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Original Article

A randomised trial of early palliative care for maternal stress in infants prenatally diagnosed with single-ventricle heart disease

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Abstract Children with single-ventricle disease experience high mortality and complex care. In other life-limiting childhood illnesses, paediatric palliative care may mitigate maternal stress. We hypothesised that early palliative care in the single-ventricle population may have the same benefit for mothers. In this pilot randomised trial of early palliative care, mothers of infants with prenatal single-ventricle diagnoses completed surveys measuring depression, anxiety, coping, and quality of life at a prenatal visit and neonatal discharge. Infants were randomised to receive early palliative care – structured evaluation, psychosocial/spiritual, and communication support before surgery – or standard care. Among 56 eligible mothers, 40 enrolled and completed baseline surveys; 38 neonates were randomised, 18 early palliative care and 20 standard care; and 34 postnatal surveys were completed. Baseline Beck Depression Inventory-II and State-Trait Anxiety Index scores exceeded normal pregnant sample scores (mean 13.76 ± 8.46 versus 7.0 ± 5.0 and 46.34 ± 12.59 versus 29.8 ± 6.35, respectively; $p = 0.0001$); there were no significant differences between study groups. The early palliative care group had a decrease in prenatal to postnatal State-Trait Anxiety Index scores (–7.6 versus 0.3 in standard care, $p = 0.02$), higher postnatal Brief Cope Inventory positive reframing scores ($p = 0.03$), and a positive change in PedsQL Family Impact Module communication and family relationships scores (effect size 0.46 and 0.41, respectively). In conclusion, these data show that mothers of infants with single-ventricle disease experience significant depression and anxiety prenatally. Early palliative care resulted in decreased maternal anxiety, improved maternal positive reframing, and improved communication and family relationships.

Keywords: CHD; hypoplastic left heart syndrome; parental stress; paediatric palliative care

CHILDREN WITH SINGLE-VENTRICLE HEART DISEASE have the highest risk of mortality of CHD patients. The majority of these patients undergo staged surgical palliation, which consists of three operations: the Norwood procedure or a systemic to pulmonary artery shunt in the first 7 days of life, a superior cavopulmonary anastomosis at 4–6 months of age, and the Fontan procedure at 12–48 months of age. The highest risk of mortality occurs during hospitalisation for the first-stage palliative surgery and the interstage period – the time between hospital discharge following the first-stage and second-stage palliative surgeries.[1,2] The initial hospitalisation for all single-ventricle patients usually involves complex care and surgery with

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cardiopulmonary bypass, a particularly stressful intervention for parents.[3] Several studies in the literature report an increased risk of psychological morbidity in mothers of children with CHD.[3–9] Providing psychosocial support may modify the development of psychosocial problems in these mothers.[4] Few studies discuss the role of paediatric palliative care in this population that receives multifaceted care.[10] Paediatric palliative care has evolved to encompass care of children with many life-threatening conditions, including CHD where survival is still a likely outcome.[11,12] Early incorporation of paediatric palliative care may improve symptom management, health-related quality of life, and communication among caregivers.[12] This study was designed to examine the benefit of early paediatric palliative care consultation on maternal stress in mothers of infants with single-ventricle heart disease. We hypothesised that maternal anxiety and depression levels would be lower in the paediatric palliative care intervention group compared with the control group, and maternal coping and health-related quality of life/family functioning would improve. This pilot study of early paediatric palliative care versus institutional standard care for neonates with single-ventricle disease is the first step in establishing standards of paediatric palliative care for infants with complex CHD, a population that accounts for a significant proportion of paediatric in-hospital deaths.[10]

Materials and methods

Study participants

Study participants included mothers referred to the University of Michigan C.S. Mott Children's Hospital Congenital Heart Center for fetal echocardiography and counselling by a paediatric cardiologist. Mothers pregnant with fetuses with single-ventricle heart disease planned to undergo the first-stage of single-ventricle palliative surgery during the neonatal period were invited to participate in the study. Participants were recruited from April, 2013 to August, 2015 by Congenital Heart Center care providers while presenting for a follow-up prenatal visit, after being referred for a fetal echocardiogram. Eligible mothers had to be English-speaking and able to comprehend and respond to the survey. Neonates born at less than 32 weeks of gestation requiring management in the neonatal ICU and neonates diagnosed with major non-cardiac anomalies requiring additional surgery beyond cardiac surgery in the neonatal period were excluded.

Study procedures

Eligible mothers were approached at a follow-up prenatal visit for recruitment, so as not to interfere with the initial diagnostic meeting/counselling, and informed consent was obtained for those mothers agreeing to participate. At the prenatal visit, a survey containing four instruments – Beck Depression Inventory-II, State-Trait Anxiety Index, Brief Cope Inventory, and PedsQL Family Impact Module – and demographic information was administered electronically to measure domains of maternal depression, anxiety, coping skills, and health-related quality of life/family functioning. The total number of survey questions was 125 with an approximate completion time of 25–40 minutes. Following survey completion, study participants' responses were reviewed for evidence of acute psychological distress, as defined by a Beck Depression Inventory-II Score of >16 or suicidal ideations, which prompted notification of the Congenital Heart Center social worker to provide additional support or referral as needed.

Upon admission for neonatal surgery – infant's date of birth, study participants were randomised by date of birth/admission in alternating blocks of three participants per block to either receive early paediatric palliative care consultation – intervention group – or institutional standard care, which included no or late palliative care consultation – control group. Nurses and physicians in the inpatient unit caring for study participants and the participants themselves were not blinded to the study arm into which they were randomised. Following birth, data were collected on all infants by chart review, including gestational age, weight, gender, cardiac diagnosis, surgery performed, co-morbidities, ICU length of stay, and hospital length of stay, for the first operative hospitalisation.

Intervention

Study participants randomised to the early paediatric palliative care intervention group were seen by the paediatric palliative care team when the neonate was admitted for planned surgery – following birth but before the first-stage palliative surgery. The initial palliative care consultation did not vary from any other palliative care consultation performed in the hospital, with a duration of 45–90 minutes. The paediatric palliative care team consisted of a physician, nurse practitioner, nurse, and social worker with paediatric palliative care specialisation and training. Although palliative care includes provision of comfort-focused end-of-life care, the intervention in this study highlights the multiple other facets of paediatric palliative care that may be beneficial to mothers of single-ventricle infants who already planned to undergo staged surgical palliation. The structured paediatric palliative care intervention specifically included evaluation of the following,

based upon accepted paediatric palliative care themes[11,13–15]: maternal understanding of their child's diagnosis and its broader impact on the child's and family's lives, concerns regarding their child's physical symptoms, social support systems and additional life stressors, expectations and hopes for their child's medical care, and fears surrounding their child's diagnosis and treatments. Each palliative care intervention addressed these themes and included particular focus on three important questions: "What is your understanding of your baby's diagnosis and how it might affect his/her and your family's lives?"; "What are you and your family hoping for?"; and "What are you most worried about?" The answers to these questions informed the paediatric palliative care team support and recommendations provided, frequency and duration of follow-up visits, and content of subsequent interactions. Follow-up visits varied from one to four ~30-minute visits per week. A standardised form was completed by a designated paediatric palliative care team provider documenting each of these areas, and the total number of contacts with the study participant was recorded.

The survey administered at the prenatal visit was administered again electronically at the time of neonatal hospital discharge (or 30 days) following the first-stage palliative surgery.

Study instruments

The following instruments, with established reliability and validity, were used.[16–20] The Beck Depression Inventory-II[16] was used to assess the severity of depressive symptoms. It includes 21 items with the score for each item on the scale ranging from 0 to 3. The State-Trait Anxiety Index[19] was used to assess current anxiety symptoms (state scale) and trait-like anxiety symptoms (trait scale). It includes 20 items for each state and trait scale (40 items total) measured on a four-point scale, with higher scores indicating higher levels of anxiety. The Brief Cope Inventory[17,18] was used to assess a broad range of coping responses. It includes 28 items consisting of subscales measuring 14 different adaptive and problematic coping reactions; scores from related items for each subscale are added together, and overall adaptive and problematic coping scores are from averaging eight and six subscales, respectively. Finally, the PedsQL Family Impact Module[20] was used to measure the overall impact of the child's cardiac diagnosis on the parent and family. It measures parent self-reported physical, emotional, social, and cognitive functioning, communication, and worry along with family daily activities and relationships. It includes 36 items measured on a five-point scale, with higher overall scores indicating

better parent functioning – less negative impact – and overall family functioning.

Data analysis

The primary outcome for this study was overall maternal stress, as reflected by measures of depression, anxiety, coping, and parental health-related quality of life/family functioning modelled after the characterisation by Rychik et al[5] and measured by the instruments as described above. Data are reported as frequency and percentage (%) for categorical variables, and mean ± standard deviation or median and interquartile range for continuous variables. Maternal and infant characteristics, prenatal and postnatal survey scores, and change in survey scores were compared between the early paediatric palliative care and the standard care groups using χ^2 or Fisher's exact test for categorical variables and t-test or Wilcoxon's rank sum test for continuous variables. Baseline prenatal Beck Depression Inventory-II and State-Trait Anxiety Index scores were also compared with normative levels from a comparative sample as published in the scale manual or from research literature using two-sample t-tests.[16,19,21,22] Changes in survey scores from prenatal and postnatal assessments were also evaluated using effect size – that is, difference in change scores between the study groups divided by standard deviation of the whole sample at baseline – in addition to p-value. All analyses were performed using SAS Version 9.4 (SAS Institute Inc., Cary, North Carolina, United States of America). p-value < 0.05 was considered statistically significant. On the basis of previous data for the PedsQL Family Impact Module,[20] an instrument designed for evaluation of a parent of a child with a chronic health condition, a sample size of 38 (19 per group) would achieve 77% power to detect a 25% mean difference in the total score between the study groups, with a two-sided significance level (α) of 0.05, for this pilot study. This study was approved by the University of Michigan Medical School Institutional Review Board. It is registered at www.ClinicalTrials.gov under the title *Impact of Early Intervention on Maternal Stress in Mothers of Fetuses Diagnosed with Single Ventricle Physiology* and identifier NCT02462434.

Results

Study participants

During the enrollment period, there were 56 eligible mothers, of whom 48 were approached and 40 consented for participation and completed baseline prenatal surveys, with an overall consent rate of 71%. Two mothers enrolled in the study were not
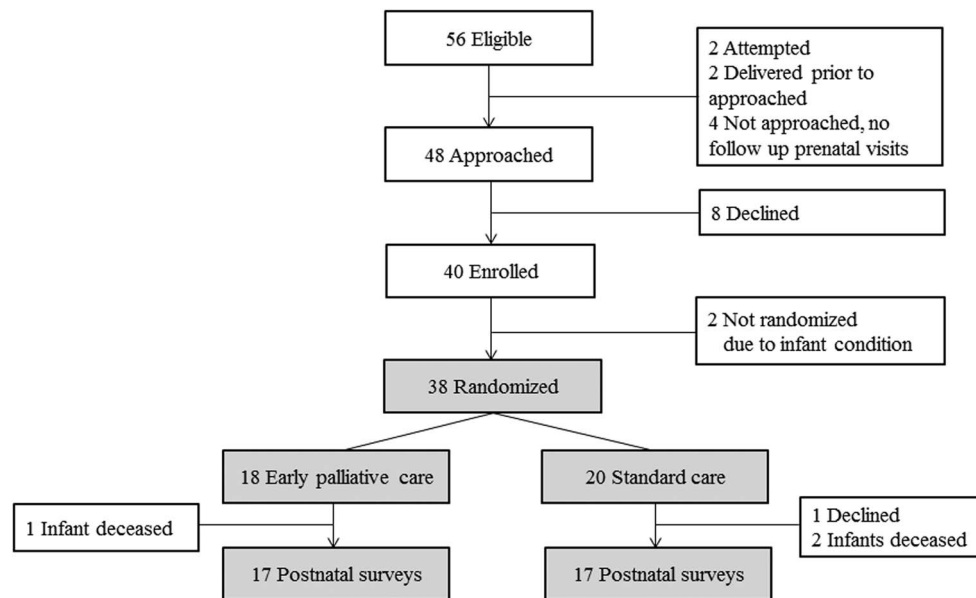
Figure 1.
Flow diagram of study participants.

randomised owing to their infants' conditions requiring immediate intervention, thus prohibiting these participants from having the early paediatric palliative care intervention as described, resulting in an actual study participant rate of 68%. In total, 38 participants were randomised, 18 in the early paediatric palliative care group, and 20 in the standard care group. In total, 17 postnatal surveys were completed in each group (Fig 1).

Baseline characteristics

There were no significant differences in the maternal demographic data between study groups (Table 1). The majority of mothers were married and had other children (71%). Baseline depression and anxiety levels, and coping mechanisms, were similar between study groups (Table 2). In total, 18 mothers (47%) had scores within the range for clinical depression on the Beck Depression Inventory-II, with 11 mothers (29%) scoring in the moderate to severe range. No mothers responded yes to the suicidal ideations question. Baseline prenatal Beck Depression Inventory-II and State-Trait Anxiety Index scores among our participants were similar in comparison with a cohort of mothers pregnant with fetuses with CHD[5] and exceeded normal pregnant sample scores[21,22] (mean 13.76 ± 8.46 versus 7.0 ± 5.0 and 46.34 ± 12.59 versus 29.8 ± 6.35, respectively; both $p = 0.0001$; Table 3). There were no significant differences in baseline maternal health-related quality of life/family functioning by the PedsQL Family Impact Module; 16 mothers (42%) scored below the clinical cut-off total score of 52.5, indicating poor parental and overall family functioning.[23]

Infants' clinical characteristics were not significantly different between the study groups (Table 4). There was a trend towards more frequent re-operations in the standard care group ($p < 0.1$). The majority of neonates (87%) had hypoplastic left ventricles and underwent a Norwood operation with a modified Blalock–Taussig shunt or central shunt (37%) or right ventricle to pulmonary artery conduit (37%) in the neonatal period. The infant sample was representative of a critically ill single-ventricle population with some infants suffering from renal dysfunction (24%), seizures (21%), stroke (18%), failed extubations (24%), cardiac re-operations or emergent sternotomies (32%), and extracorporeal membrane oxygenation (16%). There were six infant deaths (16%), three occurring before postnatal survey assessments, as seen in Figure 1, and three occurring after the study period.

Intervention data

Of the 18 mothers, 16 (89%) in the early paediatric palliative care intervention group had a "good" to "excellent" understanding of their child's diagnosis, as assessed by the paediatric palliative care team. Few mothers (17%) reported specific concerns about acute symptoms they were observing in their child, and more often discussed concerns about the future, including fears of medical instability and suffering during the infant's hospitalisation, surgical complications, poor neurodevelopmental outcome, and death. Of the 18 mothers, 11 (61%) reported

Table 1. Participants' demographics (n = 38)*.

| Characteristics | All | Intervention | | p-Value** |
		Palliative care (n = 18)	Standard care (n = 20)	
Age at enrollment (years)	27.9 ± 5.0	27.9 ± 5.8	27.9 ± 4.4	0.99
Caucasian race***	34 (89.5)	16 (88.9)	18 (90.0)	1.00
Non-Hispanic ethnicity	35 (92.1)	17 (94.4)	18 (90.0)	1.00
Highest level of education				
Less than high school	3 (7.9)	2 (11.1)	1 (5.0)	
High school diploma or equivalent (GED)	14 (36.8)	6 (33.3)	8 (40.0)	
Associate's degree (junior college)	3 (7.9)	1 (5.6)	2 (10.0)	
Bachelor's degree	12 (31.6)	6 (33.3)	6 (30.0)	0.74****
Master's degree	3 (7.9)	2 (11.1)	1 (5.0)	
Doctorate	1 (2.6)	1 (5.6)	0 (0.0)	
Other	2 (5.3)	0 (0.0)	2 (10.0)	
Marital status				
Never married	6 (15.8)	3 (16.7)	3 (15.0)	0.39*****
Married	27 (71.1)	14 (77.8)	13 (65.0)	
Living with partner	4 (10.5)	1 (5.6)	3 (15.0)	
Widowed	1 (2.6)	0 (0.0)	1 (5.0)	
Family income				
Less than $5000	6 (15.8)	3 (16.7)	3 (15.0)	
$5000–$34,999	4 (10.5)	1 (5.6)	3 (15.0)	
$35,000–$49,999	6 (15.8)	2 (11.1)	4 (20.0)	
$50,000–$74,999	9 (23.7)	5 (27.8)	4 (20.0)	0.30******
$75,000–$99,999	4 (10.5)	2 (11.1)	2 (10.0)	
$100,000 or more	7 (18.4)	4 (22.2)	3 (15.0)	
Prefer not to answer	1 (2.6)	0 (0.0)	1 (5.0)	
Unknown	1 (2.6)	1 (5.6)	0 (0.0)	
Religious preference				
Protestant	7 (18.4)	4 (22.2)	3 (15.0)	
Catholic	5 (13.2)	1 (5.6)	4 (20.0)	
Jewish	1 (2.6)	0 (0.0)	1 (5.0)	N/A
Other	12 (31.6)	7 (38.9)	5 (25.0)	
None	12 (31.6)	6 (33.3)	6 (30.0)	
Not reported	1 (2.6)	0 (0.0)	1 (5.0)	
How religious				
Very religious	7 (18.4)	4 (22.2)	3 (15.0)	N/A
Somewhat religious	16 (42.1)	7 (38.9)	9 (45.0)	
Not very religious	11 (28.9)	7 (38.9)	4 (20.0)	
Not at all religious	4 (10.5)	0 (0.0)	4 (20.0)	

GED = general educational development
*Data are presented as n (%) and mean ± standard deviation or median (interquartile range) for continuous variables
**p-Value from χ^2 test or Fisher's exact test for categorical variables and two-sample t-test or Wilcoxon's rank sum test for continuous variables
***One African American, one other (palliative care group), one American Indian, one multi-racial (standard care group)
****Comparison was made as less than or equal to high school versus associate degree or higher and p-value came from χ^2 test
*****Comparison was made as married versus all others and p-value came from χ^2 test
******Comparison was made as less than $50,000 versus $50,000 or more and p-value came from χ^2 test

additional life stressors including work, financial, family, and physical health concerns. Palliative care recommendations addressed the following: provision of resources within the hospital system such as Child Life, Spiritual Care, and simple communication between the family and healthcare providers in 15 (83%), goals of care and decision-making in 8 (44%), and complex communication in 6 (33%). The total number of contacts with the palliative care team ranged from 2 to 8 with a median of 3.

Post-intervention data

The change in survey scores from the prenatal to postnatal assessments in the early paediatric palliative care group showed a significant reduction in maternal anxiety compared with the standard care group (State-Trait Anxiety Index state scores reduced by 7.6 points versus 0.3 points in the standard care group; p = 0.02; Fig 2). By the Brief Cope Inventory, the early paediatric palliative care group had a postnatal

Table 2. Baseline Prenatal Beck Depression Inventory-II, State-Trait Anxiety Index, and Brief Cope Inventory Scores (n = 38)*.

Characteristics	All	Palliative care intervention (n = 18)	Standard care (n = 20)	p-Value**
Beck Depression Inventory-II	13.8 ± 8.5	15.0 ± 8.0	12.7 ± 8.9	0.40
Minimal depression (score 0–13)	20 (52.6)	9 (50.0)	11 (55.0)	N/A
Mild depression (score 14–19)	7 (18.4)	3 (16.7)	4 (20.0)	
Moderate depression (score 20–28)	9 (23.7)	5 (27.8)	4 (20.0)	
Severe depression (score 29–63)	2 (5.3)	1 (5.6)	1 (5.0)	
State-Trait Anxiety Index				
State anxiety	46.3 ± 12.6	48.7 ± 11.5	44.3 ± 13.4	0.29
Trait Anxiety	40.3 ± 10.3	41.3 ± 9.6	39.5 ± 13.4	0.58
Brief Cope Inventory				
Adaptive	5.6 ± 0.8	5.8 ± 0.9	5.4 ± 0.7	0.16
Active coping	5.7 ± 1.4	5.7 ± 1.5	5.7 ± 1.4	0.94
Emotional support	6.6 ± 1.4	7.0 ± 1.2	6.2 ± 1.5	0.07
Instrumental support	5.9 ± 1.6	6.2 ± 1.5	5.6 ± 1.7	0.29
Positive reframing	6.1 ± 1.5	6.2 ± 1.4	5.9 ± 1.7	0.53
Planning	6.3 ± 1.3	6.6 ± 1.0	6.0 ± 1.5	0.15
Humour	2.6 ± 0.9	2.9 ± 1.0	2.4 ± 0.7	0.06
Acceptance	6.6 ± 1.2	6.6 ± 1.2	6.6 ± 1.1	0.87
Religion	5.1 ± 1.9	5.2 ± 2.2	5.1 ± 1.8	0.79
Problematic	3.4 ± 0.9	3.5 ± 0.8	3.4 ± 0.9	0.82
Self-distraction	5.0 ± 1.8	5.4 ± 1.5	4.7 ± 2.0	0.18
Denial	3.2 ± 2.0	2.8 ± 1.6	3.5 ± 2.2	0.30
Substance use	2.0 ± 0.0	2.0 ± 0.0	2.0 ± 0.0	N/A
Behaviour disengagement	2.5 ± 0.9	2.5 ± 0.6	2.5 ± 1.1	1.00
Venting	4.3 ± 1.3	4.2 ± 1.5	4.4 ± 1.3	0.68
Self-blame	3.7 ± 1.6	3.9 ± 1.7	3.5 ± 1.6	0.41

*Data are presented as mean ± standard deviation or n (%)
**p-Value from two-sample t-test

Table 3. Comparison of prenatal psychological distress of mothers of infants with single-ventricle disease with other populations*.

Characteristics	Study cohort (n = 38)	Mothers of infants with CHD** (n = 59)	p-Value***	Mothers with healthy infants (n = 38–162)	p-Value***
Beck Depression Inventory-II	13.76 ± 8.46	11.20 ± 8.06	0.14	7.0 ± 5.0****	0.0001
State-Trait Anxiety Index					
State anxiety	46.34 ± 12.59	44.14 ± 14.69	0.45	29.80 ± 6.35*****	0.0001
Trait anxiety	40.34 ± 10.29	42.34 ± 7.72	0.28	30.92 ± 6.98*****	0.0001

*Data are presented as mean ± standard deviation
**Rychik et al[5]
***p-Value from comparison with study cohort using two-sample t-test
****Su et al[22]
*****Goodman and Tully[21]

mean score of 5.8 ± 1.0 for adaptive coping mechanisms, whereas the standard care group score was 5.1 ± 1.1 (p = 0.06) with significantly higher scores for positive reframing (6.3 ± 1.4 versus 4.9 ± 2.0 in the standard care group; p = 0.03). Finally, a positive change in perceived communication and family relationships for the PedsQL Family Impact Module was also seen in the early paediatric palliative care group – communication scores increased by 11.3 points versus 1 point in the standard care group, medium effect size of 0.46, and family relationships scores increased by 5 points versus reduced by 2.6 points in the standard care group, medium effect size of 0.41. The detailed postnatal survey results for the Beck Depression Inventory-II, Brief Cope Inventory, and PedsQL Family Impact Module are provided in the Supplementary Tables S5 and S6.

Discussion

This is the first prospective randomised clinical trial examining the effects of early utilisation of paediatric palliative care in a high-risk CHD population with risk for significant maternal psychosocial morbidity.

Table 4. Infants' characteristics (n = 38)*.

| Characteristics | All | Intervention | | p-Value** |
		Palliative care (n = 18)	Standard care (n = 20)	
Male sex	24 (63.2)	12 (66.7)	12 (60.0)	0.67
Preterm (gestational age < 37 weeks)	1 (2.6)	0 (0.0)	1 (5.0)	1.00
Birth weight (kg)	3.3 ± 0.5	3.3 ± 0.4	3.3 ± 0.6	0.73
Ventricle type				
Single left ventricle	5 (13.2)	2 (11.1)	3 (15.0)	1.00
Single right ventricle	33 (86.8)	16 (88.9)	17 (85.0)	
Fetal intervention	1 (2.6)	1 (5.6)	0 (0.0)	0.47
Surgery type (NOT mutually exclusive)				
Norwood w/mBT or central shunt	14 (36.8)	7 (38.9)	7 (35.0)	
Norwood w/RV-PA conduit	14 (36.8)	8 (44.4)	6 (30.0)	
mBT or central shunt only	3 (7.9)	0 (0.0)	3 (15.0)	N/A
Hybrid stage I palliation	2 (5.3)	0 (0.0)	2 (10.0)	
Other	6 (15.8)	2 (11.1)	4 (20.0)	
No surgery; pre-op death	1 (2.6)	1 (5.6)	0 (0.0)	
Renal dysfunction	9 (23.7)	5 (27.8)	4 (20.0)	0.71
Seizure	8 (21.1)	4 (22.2)	4 (20.0)	1.00
IVH/cerebral thrombosis or ischaemia	7 (18.4)	4 (22.2)	3 (15.0)	0.69
Chylothorax	6 (15.8)	3 (16.7)	3 (15.0)	1.00
Failed extubation	9 (23.7)	2 (11.1)	7 (35.0)	0.13
Tracheostomy	3 (7.9)	0 (0.0)	3 (15.0)	0.23
Cardiac re-operation/emergent sternotomy	12 (31.6)	3 (16.7)	9 (45.0)	0.06
Emergent sternotomy	7 (18.4)	1 (5.6)	6 (30.0)	0.09
Re-operation requiring CPB	4 (10.5)	3 (16.7)	1 (5.0)	0.33
Other cardiac re-operation	4 (10.5)	1 (5.6)	3 (15.0)	0.61
ECMO	6 (15.8)	3 (16.7)	3 (15.0)	1.00
Cardiac arrest	9 (23.7)	3 (16.7)	6 (30.0)	0.45
Initial length of intubation (days) (n = 36)	5 (4–8)	5 (4–7)	5 (3–9)	0.84
Total length of intubation (days) (n = 36)	7 (4–14.5)	5 (4–8)	8 (4–17)	0.17
Initial ICU length of stay (days)	16 (12–23)	15 (12–18)	19 (12–23.5)	0.22
Total ICU length of stay (days)	16 (12–25)	15 (12–18)	20 (12–42.5)	0.19
Hospital length of stay (days)	29 (19–44)	25 (19–31)	32 (23–79)	0.13
Deceased	6 (15.8)	2 (11.1)	4 (20.0)	0.66

CPB = cardiopulmonary bypass; ECMO = extracorporeal membrane oxygenation; HD = haemodialysis; IVH = intraventricular haemorrhage; mBT = modified Blalock–Taussig; PA = pulmonary artery; RV = right ventricle

*Data are presented as n (%) for categorical variables and mean ± standard deviation or median (interquartile range) for continuous variables

**p-Value from χ^2 test or Fisher's exact test for categorical variables and two-sample t-test or Wilcoxon's rank sum test for continuous variables

We have shown that these mothers experienced depression and anxiety and poor parental and overall family functioning prenatally. We randomised this representative group of mothers of single-ventricle infants to receive early paediatric palliative care versus standard care and subsequently found that the early paediatric palliative care intervention resulted in decreased maternal anxiety, improved maternal positive reframing, and improved communication and family relationships, suggesting decreased overall maternal stress.

In a recent study by Rychik et al, post-traumatic stress, depression, and anxiety were found to be common in mothers following prenatal diagnosis of CHD.[5] Our prenatal survey data of mothers of fetuses diagnosed with single-ventricle CHD in particular demonstrate similarly elevated levels of maternal depression and anxiety, with scores that exceed those of a normal pregnant sample. It is well known that parents of children with CHD experience increased stress at the time of their child's diagnosis and initial hospitalisation,[7] and the prevalence of parental depression, anxiety, and hopelessness, along with long-standing psychosocial morbidity, is high.[4] The added stress of heart surgery beyond the diagnosis of CHD has been determined significant for parents as well. A report of parents of children undergoing cardiopulmonary bypass surgery showed increased rates of acute surgery-related post-traumatic stress disorder (16.4% in mothers).[3] Significant determinants of overall psychological morbidity in parents of children with CHD include parental perception of, and satisfaction with, hospital care, burden of care, social support systems, and financial hardships. Providing psychosocial support and resources for parents in this environment may modify the development of significant psychosocial malfunction over time.[4]
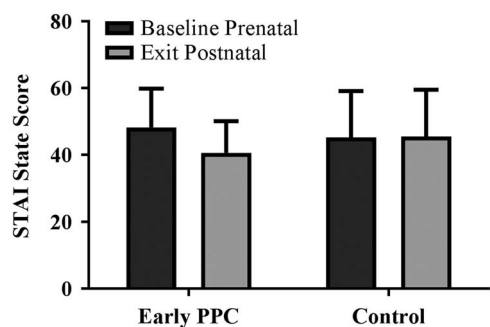
Figure 2.
State-Trait Anxiety Index State scores. Twenty-item self-report questionnaire assessing anxiety symptoms. Higher scores indicate higher levels of anxiety (score range = 20–80). Mean scores ± standard deviation are shown. There was a significant decrease in prenatal to postnatal State-Trait Anxiety Index scores in the early paediatric palliative care group (mean change −7.6 versus +0.3 in control {standard care} group; p = 0.02). PPC = paediatric palliative care; STAI = State-Trait Anxiety Index.

Despite many of the mothers in our study having a "good" to "excellent" understanding of their child's CHD and risk following birth, prenatal surveys indicated elevated levels of maternal anxiety and depression, which probably continue as they near their newborn's surgery and long after. These mothers expressed significant fears regarding the future for their children with complex CHD, with neurodevelopmental outcomes being a particular focus. These parental fears, which likely contribute to the development of anxiety and depression, are important to assess; however, they may be overlooked when providing care for neonates with complex disease. Caris et al recently showed that anxiety/stress scores among caregivers of children with hypoplastic left heart syndrome correlate with parental perception of the child's physical and developmental wellbeing, and that caregivers with concerns in these areas may need additional psychosocial support.[24]

Many features of paediatric palliative care address determinants of parental psychological morbidity, and paediatric palliative care may serve as a useful adjunct to care provided for children with complex CHD. Although several studies examine the impact of early palliative care for adult cancer patients regardless of prognosis,[25–29] few paediatric studies demonstrate the benefits of early paediatric palliative care, particularly in paediatric chronic disease populations not near the end of life and the CHD population.[30] Recent studies investigating paediatric palliative care impact in neonatal and paediatric cancer patients show improved symptom control and less child suffering by parental report, identification and help with family communication needs, and improved quality and coordination of care by addressing families' complex care needs.[31–33] To our

knowledge, studies evaluating the benefit of paediatric palliative care specifically on overall maternal stress including psychological distress, maternal coping, and overall family functioning are not available in the literature. In our study, we demonstrate a significant reduction in anxiety scores in mothers who were provided with early paediatric palliative care consultative services throughout the hospitalisation of their newborns with single-ventricle disease. Aspects of the paediatric palliative care intervention, which resulted in reduction of maternal anxiety, are difficult to understand and warrant further study. However, these mothers demonstrated improved positive reframing – an adaptive coping mechanism – which is emphasised by palliative care.[11,26] Finally, improved perceived maternal communication and family relationships were also seen in the early paediatric palliative care group by the PedsQL Family Impact Module, suggesting that emphasis in these areas may also play a role in improved overall maternal stress. We do note the trend for the standard care group neonates to have more cardiac reoperations that did not reach statistical significance, although may have been of clinical significance and could have an impact on overall maternal stress for the standard care group at the follow-up postnatal survey.

Although our study has numerous strengths, it has several limitations worth mentioning. It was performed at a single, tertiary care centre with a specialised group of paediatric cardiology and palliative care providers, and a single-ventricle population skewed towards infants with hypoplastic left heart syndrome, and in predominantly Caucasian mothers, all English-speaking, limiting the generalisability of the study results to other care centres and settings. There was a small sample size available for participation; however, with this pilot study we gained important information for a larger, randomised controlled trial. Incomplete data on eligible non-participants and variation in participant counselling provided by multiple providers during the prenatal visits – especially when the initial diagnostic meeting occurred at an outside institution – and during postnatal hospitalisation are also limitations. Additionally, the Beck Depression Inventory-II and State-Trait Anxiety Index are symptom scales, and although used to aid in clinical diagnosis of psychiatric diseases they are not considered diagnostic. We aimed to assess for the multiple demographic and socio-economic factors that contribute to the development of psychosocial morbidities; however, assessing for all of these potential factors is likely not possible. Finally, as can be seen in survey research, the potential for social desirability bias is present; however, maternal anxiety and depression levels were found to be elevated in our study, suggesting that

these components of overall maternal stress may in fact be higher than our study participants reported.

Areas of future investigation include incorporation of paternal data and the impact of early paediatric palliative care on paternal stress along with partner relationships and communication. In our study, we chose to evaluate the impact of the paediatric palliative care intervention following birth; however, evaluation of such an intervention beginning in the prenatal period would also be an area of future interest. Finally, evaluation of the impact of a palliative care intervention on infant short- and long-term neurodevelopmental outcomes is an important area of future study.

In conclusion, this is the first study to investigate the benefits of early paediatric palliative care consultation for maternal stress in the single-ventricle heart disease population, which has the highest risk of mortality and receives complex CHD care. Recognition of psychological morbidity in these mothers is extremely important when providing care. Our results in this population indicate that paediatric palliative care can lower maternal anxiety levels, increase maternal positive reframing, and improve communication and family relationships, determinants of overall maternal stress. Although these findings need to be verified in a larger population, across multiple sites, this pilot study lays the framework for designing a more definitive, multicentre randomised clinical trial to evaluate the efficacy of incorporating paediatric palliative care into the standard of care for neonates with single-ventricle heart disease.

Supplementary material

To view supplementary material for this article, please visit https://doi.org/10.1017/S10479511 17002761

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