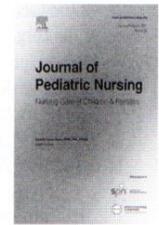




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Parents' quality of life and health after treatment decision for a fetus with severe congenital heart defect

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ABSTRACT

Purpose: This exploratory study examines differences in parents' quality of life by treatment decision and the child's survival outcome in the context of life-threatening congenital heart disease (CHD).

Design and methods: Parents of a fetus or neonate diagnosed with severe CHD enrolled in the observational control group of a clinical trial (NCT04437069) and completed quality of life (i.e., contact with clinicians, social support, partner relationship, state of mind), mental and physical health survey measures. Comparisons were made between parents who chose comfort-directed care or surgery and between those whose child did and did not survive.

Results: Parents who chose surgery and their child did not survive reported the most contact with their clinicians. Parents who chose comfort-directed care reported lower social support than parents who chose surgery and their child did not survive as well as poorer state of mind compared to parents who chose surgery.

Conclusions: Some aspects of parents' quality of life differed based on their treatment decision. Parents who choose comfort-directed care are vulnerable to some negative outcomes.

Practice implications: Decision support tools and bereavement resources to assist parents with making and coping with a complex treatment decision is important for clinical care.

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Introduction

Parents who have a fetus or neonate diagnosed with a life-threatening congenital heart defect (CHD) experience significant psychological distress (Biber et al., 2019; Wei et al., 2015; Woolf-King et al., 2017). While most CHDs can be repaired and result in good quality of life for the child, a subset of severe, life-threatening CHD diagnoses require early, intensive intervention, with low survival rates

(e.g., 12.5% 5-year survival for hypoplastic left heart) (Best & Rankin, 2016; Oster et al., 2013). Parents of a fetus or neonate diagnosed with severe CHD face an intense treatment decision with lifelong consequences – whether to pursue or forgo potentially life-sustaining interventions (Andrews et al., 2006). To inform counseling and support for parents faced with this monumental decision, it is crucial to understand what impact different treatment decisions have on parents' quality of life and health.

A severe CHD diagnosis in a fetus or neonate imposes significant stress and emotional burden on parents who must cope with lost expectations of a healthy child while simultaneously making a life-changing treatment decision (Alkan et al., 2017; Brosig et al., 2007; Connor et al., 2010; Kolaitis et al., 2017). Parents of a child with severe CHD report high psychological distress including post-traumatic stress, depression, and anxiety (Gregory et al., 2018; Woolf-King et al., 2017). While we know that parents experience poorer mental health at the time of the diagnosis and months later (Woolf-King et al., 2017), there is a limited understanding of how each treatment decision (e.g., comfort-directed care vs. surgery) may differentially affect

Abbreviations: CHD, Congenital heart defect.

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parents' quality of life. Parents who have a surviving child with a chronic illness also face a greater risk for developing a chronic physical illness themselves (Brehaut et al., 2009; Brehaut et al., 2011); however, this risk is not well understood in the context of parents who have a child with severe CHD who survives past their first surgery.

Currently, we have limited knowledge about how parents' treatment decisions contribute to their quality of life after making a life-altering decision for their fetus or neonate with severe CHD. Moreover, most available studies on parental quality of life and health focus on those who chose surgery, therefore we know less about the quality of life of parents who chose comfort-directed care (Andonian et al., 2018; Gregory et al., 2018). Without this knowledge, the development of interventions to help support and maintain parent's well-being will likely remain difficult. The aim of this exploratory study was to address these gaps in the literature by exploring potential differences in parent-reported quality of life and health by treatment decision and survival outcome (i.e., whether or not the child survived after surgery).

Methods

This study used data from a clinical trial (NCT04437069) conducted at a children's hospital in the Intermountain West and focused on evaluating the effectiveness of a decision aid intervention. Eligible participants included parents of a fetus or neonate diagnosed with a complex CHD (e.g., hypoplastic left heart syndrome) that cardiologists acknowledged as severe where termination, comfort-directed care, or surgery were viable treatment options. Details on the protocol for parent trial are published elsewhere (Delaney et al., 2021). For the purposes of this study, only participants enrolled in the observational control group (from 9/2018 to 12/2020) were included in analyses. Control group participants received standard care and were not exposed to the main study intervention testing a decision aid. This study has approval through the University of Utah Institutional Review Board. Three months after participants made their treatment decision, they were asked to complete survey measures using REDCap or via paper survey. REDCap is a secure web application for building and managing online surveys and databases. Medical information was collected through electronic health records.

Study measures

Demographic questions included self-reported age, gender identity, race, ethnicity, education, and marital status. The fetus' CHD diagnosis was extracted from medical record data. Treatment decision was self-reported by participants in a survey (termination, comfort-directed care, or surgery).

The 3 month post-decision survey included the Impact of a Child with Congenital Anomalies on Parents (Mazer et al., 2008) that is a validated questionnaire that assesses parents' quality of life, with several domains including: contact with clinicians (Cronbach's alpha [α] = 0.91; 4 items, e.g., "I am satisfied about my contacts with doctors"), social network support (α = 0.91; 6 items, e.g., "people around me support me"), partner relationship (α = 0.94; 5 items, e.g., "I feel my partner sympathizes with me"), and state of mind (α = 0.86; 4 items, e.g., "I feel guilty"). Response options ranged on a Likert-scale from 1 (strongly disagree) to 4 (strongly agree). The fear and anxiety domain was excluded in these analyses because items were only relevant if the child survived. Higher scores indicate better contact with clinicians, support from their social network, partner relationship, and state of mind.

The short-form 12-item health survey (Care, 2010; Jenkinson et al., 1997) assessed participants' mental (α = 0.87; 6 items; e.g. "have you felt calm and peaceful?") and physical health (α = 0.69; 6 items, e.g., accomplished less due to physical health). Higher scores indicate better mental and physical health.

Statistical analysis

R Studio Version 1.4.1106 (RStudio Team, 2022) was used to conduct analyses. Given the small study sample and exploratory nature of this study, formal statistical comparisons between groups were not conducted. However, mean difference estimates with 95% confidence intervals (Kassambara, 2021) are reported to highlight potential directional differences between groups and whether confidence intervals include zero (i.e., signify no effect). Comparisons were made between parents who chose comfort-directed care and parents who chose surgery (both overall and according to whether the child did or did not survive). Bivariate correlations between key study outcomes are included in Appendix Table A1.

Results

Demographic characteristics, treatment decision, and correlations

Of 35 parents enrolled in the observational control arm, 11 did not complete the 3-months post-decision survey (31% attrition) and one was excluded due to a postnatal diagnosis as their experience would likely differ than prenatal diagnoses (Fig. 1). The study sample included 23 parents who were mostly female (78%), non-Hispanic White (87%), married (91%) and had some college education (70%; Table 1). There were 16 who chose surgery, 7 who chose comfort-directed care, and no parents who chose termination.

Table 2 presents mean scores by treatment decision and survival outcome (i.e., for parents who chose surgery, whether or not their child survived) along with mean difference and 95% confidence intervals (Kassambara, 2021) for each study outcome. Bivariate correlations were also computed between quality of life domains and the health survey (see Appendix Table A1). Greater social network support, better partner relationship, and better state of mind were associated with higher mental health scores.

Quality of life

Contact with clinicians

Parents who had a child that did not survive following surgery (M = 16.00) reported higher perceived levels of contact with their clinicians than parents who had a child who survived following surgery (M = 12.91; difference = 3.09; 95% confidence interval [CI] = 0.91 to 5.27) and those who chose comfort-directed care (M = 13.43; difference = -2.57 95% CI, -4.95 to -0.19). Of note, there was no variability in the scores for parents who chose surgery and their child did not survive—all parents selected the maximum response values. Confidence intervals indicated no differences between other comparison groups.

Social network

Parents who chose comfort-directed care (M = 18.57) reported lower social network support compared to parents who chose surgery and their child did not survive (M = 23.40; difference = -4.83 ; 95% CI, -7.37 to -2.29). Confidence intervals were consistent with no difference between other comparison groups.

Partner relationship

Overall, parents rated their relationship with their partner favorably. When comparing by treatment decision, mean scores were similar for each group and confidence intervals were consistent with no difference between groups.

State of mind

Parents who chose comfort-directed care (M = 7.71) reported poorer state of mind compared to parents who chose surgery (M = 11.27; difference = -3.55 ; 95% CI, -6.15 to -0.95), particularly compared to

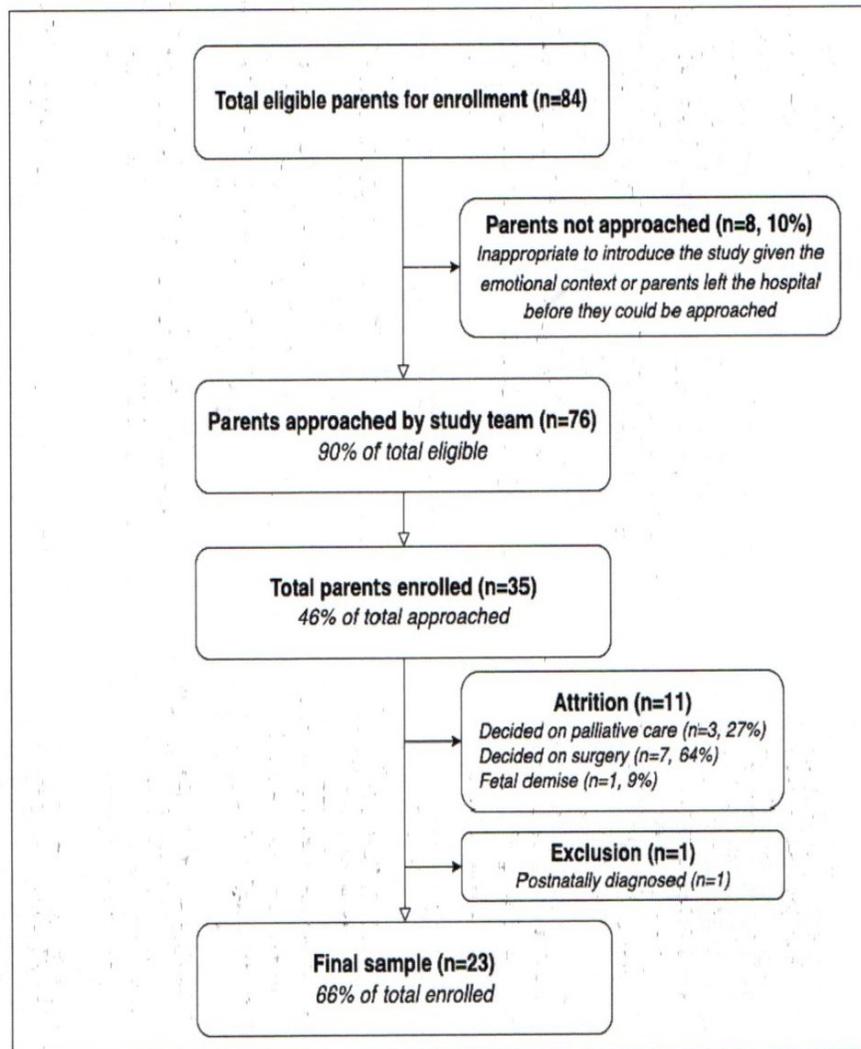


Fig. 1. Consort diagram for study recruitment, enrollment, and final analytic sample.

parents who chose surgery and their child survived ($M = 11.30$; difference = -3.59 ; 95% CI, -6.31 to -0.87). Confidence intervals were consistent with no difference between other comparison groups.

Mental and physical health

Confidence intervals indicated no differences in mental or physical health between groups (Table 2).

Table 1
Baseline characteristics of the study participants.

Characteristic	Study sample (N = 23)	Attrition sample (N = 11)
Age -- years		
Mean	27 ± 4	28 ± 4
Median	26	26
Interquartile range	26–29	26–32
Gender -- no. (%)		
Female	18 (78)	9 (82)
Male	5 (22)	2 (12)
Race/Ethnicity -- no. (%)		
Non-Hispanic White	20 (87)	10 (91)
Hispanic: White	2 (9)	1 (9)
Hispanic: Other race	1 (4)	–
Education -- no. (%)		
Some high school education or less	4 (17)	–
Some college or 2-year degree	8 (35)	7 (64)
4-year degree or higher	11 (48)	4 (36)
Marital Status -- no. (%)		
Currently married	21 (91)	9 (82)
Never married	2 (9)	1 (9)
Divorced	–	1 (9)
Fetus' CHD diagnosis -- no. (%)		
Hypoplastic left heart syndrome or hypoplastic left variant	12 (63)	5 (45)
Ebstein's anomaly/dysplastic tricuspid valve with severe regurgitation	3 (15)	2 (18)
Tetralogy of Fallot with absent pulmonary valve	1 (5)	1 (9)
Pulmonary atresia with intact ventricular septum	1 (5)	1 (9)
Heterotaxy, single ventricle	1 (5)	1 (9)
Complex single ventricle	1 (5)	1 (9)

Table 2
Outcome measures according to treatment decisions and survival outcome.

	Comfort-directed care (N = 7)	Surgery (N = 16)	Surgery, Survived (N = 11)	Did not survive (N = 5)	Comfort-directed care vs. Surgery				
<i>Mean difference estimate (95% CIs)</i>									
ICCAP Higher scores indicate better perceived status									
Contact with clinicians (Range: 4–16)	13.43 ± 2.57	13.88 ± 3.03	12.91 ± 3.24	16.00 ± 0.00	-0.45 (-3.10 to 2.21)	-2.57 (-4.95 to -0.19)	0.52 (-2.42 to 3.46)	3.09 (0.91 to 5.27)	3.09 (0.91 to 5.27)
Social network (Range: 6–24)	18.57 ± 2.70	21.38 ± 3.90	20.45 ± 4.41	23.40 ± 0.89	-2.80 (-5.79 to 0.18)	-4.83 (-7.37 to -2.29)	-1.88 (-5.44 to 1.67)	2.95 (-0.09 to 5.98)	2.95 (-0.09 to 5.98)
Partner relationship (Range: 5–20)	18.57 ± 2.44	18.50 ± 2.03	18.27 ± 2.20	19.00 ± 1.73	0.07 (-2.28 to 2.42)	-0.43 (-3.11 to 2.26)	0.30 (-2.18 to 2.78)	0.72 (-1.54 to 3.00)	0.72 (-1.54 to 3.00)
State of mind (Range: 4–16)	7.71 ± 2.36	11.27 ± 3.26	11.30 ± 2.87	11.20 ± 4.32	-3.55 (-6.15 to -0.95)	-3.49 (-8.76 to 1.79)	-3.59 (-6.31 to -0.87)	-0.10 (-5.36 to 5.16)	-0.10 (-5.36 to 5.16)
SF-12 Higher scores indicate better mental and physical health									
Mental health (Range: 0–100)	29.13 ± 13.76	40.37 ± 11.89	41.00 ± 10.86	38.97 ± 15.23	-11.23 (-24.56 to 2.10)	-9.84 (-29.53 to 9.86)	-11.87 (-25.45 to 1.71)	-2.03 (-20.58 to 16.51)	-2.03 (-20.58 to 16.51)
Physical health (Range: 0–100)	53.11 ± 5.29	50.21 ± 8.16	51.27 ± 8.58	47.89 ± 7.48	2.90 (-3.11 to 8.92)	5.23 (-4.04 to 14.5)	1.85 (-5.08 to 8.78)	-3.37 (-12.95 to 6.19)	-3.37 (-12.95 to 6.19)

Note. Plus-minus values are means ±SD; 95% Confidence Intervals which do not include zero are emphasized with bolded text; ICCAP = impact of a child with congenital anomalies on Parents, SF-12 = short-form 12-item health survey.

Discussion

Parents of a fetus or neonate diagnosed with complex CHD are at risk for experiencing significant strain on their mental health as a result of having to choose between termination, comfort-directed care, or surgery. This exploratory study begins to address the critical need to understand how to adequately support families who have made various treatment decisions and face different outcomes with their affected children to improve their quality of life. To do so, our findings illustrate some potential differences that are important to consider. Overall, parents who chose comfort-directed care reported less contact with clinicians, lacked social support and reported a poorer state of mind compared to parents who chose surgery. Notably, parents who chose surgery and their child did not survive reported the highest level of contact with clinicians. Future research needs to examine this in further detail and address if there is a need for enhanced support and counseling for parents who chose comfort-directed care and the potential for healthcare professionals to provide more equitable support across treatment decisions.

While there were differences in parents reported contact with clinicians, it is unclear what contributed to those differences. It is possible that parents who chose comfort-directed care naturally interact less within the healthcare system because they may have more contact with hospice and are not meeting with surgeons, which reduces the opportunity for contact with their clinicians. Parents have also reported that clinicians do not discuss comfort-directed care much as a treatment option (Delaney et al., 2020). Palliative care specialists are not regularly integrated into care and are underutilized, (Marcus et al., 2018) but could further enhance support to parents who chose comfort-directed care alongside cardiology specialists. Decision aids are an additional strategy that could be used to improve patient-clinician communication and shared decision making, which provide information about each treatment option, potential questions to ask clinicians, and exercises to help parents identify their values and goals related to care. Decision aids can be delivered outside of the clinic to provide additional time for families to learn about their options and facilitate shared decision making with their clinicians (Delaney et al., 2021).

Parents who chose surgery and whose child did not survive reported maximum levels of contact with their clinicians suggesting that the parents felt well-supported by their clinicians. There may be areas of improvement for parents who chose comfort-directed care and for parents who chose surgery whose child survived. Parents whose child undergoes surgery and who have to manage subsequent home care report challenges from extreme caregiving demands and having to learn disease management (Rempel et al., 2013; Sjostrom-Strand & Terp, 2019; Wei et al., 2016). This could potentially underlie the observed differences within the surgery group. Implementing video- or telephone-based homecare support programs (McCrossan et al., 2012) may be one cost-effective method for improving support for parents who have to learn home care and disease management following surgery for their child.

Parents who chose comfort-directed care reported less social network support compared to parents who chose surgery and their child did not survive. These findings are particularly striking given that the survival outcome of the child was the same in both groups, yet parents who chose comfort-directed care perceived less support than parents who chose surgery. Attitudes surrounding perceived active (i.e., surgery) vs. passive (i.e. comfort-directed care) treatment may be driving these differences (Tomlinson et al., 2011; Zikmund-Fisher et al., 2006). Increased implementation and availability of hospital-based bereavement services such as making phone calls, providing resource materials, and connecting families to appropriate support groups could be one method for supporting parents after the death of their child (Donovan et al., 2015).

There are mixed findings within the literature on the family functioning and quality of partner relationships among families of a child

diagnosed with CHD. Some studies have found negative impacts, while others have found greater family cohesion and support (Riikonen et al., 2019; Sood et al., 2021). Overall, parents within the study reported that they mostly had high-quality relationships with their partner. Exploratory findings also indicated no differences by treatment decision for partner relationship, which can offer important and reassuring information for parents as they consider these critical decisions. These preliminary findings can help with counseling parents to know that regardless of what treatment decision parents make they can still maintain the quality of the relationship with their partner.

Parents who chose comfort-directed care also reported a poorer state of mind and had mental health scores that were between 10 and 11 points lower than parents who chose surgery and their child survived. Grief and distress from losing their child could be contributing to differences between these groups. Although the confidence intervals did not meet criteria to confirm statistically significant differences, scores in this study sample ($M = 29-41$), were low relative to the general young adult U.S. population (e.g., $M = 52.9$) (Care, 2010; Jenkinson et al., 1997) Considering the lower state of mind scores among these parents and associations with mental health scores from our findings, there still appears to be clinically meaningful differences regarding their mental health. These findings suggest the need for additional research and development of interventions to support parents who choose comfort-directed care.

Although a previous study found that caregivers of a child with a chronic disease were at risk for poorer physical health themselves (Brehaut et al., 2009; Brehaut et al., 2011), parents' physical health scores did not differ and were mostly similar to the average U.S. population. Given that data were collected within a 3-month time frame and among young adults, these findings offer limited insight into the impact on physical health as such an effect may not occur that quickly in this particular age group. Future studies with a longer follow-up time point, may address the impact on physical health.

Limitations

Limitations of the present study were that we had a small sample size due to the rarity of the condition, having a single-site for the study, and challenges regarding longitudinal follow-up for parents navigating and coping with complex decisions and outcomes. While this limited our power to conduct a broader range of statistical analyses, we believe these exploratory findings are important to share as they begin to uncover parents' experiences during a critical treatment decision that have not been previously explored. Conducting a larger-scale longitudinal study, perhaps through the development and maintenance of a registry of parents who received a complex CHD diagnosis for their fetus or neonate, will be necessary to achieve robust power for statistical analyses. Lastly, the contact with clinicians subscale did not specify the type of clinician or the timing related to parents contact with clinicians. Future research should consider collecting more specific data to better understand communication deficits and at which time point parents need additional support (e.g., pre or post-decision).

Conclusions

The aim of this exploratory study was to add to the limited literature on how parent's quality of life may differ based on the life-altering treatment decision for their fetus or neonate diagnosed with complex CHD. Some aspects of parents' quality of life differed based on their treatment decision as well as the outcome of this decision. Parents who chose comfort-directed care are vulnerable to experiencing poorer social network support and some quality of life outcomes. Developing interventions and resources, such as decision support tools and bereavement resources, are important next steps to improve coping for parents who choose comfort-directed care.

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Ethical standards

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human and with the Helsinki Declaration of 1975, as revised in 2008, and has been approved by the institutional committees from the University of Utah Institutional Review Board.

Contributors' statement page

Dr. Delaney substantially contributed to the conceptualization, methodology, investigation, and writing (original draft and revisions).

Dr. Thorpe substantially contributed to the formal analysis, data curation, writing (review and editing), and visualization.

Dr. Pinto and Dr. Ozanne substantially contributed to the conceptualization, methodology, investigation, and writing (review and editing).

Ms. Hansen, Ms. Lambert, Ms. Pershing, and Ms. Tanner substantially contributed to the investigation, project administration, and writing (review and editing).

Dr. Fagerlin substantially contributed to the conceptualization, methodology, investigation, writing (review and editing), and funding acquisition.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Declaration of Competing Interest

The authors have indicated they have no financial relationships and no potential conflicts of interest relevant to this article to disclose.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.pedn.2023.02.001>.

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