Original Article

Experiences from the first 10 years of a perinatal palliative care program: A retrospective chart review

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ABSTRACT

Background: Perinatal palliative care is a relatively new component of paediatric palliative care which supports families who are expecting the birth of a child with a life-limiting condition. This study seeks to understand the characteristics of the infants and families referred for perinatal palliative care and the context for referrals in terms of diagnoses, referral characteristics, interventions, and outcomes.

Methods: A retrospective chart review of infants with prenatally diagnosed life-limiting conditions that were referred for perinatal palliative care.

Results: Eighty-five referrals were made for perinatal palliative care during the 10-year period, of which, 84 chose to continue with the service. Average gestational age at diagnosis was 23 weeks, and the mean time between diagnosis and referral to palliative care was 7 weeks. Stillbirths were common, occurring in 29% cases. Of livebirths, 59% of the infants survived for 2 days, and 80% died within 30 days. The most commonly referred conditions were trisomy 13 or 18 (24%), severe central nervous system malformations (20%), and severe congenital cardiac disease (16%). Referrals were most often made by neonatologists (39%), and maternal–fetal medicine specialists (36%).

Discussion: Our study confirms previously observed characteristics of diagnosis, referrals, and outcomes, while providing the most detailed account of lifespans for particular diagnoses to date. Our findings validate the need for perinatal palliative care, as 99% of those referred continued with the service. Future research should adopt a prospective approach to identify critical factors affecting decision making of families and physicians in the wake of a life-limiting diagnosis.

Keywords: End of life care; Palliative care; Perinatal care; Perinatal death; Pregnancy; Prenatal diagnosis

Perinatal palliative care is an emerging specialized model of care, providing palliative care for expectant parents who are facing the birth of a child with a life-limiting condition. Life-limiting conditions are incurable, and expected to shorten the lifespan of individuals (1). For parents who choose to continue pregnancy following the diagnosis of a life-limiting condition, perinatal palliative care supports them in re-framing their hopes and dreams for the pregnancy, preparing for the birth and death

of their baby, advocating for their child with care providers, and adjusting to life in the absence of their baby (2).

TIMING OF DIAGNOSIS AND REFERRAL

Advances in medical diagnostic technologies have improved the detection of a wide range of fetal conditions, leading to an increased number prenatally diagnosed life-limiting fetal

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conditions (1,3,4). Previous studies report that life-limiting conditions are typically diagnosed in the second trimester, most commonly between 18 and 20 weeks' gestation when most anatomical structures are well enough formed to permit an examination by trans-abdominal ultrasound (5,6). Consultation and support from a palliative care team is appropriate at the time of diagnosis, regardless of gestational age, and is welcomed by many parents (7). Despite this, there may be delays in referring patients for specialized perinatal palliative care; one study with 68 patients reported that perinatal palliative care referrals occurred at an average of 27 weeks' gestation, while the diagnosis of lethal fetal diagnosis most frequently occurred much earlier, generally between 18 and 20 weeks (8).

PARENTAL DECISIONS FOLLOWING DIAGNOSIS

After receiving a diagnosis of a life-limiting condition for their unborn child, parents must consider whether to continue the pregnancy (2,7). There is significant variation in the literature regarding the proportion of parents who choose to continue, ranging from 20 to 85% (5–10). A wide variety of conditions may lead to a referral for perinatal palliative care including congenital, genetic, pulmonary, cardiac, respiratory, neurologic, renal, and skeletal conditions (6,8). The medical teams which most frequently refer patients to perinatal palliative care have been reported in only one previous study as neonatologists (61%) and cardiologists (22%) (11). One fetal diagnostic centre, reported that only 11% of pregnancies with a life-limiting fetal diagnosis were referred, and 72% of those who were referred chose to continue with perinatal palliative care (6).

The creation of a birth plan helps parents to establish their goals for the care of their infant and helps them to anticipate the decisions they may be required to make. Birth plans also facilitate communication of the parental wishes and decisions to care teams. Birth plans often account for both still and live birth scenarios, describing the desired focus of care (comfort versus intensive care) and include decisions regarding resuscitation. In one review of perinatal palliative services, 82% of families created birth plans with the palliative care team prior to their child's birth, with 74% choosing to focus exclusively on comfort measures (8).

OUTCOMES

There is very limited evidence on outcomes for infants and families who receive perinatal palliative care; several studies have reported frequencies of stillbirth between 24 and 45% (5,8,11). Infants born alive often die soon after birth, with one study reporting that 69% died in the first 72 hours of life (5). There may be differences in the outcomes for infants referred

antenatally versus postnatally; one published review of a perinatal palliative care program over a 4-year period in Ireland found that for infants referred antenatally, 45% died prior to or during delivery, and 32% died within hours of birth (11). In the same study, for infants referred postnatally, 43% died within days, 34% survived weeks or months, and 23% remained alive at the time of data collection (11).

OBJECTIVE

The objective of this study is to describe the characteristics of infants with prenatally diagnosed life-limiting conditions who were referred for perinatal palliative care.

METHODS

Design

A retrospective chart review of palliative care referrals for prenatally diagnosed fetal life-limiting conditions over a 10-year period (October 2007 to December 2017) was performed. Data were extracted from clinical records and recorded in REDCap (an online database). Data analysis was performed using Microsoft Excel (Microsoft). We examined clinical records data related to life-limiting conditions, referral characteristics, interventions, and outcomes for patients and families.

Antenatal referrals included all cases when a pregnant woman was referred and seen by the palliative care team prior to the birth of the child. Postnatal referrals included cases when the referral occurred after the child's birth even though diagnosis of a life-limiting condition was made prior to birth. Unexpected inter-uterine fetal demise (IUFD) included cases where no fetal life-limiting condition was identified. These cases were excluded from analyses for birth planning, referral patterns, and life-limiting diagnosis, since these cases did not generate this information.

Setting

The Children's Hospital of Eastern Ontario (CHEO) palliative care team consists of paediatric palliative care physicians, nurses, and social workers. In 2007, the team started to provide perinatal palliative care services. CHEO is a 167-bed tertiary paediatric hospital and academic health sciences centre in Ottawa, Canada. The Ottawa Hospital (TOH), located directly adjacent to CHEO, provides tertiary obstetrical and perinatal services. Roger Neilson House (RNH) is an eight-bed free-standing paediatric palliative care hospice facility located on the grounds of CHEO. The CHEO paediatric palliative care team provides outreach consultation services at TOH, RNH, and in the surrounding community.

Patients and families who require perinatal palliative care are referred to the palliative care team, for support related to a fetal diagnosis of a life-limiting condition. The team collaborates with

the family to develop a birth plan which reflects the parents' wishes regarding resuscitation, pain and symptom management, and feeding. The birth plan also documents their wishes for memory-making (photos, and creation of other memorabilia), religious considerations (baptisms or blessings), autopsy, and funeral. [Supplementary Appendix A](#) shows a sample of this birth plan.

This study was approved by the Research Ethics Board of the Children's Hospital of Eastern Ontario.

RESULTS

Patient and family characteristics and choices

There were 85 families who were referred for perinatal palliative care during the study time frame of which 68% (n=58) were referred antenatally, 26% (n=22) postnatally, and 6% (n=5) at the time of an unexpected IUFD. All families referred antenatally were seen in consultation prior to the birth of their child. After an initial antenatal consultation, one family (1.7%) declined further perinatal palliative care services and instead chose to terminate the pregnancy.

The average maternal age was 32.4 years (range=14.4 to 51.9, n=85). The majority of deliveries were vaginal (n=65, 87%). Deliveries occurred at 34 weeks' gestation on average (range=19 to 41 weeks). For 32% (n=25) of mothers this was their first pregnancy. Previous pregnancy loss had been experienced by 28% (n=22) of mothers. The majority of families had at least one other child (n=52, 66%). [Table 1](#) shows further characteristics of the families in the study, as well as delivery methods.

Birth planning

Parents and clinicians collaborated to create birth plans for 67% (n=39) of the antenatal patients, and 14% (n=3) of postnatal

patients. The focus of the majority (n=49, 85%) of antenatal birth plans was comfort-focused care (i.e., palliative care). The majority (n=12, 55%) of those referred postnatally focused on intensive care and/or resuscitation. There were three patients (14%), in the postnatal group, who had a birth plan in place at the time of delivery. These plans had all been developed in collaboration with the neonatology team, without the involvement of palliative care.

Diagnosis, referral, and consultation

Antenatal referrals were most frequently made by a maternal-fetal medicine specialist (n=28, 48%), neonatologist (n=11, 19%), or clinical geneticist (n=11, 19%). Life-limiting conditions were most commonly diagnosed during the second trimester (n=57, 78%), at an average gestational age of 23 weeks (range=11 to 36, n=73). Referrals to palliative care were made at 30 weeks' gestation on average (range=13 to 40, n=80), with a mean gap between diagnosis and referral of 7 weeks (range=0 to 21, n=71). Further details of the referring clinical teams, timing of diagnosis, and referral are shown in [Table 2](#).

Types of life-limiting conditions and lifespans

The most commonly diagnosed conditions in our study included trisomy 13 or 18 (n=19, 24%), severe central nervous system malformations (n=16, 20%), and severe congenital cardiac disease (n=13, 17%). Among pregnancies that were not terminated, stillbirths were common (n=23, 29%). Of live births, over half died within 5 days (n=31, 55%), 71% (n=40) died within 30 days, and 84% (n=47) died within a year. At the time of data collection, six children (7.6%) were still living, and all had been discharged from the palliative care program due to significant improvements in their condition and prognosis. Of those discharged, the average age at the time of this study was

Table 1. Family and delivery characteristics

Maternal pregnancy characteristics (n=79 ^a)			
	0	1	≥ 2
Number of previous pregnancies	25 (32%)	20 (25%)	34 (43%)
Number of previous pregnancy losses	58 (73%)	19 (24%)	2 (3%)
Number of living children	27 (34%)	48 (61%)	4 (5%)
Delivery Characteristics (n=75 ^{a,b})			
Vaginal Birth	65 (87%)		
Caesarean Section	9 (12%)		
Termination	1 (1%)		

Family and delivery characteristics describe the families that were referred for perinatal palliative care in terms of prior pregnancies and outcomes, as well as the number of other children in the family. Delivery characteristics are also displayed. Data are expressed as number of instances (n), and percentages of instances.

^aExcludes pregnancies which were terminated (n=1), and cases of IUFD (n=5).

^bData could not be obtained for 4 cases.

Table 2. Referring service distribution, diagnosis, and referral timing

Referring Service (n=80)						
	Total		Antenatal (n=58)		Postnatal (n=22)	
	N	%	n	%	n	%
Neonatology	31	39%	11	19%	20	87%
Maternal–fetal medicine	29	36%	28	48%	1	4%
Genetics	11	14%	11	19%	0	0%
Obstetrics	3	4%	3	5%	0	0%
Primary care provider	2	3%	0	0%	2	9%
Cardiology	2	3%	2	4%	0	0%
Clinical Metabolics	1	1%	1	2%	0	0%
Midwife	1	1%	1	2%	0	0%
Timing of Prenatal Diagnoses (n=73)*						
	N					
		%		%		%
First trimester (Week 1–12)	1	1%				
Second trimester (Week 13–27)	57	78%				
Third trimester (Week 28–Birth)	15	21%				
Gestational Age at Time of Referral (n=80)						
Antenatal Referrals (n=58)				73%		
<22 weeks	8			14%		
22–26 weeks	12			21%		
27–31 weeks	16			28%		
32–36 weeks	18			31%		
37–40 weeks	4			7%		
Postnatal Referrals (n=22)				27%		

Referring service distribution, diagnosis, and referral timing describes the proportion, and timing of referrals to the perinatal palliative care team by type of service. Timing of prenatal diagnoses by trimester, as well as gestational age ranges at the time of referral. Data are expressed as number of instances (n), and percentages of instances.

*Data were not available for seven cases.

5 years. Further details about the types of conditions and the lifespans are shown in [Table 3](#).

Care settings following birth

The most common setting in which families received care after birth was the Labour and Delivery Ward (n=50, 63%), followed by the neonatal intensive care unit (NICU) (n=28, 35%). Just over a quarter (n=21, 26%) of the families referred to perinatal palliative care received care in the Paediatric Hospice setting. Other settings in which families received care following birth include the Mother Baby Unit (n=8, 10%), and at home (n=5, 6.2%). Data concerning place of death was not collected.

DISCUSSION

Access to perinatal palliative care

Almost all patients (84 out of 85) chose to pursue palliative care, demonstrating the demand for perinatal palliative care services

for families facing life-limiting diagnoses. In Canada, there are no legal limitations on termination, but in countries in which limitations exist, families may face significant time-pressure when deciding whether to pursue termination, as life-limiting diagnoses are frequently detected at 18 to 20 weeks' gestation, and the limit for termination is 23 weeks in many states in the USA (6,8). This may limit the opportunity for families to explore palliative care, since parents may have to decide to terminate shortly after receiving a life-limiting diagnosis.

Diagnosis and referral

The average time between diagnosis with a life-limiting condition and referral for palliative care was 7 weeks, similar to previous reports (5,6,8). This highlights the need for increased awareness of perinatal palliative care services among fetal care clinicians, as this may influence the decision to discuss the option of palliative care with a particular family (12). Furthermore, this significant gap may exacerbate the time-pressure described

Table 3. Lifespan and demise by type of condition (n=79^a)

Condition	n	% of total cases	Stillborn		<1 day		1–2 days		3–5 days		6–30 days		31–365 days		>365 days		Still alive	
			n	%	N	%	n	%	n	%	n	%	n	%	n	%	n	%
Trisomy 13 or 18	19	24%	6	32%	5	26%	1	5%	2	11%	3	16%	2	11%	0	0%	0	0%
Severe CNS malformations	16	20%	3	19%	2	13%	2	13%	1	6%	2	13%	1	6%	1	6%	4	25%
Severe congenital cardiac disease	13	16%	4	31%	2	15%	0	0%	1	8%	3	23%	0	0%	1	8%	2	15%
Multiple congenital abnormalities ^b	8	10%	3	38%	2	25%	1	13%	1	13%	0	0%	0	0%	1	13%	0	0%
Congenital renal conditions	5	6%	2	40%	3	60%	0	0%	0	0%	0	0%	0	0%	0	0%	0	0%
Congenital respiratory conditions	5	6%	0	0%	0	0%	1	20%	1	20%	1	20%	2	40%	0	0%	0	0%
Other (Intrauterine growth restriction, placental problems)	5	6%	3	60%	0	0%	1	20%	1	20%	0	0%	0	0%	0	0%	0	0%
Severe skeletal dysplasia	5	6%	2	40%	1	20%	2	40%	0	0%	0	0%	0	0%	0	0%	0	0%
Complications of prematurity	2	3%	0	0%	1	50%	0	0%	0	0%	0	0%	1	50%	0	0%	0	0%
Metabolic	1	1%	0	0%	0	0%	0	0%	0	0%	0	0%	1	100%	0	0%	0	0%
Overall	79	100%	23	29%	16	20%	8	10%	7	9%	9	11%	7	9%	3	4%	6	8%
Cumulative Mortality	79	100%	23	29%	39	49%	47	59%	7	68%	9	80%	7	89%	3	92%		

Lifespan and demise by type of condition summarizes stillbirths, lifespans, and demise of infants in the perinatal palliative care program by type of condition. Overall lifespan across all conditions and cumulative mortality over time for all conditions are also displayed. Data are displayed as the number of instances for a given category (n), as well as percentages.

^aExcludes pregnancies which were terminated (n=1), and cases of IUFD (n=5).

^bThese infants had significant medical conditions or malformations identified in 2 or more organ systems.

above, especially if, as one study reported, the first perinatal palliative care visit occurs an average of 14 days after the referral (8). Optimizing the timelines from diagnosis and referral is important to ensuring families receive high quality care, since families facing a life-limiting fetal diagnosis need support to face the grief and uncertainty of their situation and for their decision making (13).

Goals of care

Our findings describe an association between early referral and consultation by paediatric palliative care specialists and the creation of birth plans which focused entirely on comfort and palliative measures. Our findings are similar to other studies, where antenatal palliative care provision has been associated with the

creation of birth plans, which focus exclusively on a palliative or comfort-focused approach to care at the time of birth (5,8). For infants diagnosed prenatally with a lethal fetal anomaly, having goals of care which focus solely on comfort and quality of life is recommended (8,14). Despite these recommendations, we observed that some parents choose to proceed with intensive care interventions, despite the fact that these interventions may be anticipated to result in greater harm than benefit for the infant or prolong the dying process. Similar results have been reported in the literature, where some families choose to pursue intensive medical interventions as this provides them with comfort and peace of mind (8,14). In cases where the family chooses intensive interventions, elements of palliative care may also be provided, by ensuring that pain and other symptoms are treated and

the whole family is supported (15). Indeed the scope of palliative care in fetal conditions can be expanded beyond conditions deemed 'lethal' to also include conditions where the child's condition may be life-limiting, but a potentially life-saving treatment may be available, since in this situation providing palliative care in combination with intensive care can reduce suffering for the child and family and improve quality of life (15).

Referral patterns

Our perinatal palliative program received predominantly antenatal referrals (68%) while a similar program review from Ireland reported only 26% antenatal referrals (11). The observed differences may reflect variations in the medical practices and legal climate between settings. At the time of the Irish study, terminations were not legal, and there was limited emphasis on antenatal screening ultrasound, which may have been perceived as less relevant since the findings would not change the pregnancy outcome (11). In settings where termination is a legal option, one study found that discussion of palliative care decreased the likelihood of termination, suggesting that, when informed of the possibility of palliative care, some families will choose this type of care over termination (6). Laws, policies, and practices regarding prenatal screening and pregnancy termination in different settings may influence the antenatal referral pattern for perinatal palliative care.

Outcomes

Rates of stillbirth in our study (29%) fall within the range of previous findings from reviews of perinatal palliative care programs of 24 to 45% (5,8,11). We report an average gestational age at delivery of 34 weeks, consistent with previously reported findings showing averages of 33 to 36 weeks in cases where labour, if induced, was not pre-emptively scheduled (8,9). The outcomes of particular types of conditions in the perinatal palliative setting have not been thoroughly reported thus far. Our study is the first to provide a detailed account of lifespan and demise for each type of condition referred.

Limitations

Our study is limited as we were not able to identify those who would have been eligible for perinatal palliative services but were not referred or chose to terminate pregnancies upon receiving life-limiting diagnoses. Data collection relied on retrospective review of the medical records and did not gather information directly from patients or families. Furthermore, we were not able to assess the possible short- or long-term impacts of palliative care on patients or families.

Directions for future research

Increasing patient and provider awareness of the availability of perinatal palliative care has been identified as a priority,

and our findings suggest that research evaluating efforts to increase awareness and collaboration between providers of perinatal palliative care and referring providers, especially those in maternal–fetal medicine, neonatology, and clinical geneticist is useful. Studies comparing perinatal palliative care programs in a variety of settings and legal climates could help to identify critical factors affecting parent and clinician decision making. Future studies investigating the diagnosis, referral, and consultation process, as well as parental decisions about pregnancy termination and the type of care (intensive versus comfort-focused) to be provided to their infant are needed.

SUPPLEMENTARY DATA

Supplementary data are available at *Paediatrics & Child Health* Online.

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