Cardiology in the Young

cambridge.org/cty

Parents' decision-making for their foetus or neonate with a severe congenital heart defect

Rebecca K. Delaney[1], Nelangi M. Pinto[2], Elissa M. Ozanne[1], Heather Brown[3], Louisa A. Stark[3,4], Melissa H. Watt[1], Michelle Karasawa[1], Angira Patel[5], Mary T. Donofrio[6], Michelle M. Steltzer[7], Stephen G. Miller[8], Susan L. Zickmund[9,10] and Angela Fagerlin[1,9]

[1]Department of Population Health Sciences, University of Utah, Salt Lake City, UT, USA; [2]Division of Pediatric Cardiology, Department of Pediatrics, University of Utah, Salt Lake City, UT, USA; [3]Center for Clinical and Translational Science, University of Utah, Salt Lake City, UT, USA; [4]Department of Human Genetics, University of Utah, Salt Lake City, UT, USA; [5]Division of Cardiology, Ann & Robert H. Lurie Children's Hospital of Chicago, Northwestern University Feinberg School of Medicine, Chicago, IL, USA; [6]Division of Cardiology, George Washington University, Children's National Hospital, Washington, DC, USA; [7]Ann & Robert H. Lurie Children's Hospital of Chicago, Chicago, IL, USA; [8]Department of Pediatrics, Division of Pediatric Cardiology, Duke University Medical Center, Durham, NC, USA; [9]VA HSR&D Informatics, Decision-Enhancement and Analytic Sciences Center, VA Salt Lake City Health Care System, Salt Lake City, UT, USA and [10]Division of Epidemiology, Department of Internal Medicine, University of Utah, Salt Lake City, UT, USA

Original Article

Cite this article: Delaney RK, Pinto NM, Ozanne EM, Brown H, Stark LA, Watt MH, Karasawa M, Patel A, Donofrio MT, Steltzer MM, Miller SG, Zickmund SL, and Fagerlin A (2022) Parents' decision-making for their foetus or neonate with a severe congenital heart defect. *Cardiology in the Young* **32**: 896–903.
doi: 10.1017/S1047951121003218

Received: 7 April 2021
Accepted: 13 July 2021
First published online: 19 August 2021

Keywords:
Congenital heart disease; decision-making; parent; qualitative research; decision support; counselling

Author for correspondence: Dr A. Fagerlin, Department of Population Health Sciences, University of Utah, 295 Chipeta Way, Salt Lake City, UT 84108, USA.
Tel: +1 (801) 587-0049.
E-mail: angie.fagerlin@hsc.utah.edu

Abstract

Background: Parents who receive a diagnosis of a severe, life-threatening CHD for their foetus or neonate face a complex and stressful decision between termination, palliative care, or surgery. Understanding how parents make this initial treatment decision is critical for developing interventions to improve counselling for these families. *Methods:* We conducted focus groups in four academic medical centres across the United States of America with a purposive sample of parents who chose termination, palliative care, or surgery for their foetus or neonate diagnosed with severe CHD. *Results:* Ten focus groups were conducted with 56 parents (M_{age} = 34 years; 80% female; 89% White). Results were constructed around three domains: decision-making approaches; values and beliefs; and decision-making challenges. Parents discussed varying approaches to making the decision, ranging from relying on their "gut feeling" to desiring statistics and probabilities. Religious and spiritual beliefs often guided the decision to not terminate the pregnancy. Quality of life was an important consideration, including how each option would impact the child (e.g., pain or discomfort, cognitive and physical abilities) and their family (e.g., care for other children, marriage, and career). Parents reported inconsistent communication of options by clinicians and challenges related to time constraints for making a decision and difficulty in processing information when distressed. *Conclusion:* This study offers important insights that can be used to design interventions to improve decision support and family-centred care in clinical practice.

CHDs are the most common and lethal birth defect.[1–3] A subset of CHDs (25%) are considered severe and life-threatening without intervention.[3] Medical advances in cardiology have made surgical and catheter-based intervention possible for most complex CHDs. However, even with intervention, individuals diagnosed with some types of severe CHDs face a serious risk of mortality, morbidity, and lifelong complications with an uncertain quality of life.[4–7] For severe CHDs, (e.g., hypoplastic left heart syndrome), termination, palliative care, and surgery may be considered as treatment options for an affected foetus or neonate.[8–11]

Parents of a foetus or neonate diagnosed with severe CHDs are then confronted with an unexpected and challenging treatment decision about whether or not to pursue surgical intervention. After the initial diagnosis and during the decision-making process, up to 80% of parents have severe psychological distress[12] and experience high rates of post-traumatic stress symptoms, depression, anxiety, and grief.[12–15] There is a significant need to determine how to best support and counsel parents during this difficult decision-making process. Although some qualitative studies have begun to explore parents' perspectives on making treatment decisions, these previous studies have had small samples of parents from a single medical centre who chose surgery.[16–19] Therefore, there is a need to build a more robust understanding of parent perspectives by including those who chose termination or palliative care, as well as, identify information and factors that support parents making the treatment decision.

The purpose of this multisite qualitative study was to examine how parents made treatment decisions for their foetus or neonate with a severe CHD, including what factors played a role in

© The Author(s), 2021. Published by Cambridge University Press.

CAMBRIDGE UNIVERSITY PRESS

the decision-making process and identify challenges they faced during the decision-making process.

Methods

A qualitative study design was used to provide in-depth insight and robust understanding into an emotionally-laden and complex experience.[20] Focus groups were chosen based on our belief that being with others who had reached a similar treatment decision would offer a supportive and inclusive environment and elicit thoughtful reflection on this sensitive topic.[21,22] Institutional review boards at the University of Utah, Primary Children's Hospital, Ann & Robert H. Lurie Children's Hospital of Chicago, Duke Children's Hospital, and Children's National Hospital approved this study and data transfer agreements were completed. Prior research suggests that to reach at least 90% thematic saturation[23,24], five focus groups are typically necessary thus, we planned to recruit for ten focus groups to provide us with enough data to reach our study objective.

A purposive sample of parents who had a foetus or neonate diagnosed within the last 6 months to 5 years with a severe CHD, who chose between one or more of three viable treatment options (i.e., termination, palliative care, or surgery), were recruited from four academic medical centres in metropolitan cities across the United States of America (see Supplementary Table S1 for list of CHDs). Participants (biological and non-biological parents) were identified through cardiac ICUs, high-risk clinics, and/or foetal cardiology databases at each study site. Eligible individuals were sent a letter inviting them to participate in a focus group; upon agreement or initial interest, they were provided additional information by telephone or email. Because we expected thematic deviation and wanted parents to feel comfortable, focus groups were divided according to parents' treatment decisions: termination or palliative care, or surgical intervention. Prior to the focus group, parents were sent a letter with a brief description of the project goals and questions that would be discussed.

Data collection

We conducted 10 focus group discussions: 4 in Salt Lake City, UT and 2 each in Chicago, IL, Durham, NC, and Washington, DC (November, 2017-April, 2019). Equal numbers of focus groups for the two treatment categories were held in each city. One experienced moderator, trained on qualitative interviewing and the sensitive nature of the topic, conducted all focus group discussions. Most (n = 9) were conducted in person and in English; two parents participated via video conference call and one parent was supported by a Spanish interpreter. One focus group was conducted exclusively by video conferencing due to severe weather. Before each focus group discussion, the study team informed parents about the confidentiality of their statements, addressed questions or concerns, and obtained written informed consent. Parents completed a self-administered questionnaire on demographic information (e.g., age, gender, education, household income). A study team member attended most focus group discussions (n = 9) and took notes. Focus group discussions were approximately 2 hours long, audio-recorded, and transcribed verbatim by a third-party transcription service. Parents received a $50 gift card as remuneration.

The team developed a semi-structured interview guide[25,26] with open-ended questions and probes to facilitate discussion. The guide included four questions; two of which are described here: "What information did you learn after your baby was diagnosed?" and "What mattered most to you when making decisions about your baby's medical care?" Questions were asked in a general manner without referencing specific clinical situations. After each focus group discussion, the study team members debriefed, reviewed notes, discussed emerging findings, and assessed the extent to which thematic saturation was reached (i.e., the point at which little or no new information occurred).[27]

Data analysis

Data analysis was conducted using Crabtree and Miller's editing approach,[28] which involves a grounded interpretation of text and open coding to identify emergent themes. This is an inductive, or "bottom-up" approach whereby the text is reviewed and coded into meaningful categories, in order to develop overarching conclusions that are "grounded in" the data. Qualitative analysis software (Altas.ti) was used to organise and code the textual data. Analysts first conducted a deep reading of the text to develop insights on the research questions and inductively identify themes.[28] The emergent themes, together with structural codes from the interview guide, were used to create a preliminary codebook. Three transcripts were independently reviewed by two reviewers and discussed to reach a consensus on the application of codes. The codebook was updated iteratively and the team regularly discussed coding to establish consensus. A final report with findings was compiled, reviewed, and corroborated by numerous stakeholders (e.g., parents, clinicians, researchers) to establish face validity and develop additional insights on the data.[29] We largely received support and consensus regarding our findings from stakeholders. As a result, no significant changes were made to the report.

Results

Fifty-six parents participated across ten focus group discussions. Parents had an average age of 34 years (20–52 years; $SD = 5.1$) and a median education level equivalent to a 4-year college degree. Most were women (80.3%), and most identified themselves as White (89%; Table 1). Significant differences in parent characteristics between groups were only found for household income ($X^2 = 7.04$, $p = 0.007$).

Themes resulting from focus group discussions

The analysis of the focus group transcripts resulted in 11 dominant themes, which are organised within 1 of the 3 overarching domains: decision-making approaches; values and beliefs; and decision-making challenges. Representative, exemplar quotes (Q) are highlighted for each theme within the results section and Tables 2, 3, and 4.

Decision-making approaches

Four themes emerged signifying diverse approaches to making the initial treatment decision: right decision, rational-analytic, intuitive-experiential, and dependent-consultative (Table 2). Parents acknowledged the complex and multifaceted nature of the decision, with most acknowledging that there was no clear right or wrong decision (Q1), and that each parent may differ in their decision preferences (Q2). One parent highlighted that there was "an illusion of choice," (Q3) suggesting that it does not feel like a real choice is being offered when there is no clear "right" option.

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

Table 1. Parent characteristics overall and by focus group category across 10 focus groups

	Overall (n = 56; 10 FGDs)	Termination/ Palliative care (n = 17; 5 FGDs)	Surgical intervention (n = 39; 5 FGDs)
Age, M(SD)	33.9 (5.1)	33.8 (3.6)	34 (5.6)
Gender, %			
Female	80	88	77
Race, %			
White	89	100	85
African American	2	0	3
American Indian or Alaskan Native	2	0	3
Other	7	0	10
Hispanic or Latino/a, %	14	6	18
Currently employed, %	63	82	56
Education, %			
≤ High school degree	10	0	15
Some college	25	24	26
College graduate	29	29	28
Some graduate/ graduate degree	36	47	31
Annual household income, %*			
≤ $49,999	17	13	18
$50,000–74,999	20	0	29
≥ $75,000	63	88	53

FGD = focus group discussion. Independent samples t-tests (for continuous variables) and chi-square tests (for categorical variables) were computed to test for significant differences by focus group category.
*Significant differences in income were found ($X^2 = 7.04$, $p = 0.007$) but no significant differences were found among other parent characteristics.

Table 2. Decision-making approaches: summary of themes

Themes	Representative quotes
Right decision	Q1: "There isn't a right decision. It's the best decision for you and your family." (T/PC) Q2: "There are also two people making this decision. There's two parents and parents can have different decisions, right? There has to be some discussion between parents because a mom makes one decision and a dad wants a different decision, that's kind of an impasse, right? You have to have some way to get through that." (S) Q3: "Like going through this is an illusion of choice. . . . . Like you think that you have a choice in the matter, but it's really just an illusion, like this really is the right thing or the best thing whatever it is." (T/PC)
Rational-analytic approach	Q4: "I'm a numbers person, so I needed those statistics of what's the survivability of this . . . " (S)
	Q5: "If we chose to have the baby, what the process would be, statistically speaking, what would be the severity of [the heart defect]? And then, just kind of the statistics of what the treatment options are and then survival and even like long-term." (T/PC)
Intuitive-experiential approach	Q6: "You just have to go with your gut and accept that that's the right thing to do." (T/PC)
	Q7: "I just feel that at the end of the day, I strongly believe that we have a strong instinct that listens to the child. And if you listen to that, you're going to make the best decisions for [the child]. That was so personal." (S)
Dependent-consultative approach	Q8: "I think you should hear, you know, even a story like from each parent for each scenario, you know? So, like from a mom or a couple why they chose to do the surgeries, or why they chose to do homecare, or you know, why they chose to do an abortion." (S)
	Q9: (Responding to other parent discussing getting second opinion:) "[Doctors name] called me back and talked to me and said the same things, which I think is super helpful because you want to know from more than one person. I think him being willing to do that or anyone being willing to say, yes, I looked at this. I agree." (T/PC)

(S) = quote from parent in surgical intervention focus group discussion; (T/PC) = quote from parent in termination/palliative care focus group discussion.

Multiple decisional approaches emerged in the focus group discussions as identified in decision-making literature.[30–33] Some parents described a rational-analytic approach where statistics, probabilities, and numeric values were important (Q4, Q5). Others discussed an intuitive-experiential approach, relying upon gut feelings and instincts (Q6, Q7) to guide their decision. Some also described a dependent-consultative approach, where they sought advice and direction from others, like their providers or other parents who previously faced a similar decision (Q8, Q9). These approaches were not mutually exclusive, as some parents discussed using a combination of these.

Values and beliefs

Three dominant themes were found within the values and beliefs domain that parents noted as important factors for their treatment decision: religious and spiritual beliefs, quality of life for the child, and quality of life for the family (Table 3). Religious and spiritual beliefs were one of the most common values mentioned by parents in both focus group categories. For some, religious and spiritual beliefs guided their decision not to terminate (Q10). Although parents made different treatment decisions, their religious and spiritual beliefs offered support and perspective for the development of their values related to their choices (Q11).

Quality of life was a significant consideration described across all focus group discussions. First, parents considered what the quality of life, both in the short-term and long-term, would be like for their child with a severe CHD depending on each option. Common indicators of quality of life mentioned were pain or discomfort; suffering; cognitive and physical abilities; ability to attend school; independence; and adulthood (Q12–Q14).

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

Table 3. Values and beliefs: summary of themes

Themes	Representative quotes
Religious and spiritual beliefs	Q10: "I am very religious as well and that was the deciding factor to me not aborting [name]." (S)
	Q11: "And I just felt like there's a spiritual aspect that came into place and I just felt like I think my baby's mission is to come and get a breath of air and pass on to the next life." (T/PC)
Quality of life	
For the child	Q12: "Will he be able to, you know, walk and go to school and do things you think like an average child? So, I think that was a big deciding factor of how hard and fast are we going to attack this." (S)
	Q13: "The amount of discomfort (to the child) was highly important. And the idea of doing surgeries that would be open heart surgeries three times by the age of two was really hard for us to picture putting a child through." (T/PC)
	Q14: "We're doing this so [child's name] can breathe fresh air and live on our little farm and see the animals and eat food out of our garden and things that I find are quality of life and that's why – that's why this (deciding to continue with surgical treatments) is important." (S)
For the family	Q15: "I didn't want my other kids to resent the time that, you know, I'm sorry I missed this, I missed your basketball game, I missed your baseball game, I missed dance. We can't do that . . . we can't afford lessons or extra things because every spare moment, every spare dollar, every spare thing goes to (the sick child)." (T/PC)
	Q16: (responding to discussion of what parents from support group ask about how having a child with CHD will impact their other kids): "For me, it was like impact on the family. For me, it was a first time child and so it impacts your marriage. It impacts the grandparents and aunts and uncles and everybody . . . A lot of these are sort of interconnected and having a heart healthy child is going to impact the family. It's going to impact you financially. It's going to impact everything." (S)

(S) = quote from parent in surgical intervention focus group discussion; (T/PC) = quote from parent in termination/palliative care focus group discussion.

Second, parents across focus group discussions spoke about how they weighed the impact of their decision on the quality of life of their family, such as whether caring for a child with a CHD would influence care for their other children, and how each option might impact marriage, career, or finances (Q15, Q16).

Decision-making challenges

Four dominant themes within the decision-making challenges domain were found with overarching challenges discussed related to clinician communication: satisfaction with provider, incongruent discussion of treatment options and information processing, affective/emotional, and cognitive (Table 4). Although some parents expressed satisfaction with communication from clinicians (Q17), other parents noted shortcomings in how clinicians communicated diagnoses and treatment options. Across both focus group categories, parents felt clinicians did not devote equal

Table 4. Decision-making challenges: summary of themes

Themes	Quotes
Clinician communication	
Satisfaction with provider	Q17: "In my personal experience, one of the things that I most appreciated was that everyone was very clear about the range of options, whether to continue with the pregnancy or terminate." (T/PC)
Incongruent discussion of treatment options	Q18: "I think palliative (care) is really not brought up almost at all before you give birth. It was not brought up to me. They always talked about surgery. Surgery, surgery, surgery" (S)
	Q19: "They spent 45 minutes talking about the palliative surgeries for HLHS (hypoplastic left heart syndrome), and then they spend 3 minutes talking about, "Or you could choose comfort care . . . " (T/PC)
	Q20: "If your first conversation is with a pediatric cardiologist, you are probably not going to get the termination option. You're going to get potentially a palliative care, but you're going to get a surgical or medical route. If your first experience is with an OB, you're going to get termination as one of the first options." (S)
Information processing	
Affective/emotional	Q21: "It's such an emotion[al] time. It's so hard to just read the information and try to process it on your own." (T/PC)
	Q22: "It was like the most isolating experience of my life. And I felt like I had two weeks or less to make such an important decision." (T/PC)
Cognitive	Q23: "And they kept drawing the heart and I had no clue about what that meant or what it was that they were saying. It was like going to school and explain to you but you don't get it and I was crying, and I didn't understand what was going on." (S)
	Q24: "They did a great job at explaining things and drawing pictures and I heard what they said, but literally from the moment that I left their office, if you had asked me what they said when I got back in the car in the parking lot, I wouldn't have had a clue. I mean I was just jotting down little things, Tetralogy of Fallot, and then once I got home, not remembering anything that they said other than, "No one is going to do surgery," or "there is the option of doing surgery," or whatever. It was just Google." (T/PC)

(S) = quote from parent in surgical intervention focus group discussion; (T/PC) = quote from parent in termination/palliative care focus group discussion.

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

attention to the various treatment options available. Parents reported that clinicians spent the most time discussing surgical options, with less time devoted to termination and little or no discussion of palliative care as an option (Q18, Q19). One parent suggested that clinicians' specialty contributes to inconsistent communication of options (Q20).

Across both focus group categories, parents discussed how difficult it was for them to process information regarding diagnosis and treatment options. In particular, parents noted challenges in understanding and processing pertinent information related to the heightened emotions and distress associated with the diagnosis, and the short timeline to make a decision (e.g., pre- versus postnatal diagnosis, state laws regarding termination; Q21, Q22). Parents also expressed that the way clinicians communicated the diagnosis and treatment options were challenging to understand (Q23). Even if they comprehended information during their medical appointment, they often did not remember it by the time they got home (Q24).

Discussion

Our study sought to understand how parents made treatment decisions for their foetus or neonate diagnosed with a complex, life-threatening CHD. By including perspectives from parents from across the United States of America who made different decisions, our study adds to the literature by providing a comprehensive understanding of domains to leverage in the development of interventions to support parents in making a challenging choice between termination, palliative care, or surgery. Our findings highlight variability in how parents approach and process the decision, important values and beliefs that influence their decision, and challenges in clinician communication and processing information about the diagnosis and treatment options. These important insights can be used to develop interventions to support families during the treatment decision-making process for their foetus or neonate diagnosed with CHD. In addition, the domains and themes we found can be used to inform how to engage surrogate decision makers in other cardiac or end-of-life decisions in paediatric contexts.

Similar to the literature on decision-making approaches[30–33] and qualitative research on decision making after an antenatal diagnosis of CHD,[17,19] we found that parents reported varying reliance on statistics, intuition, and advice from clinicians or families with shared experiences when making their decision. Providing comprehensive information that can encompass each decision style or tailoring interventions (e.g., decision support tools) to parents' preferred approach(es) are important next steps.[19,34] For example, clinicians could ask parents what information would be most helpful to them when making this decision and provide information (e.g., statistics) congruent with their preferences, thus engaging in conversations to promote shared decision-making.[35,36]

Our study found a variety of beliefs and values that influenced treatment decisions. While our study and others[18] have found that religious and spiritual beliefs can influence parents' decisions, some clinicians may not feel comfortable addressing these with families.[37] Clinician training and education in approaches to discussing religious and spiritual beliefs could address this barrier. Engaging other professionals such as social workers and hospital chaplains already trained in facilitating these discussions may help families to make decisions that align with their belief systems.

We found that quality of life for their family and their child with a CHD was a significant consideration for parents when making treatment decisions, especially considering how surgery and subsequent caregiving of a child with a CHD can change family lifestyles and dynamics.[38,39] Providing information about how a family's daily life would be similar or different for each treatment option could guide parents in their decision. Clinicians and other parents who have gone through this treatment decision before, for example, could help provide this information. In addition, values clarification methods could be used to address the values and beliefs domain. Values clarification methods are increasingly being developed to help parents review information and identify what matters most to them to ultimately inform their preferences towards an option.[40,41] For example, items could be included about what quality of life (e.g., physical and mental health outcomes, need for lifelong medical care) would be like depending on the treatment decision and have parents reflect on what experience feels better to them and their family. Parents could then use the information generated from a values clarification exercise as a foundation for talking to family, friends, and clinicians about how their values support their treatment decision.

This study significantly adds to the limited qualitative research regarding palliative care as a treatment option for severe CHDs. While one qualitative study conducted in Canada with parents who chose surgery for their child with hypoplastic left heart syndrome reported that palliative care was discussed[18], parents from our focus group discussion reported that surgery was discussed and emphasised over palliative care. Differences in options discussed may arise due to lack of knowledge or discomfort discussing termination or palliative care,[42] institutional ethical standards, and differing opinions on the success of a surgical intervention.[43] The predominant focus on surgery, however, could influence parents' perceptions that surgery is the only option. Despite some controversy amongst medical professionals in offering palliative care for severe CHDs,[43,44] we believe that presenting balanced, unbiased information for each option is critical for ensuring parents have adequate information to make their decision. Decision aids are one method that could be used to address this barrier to clinician communication of treatment options, within the decision-making challenges domain. Decision tools have been shown to improve patient knowledge and preparation for treatment choices in other serious illnesses[45] and could be a method used to better support a standardised discussion of options and reduce bias. Within the decision aid, information about treatment options including risks and benefits of each should be included. A list of questions that parents could ask their clinician about each treatment option could also be provided so that parents are better prepared for having conversations with their clinicians, ultimately improving communication between families and clinicians. Palliative care specialists can also help families clarify the quality of life goals, provide emotional support, and navigate potential conflicts between families and clinical care teams throughout multiple medical decisions.[46] While these specialists often exist within larger healthcare systems, they can be underutilised during the foetal and newborn stage[47,48], thus it would be advantageous to improve strategies for integrating them into maternal foetal medicine. Educating and training clinicians within maternal foetal medicine and cardiology on how to communicate about palliative care will also be important for improving clinical practice and care for these families.

Parents reported significant challenges in processing information about the diagnosis and treatment options. These difficulties were further compounded by the heightened emotional distress when receiving the diagnosis and limited time frame to make a

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

decision. Parents were often confused, stating that they did not remember what clinicians said during the visit. This is common and may be why many parents report using the Internet for information,[17] which may not provide scientifically-reliable or relevant information specific to their foetus or neonate's diagnosis. Making medical decisions under emotional circumstances can also impact risk perceptions and evaluation of decisional consequences.[49] Parents from our focus group discussions, as well as other studies,[17] have requested and strongly support the development of online resources with high-quality, digestible information on how a normal versus affected heart functions and other information about the diagnosis. Having information located on a website would also facilitate families' ability to review and obtain evidence-based information as needed outside of the clinic.

Limitations of the study include the potential for recall bias since we recruited parents who had a foetus or neonate diagnosed with a severe CHD within the last 6 months to 5 years. Our inclusion criteria were intentionally broad to help with recruitment efforts, as severe CHDs are relatively rare and we wanted to be sensitive to parents who were more recently coping with their foetus or neonate's diagnosis or death. Future work could consider capturing real-time information about how parents make these decisions by using short, semi-structured interviews or surveys to help minimise parent burden and distress. Second, despite efforts to recruit similar numbers for both focus group categories, fewer parents participated in the termination/palliative care focus group discussions, which could have influenced the representativeness of our sample. This may relate to fewer parents overall who choose or are offered these options,[18,44] perceived stigma surrounding this choice,[50] and limitations of our recruitment strategies. Nevertheless, this is one of the few studies to include the insights of parents with these experiences. Thus, it represents an important addition to this literature. It is critical for future research to be conducted with families, particularly those who chose termination or palliative care who are underrepresented in the literature. Third, focus groups were not stratified based on pre- or postnatal diagnosis, which could be important for future work to consider as the timing of the diagnosis can affect parental stress and anxiety,[51] particularly for those choosing between palliative care and surgery. Furthermore, it will be important for future research to conduct studies separately for families who choose termination and palliative care as they have unique experiences that may not be fully captured when focus group discussions are conducted together. Fourth, focus group discussions were chosen to promote interactive discussion, provide greater breath, and context to understanding how parents made treatment decisions. However, they may offer less depth and detail relative to semi-structured interviews and have the potential for social desirability bias, particularly related to sensitive topics.[52] As noted previously, future work could conduct semi-structured interviews and surveys to obtain greater detail of how parents make decisions in this challenging clinical context. This research would be particularly important to obtain a more detailed understanding of how overarching domains and themes we found interact and impact treatment decisions. Lastly, although we conducted a multisite qualitative study across the United States of America and offered different methods (e.g., Spanish interpreter, video conference call) to minimise participation barriers, we had a less diverse study sample than we had intended to recruit. Thus, our sample was not representative of the United States of America population demographics which potentially limits the generalisability of our study findings. Additionally, while most demographic characteristics were similar between focus groups, parents in the surgery group reported lower income relative to the termination/palliative care group. Differences may arise related to the financial burdens of long-term care (e.g., reduced work hours to provide home care) and medical costs for surgery.[38]

Conclusion

The aim of this qualitative study was to gain an in-depth understanding of how parents across the United States of America made their initial decision about treatment for a foetus or newborn diagnosed with severe CHD, considering the options of termination, palliative care, and surgical intervention. Parents varied in their decision approaches and values and how they weighed their family's preferences and goals in their decision-making process. Parents noted significant challenges in processing the information they needed to make the decision. Improving the communication process may help reduce cognitive and emotional burdens. Future directions should include developing interventions and improving counselling that present parents with standardised information about available options, assist in clarifying their values and preferences, and prepare parents for counselling during appointments with their clinicians.

Acknowledgements. We are very grateful to our parent co-investigators who helped develop our focus group guide and who provided their perspective on the data analysis. We thank members of the Qualitative Research Core at the University of Utah, Rachel Andes, Grace Perry, Patrick Galyean, and Alex Bellenger, for their effort in coding and generating a qualitative report for the study. Finally, we thank Mingfen Xu (Duke University) and Kathleen Van't Hof (Northwestern University) for their help in preparing for the focus groups and taking notes during the focus groups. We also thank Mandy Pershing for her assistance in editing and conducting a literature search.

Supplementary material. To view supplementary material for this article, please visit https://doi.org/10.1017/S1047951121003218

Financial support. This research was supported by the American Heart Association's Children's Strategically Focused Research Network grant (17SFRN33660465) to Dr Fagerlin. Dr Delaney's effort was additionally supported by the National Institutes of Health under Ruth L. Kirschstein National Research Service Award T32HL007576 from the National Heart, Lung, and Blood Institute.

Conflicts of interest. None.

Ethical standards. The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008, and has been approved by the institutional committees at the University of Utah, Primary Children's Hospital, Ann & Robert H. Lurie Children's Hospital of Chicago, Duke Children's Hospital, and Children's National Hospital.

References

1. Hoffman JIE, Kaplan S. The incidence of congenital heart disease. J Am Coll Cardiol 2002; 39: 1890–1900.
2. Reller MD, Strickland MJ, Riehle-Colarusso T, Mahle WT, Correa A. Prevalence of congenital heart defects in metropolitan Atlanta, 1998-2005. J Pediatr 2008; 153: 807–813.

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

3. Oster ME, Lee KA, Honein MA, Riehle-Colarusso T, Shin M, Correa A. Temporal trends in survival among infants with critical congenital heart defects. Pediatrics 2013; 131: e1502–e1508.
4. Best KE, Rankin J. Long-Term survival of individuals born with congenital heart disease: a systematic review and meta-analysis. J Am Heart Assoc 2016; 5: 1208.
5. Kamphuis M, Ottenkamp J, Vliegen HW, et al. Health related quality of life and health status in adult survivors with previously operated complex congenital heart disease. Heart 2002; 87: 356–362.
6. Mellion K, Uzark K, Cassedy A, et al. Health-related quality of life outcomes in children and adolescents with congenital heart disease. J Pediatr 2014; 164: 781–788.e781.
7. Schaefer C, von Rhein M, Knirsch W, et al. Neurodevelopmental outcome, psychological adjustment, and quality of life in adolescents with congenital heart disease. Dev Med Child Neurol 2013; 55: 1143–1149.
8. Barron DJ, Kilby MD, Davies B, Wright JGC, Jones TJ, Brawn WJ. Hypoplastic left heart syndrome. Lancet 2009; 374: 551–564.
9. Bertaud S, Lloyd DFA, Laddie J, Razavi R. The importance of early involvement of paediatric palliative care for patients with severe congenital heart disease. Arch Dis Child 2016; 101: 984–987.
10. Chenni N, Lacroze V, Pouet C, et al. Fetal heart disease and interruption of pregnancy: factors influencing the parental decision-making process. Prenat Diagn 2012; 32: 168–172.
11. Zeigler VL. Pediatric ethics, issues, & commentary. Ethical principles and parental choice: treatment options for neonates with hypoplastic left heart syndrome. Pediatr Nurs 2003; 29: 65–69.
12. Woolf-King SE, Anger A, Arnold EA, Weiss SJ, Teitel D. Mental health among parents of children with critical congenital heart defects: a systematic review. J Am Heart Assoc 2017; 6: e004862.
13. Uzark K, Jones K. Parenting stress and children with heart disease. J Pediatr Health Care 2003; 17: 163–168.
14. Kolaitis GA, Meentken MG, Utens EMWJ. Mental health problems in parents of children with congenital heart disease. Front Pediatr 2017; 5: 102.
15. Lawoko S, Soares JJF. Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children. J Psychosom Res 2002; 52: 193–208.
16. McMahon E, Chang Y-S. From surviving to thriving - parental experiences of hospitalised infants with congenital heart disease undergoing cardiac surgery: a qualitative synthesis. J Pediatr Nurs 2020; 51: 32–41.
17. Bratt E-L, Järvholm S, Ekman-Joelsson B-M, Mattson L-Å., Mellander M. Parent's experiences of counselling and their need for support following a prenatal diagnosis of congenital heart disease - a qualitative study in a Swedish context. BMC Pregnancy Childbirth 2015; 15: 1–7.
18. Ellinger MK, Rempel GR. Parental decision making regarding treatment of hypoplastic left heart syndrome. Adv Neonatal Care 2010; 10: 316–322.
19. Rempel GR, Cender LM, Lynam MJ, Sandor GG, Farquharson D. Parents' perspectives on decision making after antenatal diagnosis of congenital heart disease. J Obstet Gynecol Neonatal Nurs 2004; 33: 64–70.
20. Giacomini MK, Cook DJ. Users' guides to the medical literature: XXIII. Qualitative research in health care A. Are the results of the study valid? Evidence-based Medicine Working Group. JAMA 2000; 284: 357–362.
21. Kitzinger J. Qualitative research. Introducing focus groups. BMJ 1995; 311: 299–302.
22. Lederman LC. Assessing educational effectiveness: the focus group interview as a technique for data collection. Commun Educ 1990; 39: 117–127.
23. Namey E, Guest G, McKenna K, Chen M. Evaluating bang for the buck: a Cost-Effectiveness comparison between individual interviews and focus groups based on thematic saturation levels. Am J Eval 2016; 37: 425–440.
24. Guest G, Namey E, McKenna K. How many focus groups are enough? building an evidence base for nonprobability sample sizes. Field Methods 2017; 29: 3–22.
25. Barriball KL, While A. Collecting data using a semi-structured interview: a discussion paper. J Adv Nurs 1994; 19: 328–335.
26. Kallio H, Pietilä AM, Johnson M, Kangasniemi M. Systematic methodological review: developing a framework for a qualitative semi-structured interview guide. J Adv Nurs 2016; 72: 2954–2965.
27. Guest G, Bunce A, Johnson L. How many interviews are enough? an experiment with data saturation and variability. Field Methods 2006; 18: 59–82.
28. Crabtree BF, Miller WL. Doing qualitative research. Sage Publications, Inc, Thousand Oaks, CA, 1992.
29. Tracy SJ. Qualitative quality: eight, Big-Tent, criteria for excellent qualitative research. Qual Inq 2010; 16: 837–851.
30. Stanovich KE, West RF. Reasoning independently of prior belief and individual differences in actively open-minded thinking. J Educ Psychol 1997; 89: 342–357.
31. Evans JSBT. In two minds: dual-process accounts of reasoning. Trends Cogn Sci 2003; 7: 454.
32. Scott SG, Bruce RA. Decision-making style: the development and assessment of a new measure. Educ Psychol Meas 1995; 55: 818.
33. Delaney R, Strough J, Parker AM, Bruine de Bruin W. Variations in decision-making profiles by age and gender: a cluster-analytic approach. Pers Individ Diff 2015; 85: 19–24.
34. Miller SM, Fang CY, Diefenbach MA, Bales CB. Tailoring psychosocial interventions to the individual's health information-processing style: The influence of monitoring versus blunting in cancer risk and disease. In: Baum A, Andersen BL (eds). Psychosocial Interventions for Cancer. American Psychological Association, Washington, DC, 2001, p. 343–362.
35. Charles C, Gafni A, Whelan T. Shared decision-making in the medical encounter: What does it mean? (Or it takes at least two to tango). Soc Sci Med 1997; 44: 681–692.
36. Kon AA, Ackerson L, Lo B. How pediatricians counsel parents when no "best-choice" management exists: lessons to be learned from hypoplastic left heart syndrome. Arch Pediatr Adolesc Med 2004; 158: 436–441.
37. Lee-Poy M, Stewart M, Ryan BL, Brown JB. Asking patients about their religious and spiritual beliefs: cross-sectional study of family physicians. Can Fam Physician 2016; 62: e555–e561.
38. Connor JA, Kline NE, Mott S, Harris SK, Jenkins KJ. The meaning of cost for families of children with congenital heart disease. J Pediatr Health Care 2010; 24: 318–325.
39. Daily J, FitzGerald M, Downing K, et al. Important knowledge for parents of children with heart disease: parent, nurse, and physician views. Cardiol Young 2016; 26: 61–69.
40. Fagerlin A, Pignone M, Abhyankar P, et al. Clarifying values: an updated review. BMC Med Inform Decis Mak 2013; 13: 1–7.
41. Witteman HO, Gavaruzzi T, Scherer LD, et al. Effects of design features of explicit values clarification methods: a systematic review. Med Decis Mak 2016; 36: 760–776.
42. Braun LT, Grady KL, Kutner JS, et al. Palliative care and cardiovascular disease and stroke: a policy statement from the American Heart Association/American Stroke Association. Circulation 2016; 134: e198–e225.
43. Prsa M, Holly CD, Carnevale FA, Justino H, Rohlicek CV. Attitudes and practices of cardiologists and surgeons who manage HLHS. Pediatrics 2010; 125: e625–e630.
44. Rychik J. What does palliative care mean in prenatal diagnosis of congenital heart disease? World J Pediatr Congenit Heart Surg 2013; 4: 80–84.
45. Austin CA, Mohottige D, Sudore RL, Smith AK, Hanson LC. Tools to promote shared decision making in serious illness: a systematic review. JAMA Intern Med 2015; 175: 1213–1221.
46. May R, Thompson J. The role of pediatric palliative care in complex congenital heart disease: three illustrative cases. J Palliat Med 2017; 20: 1300–1303.
47. Keele L, Keenan HT, Sheetz J, Bratton SL. Differences in characteristics of dying children who receive and do not receive palliative care. Pediatrics 2013; 132: 72–78.

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press

48. Feudtner C, Kang TI, Hexem KR, et al. Pediatric palliative care patients: a prospective multicenter cohort study. Pediatrics 2011; 127: 1094–1101.
49. DeSteno D, Gross JJ, Kubzansky L. Affective science and health: the importance of emotion and emotion regulation. Health Psychol 2013; 32: 474–486.
50. Hanschmidt F, Linde K, Hilbert A, Riedel- Heller SG, Kersting A. Abortion stigma: a systematic review. Perspect Sex Reproduct Health 2016; 48: 169–177.
51. Pinto NM, Weng C, Sheng X, et al. Modifiers of stress related to timing of diagnosis in parents of children with complex congenital heart disease. J Matern Fetal Neonatal Med 2016; 29: 3340–3346.
52. Kaplowitz MD, Hoehn JP. Do focus groups and individual interviews reveal the same information for natural resource valuation? Ecol Econ 2001; 36: 237–247.

https://doi.org/10.1017/S1047951121003218 Published online by Cambridge University Press