathing, dressing, and holding their baby as well as talking, singing, and playing music with their baby (30). Families recall that they needed encouragement and guidance to overcome their hesitation in order to spend time with their newborn (31). Some families introduce their baby to siblings and relatives to officially welcome him or her into the family. Making memories can not only validate the importance of the newborn's life and death but also create a sense of identity for individuals as parents, siblings, and grandparents.

COMFORT FOCUSED-CARE

If families choose comfort-focused care for their baby, there may be an opportunity to continue the caregiving at home with hospice support. A hospice physician and team provide expertise in end-of-life care at home and ongoing bereavement counseling for the immediate family. A designated pediatrician is customarily not needed unless the goals of care change to include life-prolonging interventions. If the baby is unlikely to survive the trip home, some parents may want to room-in with the baby in the hospital while some families may not have the financial or logistical capacity to room-in. The perinatal palliative team can provide inpatient support to the primary neonatal team who will attend to the baby's pain and symptom management as well as support the family through the dying process. Many parents have little to no experience with death and will want to know what their baby will look like and feel in the last hours of living (32). Generally, babies in the dying process will become less active, sleeping more as time goes on. Any ability to swallow saliva will become impaired. With less intake, decreased urine and stool output is expected. Cool hands and feet along with mottling of the skin is normal. Less commonly, newborns may need treatment for seizures in the dying process. Eventually, when the baby is no longer conscious, periods of apnea, or Cheyne-Stokes pattern respirations will occur. Ordinary techniques to soothe babies, such as holding, rocking, and swaddling, are usually successful to minimize discomfort. Occasionally, morphine or lorazepam may be needed for symptom management of pain or agitation, respectively. Normalizing the signs of dying for families may reduce their anxiety and distress when the time comes. It is not possible to predict when death will occur, but it is possible to prepare families for the unpredictability of death. Using words such as "weeks not months," "days not weeks," or "hours not days," can be helpful (33).

If medically possible, families can choose organ or tissue donation to create a hopeful and positive legacy. Organ procurement teams have specialized training and are the preferred communicators to maintain ethical and clinical standards while initiating organ donation discussions. Referrals to organ procurement organizations should be made in a timely manner when death is anticipated and may occur before birth (34). In addition, some families may agree to a full or limited autopsy to inform subsequent pregnancies or to improve medical knowledge for future families. Others may decline autopsy because they do not need more information, or their faith and values are not congruent with allowing an autopsy. Families may choose final arrangements based on family or faith traditions as well as their budget. Generally, cremation is less expensive than burial but the cost can vary by mortuary and location (35). Some parents will want to designate a relative or friend to do research and get quotes on available services and some families will prefer to do this work on their own.

After a baby dies, some families will want extended time with the baby and may want to use a cooling bassinet to decrease the rate that the body deteriorates (36). Others will say goodbye a few hours after the baby dies. There are physical and emotional aspects of lactation that need to be addressed. Some mothers will want to suppress breast milk production but need education on how to avoid painful engorgement and mastitis. Others will want to continue expressing and donate breast milk as a way to help other families and find meaning in the context of their loss and grief (37). It is important to recommend grief resources and counseling for families whether they go home with hospice or stay in the hospital setting. Some families may prefer to visit online bereavement resources first and others will want to start mental health therapy immediately (38, 39). The perinatal palliative team continues to support families as they make choices to honor their baby that may include a funeral, celebration of life, and donations to charities or research. Additionally, the team often maintains contact with families in bereavement. It is important to share with families that there are no right or wrong ways to grieve or feel when a baby dies.

CONCLUSION

Through the journey with a potentially life-limiting fetal diagnosis in pregnancy, a perinatal palliative team can provide the encouragement families need to make decisions based on their faith, values, and experiences. The team has the expertise to create a safe, compassionate, and caring environment for parents to process a number of intense emotions and discuss their values. Although parents must confront uncertainty and complex decision-making for both the mother and baby, perinatal palliative teams support parents to express their goals as well as find meaning and hope.

AUTHOR CONTRIBUTIONS

KM-A agrees to be accountable for and conceived, drafted, revised, and approved all aspects of the work.

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The Neonatal Comfort Care Program: Origin and Growth Over 10 Years

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The objective of perinatal palliative care is to provide holistic and comprehensive health care services to women who are anticipating the birth of a neonate diagnosed prenatally with a life-limiting condition and to continue supportive interventions for the mother and neonate after the birth. The nature of pregnancy, with two patients requiring medical care, requires clinicians from different specialties to engage with one another, the patient, and her chosen family members. Following birth, additional skill sets to treat the medical and comfort needs of the neonate, as well as the psychoemotional and medical needs of the mother, are required. An interdisciplinary team is necessary to assist families throughout the pregnancy and postnatal journey, and coordination of such care is an integral component of palliative care services. The number of palliative care programs is increasing, but little is written about the origins of such programs, their subsequent growth, and how transitions of care occur within the programs. In this publication, we will present data garnered from interdisciplinary team members of a single organization, the Neonatal Comfort Care Program at Columbia University Irving Medical Center, and how they provide care for families throughout the pregnancy and postnatal trajectory. We will address the origin and growth of the program, the development of the interdisciplinary team, and the strategies used for high-quality communication and their respective impact on care continuity. We will also provide specific recommendations from data gathered from team members, examine the role of formal and informal education, and identify barriers and future opportunities.

Keywords: interdisciplinary care, program development, life limiting conditions, perinatal palliative care (PPC), neonatal palliative care

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INTRODUCTION

There is rarely a time in a family's life that entails more transitions than pregnancy and the birth of a child. It is a time of anticipation, changes, preparation, and often great joy. When a woman is faced with news that her expected child has a life-limiting condition (LLC), the pregnancy course abruptly changes, as do the complexity and challenges she and her family experience. Historically, women were offered the options to terminate the pregnancy or prepare to have their neonate admitted to an intensive care unit. However, in 1997, the concept of perinatal hospice was introduced (1), followed by clinical recommendations for a neonatal end-of-life palliative care protocol (2). Since then, perinatal palliative care (PPC) programs, which are defined as comprehensive, interdisciplinary, coordinated services offered from fetal diagnosis through the neonatal period, have experienced significant growth (3) and national recognition (4–6). This extension of a woman's right to choose respects patients' cultural beliefs and values, can empower women, and has been met with increases in patient satisfaction (7, 8).

Clinicians from many disciplines play an integral part in supporting families by providing compassionate, evidence-based care and can offer information and services that allow families to make the most of the time with their neonate. Data show that women who opt to continue a pregnancy with an LLC appreciate opportunities to emotionally adjust and report having no regrets about their decision (9, 10). A variety of interventions, including anticipatory guidance, prenatal consults, early and continuous bereavement support, and coordination of care allow families to begin the difficult but important process of adjusting to the implications of an LLC (11, 12). Formal, integrated palliative care programs have structures in place that ensure the interdisciplinary team (IDT) can communicate well, manage the needs of families, and offer high-quality care.

The purpose of this article is to provide practical information for professionals interested in starting, sustaining, or increasing PPC services within an organization by using the model of the Neonatal Comfort Care Program (NCCP) at Columbia University Irving Medical Center (CUIMC). Aims are to (1) provide an overview of the program's origin and its subsequent growth, (2) explain the formation of the IDT, (3) outline communication strategies of team members to ensure continuity of care, (4) highlight recommendations from team members working in the program, (5) emphasize the significant role of education in culture change, and (6) identify barriers in developing the program and future opportunities.

AIM 1—PROGRAM ORIGIN AND SUBSEQUENT GROWTH

CUIMC is a large academic institution with ∼4,000 deliveries annually and about 800 pregnancies involving fetal anomalies, including LLC. The NCCP began in 2008 and grew out of a desire to provide the most comfortable and loving environment for neonates diagnosed *in utero* with LLC, help families manage and navigate the immense practical and emotional burdens associated with LLC, and offer an option of care that families identify as safe, supportive, and in sync with their personal beliefs and desires. The idea for formalizing a strategy to provide comfort care for neonates with LLC started with a neonatologist. At the beginning, obstetric and maternal fetal medicine colleagues were invited to make referrals for a prenatal palliative care consult whenever a candidate was identified. As families provided positive feedback of their experiences and as relationships between neonatology and obstetric providers advanced, referrals increased. Consults are medically oriented meetings wherein the physician reviews the patient's medical history; meets with family members to assess their understanding, desires, and values; and makes recommendations about care and treatment. The consults are reimbursable. Patient encounters are supportive services provided by appropriate IDT members and are tailored to family needs. **Figure 1** provides a timeline outlining the onboarding of the NCCP members, educational programs, acquisition of funding, and contributions to the literature.

Annual prenatal consults have grown logarithmically from 13 consults in 2008 to 64 in 2019. Patient encounters have increased in a similar fashion from 16 in 2008 to 127 in 2019.

AIM 2—DEVELOPMENT OF THE CORE AND INTERDISCIPLINARY TEAMS

In 2011, a neonatal intensive care unit (NICU) nurse self-identified an interest in partnering with the neonatologist, leading to a part-time position in 2012. By 2017, as professionals expressed an interest, a core team was established with a medical director from neonatology dedicated to, but not funded by, the NCCP program. A full-time nurse, social worker, and program manager were funded from unsolicited philanthropic donations given by individuals and foundations. The program manager is focused on program development and outreach to establish sustainability. The IDT includes collaborators/champions from neonatology and obstetrics, a child life specialist, speech language pathologist, psychologist, chaplain, obstetric nurse, and maternal–fetal medicine physician. The uniqueness of the program lies in an intentional continuity of care among team members and professionals and a desire to ensure quality services throughout the entire perinatal journey.

During this decade of growth, the NCCP established and published innovative clinical guidelines focusing on the comfort of the neonate and family bonding, leading to an impressive culture change (13). One of the key elements of bonding is that the mother and her infant should have close contact and not be separated. In 2008, in our institution, as elsewhere, neonates with LLC were admitted to the NICU for "comfort care," resulting in separation of the mother and infant. Intentional in-house education was offered, and by 2018–2020, all families followed prenatally by the NCCP received the option to room-in after birth, and all mothers who elected this option (98%) successfully kept their infant with them in a private postpartum room. The NCCP offers rooming-in for infants at the end of life, as well as infants with any kind of LLC, as long as the parent(s) desire palliative care management. Oxygen via nasal cannula, nasogastric or syringe feeding, and pain management are provided, and the NICU team is responsible for the infant. Another essential element for bonding and comfort is feeding; involvement of a speech pathologist has been key to safely and successfully feeding these fragile neonates throughout their natural lives.

AIM 3—COMMUNICATION STRATEGIES

Referrals for Consultation

Referrals for consultation to the NCCP are initiated through (1) obstetricians directly initiating a consult with the NCCP;

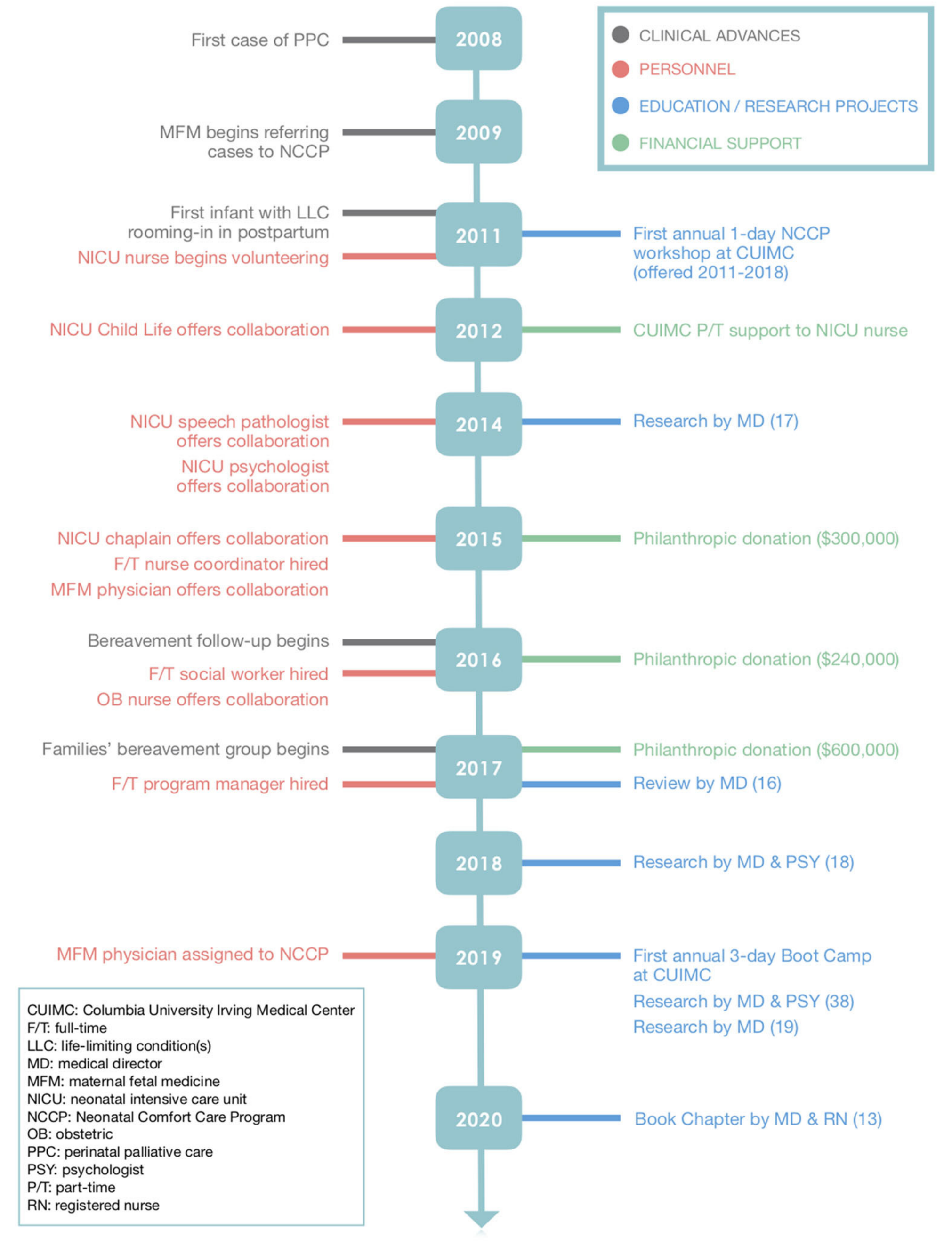

FIGURE 1 | Timeline of Neonatal Comfort Care Program (NCCP) Growth and Milestones.

(2) identification of new patients in weekly meetings among the NCCP medical director, nurse coordinator, and maternal–fetal medicine and neonatology physicians; and (3) self-referral of women requesting a PPC consultation in partnership with their obstetric provider.

Candidates

Candidates include pregnant women with potential or certain fetal LLCs and those with a complex fetal diagnosis with a potential adverse prognosis. A specific list of fetal LLC diagnoses triggers a referral to the NCCP. Included are conditions with expected early demise, such as renal agenesis, anencephaly, and limb-body-wall syndrome, as well as cases in which the burden of intensive care may exceed benefits, such as trisomy 13, trisomy 18, genetic syndromes with life-limiting prognoses, extreme prematurity at the cusp of viability, or complex congenital heart disease. Complex conditions with potential adverse outcomes also initiate a consult. Examples include uncertain diagnoses, such as multiple skeletal anomalies or severe brain anomalies, and known diagnoses with uncertain prognosis, such as bilateral renal dysplasia, severe lower urinary tract obstruction, or severe hydrops. The NCCP takes medical care of all infants with these conditions. Regardless of the family's decision to have comfort care vs. a trial of intensive care vs. full NICU support, the NCCP is available to support the family throughout the perinatal journey. At birth, if the family elects comfort care, the NCCP takes direct care of the infant.

Consults and Patient Encounters

Once a consult is initiated, the medical director is notified, and the first meeting with the family is planned. The medical director, nurse coordinator, and social worker meet with the family during the first consult to (1) probe the family's prognostic awareness, (2) clarify diagnosis, prognosis, (3) offer options for postnatal plans, (4) propose supportive interventions, and (5) establish a relationship of trust that will continue along the entire perinatal journey. Two aspects of care planning occur in tandem. First, the medical director oversees a postnatal medical care plan dependent on fetal gestational age and family need. Second, the family will meet with the nurse coordinator and the social worker, who offer supportive interventions and resources during pregnancy, delivery, and beyond. The patient encounters consist of interventions such as anticipatory guidance, discussion and documentation of a family birth plan, psychological counseling, memory making, sibling support, religious and cultural rituals, and referrals to other IDT professionals as needed. Usually two to four meetings occur.

Communication

A systematic approach of communicating patient events occurs through a weekly meeting among the core team and the maternal–fetal medicine physician. The outcome of these clinical rounds is reported in a weekly "comfort care list" emailed to the IDT and NICU and obstetric fellows. In addition, documentation from each consultation is reported in the electronic health record by each member of the NCCP team. The core team also meets monthly to review follow-up of neonates discharged home or to hospice and the follow-up of mothers/families who suffered a perinatal loss. In addition to these formal communication processes, informal conversations among team members occur through telephone calls, texting, and face-to-face meetings. The team is careful to apply patient privacy standards when communicating.

Continuity of Care—Prenatal to Postnatal

One outcome of high-quality communication among team members is the opportunity to provide continuity of care. In the NCCP, the medical director and the nurse coordinator, when available, are present at delivery to ensure continuity of care to the family or to mentor staff members in the obstetric and neonatal units. The champion obstetric nurse is also available to mentor and facilitate care in the delivery room and postpartum unit. The unpredictability of labor requires a communication system that allows pristine fulfillment of the medical and interdisciplinary birth plans.

The number of newborns with LLC is relatively small so the medical director assumes direct care of most neonates until death or discharge, along with the NICU staff (NICU fellow, nurse practitioner, resident). When the neonate is admitted to postpartum, a NICU nurse is assigned to assess the infant once a shift or more often if needed. IDT members are consulted as needed. A child life specialist helps with memory making and sibling support, and the chaplain addresses spiritual needs and coordinates specific parent-requested rituals. Standardized guidelines focused on achieving a state of comfort for the neonate are implemented and have been published (13).

The NCCP social worker collaborates with the obstetric social worker in supporting the mother and the family. The social workers provide resources and information about a coordinated discharge to home, follow-up with hospice, and disposition of the infant. Once the mother is discharged home, the care coordinator and social worker, and other IDT members if needed, continue to support the family by offering a bereavement follow-up for at least a year.

AIM 4—RECOMMENDATIONS FROM TEAM MEMBERS

Data regarding recommendations for those interested in starting or developing a program were collected from individual team members in the NCCP program. Recommendations were collated and are delineated in **Table 1**.

AIM 5—EDUCATION TO PRODUCE CULTURE CHANGE

Education has been pivotal to the advancement of PPC in CUIMC. Both informal and formal educational strategies have promoted a culture change and facilitated a positive PPC mindset among CUIMC personnel, as well as clinicians in the surrounding metropolitan areas. Neonatal palliative care advancements set the stage for the uptake of PPC with the downstreaming of literature and the increased interest among neonatologists to assist and prepare parents expecting a neonate with an LLC.

Starting in 2014, the NCCP organized and offered an annual 1-day workshop drawing 50 to 70 professionals from CUIMC and the surrounding region each year. The workshop included lectures, discussion of clinical cases, and parent interviews. In 2019, the workshop was expanded to a 3-day boot-camp training course with continuing education credits for physicians and nurses. The program agenda is available on the website and included formal lectures, role-modeling case studies, hands-on demonstrations, group discussion, parent voice, and networking opportunities. More than 80 participants from 19 states and several countries attended. Unique to the program was the opportunity for each NCCP team member to provide evidence-based strategies and share their clinical experiences with the participants (15).

TABLE 1 | Recommendations and suggested resources from NCCP team members to clinicians planning to start a PPC program.

CORE TEAM	
Neonatologist, program director	• Create a mission and vision and access it to maintain motivation (14) • Establish collaboration with OB • Look for "champion" professionals interested in collaboration • Work cohesively and regularly with team members to ensure optimal communication • Use professional encounters to teach and role model the tenets of PPC (15, 16) • Establish policy/guidelines for PPC in your institution (12) • Measure and report program outcomes and successes (17–19)
Registered nurse, program coordinator (former NICU RN)	• Identify champion RNs interested in PPC and provide teaching and mentoring • Participate in online and live educational opportunities (15) • Use online and community resources to gather bereavement supplies (20) • Develop a curriculum of Nursing PPC to create a PPC mindset
Program social worker (former PICU SW)	• Document all tasks related to PPC and build a case for SW position with data • Join perinatal hospice (20) network and network with other SWs to gain insights and information (21) • Participate in online and live educational opportunities (15, 22)
IDT TEAM	
OB registered nurse	• Develop QI projects to build a case for resources for PPC • Train and mentor OB nurses to create a PPC mindset (23, 24) • Network with OB nurses in other institutions to share ideas and resources for PPC
MFM physician	• Collaborate with the team in pregnancy management to achieve family's expectations (4) • Train OB staff to respect and support the family's goals (25, 26) • Develop research projects to demonstrate the benefits of PPC
NICU speech-language pathologist	• Contact experienced colleagues for advice (27) • Consider training champion NICU RNs to assess feeding needs for a neonate and become familiar with specific feeding equipment • Train all staff to respect and support the family's goals relative to providing nutritive or non-nutritive therapies (28)
NICU child life specialist	• Train other child life specialists to specific interventions in PPC (29) • Consider training "champion" RNs to become familiar with memory making during pregnancy and at birth
NICU psychologist	• Contact in-house psychologists and assess interest in working with bereaved parents • Develop collaboration with team SW • Be intentional about listening to parents and help them navigate their experiences during pregnancy and beyond (30, 31)
NICU chaplain	• Partner with in-house or community faith leaders • Familiarize clinicians with spiritual screening using FICA Spiritual History Tool (32) • Assess patient's spiritual preferences prenatally to prepare for spiritual interventions (33, 34)

Informal education occurs routinely by the nurse coordinator and champions within the IDT. Examples include bedside teaching in the delivery room, postpartum unit, and the NICU as mothers and their neonates present to these areas. The obstetric nurse champion conducts huddles, mentoring, and bedside teaching and is instrumental in keeping lines of communication clear and consistent. Information about the NCCP is given to all new nurses during orientation.

The medical director of the NCCP is responsible for training and mentoring obstetric and neonatal fellows and residents in the clinical setting. Additionally, a five-lesson curriculum was developed and is used to formally present essential elements of PPC.

AIM 6—BARRIERS AND FUTURE OPPORTUNITIES

Barriers to implementing workplace change were two-fold. First, PPC is an emerging field, and the evidence base was limited at the start of the NCCP program. Coupled with the development of new research knowledge were positive clinical experiences. Over time, resistance to PPC diminished, the culture improved, and the mindset of perinatologists changed. A second barrier was limited funding. Resources were allocated over time and provided support for new staff, educational initiatives, and research endeavors.

The future of the NCCP program is promising. Educational goals of the NCCP will build upon the foundation created in the last decade. Plans are in place to increase educational outreach and establish deeper collaborations with regional institutions to assist with replicating best practices related to PPC. Educational recommendations from NCCP team members include additional in-services, development of a basic curriculum for nurses centered on area of expertise, and establishment of a PPC fellowship for NICU, obstetrics, and social work. Expansion of the clinical team includes plans for a dedicated midwife who would provide continuity of care during the perinatal journey, facilitate communication during encounters with various obstetrics providers, offer one-on-one childbirth education, be present at the birth, and provide postpartum follow-up and lactation support. Involvement of additional clinicians to act as NCCP champions will reinforce expertise, assist with training and supporting colleagues, and provide more penetration of PPC culture within units. Ongoing mental health treatment in the form of groups and/or individual/couples therapy is also under consideration. Further research will provide new knowledge upon which evidence-based practices can be established.

DISCUSSION

PPC literature has grown in the last two decades and, with it, application into clinical practice. Palliative care programs are in 25 countries worldwide, with 246 in the United States and 67 globally (20). However, of the 246 perinatal hospices in the United States, only 20% offer hospital-based perinatal care, and nearly 40% offer only bereavement support. Of the 20% offering hospital care, the NCCP is unique in its prioritization of uninterrupted support of families of neonates with LLC. From the moment of diagnosis, through birth and beyond, the same physician and team members provide continuity of care to the neonate and family throughout each step of the journey.

As palliative services expand to the perinatal population, patient- and family-centered care will continue to optimize quality of life by anticipating, preventing, and treating suffering (35) through the continuum of the pregnancy and postnatal course. There are several benefits of a formal program as team members optimize communication strategies, work within standardized guidelines, and track outcomes and quality care. Systematic incorporation of evidence into practice settings can take many years, and over time, with dedicated staff and administrative support, referrals increase. The work of improving quality and effectiveness of health services requires consistent vision of individuals and teams, along with resources and administrative support, to solidify lasting change.

Culture change is an essential part of clinical advancement, especially with respect to palliative care as palliative therapies are interventions with complexities that affect the physical, psychoemotional, and spiritual aspects of human care. Advances in the NCCP program, including establishment of guidelines available to any professionals involved in perinatology, occurred through multifaceted approaches, including a mindset reorientation for leaders and frontline clinicians. As more clinicians become aware of and are supported in providing palliative care, their confidence increases, and a significant cultural shift occurs.

In line with current recommendations, an IDT should consist of a multitude of experts, all of whom play specific roles in providing tailored services to neonates, women, and their family members. The essential first step is the collaboration between the obstetric and neonatology services, which ensures that pregnancy and delivery are managed in a continuum (4). Anecdotally, medical directors are commonly neonatologists or obstetricians rather than palliative care physicians. These specialists are housed in every hospital in the USA whereas palliative care physicians are not. Essential elements of PPC come under the auspices of obstetrics and neonatology who have expertise with prenatal counseling, pregnancy management, and palliative comfort interventions for infants with a shortened life span. Communication is optimized through a systematic, consistent collaboration within the IDT. Unlike several programs in the US (3) led by a nurse coordinator or a bereavement counselor, the NCCP was initiated and developed by a neonatologist who established a collaboration with obstetricians and provides prenatal counseling and direct medical care to the neonates, promoting continuity of care for each family. The nurse leader facilitates the nursing aspects of care and coordinates interdisciplinary services, helping to form a cohesive approach to care. Thanks to professionals offering their services and prioritizing rooming-in with mother and feeding as comforting to the neonate, the NCCP has been building a mindset that contributes to the creation of a peaceful, safe, and loving environment. The life of the neonate, rather than the anticipation of his or her loss, becomes the focus of attention.

Data provided by NCCP team members offer recommendations to read appropriate discipline-focused literature, attend educational events, and listen to parents. The identification of local "champions" is key to establish the much-needed culture change toward a mindset open to PPC among professional working in the perinatal field. Professional networking affords a host of benefits by strengthening and expanding the exchange ideas and information. Networking can also facilitate change through brainstorming and problem solving. Lastly, PPC is still a very young field in medicine and practice needs to be evidence-based through prospective research.

Although the number of PPC programs has been increasing over the past years, training and education for professionals have not kept pace. Evidence-based, consistent, comprehensive training courses for professionals involved in PPC across disciplines are not widely available. A lack of knowledge is linked to clinician discomfort in providing care for families affected by an LLC (36). Since the start of NCCP, educational initiatives were a priority, leading to the changing dynamics among units that forged cultural and clinical changes. Educational approaches were diverse and included formal annual training events, group lectures, and bedside training and mentoring.

The allocation of resources is a constant challenge for health care administrators, and yet, the lack of resources can have serious consequences on patient-centered quality care (37). A variety of approaches has helped keep the NCCP operational, including the strong motivation of the core team members to provide comfort care despite scarce resources, the availability of some IDT members to offer some extra hours, and a cohesive team mentality enabling optimal communication and shared successes. Plans to continue outreach to philanthropic foundations to ensure sustainability are a future priority.

CONCLUSION

Building a new program and changing organizational culture take strong motivation, commitments of time and energy, and, eventually, adequate funding. Clinical leaders can forward a consistent vision and focus on creating an ongoing narrative related to optimal comfort care for neonates with LLC and support for their families.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

AUTHOR CONTRIBUTIONS

CW and EP conceptualized and designed the report, drafted the initial manuscript, and approved the final manuscript as submitted. Both authors contributed to the article and approved the submitted version.

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PERSPECTIVE
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“Percorso Giacomo”: An Italian Innovative Service of Perinatal Palliative Care

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The perspective proposed by this article will focus on perinatal palliative care as a strategy for improving the quality of life of neonates with life-limiting conditions when extending the patient's life is no longer the goal of care. This manuscript reports the creation of an innovative program of perinatal palliative care called “Percorso Giacomo” (Giacomo's Pathway) at Sant'Orsola Hospital in Bologna, Italy in 2013. Key features include interdisciplinary collaboration between professionals from obstetrics, neonatology and other specialties aiming to reach the most detailed fetal and neonatal diagnosis and prognosis; communication and engagement with the family to discuss goals of care and prepare a birthing plan that follows the family's desires and expectations; and personalized care to achieve comfort for each newborn and support for families according to their social, cultural, and religious backgrounds.

Keywords: perinatal palliative care, neonatal palliative care, life-limiting conditions, comfort care, interdisciplinary team

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INTRODUCTION

Perinatal palliative care (PPC) is offered in the settings of fetal or neonatal diagnoses of life-limiting conditions (LLCs) as a plan to achieve comfort of the neonate and to support the family. Given the complexity of the medical and non-medical needs of these neonates and the emotional challenges for parents, PPC is a necessarily holistic and interdisciplinary approach to care. PPC is a journey with families that begins at the moment of diagnosis, whether prenatal or postnatal, and involves a collaboration among maternal-fetal medicine specialists, neonatologists, and other professionals with the goal of establishing an individualized plan of care for the mother-baby dyad.

The American College of Obstetricians and Gynecologists (ACOG) considers PPC one of the options of care that should be offered to patients facing pregnancies complicated by fetal LLC (1). Yet, PPC is not yet routinely integrated in prenatal counseling in the United States (2, 3). This also holds true for Italy. A recent Italian national survey of neonatal intensive care unit (NICU) practitioners demonstrated that only 30% of the institutions represented offer a structured PPC program. Of these, 34% have only one program coordinator, and fewer than 30% organize PPC training for health care providers (4).

Despite technological advances in the ability to detect severe fetal anomalies, infants with LLCs are occasionally diagnosed only after birth. Moreover, critically ill neonates admitted to the NICU can face a potentially adverse prognosis or reach the end-of-life stage, at which point a redirection of goals of care is required. According to the World Health Organization (WHO), palliative care and intensive care should be provided concurrently to critically ill infants with potentially adverse prognoses (5). The American Academy of Pediatrics (AAP) and the Italian Society of Neonatology (SIN) provide recommendations for achieving comfort of terminally ill neonates and of those affected by an LLC (6–9).

The urgent need to provide the option of PPC to families with neonates prenatally or postnatally diagnosed with an LLC prompted the creation and implementation of an innovative PPC program at Sant'Orsola Hospital (SOH) in Bologna in 2013. The program was named "Percorso Giacomo" (Giacomo's Pathway) in honor of the first infant to whom SOH provided such care. This paper describes the origin and the development of the program over its first 7 years.

CLINICAL CASE

Giacomo was prenatally diagnosed with anencephaly. His parents, although aware of his short life expectancy, chose to carry the pregnancy to term. The mother was a G4 P3 woman with two healthy children and another baby with anencephaly whose delivery was complicated by failure to progress and stillbirth. As she again received an anencephaly diagnosis, she asked her provider to manage her care with the goal of having some precious time with her baby after birth. At that time there were no guidelines for PPC at SOH, so the obstetrician involved a neonatologist and a midwife to plan the delivery and the infant's postnatal care according to the mother's desire. Giacomo was delivered by cesarean section (transverse position) with Apgar scores of 9 and 9. After his scalp wound was carefully dressed, he was given to his family to hold. Per parental wish, he was baptized by the hospital chaplain. Following the family desire for bonding, Giacomo was allowed to room-in with his mother in a private postpartum room. At SOH, this was the first time that a baby with LLC was not admitted to the NICU. Comfort measures, including skin-to-skin care and holding to facilitate bonding and warmth, were offered. The infant had no sucking reflex; thus, he was given small feeds via an orogastric tube to relieve hunger and thirst. Discomfort and pain were assessed but Giacomo had no signs of distress and did not need any pharmacological treatment. Giacomo lived for only 19 hours, but he had the opportunity to meet his parents, siblings, and relatives, and he was comfortable throughout his entire life.

Written informed consent was obtained from Giacomo's parents for the publication of any potentially identifiable images or data included in this article.

PERCORSO GIACOMO

Origin

The experience with Giacomo made our perinatal team aware of the urgency to formalize a systematic approach to families who received a fetal or neonatal diagnosis of LLC, and a collaborative journey began. Another key factor that triggered the initiation of a PPC program, was that the neonatologist who provided care to Giacomo received training in PPC in 2009–2011 by participating in the Neonatal Comfort Care Program (NCCP) (10) at Columbia University Medical Center (CUMC) in New York, NY, US. Upon her return to Italy she began to investigate the feasibility of a similar program at SOH, and the meeting with Giacomo's family was the occasion to start the project.

During the debreafing of Giacomo's case, each of the professionals involved in the care expressed interest in developing a service for similar cases and the planning began. The group decided to name the program "Percorso Giacomo" (PG) in honor of Baby Giacomo.

The experience with this family was not only the trigger for the planning of a new service, but also a milestone in the understanding of the complex medical and non-medical needs of these infants and their families.

The group then opened to other professionals interested in the same goal, and a working group of champions called the "Percorso Giacomo Team" (PGT) was established. Two main objectives were identified: (1) assemble an interdisciplinary team, and (2) define guidelines and eventually establish a new hospital policy for PPC.

Gathering an Interdisciplinary Team

SOH is a large academic institution with a busy maternal-fetal medicine service that has approximately 3,500 deliveries each year. The level 3 NICU takes care of some 300 critically ill neonates annually, including infants prenatally or postnatally diagnosed with an LLC. The two services work collaboratively, but until 2013 there was limited knowledge and no direct experience of PPC.

The PGT is an interdisciplinary team that includes nine clinicians from obstetrics and neonatology who identify themselves as champions for the care of mothers and infants with a fetal or neonatal diagnosis of LLC. The program is not funded, and the team members make themselves available as primary providers for the mother-baby dyad and facilitate the execution of the plan of care in collaboration with medical staff involved in the care. Professionals from a variety of other specialties (cardiology, neurology, surgey, nephrology, genetics, etc.) are also available for consultation.

Establishing Guidelines

After carefully reviewing PPC literature, including recommendation from the ACOG (11), the AAP (6), and the SIN (8), and through networking with other national and international academic centers, local guidelines were created and implemented to standardize interventions. The essential elements are summarized in **Tables 1**, **2**.

As the focus of care is the satisfaction of neonates' basic needs for bonding, maintanance of body temperature, and relief of hunger and thirst, our guidelines reflected these recommendations. Pain and distress are assessed and treated with pharmacological interventions (12). The Neonatal Infant Pain Scale (NIPS) (13) and Echelle Douleur Inconfort Nouveau-ne' (EDIN) (14) are the preferred pain assessment tools because they are based on the infant's behavior and do not require vital signs detection, which could disrupt the bonding experience. Each neonate has unique medical and non-medical

TABLE 1 | Guidelines to provide comfort to neonates prenatally diagnosed with life-limiting conditions.

At prenatal counseling

- Discuss comfort measures in detail and assure parents that a state of comfort for their baby can always be achieved, regardless of his/her condition
- Fill out a birthplan with the family according to their preferences and cultural/social/religious background and give a copy to family
- Obtain pictures/videos of fetal ultrasounds as per family's wishes
- Discuss administration of Vitamin K, eye prophylaxis

Before delivery

- Review birthplan with family
- Collect items for special dressing if needed (anencephaly, limb-body-wall complex, etc.) and clothing appropriate for the baby's clinical condition and size
- In case a blood test is needed to confirm the diagnosis, plan for cord sample

After delivery

- Evaluation of baby's clinical status, provide gentle suction if needed
- Place hat, place dressing as needed and wrap in clean warm blanket
- Allow mother and/or father for skin-to-skin or holding
- Facilitate bonding with siblings and extended family as per family's wishes
- Involve chaplain as per family's wishes
- Obtain pictures, videos and memories as per family's wishes
- Transfer mother and baby to post-pastum for rooming-in in a private room

Neonatal plan of care

- Vital signs: heart rate, respiratory rate, temperature and pain score (NIPS or EDIN) every 3 h or as needed according to the baby's status
- No blood or other tests should be obptained, unless needed for confirmation diagnosis
- Encourage breastfeeding or gavage feeds as appropriate. Establish a minimum intake to guarantee relief of hunger/thirst (breast milk or formula). Consider colostrum care for babies at end of life
- Assessment of respiratory distress (air hunger, agitation, increased work of breathing, gasping) and use of non-pharmacological strategies (gentle suctioning upper airways, positioning) or pharmacological treatment (morphine sulfate PO, lorazepam PO)

NIPS, Neonatal Infant Pain Scale; EDIN, Echelle Douleur Inconfort Nouveau-né; PO, per os.

TABLE 2 | Guidelines to provide comfort to neonates with life-threatening or terminal conditions in the NICU.

Neonates critically ill but still receiving full intensive care

- Discuss baby's clinical status and potential comfort interventions with medical/nursing team
- Discuss with parents a plan of comfort interventions to improve quality of life
- Provide privacy in the NICU
- Facilitate baby/parents bonding by holding, gentle touch, massage
- Encourage breast pumping if mother desires to give her baby breast milk
- Provide parents the opportunity to take care of their baby's needs (colostrum care, non nutritive sucking, diaper change, sponge bath, dressing, etc.)
- Involve siblings and extended family as per family's wishes
- Involved chaplain as per family's wishes
- Obtain pictures, videos and memories as per family's wishes

Neonates at end-of-life stage after redirection from intensive to palliative care

- Discuss plan for weaning of life-sustaining support with parents
- Assure parents that a state of comfort for their baby can always be achieved, regardless of his/her condition
- Encourage parents to stay/hold the baby
- Involve chaplain as per family's wishes
- Involve siblings and extended family as per family's wishes
- Obtain pictures, videos and memories as per family's wishes
- Provide private settings with intermittent supervision of medical/nursing team
- Wean life-sustaining support, silence monitors'alarm, remove unnecessary devices and IV/arterial lines in appropriate fashion
- Maintain an IV access, if present, for pain management
- Facilitate bonding and warmth by skin-to-skin, holding, colostrum care
- Assess signs of respiratory distress (air hunger, agitation, increased work of breathing, gasping) and assess pain by validated clinical scores (NIPS or EDIN)
- Use non-pharmacological strategies (gentle suctioning upper airways, positioning) or pharmacological treatment (acetaminophen PO; morphine sulfate PO/IV; fentanyl IV; lorazepam PO/IV; midazolam PO/IV)
- After the baby dies, help parents to sponge bath and dress their baby
- Discuss with family in regard to autopsy consent

IV, intravenous; NIPS, Neonatal Infant Pain Scale; EDIN, Echelle Douleur Inconfort Nouveau-né; PO, per os.

needs; thus, we personalized the care plan in consideration of diagnosis, prognosis, gestational age, and family preferences. Additionally, as a result of a policy change in our hospital, when a neonate, prenatally diagnosed with an LLC, is not admitted to the NICU, is directly transitioned from delivery room to a post-partum for rooming-in with the mother in private setting. The midwife in charge of the mother-baby dyad provides support and care in collaboration with NICU nurses and neonatologist is available for medical evaluation and interventions.

Criteria for Enrollment

PG offers PPC to neonates prenatally or postnatally diagnosed with an LLC that is expected to render their life brief, such as renal agenesis, anencephaly or extreme prematurity. PPC is also offered to neonates diagnosed with severe genetic, metabolic, skeletal, cardiac, neurological or oncological conditions and life-limiting syndromes. The provision of intensive care can prolong the life of these neonates, but when the burden of invasive and aggressive treatment might exceed the benefit in terms of length of life, an option of PPC is offered.

Perinatal Counseling

The PGT can be accessed in different ways. If a mother receives a diagnosis of fetal LLC at SOH, the obstetric provider offers available options, including consultation with the PGT. Patients might also be referred by outside institutions. After consultation, the mother has the option to transfer her care to SOH, or to continue care with her current provider, with the PGT functioning as consultant.

Members of the PGT—including a neonatologist, an obstetrician, and a midwife—are involved in the encounter with the family. Before meeting the family, team members meet to review available information about the fetal diagnosis, discuss the need to obtain further tests and/or to involve more specialists, and identify care options to be presented to the family. It is essential that the family receives clear and honest information from both obstetricians and neonatologists.

The conversation with the family focuses on communicating of detailed information about the presumed or certain diagnosis and prognosis and, if needed, any further medical investigations. Team members listen attentively to the family to identify concerns, hopes, and expectations that will help them prepare a comprehensive plan for the reminder of the pregnancy, labor and delivery, and postnatal birthing plan. At the end of the meeting the parents are also given the option to meet a psychologist to assess mental health and to provide grief support, resources, and referrals for the family at large. Depending on the complexity of the diagnosis and the gestational age, one or more meetings are needed to complete a comprehensive plan.

The outcome of the conversation/s is documented on the mother's health record to facilitate communication with all clinicians involved in mother-baby care. Moreover, providers and family complete a document -the "birthing plain"- that is kept by the family and presented upon hospital admission.

Members of the PGT follow labor and delivery, and the neonatologist present at birth either confirms the neonate's diagnosis or obtains further tests for confirmation. The team ensures that the birth plan formulated during prenatal counseling is included in the overall management care plan.

When neonates admitted to the NICU are postnatally diagnosed with an LLC or a potentially adverse prognosis, the medical team attentively evaluates each case and may involve the PGT as an extra layer of support to families. Different options can be proposed including the continuation of intensive care with focus on quality of life, or the redirection of goals of care with transition to palliative care.

Pregnancy and Neonatal Outcomes

The PGT cared for 24 cases in its first 7 years. The team followed 20 pregnancies, the outcome of which included two intrauterine fetal deaths, two second trimester spontaneous abortions, and 16 live births. Three of these infants are currently alive and stable, and PPC was not considered medically appropriate for these infants because their postnatal diagnoses were more favorable than had been expected. The PGT also facilitated palliative care plans for four neonates admitted to the NICU with severe brain injury secondary to severe metabolic conditions or hypoxic-ischemic insults, all of whom died.

Education and Program Development

Teaching and training are essential to fostering a mindset that includes palliative care as an essential component of medical practice in perinatology. Over time the PGT built several initiatives for PPC training at SOH. Formal teaching, including a curriculum of six lessons for pediatric residents and fellows, was established in 2017. Bedside training and clinical case discussion for physicians, midwives, and nurses also have been introduced over the past 7 years.

In 2014, the concern for education and program development inspired the PGT to organize a National Workshop featuring national and international experts in PPC. The workshop goals included presentation of cutting-edge experiences of PPC and network development. The workshop was attended by some 300 clinicians from all across Italy and laid the foundation for collaboration with several academic institutions. The meeting was also attended by the Maternal Infant Department leadership and administrators. They expressed their interest and encouraged development of a PPC service at SOH. The PGT then worked tirelessly with administrators and leadership to establish a new hospital policy for PPC, which was approved in 2019.

In our region of Italy, Emilia-Romagna, a new project for the organization of a regional palliative care network has begun (15). This pediatric hospice network offers inpatient care and home care for children with complex medical conditions and chronic health problems. PG has been included in this network by the project committee. This is an essential step to build a continuity of care for neonates born with LLC or for those with complex diseases and potentially adverse prognoses. After hospital discharge, infants can be supported either at home or in a rehabilitation facility by specialists in palliative care. This project is also an opportunity to promote the palliative care mindset in our region and, hopefully, in the entire nation.

DISCUSSION

Over the past years, PPC has been gaining interest in Italy, and clinicians involved in perinatology have been working to define the best care practice (16) and to establish adequate pathways to care for neonates diagnosed with LLCs (17). Moreover, the SIN has offered suggestions for PPC in a document focused on guidelines for prevention and treatment of neonatal pain (8, 9) and Italy has enacted legislation promoting the use of palliative care (18).

PG has positioned itself as an innovative pathway for PPC origins and guidelines.

What triggered the actualization of this program was the desire of a family (Giacomo's parents) to be accompanied along the perinatal journey. The professionals involved in the care were inspired to create a standardized program for families with similar needs. The experience of PG show that a strong motivation is pivotal to the birth of a new program.

PG was modeled after the NCCP in New York. In the planning process the PGT utilized evidence-based recommendations (6, 8, 9, 11); however, the innovative aspects of the PG guidelines came largely from direct experience and ongoing collaboration with the NCCP. An important step for "culture change" at SOH is the transition of neonates with LLC from delivery room directly to post-partum to room-in with the mother, instead of admission to the NICU (**Table 1**).

Another innovative element includes the introduction of early palliative care interventions to improve quality of life of babies still receiving full intensive care (**Table 2**). Both innovations have been supported by studies of parental satisfaction (19) and stress evaluation (20) published by the NCCP.

In pregnancy care, two patients are at stake. Thus, tight collaboration of specialists in obstetrics and neonatology is essential to propose available options (1), to plan a safe pregnancy and delivery, and to provide a personalized care for each neonate in accord with the family's preference (21). Pregnancy is a time of hope and expectation, but with the discovery of severe anomalies, fear can quickly overcome hope as expectations are no longer the norm. The option of PPC supports the family and helps in the decision-making process (22, 23). Moreover, the 7-years experience of PG shows that families felt welcomed and embraced along the perinatal journey. Parents expressed of PG gratitude and praise for the program, as clearly illustrated by parental feed-back messages. *"PG was the light at the end of the tunnel, PG was the hope that our baby would have been able to be embraced by mom, dad an her siblings"*; *"PG taught us that our life cannot be measured by its lenght, rather by the mark we leave"*; *"The program filled somehow the void we experienced and we felt serene."*

The greatest desire of parents of neonates with LLC is not wanting them to suffer. Thus, pain and distress must be attentively assessed and treated (12). In the population we served any degree of discomfort was successfully managed with the use of non-pharmacological strategies, such as skin-to-skin holding, and colostrum care (24) or proper medications. This observation has been validated by studies of parental perception of their infants' state of comfort, when infants were treated with standardized comfort measures (19).

PPC not only addresses the population of neonates expected to have early perinatal demise, but also can be integrated with the intensive care of critically ill neonates at any stage of the illness (20, 25). In its first years, PG focuses largely on the care of mothers with fetal life-limiting diagnoses and followed neonates with short life expectancy however, in time the PGT began to be consulted regarding the plan of care of critically ill neonates with unexpected life-threatening diagnoses. Thus the PGT developed guidelines to facilitate quality of life during the NICU admission and/or to redirect goals of care at end of life (**Table 2**).

FUTURE DIRECTION

PG has been offering perinatologists the opportunity to improve their confidence in this growing field in medicine. Within 1 year of the program's establishment, the PGT organized a national conference. Since that time many other educational initiatives have been proposed to guide and educate providers. When PPC knowledge is lacking, clinicians are uncomfortable and reluctant to offer this much-needed care (26). Thus, PG is planning to continue to offer training courses for medical and nursing professionals built on the knowledge and experience acquired over the past 7 years.

Throughout the years, PG has followed infants with a variety of severe life-threatening conditions, including rare and complex diseases. Collaboration with other academic centers has been essential to share experiences, to discuss challenging cases, and to provide the best possible care to each infant. Further networking opportunities will support the implementation of new programs in tertiary NICUs and will provide the necessary tools to professionals working in small institutions where programs cannot be implemented.

PG intends to facilitate the spreading of a mindset that integrates palliative care in the medical plan for neonates with LLC or critically ill. Being aware that further studies measuring outcomes and quality of care are needed, our group is in the process of analyzing the efficacy of this program.

As recommended by national and international organizations (1, 6, 27), PPC is a crucial option to be offered to families facing a fetal or neonatal diagnosis of LLC. Essential components include early involvement with families at the time of diagnosis, interdisciplinary collaboration of perinatologists, and promotion of education to facilitate a culture change.

DATA AVAILABILITY STATEMENT

The original contributions presented in the study are included in the article/supplementary material, further inquiries can be directed to the corresponding author/s.

ETHICS STATEMENT

Written informed consent was obtained from the participant's parents for the publication of any potentially identifiable images or data included in this article.

AUTHOR CONTRIBUTIONS

CL conceptualized and designed the report. MB, LC, GF, LP, and GS drafted the initial manuscript and approved the final manuscript as submitted. All authors contributed to the article and approved the submitted version.

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REVIEW
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Summary of the Key Concepts on How to Develop a Perinatal Palliative Care Program

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Purpose of review: The aim of this study is to assess the most significant Perinatal Palliative Care (PnPC) development projects in the literature and summarize the shared key principles.

Recent findings: PnPC is a new concept in neonatal intensive care approach. Advancements in perinatal diagnostics and medical technology have changed the landscape of the perinatal world. The threshold of viability continues to decrease, and diagnostic information is available earlier in pregnancy and more rapidly at the bedside; overall outcomes continue to improve. This rapid technological improvement brings ethical debates on the quality of life of patients with life-limiting and life-threatening conditions and the need to involve the family in the decision-making process, according to their wishes and cultural beliefs. Although the Perinatal Hospice concept was developed in the 1980s in the US, the first recommendations on how to develop a PnPC pathway were published in the early 2000s. We considered the most relevant position statements or guidelines on PnPC published in the last two decades. Some of them were more pertinent to pediatrics but still useful for the fundamental concepts and PnPC project's development.

Summary: Health care providers and institutions are encouraged to develop PnPC programs, which have the goal of maximizing the quality of life of infants with non-curable conditions. These may generally include the following: a formal prenatal consultation; development of a coordinated birth plan between obstetrician, newborn care, and family; access to other neonatal and pediatric specialties, as needed; comfort palliative care during the prenatal, birth, and postnatal periods; and psychosocial and spiritual support for families, siblings, and staff.

Keywords: perinatal palliative care, limit of viability, life-limiting condition, life-threatening condition, program

KEY MESSAGES

- PnPC principles and guidelines have been well defined and standardized in the last two decades;
- The candidates to PnPC are well identified: fetuses/infants with life-limiting or threatening conditions that seem incompatible with long-term survival and/or that carries the risk of severe impairment of quality of life as well as prematurity that limits viability;
- Preterm infants born at the limit of viability may seem to be the most challenging candidates because there is often no time to go through the perinatal consultation process and create a birth plan; thus, conflicts could easily emerge between clinicians and staff;
- The key issue is currently the implementation of PnPC into the NICU where we can consider two models: integrative vs. consultative;
- Regardless of the strategy adopted to implement a PnPC pathway in clinical practices, it is mandatory to identify the quality indicators/outcome data and perform regular audits and case debriefing to assure quality improvement;
- Many papers are available on end-of -life care and management of pain and other distressing symptoms;
- Settings for the PnPC program are also important; home discharge and perinatal hospice should be considered in specific cases;
- Staff education is another key rule for a successful implementation of PnPC and should be constantly addressed in the PnPC program.

INTRODUCTION

A Perinatal Palliative Care (PnPC) program aims to optimize quality of life for fetus and infants with life-limiting or life-threatening conditions and their families. The goal of the program is to ensure that infants who are expected to survive only hours, days, or months do not suffer unnecessary pain and discomfort in a family-oriented environment in which parents are allowed to stay with their infant all the time with a grade of involvement based on their wishes and expectations (1).

A dedicated multi-disciplinary team that is comprehensive of all the specialists involved in the perinatal care path should continually reassess all the needs (of the body, mind and spirit) of the newborn and his family while trying to give timely and concrete answers to each of them. This is in accordance with the recent ACOG Committee Opinion #786 entitled Perinatal Palliative Care that is also endorsed by the American Academy of Pediatrics (2, 3).

Perinatal Palliative Care integrates the clinical care of the infant starting from the diagnosis of a life-limiting conditionor from a decision to withholding or withdrawn life support. This also offers an advanced care plan when the transition from intensive care to comfort care is proposed because the best interest of the infants is not prolonging life (2, 3).

Thereafter PnPC may start before birth and continue through comfort palliative care and end-of-life care. After death, there is bereavement care in a continuum of activities to support infant and parents based on their wishes, values, preferences, and spiritual needs. Counseling, debriefing, and emotional support for the staff should also be part of the care path (4).

In all of these steps, it is possible to share an integrated care plan and bundles between the professionals involved to make the process more effective always with respect for the unique individuality of the infants and its family. This study summarizes the development of a PnPC program via a narrative review of the evidence on perinatal management in the world's most industrialized countries.

METHODS

While not adhering to strict PRISMA standards, this review utilized a thorough review of Embase, PubMed, and Google Scholar and the National Bioethics Committed websites via a combination of the following search strings: practice guidelines; AND perinatal palliative care; AND limit of viability. No language restrictions were applied. All potentially relevant titles and abstracts were retrieved and assessed for eligibility.

RESULTS

We retrieved 34 full text articles from electronic databases and six from National Bioethics Committed websites. We excluded 10 articles as no pertinent with the review. Six papers manually searched were included.

Evidence From the Literature on PnPC

The application of palliative care concept to the newborn was described for the first time in the United States in 1982 (5). Brian Carter first developed a neonatal end-of-life palliative care protocol in 2002. The protocol was prepared by a 101-member panel using the Delphi research method to build a consensus document. The data collection included both participant input over an 18-month period and synthesis of 16 published and non-published end-of-life protocols developed by international, national, regional, institutional, and parent organizations. While contributors to the project mainly come from the USA, the principles and processes discussed here may be applicable to those in other countries. This work still represents a milestone in the Perinatal Palliative Care program developed all over the word although it focuses mainly on end of life care that is a part of the PnPC program (6).

In 2007, the core standards for Pediatric Palliative Care in Europe were published: IMPaCCT International meeting for Palliative Care in children. Those standards referred to pediatric populations and could also be validated for PnPC in the major points. However, PnPC is a pathway dedicated to fetus and newborns with life-limiting or life-threatening conditions, and thus this standard should be integrated with

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Abbreviations: AAP, American Academy of Pediatrics; ESPNIC, European Society for Paediatric & Neonatal Intensive Care Society; EFCNI, European foundation for the care of newborn infants; IMPaCCT, International meeting for Palliative Care in children; NICE, National Institute for Health and Care Excellence; NICU, neonatal intensive care unit; PnPC, perinatal palliative care; PPC, pediatric palliative care.

prenatal consultation and birth plans as well as decision-making on extreme prematurity and the perinatal hospice (7).

In 2013, the American Academy of Pediatrics (AAP) published a Policy Statement on Pediatric Palliative Care and Hospice Commitments, Guidelines, and Recommendations. This team considered children with life-threatening and life-shortening conditions, and they summarized 12 recommendations that should be respected when developing a PnPC program. Briefly, the transversal standards for pediatric palliative care (PPC) include the availability of a dedicated interdisciplinary team that should have sufficient expertise to address all the basic needs of the child and his family (physical, psychological, emotional, practical, and spiritual). The PPC team should provide collaborative integrated multimodal care in relation to the regional pediatrics hospices while maintaining a core level of competency. The PPC team should be consulted frequently if there is a child with life-limiting condition to provide the best and most prompt care possible and should provide communication and decision support, family support, sibling support, and health care professional support. PPC should be a core part of medical school, residency, fellowship, and continuing education curricula. The PPC team should support and engage in research, and the program should have a quality improvement agenda. PPC teams should address ethical issues and engage in relationships with the hospital's ethics committee. The PPC program may have full financial support (8).

The AAP (3), European Foundation for the Care of Newborn Infants (EFCNI) (9), Pediatric Palliative Care Group Australia (10), and national neonatal societies (11–14) have published guidelines and statements specifically regarding PnPC. **Table 1** summarizes the most important guidelines.

Categorization

Guidelines have identified three types of patients for whom PnPC may be anticipated and offered in the newborn period: (a) those born at the threshold of viability; (b) those with birth anomalies that may threaten vital functions, and (c) those for whom intensive care has been appropriately applied but developed an incurable disease (1, 6).

Multidisciplinary Approach

Most guidelines suggest a multidisciplinary approach involving an interdisciplinary team who can address all the needs of the child and family. It is important to develop an action plan via a multidisciplinary project group with the steps needed to put guidelines into practice at the local level. This may differ from setting to setting. Basically, an effective perinatal palliative care team may include obstetricians, neonatologists, health care providers with expertise in pediatric palliative care, nurses, social workers, mental health professionals, religious counselors, and experts on the child's disease (2, 9, 11, 14). The goal of this team is to provide support and continuity of care throughout pregnancy, delivery, as well as postpartum and neonatal periods. It is important to monitor the data more indicative of the requested changes as in a quality improvement process. This work should review and audit to give evidence of the guideline's implementation.

Prenatal Diagnosis

Advanced technology can suggest precociously if the fetus is carrying a life-limiting condition. The diagnosis can be made early in pregnancy with invasive or non-invasive prenatal investigations (fetal ultrasound, non-invasive prenatal test, chorionic villous sampling, amniocentesis). The condition may also be suspected and confirmed at birth.

In such cases, antenatal counseling should occur with a multidisciplinary team including at least the gynecologist, the neonatologist, the palliative care team when available, and the consultant with expertise in the child's disease (2, 9, 11, 14). The results of genetic and other diagnostic evaluations, and information about the condition of the fetus and its prognosis should be shared among the group. Different options from termination of pregnancy to palliative comfort care at birth can be presented to the family. A plan of action at birth can be created according to the family's wishes and beliefs (15).

Birth Plan

A death that is anticipated before birth affords the family the opportunity to work with the health-care team to develop both a birth plan that specifies their desires for how they want the birth to evolve and a palliative care plan. The family's participation in making these plans helps to provide them with a sense of control, and the plan honors their choices for their baby. This is worthwhile in creating a therapeutic alliance for ongoing care. This individualized proposal for delivery and neonatal care could be reviewed during the following consultations until birth. When the birth occurs, the plan should be followed regardless of which health professionals are present at the time (1, 15).

In 2016, the National Institute for Health and Care Excellence (NICE) published guidelines on end-of-life care for infants, children, and young people with the life-limiting conditions: planning and management (16).

NICE's guidelines indicated the modern general principles for developing plans for the care of the child with life-limiting or threatening conditions considering the Advance Care Plan—the preferred tool that considers the preferences and values of parents and caretakers. In the recommendations for research, the NICE committee indicated perinatal palliative care as one of the goals for future research when there is a perinatal diagnosis of life-limiting condition (16).

Postnatal Diagnosis

There is no opportunity to prepare a birth plan in the case of unanticipated loss, for an unexpected preterm birth, for a life-limiting condition diagnosed postnatally, or in case of a development of an incurable disease.

Limit of viability is a unique condition of PnPC. A systematic review published in Pediatrics in 2015 summarized international recommendations on management of preterm newborn born between 22- and 25 weeks of GA. Although there is a wide variation in clinical practice; the paper identified a gray zone between 23- and 24-weeks GA, general consensus for comfort care at 22 weeks' GA, and agreement for active care after 25 weeks' GA. While this statement may pertain to well-resourced countries, periviability may be referenced in developing or

TABLE 1 | Guidelines, position statements and opinion on perinatal palliative care worldwide.

Country	Year	Author	Methodology	Area	Topics
Australia	2018	Paediatric Palliative Care Group Australia	Guidelines	PC	- Family-centered communication and decision-making - Teamwork and coordination of care - Components of care - Use of triggers to recognize children approaching the end of life - Response to concerns - Leadership and governance - Education and training - Supervision and support for interdisciplinary team members - Evaluation, audit and feedback - Systems to support high-quality care
Canada	2016	Limbo et al.	Position statement	PnPC	- Perinatal bereavement - Care coordination - Interprofessional teamwork - Birth and advance care planning - Follow-up
Europe	2007	IMPaCCT: standards for pediatric palliative care in Europe.	Position statement	PPC	- Palliative team - Care coordinator - Pain management - Grief management - Training and education - Euthanasia
Europe	2019	EFCNI (European foundation for the care of newborn infants)	Committee opinion	PnPC	- Decisions of withholding or withdrawing life support - Communication in ethically complex decisions - Rights of infants, parents, and families in difficult decisions - Palliative care
France	2011	French Society of Neonatology and the Working Group on Ethical Issues	Position statement	PnPC	- Euthanasia is forbidden by French law - Physician may lawfully decide to withhold or withdraw a life-sustaining treatment that is unanimously considered unreasonable obstinacy. - Medical decisions must sometimes be taken for a newborn baby whose survival depends on intensive care but whose future quality of life is predicted as poor. - Reasonable treatments that should be started or continued must be distinguished from unreasonable treatments that should be stopped. - This requires assessment of the limit between a quality of life judged as acceptable for the patient and his family and a quality of life judged as unbearable.
Netherlands	2006	Verhagen et al.	Protocol	PnPC	The Groningen Protocol for Euthanasia in Newborns. Requirements that must be fulfilled: - The diagnosis and prognosis must be certain - Hopeless and unbearable suffering must be present - The diagnosis, prognosis, and unbearable suffering must be confirmed by at least one independent doctor - Both parents must give informed consent - The procedure must be performed in accordance with the accepted medical standard
UK	2014	Northern Neonatal Network UK	Guidelines	PPC	- Care before death - Care at the time of death - Care after death - Bereavement support - Staff support and supervision
UK	2015	NANN (National Association of Neonatal Nurse)	Position statement	PnPC	- Palliative care should be offered at any period in which the infant's life may be limited - When a prenatal diagnosis is made, palliative care should be offered prior to delivery. - When continuation of the pregnancy is chosen, planning and decision making for the birth will be present - All care goals should be developed as a team and include the parents. - Family conferences are essential to the providers' understanding of families' needs and their hopes and goals for their infant - An advocate or palliative care provider should be identified prenatally or at delivery/diagnosis for each infant and family in need of palliative services. - Palliative care team - Private home-like environment - Parental preferences - Local hospice - Artificial nutrition - Bereavement care - Support services to all members of the healthcare team

(Continued)

TABLE 1 | Continued

Country	Year	Author	Methodology	Area	Topics
UK	2016	NICE (National Institute for Health and Care Excellence)	Guidelines	PPC	- Advance Care Planning - Emotional and psychological support and interventions - Managing distressing symptoms, such as pain, agitation, seizures or respiratory distress - Hydration and nutrition - Recognizing that a child or young person is likely to die within hours or days - Care and support for parents, carers and healthcare professionals after the death of a child or young person - Care at home
UK	2019	Mancini et al.	Guidelines	PnPC	- Support for Staff: Building Resilience in Nurses - The Importance of Effective Communication on a Neonatal Unit - Spiritual, Cultural and Religious Care for the Baby and Family - Ethical Concepts in Neonatal Palliative Care - Legal Issues in Neonatal Palliative Care - The Principles of Genetics Within Neonatal Palliative Care - Antenatal Care for the Mother and Baby in the Context of Neonatal Palliative Care - Care of Twins, Multiple Births and Support for the Family: A Detailed Background - Care of Twins, Multiple Births and Support for the Family: The Butterfly Project - The Decision-Making Process and the Role of the Neonatal Nurse - Advance Care Planning - Organ and Tissue Donation - Continuing Emotional and Practical Support for the Whole Family - Fundamental Principles of Effective Symptom Management in Neonatal Palliative Care - Collaboration Between Hospices and Neonatal Units - Interdisciplinary Working in Neonatal Palliative Care - Developing Knowledge and Competence in Neonatal Palliative Care Practice - Care after Death
USA	2019	ACOG (American College of Obstetricians and Gynecologists)	Committee opinion	PnPC	1. Decisions about non-initiation or withdrawal of intensive care should be made by the health care team and the parents of a high-risk infant working together. This approach requires honest and open communication. Ongoing evaluation of the condition and prognosis of the high-risk infant is essential, and the physician, as the spokesperson for the health care team, must convey this information accurately and openly to the parents of the infant 2. Parents should be active participants in the decision making process concerning the treatment of severely ill infants 3. Compassionate basic care to ensure comfort must be provided to all infants, including those for whom intensive care is not being provided 4. The decision to initiate or continue intensive care should be based only on the judgment that the infant will benefit from the intensive care. It is inappropriate for life-prolonging treatment to be continued when the condition is incompatible with life or when the treatment is judged to be harmful, of no benefit, or futile
USA	2019	AAP (American Academy of Pediatrics)	Committee opinion	PnPC	
Germany	2020	PaluTIN group	Committee opinion	PnPC	10-key elements: - Focus on Needs and Hope - Empowering Parenthood - Communication - Evaluating Therapeutic Goals and Treatment Choices Together - Advance Care Planning - Comfort Care Toward the End of Life - Grief Counseling - Spirituality, Religion, and Pastoral Guidance - Support Systems - The Team Duality

PC, palliative care; PPC, pediatric palliative care; PnPC, perinatal palliative care.

middle-to-poorly resourced countries at gestational ages above 24–25 weeks EGA (17).

Guidelines usually describe a risk-based approach for interventions in the "peri-viable" period by dividing preterm infants into three categories: "*beneficial*" where intervention is generally recommended; "*gray zone*" where interventions follow the decision of parents; and "*futile*" where comfort care is recommended (18–20). This decision cannot be based only on gestational age. For example, across similar time periods, survival rates at 22 weeks' gestation in different countries vary dramatically: from zero in Switzerland and France to as high as 34% in Japan (18, 21). These differences are attributed to either a more active interventionist approach or to underreporting of perinatal deaths. The British Association of Perinatal Medicine (BAPM) proposed a risk-based approach for neonates born <27 gestational weeks based on unmodifiable (i.e., gestational age, fetal growth, sex, plurality) and modifiable risk factors (i.e., prenatal steroids, setting for birth) (22). Some guidelines suggested a more active involvement of families in the decision-making process (2). Others suggested not to resuscitate below some gestational age limit independently of the parents' wishes (17).

In the case of a life-limiting condition diagnosed or confirmed postnatally, it is important to note that the list of diseases could change from country to country and from year to year. As novel therapies emerge and change the disease outcome, management may shift from a palliative care to a more intensive management. For example, in some cases, heart surgery could be offered to children with trisomy 18 (23) and cases of children affected from trisomy 18 who survived longer than a few years have been reported (24). Nevertheless, palliative care teams remain a valuable resource in these settings because they can assist families with decision-making and provide ongoing support since some of these novel therapies are experimental, and the outcomes may be variable.

Similar recommendations are given for children who developed a poor outcome condition after experiencing prolonged intensive care treatment (e.g., severe bronchopulmonary dysplasia or massive necrotizing enterocolitis) or those newborns that cannot improve despite massive medical treatment (Apgar score of 0 at 10 min, refractory pulmonary hypertension, or refractory septic shock). In such cases, guidelines suggest creating a trusting relationship between family and team. The family should be included into the decision-making process without overwhelming them; this can shift the intensive care approach to a palliative one. These transitions may include discontinuation of mechanical ventilator support, artificial circulatory support, and/or artificial nutrition and hydration (2, 9).

Timing and Setting

Newborns can survive less than a few hours or much depending on the disease. If the diagnosis was made prenatally, then the parents are aware of the child's condition and have a birth plan. The PnPC may be applied in the delivery room. PnPC is generally offered by the neonatologist to guarantee comfort, maintain temperature, and prevent respiratory distress to the newborn (2, 15). Parents should be offered the opportunity to hold and to spend as much time as they wish with their baby in a quiet and private location. After the baby's death, a bereavement care plan should be organized for the family including communicating with parents and creating memories, giving the opportunity to dress the baby and take pictures, and making molds of handprints or footprints—of course while considering the family's spiritual, religious and cultural beliefs (15).

If a life-limiting condition is suspected or confirmed postnatally or there is a shift from intensive care to PnPC, then comfort care may be provided in the neonatal intensive care unit (NICU). Such care includes pain and symptom management, prenatal palliative care consultations, end-of-life care, communication and conflict resolution, collaboration in the care of the medically complex infant, and discharge to hospice where available (2, 9, 14, 16). Amy Kuebelbeck in her website (www.perinatalhospice.org) reported 226 perinatal hospices in the USA and 57 international programs. Perinatal hospice is a family-centered, safe, and quiet place where all of the care is focused on the quality of life of the baby and honoring parent values and wishes; in the case of discharge to home, the focus is on the education of the family on the management of the newborn (25).

Management

In 2014 Alexandra Mancini et al. "Practical guidance for the management of palliative care on neonatal units" on behalf of the Royal College of Pediatrics and Child Health (26). However, these practical guidelines do not cover the process of reaching the decision to withhold or withdraw life-sustaining treatment and has limited information on decision-making. Other toolkits and practical guidelines are available in this contest. This text focuses on the practical aspect of care including symptoms alleviation and comfort care while providing support to families and staff.

Palliative care has two fundamental concepts: withholding and withdrawing treatment (9, 27). While there may be an emotional difference between not initiating an intervention at all (withholding) and discontinuing it later in the course of care (withdrawing), there is no ethical difference between the two options (28). When an intervention no longer helps to achieve the patient's goals for care or desired quality of life, it is ethically and legally appropriate for physicians to withdraw it and redirect the care. This means that in end-of-life care, the physician can decrease and discontinue the mechanical ventilation and stop any vasoactive drugs, antibiotics, and fluid therapy. Even nutrition, although debated, can be suspended (27–29). The AAP stated that withholding or withdrawing artificial nutrition is acceptable, particularly when it's prolonging the process of dying but children capable to eat and drink safely, who wanted to eat and drink, should be given food and fluids by mouth (28).

This approach is applicable in high resource countries. In a retrospective multicentric study performed in Latin-American NICUs showed a completely different approach in end of life decision making. Withdrawing the support was no existent and usually the baby died with full life support. Preterm babies <27 gestational weeks were not admitted to the NICUs (30).

The European Standards of Care for Newborn Infant Health by EFCNI were published in 2018 and were meant as a benchmark for the development of standards in each individual country (9). They are comprehensive of evidence-based practices and qualitative outcome indicators that will significantly improve health outcomes in preterm infants in Europe and beyond. They developed four standards for key ethical decisions: decision to withhold or withdraw life support; palliative care, communication in ethically complex decisions; and the rights of infants, parents, and families in difficult decisions. They also found that the indicators of meeting the standard were aside from the evidence of a guidelines or formal setup pathway, patient information sheet, training documentations, annual report, or minutes of debriefing. Clearly, parental feedback is a critical issue/factor.

The National Association of Neonatal Nurses (NNAN) published a position statement on palliative and end-of-life care for newborn and infants in 2015 focusing on transport of infant when a redirection of care is chosen as well as nutrition and hydration and on nurse education (31).

Bidegain and Younge proposed two models of PnPC, the "*integrative model*" and the "*consultative model*" (32). The integrative model can be offered early in the pathway of every life-limiting condition or extreme prematurity. Here, the principles of "palliative care" are integrated as core element of the intensive care and after. This perspective could start before birth when a prenatal diagnosis of incurable condition has been made. It could then continue at home or in a hospice context if the child is going to survive longer. The availability of protocols, bundles, and order sets guide clinicians in the process.

In the consultative model, the palliative medicine specialist provides expert advice to the NICU staff on a child with palliative needs. Consultation may focus on same aspect of care as symptom management, consultation, and communication within the primary care team and with families. There is also withdrawal of intensive care and transition to hospice.

Thinking to the implementation of PnPC guidelines, one should consider a combined approach with palliative care principles/resources integrated within the NICU and consultation services available in situations of higher complexity (32).

Conflicts

In most situations, the care team and family agree about a desire to mitigate pain and suffering including what is in the infant's best interest and how to proceed (2, 9, 22, 26). In some situations however, ethical conflict can arise regarding the provision of potentially inappropriate or no longer beneficial care. This conflict can arise between staff members or between physicians and the family.

Conflicts between parents and staff could be avoided or mitigated involving parents from the starts, allowing time for reflexion (33) or for additional meetings, additional diagnostic tests, and second opinions (34). When the conflict cannot be mitigated, consulting the local ethics committee is suggested (2, 15).

Milk Donation and Lactation Suppression

Pregnant women with an intrauterine diagnosis of a lethal condition can plan to practice skin to skin contact and breastfeeding with their babies at the time of birth. During the postmortem period, the mothers can be supported if they decide to engage in milk donation (31, 35). Donating breast milk can have a profound influence on how bereaved women make meaning of their experience, and some women described milk donation after perinatal loss as a mechanism to facilitate their progression through grief. However, if a mother decides not to donate milk, then the option of suppressing lactation via pharmacotherapy should be offered (36).

Following the Death

Although difficult, it is mandatory to give information to the family regarding burial arrangements and final disposition of the infant's body. Verbal and written information about hospital services should be provided to the parents (2, 15). A meeting with the healthcare professionals who actually cared for their baby and parents should be arranged 1–2 months after the loss. During the meeting the clinical report should be provided (15).

Organ Donation

The issue on organ donation during the neonatal period is controversial. First, it is extremely difficult to ascertain brain death in newborns. It is unclear if giving heparin and intubation to ensure the viability of organs in a baby diagnosed with a life-limiting condition is ethical acceptable. In Italy, organ donation may be considered only term newborns more than 7 days old (>38 gestational weeks) in which brain death is demonstrated (37, 38). In UK organ donation for infants <2 months of age could be performed only if there is a neurological determination of death, which is rarely possible to confirm at this age (39). Ancillary tests, which included electroencephalogram and radionuclide cerebral blood flow study to confirm neurological death could be used in the USA, making the organ donation possible (40).

The strength of our study is that it provides an overview of the most significant aspects of PnPC and aims to provide key principles to create or implement a PnPC program. We recognize some limitation. We did not perform a systematic review but only a narrative review of the most relevant evidence on PnPC in literature and we did not include internal or unpublished guidelines of PnPC that many hospitals might have.

DISCUSSION

Advancements in perinatal diagnostics and medical technology have changed the landscape of the perinatal world. The threshold of viability continues to decrease, and diagnostic information is available earlier in pregnancy and more rapidly at the bedside. The overall outcomes continue to improve. This rapid technological improvement also brings ethical debates on the quality of life of these patients and the need to involve the family in the decision-making process.

Many professional organizations and scientific societies in industrialized countries promulgate management guidelines for these infants and newborns with poor prognosis pathologies, but the management of these patients, as well as their survival, is very heterogeneous. Survival depends on various factors including social, religious beliefs, law restrictions, and national ethics opinions (41).

In the last two decades, the principles and best practices in a PnPC program were mostly defined along with an overarching palliative care paradigm. However, the literature suggests that there are still many challenging aspects surrounding the implementation of PnPC. These are still lacking in many places were perinatal care are delivered. In our opinion, this is due to several reasons.

First, the key principles of PnPC include advancing care planning, symptom management, and psychosocial support for families and NICU staff even in the neonatal period. Help with the decision-making process and how to manage conflicts and bereavement care are quite definitely defined (2, 9, 15, 41). All of this knowledge is reported as guidelines or evidence-based practices. It is mostly "opinion of the expert" or consensus (based on Delphi methodology) with low levels of evidence. These low levels of evidence may make this research "unconvincing" and may interfere with the implementation of a PnPC.

Second, the outcomes of a "standardized" program and quality improvement initiatives are inconsistently reported. This may due to difficulties in measuring the quality of care/improvement in this setting.

Third, the fact that the PnPC is strongly focused on end-of life care limits the use of PnPC encouraging the misconception that palliative care should be reserved for only end-of-life situations. Rather, it should be considered a comprehensive and coordinated model of care—a curative treatment for fetus/neonates with life-limiting or life-threatening conditions with the goal the quality of life of the infant and family according to their wishes and values.

It is essential to give evidence of rigorous PnPC implementation process as well as evaluation of a wide range of outcome data. Only in this way we can give evidence for successful implementation of PnPC, which is still very low.

The PnPC and its benefits have been endorsed by many institutions and scientific societies (2, 9, 12, 16); however, the PnPC still lags behind other areas of palliative care when it comes to successful implementation of a quality program. Organized programs that include protocols, orders, staff education, and dedicated palliative care teams need to be addressed. The parents' experience is another area to be monitored because this might give the health care professionals insight into the benefits of the best practices in this context.

The continuity of the incurability pathway should be advised because in some situations the infants may survive months and need a different allocation of care. It is also essential that the link between regional pediatric hospice and territorial (primary) care be clear (25). During the life-limiting trajectory, discharge home with hospice support is often in the best interest of the child and their family instead of remaining in the NICU.

PnPC is a new concept in neonatal intensive care approach. Withholding or withdrawing the intensive care does not mean stopping treatments. Rather, it means redirecting active treatment to a different approach (27, 32). PnPC needs education and training of the staff to this new field of care (2, 41). All members of the health care team need to be part of the decision-making process and disagreements between staff or parents and staff needs to be resolved. Opinions from an independent clinical ethics committee may be helpful in resolving conflicts. Organ donation, disposition of the infant remains, and milk donation are elements that should be discussed in a PnPC protocol.

CONCLUSION

PnPC is a new field of newborn care. It aims to ameliorate the health status of children with life-limiting conditions. Creating a PnPC program is mandatory to guide physicians and parents through this difficult journey.

AUTHOR CONTRIBUTIONS

PL conceptualized and designed the review and drafted the initial manuscript. MC drafted the final version of the manuscript. FB aided in designing the manuscript and contributed to the content. FR reviewed the literature. All authors contributed to the article and approved the submitted version.

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BRIEF RESEARCH REPORT
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Development of a Perinatal Palliative Care Model at a Level II Perinatal Center Supported by a Pediatric Palliative Care Network

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Objective: To describe the model build up to take care of fetuses and newborns eligible to perinatal palliative care (PnPC) followed in an Italian II level perinatal center.

Methods: Retrospective chart review of all fetuses and newborn infants eligible to PnPC admitted to level II perinatal center within a 4 years period.

Results: Forty-five of 848 infants (0.5%) referred to II level NICU were eligible to PnPC. Twenty-seven percentage had fetal diagnosis. Twenty percentage were preterm infants at the limit of viability, 35% were newborns with life limiting or life threatening disease diagnosed *in utero* or at the postnatal ward, 45% were newborns not responding to intensive care intervention with high health care needs or medical complexity. Fifty-seven percentage of neonates admitted to NICU died before discharge, while 16 (35% of population considered) were discharged home. Median age at death was 4 days after birth, and delivery room death immediately after birth occurred in six patients (13%).

Conclusions: Despite the paucity of our population and the high variability in disease trajectories the perinatal palliative care program build up in our region provides a reproducible method for a structured taking in charge of fetuses and neonates eligible to PnPC and their families, from the time of diagnosis to bereavement, in both outpatient and inpatient settings.

Keywords: life limiting conditions, pediatric palliative care, perinatal palliative care, life threatening conditions, fetal diagnosis

INTRODUCTION

Perinatal palliative care (PnPC) focuses on coordinated care strategies for babies born at the edge of viability, fetuses and neonates with confirmed or potential diagnosis of life-limiting and life-threatening conditions, neonates who become critically ill during a neonatal intensive care unit (NICU) stay and are not responding to aggressive medical management (1). PnPC implies multidisciplinary, global and continuous care of the family from the time of diagnosis to the end-of-life. Initial combination of life-prolonging and disease-modifying treatment with palliative care could afterwards turn to exclusive attention to quality of life and comfort care because of inappropriateness and futility of previous treatments (2).

Lack of evidence-based empirical studies to identify the best model for PnPC (2) and the insufficient diffusion of structured PnPC programs leaves many aspects challenging in an otherwise growing field of evidence. A recent national survey reports that only 30% of Italian NICUs offer a structured PnPC program (3) despite a national law of 2010 which protects the citizen's right to access palliative care (4).

The pediatric palliative care (PPC) Network in our region, since 2016 has started to develop a program for PnPC in collaboration with level II perinatal center, considering national and international suggestions and experiences (5–10). Palliative care is disposable at any time in the pregnancy cycle: when the fetus is *in utero*, when the pregnancy is ended, when there is an early induction, after a live birth and transition to palliative care in the NICU or upon discharge home (5). PnPC planning and care is provided at the time of diagnoses on through the whole trajectory of life, and is applied indifferently in hospital settings and at home. PPC team coordinates care with hospital and local staff in a collaborative, community-based and family centered frame.

OBJECTIVE

The aim of this study was to describe the model build up to take care of fetuses and newborns eligible to perinatal palliative care and followed in an Italian II level perinatal center.

METHODS

The study population consisted of all fetuses and neonates eligible to PnPC who were cared for in the Trentino II level perinatal center from January 1, 2016 to May 31, 2020. All eligible participants were identified by examining retrospectively hospital documentation and were subsequently distributed in three groups according to current literature about eligibility to PnPC (1, 10, 11).

Group 1: newborns at the threshold of viability (birth weight $<$500 g or gestational age under 24 weeks).

Group 2: newborns with life limiting or life threatening disease diagnosed *in utero* or at the postnatal ward.

Group 3: newborns not responding to intensive care intervention with high health care needs or medical complexity.

PnPC Model Description

The PPC team can be activated at any time from diagnosis of a fetal or neonatal condition eligible to palliative care (**Figure 1**). This first contact serves to meet the parents, describe PPC network, establish a therapeutic alliance and focus on exploring parental goals.

Thereafter a multidisciplinary assessment of problems and needs takes place involving: parent(s) and members of their support system, obstetrician, neonatologist, PPC team (pediatrician, nurse, and psychologist), other specialists (neuropsychiatrist, pediatric cardiologist, and geneticist) if appropriate. Problems, needs and goals are reassessed through a model utilized in PPC (12), exploring also specific perinatal needs and tailored on each family, considering every available resource in order to achieve the endorsed goals.

The PPC team elaborates subsequently an individualized advanced birth and care plan which contains anticipatory management (**Table 1**).

A family conference is than arranged to review the individualized birth and care plan and introduce PPC network team members. Parents are given the opportunity to re-express their goals of care and they are told that the plan will be continuously reassessed. These meetings range in length from 1 to 3 h.

The plan is finally discussed with all the health care providers involved and targeted training on PPC is offered and provided to all the nodes of the network involved (obstetrician, neonatologist, nurses, psychologist, emergency services, social service, and home care service).

During prenatal, birth and postnatal periods the PPC team has repeated interactions with parents and the health care providers involved. Psychological support is provided for parents, siblings and other family members and the team caring for the fetus or neonate. This support proved to be particularly valuable in providing age-appropriate, honest and concrete explanations to young children to help them understand.

At time of delivery one of the members of the PPC team is present or available for consultation.

After delivery for neonates who survive the immediate newborn period rooming-in–if appropriate and desired–is ensured. Infants who survive long enough to be discharged from the hospital are taken care of at home, were planned physician visits take place once a fortnight and planned nurse visits twice a week. Visits of psychologists, social workers, physiotherapists are planned according to individual needs of each family. Patients and parents have a round-the-clock access to telephone consultation service, and intervention visits are a standard procedure in case of a patient's health deterioration. PPC team takes care of bereavement counseling and provides support for health workers involved who are dealing with emotional distress. PPC is free of charge and all medical equipment is loaned to the families free of charge.

RESULTS

Between January 2016 and May 2020, 9.849 neonates were born at the Trentino II level perinatal center "S Chiara" and 848 neonates were cared for at the II level NICU. The center counts an annual working volume of 2,200 prenatal diagnostic ultrasound investigations, 1,400 screening techniques for chromosomal abnormalities, 80 villocentesis and 40 amniocentesis. During the period in exam 62 voluntary interruptions of pregnancy, 60 spontaneous abortions and 15 fetal deaths took place.

Forty-five neonates (0.5% of all NICU admissions and 0.05% of all newborns) eligible to PnPC were retrospectively identified, 12 (27%) of them had fetal diagnoses, two stillbirths and 43 livebirths. Median (range) gestation at delivery was 31 (23–41) weeks. Babies survived for 1 h to 565 days (median 4 days). Deaths in the delivery room shortly after birth accounted for

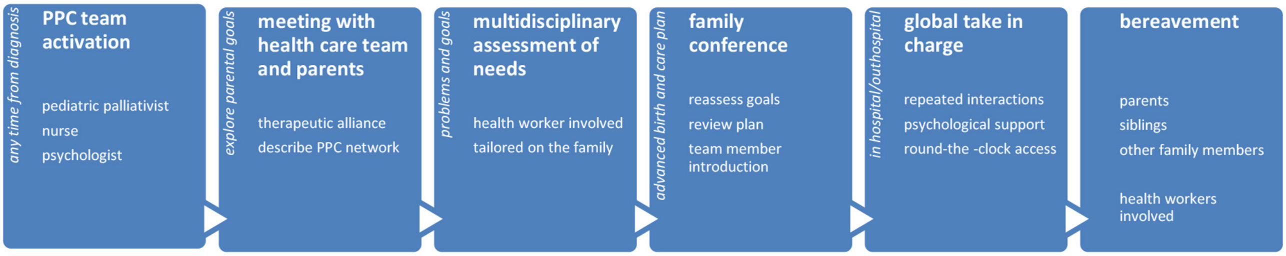

FIGURE 1 | PnPC model description.

TABLE 1 | Content of advanced birth and care plan.

Birth care plan	• Place • Timing • Mode • Monitoring in labour • People to be present at delivery • Care provided at delivery • Which (if any) diagnostic interventions to be done • Postnatal care • Skin to skin
Palliative care plan	• Pain • Medical system access and quality • Family oriented care • Dignity and respect • Decision making • Psychosocial • Spiritual symptoms treatment plan • Family support (siblings and grandparents)
Bereavement care plan	• Arranging for spiritual/cultural care • Psychosocial support • Supporting memories

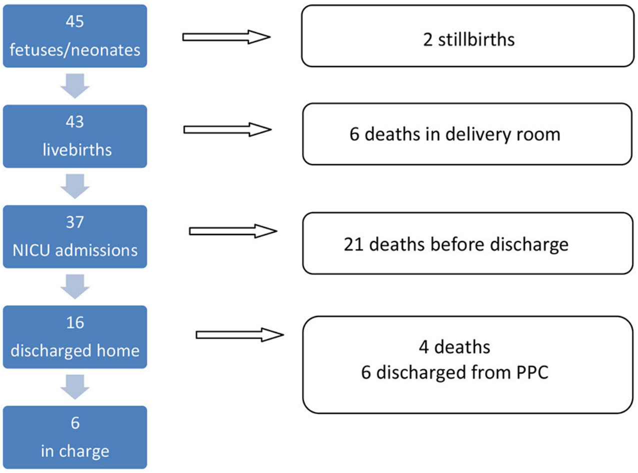

FIGURE 2 | Outcome of 45 neonates.

13%. Twenty one out of 37 neonates (57%) admitted to the NICU died before discharge (median age at death was 4 days). Of the 21 patients who died, 12 (57%) died while receiving maximal support measures (including cardiopulmonary resuscitation), and 9 (44%) died under primary comfort care or after withholding or withdrawal of life support measures. 16 (35%) were discharged home. Outcome is reassumed in **Figure 2** and main characteristics of the 45 fetuses and neonates analyzed in the study are summarized in **Table 2**.

In group 1, two out of nine newborns died in delivery room, while seven died in intensive care unit receiving support measures (median age at death was 3 days). Only in one case PPC team has been involved.

In group 2, besides two stillborns, four out of 16 newborns died in delivery room, two in NICU and eight were discharged home. Four of these eight infants died in hospital (place of death chosen by the parents) receiving comfort care at a median age at death of 111 days. Four to date are in charge of CPP network.

Group 3 includes 20 neonates, 12 of them died in NICU and eight were discharged home and taken care of by the PPC network. For six of these it has been possible to discharge them from PPC network and entrust them to the family pediatrician because of the improvement of clinical conditions and the reduction of complexity of care. Two to date are in charge of CPP network.

Altogether for 30 of the 45 fetuses and newborns eligible to PPC (67%) PPC team has been involved. The involvement grew during the period analyzed (from 50% in 2016 to 88% in 2019) and differed between group 1 and 2–3, respectively (11 vs. 80%).

The duration of PPC among all fetuses/neonates varied from 1 to 853 days with a median duration of 99 days. For those discharged home instead from 18 to 853 days with a median duration of 316 days. In this group of 16 infants the most frequent clinical problems included neurological symptoms (69%) and dysphagia (81%). A high percentage of the infants (62%) had a feeding tube (naso-orogastric) inserted, 20% percutaneous endoscopic gastrostomy and 12% tracheostomy, 40% of patients needed mechanical ventilation at discharge one patient had a central venous catheter inserted. Fifteen of all the 30 families

TABLE 2 | Distribution of neonates eligible to PnPC.

Group	1	2	3
	Newborns at the threshold of viability (birth weight < 500 g or gestational age under 24 weeks)	**Newborns with life limiting or life threatening disease diagnosed *in utero* or at the postnatal ward**	**Newborns not responding to intensive care intervention with high health care needs or medical complexity**
nr (%)	9 (20)	16 (35)	20 (45)
Died before discharge			
nr (%)	9 (100)	8 (50)	12 (60)
Age at death	*4.5 ± 4.6 (3) 0.04–13 days	*0.8 ± 1.5 (4) 0.04–4 days	*5 ± 7 (2.5) 0.4–21 days
In charge of PPC			
nr (%)	1 (11)	13 (80)	16 (80)
Duration	1day	*257 ± 205 (316) 18–494 days	*163 ± 240 (47) 1–853 days
After discharge			
Died at home	0	0	0
Died in hospital	0	4	0
Discharged from PPC	0	0	6
In charge to PPC	0	4	2
	BW 490 (415–522)	Trisomy 13, 18 $n = 4$	HIE $n = 8$
	GA 23 (23–25)	Thanatophoric dwarfism $n = 1$	Severe brain injury $n = 3$
		Potter's syndrome $n = 2$	MOF $n = 2$
		Metabolic disease $n = 2$	Dysphagia and CLD $n = 7$
		Multiple malformations $n = 4$	
		Epidermolysis bullosa $n = 1$	
		Cardiomyopathies $n = 2$	

BW, birth weight; GA, gestational age; HIE, hypossic-ischemic encephalopathy; MOF, multiorgan failure; CLD, chronic lung disease.
**Mean ± standard deviation (median) min-max.*

taken in charge agreed and received permanent psychological care and every family received social supports needed.

Eight of the 45 pathways begun at prenatal diagnosis of a life limiting or life threatening condition, three out of eight were achieved beyond 20 weeks of gestational age (13), whereas the other five diagnoses were obtained within 22 weeks of gestation but parents decided to continue pregnancy.

DISCUSSION

This study retrospectively analyzes data of all fetuses and neonates cared for in the II level perinatal center of Trento during a 4 year period from January 2016 to May 2020. Regional PPC network began to elaborate a PnPC program since 2016.

In our experience newborns belonging to group 1 rarely were notified to PPC team thus reproducing results of other experiences (11). Reasons for this may be the difficulty in acknowledging prognosis in this subgroup of patients and the reduced exposure of treating team to this kind of patient (one every 5–6 months), moreover there is to consider lack of palliative care education in medical and nursing school in our country.

Whereas, the involvement of the PPC network is growing in regard of fetuses and infants of group 2 because health professionals were previously unfamiliar with alternative management such as PnPC and the growing evidence of the importance of early referral to PnPC has been shared and recognized. Even if lethal fetal abnormalities are uncommon and most couple request termination after a diagnosis of lethal abnormality, late diagnoses will continue to arise as incidental discoveries and many of these babies will be life born with a variable and unpredictable life course (14) thus requiring adequate and comprehensive care.

Outcome of group 3 results very heterogeneous but takes in account a group of patients which can take advantage of an early referral to PnPC because of the clinical complexity of their condition. Health care providers are faced with the difficult task to revise treatment from aggressive cure-oriented intervention to palliative care goals coping with personal values and feelings.

Since the implementation of the model described, an improvement has been noted by health workers involved in terms of facilitated decision making and consensus building, improvement of therapeutic alliance, better flow of information in a framework characterized by difficult and complex matters and many "gray zones."

Taking inspiration from this first years of networking a formal education plan for possible staff members involved is in a planning stage and team support in order to manage properly communication management and emotional containment has been completed (15, 16).

NICU may not be the optimal place for provision of palliative care, but according to the evidence that location is not as important as the "mind set" of persons involved (1) and considering our experience appropriate levels of PnPC can be accomplished through attitudes, appropriate skills training and shared birth and advanced care plans.

CONCLUSIONS

At a first glance our data depict the broad variability health workers have to face in managing patients and families in need of PnPC. The vast clinical conditions with different and unpredictable trajectories of life in terms of length and burden of care requires multidisciplinarity, communication between services, forecasting of problems, flexibility and adaptation to different settings. Despite our small population and the high variability in disease trajectories the perinatal palliative care program developed in our region provides a replicable method for a structured responsibility of fetuses and neonates eligible to PnPC and their families, from the time of diagnosis to bereavement, in both outpatient and inpatient settings. Outcomes from our program support development of similar programs to meet the needs of the families who do not wish to terminate pregnancies or who do not wish to use intensive care services for babies, if born alive and as near to their home as possible.

DATA AVAILABILITY STATEMENT

The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

ETHICS STATEMENT

Ethical review and approval was not required for the study on human participants in accordance with the local legislation and institutional requirements. Written informed consent from the participants' legal guardian/next of kin was not required to participate in this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

MB, PM, and IS contributed in collecting data. CD, VF, and FU contributed in building the PnPC program. SG, BS, and FZ participated in defining the pathways. EB, GG, ST, and MS participated in manuscript review. All authors contributed to the article and approved the submitted version.

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REVIEW
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Perinatal Palliative Care: Cultural, Spiritual, and Religious Considerations for Parents—What Clinicians Need to Know

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For perinatal palliative care (PPC) to be truly holistic, it is imperative that clinicians are conversant in the cultural, spiritual and religious needs of parents. That cultural, spiritual and religious needs for parents should be sensitively attended to are widely touted in the PPC literature and extant protocols, however there is little guidance available to the clinician as to how to meet these needs. The objective of this review article is to report what is known about the cultural, spiritual and religious practices of parents and how this might impact neonates who are born with a life-limiting fetal diagnosis (LLFD). The following religions will be considered—Islam, Buddhism, Hinduism, Judaism, and Christianity—in terms of what may be helpful for clinicians to consider regarding rituals and doctrine related to PPC. Data Sources include PubMed, Ovid, PsycInfo, CINAHL, and Medline from Jan 2000–June 2020 using the terms "perinatal palliative care," "perinatal hospice," "cultur*," and "religiou*." Inclusion criteria includes all empirical and research studies published in English that focus on the cultural and religious needs of parents who opted to continue a pregnancy in which the fetus had a life-limiting condition or had received perinatal palliative care. Gray literature from religious leaders about the Great Religions were also considered. Results from these sources contributing to the knowledge base of cultural, spiritual and religious dimensions of perinatal palliative care are considered in this paper.

Keywords: perinatal palliative care, parents, religious needs, cultural needs, spiritual needs

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INTRODUCTION

Perinatal palliative care (PPC) is a specialized branch of pediatric palliative care that considers an interdisciplinary strategy for the care of newborns with life-limiting or life-threatening conditions. The World Health Organization defines palliative care as the "primary goal for the provision of a good quality of life for those with life-threatening diseases." This expands to palliative care for children and infants as a specialized field that extends to the family of children and infants. The WHO has set up principles of palliative care for children, and it includes the following points: "(a) Complete care of the infant must be taken including mind, soul, and body; (b) Moral support to the family should be provided;

(c) Palliative care should start when the decision for not providing any more intensive care has been made; and (d) Care should be implemented even when resources are limited" (1).

For PPC to be truly holistic, it is imperative that clinicians must be conversant in the cultural, spiritual, and religious needs of parents. The need for cultural, spiritual, and religious considerations to be sensitively attended to is widely touted in the literature on PPC and extant protocols; however, there is little guidance available to the clinician on meeting these needs.

In recent times, most life-limiting fetal diagnoses (LLFD) are made during the prenatal period at a time that should represent hope and joy for parents and families. When faced with an LLFD, however, these hopes and dreams of a healthy child fade. It is at the margins of life and death that people consider the existential questions of life and their spirituality, and, at times, they may even question their religious beliefs. The death of their child will lead to maladaptive grief, long-term diminished quality of life, and symptoms linked to psychological morbidity if parents are unable to reconcile these very personal needs (2). Religious, cultural, and spiritual beliefs can bring profound comfort and healing for parents faced with an LLFD, and the majority of parents want the healthcare team to have the sensitivity and skills to discuss and tend to these needs (2).

While it is not possible for the clinician to be knowledgeable about all religious tenets and cultural norms encountered in practice, having the knowledge of the most common religions and how these religious tenets apply to infants with an LLFD or that are born at the margins of viability can assist parents greatly. Sadeghi et al. (3) identified that belief in the force of the supernatural, the need for comfort of the soul, and human dignity for the newborn were the important dimensions for parents of the infants who were not expected to survive (3).

The large variations in religious practice are known from the outset, and it is beyond the reach of this review to identify and accept all of these variations exhaustively. The objective of this review is to report on what is known about the cultural, spiritual, and religious practices of parents and how this might impact the neonates who are born with an LLFD. The following religions will be considered in terms of what may be helpful for clinicians to consider regarding rituals and doctrine related to PPC: Islam, Buddhism, Hinduism, Judaism, and Christianity.In addition, cultural and spiritual needs will be addressed.

MATERIALS AND METHODS

Data sources for this review included PubMed, Ovid, PsycInfo, CINAHL, and Medline. The data from January 2000 to August 2020 were collected using the terms "perinatal palliative care," "perinatal hospice," "cultur*," "spiritual*," and "religiou*." The inclusion criteria included all empirical and research studies published in English that focused on the cultural, spiritual, and religious needs of parents who opted to continue with their pregnancy, where the fetus had a life-limiting condition or had received PPC. Results from these studies contributing to the knowledge base of cultural and religious dimensions of PPC are considered in this review. **Table 1** provides details of the types of papers included and their level of evidence.

In addition to these published papers, sources discussing the principles of the Great Religions were also included to provide a background about religious doctrine, beliefs, and current global demographics.

DISCUSSION

Religious Considerations for Parents

We live in a multicultural, multi-faith world, where religion is seemingly more important than it has ever been, with four out of five people identifying as belonging to an organized faith (16). Christianity is the fastest growing religion, with Islam and Hinduism also experiencing global growth (16). Medicine, in particular, does not accept religious concepts and practices as part of the medical model (17); however, it is increasingly important in considering how faith intersects with the biomedical and psychosocial model of health. Having one's religious and spiritual beliefs recognized brings comfort and may also promote optimism, even when facing a complex prenatal diagnosis with uncertain outcomes (14).

Religion is somewhat simpler to define than spirituality. It considers beliefs, practices, and rituals and may also include beliefs about spirits, both good (angels) and bad (demons). Religion can be observed either publicly, as part of a community, or privately, and often as both (18). It is estimated that 78% of Americans have belief in God, with another 15% believing in a "higher power" or something "bigger than themselves." In terms of prayer, 84% of Americans believe that their chance of recovery is increased by the act of praying for the sick; 79% believe in the presence of "miracles"; and 72% perceive that God can cure incurable conditions (2). Having religious beliefs can assist people by bringing some order to life when events make life untenable with the offering of rituals, traditions, and guidance. Further, the concept of an "afterlife," or existential heaven, may bring great comfort to parents faced with an infant with an LLFD.

In a discussion paper, McGuirl and Campbell (12) claimed that knowledge of the most common religious beliefs is helpful for clinicians in having end-of-life conversations with parents who are making decisions for their children, including the decision on withholding and/or withdrawing from active involvement in interventions. However, there are also studies that associate religious beliefs with an increased propensity toward life-prolonging care that may prolong the suffering of infants with life-limiting conditions (2).

In the Southeast and East Asian community, religious and spiritual values may also directly affect the pregnancy decisions linked to genetics. Leung et al. (19) stated that while Chinese women in Hong Kong generally support the termination of pregnancy in the early stages for chromosomal defects and non-medical reasons, religious context is a major contributor to the negative attitudes toward termination. Notably, most women in the study also accepted that for an undesired fetal gender, termination should not be performed. Meanwhile, the health values of some strongly traditional Southeast Asian groups concentrate

TABLE 1 | A summary of reviewed studies.

Authors/year	Purpose	Method (study design, sample, and data collection)	Main findings	Level of evidence[1]
Ayed and Ayed (4)	To provide an overview of palliative care and challenges faced by the health care system during treatment of Muslim infants.	Debate paper based in a neonatal unit in Kuwait City.	The questions posed by medical advances related to fetal and newborn palliative care require a degree of interpretation and application of the Quran by authoritative teachers (Imams).	C
Boss et al. (5)	What categorizes parental decision-making regarding delivery room resuscitation in terms of "religion," "spirituality," and "hope."	Qualitative multicenter study; Parents of infants who had died as result of prematurity or congenital anomalies. Interviews and maternal medical charts.	Some parents felt that they had not made any decisions regarding resuscitation and instead "left things in God's hands." These parents typically were documented by staff members to "want everything done." Parents and physicians had different interpretations of what was discussed and what decisions were made.	B
Caulfield et al. (6)	To evaluate trends, outcomes and characteristics of newborn infants of Catholic parents baptized over a 15-year period in an Irish maternity hospital.	Retrospective study. Patients were identified from the "register of baptisms" from the years 2002–2016.	A gradual decline of emergency baptisms over the 15-year period was reported. Maternity hospitals and neonatal units need to have access to emergency baptism service or other equivalent "spiritual blessings" as appropriate to the faiths followed by the family in an emerging multi-faith population.	B
Chichester and Wool (7)	Explores feeding issues for infants who are not expected to survive across a variety of cultures and religions: Amish, Judaism, Hindu, Muslim, African American, Hispanic-Latino and Chinese American.	Discussion paper.	Customs, beliefs and religious laws and ceremonies often involve food and are deeply connected with events, such as birth and death.	C
Das (8)	Belief in reincarnation is widespread amongst Hindus. A newborn is not a new creation, or a by-product of its parents' chemistry—it is an eternal soul continuing its progression of lives within the material world in a new body.	Best-practice guidelines.	Palliative care welcomed for newborns not expected to survive, however Hindus are wary of any intervention that might hasten death or cause it artificially. The Hindu ideal is to remove all active treatments and allow death to come to the baby in its own time.	C
El Sayed et al. (9)	To explore the challenges for trainees when EOL decisions are undertaken and to encourage them to reflect on how they might influence decision-making.	In-depth, semi-structured interview guide developed to address trainees' physician beliefs, attitudes, preferences and expectations regarding discussions of EOL neonatal care.	Dealing with different cultures and an inability to interact effectively with people of different cultures was identified as a barrier.	B
Hawthorne et al. (10)	To examine differences in parents' use of spiritual and religious coping practices by gender, race/ethnicity, and religion at 1 and 3 months after infant/ICU death.	The Spiritual Coping Strategies Scale was used to measure religious and spiritual coping practices, separately. 165 bereaved parents, 78% minority.	Black non-Hispanic mothers used more religious coping practices at 3 months than White non-Hispanic mothers. Protestant and Catholic parents used more religious coping practices than the "no" and "other" religion groups at 1 and 3 months. Within the 30 mother–father dyads, mothers reported significantly greater use of religious coping practices at 1 and 3 months and spiritual coping practices at 3 months than fathers.	B
Kain (11)	To examine the notion of being born dying and karma for Buddhists and non-Buddhists (given that karma is a belief held by both).	Discussion paper.	Spirituality and culture go hand in hand for Buddhists, and Western medicine is beginning to acknowledge the importance of spirituality in healthcare. Spiritual care for dying infants and their families is an essential component of this type of care and is not only the domain of chaplaincy services but of the entire interdisciplinary team.	C

(Continued)

TABLE 1 | Continued

Authors/year	**Purpose**	**Method (study design, sample, and data collection)**	**Main findings**	**Level of evidence**[1]
McGuirl and Campbell (12)	To provide a foundation about prominent religions so that clinicians can better understand and assess the potential religious/ moral complexities that families face in the process of making treatment decisions for their infants, including decisions regarding withholding and/or withdrawing active intervention.	Discussion paper.	Knowledge of prominent religious beliefs can be helpful as clinicians enter end of life conversations with parents. Clinicians can use this information to better understand a family's perspectives and strongly held views and can foster clerical and other spiritual support to assist families in exploring, detailing and clarifying their beliefs, values and goals.	C
Raingruber and Milstein (13)	To identify which interactions with health care providers were and were not helpful. Parents discussed what it was like to have an infant with a life-threatening illness and what helped them to cope.	Interpretive phenomenological investigation; purposive sample of 7 parents.	Parents indicated that they benefited from identifying "circles of meaning," or ways in which their infant touched another life. They appreciated hearing from health care providers that they felt close to their infant.	B
Rosenbaum et al. (2)	To increase practitioners' awareness of spiritual and existential distress and to provide strategies to address such needs, particularly at the end of life.	Literature review of the spiritual care literature and narratives of parents who have experienced the loss of their baby in our NICU.	It is important to support parents through loss by providing comprehensive care that focuses not only on the infant's physical needs, but also addresses parents' and families' spiritual, religious, and existential needs.	C
Sadeghi et al. (3)	To explore the spiritual needs of families in Iran at the end of life and through bereavement in the NICU.	An exploratory qualitative study using purposeful sampling and semi-structured interviews with 24 participants.	Data analysis revealed three main themes: spiritual belief in a supernatural power, the need for comfort of the soul, and human dignity for the newborn.	B
Seth et al. (14)	To describe the role structured religion and spirituality plays in Latinas' daily lives and to evaluate how religiosity and spirituality influences health care decisions, specifically in prenatal diagnosis.	Cross-sectional qualitative study. Semi-structured interviews with 11 women to describe religious beliefs and thoughts while considering the option of amniocentesis for prenatal diagnosis.	Latino women found their religious and spiritual beliefs comforting, providing validation for their decision regarding prenatal diagnosis, as opposed to a direct influential factor in their decision-making process. Their spiritual faith helped maintain optimism even when faced with the complexity of the amniocentesis decision-making process.	B
Shukla et al. (15)	To identify and formulate recommendations regarding challenges faced while considering redirection of care for Muslim infants based on experiences of neonatologists.	Cross-sectional survey using a web-based questionnaire: Neonatologists practicing in countries with predominantly Muslim population (Kuwait, Oman, Saudi-Arabia and Egypt).	Redirection of care consideration for Muslim infants has many socio-cultural and religious barriers. Comprehensive ethical codes conforming to Islamic and legal standards are required to aid decision-making.	B

[1] *Level of evidence.*
[A] *Body of evidence can be trusted to guide practice.*
[B] *Body of evidence can be trusted to guide practice in most situations.*
[C] *Body of evidence provides some support for recommendation(s), but care should be taken in its application.*
[D] *Body of evidence is weak, and recommendation must be applied with caution.*

on the unity of mind and body and may clash with the biomedical model practiced in the United States. Surgery, for instance, is considered a breach of the "conscience"; blood is considered irreplaceable once drawn; and prenatal treatment is considered unnecessary because pregnancy is not considered a disease (20).

It is critical that clinicians are aware of the importance of inquiring about the social and religious affiliations of a patient, as these do not always correlate with the specified religious affiliation of the patient and these are important guiding principles in perinatal decision-making.

Islam

Islam impacts all aspects of daily living: from eating to clothing to health practices. Islam has been pursued for more than 1,400 years, originating from Mohammad in Mecca, Arabia, who is believed to be the last prophet sent by God (*Allah*, translated literally as "God"). Clinicians should be aware of the Five Pillars

TABLE 2 | Islam—what clinicians need to know.

For the infant	For the parents and family
• Breastfeeding is encouraged: a Muslim child has the right to be breastfed and raised "with kindness and respect." • Anything that hastens the death of the baby, including withholding sustenance is unacceptable because the time of death is predestined by Allah. • Suffering can be interpreted as a test of faith; however, pain relief is considered acceptable. • The dying infant needs to face east, and to die facing east. • Muslims do not like to die in hospitals—the same may be true for infants and this requires discussion with parents. • Muslim adults with terminal illness prefer to stay at home with their families until they die—again, discussion with parents is required for infants. • Muslims may not consider morphine a treatment option and may believe more in Allah's potential to heal and reduce pain then on conservative medicines (i.e., Allah has the power to cure ailments.) • The burial (if the baby is viable) should normally take place as soon as possible. • Upon death, enshroud the baby's body in a white cotton or linen cloth.	• Prayer occurs five times a day: dawn to sunrise, noon, afternoon, sunset, and evening. • Prayers are performed facing east. • Muslim law permits termination of pregnancy only if continuing the pregnancy seriously impacts the mother's life. • After 19 weeks of gestation, Muslim law accepts maternal health as the only reason for terminating the pregnancy. • Muslim parents may lean more toward following the Quran rather than seeking medical advice. • Parents may be less likely to question health care providers about their child because this is interpreted as a sign of mistrust. • Muslim families may not like to be reminded of their child's illness repeatedly, or that their child may die due to a LLFD. • The Quran provides reassurance that the baby will be received into paradise and is increasingly consulted as death approaches.

Ayed and Ayed (4).

of Islam and work with families to understand how this belief may influence their decision-making when having a baby with an LLFD. For Muslims, the Five Pillars are five broad principles that allow them to live a good and responsible life. These include the following: The Declaration of Faith (*Shahada*); praying five times a day (*Salat*); giving charity (*Zakah*); fasting during the month of Ramadan (*Sawm*); and a pilgrimage to Mecca at least once (*Hajj*). It is important that healthcare providers are cognizant and do not confuse Islamic traditions with cultural traditions.

Comprised of 24.1% of the global population (21), Muslims believe that Allah is in control of both the beginning and the end of life. All outcomes, including death, are predetermined by Allah (21). The teachings of their holy book (the Quran) influences healthcare practices and the pivotal moments of life, such as birth, death, and illness (4). **Table 2** provides guidance to clinicians caring for infants of Muslim families.

For practitioners of Islam (Muslims), the pervasive belief is that curative medical interventions come from Allah, and healthcare professionals are the medium for delivering the will of Allah. The Quran provides guidance and is considered the source of healing for psychological and spiritual distress. If family members, including infants and children, require surgical or medical measures, the Quran can be used as an addendum to resolve theological, social, and cultural needs (4).

To facilitate decision-making, rigorous ethical codes are required that adhere to the Islamic social and legal standards (15). When an LLFD is made, the Quran states quite clearly that a pregnancy should not be terminated for reasons that might include financial fears or fears that the parent/s will not be able to care for their infant after birth. However, when considering extremely premature infants born at <25 weeks of gestation (12), the "legal opinion" of the Islamic society is that two specialist physicians need to be involved in the decision-making, and while for some Muslims the sole decision-maker might be the physician who is treating the infant, the parents should always be involved in the decision-making with as much autonomy afforded to them in their decision-making role. The treating physician should, however, freely consult with other pediatricians/neonatologists and caregivers.

The steps of the legal process following the diagnosis of an LLFD according to the Islamic society are as follows: confirmation of lethal malformations by ultrasound and/or chromosomal analysis; approval of the malformation by at least two neonatology and perinatology specialists; documentation of the type of malformation in the medical records of the mother; obtaining the written consent from parents or their delegates; termination of the pregnancy is permitted if the gestational age is longer than 19 weeks, but only if the continuation of the pregnancy is expected to result in the death of the mother and if fetal death *in utero* is confirmed. The infant can receive PPC if born alive (4).

In summary, clinicians need to take care in exploring with families the role of their religion in decision-making and to have some awareness of the teachings and beliefs that guide their daily lives. An understanding of their religious needs provides a framework for discussion and allows the healthcare team to support the parents through difficult decision-making processes (12).

Buddhism

Buddhism is a faith that was established more than 2,500 years ago in India by Siddhartha Gautama ("the Buddha"). Comprised of 6.9% of the global population, Buddhists recognize suffering as central to human existence. Suffering is unavoidable and life-limiting illness serves to prompt reflection on the ultimate meaning of life. Death is associated with rebirth (reincarnation), and having serene surroundings at the time of death and during the dying process is important to the dignity of the person and for their rebirth (21). Although Buddhists believe that death is a part of life, the religion is not somber, but it is full of serenity, hope, and wisdom, but recognizing that suffering is unavoidable, just as death is unavoidable. Central to this is a belief in *karma* (as is also in Hinduism), or the sum of one's actions (and inactions) in their current and previous states of existence. The sum of these karmic actions decides their fate in future existences, influencing how they will be reincarnated and what they will be reincarnated as. Karma, however, is not isolated to Hinduism and Buddhism: in America, 27% of the population were reported to have belief in reincarnation and "some form of karma" (11). In 2014, Kain

TABLE 3 | Buddhism—what clinicians need to know.

For the infant	For the parents and family
• As in Hinduism, the illness of the baby is the result of past and current actions (karma). • At death, Buddhist's believe that the person will go through a process known as "samsara," or reincarnation. Ensuring a serene setting at the time of death will be important. • Buddhists may not practice specific death rituals, however, rituals that do take place focus on helping the baby achieve a better station in their next life. • In Buddhism, burial and cremation are both practiced, so it is important not to make assumptions of this. • When creating an environment of serenity, white or yellow flowers are acceptable; red flowers are to be avoided as they symbolize happiness. • There are no rules or specific timeframe that determines when the burial or cremation of the baby should take place.	• Parent's will believe their baby could be reborn (reincarnated) as a god, demigod, human, animal, "hungry ghost" or "hell creature," depending on the baby's karma and actions during their brief life. • The parents may wish to place images of Buddha and flowers near their infant to maintain a sense of calm in the face of death. • A calm, serene environment helps to maintain the focus on religious thoughts and the "good deeds" performed during the infant's short life. • Parents may also consider the karma of their infant, and their own karma when faced with the death of their infant.

Kain (11) and Lewis and Foley (21).

explored the notion of being born after dying and karma for Buddhists and non-Buddhists, in a discussion paper, reported the dilemma of the karmic "baggage" of being born after dying. Because Buddhist scripture does not delineate at which point death occurs along the continuum, whether at the beginning of life or at the end, Buddhist parents can take comfort by realizing that their infant being reborn as a human suggests that their child already had a degree of untarnished karmic "baggage." Buddhist traditions accept human birth as a beneficial rebirth since the conditions for a sentient being to attain enlightenment are ideal only as a human being, which is the ultimate objective for the Buddhist practitioner (11).

Table 3 summarizes what clinicians need to know in terms of caring for infants of Buddhist families, especially for both the infant and the parents.

Essentially, when the death of their infant becomes imminent, Buddhist parents need to be able to focus on caring for the spiritual state of the infant rather than unnaturally prolonging the life of their child, as this will encourage a good rebirth. It is important for medication to not interfere with consciousness. When caring for Buddhist families, the clinician will benefit from demonstrating the "right understanding" (this is an appreciation of the four noble truths: the truth of suffering; the truth of the cause of suffering; the truth of the end of suffering; and the truth of the course that frees us from suffering) and an understanding of mindfulness (the whole-body-and-mind awareness of the present moment), which in turn creates good karma for the child and encourages the well-being of the child. In both its present state and as a living being, this will benefit the child in the current life and for the next life to come (11).

TABLE 4 | Christianity—what clinicians need to know.

For the infant	For the parents and family
• Baptism by water is considered necessary for salvation using a formulaic prayer, depending upon the sub-denomination. • For infants who die before baptism, the Order of Christian Funerals contains a special rite for these infants.	• Christians of all sub-denominations often have a belief in miracles, especially through prayer. • There is no firm, detailed Dogma regarding consolation of parents, however, parents will find comfort in the simple faith that God wants "what is good" for all His Children. • Emergency baptism can be carried out by staff at the request of parents if the infant is at risk of dying before the arrival of a priest/pastor.

Birgit (22), Caulfield et al. (6), and Lewis and Foley (21).

Christianity

Christianity is a monotheistic Abrahamic faith. Its ideology is based on the life and teachings of Jesus Christ of Nazareth, practiced by over 30% of the global population. Its followers, known as Christians, include many denominations: in the Western world, Roman Catholic; Protestant (Adventist, Anabaptist, Anglican, Baptist, Calvinist, Evangelical, Holiness, Lutheran, Methodist, and Pentecostal); in the East, Eastern Catholic; Eastern Orthodox; Oriental Orthodox Church of the East (Nestorian); and in Nontrinitarian, Jehovah's Witness; Latter Day Saint; and Oneness Pentecostal.

In Christianity, Jesus is considered the savior, and although beliefs vary between sub-denominations, most Christians view illness as a natural process of the body and even as a testing of their faith. While the death of an infant represents profound loss, most Christians view death as being part of the will of God (7). **Table 4** provides guidance for clinicians caring for infants of Christian families, especially for both the infant and the parents.

In a qualitative multicenter study, Boss et al. (5) described some parents whose infants had died due to severe prematurity or congenital defects as being reluctant to make any decisions regarding resuscitation. These parents were described as wanting matters "left … in God's hands," yet typically they were documented to be most likely to "want everything done." In this study, for most parents, faith, spirituality, and hope have influenced the decision-making process.

Prayer is an important component for Christians, regardless of the sub-denomination, and may be directed to one or all the holy trinity: God, the Holy Spirit, and/or Jesus Christ. Baptism, which is a Christian rite of entry into Christianity, almost always through the use of water, is another important ritual, usually performed near the time of birth. According to Birgit (22), the onus is upon parents to seek baptism if their infant dies at birth or is imminently dying as the unbaptized infant cannot "seek truth" or choose to behave in an "ethical way." Christians believe that the baptized child who dies will end up in heaven, but there are reasons for hope that, in the presence of God, children who die without baptism will still feel the joy of everlasting life (i.e., enter Heaven) (22).

Caulfield et al. (6) have recommended that, in order to fulfill the needs of families in an emerging multi-faith community, maternity hospitals and neonatal units need to have access to emergency baptism services or other similar "spiritual blessings." In a study to evaluate trends of infants of Catholic parents baptized over a 15-year period at an Irish maternity hospital, Caulfield et al. (6) reported a gradual decline of emergency baptisms.

Access to emergency baptism services is important, because, in the early stages of grief of parents whose infant had died, 37% of the parents (who were all Christian) said that their religious beliefs were challenged and that they questioned their faith (10). Parents in a study by Hawthorne et al. (10) expressed their anger and betrayal toward God (to whom they had prayed for help) for being cruel and unjust and the perception that they were being punished by His failure to protect their child; 9% said that they had lost their trust in God. While one of the most important early sacraments in Christianity is Baptism (22), it is important for clinicians to be mindful that many Christians may not subscribe to the point of view outlined in the study by Hawthorne et al.

Hinduism

Hinduism, with its origins and traditions dating back more than 4,000 years, is considered the oldest religion in the world. Hinduism is practiced by 15.1% globally and is a broad term that describes many subgroups of the religion, which influence its practice and customs. Most forms of Hinduism are henotheistic, adulating a single deity (*Brahman*), but other gods and goddesses are still acknowledged by their followers. In the scriptures and wisdom heritages of the Vedic tradition, however, all of them have their individual origins (8). Hinduism, irrespective of tradition, offers a systematic explanation of life and death that most Hindus accept. The term "life" translates to "*the atma*," which can be interpreted as the "soul" (8). In Hinduism, achieving a state of *Nirvana* (or "oneness with God") is the primary purpose. Similar to Buddhism, disease and illness are considered the result of karmic actions, and death is simply a passage because the atma has no beginning or end (21). Hindus believe in reincarnation, so a newborn is not a "new" creation but a perpetual being continuing its progression of lives in a new body.

In terms of infants born with an LLFD, the *Bhagavat Purana* (a revered Hindu text) affords personhood to the fetus at 180 days of gestation as the embryo begins to develop awareness and beings to feel pain (8). The *Dharmashastras* (the treatises of Hinduism) view the termination of pregnancy (the "killing" of a human fetus is referred to as "*bhrunahatya*") as a sin; however, in less traditional Hindu families, termination may not be seen this way.

Clinicians should be aware that more importance is placed on the male child in Hinduism, given that they continue the lineage of the family, including the name. Therefore, it is more likely that the death of a male infant will be more acutely felt in traditional Hindu families than the death of a female infant (23). In the Hindu tradition, palliative care is considered appropriate for infants who are not expected to survive. Any interventions that might hasten death are not considered appropriate, and where possible, all active treatments should be ceased and the death of the infant should be allowed to happen in its own time (8). **Table 5** provides guidance for clinicians caring for infants of Hindu families, especially for both the infant and the parents.

TABLE 5 | Hinduism—what clinicians need to know.

For the infant	For the parents and family
• As in Buddhism, the illness of the infant is the result of past and current actions (karma). • Food is viewed as a gift from God and is often part of religious rituals. Not taking nutrition near the time of death is part of the preparation for the end-of-life. • The mother may pump and discard the colostrum, as it's believed to be unsuitable for infants (in Hinduism, most infants are fed sugar water instead.) • Avoid cannulation of the right hand/arm as the right hand is considered holy to promote self-cleaning. • The parents may wish to place holy water (from the Ganges) and basil leaves on the infant's body • Once deceased, the infant's arms should be straightened. • Parents may wish to tie sacred threads around the infant's wrists or neck. • Cremation is the norm in Hinduism. However, this norm generally doesn't apply to children who die within their first year. These infants are usually buried rather than cremated, or their bodies may be consigned to rivers.	• Hindus may place more importance on male children (due to the family name and lineage); the death of a male child may be more acutely felt than the death of a female child. • The father is the head of the household, and decision making is often made at the family leve.l • Hindus believe in reincarnation, believing their child may be reborn in the same family to their children or grandchildren and return to earth. • Death should be a peaceful transition from life to death, ensuring the soul will rest in peace. • Like most, Hindus view death of a child in any family as one of the worst misfortunes imaginable.

Chichester and Wool (7), Jayaram (23), and Lewis and Foley (21).

Judaism

Judaism is practiced by <0.2% of the global population. Judaism is the oldest monotheistic religion, dating back nearly 4,000 years. The followers of Judaism believe in one God who, through ancient prophets, revealed himself. Its primary religious beliefs revolve around a code of ethics with four groupings of Jewish beliefs: reform (largest affiliation [35%] of American Jews), reconstructionist (an evolving civilization of the Jewish people), conservative ("traditional" Judaism outside of North America), and orthodox (adherence to a traditional understanding of Jewish law; this includes Haredi and Hasidic). Jewish clergy are known as *Rabbi*. Psalms, as the last prayer of confession (*vidui*), are conducted at the bedside of the person (21).

Table 6 provides guidance for caring for infants of Jewish families, especially for both the infant and the parents.

A prematurely born infant is referred to as a "*Nefel*": the same word is often applied to an abortus or an infant born with major life-incompatible defects. The word Nefel, in Jewish

TABLE 6 | Judaism—what clinicians need to know.

For the infant	For the parents and family
• Prayer shawls are common and are often passed between generations. • Food and water are considered basic needs. Withholding feeds may not be considered acceptable. However, it is appropriate to remove artificial nutrition and hydration to allow death to take its natural course if death is imminent. • Pain management is acceptable and preferable (and may be mandated) even if death is hastened. • At death, the infant's arms should not be crossed. • Where possible, any clothing or dressings with blood on them should also be prepared for burial with the infant. Jewish people believe it is important that the whole person be buried together. • Encourage the parents to see and touch their infant, photos are appropriate, and it is appropriate to collect mementos. • If the parents have not named their infant, it is appropriate to encourage them to do so. • Autopsy is appropriate (and even encouraged) if it can help determine the future pregnancies for the couple. Remember that the whole person needs to be buried, however. • The infant should be buried as soon as possible, in accordance with Jewish practice.	• All human life has value, principles such as quality of life may not contribute to the decision for curative or palliative care. • It was once thought that if the infant died before 30 days, including the full 30th day the infant is not mourned (Shulchan Aruch Yoreh De'ah 374:8), however nowadays this is mostly untrue. [The Yoreh De'ah is a section of Rabbi Jacob ben Asher's compilation of halakha, Arba'ah Turim]. • If the pregnancy was full-term, parents and other family members are generally obligated to have full bereavement practices, just as for any other child. • The parents will regard the healthcare team as being in a place of authority and may look to their doctors for their opinions regarding treatment options. • Following the death of the term infant, parents will recite Kaddish [hymn of praises about God found in Jewish prayers] for 30 days and may observe yahrzeit [the first anniversary of the death]. • In the case of premature birth, there is still debate within the Jewish community on how to define "viability." Consult with parents and their Rabbi to guide decision making. • No work can be done on the Sabbath (Friday sunset to Saturday sunset). This may result in parents delaying important decisions, including driving to the hospital to speak with the health care team.

Chichester and Wool (7) and Wolfson (24).

culture, refers to an infant who lacks sufficient hair and nail growth; is born in the eighth month of pregnancy; and survives for <30 days. Relevant social rules are different for a Nefel in Judaism compared to a well term infant (12).

It was once thought that if the infant dies, up to and including, on the full 30th day, then the infant is not mourned (Shulchan Aruch Yoreh De'ah 374:8); however, nowadays this is mostly untrue. [The Yoreh De'ah is a section of Rabbi Jacob ben Asher's compilation of halakha, Arba'ah Turim]. It has been reported that Jewish parents still feel a keen sense of loss and grief when an infant dies before the thirty-day benchmark (24).

Spiritual considerations for parents

Though used interchangeably, faith and spirituality are different from each other. Spirituality is more difficult to define because there are no common characteristics that were agreed upon. It is a broader concept that each individual defines for themselves (18). Spiritual care is distinctly different from identifying and resolving specific health problems: It is about being present in the journey of "making meaning" at the pivotal times in one's life.

In an explorative study about relevant sense-making activities, whether through prayer and/or meditation, first-time mothers established two key themes to help them deal with the life-threatening illness of their infants: (a) looking for meaning circles and (b) using intuitive/spiritual experiences to find meaning (13). Similarly, Sadeghi et al. (3) found three spiritual themes identified by parents that had lost an infant following an LLFD: a philosophical belief in the forces of the supernatural; the need for the comfort of the soul; and human dignity for the newborn (3).

To help parents make meaning and meet their spiritual needs when faced with perinatal death, the parents should be encouraged to consider how their infant has touched other lives besides their own; this includes other family members, particularly grandparents, who would have had their own hopes and dreams for the child. Raingruber and Milstein (13) propose that parents imagine the waves that circle around just one pebble tossed into a pool and that this enables parents to recognize how the life and/or death of their child form others. Parents will gain an insight into the life and death of their baby by focusing on these waves (13). These simple practices do not require parents to follow a particular religion, and comfort can be found in simple acts of making meaning. It is important to support parents through their loss by providing care that also addresses the spiritual, religious, and, existential needs of the parents and families (2).

In 2017, Hawthorne et al. explored the disparities between gender, race/ethnicity, and religion in the use of spiritual and religious coping strategies by parents after the death of a child. The study found that non-Hispanic black mothers were more likely to use religious coping strategies than non-Hispanic white mothers. Christian parents used more religious coping practices than the other groups, and mothers reported considerably more use of religious and spiritual coping practices than fathers (10). This study concluded that black mothers were more religious than they were spiritual, and the mothers were more spiritual and religious than the fathers in coping with the loss of an infant, providing some guidance to healthcare professionals on how to support bereaved parents of diverse races/ethnicities and religions. It has been identified in Latino women that their religious and spiritual beliefs were found to be intensely comforting. Their combined beliefs helped them to validate the decisions they had made in response to an LLFD. Their spirituality helped them to maintain a sense of optimism even when faced with difficult decisions following complex amniocentesis findings (14).

Cultural Considerations for Parents

Culture refers to the collective deposit of information, experience, opinions, principles, behaviors, meanings, hierarchies, faith, notions of time, responsibilities, spatial relationships, concepts of the world, and material objects and belongings acquired through person and community by a group of people over different periods of time (25).

In 2008, Davies et al. conducted a survey demonstrating that almost 40% of healthcare professionals cited "cultural differences" as a barrier to providing palliative care. This was particularly so among ethnic minorities whose cultures were poorly understood, including Latino, Indian, and Native and African Americans. Cultural gaps extended to language barriers, religious differences, and mistrust of healthcare providers and care of health practitioners who did not have the same racial or cultural context and who did not recognize these differences (26). In a study that explored the challenges for trainee neonatologists, dealing with different cultures and the inability to interact effectively with people of different cultures were identified as significant when the trainee had different faith and religious beliefs to those of the parents and families. The trainees were more likely to respond with anxiety, wariness, and even anger or fear to unknown cultures or unfamiliar cultures when working with parents that are making the end-of-life decisions about their infant (9).

No matter how committed, highly trained, and dedicated the healthcare team is, when caring for parents and their infant with an LLFD, an expert understanding of how families function, spiritually, cross-culturally, and existentially, presents a considerable challenge in PPC.

CONCLUSIONS

It is not surprising that families rely on their faith and spirituality in times of crisis to help them make sense of the path to the end of life. This analysis revealed why healthcare practitioners need to become familiar with the definitions of the moral and religious ideas that arise in their clinical field or specialization for parents and families. Regardless of the religious, spiritual, or cultural beliefs, all parents facing the death of their infant due to an LLFD require a compassionate, supportive, and knowledgeable healthcare service. Life and death are momentous occasions in the life cycle, and when the two occur in close proximity, beliefs and religious ceremonies become even more profound. All members of the healthcare team providing care to parents from the moment of the diagnosis through the palliation of to death of the infant require a unique skill set, including the ability to pay attention to, and the knowledge of, religious, spiritual, and cultural needs of the parents.

AUTHOR CONTRIBUTIONS

VK conceptualized and designed the review, drafted the manuscript, and is solely responsible for data collection and interpretation of the literature data.

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