COPING, PARENTING AND DECISION MAKING FOR MOTHERS OF INFANTS RECEIVING TREATMENT FOR A CONGENITAL ANOMALY IN THE NICU

by
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Abstract

Background: A prenatal or postnatal congenital diagnosis is a known source of parental distress and disrupts parental adaptation, often to a greater degree in mothers. The purpose of this study was to explore the coping, parenting, and decision-making adaptation processes for mothers who receive a prenatal vs. postnatal congenital diagnosis for their infant who requires treatment immediately after birth in the NICU.

Methods: This study utilized a cross-sectional, sequential explanatory mixed methods design. Quantitative surveys and in-depth semi-structured qualitative interviews were conducted with mothers faced with decision making for their infant in the NICU diagnosed with a congenital anomaly between July 2016 and January 2018. T-tests and correlations were used to analyze quantitative data, qualitative content analysis was used to analyze qualitative data, and data matrices were used to combine quantitative and qualitative data.

Results: There were 37 participants in the quantitative sample (mean age 30.6 ±6.0) and 20 of these participants (mean age 31.25 ±5.73) agreed to qualitative interviews; approximately two-thirds of participants were White, 25% African American, and 10% were Hispanic. On quantitative surveys, mothers in the postnatal group demonstrated more engaged decision making (86%), compared with mothers in the prenatal group (52%). Mothers in the prenatal group utilized significantly more acceptance coping than mothers in the postnatal group (U=78; p=0.01). Parenting values among mothers in the prenatal and postnatal groups were similar; both groups placed greatest priority on the parenting values of making medical decisions and focusing on their child's health, and

lowest value on keeping a positive attitude. Qualitative findings revealed barriers and facilitator themes of coping, parenting, and decision making, including access, behavioral, cognitive, communication, emotional, environmental, relational, and spiritual barriers and facilitators. Mixed methods findings explained that mothers in both the prenatal and postnatal groups were engaged in decision making, but at different time points and in different ways.

Conclusions: Overall, the findings show trends in NICU coping, parenting and decision-making for mothers who have received a prenatal vs. postnatal diagnosis. Future researchers should explore study variables in specific congenital anomaly populations and racial/ethnic minority populations where congenital anomalies are more prevalent.

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Dedication

For the NICU parents and infants who have inspired this work.

Preface

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CHAPTER ONE: INTRODUCTION

Background

Congenital anomalies account for over a quarter million neonatal deaths and 3.2 million birth-defect related disabilities worldwide each year (World Health Organization, 2016). In the U.S. context, congenital anomalies impact one in 33 infants (CDC, National Center for Health Statistics, 2017). Moreover, congenital anomalies are the number one cause of infant death (CDC, National Center for Health Statistics, 2017), and frequently result in neonatal intensive care (NICU) hospitalization.

In the NICU, parents must quickly adapt to be part of treatment decisions related to congenital anomalies. Recommendations by the American Academy of Pediatrics (AAP) stress the need to engage parents of high risk newborns in shared decision making (SDM) with health care providers in order to include parent values and preferences, based on culture, religion, and other factors, as treatments are considered (American Academy of Pediatrics Committee on Fetus and Newborn & Bell, 2007). Parent engagement in decision making is imperative, given that health care providers have their own biases about treatment options and often overestimate the burden of long-term outcomes, like disability, which may not reflect the values of parents (Penticuff & Arheart, 2005; Saigal, 2000; Saigal et al., 2006).

Despite this AAP recommendation that parents of high risk newborns engage in shared decision making, parent engagement in decision making has been shown to vary from independent decision making (i.e. parents seek information from, but make decisions independent of health care providers), to shared decision making (i.e. decisions

are made with input from both parents and health care providers), to reliance upon health care providers for decision making (Gagnon & Recklitis, 2003). In addition, several barriers may limit parent engagement in decision making, including young parent age, low educational attainment, low levels of coping, and parent perceptions regarding infant illness severity that differ from those of the health care team. Certain infant factors, such as timing of congenital anomaly diagnosis, low birth weight and early gestation age, are other contextual variables that may impact parent decision making.

In previous studies, mothers display more difficulty than fathers following a congenital diagnosis, exhibiting greater anxiety, depression, and reporting a lower quality of life, (Fonseca, Nazare, & Canavarro, 2012). While these previous studies have explored the psychological distress of mothers who receive news of a congenital diagnosis for their infant, no studies, to our knowledge, have explored the coping, parenting, and decision making of mothers of infants necessitating treatment decision making for a congenital anomaly during NICU hospitalization. Exploring the processes, barriers, and facilitators to coping, parenting and decision making for mothers of NICU infants receiving treatment for a congenital anomaly could guide the development and implementation of future decision support interventions. These interventions could improve engagement of parents in decision making with providers, subsequently optimizing treatment and health outcomes for children, and family-centered care for parents.

Purpose

The purpose of this study was to explore NICU mothers' coping, parenting, and decision making related to NICU treatments for their infant's congenital anomaly.

Since previous studies have suggested adaptation differences between mothers who receive a prenatal vs. postnatal congenital diagnosis for their infant, we focused our study on differences in mothers of infants in the NICU who received a prenatal vs. postnatal diagnoses. A cross-sectional, sequential explanatory mixed methods design was used, guided by the Roy Adaptation Model.

Specific Aims

Quantitative Aim: 1. To estimate differences and associations of coping factors (infant illness severity perception; general coping; religious coping), parenting values (i.e. "good parent" attributes), and decision making engagement for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth.

Hypothesis 1.1: Mothers who learn of their infant's congenital diagnosis prenatally will exhibit higher levels of positive coping than mothers who learn of their infant's congenital diagnosis postnatally.

Hypothesis 1.2: Mothers who learn of their infant's congenital diagnosis prenatally will exhibit greater decision making engagement than mothers who learn of their infant's congenital diagnosis postnatally.

Qualitative Aim: 2. Explore barriers and facilitators related to coping, parenting, and decision making for mothers of NICU infants with a congenital anomaly.

Mixed Aim: 3. To describe and compare the barriers and facilitators of coping, parenting, and decision making for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth.

Conceptual Framework

The Roy Adaptation Model (RAM), developed by Sr. Callista Roy, informs this research study. The RAM describes an individual's internal and external systems used to adapt to a stressor and posits that one form of adaptation to stressful stimuli is functioning within an established role (C. Roy, 2009). Variables in all of the RAM's four concepts (Stimuli, Coping Process, Adaptive Modes, and Adaptive Responses) were identified for this study; portions of the model highlighted in grey will be addressed (See Figure 1). The focal stimulus, or stimulus confronting the mother, are defined in this study as the congenital diagnosis and NICU admission. The contextual stimuli, or stimuli that add meaning to the focal stimuli, are defined in this study as parent characteristics (i.e. age and education) and infant characteristics (i.e. timing of congenital diagnosis, birth weight, gestational age). Cognator variables, which are an individual's cognitive and emotive processing (C. Roy, 2009), as well as perceptions, are defined in this study as parents' perception of infant illness severity and general/religious coping; religious/spiritual factors were identified in prior studies as important dimensions of being a 'good parent' (Feudtner et al., 2015; Hinds et al., 2009; October, Fisher, Feudtner, & Hinds, 2014). Under the Role Function Mode, which describes how an individual functions within an established role, we've identified dimensions of being a 'good par(ent'(C. Roy, 2009). Finally, the Adaptive Response, which is the outcome of an individual's adaptation to a stressful stimuli, is defined in this study as decision making engagement, and is either engaged (independent or shared) or unengaged (reliant on physician). The RAM has guided at least 12 prior studies related to coping with serious life events, including a study of parents' coping with the care of their child with

cancer (Yeh, 2003). Specific concepts as they relate to the RAM in this study are described below.

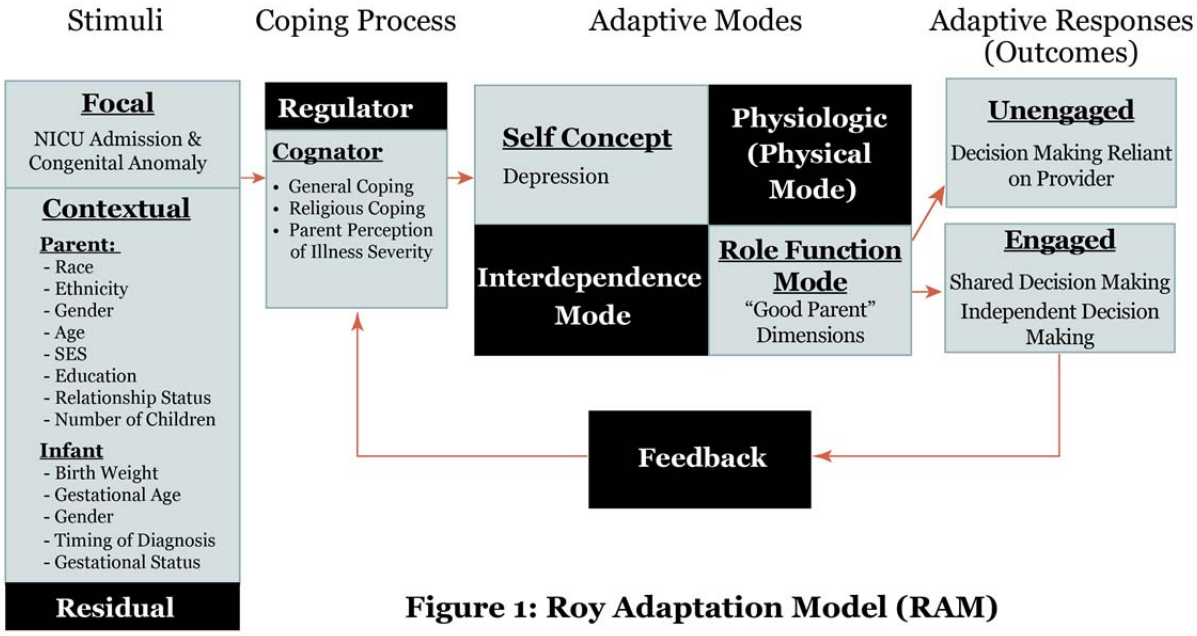

Figure 1: Roy Adaptation Model (RAM)

Key Conceptual and Variable Explanations

Stimuli Variables

Congenital anomalies

Congenital anomalies are structural or functional anomalies that occur during the fetus' development before birth (World Health Organization, 2016). These anomalies may be caused by genetic factors or environmental influences, or a combination of both. Congenital anomalies can also be caused by conditions impacting the mother during her pregnancy, including maternal chronic illnesses (i.e. diabetes, hypertension, autoimmune diseases), maternal infections (i.e. chicken pox, rubella, cytomegalovirus), substance abuse, ingestion of certain medications, and raw or undercooked foods. Although the

cause of congenital anomalies can be identified in certain cases, in other cases the cause is unclear (World Health Organization, 2016). The severity of congenital anomalies ranges from mild to severe. Congenital anomalies are defined as the focal stimuli for this study.

Parent demographics

Several parent demographics are reported as potential limiting factors to parent engagement in decision making. Young parent age and low educational attainment may limit parents' cognitive processing or be associated with less active involvement in decision making, both of which challenge decision making (Boss, Donohue, & Arnold, 2010; Kraetschmer, Sharpe, Urowitz, & Deber, 2004; Street, Gordon, Ward, Krupat, & Kravitz, 2005; Tarini, Christakis, & Lozano, 2007). Additionally, parents with low levels of coping may be unable to cognitively process information about their infant's congenital anomaly, which can limit a parent's ability to be part of decision making (J. Lalor, Begley, & Galavan, 2009; J. G. Lalor, Begley, & Galavan, 2008). More specifically, high levels of negative religious coping (i.e. feeling abandoned by or anger at God) have been shown to be associated with low parental investment and high distress, which can limit engagement in decision making (Dumas & Nissley-Tsiopinis, 2006). Several studies indicate that mothers of NICU infants have more difficulty than fathers adapting to a NICU admission, resulting in more long-term mood disturbances, lower levels of long-term attachment with their child, and potential child behavior problems and child cognitive deficits later in life (Affleck, Tennen, & Rowe, 1991; Doering, Dracup, & Moser, 1999; Doering, Moser, & Dracup, 2000; Pinelli, 2000). Together, these factors suggest a need to support the decision making role of mothers with a NICU infant

(Obeidat, Bond, & Callister, 2009). Parent demographics, such as relationship status, are identified as a contextual stimuli for this study.

Infant demographics

Infant characteristics may also influence parent engagement in decision making. Studies indicate that when infants are diagnosed prenatally with a congenital anomaly, parents may experience increased burden and grief, compared to when infants are diagnosed with a congenital anomaly postnatally (Brosig, Whitstone, Frommelt, Frisbee, & Leuthner, 2007). An increase in burden and grief may impact parents' ability to make decisions. Moreover, infants born with low birth weight and at an early gestational age pose complex medical, social and ethical challenges, which make decision making for parents less certain, and potentially more complicated, since prognostic outcomes are unpredictable (Nadroo, 2011). Infant demographics are also identified as contextual factors for this study.

Coping Process Variables

Parent perception of infant illness severity

Illness perceptions are comprised of interrelated cognitive beliefs about the cause of an illness, the types and timeline of symptoms, consequences of the illness, and beliefs about control or cure (Petrie & Weinman, 2006). Differences in parental and health care provider perceptions about the severity of an infant's illness can complicate shared decision making. For example, parents who perceive the severity of their infant's illness differently than the health care team may have difficulty understanding the need for specific treatment interventions (Brooks, Rowley, Broadbent, & Petrie, 2012). For parents with high-risk newborns, the perception of their infant's illness severity is

associated with the level of stress they experience during their infant's hospital admission, which can subsequently influence ability to engage in decision making (Brooks et al., 2012). Parents' illness perceptions, as they pertain to their infant, are considered a coping process variable in this study and were measured using the Brief Illness Perception Questionnaire.

General coping

A parent whose infant is admitted into the NICU may cope in a variety of ways. Appraisal-focused coping is a cognitive adaptive form of coping, which involves the parent challenging his/her own thinking or assumptions (Weiten & Lloyd, 2008). A second type of coping, problem-focused coping, is also an adaptive form of coping, whereby a parent attempts to eliminate a stressor (Weiten & Lloyd, 2008). A parent using problem-focused coping may seek out further information, a new skill, or plan in some other way to eliminate the stressor. A third form of coping is emotion-focused coping. Emotion-focused coping is targeted at managing the emotions that arise due to stressors. One common form of emotion-focused coping includes seeking emotional social support (Carver, Scheier, & Weintraub, 1989). Understanding these different forms of coping is important, since parents may tend to utilize certain types of coping over others in particular situations. Items on the Brief COPE scale, which were used in this study, assessed these various forms of coping.

Religious and spiritual coping

Prior literature on parents' use of spiritual and religious coping does not differentiate between the roles of each. In the present study, religious coping was defined as coping that involves shared faith, beliefs and adherence to practices and rituals that

enable individual expression of a connectedness to a Higher Power or to God (Davies, Brenner, Orloff, Sumner, & Worden, 2002; Koenig, 2007). Spiritual coping, on the other hand, will be defined as coping focused on "one's personal search for meaning and purpose, and a trusting relationship to something greater than oneself that is significantly meaningful" (Robinson, Thiel, Backus, & Meyer, 2006).

Several studies report that religious and/or spiritual coping are resources for parents in and beyond the NICU, especially in end-of-life and grief situations. For instance, parents who drew upon their belief in an afterlife and felt it was a reality for their own infant coped with their infant's death, responding with less fear, depression and more acceptance (Armentrout, 2009; Carroll, Mollen, Aldridge, Hexem, & Feudtner, 2012; Hexem, Mollen, Carroll, Lanctot, & Feudtner, 2011). Religious and/or spiritual coping, like other means of coping, can be either positive or negative. The RCOPE instrument in this study explored parents' use of both positive and negative forms of religious coping. The qualitative portion of this study will help inform differences between parents' religious and/or spiritual coping.

Adaptive Mode Variables

Depression

While no studies were found that examined depressive symptoms and parent decision making in the NICU setting, numerous studies report the prevalence of depressive symptoms in mothers of infants hospitalized in the NICU, which has been reported as high as 63% (Davis, Edwards, Mohay, & Wollin, 2003; Lefkowitz, Baxt, & Evans, 2010; Miles, Holditch-Davis, Schwartz, & Scher, 2007; Pinelli, 2000; Singer et al., 1999). Mothers of preterm infants are at even higher risk of depressive symptoms

than mothers of term infants (Ballantyne, Benzies, & Trute, 2013). Other risk factors for depressive symptoms in mothers of NICU infants include immigrant status, single parent status, and low level of social support (Ballantyne et al., 2013). Fathers of NICU infants, although at lower risk of depression, have also reported such symptoms (Gonulal, Yalaz, Altun-Koroglu, & Kultursay, 2014). The present study will add to the NICU literature by describing how depression impacts parents, as they seek to achieve their ideal of deciding as a 'good parent' for their ill child.

The role of parenting

Four descriptive treatment decision-making studies by Hinds and colleagues describe the parent-reported theme of being, or acting as, a 'good parent.' This theme was identified in up to 84% of interviews with parents making decisions for their ill child with incurable cancer (Hinds et al., 1997; Hinds et al., 2000; Hinds et al., 2001; Hinds, Burghen, & Pritchard, 2007; Pritchard et al., 2008). Parents in these studies reported that discussing their parenting values, or their perception of "doing what a 'good parent' would do," facilitated decision making at the end of their child's life by strengthening parent-provider communication and improving decision making satisfaction. The relative importance of each of these 'good parent' dimensions may vary for individual parents; however, a ranking of these 12 dimensions has been used to identify "good parent" values that are important for parents in particular pediatric health care settings (Feudtner et al., 2015). For example, in previous 'good parent' studies, religious or spiritual coping has been cited as a key dimension that helped facilitate parents' decision making and provide psychological support during the decision making process (Allen, 2014; Einarsdottir, 2009; Hinds et al., 2009; Kavanaugh, Savage, Kilpatrick, Kimura, &

Hershberger, 2005; Michelson et al., 2009; October et al., 2014; Peng, Liu, Chen, & Bachman, 2012). Despite this previous work, what is still unknown is which 'good parent' values are specific to NICU parents, and how such values may help parents engage in decision making for their infant. Previous good parent studies suggest that identifying factors, which help parents achieve their definition of deciding as a 'good parent' could be protective, in terms of parent health outcomes (Hinds et al., 2012), yet to date, these factors remain unexplored. More in-depth knowledge about parenting values of NICU parents and how these values influence parent decision making will inform the development of decision support interventions that promote parent engagement in complex NICU decision making. The Good Parent Ranking Exercise will identify which "good parent" values are important to NICU parents and qualitative interviews will further explore these, and other values, that may be important to NICU parents.

Adaptive Response Variable

Decision making engagement

Decision making engagement is defined as the adaptive response and study outcome variable. Parents' decision making style, specific to treatment decision making for their infant's congenital anomaly, served as a proxy measure for decision making engagement. The Control Preference Scale was used to identify parents' decision making style as either engaged (i.e. decision making performed either in an independent or shared fashion) or unengaged (i.e. decision making was delegated to the health care team) (Degner & Sloan, 1992).

Significance/Innovation

The proposed study informs the approach to treatment decision making for infants with unpreventable congenital anomalies, the numbers of which are significant, given that the cause of over 70% of congenital anomalies are still unknown (March of Dimes., 2017). Until a wider portfolio of prevention approaches can be identified, infants will continue to be born with congenital anomalies and require complex decision making.

This study facilitated a systematic integration of previous findings about parenting an acutely ill child (i.e. "good parent" studies) in a new study population, NICU mothers. The cross-sectional approach allowed for a more in-depth exploration of general and religious coping, which in previous NICU studies has been retrospective, qualitative, and focused on end-of-life decision making (Armentrout, 2009; Brosig, Pierucci, Kupst, & Leuthner, 2007; Hexem et al., 2011; Michelson et al., 2009; Peng et al., 2012; Robinson et al., 2006; Sutan & Miskam, 2012). Additionally, while prior studies that explore decision making in the NICU include predominantly Caucasian samples (Armentrout, 2009; Brosig et al., 2007; R. Roy, Aladangady, Costeloe, & Larcher, 2004), this study aimed to include a more racially diverse sample.

Dissertation Organization

This dissertation consists of five chapters. Chapter one provides an overview of the study, with a description of the purpose, aims, conceptual framework and important study concepts.

Chapter two (manuscript one) is an integrative review of the relevant qualitative, quantitative, and mixed methods literature on the influence of parents' religion and/or spirituality on decision making for their critically ill child. In addition to being a review

of the critical articles, this manuscript provides recommendations for future research in the area of religion, spirituality, and parent decision making. This manuscript is being prepared for submission to the Journal of Advanced Nursing.

Chapter three (manuscript two) presents a subset of the qualitative findings from semi-structured interviews, which explored mothers' perceptions of barriers and facilitators to coping, parenting, and decision making in the NICU. The manuscript reports decision-making findings from the prenatal and postnatal groups.

Chapter four (manuscript three) reports the mixed methods findings from the quantitative surveys and qualitative surveys, which were part of this study. The manuscript compares participants based on timing of when they learned of their infant's congenital diagnosis (prenatal vs. postnatal) and reports barriers and facilitators to decision making, coping, and parenting.

Chapter five presents 1) a concise summary of the dissertation findings reported in chapters three and four, 2) study strengths and limitations, and 3) study implications.

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CHAPTER TWO: MANUSCRIPT ONE

The Influence of Parents' Religion or Spirituality on Decision Making for their Critically Ill Child: A Mixed Methods Integrative Review

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Abstract

Aim: To systematically review the literature describing the influence of parents' religion and/or spirituality on decision making for their critically ill child.

Background: Parents' religion and/or spirituality can influence which treatments they accept or decline for their child or how they respond to significant health care events. A greater understanding of the influence of parents' religion and spirituality on their decision making for their critically ill child may lead to the development and testing of decision support interventions, which could improve shared decision making between parents and providers, subsequently optimizing treatment and health outcomes for children and families.

Design: An integrative review incorporating quantitative, qualitative, and mixed methods data.

Data sources: Keyword searches of literature investigating the relationship between parents' religion or spirituality and their medical decision making for a critically ill child were conducted (origin –January 2017) using five databases: PubMed, CINAHL plus, PsychInfo, Scopus, and Embase.

Review methods: The integrated review method described by Whittemore and Knafl was used to guide this review. Titles and abstracts were screened for inclusion and exclusion criteria and the remaining full-text articles were then reviewed. Study quality was assessed using standardized critical appraisal instruments from The Joanna Briggs Institute. Data were extracted using the constant comparison method.

Results: A total of 24 studies, including 15 qualitative studies, six quantitative studies and three mixed methods studies were included in this review from 1,642 non-duplicated

records initially retrieved. Three themes related to how parents' religion or spirituality influences decision making for their critically ill child were generated: 1) Religion and spirituality as guidance during decision making, 2) Religion and spirituality as forms of comfort and support during the decision making process, and 3) Religion and spirituality as a source of meaning, purpose, and connectedness in the experience of decision making.

Conclusion: This review suggests that parents' religion and/or spirituality is an important and primarily positive influence in their medical decision making for a critically ill child. Future research should explore how to facilitate health care providers' understanding of parents' religious and/or spiritual beliefs and how to incorporate these beliefs into medical decision making.

Key Words: parents, medical decision making, critical care, spirituality, religion

Introduction

Children admitted into acute health care settings receive treatment for a variety of disease states, many of which are complex or chronic in nature and require involvement of surrogate decision makers (Society of Critical Care Medicine., 2018). Parents are typically understood to be the appropriate surrogates for a hospitalized child, making decisions based on the best interests of the child, and are therefore asked to engage in decision making with the health care team when their child becomes ill (Birchley, 2014; Muirhead, 2004). In fact, the recommended approach to decision making in pediatrics, as in other health care contexts, is the shared decision making model (SDM) (Birchley, 2014; Opel, 2017). SDM implies that both parties, parents and providers, share information and work together to develop an approach to a preferred treatment plan (Opel, 2017). Part of SDM in pediatrics includes exploring and incorporating parents' values, preferences and beliefs, including religious and spiritual beliefs (Opel, 2017). For many parents of critically ill, hospitalized children, their religion and spirituality may influence which treatments they accept or decline for their child or how they approach significant events, such as the death of a child while hospitalized (Puchalski, 2004). To inform understanding of how parents' religion and spirituality influence their decision making for their critically ill child, an integrative review was undertaken.

Background

In this review, we conceptualize religion and spirituality as two distinct, although related concepts (Koenig, King, & Carson, 2012). Religion is conceptualized as shared faith, beliefs and adherence to practices and rituals that enable individual expression of

connectedness to a Higher Power or to God (Davies et al., 2002; Koenig, 2007). The language of religion is also used interchangeably with religiosity or religious involvement and those beliefs and behaviors associated with organized religious institutions (Koenig, 2007). For clarity, we use the terminology religion throughout this paper when referencing a set of beliefs, practices, or rituals associated with organized religious affiliations.

Spirituality, on the other hand, is one's personal search for meaning and purpose, and a trusting relationship to something greater than oneself that is significantly meaningful (Robinson et al., 2006). Spirituality is further conceptualized as deriving meaning of individual experiences through dimensions of connectedness (Reed, 1992). These dimensions of connectedness that empower individuals to move beyond stressful situations can be intrapersonal (within oneself), interpersonal (between others and the environment), or transpersonal (beyond the self, extending to God or another higher power) (Reed, 1992).

Parents, particularly those who have critically ill children in the intensive care setting, report significant spiritual needs and cite the importance of their faith during their child's inpatient stay; yet few parents experience the incorporation of values related to their religion and spirituality by the health care team during their child's inpatient stay (Meert, Thurston, & Thomas, 2001; Meert, Thurston, & Briller, 2005). Several studies report the lack of elicitation of parents' religious or spiritual beliefs, due to provider discomfort or lack of provider training in performing spiritual assessments (Catlin et al., 2001; Lo et al., 2002). When providers fail to elicit parents' beliefs related to their

religion and spirituality, treatments for children risk being uninformed by parents' values. Furthermore, treatments may also run the risk of being biased by health providers,' who have been shown to overestimate the burden of certain outcomes that parents are willing to accept for their child (Penticuff & Arheart, 2005; Saigal, 2000; Saigal et al., 2006). A better understanding of how parents' religious and spiritual beliefs influence their decision making when their child is hospitalized may lead to the development and testing of decision support interventions, which could improve shared decision making between parents and providers, subsequently optimizing treatment and health outcomes for children, and family- centered care for parents.

Methods

Aim

The aim of this mixed methods systematic review was to summarize and critique the existing literature exploring the influence of parents' religion and/or spirituality on their decision making for their critically ill child age 0 to 18. The review questions were: 1) Which religious and spiritual factors influence parent decision making? 2) How do parents of critically ill children use religious and/or spiritual factors in their health care decision making? 3) Who supports the use of parents' religion or spirituality during decision making for their critically ill child?

Design

This integrative review of the literature was conducted using Whittemore and Knafl's updated integrative review method (Whittemore & Knafl, 2005). PRISMA reporting guidelines for systematic reviews and meta-analysis were also followed (Moher, Liberati, Tetzlaff, Altman, & PRISMA Group, 2009). An integrative review

approach was selected because it allows for inclusion of diverse sources (i.e. qualitative, quantitative, mixed methods) to understand a phenomenon analyzed, and to then inform evidence-based practice initiatives (Whittemore & Knafl, 2005). Whittemore and Knafl's integrative review approach includes five stages: problem identification, literature search, data evaluation, data analysis, and presentation of the resulting data integration. Using this integrative approach, this review aims to present the state of the science pertaining to the influence of parents' religion and spirituality on their decision making for their critically ill child, with the goal of contributing to policy, theory development, and clinical practice guidelines (Whittemore & Knafl, 2005).

Search Methods

In consultation with a health librarian, the lead author conducted a review of the literature using PRISMA guidelines to identify relevant peer-reviewed literature on parent decision-making and religion/spirituality. The following five databases were searched from database inception to January 2017: PubMed, Cumulative Index to Nursing and Allied Health Literature (CINAHL plus), Embase, Scopus, and PsychInfo. Search criteria were developed using the following medical subject headings (MeSH) and non-MeSH search terms: *religion, spirituality, parents/mother/father/surrogate, and parental decision making*. Limits were set to include only articles published in English. Additionally, a manual search of relevant literature and references was performed to identify additional studies applicable to the review.

Inclusion and Exclusion Criteria

Articles were included in the review if they satisfied the following eligibility criteria: 1) were primary, peer-reviewed research articles; 2) addressed the relationship

between parents' religion and/or spirituality and medical decision making for their child 21 years of age and under; 3) discussed decision making in the intensive or critical care setting. Studies were excluded if the research focus was on 1) public health topics (i.e. decisions related to health screening and vaccinations); 2) decisions of a less critical nature (i.e. circumcision and breastfeeding); 3) decision making from the health provider's perspective; 4) strictly hypothetical parent decision making 5) decisions about the use of complimentary or alternative medicine; or 6) decisions related only to participation of a child in research.

Search Outcome

The search strategy returned 3,027 potentially relevant citations from the five databases and 11 articles from a hand search. All citations were imported into Covidence®, a web-based software for conducting systematic reviews, and duplicates removed, resulting in 1,642 citations. Two reviewers (M.K.U. and K.D.) independently reviewed the 1,642 titles and abstracts of studies against inclusion and exclusion criteria, which resulted in the exclusion of 1,507 articles. Conflicts about eligibility/exclusion of articles were discussed until consensus was reached on whether the criteria were met for a particular article. The full text of the 135 remaining articles was assessed by the same two researchers for eligibility, again resolving conflicts through consensus. Of these 135 articles, 111 articles were excluded because they either did not utilize a research approach; failed to focus on parent decision making; discussed decision making by parents during the child's postmortem state or before the child was born; focused on non-pediatric or outpatient populations; or mentioned spirituality and/or religion, but did not explore either of these as variables relative to parents in the research study. A total of 24

articles were included for this review (Figure 1).

Quality Appraisal

Two reviewers (M.K.U. and K.D.) independently assessed the 24 full-text articles using standardized Joanna Briggs Institute (JBI) critical appraisal instruments. Evaluating the quality of studies in an integrative review where diverse sources are included is complex. We chose to use quality appraisal tools from JBI given that the 24 studies in our review utilized two main study designs and JBI had quality appraisal tools specific to these designs. Qualitative studies were assessed using the JBI Critical Appraisal Checklist for Qualitative Research and quantitative studies using the Critical Appraisal Checklist for Analytical Cross Sectional Studies (Joanna Briggs Institute 2017). For the mixed methods studies, the qualitative and quantitative sub-study sections were appraised separately using the appropriate critical appraisal tool. Disagreements between quality appraisals were resolved through discussion. No study was excluded on the basis of the quality evaluation given the relative lack of information on our topic of interest, although studies with low rigor contributed less to the analytic process (Whittemore & Knafl, 2005).

Overall, the quality of qualitative studies was moderate to high while the quality of quantitative studies fluctuated between weak to moderately high and consisted of only cross-sectional descriptive designs. Two mixed methods studies had a higher appraisal score for the quantitative portion of the study compared to the qualitative portion (Coughlin, Hernandez, Richardson, & da Silva, 2007; Tamburro, Shaffer, Hahnlen, Felker, & Ceneviva, 2011); the third mixed methods study had relatively equal appraisal scores for the quantitative and qualitative portion (Brosig et al., 2007). Qualitative data

analysis methods often had inconclusive or no statements about reaching data saturation and most failed to orient the study by stating a philosophical perspective or research methodology. The influence of the researcher on the research was also largely unaddressed. Quantitative studies, overall, had limited generalizability and power, lacked diverse samples, utilized mostly self-report measures, and many introduced recall bias due to their retrospective nature.

Data Abstraction

Data from the 24 included articles in the review were extracted by one researcher (M.K.U.) using a data extraction form, capturing study design, sample, setting, decision type, and summary of religion and spirituality and decision making findings (see Table 1). A second researcher (K.D.) was trained on the data extraction form and independently confirmed extraction of these data. Disagreements were resolved through discussion. For one mixed methods study, only data from the qualitative sub-study was relevant to this review, so abstraction of data from this article was limited to the qualitative data (Brosig et al., 2007).

Data Synthesis

We utilized the five stages of data analysis, as outlined by (Whittemore & Knafl, 2005), in performing our data synthesis: data reduction, data display, data comparison, conclusion drawing, and verification. Table 1 summarizing the papers included in this review, accomplished data reduction, or the division of studies by subgroups according to their types of evidence (qualitative, descriptive), and created a data display matrix that allowed for further synthesis using a constant comparison method. A constant comparison method of analysis is compatible with integrative reviews that synthesize

diverse methodologies and allows for movement among codes found in the qualitative and quantitative data (Whittemore & Knafl, 2005). Using the data display matrix (Table 1), the first (M.K.U.) and second author (J.B.H.) compared religion/spirituality and decision making data from each article's data extraction form, and identified patterns and major themes. These themes were then verified by a third author (M.T.N.).

Results

Study Characteristics

Study designs. The majority (n=15) of the 24 eligible studies utilized qualitative designs, with authors applying the following data collection methods: in-person/telephone interviews (Armentrout, 2009; Bluebond-Langner, Belasco, Goldman, & Belasco, 2007; Boss, Hutton, Sulpar, West, & Donohue, 2008; Carroll et al., 2012; Einarsdottir, 2009; Ellinger & Rempel, 2010; Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Meyer, Ritholz, Burns, & Truog, 2006; Pepper, Rempel, Austin, Ceci, & Hendson, 2012; Prows & McCain, 1997; Robinson et al., 2006; Sharman, Meert, & Sarnaik, 2005); chart data analysis (Peng et al., 2012); and participant observation (Bluebond-Langner et al., 2007). Quantitative study designs (n=6) were either descriptive or correlational, and data collection in these studies involved surveys and questionnaires (Arutyunyan, Odetola, Swieringa, & Niedner, 2018; Madrigal et al., 2016; Meyer, Burns, Griffith, & Truog, 2002) or retrospective chart reviews (Hileli, Weyl Ben Arush, Hakim, & Postovsky, 2014; R. Roy et al., 2004; Schimmel, Steinberg, Mimouni, & Yekel, 2016). Three of the 24 eligible studies utilized a mixed methods design (Brosig et al., 2007; Coughlin et al., 2007; Tamburro et al., 2011). Data were collected through surveys and semi-structured interviews (Brosig et al., 2007), chart review (Coughlin et al., 2007), or

discussions about goals of care/importance of religion/spirituality and chart review (Tamburro et al., 2011). For one of these mixed methods studies (Brosig et al., 2007), only the qualitative component was applicable to this review based on the defined inclusion criteria. Therefore, given mixed methods synthesis guidelines, only the qualitative data for this study were included (Joanna Briggs Institute 2014 Manual, p. 19). Sample sizes in qualitative studies ranged from 7 to 73, while those in quantitative studies ranged from 33 to 162. Mixed methods studies ranged from 19 to 130 participants. Table 1 presents details of all 24 studies.

Race/ethnicity of parent participants. In 13 of the 24 studies reviewed, the majority (> 60%) of parent participants were White (Armentrout, 2009; Arutyunyan et al., 2018; Bluebond-Langner et al., 2007; Boss et al., 2008; Brosig et al., 2007; Carroll et al., 2012; Hexem et al., 2011; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Prows & McCain, 1997; Robinson et al., 2006; Tamburro et al., 2011). One study reported 45% White participants, but the race of 40% of participants was unknown (Hinds et al., 2009). One study noted that the majority of participants (>60%) were African American (Sharman et al., 2005). Another study reported the percentage of White (38%), African American (33%), Afro-Caribbean (13%) and Other (15%) participants who made a decision to withdraw life-sustaining treatment for their child for a subset of total participants, yet the overall racial breakdown of participants was not reported (R. Roy et al., 2004). Neither race nor ethnicity of participants was reported in the remaining eight studies (Coughlin et al., 2007; Einarsdottir, 2009; Ellinger & Rempel, 2010; Hileli et al., 2014; Kirschbaum, 1996; Peng et al., 2012; Pepper et al., 2012; Schimmel et al., 2016); these studies took place in Israel, Canada, or Taiwan.

Settings where decision making occurred. Fifteen of 24 studies (63%) included in the review took place in the U.S. (Armentrout, 2009; Arutyunyan et al., 2018; Boss et al., 2008; Brosig et al., 2007; Carroll et al., 2012; Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Prows & McCain, 1997; Robinson et al., 2006; Sharman et al., 2005; Tamburro et al., 2011). The remaining nine studies (38%) took place in Canada (Coughlin et al., 2007; Ellinger & Rempel, 2010; Pepper et al., 2012), Iceland (Einarsdottir, 2009), Israel (Hileli et al., 2014; Schimmel et al., 2016), the U.K. (R. Roy et al., 2004), Taiwan (Peng et al., 2012), and one in both the U.K. and the U.S. (Bluebond-Langner et al., 2007). Decision making by parents in the reviewed studies occurred in four major clinical settings: the perinatal/neonatal intensive care setting (Armentrout, 2009; Boss et al., 2008; Brosig et al., 2007; Coughlin et al., 2007; Einarsdottir, 2009; Ellinger & Rempel, 2010; Kirschbaum, 1996; Peng et al., 2012; Pepper et al., 2012; R. Roy et al., 2004; Schimmel et al., 2016); pediatric intensive care (Arutyunyan et al., 2018; Brosig et al., 2007; Kirschbaum, 1996; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Robinson et al., 2006; Sharman et al., 2005); hematology/oncology/bone marrow transplant unit (Bluebond-Langner et al., 2007; Hileli et al., 2014; Hinds et al., 2009; Kirschbaum, 1996; Prows & McCain, 1997); and the inpatient palliative care setting (Carroll et al., 2012; Hexem et al., 2011; Tamburro et al., 2011). Table 2 lists specific decisions made in each setting.

Study Outcomes

Findings from the data of the 24 studies were grouped according to three main themes. More than one area of focus was captured in several studies (n=14). The three

themes related to the influence of religion and spirituality on medical decision making among parents of critically ill children derived from the reviewed articles include: 1) Religion and spirituality used as guidance during decision making, 2) Religion and spirituality as forms of comfort and support during the decision making process, and 3) Religion and spirituality as a source of meaning, purpose, and connectedness in the experience of decision making.

Decision Making Outcomes

Religion and spirituality as guidance during decision making.

Sixteen articles examined parents' reliance upon their religion and spirituality for guidance when making decisions for their critically ill child (Arutyunyan et al., 2018; Boss et al., 2008; Coughlin et al., 2007; Einarsdottir, 2009; Ellinger & Rempel, 2010; Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Peng et al., 2012; Pepper et al., 2012; R. Roy et al., 2004; Sharman et al., 2005; Tamburro et al., 2011). These religious and spiritual influences included those from religious communities, spiritual leaders, higher powers, health care professionals, rituals, spiritism, and religious teachings (Arutyunyan et al., 2018; Einarsdottir, 2009; Hexem et al., 2011; Madrigal et al., 2016). The percentage of parents who have reported the influence of their religious and/or spiritual beliefs on their child's medical treatment or end-of-life decision making is between 31% (Meyer et al., 2002) and 49% (Arutyunyan et al., 2018). Moreover, parents who classified themselves as moderately or very religious or spiritual were more likely to be influenced by their religious or spiritual values when making decisions for their critically ill child (Arutyunyan et al., 2018).

Guidance with life-sustaining treatment or end-of-life care decisions. The majority of studies reviewed reported a reliance on a specific religious affiliation (Coughlin et al., 2007; Peng et al., 2012; R. Roy et al., 2004) or religious belief (Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Tamburro et al., 2011) for guidance in decisions related to life-sustaining treatments or end-of-life care. The first study describing the influence of religious affiliation on decision making was by Coughlin et al. (2007). In this study, parents who declined or limited resuscitation for their extremely premature infant, whose death was imminent despite treatment, had a higher proportion of mothers identifying as Catholic (63%) in comparison to those non-Catholic (28%) parents who opted to continue treatment despite imminent death (Coughlin et al., 2007). However, families in which the mother was non-Catholic (63%) were more likely to withdraw life-sustaining treatment based on a poor long-term prognosis compared to families where the mother was Catholic (17%). In the second study by Roy (2004), parents who were religiously affiliated with the Orthodox Jewish faith, a religion opposed to stopping treatment that sustains life, reported that their religious beliefs were the primary reason for refusing life-limiting treatment (R. Roy et al., 2004). Although the decisions under consideration were related to withdrawal of life-sustaining treatment, the specific religious beliefs associated with those religious affiliations that guided these decisions were not detailed. In the third study by Peng et al., 2012, parents were predominantly Buddhist and religious beliefs pertaining to life and death issues influenced parents' planned timing of the discontinuation of artificial ventilation for their child, and subsequent death (Peng et al., 2012). In this study, beliefs about reincarnation, specifically how one's situation at death can influence stages in

reincarnation, may have contributed to the parents' preference for when the child's life would end.

Four studies explored the influence of parents' religious beliefs on their decision making related to life-sustaining treatments or end of life (Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Tamburro et al., 2011). In a study by Hexem (2011), one parent reported that religious teachings on pediatric-palliative-care-related ordinary versus extraordinary measures, especially those related to end-of-life care, guided their decision making choices. Such religious teachings also helped parents think through the decision making process, rather than try to plan or control events (Hexem et al., 2011). In the Einarsdottir (2009) study, many parents reported a reliance on Spiritism, a belief that communication between the spirits of the deceased and those still living are transmitted through a medium (Einarsdottir, 2009). This medium is described as an individual capable of contacting and communicating with deceased spirits. In this study, Spiritism in the form of information communicated from the spiritual world through dreams or mediums, led parents making life-sustaining treatment withdrawal decisions about their extremely low birth weight (< 1,000 grams) infant to conclude that whatever outcome occurred was their infant's destiny. Parents also believed that mediums had the capacity to mobilize help outside of the care delivered by the health care team. For example, mediums were asked to seek the help of physicians in the "other world" for the purposes of aiding surgeons in a planned operation. Parents in this study also reported that faith in God or a superior power guided decision making for their infant, but no further explanation about the role of faith was provided (Einarsdottir, 2009).

The final two studies, focused on influences of religious beliefs on parents'

decision making related to life-sustaining treatments or the end of life, examined parental religion or spirituality as reasons that parents selected a particular life-sustaining treatment over another (Kirschbaum, 1996; Tamburro et al., 2011). The first study by Tamburro (2011) described the importance of limiting life-sustaining treatment among parents of children who identified religion as important or not important to the family unit. Families making resuscitation decisions for their child with a complex medical condition referred to palliative care, and who identified religion as important, had a higher likelihood of opting for limitations in life-sustaining support, compared to parents who reported that religion was not important (Tamburro et al., 2011). In the second study, Kirschbaum (1996) described parents with a "spiritual view of their existence" who believed they were "called" to remove life-sustaining treatment for their ill child (Kirschbaum, 1996).

Religion as guide to being a passive or active decision maker. Religion influenced parents' role as a passive versus active decision maker. In five studies reviewed, parents clearly conceptualized their religious beliefs as trust in a higher power or God; parents also reported their belief that God had ultimate authority over the child's health outcome or situation (Boss et al., 2008; Ellinger & Rempel, 2010; Hexem et al., 2011; Pepper et al., 2012; Sharman et al., 2005). In two of these studies, parents were passive decision makers, believing that since their child's health outcome was under God's control, there were either no decisions to make or they had no role in decision making (Boss et al., 2008; Pepper et al., 2012). Instead, parents deferred to physicians, asking them to do "everything they could," when making treatment decisions for their child (Boss et al., 2008). In two other studies, parents were active decision makers, believing that although

their child's health outcome was controlled by God, their role was to be actively involved (Ellinger & Rempel, 2010; Sharman et al., 2005), making decisions that would preserve or extend the life of their child (Ellinger & Rempel, 2010). In the fifth study, parents were generally active in decision making, although they sought the help of God in making more difficult decisions for their child (Hexem et al., 2011).

Religion and spirituality guides shared decision making between parents and the health care team. In addition to being a direct guiding influence to parental decision making, parents believed it important to share their religious or spiritual beliefs with the health care team, particularly physicians (Arutyunyan et al., 2018; Hinds et al., 2009). In one study, 46% of parents reported that sharing their religious affiliation or spiritual beliefs related to faith, meaning, and peace with providers could facilitate common ground for shared decision making (Arutyunyan et al., 2018). Seventy-two percent of parents who reported the influence of religious affiliation or spiritual beliefs on their decision making also reported that they would welcome discussions about their beliefs by their child's physician (Arutyunyan et al., 2018). In fact, parents were more likely to disclose their religious or spiritual beliefs if asked by the physician (Arutyunyan et al., 2018). One-third of parents expressed a desire for their child's physician to ask about their religious or spiritual beliefs, and this number increased to 50% when the child was seriously ill (Arutyunyan et al., 2018). Even more (62%) parents felt comfortable sharing religious or spiritual beliefs when interacting with a chaplain (Arutyunyan et al., 2018). In a second study, parents reportedly wanted the clinical team to show an awareness of and respect for religious beliefs and incorporate them as health care decisions were made (Hinds et al., 2009).

Guidance with prognostic decision making. In one study, parents deferred to their religious and spiritual beliefs, which were not specified, for guidance with decision making related to their child's prognosis, over information provided by the health care team (Boss et al., 2008). For these parents, maintaining hope that all would be fine drove their decision making (Boss et al., 2008). For instance, parents who were given a poor prognosis for their infant reported choosing or being encouraged by friends and family to pray and trust for a miracle rather than depend solely on medical information provided. In this same study, another family's religious beliefs guided their decision to transfer their young child to a hospital they believed capable of miracles (Boss et al., 2008). In a second study, parents' religious or spiritual beliefs were more important to their end-of-life decision making than advice provided by hospital staff or family or friends (Meyer et al., 2002).

No guidance. In three studies reviewed, religion was not an influence on decision making during life-sustaining treatments or end-of-life care (Bluebond-Langner et al., 2007; Hileli et al., 2014; Schimmel et al., 2016). In the first study by Bluebond-Langer (2007), parents of children with incurable cancer did not differ by religious affiliation when considering cancer-directed vs. symptom directed interventions. Regardless of religious affiliation, parents perceived that their continual involvement in decision making related to cancer treatment and supportive care was part of, and a priority to, their parental role as decision maker, care taker, and advocate (Bluebond-Langner et al., 2007). In a second study by Hileli (2014), parents of children with incurable cancer were not influenced by specific religious doctrine when considering end-of-life decisions. Although parents affiliated with the Druze religion, a religion characterized by

determinism (i.e. a belief that one's destiny is predetermined by God and reincarnation) consented to a Do Not Resuscitate (DNR) order less frequently than parents with a Jewish, Islamic, or Christian affiliation, the finding was not statistically significant (Hileli et al., 2014). In the third study by Schimmel (2016), parents' decision making for full medical care vs. comfort care for their infant born at 23 weeks gestation was not significantly influenced by religious affiliation; no differences were found among parents who identified as Jewish, Muslim, or Catholic (Schimmel et al., 2016).

Religion and Spirituality as forms of comfort and support during the decision making process

Religion and spirituality as comfort.

Twelve of the reviewed studies highlighted ways in which parents drew upon their religious or spiritual beliefs to find comfort during decision making for a critical illness or death of a child (Armentrout, 2009; Brosig et al., 2007; Carroll et al., 2012; Einarsdottir, 2009; Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Meyer et al., 2006; Peng et al., 2012; Pepper et al., 2012; Prows & McCain, 1997; Robinson et al., 2006). Comfort was derived through parents' connectedness to God (Armentrout, 2009; Brosig et al., 2007; Carroll et al., 2012; Kirschbaum, 1996; Meyer et al., 2006; Pepper et al., 2012; Robinson et al., 2006); prayer (Carroll et al., 2012; Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Meyer et al., 2006; Peng et al., 2012; Prows & McCain, 1997; Robinson et al., 2006); religious names given to their child (Einarsdottir, 2009); Bible reading (Hexem et al., 2011); journal writing (Hexem et al., 2011; Kirschbaum, 1996); counsel from religious personnel (Meyer et al., 2006); support from faith communities (Meyer et al., 2006); meaningful religious artifacts (Peng et al., 2012) and

belief in an afterlife (Armentrout, 2009; Hexem et al., 2011; Kirschbaum, 1996; Robinson et al., 2006).

Comfort achieved through a connectedness with God. Nine studies described how parents' close and personal relationship with God gave them relief from helplessness, provided peace and strength, and even permission to cease life-sustaining treatments during their child's illness (Armentrout, 2009; Brosig et al., 2007; Carroll et al., 2012; Einarsdottir, 2009; Hexem et al., 2011; Hinds et al., 2009; Meyer et al., 2006; Peng et al., 2012; Pepper et al., 2012). Parental connection with God was particularly comforting during the course of end-of-life decision making, when some parents returned to previously established religious beliefs, which filled a void in their life (Brosig et al., 2007). On the other hand, parents who had not previously believed religion important, found comfort during end-of-life decision making through religious practices, such as prayer (Einarsdottir, 2009).

Comfort through hopefulness. Some parents characterized the comfort they experienced as "hope." While the meaning of hope was not clearly articulated in every study (Robinson et al., 2006), parents generally defined hope as the desire for a miraculous cure or recovery for their child (Armentrout, 2009; Boss et al., 2008; Carroll et al., 2012; Einarsdottir, 2009; Hinds et al., 2009). Parents perceived hopefulness that their child could survive when the health care team communicated emotion, in the form of compassion (Boss et al., 2008). Parents experienced a lack of hopefulness when health care team members failed to give any good news (Pepper et al., 2012) or lacked an emotional response to parental grief (Boss et al., 2008). Parents reported hope as

something that could be present or absent; if present, parents could have it in varying amounts, (Carroll et al., 2012).

Parents also described hope in being a "good" parent and doing everything to ensure the child's health and safety (Pepper et al., 2012). For other parents, hope was derived through learning that a medical treatment was available to their child (Ellinger & Rempel, 2010; Prows & McCain, 1997). While some parents focused on hope to aid their decision making (Pepper et al., 2012), other parents rejected the notion of hope, defined as their child's return to health, feeling hope was too passive (Hexem et al., 2011). These parents' preference to be more active in the decision making process and their child's treatment led them to reject the notion of hope (Hexem et al., 2011).

Comfort through prayer. Prayer was another commonly used religious practice that provided parents comfort during and following decision making and resulted in an increased connectedness to God (Carroll et al., 2012; Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Meyer et al., 2006; Peng et al., 2012; Prows & McCain, 1997; Robinson et al., 2006). Whether prayer was used occasionally or more frequently, practiced alone or with a group, this religious practice provided parents a source of peace and the opportunity for reflection during the decision making process (Carroll et al., 2012; Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Peng et al., 2012). In the studies reviewed, the content of parental prayers was not disclosed; however, parents that prayed for strength, help or guidance, or the miraculous recovery of their child found praying for God's help comforting (Kirschbaum, 1996; Meyer et al., 2006; Robinson et al., 2006). During the final days of a child's life, parents' prayers were a comforting religious practice and reduced parents' pain, anxiety, and overall suffering (Peng et al.,

2012; Robinson et al., 2006). After the death of a child, prayer filled a void created by grief, and served to facilitate a close relationship among family members (Brosig et al., 2007). Prayer was a religious practice that parents could use in any location, not just a religious institution, so was readily available to parents (Hexem et al., 2011).

Comfort through religious rituals, texts, music, and artifacts. In some studies, parents attributed their infant's recovery to religious or spiritual rituals performed by themselves or others; these rituals eased parents' distress and brought comfort. Einarsdottir (2009) notes that several parents attributed the sudden recovery of their infant to the child's baptism (Einarsdottir, 2009). Another mother in this study expressed belief that a name with religious origin, which she bestowed on her infant, may have been responsible for the improvement of her infant's respiratory status and her own contentment.

Christian Bible stories that referenced the trials of Job and Abraham were helpful to parents and reminders that God could bring them through a difficult experience (Hexem et al., 2011). Parents also turned to religious music or religious artifacts (i.e. Buddhist charms) for comfort at the end of their child's life (Peng et al., 2012).

Parents who turned to religion for comfort were generally accepting of their child's health outcome after decision making, even if the outcome resulted in death or disability (Brosig et al., 2007; Hexem et al., 2011; Robinson et al., 2006). While parents may have been sorrowful about such outcomes, relying on religious beliefs and practices, like praying (Brosig et al., 2007; Hexem et al., 2011) and reading the Bible (Hexem et al., 2011), eased their emotional distress and provided a positive element to what would otherwise have been an entirely negative experience (Hexem et al., 2011).

Lack of comfort experienced as spiritual distress. Not all parents experienced comfort provided through their religion or spirituality during the decision making process. In five studies, parents expressed spiritual distress or disconnection from their spirituality during the course of their child's critical illness or at the time of the child's death (Armentrout, 2009; Einarsdottir, 2009; Hexem et al., 2011; Meyer et al., 2006; Robinson et al., 2006). During their child's illness, parents questioned God (Armentrout, 2009; Hexem et al., 2011), felt deceived by faith (Robinson et al., 2006), or believed that the child's illness might be punishment for their own or their child's moral wrongdoing (Hexem et al., 2011; Kirschbaum, 1996). Parents also declined participation in religious practices such as baptism, believing this to be a surrender to the child's imminent death, which was not comforting (Einarsdottir, 2009). After a child's death, parents continued to believe that God existed (Armentrout, 2009; Hexem et al., 2011); however, parents were angry with God and questioned God's omnipotence (all-knowing) (Armentrout, 2009; Hexem et al., 2011), given that their child did not survive. At the same time, some parents kept attending church because they felt a sense of solidarity with God, who also lost His Son [Jesus] (Armentrout, 2009).

Support through religion and spirituality. Twelve studies discussed parental use of religion or spirituality as sources of support during the decision making process (Armentrout, 2009; Arutyunyan et al., 2018; Boss et al., 2008; Brosig et al., 2007; Hexem et al., 2011; Hinds et al., 2009; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Pepper et al., 2012; Robinson et al., 2006; Sharman et al., 2005). Religious and spiritual support was received through chaplains, clergy, or fellow church members; God or Higher Power; health care practitioners (Arutyunyan et al., 2018; Boss et al., 2008;

Brosig et al., 2007; Hexem et al., 2011; Madrigal et al., 2016). Alternatively, parents described offering religious and spiritual support to family members of other critically ill children (Armentrout, 2009; Prows & McCain, 1997; Robinson et al., 2006).

Chaplains, clergy, and fellow church members as sources of support. In seven studies, chaplains, clergy or fellow church members were sources of support (Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Madrigal et al., 2016; Meyer et al., 2002; Meyer et al., 2006; Robinson et al., 2006). For example, in the study by Meyer (2002), of the 58% of parents who reported having the support of their minister, priest, or rabbi when making decisions about removal of life support, 78% found their support to be helpful (Meyer et al., 2002). In two studies, clergy and fellow church members offered prayers, which felt supportive (Hexem et al., 2011; Robinson et al., 2006). Clergy offered counsel and engaged in helpful discussions with parents about Bible texts or formal church teachings that eased parental burden both during and after decision making(Meyer et al., 2006). Biblical texts and church teachings were particularly helpful in supporting parental decisions to remove life-sustaining treatment or not resuscitate their child (Hexem et al., 2011; Robinson et al., 2006). Chaplains performed family-requested religious rituals, such as baptism, and provided a supportive presence, particularly at the time of a child's death (Brosig et al., 2007). Parents who relied on more sources of spiritual support identified their church community and clergy as key sources of this support and, African American parents were more likely to identify more sources of spiritual support compared to White parents (Madrigal et al., 2016). Two studies described the importance of faith and hope as a source of emotional support during palliative care or end-of-life decision making (Meyer et al., 2006; Robinson et al., 2006).

Support from a higher power or God. Some parents reported that support received was directly from God (Hexem et al., 2011). In particular, when parents relinquished their concerns to God, their sense of isolation was decreased (Hexem et al., 2011).

Support from healthcare practitioners. Parents valued support or encouragement from healthcare practitioners that their child could survive (Boss et al., 2008; Pepper et al., 2012). On the other hand, when physicians communicated to parents what was interpreted as negative information, parents felt unsupported and advocated on their child's behalf in ways that were in contrast to these healthcare practitioners' decision preferences (Boss et al., 2008).

In one study, parents felt support when physicians prayed for their child, who was at the end of life (Peng et al., 2012). In another study, 75% of parents welcomed the support of physicians' personal prayers for their child; however, fewer parents (38%) wanted physicians to pray with them (Arutyunyan et al., 2018), and even fewer parents (13%) would request physicians to initiate prayer (Arutyunyan et al., 2018).

In three studies, parents described the support provided by nurses, who were key healthcare practitioners (Brosig et al., 2007; Meyer et al., 2002; Robinson et al., 2006). In one of these studies, nurses provided crucial spiritual support by referring parents to a chaplain at their child's end of life (Robinson et al., 2006). In another study, parents ranked nurses (64%), over family (54%), physicians (44%), friends (23%), and ancillary staff (20%), as most frequently present at the child's time of death following life-support withdrawal (Meyer et al., 2002).

Other supportive health care team members included palliative care providers and child life specialists. Palliative care providers offered support through problem-solving

assistance to parents who were going through a difficult time (Brosig et al., 2007). Child life specialists offered critical support to siblings of ill patients. This support was evidenced through answering questions about the impact of the death of a child on siblings, which resulted in a more positive grieving experience for parents involved in end-of-life decision making (Brosig et al., 2007).

Support to other families. Spirituality and a connectedness to others was a rationale given by parents for offering support to parents of other children with a similar illness experience (Armentrout, 2009; Prows & McCain, 1997; Robinson et al., 2006). In two studies, parents who wanted ultimate good to come out of their child's death (Armentrout, 2009), chose to use their end-of-life decision making experience to develop more compassion and subsequently support other parents going through the same decision (Armentrout, 2009; Robinson et al., 2006). In the Armentrout study, one parent started a ministry within her own church, which provided support to other parents whose children had died (Armentrout, 2009). In the Robinson study, parents offered their support to other families, encouraging a reliance on the belief in life after death, that a deceased child was in heaven and one day, the parent would see their child again, pain-free and happy (Robinson et al., 2006). In a third study by Prows, parents used what was learned from their illness experiences with bone marrow transplant to provide support to other parents faced with similar decisions (Prows & McCain, 1997).

Religion and spirituality as a source of meaning, purpose, and connectedness in the experience of decision making

In nine studies, researchers reported that religion and spirituality enabled parents seeking to find meaning, purpose, and a sense of connectedness in the decision making

experience of their critically ill child(Armentrout, 2009; Brosig et al., 2007; Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Meyer et al., 2002; Meyer et al., 2006; Prows & McCain, 1997; Robinson et al., 2006). Meaning and purpose were evidenced when parents accepted the child's existence, however brief, as serving a purpose. One study that explored parents' end-of-life decision making found that for 73% of parents, religious and spiritual beliefs were important to parents finding meaning during their child's acute hospitalization when decision making occurred, and also after their child's death (Meyer et al., 2002). In other studies, parents identified with the concept of goodness in their spirituality, which dignified the child's existence, and was tied to belief in an afterlife for their child (Armentrout, 2009; Hexem et al., 2011). Although parents would rather have their child alive, they were able to see dignity or goodness in the child's death and to view the life of their deceased child in a positive manner (Armentrout, 2009; Hexem et al., 2011). In other studies, parents found meaning in their decision making experiences with supernatural explanations around the child's illness or healing, a sense that "everything happens for a reason," or a sense that the child's illness served a specific purpose in the parent's lives (Einarsdottir, 2009; Hexem et al., 2011; Kirschbaum, 1996; Prows & McCain, 1997).

Spiritually, the child's illness contributed to parents' and families' connectedness to God. In six studies, parents seeking meaning through their spirituality or religious beliefs expressed a connectedness to God and others during the decision making experience (Armentrout, 2009; Brosig et al., 2007; Hexem et al., 2011; Kirschbaum, 1996; Prows & McCain, 1997; Robinson et al., 2006). For example, a belief in God, faith, and prayer served to strengthen family relationships or to provide a watchful presence

over siblings of the deceased child (Armentrout, 2009; Brosig et al., 2007). In these studies, parents were able to identify lessons in their decision-making experience, such as developing greater compassion for others and serving as an example to other parents experiencing similar events (Armentrout, 2009; Brosig et al., 2007; Hexem et al., 2011; Kirschbaum, 1996; Prows & McCain, 1997).

Even parents who experienced anger and questioning around their child's illness were able to overcome their emotions to make difficult end-of-life decisions and subsequently view their child's death in a positive manner. For example, parents viewed their child's death as having purpose (Armentrout, 2009), felt their child would have normal functioning (Hexem et al., 2011), be in a happier place (Hexem et al., 2011; Kirschbaum, 1996; Robinson et al., 2006), and free from suffering in life after death (Kirschbaum, 1996; Meyer et al., 2006; Robinson et al., 2006). Parents also felt they could maintain connectedness with their deceased child and see their child again in an afterlife (Armentrout, 2009; Hexem et al., 2011; Kirschbaum, 1996; Robinson et al., 2006).

At the same time, parents questioned long-held religious and spiritual beliefs after the decision-making experience and upon the death of their ill child. In one study, parents described a new religious reality, one where they maintained their belief in God but the child's death led to an altered view (Armentrout, 2009). Specifically, these parents no longer identified with an omnipotent (i.e. all-knowing) Higher Power because they felt such a God wouldn't allow a child to die (Armentrout, 2009).

Discussion

This integrative review was undertaken to examine and appraise the current evidence of parents' religion and spirituality and its influence on decision making for a critically ill child. A detailed review of 24 studies revealed that religion and/or spirituality are generally perceived by parents across critical care pediatric settings to be positive influences when making intensive care, life-sustaining, or end of life decisions. This review highlighted three analytic themes that summarize the influence of parents' religion and/or spirituality on their decision making, including: 1) Religion and spirituality used as guidance during decision making, 2) Religion and spirituality as forms of comfort and support during the decision making process, and 3) Religion and spirituality as a source of meaning, purpose, and connectedness in the experience of decision making.

Our finding that parents' religious and spiritual beliefs are important in guiding their decision- making process is consistent with other perinatal and pediatric studies (Keenan, Doron, & Seyda, 2005; Kharrat et al., 2017). Our review reports both the percentage of parents who rely on religious beliefs during decision making, and also the types of religious or spiritual beliefs that parents use to guide their decision making. The majority of studies reviewed discussed parents' reliance upon a specific religious affiliation or religious belief to guide decision making; however, how such affiliations or beliefs guided parents' decision making was not always explicit. One interesting finding in a study by Coughlin et al. was the different approach to life-sustaining treatment by families with a Catholic vs. non-Catholic mother. Families with Catholic mothers tended to decline or limit resuscitation for their extremely premature infant, more than families

of non-Catholic mothers, when death was imminent (Coughlin et al., 2007). This study also reported that families with non-Catholic mothers were more likely to withdraw life-sustaining treatment based on a poor long-term prognosis compared to families where the mother was Catholic. A predominant belief in Catholicism is the understanding that human life is sacred (United States Conference of Catholic Bishops., 2018). Therefore, the finding that families with Catholic mothers were less likely to withdraw life-sustaining treatment based on poor long-term prognosis may not be surprising. However, in other research, religion has been used to rationalize continuing treatment at the end of life, citing belief in a miraculous recovery, a view that the health care team is an instrument of God for healing, or mistrust in the healthcare team (Mansfield, Mitchell, & King, 2002; Moseley et al., 2004; Moseley, Freed, Bullard, & Goold, 2007; Rushton & Russell, 1996; Sulmasy, 2006; Wagner & Higdon, 1996). Therefore, in the Coughlin study, it's possible that families with Catholic mothers accepted that their infant's death, despite treatment, was imminent and turned to their religion for meaning and comfort as death approached, rather than as a rationale for further medical interventions. Findings from another study by Einarsdottir, which describe parents' reliance on Spiritism, may lead some providers to feel discomfort and to equate such beliefs with supernatural forces that are opposed to religious doctrine associated with more established religious traditions (Einarsdottir, 2009). However, as health providers, our practice is in settings that are increasingly diverse; therefore, it's important to understand that such beliefs exist and may be influential for some parents in end-of-life decision making.

Findings from this review also suggest the specific contexts in which parents may draw upon their religious and/or spiritual beliefs, including during life-sustaining, end-of-

life, or prognostic decision making; during shared decision making with the health care team; and when considering their level of involvement in decision making. Although religious or spiritual beliefs are likely not important in guiding all parents' decision making, two-thirds of the reviewed articles reported religious and/or spiritual beliefs as important to parents. As our findings show, some parents defer to their religious or spiritual beliefs over medical information when making decisions (Boss et al., 2008; Meyer et al., 2002). Other parents feel that sharing their religious beliefs with providers could, in fact, provide common ground for decision making about their critically ill child (Arutyunyan et al., 2018). Therefore, these findings are instructive for providers who seek to incorporate parents' religious or spiritual values into specific health-related decisions.

Half of the reviewed studies discussed parents' use of religion or spirituality to find comfort during decision making. Parents found comfort through aspects such as their personal connectedness to God, hopefulness, prayer, and religious rituals, texts, music, or artifacts (Armentrout, 2009; Boss et al., 2008; Brosig et al., 2007; Carroll et al., 2012; Einarsdottir, 2009; Ellinger & Rempel, 2010; Hexem et al., 2011; Hinds et al., 2009; Kirschbaum, 1996; Meyer et al., 2006; Peng et al., 2012; Pepper et al., 2012; Prows & McCain, 1997; Robinson et al., 2006). Many parents draw upon established religious or spiritual beliefs during the course of their child's critical illness, while other parents who had not previously identified religion or spirituality as important, newly turn to religious or spiritual beliefs expressed through prayer, for comfort (Einarsdottir, 2009). Given that the reviewed articles did not discuss the content of parental prayers, further studies that explore what types of prayers comfort parents during decision making, and how these

prayers can be supported, are warranted. Interestingly, some parents characterized the comfort they experienced as "hope," not only hope in the child's recovery, but hope that they were being a "good" parent or ensuring the child's health and safety (Hinds et al., 2009; Pepper et al., 2012). Hope has been reported in other pediatric studies and is often associated with parents' wish for a miracle, or desire to provide maximal medical treatments with the goal of cure (Granek et al., 2013; Green, 2015). Our review may highlight the multidimensional nature of some parents' hope, specifically in the context of critical care decision making, and guide providers in this context in focusing parents' hope on their parental role, especially at the end of a child's life, instead of on treatments that may not achieve recovery or the child's survival.

Similarly, parents' felt supported in their religion and/or spirituality by chaplains, clergy, and fellow church members; A Higher Being or God; and healthcare practitioners. A significant finding in our review is the supportive role of nurses, as perceived by parents, at the end of a child's life; some parents ranked nurses as the persons most frequently present at a child's bedside at the time of death (Brosig et al., 2007; Meyer et al., 2002; Robinson et al., 2006). Other studies have cited the role of nurses in advocating for parents making decisions and facilitating a positive parent-child relationship at the end of life (Butler, Hall, & Copnell, 2018; Hinds et al., 2001). These findings may have implications in terms of involving nurses as assessors of parents' spiritual needs, especially in the context of end-of-life cases, which aligns with the Hospice and Palliative Nurses' Association (HPNA) position paper on the value of the processional nurse in palliative care, part of which is to provide spiritual support (Hospice and Palliative Nurses Association, 2015). Additionally, parents cited spirituality and connectedness to

others as a rationale for offering support to parents of other children facing similar health care decisions (Armentrout, 2009; Prows & McCain, 1997; Robinson et al., 2006). This finding confirms what other studies have found, which is that pediatric health care experiences, while they can be traumatic for parents, may also spur positive growth and facilitate greater personal strength and recognition of new possibilities in life (Picoraro, Womer, Kazak, & Feudtner, 2014), such as providing support to others in distress.

Our finding that parents looked to religion and/or spirituality to find meaning, purpose, and connectedness during decision making is consistent with other studies. One related study, which explored the spirituality of parents of children with cancer who had a poor prognosis, found that spirituality helped parents generate meaning in the midst of uncertainty and current or anticipated loss and grief (Nicholas et al., 2017). Our finding that parents derive meaning through religious or spiritual beliefs may be particularly noteworthy in that meaning-making drew parents closer to family and others, rather than creating feelings of disconnectedness and isolation. In previous studies, parents have reported feeling isolated due to the demands of decision making in critical care environments (Carter & Leuthner, 2002). Methods for reducing parents' isolation have been suggested, including use of good communication, transparency and shared determination during decision making, advocacy and support groups, and family resources centers (Carter & Leuthner, 2002). Our findings add to these interventions by suggesting that facilitating parents' religious or spiritual beliefs can be an additional means for promoting parents' well-being and deterring isolation during their critical care decision making process.

Suggestions for Future Research

The field of religion, spirituality and health research is young and developing (Koenig & Bussing, 2010). In considering the role of religion and spirituality in decision making, future researchers should clearly distinguish the concepts of religion and spirituality, since religion and spirituality are two different concepts with differing influences on health outcomes (Lucchetti, Koenig, Pinsky, Laranjeira, & Vallada, 2015). Additionally, research involving the concepts of religion, spirituality, and decision making should ideally be conducted longitudinally in order to capture how these variables change for parents over time, throughout the course of the child's critical illness. As more becomes known about the relationship between religion, spirituality, and decision making, studies should explore who may best assess parents' religion and spirituality, and which interventions might facilitate the role of religious and/or spiritual factors in specific health decisions. Specifically, our review highlights the potential role that nurses may play in performing general spiritual assessment, particularly in end-of-life cases, since once death of a child is imminent, the role of providing comfort care traditionally rests with nursing staff (Meyer et al., 2002). Our review also highlights the comforting nature of prayer for parents, but suggests a need for further investigation into the types of prayers that are most comforting and how these prayers can be supported.

In general, recruitment of parents in decision-making research can be difficult, because some parents, as mentioned in this review, don't view themselves as having made a decision for their critically ill child. Strategies for obtaining significant samples include expanding inclusion criteria to include parents who do not speak English,

conducting multi-center studies, and utilizing intensive case study or multiple case study research designs (Hill et al., 2014).

Limitations

We utilized a systematic, rigorous approach in undertaking this review. However, the possibility exists that we did not retrieve all relevant studies. Limitations of studies in general include the use of explorative and descriptive approaches, which do not add to statistical support for associations between religion, spirituality, and decision making, but do provide conceptual and parent-reported support. Furthermore, some studies did not have 'religion,' 'spirituality,' or 'decision making' as their primary focus, and therefore may not have discussed detailed information about these topics in their findings, impacting what could be reported.

Several methodological issues pertaining to the included studies are worth noting. Some studies had a retrospective component, increasing the risk of recall bias. The majority of studies included mostly White participants, and eight studies did not detail racial demographics of participants. While these racial demographics reflect the current state of pediatric decision-making evidence, the disproportionate racial make-up of participants in many studies may not accurately represent decision making by all parent groups. Different sampling techniques could increase racial diversity in future studies. Limitations for studies that had a qualitative component in this review include limited transferability, as many of the participants were drawn from clinically and/or geographically homogenous samples. Also, qualitative studies, in general, lacked credibility, given that researchers failed to mention when or if saturation was reached. Limitations for the quantitative studies in this review included the use of convenience

sampling, and some of the quantitative studies had small sample sizes, increasing the likelihood of low statistical power.

Despite these limitations, this integrative review reflects the current state of evidence for the influence of parents' religion/spirituality on decision making for their critically ill child. The integration of qualitative, quantitative and mixed methods data strengthens the review findings, while also highlighting additional areas of future research.

Conclusions

This is the first integrative review, of which we are aware, that provides a synthesis of the evidence about the influence of parents' religion and spirituality on decision making for a seriously ill child. We found that parents view religion and spirituality as a primarily positive and helpful influence when making decisions for their ill child and use religion and/or spirituality to help guide decision making, find comfort and support, and derive meaning, purpose, and connectedness from their decision making experience. In addition to identifying these outcomes reported in the literature, this review offers guidance for the future research agenda into how religion and spirituality contribute to decision making for parents in a stressful critical care environment.

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Figure 1: Literature Review and Flowchart of Included Studies

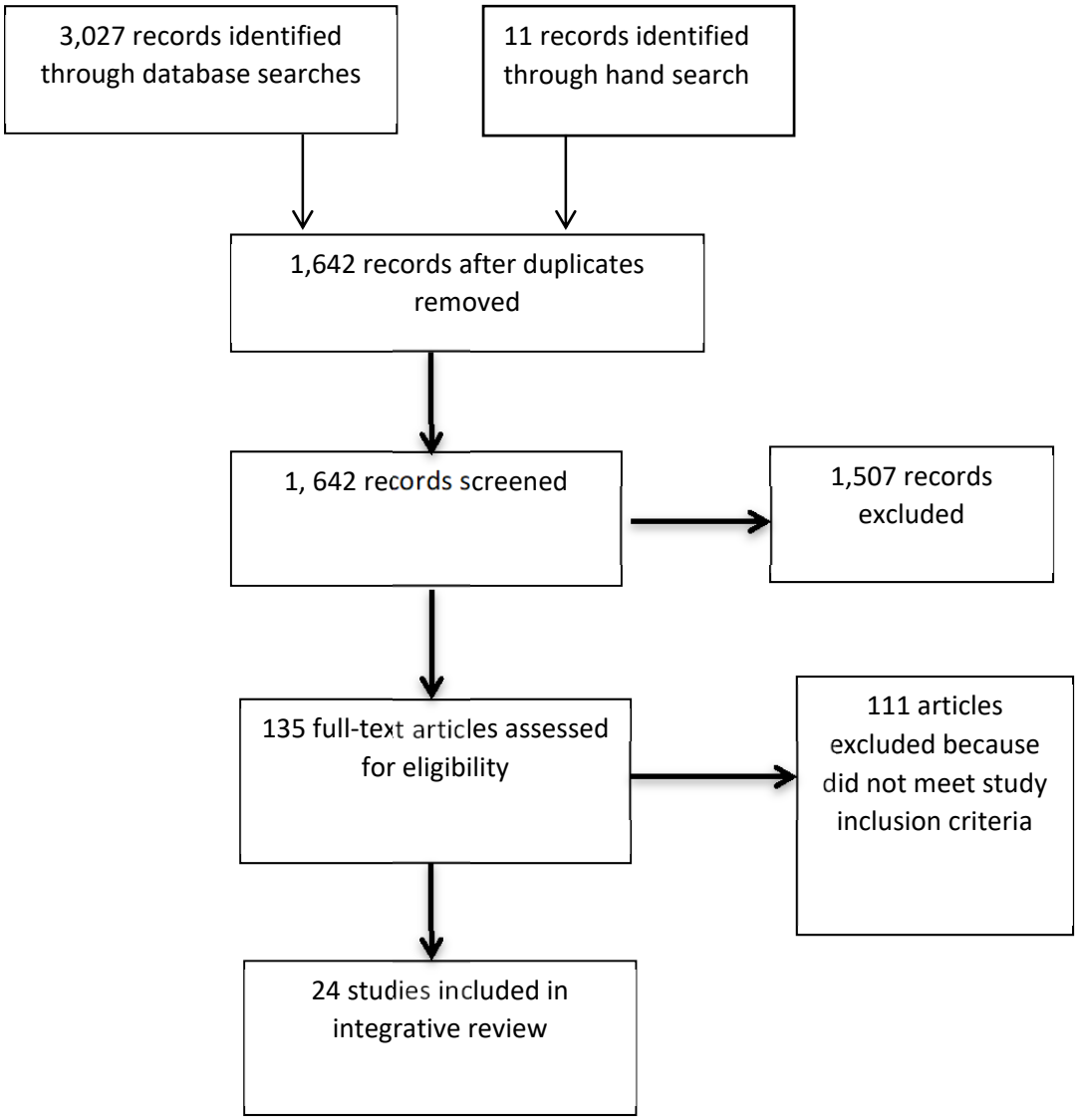

Table 1: Characteristics of Decision-making Studies Involving Religion and/or Spirituality

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
Qualitative				
Armentrout (2009) USA	Qualitative, Grounded theory Narrative interviews	NICU 15 parents, (4 couples & 7 mothers)	Removal of life support	-- Faith: a source of comfort following decision making; gave parents strength -- Despite feeling angry, some parents kept attending church; felt God understood b/c He also lost Son -- Belief in higher being was maintained, but not God of childhood -- Belief in afterlife: made decision making easier; brought parents hope of seeing child again -- Religion/spirituality: helped parents view life of child as positive; informed support of other parents making similar decisions; prompted use of experience for good -- Lessons: parents saw lessons in experience including being an example, having more compassion/ sensitivity, personal betterment
Bluebond -Langner (2007) USA & UK	Qualitative, ethnographic Observation; open-ended, semi-structured interviews; record review	Pediatric oncology Parents of 34 children, (17 USA, 17 UK)	Treatment versus supportive care after standard therapy has failed	--Religious background of child was reported: 14 Protestant, 6 Catholic, 8 unspecified, 5 Other --Parents of children with incurable cancer did not differ by religious affiliation when considering cancer-directed vs. symptom directed interventions --Regardless of religious affiliation, parents decision making was part of, and a priority to, their parental role as decision maker, care taker, and advocate
Boss (2008) USA	Qualitative Semi-structured interviews	Perinatal 26 mothers	Delivery room resuscitation	--Religion/ spirituality, & hope guided DM for most parents, regardless of medical information --Parents were encouraged by friends/family to pray for miracle --Some parents felt they made no decision; “left things in God’s hands” & asked physicians to do everything they could --Parents felt abandoned without hope and compassion of physicians
Carroll (2012)	Qualitative	Palliative Care	Range of pediatric	--Praying to God/knowing that there is a God gave parents peace & helped them deal with difficult decisions

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
USA	Semi-structured interviews	16 parents of 15 patients, (56% moms, 44% dads)	palliative care decisions	--Parents expressed hope that child would improve, quantified as having more or less of hope, expressed as something that could be present or absent, hope could be expressed in conflicting ways
Einarsdottir (2009) Iceland	Qualitative Open-ended, semi-structured interviews	NICU 29 parents, (28 mothers, 25 fathers)	Withdrawal of treatment decisions	--Knowledge about the infant's future & additional help was sought from various sources, such as mediums and dreams --Parents belief in Spiritism meant details of events known to the dead person and the supplicant were transmitted by a medium. -- Mediums were thought to mobilize help classified as supplementary to the help given by professionals in the NICU --Parents expressed their faith in God or some superior power. Individuals who normally did not consider themselves religious began praying occasionally or regularly --Parents, relatives, or friends of the family sent a request with the infant's name to prayer circles at churches --A few parents stated whatever happened to their infants was their destiny or course of nature. --Some parents hinted their infant's sudden recovery was related to emergency baptism; other parents declined to have their infant baptized because they interpreted it as a sign of surrender or an acceptance of imminent death --Parents felt well at child's baptism because of names chosen for child
Ellinger (2012) Canada	Qualitative In-depth interviews	Perinatal/ NICU Parents of 15 children (15 mothers, 10 fathers)	Norwood procedure	--Parents were willing to accept any outcome as long as they did everything they could to preserve the life of their child. --Some parents questioned the kind of life the child would have, but went through with Norwood & put child in God's hands; faith played a key role in these parents' decision making process

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
Hexem (2011) USA	Qualitative Open-ended interviews	Palliative Care 73 parents	Range of pediatric palliative care decisions	--Parent reported decisions were less difficult when they knew or accepted God/God's will. --Parents associated their religions with the statement "everything happens for a reason, but weren't always happy about it. --Parents associated religion/spirituality with goodness; it dignified their child's existence & specialness & was tied to parent's belief in an afterlife for their child --Some parents wanted to know church teachings on decisions; these teachings gave parents patience & the ability to think things through, rather than plan/ control --Parents read the Bible in response to stressful life events --Religion/spirituality provided support, peace, comfort and moral guidance --Church community provided a network and source of unconditional love/ support --Religion helped parents deal with anger, gave them guidance on their parental role --Parents who held to faith did not think this was incompatible with other emotions
Hinds (2009) USA	Qualitative Semi-structured interviews	Peds oncology 62 parents (of 58 patients)	Noncurative treatment decision (DNR, initiate terminal care, enroll in phase I trial)	--When making decisions, parents wanted clinicians to know about & respect their religious beliefs/practices --Prayer, faith, access to clergy & belief the parent-child relationship endures beyond death were themes
Kirschbaum (1996) USA	Qualitative, phenomenology Open-ended, semi-structured	PICU, NICU, peds oncology 39 parents of 20 children (20 mothers, 19 fathers)	Life-sustaining treatment	--Some parents had supernatural explanations for why child had become ill or died --Some parents thought they had done something wrong to deserve child's illness --Some parents were conscious their child's illness had been part of a larger plan & a higher power was in control & making good come from it -- Parents spoke of hoping for, praying for, or expecting a miracle --Parents felt medical know-how was God-given --Parents who felt strongly about the spiritual nature of their experience also described being at peace after having accepted their loss --Parents turned to reflection through prayer and writing in journals for strength, guidance and a source of hope. Prayer was also a way to find community

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
				--Parents spoke of the support of chaplains, pastors, spiritual counselors or their church community --The experience of their child's illness or death, strengthened parents' faith --Parents saw others' faith life affected by their situation as well. --Parents were comforted their child would go to a happier place (afterlife) and that they'd one day be reunited. Hope, faith and connectedness with their child figured importantly into their decision making
Meyer (2006) USA	Qualitative Open-ended questionnaires	PICU 56 parents, (36 mothers, 20 fathers)	Withdrawal of life support	--Faith was central to parents' efforts in making meaning of their child's situation, to provide guidance and permission around EOL decision making and to cope better --Faith helped sustain some parents and offered comfort in the act of praying for God's help and guidance, seeking counsel from religious personnel, and receiving social and emotional support from faith communities --Parents emphasized faith's positive aspects; some acknowledged spiritual distress
Peng (2012) Taiwan	Qualitative Retrospective chart review	NICU 50 charts of NICU patients	Resuscitation (DNR); removal of ventilator	-- Religion (Buddhism) was an important aspect for families of dying infants -- Some religious beliefs caused parents to choose timing of child's death --Some families played Buddhist music to their dying child; prayed with the physician for the dying infant; put a good luck charm inside the incubator
Pepper (2012) Canada	Qualitative Semi-structured interviews	NICU 7 parents (2 couples, 3 mothers)	Treatment for extremely premature infants	--Some parents reported letting God decide (after having initial consult with neonatologist) -- Some couples felt helpless after birth of premature infant and found comfort connecting with God. Parents placed perilous decisions in God's hands --One couple focused on the value of hope to aid their decision making & identified reluctance of the health care team in providing hope --Parents defined hope as hoping child would live; also being a "good mother" & hoping she was doing everything to ensure her son's health and safety
Prows (1997) USA	Qualitative In-depth interviews	Bone Marrow Transplant (BMT) center	BMT vs. natural death	--In BMT decision making, parents searched for information and spiritual meaning -- Parents going through the BMT decision made them more able to support others going through the same decision -- Some parents reported prayer was helpful

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
		10 parents (6 mothers, 4 fathers) of 7 children		
Robinson (2006) USA	Qualitative Open-ended questionnaire	PICU 56 parents, (36 mothers, 20 fathers)	EOL decision making	--Comfort was found through personal faith, belief in God, prayer, belief in afterlife --Religion helped parents accept child's health outcome --Parents characterized the comfort they felt as hope -- Some parents expressed spiritual distress or disconnect from their spirituality, or felt deceived by faith -- Chaplains, clergy or fellow church members were sources of support—clergy & fellow church members offered prayers, which felt supportive -- Biblical texts and church teachings were particularly helpful in supporting parental decisions to remove life-sustaining treatment or not resuscitate their child --Faith and hope were sources of emotional support during EOL DM --Spirituality was a rational for offering other families support; parents encourage other families to rely on belief in afterlife & that parent would see their child again
Sharman (2005) USA	Qualitative In-depth, semi-structured interviews	PICU 14 parents of 10 children, (64% mothers, 36% fathers)	Limit or withdraw life support	--Parents relied on a higher power for strength, wisdom, divine intervention, and a final answer as to when death would occur --Parents felt God/belief in God and sometimes prayer helped them through --God was described as an anchor for some parents; some felt God had the final decision-making authority.
Quantitative				
Arutyunyan (2016) USA	Quantitative Cross-sectional survey	PICU 162 Parents (81%	Medical treatment	-- 49% of parents said religious/spiritual beliefs influence child's care decisions -- 46% of parents agreed sharing religious/spiritual views may provide common ground for medical decision making -- Parents who regarded selves as moderately/ very religious/spiritual were more likely to state their decisions for their child were influenced by their beliefs

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
		mothers, 15% fathers)		-- Parents whose beliefs influenced decisions were also more likely to welcome the physician's inquiry into their beliefs (72%)
Hileli (2014) Israel	Quantitative Chart review	Peds Oncology, Hematology 73 charts	DNR order	--Positive responses to a DNR request were approximately equal among Jews, Muslims, Christians --Consent to a DNR was significantly lower among Druze families, but not statistically significant --Those with higher educational level and higher monthly income gave their assent to DNR more often
Madrigal (2016) USA	Quantitative, prospective cohort study Survey	PICU 86 parents of 75 children, (60 mothers, 26 fathers)	The most difficult medical decision regarding the care of the child (a decision already made (past), one presently being made, one that is foreseeable or unlikely (future))	--Parents with more spiritual forms of support ranked church community ($p<0.001$), spiritual leader($p<0.001$), higher power ($p<0.001$), prayer ($p<0.001$), spouses ($p=0.002$), support group ($p=0.003$), and what my child wants ($p=0.023$) as higher than the parents with less forms of spiritual support --African American participants were more likely to have more spiritual forms of support (n=10/12, 83%) and Whites fewer (n=32/59, 54%, $p=0.032$)
Meyer (2002) USA	Quantitative Survey	PICU 56 parents, (36 mothers, 20 fathers)	EOL (i.e. Foregoing life-sustaining treatment)	--58% of parents reported that their ministers, priests, rabbis were available to them before their child's death --Those with access to religious support persons found them to be helpful 78% of the time --73% parents reported their religious/spiritual beliefs were important/very important during child's hospitalization & remained so after death

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
Roy (2004) UK	Quantitative Retrospective chart review	NICU Analysis of 39 charts	Withdrawal/ withholding of Life-sustaining treatment, DNR	--Decisions to withdraw life-sustaining treatment were determined by parents' religious beliefs --Of 11 sets of parents who refused withdrawal of LST, 3 (including 2 orthodox Jewish families) gave religion as the primary reason --Nearly all white parents, but only 54% of Black African parents agreed to withdrawal of LST, with religious and personal beliefs being a factor
Schimmel (2016) Israel	Quantitative Retrospective chart review	Perinatal 26 families of 33 infants	Full medical care vs. comfort care for infants born at 23 weeks gestation	--No differences in approach determined parental attitudes regarding the option to choose full medical care vs. comfort care for babies at the edge of viability among Jewish, Muslim, or Catholic, religious or secular couples
Mixed Methods				
Brosig (2007) USA	Mixed methods Survey, semi-structured interviews	Palliative care 19 Surrogates (11 mothers/ fathers, 7 mothers, 1 aunt)	EOL	--All families reported finding comfort in their beliefs in God --Religion provided sense of meaning for child's child --Deceased child brought family together/ watched over other children
Coughlin (2007) Canada	Mixed methods Chart review, thematic analysis of documented multidisciplinary team discussions	NICU Families of 130 infants	Resuscitation, withdrawal of treatment	--Of 44 patients (out of 130) in whom resuscitation was declined or limited, there was a higher proportion of families documenting maternal religion as Catholic than non-Catholic (63% vs. 28%) --The overall rate of treatment withdrawal did not differ statistically between families in which maternal religion was identified as Catholic compared with non-Catholic (29% vs 48%). --Families in which the mother self-identified as non-Catholic were more likely to withdraw based on the prediction of poor long-term outcomes than those families in which the mother self-identified as Catholic (63% vs. 17%)

Author (Year) Country	Research Design & Methods	Setting & Sample	Parent Decision	Summary of Religion/ Spirituality & Decision Making findings
Tamburro (2011) USA	Mixed methods Goals of care discussion, chart review	Palliative Care 50 patients and their families	Pediatric palliative care, resuscitation decisions	--38% of patients/ families that acknowledged that spirituality was important to them opted for some limitation of support compared with only 22% of families that expressed that spirituality was not important to them

CHAPTER THREE: Manuscript Two

NICU Decision Making for Mothers of Infants with a Congenital Anomaly

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Abstract

Objective: To explore the characteristics of medical treatment decision making for mothers of infants receiving treatment for a congenital anomaly in the neonatal intensive care unit (NICU). This article will report a subset of the qualitative findings from a larger mixed-methods study.

Study Design: Using semi-structured qualitative interview methods, we explored 20 mothers' decision-making for their infant with a congenital anomaly who was receiving treatment in an urban, academic-center NICU. Interviews were uploaded into NVivo 11 and coded using a qualitative content analysis approach. Mothers' themes were grouped based on the timing (prenatal vs. postnatal) of diagnosis of the infant's congenital anomaly.

Results: The primary themes that emerged from these data were decisional features (i.e. types of decisions, appropriateness of decisions), decisional involvement, and NICU decision-making processes. The decision that mothers most commonly faced in the NICU was surgery and many mothers describe the importance of making a decision that was appropriate for their infant. In terms of decision involvement, mothers who received a prenatal diagnosis and those who received a postnatal diagnosis for their infant were involved in decision making, but at different times with different manifestations of involvement based on timing of diagnosis. In terms of NICU decision making processes, mothers who received a prenatal versus postnatal diagnosis for their infant identified similar barriers and facilitators to their decision making process.

Conclusions: Understanding mothers' NICU decision making for their infant with a congenital anomaly may help guide clinicians' care of families and their use of pediatric-specific skills and tools to promote effective shared decision making in the neonatal setting.

Keywords: Neonatal Intensive Care, congenital anomalies, parent decision making

Introduction

Worldwide, congenital anomalies account for over 3.2 million birth defect related disabilities and a quarter million deaths each year (World Health Organization, 2016). In the U.S. alone, congenital anomalies impact one in 33 infants and are the number one cause of infant death (CDC, National Center for Health Statistics, 2017). Due to the association of congenital anomalies with illness severity, disability, and increased risk of death, many infants born with a congenital anomaly require neonatal intensive care (NICU) hospitalization immediately after birth and subsequent decision making by parents (Peller, Westgate, & Holmes, 2004).

In the NICU, shared decision making between parents and providers is recommended when determining which treatments are best for infants with congenital anomalies. However, not only is it difficult for NICU providers to accurately predict outcomes for these infants (Meadow et al., 2012), but involvement of parents in decision making within the NICU can be challenging, and can vary based on factors, such as parents' demographic characteristics, coping processes, and parenting values. For example, young parent age and low educational attainment have been demonstrated to limit parents' cognitive processing or be associated with less active involvement in decision making (Boss et al., 2010; Kraetschmer et al., 2004; Street et al., 2005; Tarini et al., 2007). Additionally, parents with low levels of coping may be unable to cognitively process information about their infant's congenital anomaly, which can limit a parent's ability to be part of decision making (J. Lalor et al., 2009; J. G. Lalor et al., 2008). Furthermore, parents in previous studies have been shown to value different aspects of

being a ‘good parent’ when making difficult decisions for their ill child (Feudtner et al., 2015; Hinds et al., 2009; October et al., 2014).

Other studies offer guidance on which groups of parents of children with a congenital anomaly require further study. Several studies indicate that parents with prenatal foreknowledge of a congenital anomaly experience increased burden and grief, compared to parents who learn of their infant’s congenital anomaly after birth. Studies also indicate that mothers of NICU infants have more difficulty than fathers adapting to a NICU admission, resulting in more long-term mood disturbances, lower levels of long-term attachment with their child, and potential child behavior problems and child cognitive deficits later in life (Affleck et al., 1991; Doering et al., 1999; Doering et al., 2000; Pinelli, 2000). However, how the burden and grief that can accompany the diagnosis of a congenital anomaly is compounded by the stress of a NICU admission, especially when treatment decision making is necessary, remains unexplored. Therefore, the purpose of this study was to investigate the decision making of mothers involved in treatment decisions for their infant with a congenital anomaly receiving care in the neonatal intensive care unit (NICU).

Theoretical Framework

The Roy Adaptation Model (RAM) was used as the theoretical framework for this study (see Figure 1). According to this model, adaptation is both a process and an outcome. As a process, adaptation is an individual’s continual response to environmental stressors (focal stimuli) using coping processes and adaptive modes (such as their role function) with the goal of adapting to their environment. Adaptation as an outcome is an

individual's positive response or lack of positive response in a situation (adaptation response or outcome). In this study, the RAM provides a model to explore the process of how mothers, when faced with the congenital diagnosis of their infant and a NICU hospitalization, use coping processes (general coping, religious coping, and illness perception) and adaptive modes (depressive symptoms and their role as a parent) to respond by engaging in decision making for their infant who requires treatment in the intensive care setting (C. Roy, 2009).

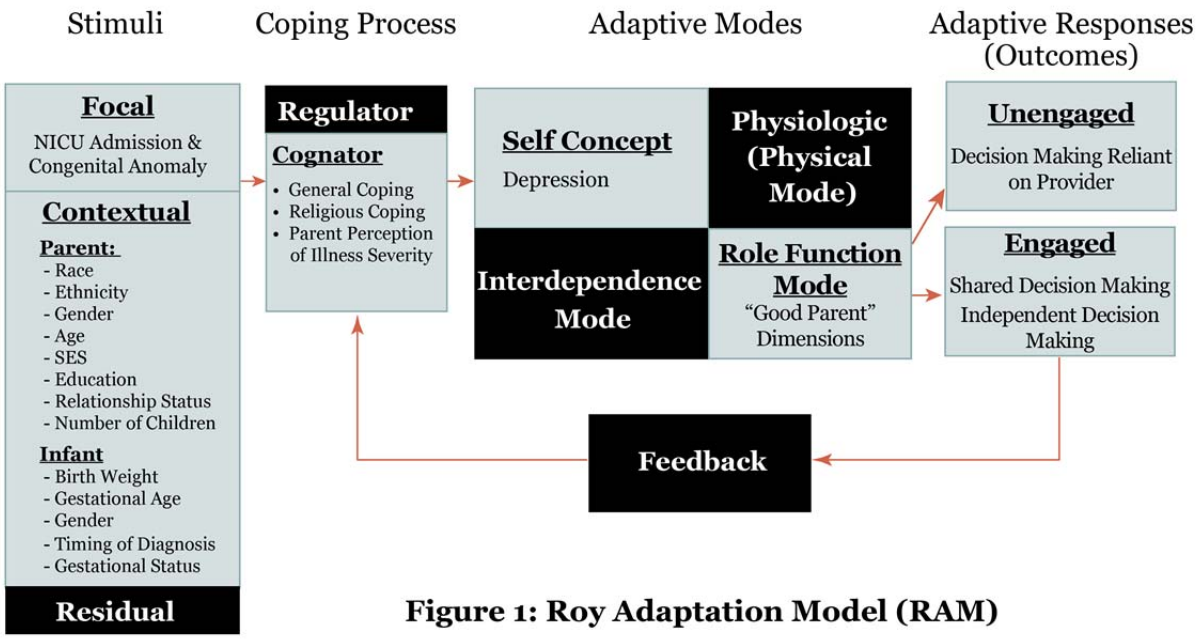

Figure 1: Roy Adaptation Model (RAM)

Design and Methods

Study Design

This article describes a subset of the qualitative findings from a larger, mixed-methods NICU study, in which mother participants completed a quantitative survey and qualitative semi-structured interview. The qualitative interviews aimed to further explain

and clarify the coping, parenting, and decision-making processes of mothers of infants with a congenital anomaly in the NICU. This paper presents the qualitative findings on mothers' decision making.

Sample

This study was conducted in a large urban neonatal intensive care unit, Johns Hopkins Children's Center NICU (JHNICU) in Baltimore, Maryland. The JHNICU is a 45-bed level IIIc NICU with 710-730 annual admissions; approximately 135-155 infants with severe congenital anomalies are admitted each year; 60% of these infants are Caucasian, 25% African American, and 15% other races. The JHNICU is the only facility in the state that offers pediatric cardiac surgery, neurosurgery, and general surgery.

The institutional review board of Johns Hopkins Medicine (Baltimore, Maryland) approved this study. Participants were recruited from the neonatal intensive care unit. Mother participants were eligible if they were 17 or older, biologically related to or had primary caretaking responsibility of an infant 0-6 months of age hospitalized in the NICU, had an infant with a heart, lung, brain/nervous system, diaphragm, renal, musculoskeletal, digestive, or chromosomal anomaly, had made or deferred a treatment decision as identified through a Decision Checklist (a study-team constructed checklist with common NICU decisions, from which mothers could identify an important decision they had been part of), and had the ability to speak and write English. Exclusion criteria included surrogates representing Child Protective Services (CPS), as decision-making processes of these surrogates may be limited by CPS regulations. Mother participants gave consent to include their infant's relevant medical record data pertinent to the study.

Data Collection and Procedures

Sampling. To reduce the risk of participant burden, we only approached mothers after the fourth day of their infant's NICU stay. This timing of approach was chosen based on prior interviews with NICU parents, which revealed that parents prefer a few days of adjustment to the NICU environment before participating in research (Hinds and colleagues, correspondence). After providing consent, mother participants completed a quantitative survey consisting of a demographics questionnaire and coping, parenting, and decision-making variables. A subset of mothers was purposefully selected to participate in the qualitative interviews based on the timing of when they learned of their infant's congenital diagnosis (prenatal vs. postnatal). We aimed to recruit ~50% of the sample from mothers who learned of their infant's congenital diagnosis before birth and ~50% from mothers who learned of their infant's congenital diagnosis after birth, to capture a range of views about decision making in the NICU and provide a thorough understanding of facilitators and barriers to coping, parenting, and decision making. Based on prior research, it was anticipated that at least 12-20 participants total needed to be interviewed before data redundancy was achieved (Hinds et al., 2009; Johnson & Christensen, 2008; October et al., 2014). We report a subset of the qualitative findings in this article.

Procedures. Interviews were conducted by MKU in a private room on the NICU or by phone. Before beginning the interview, the investigator advised the mother that she could stop the interview at any time, if desired. In addition, NICU social workers were on call, in case the mother became upset and required additional support or follow-up. To give context to interview questions, the investigator reviewed with the mother the most

important decision she had made (or deferred) for her infant using the Decision Checklist. On the Decision Checklist, mothers were asked to select all decisions they had made, and then circle the most important decision. The most important decision was the focus of qualitative discussion. A semi-structured interview guide was developed by the research team based on variables of coping, parenting, and decision making selected through the Roy Adaptation Model. This interview guide was used during interviews to elicit mothers' decision-making perceptions (see Appendix B). Additional probing questions were used to further understand a mother's perceptions of barriers and facilitators related to decision making. Most mothers were interviewed alone; however, in the case of four mothers, their decision partner (husband or significant other) was present and expressed a wish to join the interview.

During the interview, the investigator took notes and memos to add contextual information. As a form of member checking, the investigator summarized interview responses with mothers prior to ending the interview, and amended content as necessary. Interviews were audiotaped and transcribed prior to analysis. Personal identifiers were removed from the transcribed verbatim interviews, after checking for accuracy against the original audiofile (DiSantis et al., 2013). Each interview lasted 30-60 minutes. At the completion of the interview, each participant received a $15 gift card for her time/effort. The research team met on a bi-weekly basis to monitor and discuss qualitative data collection.

Data Analysis

We utilized a qualitative content analysis approach to analyze interview data. Qualitative content analysis is utilized when describing a particular phenomenon (coping,

parenting, and decision making, in this case), and when research literature on the study phenomena is limited (Hsieh & Shannon, 2005). Interview transcripts were reviewed prior to coding to verify accuracy. Interviews were uploaded into NVivo 11.0 (QSR International Melbourne, Australia), a qualitative software analysis package. A consensus method between the first author and senior author was used to develop a priori codes from the qualitative interview guide, which was developed from the main concepts of the Roy Adaptation Model. These a priori codes were collected into a codebook, which was used to code subsequent interviews. Steps in the qualitative analysis included: (1) reading the interview transcripts 2-3 times to gain a general understanding of the content, (2) coding passages as either a facilitator (defined as any person, resource, or attribute that helps a parent) or barrier (defined as any person, resource or attribute that deters or prevents a parent), (3) coding passages into barrier and facilitator categories, including access, behavioral, cognitive, communication, emotional, environmental, illness perception, instrumental, relational, and spiritual, and (4) examining each factor to identify themes. The first three steps of the qualitative analysis process were conducted deductively by MKU and MW. In the fourth step of analysis, an inductive approach was utilized. The first author developed the primary themes, which were discussed with the senior author (MTN). The transferability (applicability) of study findings was aided through the use of purposive maximum variation sampling. Multiple coders were employed to increase reliability (Creswell & Plano Clark, 2011). Credibility of data was increased through the presentation of representative quotes with the main study themes. Finally, an audit trail was used to ensure the analysis process was acceptable and the findings and interpretations were supported by the data.

Results

A total of 20 mothers (and 4 of their decision partners) participated in interviews. Twelve mothers received a prenatal congenital diagnosis and eight mothers a postnatal diagnosis for their infant. Table 1 summarizes demographics of mother participants and Table 2 summarizes demographic data extracted from infant participants' charts. Participants were predominantly White, well educated, middle income mothers. Infants were predominantly full-term, female with a digestive congenital anomaly. Three main themes emerged from interview data: Decisional Features, Decisional Involvement, and NICU Decision Making Processes. These themes and related subthemes are described below.

Decisional Features

Types of decisions. Mothers in our sample chose an important decision from the Decision Checklist: surgery (n=13), medication administration (n=3), withdrawal or withholding of life-sustaining treatment (n=2), blood transfusion (n=1), and radiology testing (n=1). While mothers faced different decisions, they consistently described the types of decisions they were making as "big" and "life altering." Descriptions of decisions are depicted below.

Surgery. Two-thirds of mothers described being part of a major surgical decision for their infant with a congenital anomaly. For most mothers, surgery was seen as the next logical step, rather than an actual decision. Mothers described surgery as a "no brainer," or a decision that they did not hesitate to make or agree to because not performing surgery typically meant their infant's outcome would be death. Mothers derived comfort while making surgical decisions in the reputation of the facility where

they had sought care and the expertise of surgeons, who had vast, specialized experience. Most mothers felt prepared to make a surgical decision for their infant once admitted to the NICU because their infant had received a prenatal congenital diagnosis and mothers had been prepared by surgeons beforehand about what type of surgical decision to expect.

> Um, like, we pretty much knew, like, she couldn't eat. Cuz her intestines weren't working. So, like, if she didn't have surgery, like, that would be it. I felt like it wasn't, like, elective, like, oh, you could maybe do this now, or do it when she's six months old or something. It was like, well, if she doesn't have the surgery, she can't eat. I felt like that decision was pretty, like, you had to do it. Like, there was really no option. Um—cuz that was, like, a life decision for her, you know. I mean—Or death. yeah, it wasn't like, oh, let me fix your toe or something, you know. It was like you have to eat. So, I really didn't even consider it, like, a decision almost. It was just, like, yeah, we're doing it (Participant 1003-Postnatal).

Medications. Three mothers faced decisions about whether or not to treat their infant with an intermittent or continuous medication. Mothers opted for the administration of medication to their infant, since it was deemed the best treatment by the health care team, whom the mothers trusted, even though medication side effects were sometimes difficult to witness.

> I put trust in the doctors about the decision, "they're the experts; they've seen it more; surgery wasn't an option, this was next best thing (Participant 1021-Prenatal).

> Um—it—once I found or realized that her PDA was gonna potentially close, and that [medication] was pretty much the only thing that was keeping the oxygenated blood or flow to the lungs, it-it was kind of already determined for me, I guess. You know—it was—it was a necessity for her. Um—the quality of life and what was gonna make it easier for her. So it was—it was a no-brainer—but, at the same time, a-a concern because there's always side effects (Participant 1021-Prenatal).

Life-sustaining treatments. Two mothers in our sample received congenital diagnoses for their infant that were incompatible with life, so were facing withholding or withdrawal of life-sustaining treatment decisions. These mothers described feeling that

they didn't have a choice, rather their decision was chosen for them. Withdrawal of life-sustaining treatment decisions re-focused mothers to important people in their life and slowed down what mothers described as a busy life. Mothers deciding whether to withhold life-sustaining treatments based their decision on how much discomfort such treatments would cause their infant, and how successful these treatments would be in giving their infant a longer life.

> So once we got that information, realizing that, oh, so his EEG was-was not normal. And then the MRI was not normal. And then going through the different slides of the MRI and actually seeing what a normal brain should look like, versus what our child's brain looks like, that was very telling for us… I think that once-once we got that information, I was explaining to my husband, like, what this means, 'cause it's not about his initial diagnosis. It's the complications, or is what going—what is going to actually affect him the most. And to watch him go through that, like, why? I mean, I don't-I don't want to do that. Like, that would be painful to see revolving in and out of the hospitals, being—have to be in a place where, you know, our child would not be our child. We would have to sit here and have someone else do basic care for him, because we wouldn't be able to do those cares, because he would have to be in a place where they provide round-the-clock care (Participant 1018-Postnatal).

Blood and radiology procedures. Mothers making decisions about blood or radiology procedures were mainly concerned with potential side effects of these treatments for their infant. Mothers only wanted these treatments done if necessary, not just as a precautionary measure.

> To me, it's more like to receive blood, I feel like it has to be life or death type of thing. And just to do it just to do it didn't sit well with me. They were gonna do it just to boost up his count because of possible blood loss. He [the physician] didn't know how much blood he was gonna lose. He's tiny. It was a precautionary thing. And it was—that didn't sit well with me (Participant 1029-Prenatal).

Appropriateness of decision. Many mothers discussed the importance of making a decision that was "good" or "right" for their infant. Mothers determined goodness or rightness based on the success of the treatment, the number of unnecessary procedures or

unattractive alternatives that the treatment would help avoid, or a parental intuition or feeling that it was the best decision.

> Participant: So we relied on, a lot of our information relied on what they were telling us, like what we were reading online and stuff like that. And then what they were telling us. So we made sure it was the right decision for him.
>
> Interviewer: How did you know what the right decision was?
>
> Participant: I think it was just what felt right. I mean, because obviously, we don't know. Like we have no idea. I mean, we don't know, you know a hundred percent. We were really going with what felt right. Cuz that was all we had to go off of (Participant 1026-Prenatal).

Decisional involvement. Mothers in both the prenatal and postnatal group described being involved in decision making for their infant; however, the timing and type of involvement differed. Mothers in the prenatal group were more involved in decision making before their infant was born, choosing a hospital and/or surgeon who could address their infant's congenital anomaly treatment needs, and then relinquished decision making to the health care team upon their infant's NICU admission. While some mothers in the prenatal group described having "pre-made" a decision for their infant before birth, other mothers in the prenatal group described a "wait and see" approach, deciding, based on doctors recommendations, which decision was the best decision once the infant was born and the severity of the congenital anomaly could be evaluated. Even if the decision making for the mothers in the prenatal group was not certain at the time of their infant's delivery, mothers had engaged in multiple conversations with specialists such that at the time of delivery, mothers felt comfort with decisions that needed to be made.

> We already decided that this was the right team for the job, and the right facility for the job. Their knowledge is much higher than ours. If you trust someone, it's easy to follow their lead (Participant 1004-Prenatal).

> I chose this hospital from, you know, a lotta people say it's one of the best hospitals and stuff like that. On Google, for instance, if you search, um, "best hospital in the United States," this hospital comes up fourth. So uh, that—that was when I was like, "Oh, okay, good point." And, uh, people that I've met around, um, inside John Hopkins. My therapist had a baby that was in the NICU at John Hopkins. So she was like, "He'll be in great care there," –so I was like, "That's how I chose to bring him here." (Participant 1023-Prenatal)

Mothers in the postnatal group were also involved in decision making, but more often at the time of their infant's NICU admission. This group of mothers described a desire to be involved in decision making as part of their parental role. Overall, mothers in the postnatal group described decision making as being difficult, but were less likely to relinquish their decision-making role to the NICU health care team.

> I think we'd always want to be at least in— involved in the decision making process. Yeah, I don't—I wouldn't be able to let anybody else make the decision— just cuz one, that's not me, and two, I would, uh—yeah, I don't—I couldn't put that on somebody else when I know it's, you know, my child and, you know, you live with it, and that's—That's our little human. (Participant 1007-Postnatal)

Other mothers in the postnatal group expressed being glad for a postnatal diagnosis, which they viewed as more clear-cut than receiving a prenatal decision.

> Now, I will say had this been found in utero at a certain point in time, this decision probably would not be as clean for the two of us. I think it would have been much more difficult in terms of the discussion and the impact, and, um—Well, I think—- how we would reach a decision (Participant 1018-Postnatal).

Involvement of decision partners. Ninety percent of mothers were married or partnered and named their husband or significant other as their primary decision partner in the NICU. Significant others frequently acted as the primary decision maker for an infant during mothers' recovery from delivery, relaying information between the NICU health care team and mother until the mother was able to be present at the infant's bedside. Significant others also helped mothers stay calm during decision making,

offering reassurance about a chosen decision when mothers expressed doubt. Some mothers, despite having a decision partner, felt more decision responsibility, as the mother. Mothers infrequently depended on friends and family as decision partners, citing friend/family's unfamiliarity with the NICU and unfamiliarity with types of decisions proposed by the health care team. Decision making could feel isolating to some mothers.

> I think I definitely relied on my husband to kind of push me towards making a decision... Um, I didn't have time to kind of waffle like I usually would. I would go back and forth and change my mind a million times, but um—and then once you commit to something like that, once you're like, "Yeah, okay, let's do it," then it starts moving really fast and you can't pull back and say, "No, no, no, no, wait. I change my mind." He's [my husband] very, like he can make a decision and he's very, you know—he's very calm and reasonable and logical about it. So, he'll, you know, "Okay, well," h-his thought can't be saying, "Well, what else are we gonna do?" You know? "What else is gonna help her? "as much as this?" ⋯ Um, so it was good to have him to kind of be the rationale, like logical piece—to my emotional, nervous self (Participant 1011-Postnatal).

> With my other kids, like I've relied on um, you know, advice from my parents. I've relied on um, advice from like my husband's family. You know, and um, finding other moms to—that I could talk to and get advice from, and you know, just kind of vent stuff too. Like, you know, my kid ate a crayon today. Like um, but when you're making decisions here, it's I mean, you don't—their—with this situation, we had—we didn't have anybody that we actually knew. Um, you know, but it's kind of—you feel kind of, I don't want to say alone, but like, you have to dig harder to be able to find the- the um, connections that you want, in order to be able to make more informed decisions about, you know, what we're doing. (Participant 1026-prenatal)

NICU decision making process. When asked whether the decision-making process in the NICU was the same or different than in other parts of life, mothers were split in terms of their response. Approximately half of the sample, consisting of mothers from both the prenatal and postnatal group, perceived that NICU decision making resembled decision making in other parts of their life. These mothers perceived both NICU and life decision making to entail gathering information, considering options, weighing risks and benefits,

and sometimes asking friends or others who have gone through the same decision.

Several mothers, while indicating that decision making was the same in the NICU and in life, felt that NICU decision making was harder because it impacted their child's life.

> Interviewer: Do you think how you make decisions in the NICU, is that similar how you make decisions outside of the NICU?
> *Participant:* I feel like we would have all the information before we made a decision. I just feel like it's harder. But other than that, yeah. We would still be get—we would never make a decision unless we had all the— information and all the—it's just not— not us.
>
> *Interviewer:* So having information is really important to you no matter what.
>
> *Participant:* Knowing—yeah, knowing every step of the way everything that's gonna happen. That is our number one thing before even saying or doing something (Participant 1007-Postnatal)

The other half of the sample, also consisting of mothers from both the prenatal and postnatal group, perceived decision making was different in the NICU than in the rest of life. These mothers felt decision making in life involved choice between options, but NICU decision making lacked options. Other mothers described feeling isolated during NICU decision making and perceiving that "wrong" decision making could cost their child's life. Decisions in the NICU also seemed more time-sensitive with less room for deliberation and more pressure to make the right decision.

> *Interviewer*: Is your involvement in his surgery similar to how you approach different decisions you make in life, or do you feel like it is different?
> *Participant*: No, this is—it's—no, yeah. It's not really a decision you hafta—there's only one option. The other—the other option is a non-option. You can't let the child die. So it's-it's not really a decision…Because it's life and death. Do the surgery—do the surgery or he dies (Participant 1020-Postnatal)

NICU decision making barriers. Mothers in the prenatal and postnatal groups identified cognitive, communication, emotional, and relational decision making barriers. Cognitive, or knowledge-acquiring, decision making barriers included receiving

information from the NICU health care team about potential complications or negative side effects to proposed treatments.

> It's[surgery] a harder decision—Cuz it's—anything can go wrong—in that medical. Anything. You know, you sign the papers and just like they tell you, it's a one in such-and-such chance that she can die, but it- it can happen. You know. Anything can happen on a- on a surgery table, so you just pray and think that everything go well when she was in surgery. (Participant 1008--Prenatal)

Mothers in the postnatal group identified two additional cognitive decision making barriers, including lack of pertinent information to make a decision and reading "worst case" information on the internet.

> The internet makes it hard. You can, you know—We get—we're in the process of making this decision, so you can go online and read about anything. Um, good or bad. And the bad things that you read about and you hear about, um, make it very hard. (Participant 1011--Postnatal)

Mothers in the prenatal and postnatal groups also identified decision-making barriers related to communication, including receiving conflicting information from health care team members.

> And so, it was just a lot of mixed messages, and that's hard. Um, we would hear from the surgeon, you know, "Oh, three weeks from start to finish." And then we came and some of the nurses were like, "Oh, you'll be here at least a month." It's like, "Oh geez." [Laughter] So, when you're making decisions about—are we gonna go through with the surgery or not—and you're hearing, "Oh, it could be two weeks, could be two months that you're here," that's hard, um—to just not know. And I think a lot of our struggle was, we just didn't know. (Participant 1011-Postnatal)

Mothers in the prenatal group identified an additional communication barrier during decision making, which was receiving insufficient preparation for decisions that would need to be made for their infant after birth. Mothers in the postnatal group reported that they sometimes misunderstood the medical language used by the health care team.

Mothers in the prenatal and postnatal groups also identified emotional barriers to decision making, which included being uncertain of their infant's health outcome following a procedure and concern related to prior negative health care experiences that were similar to the current decision being made for their infant.

> My 11-year old daughter has had like a outpatient surgery. She had a hemangioma on her, like right inner lip. So when she was three they removed it. When she came out of anesthesia she was very combative, very scared, very—they say it's common for some children when they come-come to, but that was very scary for me. I was about 20, 21 maybe. To see her, um, come outta anesthesia like that. So maybe there— maybe that weighed a lot on my reservations for the anesthesia [for my infant]. (Participant 1022--Postnatal)

> Interviewer: Did you feel when you were making the decision for surgery there was anything that made it harder?
>
> Participant: The uncertainty was difficult. (Participant 1013-Prenatal)

Mothers in the prenatal and postnatal groups identified relational barriers to decision making, which included making decisions that opposed family members' preferences, having uncertainty about whether to trust doctors to recommend decision options, and physicians conveying a limited timetable to make decisions.

> Like, it's not like it's somethin' like, oh, it's a common cold type of issue. No, it's surgery, so, like, you have to kinda sort of trust what the doctor's saying at that point in time. You're putting all your faith and trust in them. You know, so it's hard. Cuz I don't trust a lotta people., not a lot, especially when it comes to my kids. It's like— Like I really have to trust [laughter] you in order to let you, like, even just take care of my kid for a few hours, you know? That's, like, really hard for me. So, um, yeah. It's just a lack of control cuz with that I guess I have to kinda sorta depend on the doctor, trust them that they're doing their thing and they're gonna do it okay and you know—(Participant 1029--Prenatal)

NICU decision making facilitators. Mothers in the prenatal and postnatal groups identified decision making facilitators pertaining to cognitive, emotional, and relational aspects. Mothers in the prenatal group also identified spiritual decision-making

facilitators.

Cognitive decision-making facilitators for mothers in the prenatal and postnatal groups included receiving information from the health care team about procedures planned for their infant and reading relevant, informative online information.

> I trusted the doctors here after I saw them the first couple of times cuz they knew what they were talking about. They gave me more information on the types of procedures that I could have done in regards to seeing if it could possibly be linked or if it could happen again. And it just made a lot of sense. It helped make it [i.e. coming to a decision] easier. Because at first I was a little skeptical in regards to all of it, knowing that not many people knew about it to begin with. And also knowing that it's more severe, well as they were saying, it was more severe. Um, I don't know. I think them giving me information made it easier because they knew they could do something to help her. And I guess that's all I wanted was for someone to say that they could help her. (Participant 1017--Prenatal).

> So we really—a lot of our information relied on what they were telling us, like what we were reading online and stuff like that. And then what they were telling us. So- so we made sure it was the right decision for him. (Participant 1026--Prenatal)

Mothers in the prenatal group also identified having foreknowledge of the decision and using their parental intuition as decision making facilitators.

> Um, as far as the decision for surgery, I think that it was premade before we got here—because we did understand um, what we were up against and had about 15 to 18 weeks to make the decision. Um, so we spent that time educating ourselves and researching the defect that we're up against. And decided on who was the best. And then after we made that decision, have that confidence level in that surgeon, you know, we were taking their lead. We already decided that this was the right team for the job, and the right facility for the job. So whatever they recommended, really, we just—our only decision was we're gonna go with whatever they recommend. (Participant 1004-Prenatal)

> That's been the hardest thing is just kinda trusting your instinct a little bit, that you're—It's-it's a new instinct if, you know—Especially she's our first, so—- to have this instinct about like just knowing, like feeling like you just know what you need to do or what decision you have to make. Um, so I think it's hard to trust that as a parent—- in—and when you're making a medical decision, but I think it's been really important for us to kind of get more in touch with that instinct and

> be willing to kind of let it like, “Okay, this is what we need to do based on how I feel—“and the information that we have,” (Participant 1011-Postnatal)

Mothers in the postnatal group identified receiving information from other NICU families and having information about treatment alternatives as facilitators to their decision making.

> Interviewer: Yeah. So you had a choice of either doing the jaw distraction surgery or letting her go to another hospital where they would do the trach? And you thought the trach was better?
>
> Participant: Right. She [i.e. my daughter who died] had to have, um, a breathing tube and it wouldn’t go all the way down to her stomach or somethin’, so they was thinkin’ about doing a trach and I don’t think it was- I don’t- I don’t like the trach. My son [i.e. who died] had the trach. I don’t like it. The other hospital just goin' give her a trach and they can do the surgery to make it better, so when they told me they can fix it in less than a week, I was okay. Somethin' goin' on instead of just lettin' her lay there with the stuff all in her. (Participant 1024-Postnatal)

Mothers also described that information directly from the surgeon carried more weight than if information was relayed through another health care team member.

> I mean, just when you think about that, like—in my mind, I just think of, like, a surgeon, like, probably knows more about her intestines than anyone else. Like, he’s gonna go in there and open him up. So, when he explained to me, I just felt like he really knew what he was talking about. That carried a lot of weight. Yeah, because he’s gonna be the one that, like, goes in there and sees them. He also gave me an update after surgery. Not to sound mean, but it was like I don’t want the information from, like—someone who was just in the room, watching him. Like, that—I wanted the information from him. (Participant 1003-Postnatal)

Mothers in the prenatal and postnatal groups identified emotional decision making facilitators, including having hope or certainty in their decision making process due to previous decision making experiences.

> Participant: I think it would only be right to really give her the best chance that she, you know—that we can give her without—just givin’ up hope. Cuz we done—we done pushed this far. Past ter—past termination and past, um, everything else. So.

Interviewer: Sounds like keeping her was a big decision, and now these are big decisions.

Participant: Yes. But we knew it would be a long road of decision-making. (Participant 1008--Prenatal)

Interviewer: Was there something inside you that helped you make that decision or how did you come to that decision? Mostly based on—

Participant: Right, based on the past experience. My son had the trach. I don't like it..

Interviewer: So you had that experience to sort of base—help make your decision?

Participant: Right. Yeah. To have the trach or I rather try somethin' different. (Participant 1024--Postnatal)

Mothers in the prenatal and postnatal groups identified relational facilitators to their decision making, including having trust in the health care teams' competence or the hospital's reputation and decisional support from family members.

Interviewer: It sounds like you—mostly you let the providers make that decision. Can you say a little more about that, what that was like?

Participant: Well, I think a lot of it had to do with like um, I mean being from so far away and like, you know, we originally were going to get treatment close to home. [We] started doing the research and kept seeing the same, names over and over. Within a week, you know, we were up here meeting with the [surgeons]. Meeting like, especially with this main surgeon, him having such a plan. The comfort level that they give you in their confidence—that's not an arrogance. It's just they've been there. (Participant 1004--Prenatal)

Interviewer: When you think about that decision, can you think of other things around trying to make that decision that were supportive for you in that time?

Participant: Just knowing that she's gonna get the best care at [this hospital]. I mean, [this hospital] is known to be one of the most amazing places for, um, NICU babies and just—you know, just knowing that they have that great rapport was—you know, I knew she was gonna be in good hands. (Participant 1014--Postnatal)

Finally, mothers in the prenatal group identified spiritual facilitators to their

decision making, which included prayer, trust in God, and receiving spiritual support from clergy.

> But I felt like once we—I had—what I had to keep telling myself, which got me through it, was we had prayed and asked God for clarity. The surgeons came in right after that, and we felt instantly okay with our decision, whereas before we were saying maybe we should just give in (Participant 1025--Prenatal)

> Participant: I'll probably call—discuss everything with my pastor. She hasn't been up there. Um, to the hospital yet. She doesn't have transportation, but um, well, people, they take her to different places, but she always calls and- and um—and asks about how he's doin', and she always prays—um, over the phone and prays uh, for him, well, on a daily basis, but she always calls and checks in on him to see how he is. She calls me. Of course, she can't call the hospital, but she calls, always calls me. (Participant 1027--Prenatal)

Discussion

This study explored the decision making of mothers of infants receiving treatment for a congenital anomaly in the NICU setting. The primary themes that emerged from these data were Decisional Features, Decisional Involvement, and NICU Decision Making Processes. In comparing decision making of mothers who had received a prenatal versus postnatal diagnosis for their infant, we found that mothers who received a prenatal diagnosis were often more actively involved in making decisions for their infant prior to delivery, choosing specific facilities and specialists to provide care, and frequently deferring treatment decision making to providers once their infant was admitted into the NICU. Conversely, mothers who received a postnatal diagnosis were more actively involved in treatment decision making in the NICU, infrequently relinquishing their decision-making role to the health care team.

While current recommendations from the American Academy of Pediatrics stress the need to involve parents of high-risk newborns in the shared decision making process,

the findings of this study may shed light on how involvement in treatment decision making may differ based on whether the diagnosis is made before or after birth (American Academy of Pediatrics Committee on, Newborn, & Bell, 2007). Previous studies have distinguished parents' active decision making from passive decision making and identified factors important to each decision-making role. Authors of these studies have described that active decision making involves parents' gathering of information, weighing of consequences, and considering alternatives. Conversely, these authors have identified what they term passive decision-making factors, which include parents' value of medical expertise and physician recommendations (Weiss, Bard, Cook, Black, & Joffe, 2016). Our study suggests that mothers who receive a prenatal congenital diagnosis may be more passively involved in decision making in the NICU setting, but may adopt an active decision-making role at other points along the decision-making timeline. For example, mothers in our study who received a prenatal congenital diagnosis for their infant were actively involved before delivery gathering information about facilities and providers who specialized in the care of their infant's condition. These mothers weighed the consequences of obtaining care closer to home versus uprooting their lives to receive care at a facility they felt would be ideal. Once mothers had considered alternatives and chosen a hospital and provider, they built trust with these providers by having multiple conversations where parental values and preferences were shared. When it came time to enact the important decision in the NICU, mothers engaged in a more passive fashion, entrusting providers to manage the nuances of the decisions, and even authorizing providers to do what they thought would be best if the decision-making plans required a change of course. These findings exemplify how it could be reasonable for a parent to

defer a treatment decision to NICU providers who have the expertise to manage the outcome of the treatment.

An interesting finding of this study is the limited decision-support role that family and friends played for mothers of infants with congenital anomalies in the NICU. Because of the rare nature of many congenital anomalies and the specialized treatments that infants with these diagnoses require, mothers' had difficulty drawing on the knowledge and experience of friends and family as they would in other decision making situations, because friends and family were largely unacquainted with the complexities of their infant's needs. Consequently, mothers who were partnered (90%) relied heavily on their significant other for decision support, as well as the health care team for information and recommendations pertaining to treatment options. These findings align with previous studies, which have also found that parents often assume the role of primary decision maker for their critically ill infant in the NICU and depend on nurses and physicians to share information so that they can participate in the decision making process (Currie et al., 2016). In the Currie study, providers' communication also seemed to lessen mothers' feelings of isolation during the decision making process. This finding is consistent with other studies, which show that the level of provider communication is decisive in parents' experience of inclusion in the NICU (Wigert, Blom, Bry, 2014). In our study, mothers reported that nurses were particularly crucial in providing emotional and instrumental support and fostering inclusion.

Our finding that mothers drew primarily upon their significant other as a decision partner during NICU treatment decision making indicates that providers should ask intentional questions to make sure they are involving the appropriate decision partner(s).

In addition, this study informs NICU providers' care of mothers of infants with congenital anomalies, offering data on what type of information would be beneficial in the neonatal period. Given that family and friends, due to lack of knowledge about or experience with congenital anomalies, are less likely to serve as a support to mothers of infants receiving treatment for a congenital anomaly, NICU providers should be aware that they may need to provide more decision making support to this population. Mothers who receive a prenatal or postnatal diagnosis benefit from receiving information from providers about the functioning of the neonatal health care team, as soon after NICU admission as possible. Providers who care for infants who are born at outlying hospitals, which serve as catchment areas for major, medical center NICUs where congenital anomaly treatment occurs, can provide information, potentially in the form of brochures, to mothers that offer information about treatment resources for specific congenital anomaly diagnoses. Training NICU health care personnel in how to offer various types of support, including information, emotional, and instrumental support, could decrease the risk of adverse psychological responses in mothers and maximize health outcomes for infants.

Mothers who received a prenatal or postnatal congenital diagnosis experienced similar barriers and facilitators during their NICU decision making. As other studies have noted, strategies that identify specific barriers to parents' decision making may be necessary to promote effective shared decision making in the neonatal setting (Boland, McIsaac, & Lawson, 2016). A parental shared decision-making preferences (PSDM) instrument being developed by researchers from the Children's Hospital of Philadelphia

holds great promise for assessing decision-making barriers and identifying facilitators for individual parents (Children's Hospital of Philadelphia Research Institute., 2018).

Limitations and Strengths

This study had some limitations. Mothers were recruited from a single NICU and the demographics of our sample, while representative of the population of mothers with infants diagnosed with a congenital anomaly at the recruitment institution, were mostly White, non-Hispanic, married, college-educated, and English-speaking mothers, which limits the representativeness of our findings. It's possible that other key concepts not discussed in interviews are important to other demographically diverse groups. In particular, this study focused on mothers' decision making, but the under-representation of fathers in research is an important area of future research. Future studies would also benefit from exploring decision making of culturally and ethnically diverse parents with different belief systems that may guide decision making, as many congenital anomalies are prevalent among minorities groups.

Despite these limitations, the study had several strengths. Many prior pediatric decision making studies explore parent decision making retrospectively, using interview data from conversations weeks, months, or years after their child's treatment. Our study was prospective, exploring mothers' decision making at the time it actually occurred in the NICU. We utilized a Decision Checklist, which allowed mothers an opportunity to specify specific decisions that were most important related to treatment decision making for their infant, and this checklist subsequently served as a reference point for other survey completion and the qualitative analysis.

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Table 1: Mother Participant Characteristics for the Qualitative Sample by Prenatal and Postnatal Group

Variable, N (%)	**Qualitative Sample (n=20)**	**Prenatal (n=12)**	**Postnatal (n=8)**	**p value**
Age (yrs), mean ± SD (range)	31.25 ±5.73 (19-40)	30.42±6.74 (19-40)	32.50±3.85 (26-38)	p=0.143[T]
Race				
White	14 (70.0)	10 (8330)	4 (50.0)	p= 0.188[+]
Black	5 (25.0)	2 (16.7)	3 (37.5)	
Other	1 (5.0)	0 (0.0)	1 (12.5)	
Ethnicity				p= 0.653[+]
Hispanic	2 (10.0)	1 (8.3)	1 (12.5)	
Non-Hispanic	18 (90.0)	11 (91.7)	7 (87.5)	
Education				p= 0.176[+]
High school grad or less	4 (20.0)	2 (16.7)	2 (25.0)	
College grad or less	10 (50.0)	8 (66.7)	2 (25.0)	
Some grad school	6 (30.0)	2 (16.7)	4 (50.0)	
Income				p= 0.591[+]
<$45,000	6 (30.0)	4 (33.3)	2 (25.0)	
>$45,000	13 (65.0)	8 (66.7)	5(62.5)	
Decline/don't know	1 (5.0)	0 (0.0)	1 (12.5)	
Employment				p= 0.368[+]
Full-time	11 (55.0)	5 (41.7)	6 (75.0)	
Part-time	4 (20.0)	3 (25.0)	1 (12.5)	
Unemployed/stay at home parent	5 (25.0)	4 (33.3)	1 (12.5)	
Health Insurance				p= 0.650[+]
Private	10 (50.0)	5 (41.7)	5 (62.5)	
Medicaid/Other	10 (50.0)	7 (58.3)	3 (37.5)	
Partner status				p= 0.653[+]

Variable, N (%)	Qualitative Sample (n=20)	Prenatal (n=12)	Postnatal (n=8)	p value
Married/partnered	18 (90.0)	11 (91.7)	7 (87.5)	
Divorced/Separated	0 (0.0)	0 (0.0)	0 (0.0)	
Single	2 (10.0)	1 (8.3)	1 (12.5)	
Religion*				p= 0.034+
Protestant	12 (60.0)	10 (83.3)	2 (25.0)	
Catholic	4 (20.0)	1 (8.3)	3 (37.5)	
Other	2 (10.0)	0 (0.0)	2 (25.0)	
None	2 (10.0)	1 (8.3)	1 (12.5)	

+=Fisher's Exact; λ=Chi Square; U= Mann Whitney U; T=t-test

*Significant different between prenatal and postnatal groups, $p<0.05$

**Significant difference between prenatal and postnatal groups, $p<0.01$

Table 2: Infant Characteristics for the Qualitative Sample by Prenatal and Postnatal Group

Variable, N (%)	Infant Sample (n=20)	Prenatal (n=12)	Postnatal (n=8)	p value
Age at birth, Mean,g ± SD (range) Median, IQR	36.62 ±2.86 [37.2, 3.2]	36.77 ±1.63 [36.5, 2.7]	36.39 ±4.24 [37.9, 3.6]	p= 0.779[T]
≥ 37 weeks (full-term)	11 (55.0)	8 (66.7)	4 (50.0)	p= 0.648[+]
< 37 weeks (preterm)	9 (45.0)	4 (33.3)	4 (50.0)	
Gender				
Male	9 (45.0)	7 (58.3)	2 (25.0)	p= 0.197[+]
Female	11 (55.0)	5 (41.7)	6 (75.0)	
Gestational status				
Single birth	17 (85.0)	10 (83.3)	7 (87.5)	p= 0.656[+]
Multiple birth	3 (15.0)	2 (16.7)	1 (12.5)	
Anomaly Type				
Digestive	11 (55.0)	7 (58.3)	4 (50.0)	p= 0.103[+]
Heart	3 (15.0)	3 (25.0)	0 (0.0)	
Brain/Nervous System	2 (10.0)	1 (8.3)	1 (12.5)	
Musculoskeletal	3 (15.0)	0 (0.0)	3 (37.5)	
Diaphragm	1 (5.0)	1 (8.3)	0 (0.0)	
Birthweight				
Mean,g ± SD	2568 ±176	2524 ±668	2635 ±987	p= 0.767[T]
Average birth weight	12 (60.0)	7 (58.3)	5 (62.5)	p= 0.764[+]
Low birth weight (<2500g)	6 (30.0)	4 (33.3)	2 (25.0)	
VLBW (<1500g)	1 (5.0)	1 (8.3)	0 (0.0)	
ELBW (<1000g)	1 (5.0)	0 (0.0)	1 (12.5)	
Age at Interview, Days Mean ± SD (range) Median, IQR	15.2 ±11.2 (5-46) [13.0, 15.0]	12.83 ±7.61 (5-29) [12.5, 10.0]	18.63 ±14.95 (5-46) [14.0, 26.0]	p= 0.266[T]

+=Fisher’s Exact; λ=Chi Square; U= Mann Whitney U; T=t-test

Table 3: Frequencies of Decisions Made by Mothers in the Qualitative Sample on Behalf of their Infant by Diagnosis timing

Mother's Decision	**Qualitative Sample (n=20) n, %**	**Prenatal (n=12)**	**Postnatal (n=8)**	**p value**
Surgery	13 (65.0)	8 (66.7)	5 (62.5)	p= 0.918+
Intermittent or Continuous Medications	3 (.0)	2 (16.7)	1 (12.5)	
Withdrawal or withholding of life-sustaining treatment	2 (10.0)	1 (8.3)	1 (12.5)	
Blood	1 (5.0)	1 (8.3)	0 (0.0)	
Radiology Testing	1 (5.0)	0 (0.0)	1 (12.5)	

+=Fisher's Exact

CHAPTER FOUR: Manuscript Three-Mixed Methods

Processes, barriers, and facilitators of coping, parenting, and decision making for mothers of NICU infants receiving treatment for a congenital anomaly: Results of a mixed methods study

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Abstract

Background: A pre- or postnatal diagnosis of a congenital anomaly in an infant is a known source of parental distress and is particularly disruptive to mothers' adaptation after delivery. What remains unexplored is how the timing of a congenital diagnosis (prenatal or postnatal) and other factors may influence mothers' coping, parenting, and decision making for their infant requiring treatment in the NICU. Therefore, the purpose of this study was to describe how timing of a congenital diagnosis and other parent and infant factors influence coping, parenting, and decision making in mothers of infants in the NICU with a congenital anomaly, and the barriers and facilitators to coping, parenting, and decision making.

Methods: A cross-sectional, sequential explanatory mixed methods design was used to survey a convenience sample of mothers with infants receiving treatment in one urban neonatal intensive care unit (NICU). A subsample of mothers was interviewed to explain survey responses. Chi-square (or Fisher's exact) and t-tests (or Mann Whitney U) statistics were used to identify associations between coping and decision making variables. Parenting variables were analyzed using maximum difference scaling and Hierarchical Bayes estimation. Interviews were analyzed using qualitative content analysis. Qualitative data was used to further explain quantitative findings.

Results: Thirty-seven mothers of NICU infants completed cross-sectional quantitative surveys and 20 of these mothers participated in in-depth qualitative interviews. On quantitative surveys, mothers in the postnatal group demonstrated more engaged decision making (86%), compared with mothers in the prenatal group (52%). Mothers in the

prenatal group utilized significantly more acceptance coping than mothers in the postnatal group (U=78; p=0.01). Parenting values among mothers in the prenatal and postnatal groups were similar; mothers in both groups placed greatest priority on parenting values such as making medical decisions and focusing on their child's health, and lowest value on keeping a positive attitude. Qualitative findings revealed person-related (behavioral, cognitive, emotional, & spiritual) and contextual-related (access, communication, environmental, relational) barriers and facilitator themes of coping, parenting, and decision making adaptation. Mixed methods findings explained that mothers in the prenatal group, while deferring decisions to the health care team in the NICU, were engaged in decision making about their infant's congenital anomaly before delivery.

Conclusion: Findings, which highlight differences in coping, parenting, and treatment decision making between mothers of NICU infants who learn of the congenital diagnosis prenatally vs. postnatally, guide providers in offering support to this specialized population.

Keywords: Congenital anomaly, neonatal intensive care, coping, parenting, decision making

Introduction

Congenital anomalies are structural or functional anomalies that arise during intrauterine life and are present at the time of birth (CDC, National Center for Health Statistics, 2017). Each year, approximately 3.2 million infants are diagnosed with a congenital anomaly, which can lead to life-long disability or death (World Health Organization, 2016). A pre- or postnatal congenital diagnosis can be a source of parental distress and can disrupt parental adaptation, particularly when neonatal intensive care (NICU) hospitalization of the infant is necessary to address a health challenge in the immediate postnatal period (Fonseca et al., 2012; Mazer et al., 2008).

Previous studies have examined the psychological impact of an infant's prenatal versus postnatal congenital diagnosis on parents. Parents who learn of a congenital diagnosis either prenatally or postnatally report clinically significant levels of psychological distress at the time of their infant's birth (Brosig et al., 2007). Mothers tend to present with more anxiety, depression, and lower quality of life than fathers at the post-diagnosis stage, which suggests a greater impact of the diagnosis on mothers (Bevilacqua et al., 2013; Fonseca et al., 2012). In fact, in one study, mothers receiving a prenatal congenital diagnosis had anxiety and depression scores similar to patients with a major depressive episode (Leithner et al., 2004). While the distress of mothers following a congenital diagnosis is well documented, what remains unexplored is how the timing of a congenital diagnosis and other factors may influence mothers' coping, parenting, and decision making for their infant requiring treatment for a congenital anomaly in the NICU context.

According to the Roy Adaptation Model (RAM), adaptation is both a process and an outcome, whereby focal and contextual stimuli influence one's ability to cope, which in turn influences one's adaptation (C. Roy, 1997). The RAM has been used in prior studies to explore individuals' efforts to adapt to a perceived challenge. In this study, the RAM provides a model to explore how mothers, when faced with the congenital diagnosis of their infant and a NICU hospitalization (focal stimuli), are able to cope, parent, and make decisions for their infant who requires treatment in the intensive care setting. Understanding these processes of adaptation will inform decision and family support interventions for NICU mothers facing their infant's treatment for a complex congenital anomaly.

Purpose

The purpose of this study is to explore the coping, parenting, and treatment decision- making adaptation of mothers of NICU infants with a congenital anomaly. The specific aims were: 1) To estimate differences and associations of coping factors (infant illness severity perception; general coping; religious coping), parenting values (i.e. "good parent" attributes), and decision-making engagement for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth. *Hypothesis 1.1*: Mothers who learn of their infant's congenital diagnosis prenatally will exhibit higher levels of positive coping than mothers who learn of their infant's congenital diagnosis postnatally. *Hypothesis 1.2*: Mothers who learn of their infant's congenital diagnosis prenatally will exhibit greater decision making engagement than mothers who learn of their infant's congenital diagnosis postnatally, 2) Explore barriers and facilitators related to coping, parenting, and decision making for mothers of NICU infants with a congenital anomaly,

3) Integrate quantitative and qualitative data to describe the barriers and facilitators of coping, parenting, and decision making for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth.

Methods

Overview of Methods

Study design. This study utilized a cross-sectional, sequential explanatory mixed methods design [QUAN+QUAL] (Creswell & Plano Clark, 2011). This design allowed us to gain an in-depth understanding of the participant's coping, parenting, and decision-making adaptation processes by using the qualitative data to explain the statistical results from the quantitative data, while providing greater insight into potential differences in coping, parenting, and decision making between mothers in the prenatal vs. postnatal group (Creswell & Plano Clark, 2011). We adapted the Roy Adaptation Model to select study variables and determine which relationships to analyze (C. Roy, 2009)(Figure 1).

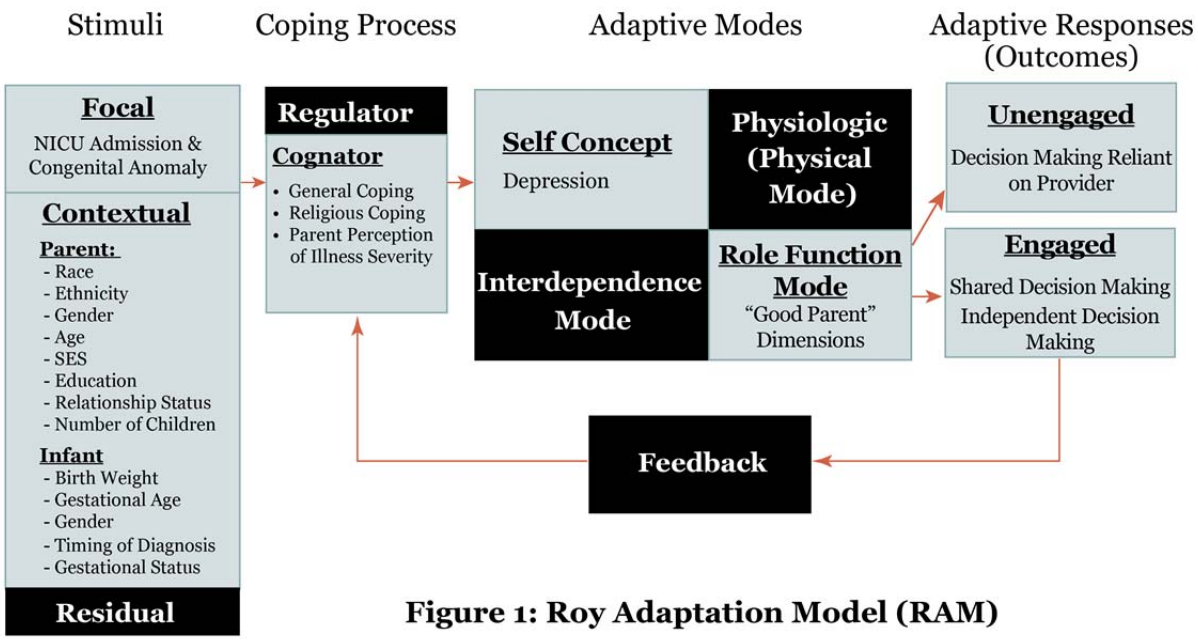

Figure 1: Roy Adaptation Model (RAM)

Sample. The institutional review board of Johns Hopkins Medicine (Baltimore, Maryland) approved this study. Participants were recruited from the neonatal intensive care unit (NICU). Mother participants were eligible if they were 17 or older, biologically related to or had primary caretaking responsibility of an infant 0-6 months of age hospitalized in the NICU, had an infant with a heart, lung, brain/nervous system, diaphragm, renal, musculoskeletal, digestive, or chromosomal anomaly, had made or deferred a treatment decision as identified through a Decision Checklist, and had the ability to speak and write English. Exclusion criteria included surrogates representing Child Protective Services (CPS), as decision-making processes of these surrogates may be limited by CPS regulations. Mother participants gave consent to include their infant's relevant medical record data pertinent to the study. We sought to include a racially diverse sample, representative of the population at the hospital. Additionally, to reduce the risk of participant burden, we only approached mothers after the fourth day of their infant's NICU stay. This timing of approach was chosen based on prior interviews with NICU parents that revealed parents prefer a few days of adjustment to the NICU environment before participating in research (Hinds and colleagues, correspondence).

Quantitative Data Collection

Sample size. Based on a publication by Brosig, Whitstone, Frommelt, Frisbee, & Leuthner, which reports a medium effect size for psychological distress in parents of infants who learn of a congenital diagnosis prenatally vs. postnatally, and based on a publication by Hertzog (2008), which provides guidelines on adequate pilot sample sizes to detect a medium effect size (η^2=0.14, which corresponds to a Cohen's D value of a medium effect) at a significance of 0.05, a convenience sampling method was used to

attempt recruitment of 40-50 mothers of infants with a congenital anomaly hospitalized in the Johns Hopkins NICU (Baltimore, MD) for this exploratory study (Brosig et al., 2007; Hertzog, 2008). An attempt was made to recruit 20 mothers in each group (i.e. the two groups being mothers who learned of their infant's congenital diagnosis prenatally vs. postnatally).

Procedures. Surveys were pilot tested in a sample of 3 NICU mothers to test for feasibility before recruitment began. The Johns Hopkins Medicine IRB approved a HIPAA waiver to search the electronic patient records for mothers of infants who met inclusion criteria. Once a potentially eligible mother was identified, a study team member consulted with a NICU clinician and prescreened the mother to ensure she met eligibility criteria. NICU clinicians used a written script and study information sheet when seeking permission for a study team member to attempt recruitment. The investigator (MKU) scheduled a mutually agreeable time to meet with mothers who indicated interest in the study. Before beginning data collection, the investigator described the study to the mother, confirmed inclusion eligibility with the mother, and asked the mother to provide written consent to participate.

Once consent was obtained, mothers were invited to a private sitting area of the NICU, where a brief explanation of survey measures was provided. A study team member offered mothers with low literacy one-on-one assistance to complete surveys. All quantitative surveys were completed using paper and pen. The investigator used medical records to gather infant data. After survey completion, the investigator reviewed surveys for completeness. In cases where responses were incomplete or unclear, the study team

member sought clarification from the respondent. Participants were offered a $15 gift card for their time/effort.

Measures.

Parent and infant characteristics. Individual mother characteristics were assessed via the study questionnaire, which included questions about sociodemographics (e.g., age, sex, race, education, income, employment status), as well as questions about timing of the infant's congenital diagnosis and number of other children. Infant characteristics were collected using the electronic medical record and included birth weight (grams), gestational age (weeks and days), gestational status (single vs. multiple gestation), sex, and type of congenital anomaly. Other main study variables were measured via established instruments, which are listed in detail below.

***Infant illness perception*.** The Brief Illness Perception Questionnaire, developed by Broadbent et al. was used to measure mothers' perceptions of their infant's illness severity (Broadbent, Petrie, Main, & Weinman, 2006). The Brief Illness Questionnaire consists of 9-items that capture the main dimensions of an individual's illness perception, including cognitive and emotional representations (Broadbent et al., 2006). Each of the nine items is rated on a 10-point Likert-type scale (1 to 10). An example of a survey question is, "how sick is your baby?" Relevant anchors for this question are "not sick at all" (1) to "extremely sick" (10). Although normally completed by a patient in response to their illness, the survey has been modified in previous studies to measure parents' perceptions of their infant's illness (Brooks et al., 2012). The scale has good test-retest reliability and has been associated with a variety of chronic illness outcomes in adults (Broadbent et al., 2006).

***General coping*.** The Brief COPE survey was used to assess generalized coping among mothers. This 28-item survey developed by (Carver, 1997) is an abbreviated version of the COPE inventory (University of Miami, Department of Psychology, 2007). The Brief COPE represents a multi-faceted coping-style inventory with 14 coping-strategy subscales. Each coping-strategy subscale is comprised of two questions measured using a 4-point Likert-type scale, indicating the extent to which an individual uses the particular coping strategy (1="I haven't been doing this at all"; 4= "I've been doing this a lot"). Subscale totals range from a minimum score of two to a maximum score of eight (Carver, 1997). We grouped the Brief COPE subscales according to positive (or beneficial) coping mechanisms and negative (or inhibitory) coping mechanisms, in order to compare our data with previous studies that have classified the Brief COPE subscales into positive and negative coping categories (Huenink & Porterfield, 2017). The reliability and validity of the Brief COPE has been demonstrated and the tool has been utilized among NICU parent populations (Brelsford, Ramirez, Veneman, & Doheny, 2016; Carver, 1997; Huenink & Porterfield, 2017; Shaw, Bernard, Storfer-Isser, Rhine, & Horwitz, 2013).

Religious coping. The Brief RCOPE was used to assess religious coping among mothers. This 14-item survey includes two subscales of positive and negative religious coping (7-items each). Positive religious coping can be defined as "feeling as though God is supporting one through a difficult time or praying to God for love and grace" (Brelsford et al., 2016; Pargament, Feuille, & Burdzy, 2011). Alternatively, negative religious coping may be defined as "feeling God has abandoned the individual or feeling anger at God" (Brelsford et al., 2016; Pargament et al., 2011). Each of the 14 items on

the Brief RCOPE is assessed using a 4-point Likert scale, indicating the extent to which the individual uses the coping strategy (1=”not at all”; 4= “I did this a lot”). Both the positive and negative religious coping subscales have demonstrated high internal consistency of 0.90 and 0.81 (Pargament et al., 1998). Additionally, studies have demonstrated that positive and negative religious coping have high levels of incremental validity compared to measures of general religiousness and secular factors, such as mood and social support (Pargament et al., 2011). The RCOPE scale has been used in the NICU parent population (Brelsford et al., 2016).

Depression. The Edinburgh Postnatal Depression Scale (EPDS) was used to assess depressive symptomatology among mothers. This 10-item scale was developed to screen for significant depressive symptoms in perinatal women and is one of the most widely used self-report instruments for identifying postnatal depression in research and the clinical setting (Matthey, Henshaw, Elliott, & Barnett, 2006). Based on their mood over the last seven days, respondents are asked to consider each of the 10 survey items, which are assessed using a 0 to 3 Likert-type scale, indicating the extent to which each statement applies to the individual, for a composite total score of 0-30. The EPDS has been validated among adult and adolescent women and demonstrates moderate to good reliability across populations (Boyd, Le, & Somberg, 2005; J. L. Cox & Holden, 2003). We utilized a score of greater than 13 as a cutoff, since scores greater than 13 are a possible indication of significant depressive symptomatology (J. L. Cox, Holden, & Sagovsky, 1987; Wisner, Parry, & Piontek, 2002). The EPDS has been used to assess depressive symptoms in various studies that involve the NICU mother population (Neri,

Agostini, Salvatori, Biasini, & Monti, 2015; Stasik-O'Brien, McCabe-Beane, & Segre, 2017).

Decision checklist. A decision checklist identifying decisions commonly required of parents of an infant with a congenital anomaly in the NICU was developed with the input of a neonatologist on the study team to determine what decisions mothers had made in the NICU related to their infant's congenital anomaly. Mothers were encouraged to review decisions they had made related to their infant's congenital anomaly. Mothers were then asked to identify which single decision was most important for them, out of those decisions considered. The decision identified as most important was used to complete the Control Preference Scale-Pediatrics (CPS-P), described below.

Decision making engagement. The Control Preference Scale-Pediatrics (CPS-P) was used to measure decision-making engagement by mothers. The adult version of the CPS-P, the Decision Control Preference (DCP) scale, was developed by Degner, Sloan, and Venkatesh and asks respondents to identify their level of involvement in a specific decision (Brelsford et al., 2016; Degner & Sloan, 1992; Degner, Sloan, & Venkatesh, 1997). Our study utilized an adapted version of the DCP scale, the CPS-P, which assessed parents' actual decision control preferences related to health care decisions for their child (Pyke-Grimm, Degner, Small, & Mueller, 1999). On the CPS-P, parent participants' were presented with 5 levels of decision control, which were then scored into three decision control categories, ranging from 1) independent decision making, in which parents' decisions are made independent of health care providers, to 2) shared decision making, in which decisions are shared between the parent and health care providers, to 3) reliant decision making, in which parents give health care providers

decision making control. In this study, we presented parents with all five options of the CPS-P, and asked them to identify the extent to which they were engaged in decision making for their infant. Their responses were categorized as either engaged (independent or shared decision making) or unengaged (reliant on the health care provider). The CPS-P has been validated in the pediatric setting and used with parents of pediatric cancer patients with a reliability estimate of 0.69, based on Gulliksen and Tukey's index of reliability (Mack et al., 2011; Pyke-Grimm et al., 1999; Pyke-Grimm, Stewart, Kelly, & Degner, 2006).

Parenting values. The Good Parent Ranking Exercise (GPRE) is a discrete choice experiment, which allowed parents to select values and goals they believed to be most or least important in parenting their infant in the NICU with a congenital anomaly. Items on the GPRE were based on previous qualitative research with parents of children with other life threatening illness (Hinds et al., 2009). Items were revised in a subsequent pediatric palliative care study, based on input from interdisciplinary palliative care professionals and a parent focus group, resulting in 12 items, which have been used in subsequent studies (Feudtner et al., 2015; Mooney-Doyle, Deatrick, Ulrich, Meghani, & Feudtner, 2018; October et al., 2014). Parents selected choices among 12 permutated sets of four good parent beliefs based on rank-ordering, meaning items are ranked from most to least important (Mooney-Doyle et al., 2018). Maximum difference scaling is used to calculate a point value for each parenting value. Each point value represents the relative importance of the parenting value in comparison with other parenting values. Point values for each of the 12 parenting values when summed equal 100 points (Feudtner et al., 2015).

Qualitative Data Collection

Sampling. Participants from the quantitative sample who agreed to join the qualitative portion of the study were interviewed upon completion of surveys. A purposive sampling technique was used to deliberately sample ~50% of mothers who learned of their infant's congenital diagnosis before birth and ~50% from mothers who learned of their infant's congenital diagnosis after birth, to capture a range of views about decision making in the NICU and provide a thorough understanding of facilitators and barriers to coping, parenting, and decision making. Based on prior research, it was anticipated that at least 12-20 participants total needed to be interviewed before data redundancy was achieved (Hinds et al., 2009; Johnson & Christensen, 2008; October et al., 2014).

Procedures. Interviews were conducted in a private room on the NICU or by phone. Before beginning the interview, the investigator advised the mother that she could stop the interview at any time, if desired. In addition, NICU social workers were on call, in case the mother became upset and required additional support or follow-up. To give context to interview questions, the investigator reviewed with the mother the most important decision she has made (or deferred) for her infant using the Decision Checklist. On the Decision Checklist, mothers were asked to select all decisions they had made, and then circle the most important decision. The most important decision was the focus of qualitative discussion. A semi-structured interview guide was developed by the research team based on variables of coping, parenting, and decision making selected through the Roy Adaptation Model. Additional probing questions were used to further understand a

mother's perceptions of barriers and facilitators related to coping, parenting, decision making.

During the interview, the investigator took notes and memos to add contextual information. Interviews were audio recorded and transcribed verbatim. Personal identifiers were removed from the transcribed interviews, after checking for accuracy against the original audio file (DiSantis et al., 2013). As a form of member checking, the investigator summarized interview responses with mothers prior to ending the interview, and amended content as necessary. Each interview lasted 30-60 minutes. At the completion of the interview, each participant received an additional $15 gift card for her time/effort. The investigator and mentoring team met on a bi-weekly basis to monitor and discuss qualitative data collection.

Data Analysis

Our analytic approach consisted of three phases. In phase one, we conducted descriptive statistics to summarize our data, using medians and interquartile range (IQR) or means and standard deviations (SD), depending on the distribution pattern of the variable. Dichotomous and categorical variables were summarized using frequencies and percentages. Parenting characteristics were analyzed using maximum difference scaling and Hierarchical Bayes estimation via Sawtooth MaxxDiff and Choice-Based Conjoint/HB modules (v. 6.0), which uses multinomial logistic regression modeling to calculate the probability of a parent choosing a specific 'good parent' attribute as the best or worst given attributes shown in a set, transforming raw scores to a 0 to 100 scale (Feudtner et al., 2015; October et al., 2014). Independent sample t-tests or Mann-Whitney U tests were used to estimate differences in coping factors (infant illness severity

perception, general coping, religious coping, and depression) between the two diagnosis groups (prenatal vs. postnatal), depending on the distribution pattern of the variable. A Fisher's Exact test was used to compare the decision making engagement (engaged vs. unengaged) of mother's in each diagnosis group (prenatal vs. postnatal), given that expected frequencies were <5 in some crosstab cells. Spearman's rank-order correlation was used to analyze associations between coping variables for mothers in each diagnosis group (prenatal vs. postnatal). Effect sizes of coping variables were calculated using r type estimates or Cohen's D. SPSS 25 was used for all analyses. Level of significance was set at 0.05.

In phase two, we utilized a qualitative content analysis approach to analyze interview data. Qualitative content analysis is utilized when describing a particular phenomenon (coping, parenting, and decision making, in this case), and when research literature on the study phenomena is limited (Hsieh & Shannon, 2005). Interview transcripts were reviewed prior to coding to verify accuracy. Interviews were uploaded into NVivo 11. A consensus method between the first author and senior author was used to develop a priori codes from the qualitative interview guide. These a priori codes were collected into a codebook, which was used to code subsequent interviews. Steps in the qualitative analysis included: (1) reading the interview transcripts 2-3 times to gain a general understanding of the content, (2) coding passages as either a facilitator (defined as any person, resource, or attribute that helps a parent) or barrier (defined as any person, resource or attribute that deters or prevents parents), (3) coding passages into barrier and facilitator categories, including access, behavioral, cognitive, communication, emotional, environmental, illness perception, instrumental, relational, and spiritual, and (4)

examining each factor to identify themes. The first three steps of the qualitative analysis process were conducted deductively by MKU and MW. In the fourth step of analysis, an inductive approach was utilized. The first author developed the primary themes, which were discussed with the senior author (MTN). The transferability (applicability) of study findings was aided through the use of purposive maximum variation sampling. Multiple coders were employed to increase reliability (Creswell & Plano Clark, 2011). Credibility of data was increased through the presentation of representative quotes with the main study themes. Finally, an audit trail was used to ensure the analysis process was acceptable and the findings and interpretations were supported by the data.

In phase three, we utilized Onwuegbuzie and Teddlie's analytic process of synthesizing qualitative and quantitative findings to form conclusions (Onwuegbuzie & Teddlie, 2003). Combining quantitative and qualitative data about mothers' coping, parenting, decision making was facilitated through a data display table. Data were then compared and integrated to form meta-inferences (Onwuegbuzie & Teddlie, 2003).

Results

The final analysis sample included 37 mothers who completed quantitative surveys and 20 mothers who completed follow-up qualitative interviews. Results are presented in three sections. First, we present quantitative descriptive findings for mothers' demographic, coping, parenting, and decision making results. Next, qualitative findings are presented. Finally, we present the integration of significant quantitative results with qualitative data, facilitated through a mixed methods data display matrix.

Quantitative Data.

Sample characteristics. A total of 45 mothers were approached and 37 (82%) consented to participate in the study. Those declining stated too much going on (n=3), general disinterest (n=2), imminent discharge of their infant (n=1), protection of infant's diagnostic data (n=1), or already participating in multiple research studies (n=1) as reasons for not participating. Of the 37 mothers who agreed to participate, all completed and returned surveys. Table 1 summarizes the sample characteristics of the quantitative sample (n=37) and the qualitative subsample (n=20) of mothers. Demographics of the quantitative and qualitative samples were similar. Overall, the quantitative sample consisted of predominantly White (68%), partnered (87%) mothers with a mean age of 30.6 (±6.0) who were employed full-time (60%). Mothers in the postnatal group had significantly higher education (Fisher's Exact p=0.016) and significantly higher Catholic religious affiliation (Fisher's Exact p=0.008). The qualitative subsample (n=20) was also predominantly White (70%), partnered (90%), employed full-time (55%) with a mean age of 31.25±5.73. Table 2 summarizes characteristics of infant demographics.

Decision making results. Tables 3 and 4 summarize mothers' quantitative decision making data. In the overall and qualitative sample, surgery was the most common decision among mothers (57% and 65% respectively) (Table 3). Decision making engagement was significantly correlated with education (V=0.473; p=0.016) and religious affiliation (V=0.558, p=0.009); parents with more education and identifying with a religious affiliation were more likely to be actively engaged in decision making. Engaged decision making was used more frequently in the postnatal group (86%) than

the prenatal group (52%), but did not reach statistical significance (Fisher's Exact p=0.07).

General coping results. Tables 4 and 5 summarize mothers' illness perception and general coping data. We found a significant negative correlation between infants' birth weight and mother's overall illness perception (r_s=-0.347; p=0.035), indicating that the lower the infant's birth weight the more seriously ill the mother perceived the infant's status. Mothers utilized positive coping strategies (emotional support, acceptance, active coping, planning, instrumental support, religion, and reframing) most frequently (mean score >6; scores can range 0-8). We demonstrated that acceptance coping was positively correlated with number of other children (r_s=0.334; p=<0.05) and reframing (r_s=0.522; p=<0.01). We also found that mothers in the prenatal diagnosis group utilized significantly more acceptance coping (7.04 ±1.15), compared to mothers in the postnatal diagnosis group (6.00 ±1.04), (U=78; p=0.01).

Religious coping results. Tables 4 and 5 summarize mothers' religious coping. Mothers reported utilizing more positive religious coping (19.7 ±6.5) compared to negative religious coping (9.5 ±3.5) and 92% of mothers rated religion as somewhat or very important (positive and negative subscale scores can range 7-28). In the overall sample, a significant relationship was found between positive religious coping and denial (r_s=0.337, p=<0.05) and positive religious coping and substance use (r_s=0.326, p=<0.05). A significant relationship was also found between negative religious coping and self blame (r_s=0.608, p=<0.05) and negative religious coping and depression (r_s=0.549, p=<0.01).

Depression results. Table 4 and 5 summarize mothers' depression data. The mean depression score for the overall sample was 9.35 ±4.6 and 8.45±4.9 for the qualitative

sample (scores range from 0-30). Significant correlations were found between depression and the following five coping strategies used by the mother: instrumental support (r_s=0.364, p=<0.05), denial (r_s=0.359, p=<0.05), self blame (r_s=0.802, p=<0.01), venting (r_s=0.330, p=<0.05), and negative religious coping (r_s=0.549, p=<0.01).

Parenting results. For the overall sample and the prenatal and postnatal groups, the two most highly ranked parenting values were "putting my child's needs above my own when making medical care decisions" and "Focusing on my child's health" (Table 6). Overall rankings for the prenatal and postnatal groups were similar except the postnatal group ranked "focusing on my child's quality of life third, while the prenatal group ranked this value sixth.

Qualitative Results.

In performing the qualitative analysis, we identified several themes that further elucidated mother participants' views regarding barriers and facilitators to coping, parenting, and decision making for an infant with a congenital anomaly in the NICU. Themes associated with barriers and facilitators were categorized as person-related (behavioral, cognitive, emotional, & spiritual) or contextual-related (access, communication, environmental, relational) factors. Table 7 provides definitions of these themes and a summary of the qualitative findings across themes.

Mixed Methods Results

Data from the qualitative interviews were used to clarify or further explain the quantitative survey findings. Although quantitative findings showed more decision engagement among the postnatal group of mothers, qualitative findings clarified that mothers in the prenatal group were also engaged in decision making, but frequently

during the time before their infant's birth; their decision making was focused on selecting specific providers and institutions to obtain care once the infant was born. Qualitative data also further described that mothers who learned of their infant's congenital anomaly prenatally had time before delivery to consider and understand probable treatment options, which seemed to explain the significant difference in use of acceptance coping for this group. Mothers who learned of their infant's diagnosis postnatally described being more involved in decision making in the NICU, which is when they learned of their infant's diagnosis and potential treatment options. Mothers gave further descriptions of other quantitative findings through their interview dialogue, as displayed in table 8.

Discussion

To our knowledge, this is the first study to examine the potential impact of the timing of a congenital diagnosis on parents' coping, parenting, and decision making adaptation for an infant receiving treatment in the NICU context. Integrating the quantitative data and qualitative interviews revealed that mothers who learned of their infant's congenital anomaly prenatally vs. postnatally perceived similar barriers and facilitators to their coping, parenting, and decision making in the NICU, but some differences in their adaptation were apparent and will be discussed.

Our finding that surgical treatment decisions were most significant (57%) for mothers in the overall sample cohered with our data that the majority of infants in our sample had digestive anomalies and required surgery soon after birth. Our statistical analyses also displayed that mothers in the postnatal diagnosis group displayed more engaged decision making than mothers in the prenatal group. We originally hypothesized that mothers in the prenatal diagnosis group would display more engaged decision

making due to their ability to obtain prenatal counseling or education, and identify resources for the birth and postnatal care of their infant. Our demographic findings revealed that the postnatal diagnosis group had significantly higher education and significantly more mothers identifying a religious affiliation. These demographic variables were positively correlated with decision-making engagement, so our finding that mothers in the postnatal group demonstrated more decision-engagement could be a result of confounding. What we know about the impact of education and religion on decision from other studies includes that parents' preferred decision-making role (i.e. engaged vs. unengaged) may be influenced by their knowledge of the child's disease or proposed treatments, with higher knowledge/education being associated with more engaged decision making (Lipstein, Brinkman, & Britto, 2012). Additionally, in previous studies religious beliefs have been shown to influence the degree to which parents' are involved in decision making. Studies by (Ellinger & Rempel, 2010) and (Sharman et al., 2005) report that parents were active decision makers despite their belief that God controlled their child's health outcome. Parents in these studies were involved in making decisions that would preserve or extend the life of their child, which may be indicative of religious beliefs that prioritized the dignity or sanctity of life (Ellinger & Rempel, 2010; Sharman et al., 2005). Our qualitative interviews further clarified our statistical findings, showing that mothers in the prenatal and postnatal groups both engaged in decision making, but at different time points and in different ways. Mothers in the prenatal group engaged in decision making before their infant was born, choosing a facility and/or physician to treat their child after birth, and then relinquished decision making to the healthcare team, with whom they had built a trusted relationship, once their infant was

born. Mothers in the postnatal group more more engagement in specific NICU decisions (i.e. surgery, etc.), since the infant's diagnosis was new and information was still being gathered and decided.

Overall, mothers' infant illness severity perception scores were more elevated in our sample, compared to the illness severity perception scores of parents reported in previous neonatal and pediatric studies (Brooks et al., 2012; Michel, Taylor, Absolom, & Eiser, 2010). In these other studies, children had fewer (29%) congenital anomalies (Brooks et al., 2012) and were older children (age 12-15) and in cancer remission (Michel et al., 2010). This may account for illness perception severity score differences between previous samples and ours. Our samples' elevated illness perception scores are significant since parents' illness perceptions of their infant, rather than objective information about the infant's illness severity, have been shown to influence parents' notion of how ill their infant actually is (Brooks et al., 2012). In short, mothers who perceive their infant to be more severely ill are likely to use this perception in making decisions for their infant, rather than objective information from the health care team (Brooks et al., 2012). At the same time, an elevated illness severity perception may not be entirely negative. In several studies, elevated illness severity perception, particularly for those who perceive that an illness impacts them on a daily basis, have reported more benefit-finding and post-traumatic growth (Barakat, Alderfer, & Kazak, 2006; Michel et al., 2010).

In our sample, mothers most commonly utilized positive forms of coping. This finding is consistent with a study, which reported that parents of a general NICU population most commonly utilized positive coping, including emotional support, acceptance, active coping, planning, instrumental support, religion, and reframing, when

coping with a NICU stay (Huenink & Porterfield, 2017). Other studies have reported that helpful coping mechanisms, including active coping, planning, instrumental support, and reframing, can improve psychological outcomes in NICU parents, such anxiety (Greening & Stoppelbein, 2007; Shaw et al., 2013; Young Seideman et al., 1997). We found that mothers who learned of their infant's congenital diagnosis prenatally displayed significantly higher acceptance coping. This high use of acceptance coping among mothers in the prenatal diagnosis group was further explained in the qualitative interviews. Mothers who learned of their infant's diagnosis prenatally had time to adjust to their infant's congenital diagnosis before birth, so felt prepared once the infant was born. Other studies that have explored parents' acceptance of a difficult diagnosis for their child describe how, with time, families acquire tools, which help them frame their reality more positively (Kandel & Merrick, 2007). Parents in the postnatal diagnosis group, who did not have the benefit of time to adapt to their infant's diagnosis may have struggled more to accept their child's condition.

We found a significant correlation between negative religious coping and both self-blame and depression. Examples of these negative coping mechanisms were conveyed in qualitative interviews. One author has suggested that the correlation between negative religious coping and self blame can manifest because the individual views God as a vengeful Higher Power who is punishing them or believes that an action in their past has produced their current suffering (Pargament et al., 1998). Authors of other NICU studies have reported significant correlations between negative religious coping and denial, another negative coping mechanism (Brelsford et al., 2016). A second significant correlation we found was between negative religious coping and depression.

Negative religious coping has been shown to predict or be associated with depressive symptoms in other studies of parents of children with autism (Tarakeshwar & Pargament, 2001) and parents of children with cystic fibrosis (Szczesniak, Zou, Stamper, & Grossoehme, 2017).

The mean depression scores for mothers participating in our study were well below the typical cutoff used to detect depressive symptomatology (Stasik-O'Brien et al., 2017). Many factors contribute to depressive symptomatology and it's possible that our sample was not predisposed or that parents who were not depressed did not enroll in the study. Some studies indicate that partner relationships, especially if healthy, and positive coping mechanisms, as our sample demonstrated, can act as buffers against depression (Rychik et al., 2013). We did find, in both the prenatal and postnatal diagnosis groups, a significant correlation between negative religious coping and depression. However, negative coping in our sample was low, a finding that is consistent with the low levels of depressive symptomatology found in our sample. Another possibility is that depressive symptomatology was low in our sample due to the timing at which mothers were surveyed. Some studies have suggested that depression, while not significant in the perinatal period, becomes significant as the child with a severe congenital anomaly ages. For instance, (Solberg et al., 2011) found that mothers of infants with severe congenital heart defects did not have significant levels of depression at 30 weeks gestation (during pregnancy), but did have significant depression at 6 and 18 months. Another study that reported systematic review findings on the mental health of parents of children with heart defects found that parents of children with critical heart defects are at elevated risk for psychological issues, including depression, in the weeks and months following surgery

(Woolf-King, Anger, Arnold, Weiss, & Teitel, 2017). While just over half the mothers in our sample were surveyed in the days or months following a surgical decision, the other mothers were surveyed following a medical decision, which may not have contributed less to depressive symptomatology. While our sample did not display concerning depressive symptomatology at the time of study participation, mothers who participated could benefit from careful follow up, since depressive symptomatology could develop later, and studies indicate that elevated depression and a preference for a passive role in decision making are correlated (Schweingruber & Kalil, 2000; Woolf-King et al., 2017). While some parents with depressive symptomatology may prefer a passive decision making role, others' decision-making preferences could be altered by depressive symptomatology and could result in incongruence between the parents' preferred decision making role and actual decision making role, which has been shown to result in decision-making dissatisfaction (Ranchod et al., 2004).

Findings from the Good Parent Ranking exercise represent what we understand to be the first exploration of the "good parent" concept in an exclusively NICU parent population. Previous studies have explored what being a "good parent" means for parents of critically ill children with cancer and children in the PICU (Hinds et al., 2009; October et al., 2014). Two additional studies recruited parents from a combination of critical care settings, including the pediatric, cardiac, neonatal intensive care, oncology, and bone marrow units (Feudtner et al., 2015; Mooney-Doyle et al., 2018). Parents in our prenatal and postnatal diagnosis samples endorsed "putting my child's needs above my own when making medical decisions" and "focusing on my child's health" as the top two "good parenting" values when making decisions for their infant with a congenital anomaly.

These values represent mothers' perception that illness management is a key parenting responsibility. These findings are consistent with the other pediatric studies measuring good parent attributes in parents of children with critical illnesses (Feudtner et al., 2015; Mooney-Doyle et al., 2018; October et al., 2014).

One interesting "good parent" finding is that mothers who learned of their infant's congenital diagnosis postnatally ranked the value "focusing on my child's quality of life" higher than mothers who learned of their infant's congenital diagnosis prenatally. It is well documented that parents who learn of a congenital diagnosis prenatally are often devastated (Miquel-Verges et al., 2009). However, while these parents may experience uncertainty up until the time of their infant's birth, they may also view the prenatal period as time to prepare for their infant's specific needs after birth, as other studies show and our qualitative sample confirmed (Brosig et al., 2007; Nusbaum et al., 2008). Mothers in the postnatal group who did not have time to prepare for their infant's needs may have been more concerned about their infant's quality of life in the immediate neonatal period. It's also possible that the postnatal group's higher ranking of quality life on the Good Parent Ranking Questionnaire reflects these mothers' own concern with quality of life immediately after learning of their child's congenital diagnosis. Studies that have explored parents' quality of life following their child's congenital diagnosis indicate that some aspects of mothers' quality of life are low in the early post diagnosis stage, but increase/normalize over time (Fonseca, Nazare, & Canavarro, 2014). Studies comparing quality of life between mothers who learned of their infant's congenital anomaly prenatally versus postnatally report that mothers who receive a prenatal diagnosis have a higher psychological quality of life (Fonseca et al., 2012). Mothers in the postnatal group,

if experiencing low quality of life themselves, may have similarly been concerned about low quality of life for their infant.

Strengths & Limitations

Although previous studies examine the psychological impact of a prenatal vs. postnatal congenital diagnosis on parents (Brosig et al., 2007; Fonseca et al., 2014; Skari et al., 2006), this is the first study, of which we are aware, that examines decision making for parents of infants with a prenatally vs. postnatally diagnosed congenital anomaly receiving treatment in the NICU. There are potentially many decision points for parents of children with congenital anomalies, and our study identifies the types of anomalies that may necessitate decision making during the immediate neonatal period, as well as the decisions that parents deem important at this stage in their child's treatment. We have identified important coping processes and parenting values that may assist parents' decision making at this juncture. Additionally, our participation rate of 82% was higher than participation rates (73%) reported in similar studies (Fonseca et al., 2014). To expand these findings, next steps could include conducting a large, multi-center study using block or stratified sampling, so that differences in decision making for specific congenital anomaly groups could be detected. Additionally, a longitudinal study could explore other time points where parents are faced with decision making for congenital anomalies and how specific coping, parenting, and decision making needs change over time.

There are several limitations to this study. The cross-sectional design precludes drawing causal inferences within the quantitative data and generalizability is limited due to the use of a convenience sample from a single acute care center. Mothers who declined

to participate may have had more experience making decisions, such as mothers with infants ready for discharge, or a different decision-making process, such as mothers too involved in their infant's care to participate, potentially limiting the data captured by this study. The small sample size precluded performing more detailed statistics to test the relationships among variables identified through our conceptual model, the Roy Adaptation Model (RAM), because they lacked power. Although our qualitative data seemed to affirm relationships among variables, future studies could utilize the RAM to identify variables, which could be tested more in depth quantitatively. The small sample may have also increased the likelihood that a significant difference in decision-making engagement between the prenatal and postnatal diagnosis groups was not detected. Utilizing surveys that relied on self-report introduced the potential for self-report, social desirability, and shared methods variance bias. We attempted to minimize the social desirability bias by reminding participants that survey responses had no right or wrong answers, and by encouraging participants to answer based on their own experiences. Because we utilized non-probability sampling, self-selection bias is possible and may have led to a sample of parents who could be present in the NICU (i.e. we may not have captured parents who were caring for other children at home and/or parents holding multiple jobs which prevented them from being present) and had reached a level of acceptance that enabled them to talk more openly about their experience. Although demographically our study sample was racially representative of the infants with congenital anomalies admitted to the recruitment hospital and more diverse than many other NICU decision making studies, which include primarily White participants (Rosenthal & Nolan, 2013), the sample was still homogenous based on race, education,

and other demographics. It's possible that parents with higher education may have been able to access and understand information related to their infant's decision making more easily than parents without such education. Furthermore, infant participants had a variety of congenital diagnoses, which precluded us from exploring the influence of specific features and severity of different congenital anomalies on study variables; however, we attempted to recruit mothers of infants with severe congenital anomalies, as defined by the March of Dimes, with the goal of capturing similar congenital anomaly experiences. It's possible that utilizing a decision checklist to identify mothers' decisions could have introduced decisions that mothers would not have reported on their own; however, the decision checklist was developed with input from neonatal specialists who support parents in their decision making processes in the NICU, which increased the likelihood that the decisions presented were representative. Finally, we recruited only English-speaking mothers, which means data from mothers of minority groups, such as Hispanics, where some congenital anomalies are more prevalent were not included.

Conclusions

While mothers of infants with a congenital anomaly receiving treatment in the NICU were quite similar in coping, parenting and decision making, despite the timing of their infant's congenital diagnosis, some differences were noted. This study further informs how to support mothers' adaptation following a congenital diagnosis. Effective health communication related to specific coping, parenting and decision-making needs or preferences of mothers engaged in congenital anomaly treatment decision making can facilitate shared decision making with the health care team and optimize infants' health outcomes.

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Table 1: Mother Participant Characteristics by Study Phase

	Quantitative Sample				Qualitative Subsample			
Variable	***Quantitative Sample (N= 37)*** **N (%)**	**Prenatal (n=23) n (%)**	**Postnatal (n=14) n (%)**	**p value**	**Qualitative Subsample (n=20) n (%)**	**Prenatal (n=12) n (%)**	**Postnatal (n=8) n (%)**	**p value**
Age (yrs), mean ± SD (range)	30.6 ±6.0 (19-40)	30.39 ± 6.7 (19-40)	31.00 ± 4.88 (20-38)	p=0.13[T]	31.25 ±5.73 (19-40)	30.42±6.74 (19-40)	32.50±3.85 (26-38)	p=0.143[T]
Race								
White	25 (67.6)	15 (65.2)	10 (71.4)	p=0.86[+]	14 (70.0)	10 (8330)	4 (50.0)	p= 0.188[+]
Black	10 (27.0)	7 (30.4)	3 (21.4)		5 (25.0)	2 (16.7)	3 (37.5)	
Other	2 (5.4)	1 (4.3)	1 (7.1)		1 (5.0)	0 (0.0)	1 (12.5)	
Ethnicity								
Hispanic	4 (11)	1 (4.3)	3 (21.4)	p=0.142[Λ]	2 (10.0)	1 (8.3)	1 (12.5)	p= 0.653[+]
Non-Hispanic	33 (89)	22 (95.7)	11 (78.6)		18 (90.0)	11 (91.7)	7 (87.5)	
Education*								
High school grad or less	6 (16.2)	4 (17.4)	2 (14.3)	p=0.016[+]	4 (20.0)	2 (16.7)	2 (25.0)	p= 0.176[+]
College grad or less	22 (59.4)	17 (73.9)	5 (35.7)		10 (50.0)	8 (66.7)	2 (25.0)	
Some grad school	9 (24.4)	2 (8.7)	7 (50)		6 (30.0)	2 (16.7)	4 (50.0)	
Income								
<$45,000	10 (27.0)	5 (21.7)	5 (35.7)	p=0.64[+]	6 (30.0)	4 (33.3)	2 (25.0)	p= 0.591[+]
>$45,000	22 (59.5)	14 (60.9)	8 (57.1)		13 (65.0)	8 (66.7)	5(62.5)	
Decline/don't know	5 (13.5)	4 (17.4)	1 (7.1)		1 (5.0)	0 (0.0)	1 (12.5)	
Employment								
Full-time	22 (59.5)	13 (56.5)	9 (64.3)	p=0.89[+]	11 (55.0)	5 (41.7)	6 (75.0)	p= 0.368[+]
Part-time	5 (13.5)	3 (13)	2 (14.3)		4 (20.0)	3 (25.0)	1 (12.5)	
Unemployed/stay at home parent	10 (27)	7 (30.4)	3 (21.4)		5 (25.0)	4 (33.3)	1 (12.5)	

	Quantitative Sample				Qualitative Subsample			
Variable	***Quantitative Sample (N= 37)*** **N (%)**	**Prenatal (n=23) n (%)**	**Postnatal (n=14) n (%)**	**p value**	**Qualitative Subsample (n=20) n (%)**	**Prenatal (n=12) n (%)**	**Postnatal (n=8) n (%)**	**p value**
Health Insurance								
Private	20 (54.1)	12 (52.2)	8 (57.1)	p=0.77$^{\lambda}$	10 (50.0)	5 (41.7)	5 (62.5)	p= 0.650^{+}
Medicaid/Other	17 (45.9)	11 (47.8)	6 (42.9)		10 (50.0)	7 (58.3)	3 (37.5)	
Partner status								
Married/partnered	32 (86.5)	19 (82.6)	13 (92.9)	p=0.78^{+}	18 (90.0)	11 (91.7)	7 (87.5)	p= 0.653^{+}
Divorced/Separated	1 (2.7)	1 (4.3)	0 (0.0)		0 (0.0)	0 (0.0)	0 (0.0)	
Single	4 (10.8)	3 (13)	1 (7.1)		2 (10.0)	1 (8.3)	1 (12.5)	
Religion/***								
Protestant	21 (56.8)	15 (65.2)	6 (42.9)	p=0.008^{+}	12 (60.0)	10 (83.3)	2 (25.0)	p= 0.034^{+}
Catholic	6 (16.2)	1 (4.3)	5 (35.7)		4 (20.0)	1 (8.3)	3 (37.5)	
Other	2 (5.4)	0 (0.0)	2 (14.3)		2 (10.0)	0 (0.0)	2 (25.0)	
None	8 (21.6)	7(30.4)	1 (7.1)		2 (10.0)	1 (8.3)	1 (12.5)	
Religious Importance								
Not important	3 (8.1)	3 (13.0)	0 (0.0)	p=0.293^{+}	1 (5.0)	1 (8.3)	0 (0.0)	p= 0.795^{+}
Somewhat	12 (32.4)	6 (26.1)	6 (42.9)		8 (40.0)	4 (33.3)	4 (50.0)	
Very	22 (59.5)	14 (60.9)	8 (57.1)		11 (55.0)	7 (58.3)	4 (50.0)	
Religious service attendance								
2x/year or less	18 (49)	10 (44)	8 (57)	p=0.42$^{\lambda}$	9 (45.0)	5 (41.7)	4 (50.0)	p= 0.535^{+}
1x/month or more	19 (51)	13 (56)	6 (43)		11 (55.0)	7 (58.3)	4 (50.0)	
# Other Children								
0	13 (35.2)	5(21.7)	8(57.1)	p=0.105^{+}	7 (35.0)	3 (25.0)	4 (50.0)	p= 0.594^{+}
1-2	14 (37.8)	11(47.8)	3(21.4)		4 (20.0)	3 (25.0)	1 (12.5)	
3 or more	10 (27)	7(30.4)	3(21.4)		9 (45.0)	6 (50.0)	3 (37.5)	

	Quantitative Sample				Qualitative Subsample			
Variable	***Quantitative Sample (N= 37)*** **N (%)**	**Prenatal (n=23) n (%)**	**Postnatal (n=14) n (%)**	**p value**	**Qualitative Subsample (n=20) n (%)**	**Prenatal (n=12) n (%)**	**Postnatal (n=8) n (%)**	**p value**
Other Children Hospitalized								
Yes	10 (27)	7(30.4)	3(21.4)	p= 0.71⁺	7 (35.0)	5 (41.7)	2 (25.0)	p= 0.642⁺
No	27 (73)	16(69.6)	11(78.6)		13 (65.0)	7 (58.3)	6 (75.0)	

+=Fisher's Exact; λ=Chi Square; U= Mann Whitney U; T=t-test

*Significant different between prenatal and postnatal groups, $p<0.05$

**Significant difference between prenatal and postnatal groups, $p<0.01$

Table 2: Infant Characteristics by Study Phase

	Quantitative Sample				Qualitative Subsample			
Variable	***Quantitative (N= 37) N (%)***	**Prenatal (n=23) n (%)**	**Postnatal (n=14) n (%)**	**P Value**	**Qualitative Subsample (n=20)**	**Prenatal (n=12)**	**Postnatal (n=8)**	**p value**
Age at birth, Mean,g ± SD (range) Median, IQR	36.12 ± 3.57 [37.1,0.9]	35.89 ± 3.43 [36.7,4.0]	36.49 ± 3.90 [37.4,4.4]	p= 0.56[u]	36.62 ±2.86 [37.2, 3.2]	36.77 ±1.63 [36.5, 2.7]	36.39 ±4.24 [37.9, 3.6]	p= 0.779[T]
≥ 37 weeks (full-term)	20 (54.1)	11 (47.8)	9 (64.3)	p= 0.330[ʌ]	11 (55.0)	8 (66.7)	4 (50.0)	p= 0.648[+]
< 37 weeks (preterm)	17 (45.9)	12 (52.2)	5 (35.7)		9 (45.0)	4 (33.3)	4 (50.0)	
Gender								
Male	17 (45.9)	9 (39.1)	8 (57.1)	p= 0.286[ʌ]	9 (45.0)	7 (58.3)	2 (25.0)	p= 0.197[+]
Female	20 (54.1)	14 (60.9)	6 (42.9)		11 (55.0)	5 (41.7)	6 (75.0)	
Gestational status								
Single birth	33 (89.2)	21 (91.3)	12 (85.7)	p= 0.630[+]	17 (85.0)	10 (83.3)	7 (87.5)	p= 0.656[+]
Multiple birth	4 (10.8)	2 (8.7)	2 (14.3)		3 (15.0)	2 (16.7)	1 (12.5)	
Anomaly Type								
Digestive	15 (40.5)	8 (34.8)	7 (50.0)		11 (55.0)	7 (58.3)	4 (50.0)	p= 0.103[+]
Heart	7 (18.9)	6 (26.1)	1 (7.1)	p= 0.113[+]	3 (15.0)	3 (25.0)	0 (0.0)	
Brain/Nervous System	7 (18.9)	5 (21.7)	2 (14.3)		2 (10.0)	1 (8.3)	1 (12.5)	
Musculoskeletal	5 (13.5)	1 (4.3)	4 (28.6)		3 (15.0)	0 (0.0)	3 (37.5)	
Diaphragm	3 (8.1)	3 (13.0)	0 (0.0)		1 (5.0)	1 (8.3)	0 (0.0)	
Birthweight								
Mean,g ± SD	2505 ±871	2362 ±812	2739 ±945	p= 0.210[T]	2568 ±176	2524 ±668	2635 ±987	p= 0.767[T]
Average birth weight	19 (51.4)	11 (47.8)	8 (57.1)	p= 0.883[+]	12 (60.0)	7 (58.3)	5 (62.5)	p= 0.764[+]
Low birth weight (<2500g)	13 (35.1)	8 (34.8)	5 (35.7)		6 (30.0)	4 (33.3)	2 (25.0)	

	Quantitative Sample				Qualitative Subsample			
Variable	***Quantitative (N= 37) N (%)***	**Prenatal (n=23) n (%)**	**Postnatal (n=14) n (%)**	**P Value**	**Qualitative Subsample (n=20)**	**Prenatal (n=12)**	**Postnatal (n=8)**	**p value**
VLBW (<1500g)	2 (5.4)	2 (8.7)	0 (0.0)		1 (5.0)	1 (8.3)	0 (0.0)	
ELBW (<1000g)	3 (8.1)	2 (8.7)	1 (7.1)		1 (5.0)	0 (0.0)	1 (12.5)	
Age at Interview, Days Mean ± SD (range) Median, IQR	14.1±9.7 (5-46) [12.0, 12.0]	12.00 ±6.91 (5-29) [11.0, 10.0]	17.43 ±12.68 (5-46) [14.0,19.0]	p= 0.210[u]	15.2 ±11.2 (5-46) [13.0, 15.0]	12.83 ±7.61 (5-29) [12.5, 10.0]	18.63 ±14.95 (5-46) [14.0, 26.0]	p= 0.266 [T]

+=Fisher's Exact; λ=Chi Square; U= Mann Whitney U; T=t-test

Table 3: Frequencies of Decisions Made by Mothers on Behalf of their Infant by Study Phase

	Quantitative Sample				Qualitative Subsample			
Parent Decision	**Quantitative Sample (N= 37) *n, %***	**Prenatal (n=23)**	**Postnatal (n=14)**	**p value**	**Qualitative Subsample (n=20) n, %**	**Prenatal (n=12)**	**Postnatal (n=8)**	**p value**
Surgery	21 (56.8)	12 (52.2)	9 (64.3)	p= 0.697+	13 (65.0)	8 (66.7)	5 (62.5)	p= 0.918+
Intermittent or Continuous Medications	7 (18.9)	4 (17.3)	3 (21.4)		3 (15.0)	2 (16.7)	1 (12.5)	
Blood	3 (8.1)	3 (13.0)	0 (0.0)		1 (5.0)	1 (8.3)	0 (0.0)	
Withdrawal or Withholding of Life-sustaining Treatment	3 (8.1)	2 (8.7)	1 (7.1)		2 (10.0)	1 (8.3)	1 (12.5)	
Evaluation by Specialist	1 (2.7)	1 (4.3)	0 (0.0)		0 (0.0)	0 (0.0)	0 (0.0)	
Radiology Testing	1 (2.7)	0 (0.0)	1 (7.1)		1 (5.0)	0 (0.0)	1 (12.5)	
Ventilator	1 (2.7)	1 (4.3)	0 (0.0)		0 (0.0)	0 (0.0)	0 (0.0)	

+=Fisher's Exact

Table 4: Parent Coping and Decision Making Findings by Study Phase and Prenatal vs. Postnatal Diagnosis

	Quantitative Sample					**Qualitative Subsample**			
Variable, N (%)	***Quantitative Sample (N= 37) N (%)***	**Prenatal (n= 23) n (%)**	**Postnatal (n=14) n (%)**	**p Value**	**Effect Size (r)**	**Qualitative Subsample (n=20)**	**Prenatal (n=12) n(%)**	**Postnatal (n=8) n(%)**	**p value**
Illness Perception (total) Mean ± SD (range)	54.1 ± 12.6 (27-81)	53.30 ±13.78 (27-72)	55.36 ±10.85 (39-81)	p= 0.64[T]	0.11[d]	54.4 ±13.3 (29-81)	53.08±13.84 (29-68)	56.25±13.20 (39-81)	p=0.616[T]
	Illness Perception Subscale Items (Mean±SD), [Median, IQR]								
IP1: Consequences	6.27 ±2.912 [6.0, 5.0]	6.39 ±3.1 [7.0, 6.0]	6.07 ±2.674 [5.0, 5.0]	p=0.739[u]	0.055	5.95 ±3.10 [5.0, 6.0]	6.00 ±3.10 [6.0, 6.0]	5.88 ±3.31 [5.0, 7.0]	p=0.932[u]
IP2: Timeline	5.86 ±3.63 [5.0, 8.0]	5.96 ±3.674 [6.0, 8.0]	5.71 ±3.688 [5.0, 8.0]	p= 0.834[u]	0.035	6.05 ±3.66 [6.0, 8.0]	6.83 ±3.54 [8.0, 7.0]	4.88 ±3.76 [4.0, 8.0]	p=0.200[u]
IP3: Personal Control	3.76 ±2.733 [3.0, 4.0]	3.61 ±2.675 [3.0, 4.0]	4.00 ±2.909 [4.0, 5.0]	p=0.747[u]	0.053	3.05 ±2.42 [2.5, 4.0]	3.08 ±2.23 [2.5, 4.0]	3.00 ±2.83 [2.0, 4.0]	p=0.745[u]
IP4: Treatment Control	9.14 ±1.946 [10.0, 1.0]	9.30 ±1.579 [10.0, 0]	8.86 ±2.476 [10.0, 2.0]	p=0.632[u]	0.079	7.77 ±2.40 [10.0, 2.0]	9.25 ±1.87 [10.0, 0]	8.38 ±3.11 [10.0, 2.0]	p=0.361[u]
IP5: Identity	5.68 ±2.759 [6.0, 5.0]	5.17 ±2.902 [5.0, 6.0]	6.50 ±2.378 [6.5, 3.0]	p=0.290[T]	0.503[d]	5.45 ±2.70 [5.5, 5.0]	4.75 ±2.86 [4.5, 6.0]	6.50 ±2.20 [6.5, 3.0]	p=0.162[T]
IP6: Concern	8.05 ±2.677 [10.0, 3.0]	7.52 ±3.058 [9.0, 6.0]	8.93 ±1.639 [10.0, 2.0]	p=0.172[u]	0.225	8.05, ±2.48 [9.5, 5.0]	7.58 ±2.78 [8.5, 6.0]	8.75 ±1.91 [10.0, 3.0]	p=0.321[u]
IP7: Coherence	8.51 ±1.574 [9.0, 3.0]	8.48 ±1.504 [9.0, 2.0]	8.57 ±1.742 [9.5, 3.0]	p=0.661[u]	0.072	8.25 ±1.68 [8.0, 3.0]	8.42 ±1.56 [8.5, 3.0]	8.00 ±1.93 [8.0, 4.0]	p=0.601[u]
IP8: Emotional	8.57 ±1.444 [9.0, 3.0]	8.48 ±1.534 [9.0, 2.0]	8.71 ±1.326 [9.0, 3.0]	p=0.770[u]	0.048	8.40 ±1.64 [9, 3]	8.08 ±1.78 [8.5, 3.0]	8.88 ±1.36 [9.5, 3.0]	p=0.320[u]
IP9: Serious	8.05 ±1.957 [8.0, 3.0]	8.17 ±2.059 [9.0, 3.0]	7.86 ±1.834 [7.5, 4.0]	p=0.521[u]	0.105	7.65 ±2.28 [8.0, 4.0]	7.58 ±2.50 [8.5, 4.0]	7.75 ±2.05 [7.5, 4.0]	p=0.878[u]

	Quantitative Sample					Qualitative Subsample			
Variable, N (%)	***Quantitative Sample (N= 37) N (%)***	**Prenatal (n= 23) n (%)**	**Postnatal (n=14) n (%)**	**p Value**	**Effect Size (r)**	**Qualitative Subsample (n=20)**	**Prenatal (n=12) n(%)**	**Postnatal (n=8) n(%)**	**p value**
	Positive General Coping subscales (Mean ±SD), [Median, IQR]								
Reframing	6.05 ± 1.7 [6.0, 3.0]	6.30 ± 1.52 [6.0, 2.0]	5.64 ± 2.06 [5.5, 3.0]	p= 0.32[u]	0.163	6.30 ±1.5 [6.0, 3.0]	6.33 ±1.50 [6.5, 3.0]	6.25 ±1.58 [6.0, 3.0]	p=0.906[T]
Planning	6.24 ± 1.6 [6.0, 3.0]	6.48 ± 1.38 [7.0, 2.0]	5.86 ± 1.96 [6.0,4.0]	p= 0.39[u]	0.140	6.35 ±1.8 [7.0, 3.0]	6.83 ±1.12 [7.0, 2.0]	5.63 ±2.39 [6.0, 5.0]	p=0.340[u]
Humor	2.46 ± 1.0 [2.0, 1.0]	2.26 ± 0.62 [2.0, 0]	2.79 ± 1.42 [2.0,1.0]	p= 0.189[u]	0.216	2.55 ±1.23 [2.0, 1.0]	2.08 ±0.289 [2.0, 0]	3.25 ±1.75 [2.5, 2.0]	p=0.029[u]
Acceptance	6.65 ± 1.2 [7.0, 3.0]	7.04 ± 1.15 [7.0,1.0]	6.00 ± 1.04 [6.0, 2.0]	p= 0.01[u]	0.444	6.65 ±1.2 [7.0, 3.0]	7.25 ±0.965 [7.5, 1.0]	5.75 ±0.886 [5.5, 2.0]	p=0.005[u]
Religion	6.11 ± 2.1 {7.0, 3.0]	6.13 ± 2.26 [7.0, 3.0]	6.07 ± 1.73 [6.5, 3.0]	p= 0.63 [u]	0.080	6 ±1.9 [6.0, 4.0]	6.25 ±1.87 [6.5, 3.0]	5.63 ±1.92 [5.5, 4.0]	p=0.477[T]
Active	6.27 ± 1.3 [6.0, 3.0]	6.17 ± 1.30 [6.0, 2.0]	6.43 ± 1.45 [7.0, 3.0]	p= 0.54[u]	0.100	6.20 ±1.4 [6.0,3.0]	6.08 ±1.24 [6.0, 2.0]	6.38 ±1.60 [6.5, 3.0]	p=0.651[T]
Emotional Support	7.08 ± 1.1 [8.0, 2.0]	6.96 ± 1.07 [7.0, 2.0]	7.29 ± 1.07 [8.0, 2.0]	p= 0.31[u]	0.166	7.10 ±1.1 [8.0, 2.0]	7.00 ±1.13 [7.5, 2.0]	7.25 ±1.17 [8.0, 2.0]	p=0.609[u]
Instrumental Support	6.22 ± 1.5 [6.0, 2.0]	6.30 ± 1.42 [6.0, 2.0]	6.07 ± 1.59 [6.5, 2.0]	p= 0.70[u]	0.063	6.00 ±1.6 [6.0, 2.0]	6.00 ±1.41 [6.0, 2.0]	6.00 ±1.85 [6.5, 2.0]	p=1.0[T]
	Negative General Coping Subscales (Mean ±SD), [Median, IQR]								
Self-distraction	4 ± 1.4 [4.0, 2.0]	3.78 ± 1.35 [4.0, 1.0]	4.21 ± 1.58 [4.0, 3.0]	p = 0.46[u]	0.121	3.8 ±1.0 [4.0, 1.0]	3.83 ±0.937 [4.0, 0]	3.75 ±1.17 [4.0, 1.0]	p=0.541[u]
Denial	3.22 ± 1.8 [2.0, 3.0]	3.22 ± 1.95 [2.0, 1.0]	3.21 ± 1.67 [2.0, 3.0]	p= 0.97[u]	0.006	3.10 ±1.6 [2.0, 3.0]	2.75 ±1.36 [2.0, 1]	3.63 ±1.92 [3.0, 3.0]	p=0.337 [u]
Substance Use	2.1 ± 0.5 [2.0, 0]	2.17 ± 0.58 [2.0, 0]	2.00 ± 0 [2.0, 0]	p= 0.23[u]	0.184	2.10 ±0.5 [2.0, 0]	2.17 ±0.577 [2.0, 0]	2.00 ±0.00 [2.0, 0]	p=0.414 [u]

	Quantitative Sample					Qualitative Subsample			
Variable, N (%)	***Quantitative Sample (N= 37) N (%)***	**Prenatal (n= 23) n (%)**	**Postnatal (n=14) n (%)**	**p Value**	**Effect Size (r)**	**Qualitative Subsample (n=20)**	**Prenatal (n=12) n(%)**	**Postnatal (n=8) n(%)**	**p value**
Behavioral Disengagement	2.05 ± 0.3 [2.0, 0]	2.09 ± .417 [2.0, 0]	2.00 ± 0 [2.0, 0]	p = 0.44[u]	0.128	2.00 ±0 [2.0, 0]	2.00 ±0.00 [2.0, 0]	2.00 ±0.00 [2.0, 0]	p=1.0 [u]
Venting	4.03 ± 1.6 [4.0, 2.0]	3.70 ± 1.36 [3.0, 2.0]	4.57 ± 1.87 [4.0, 3.0]	p= 0.15[u]	0.236	4.15 ±1.7 [4.0, 2.0]	3.58 ±1.24 [3.0, 2.0]	5.00 ±1.93 [4.5, 3.0]	p=0.060 [T]
Self-Blame	3.57 ± 1.5 [3.0, 3.0]	3.65 ± 1.53 [4.0, 2.0]	3.43 ± 1.51 [3.0, 3.0]	p= 0.66[u]	0.072	3.30 ±1.3 [3.0, 2.0]	3.08 ±1.08 [3.0, 2.0]	3.63 ±1.51 [3.0, 3.0]	p=0.424 [u]
Religious Coping									
Positive	19.7 ± 6.5 [19.0, 11.0]	20.61 ± 7.21 [23.0, 11.0]	18.21 ± 4.87 [17.5, 8.0]	p = 0.158[u]	0.232	19.15 ±5.5 [18.0, 10.0]	20.75 ±5.74 [21.5, 10.0]	16.75 ±4.46 [15.5, 8.0]	p=0.114[T]
Negative	9.5 ± 3.5 [8.0, 4.0]	9.30 ± 3.02 [8.0, 4.0]	9.93 ± 4.38 [8.0, 3.0]	p = 0.604[u]	0.085	9.4 ±3.1 [8.0, 4.0]	9.00 ±2.66 [8.0, 4.0]	10.00 ±3.74 [8.0, 6.0]	p=0.404[u]
Depression	9.35 ± 4.6	9.57 ± 4.73	9.00 ± 4.54	p = 0.72[T]	0.787[d]	8.45±4.9	7.92 ±4.87	9.25 ±5.12	p=0.564 [T]
	Control Preference Scale (CPS) in 2 categories (Engaged vs. Unengaged)								
Shared/Active	24 (64.8)	12 (52.2)	12 (85.7)	p= 0.074[+]		16 (80.0)	8(66.7)	8(100)	p=0.117[+]
Deferred	13 (35.1)	11 (47.8)	2 (14.3)			4 (20.0)	4(33.3)	0(0.00)	

+=Fisher's Exact; λ=Chi Square

U= Mann Whitney U; T=t-test, d=Cohen's D

Table 5: Spearman's Rank-ordered Correlations Comparing Illness Perception Subscale Items and Other Coping Variables for the Quantitative Sample

Variables	Illness Perception Consequences	Illness Perception Timeline	Illness Perception Personal Control	Illness Perception Treatment Control	Illness Perception Identity	Illness Perception Concern	Illness Coherence	Illness Perception Emotional Representation	Illness Perception Causal
Positive Coping Mechanisms									
Acceptance	0.223	0.261	-0.114	0.032	-0.182	-0.132	-0.103	-0.227	-0.033
Emotional Support	0.024	0.213	-0.285	-0.094	0.042	0.331*	0.434**	0.006	0.140
Humor	0.195	-0.138	0.074	-0.175	0.189	-0.186	-0.068	-0.150	-0.166
Reframing	0.195	0.085	0.025	-0.085	-0.056	-0.166	0.224	-0.252	-0.099
Religion	0.014	0.179	0.279	0.164	0.147	0.080	0.369*	0.160	0.065
Active Coping	0.241	0.028	0.218	0.141	0.142	-0.066	0.005	-0.086	-0.114
Instrumental Support	0.304	0.282	-0.098	0.036	0.023	0.215	0.411*	0.130	-0.103
Planning	0.383*	0.304	-0.269	0.009	0.139	0.296	0.193	0.231	0.167
Positive Religious Coping	-0.053	0.146	0.262	0.124	0.130	0.142	0.462**	0.175	0.019
Negative Coping Mechanisms									
Behavioral Disengagement	0.087	0.185	0.104	-0.332*	0.157	0.151	-0.194	0.187	0.056
Denial	0.232	0.045	0.144	-0.247	0.410*	0.269	0.213	0.393*	0.100
Self-distraction	0.097	-0.46	0.104	0.128	0.009	-0.036	-0110	0.024	-0.090
Self-blame	0.231	0.132	-0.158	-0.071	0.320	0.244	0.067	0.377*	0.210
Substance Use	0.187	0.173	-0.023	-0.431**	-0.068	0.084	0.267	-0.134	0.011
Venting	0.277	0.297	-0.158	-0.246	-0.357	0.429**	0.126	0.307	0.310
Negative Religious Coping	0.071	0.111	-0.437**	-0.136	0.109	0.129	0.136	0.149	0.050

Depression	0.044	0.005	-0.227	0.020	0.073	0.311	0.181	0.348*	0.140

*Significant different between prenatal and postnatal groups, $p<0.05$

**Significant difference between prenatal and postnatal groups, $p<0.01$

Table 6: Relative Ranking and Importance of Good Parent Attributes for Children with Congenital Anomalies by Pre vs. Postnatal Diagnosis

Relative Ranking	Good parent attribute: Quantitative Sample (n=37)	Relative Importance	Good parent attribute: Prenatal (n=23)	Relative Importance	Good parent attribute: Postnatal (n=14)	Relative Importance
1	Putting Child's needs above my own when making medical decisions	16.1	Putting my child's needs above my own when making medical care decisions	17	Focusing on my child's health	15.8
2	Focusing on my child's health	15.9	Focusing on my child's health	15.7	Putting my child's needs above my own when making medical care decisions	14.7
3	Making sure my child feels loved	12.6	Making sure my child feels loved	12.0	Focusing on my child's quality of life	13.6
4	Making informed medical care decisions	11.5	Making informed medical care decisions	10.7	Making sure my child feels loved	13.4
5	Focusing on my child's quality of life	10.0	Advocating for my child with medical staff	9.0	Making informed medical care decisions	13.1
6	Advocating for my child with medical staff	9.1	Focusing on my child's quality of life	8.3	Advocating for my child with medical staff	8.9
7	Focusing on my child's comfort	7.5	Focusing on my child's comfort	7.7	Focusing on my child's comfort	7.2
8	Focusing on my child having as long a life as possible	7.1	Focusing on my child having as long a life as possible	7.1	Focusing on my child having as long a life as possible	7.1
9	Staying at my child's side	5.4	Staying at my child's side	6.3	Staying at my child's side	3.6
10	Keeping a positive outlook	1.9	Keeping a positive outlook	2.4	Keeping a realistic outlook	1.3
11	Focusing on my child's spiritual well-being	1.5	Focusing on my child's spiritual well-being	2.4	Keeping a positive outlook	1.1
12	Keeping a realistic outlook	1.4	Keeping a realistic outlook	1.5	Focusing on my child's spiritual well-being	0.12

Table 7: Qualitative Findings of Coping, Decision Making, and Parenting Barriers and Facilitators

		Barriers		Facilitators	
		Prenatal	Postnatal	Prenatal	Postnatal
		1) Access—The ability to reach or approach, or the financial, material, or service aspects of, healthcare.			
Contextual-related Factors	Coping	• Distance from home to hospital (4) • Economic needs during NICU stay (2)			
	Parenting	• Parking Expenses (1)	• Parking Expenses (1)	Staffs' assistance with obtaining care supplies & completing needed services for infant (3)	
		2) Communication—The exchange of information between individuals or groups.			
	Coping	• Delays in NICU procedures (1) • Poor health care team communication (1)	• Poor health care team communication (2) Unexpected/missed diagnosis (2)		
	Decision Making	• Different description of decision making before vs. after birth by medical team (1) • Insufficient prep for decisions that would need to be made after birth (1)	• Conflicting information about decision (1) • Misunderstanding medical language (1)		

		Barriers		Facilitators	
		Prenatal	Postnatal	Prenatal	Postnatal
	Parenting		• Unclear understanding of health care team members' roles (1)	Clear/responsive communication from health care team (7)	
		3) Environment-The physical surroundings impacting an individual.			
	Parenting	• Non-conducive NICU accommodations (2) • Technology interference (2) NICU routine mismatch with parenting style (1)	• Non-conducive NICU accommodations (2)	• Privacy of patient rooms (2) Proximity of family lounge for eating (1)	
		4) Relational-Describes the way in which persons are connected.			
Contextual-related Factors	Coping	• Separation from family/friend support system (2) • Separation from NICU Infant (1) Unwelcome advice from family (1)	• Separation from NICU infant (3)	• Emotional support from family/friends (15) • Emotional support from health care team (9) • Instrumental support through community fundraisers (3) Informational/Emotional support from other NICU families (2)	• Emotional support from family/friends (6) • Emotional/informational support from health care team (5) Informational/Emotional support from other NICU Families (1)
	Decision Making	• Uncertainty of whether to trust doctors to recommend appropriate decision options (1) • Physicians limiting time allowed to make decision (1)	• Deciding against other family members' preferences (1)	• Family decisional support (4) • Trust in medical team (4) • Trust in facility's reputation (3)	• Family decisional support (3) • Trust in facility's reputation (1) • Trust in health care team (2)

		Barriers		Facilitators	
		Prenatal	Postnatal	Prenatal	Postnatal
	Parenting	• Others questioning mothers' choices/intuition (2) • Struggle to find role as NICU parent (1) • Involuntary revoking of parenting duties (5) • Struggle to balance home & NICU parenting (2)	• Struggle to balancing home & NICU parenting (3) • Involuntary revoking of parenting duties (2) • Struggle to find role as NICU parent (3) • Struggle to balance personal recovery & NICU parenting (1)	• Trust in health care providers (3) • Practical support from family (1)	• Emotional support from staff (5) • Staff advocacy for parents/infant (1) • Emotional/practical support from family (1)
		1) Behavioral-Describes the way in which one acts or conducts oneself.			
Person-related Factors	Coping			• Encouraging/Helping other NICU families (3) • Listening to Music (1) • Involvement in care of NICU infant (4) • Expressing breast milk for NICU infant (2) • Psychotherapy (1)	• Involvement in care of NICU infant (3) • Expressing breast milk for NICU infant (1) • Taking breaks in NICU family lounge • Psychotherapy (1) • Humor (1)
Person-related Factors	Parenting			• Breastfeeding/pumping (5) • Mother's involvement in NICU care (4)	• Mother's involvement in NICU care (4)
		2) Cognitive-Describes the mental action or process of acquiring knowledge or understanding.			

		Barriers		Facilitators	
		Prenatal	Postnatal	Prenatal	Postnatal
	Coping			• Disease education received from health care team (1)	• Disease education by health care team (1) • Mothers' professional knowledge of health care field (1)
	Decision Making	• Information about potential negative side effects (1)	• Information about risk of complications (1) • Lack of pertinent information needed to make decision (3) "Worst case" information from internet	• Information/education from health care team about procedure (3) • Online information (1) • Foreknowledge of decision (1) • Parents' intuition (2)	• Information/education from health care team about procedure (4) • Relevant internet information (1) • Information from other NICU families (1) • Information about unattractive alternatives (1)
	Parenting			• Teaching/coaching from health care team on parenting a NICU infant (3)	Being informed by health care team (2)
		3) Emotional-Describes instincts or feelings deriving from one's mood, circumstances, or relationships with others.			
	Coping	• Mothers' self blame/depression related to infant's anomaly (2) • Guilt over leaving infant & not being one to provide care (2) • Fear of infant death (1)	• Emotional upset of mother over NICU hospitalization (1) • Guilt over leaving infant (2)	• Mothers' use of optimism (1)	

		Barriers		Facilitators	
		Prenatal	Postnatal	Prenatal	Postnatal
Person-related Factors	Decision Making	• Uncertainty about outcome (1)	• Concern related to prior negative health care experiences (1)	• Hope for longer life based on previous positive decision making experience (1)	• Certainty based on previous decision making experience (1)
		4) Spiritual—Describes how one derives meaning of individual experiences through dimensions of connectedness.			
	Coping	• Feeling tested by God (1)	• Questioning God (1)	• Belief in God/Higher Power, prayer (12) • Spiritual support from church/chaplain (8) • Serving as spiritual examples to community during infant's illness (2) • Singing spiritual songs (1)	• Belief in God/Higher Power, prayer (7) • Spiritual support from church/chaplain (2) • Spiritual support from friends/social media (4) • Singing spiritual songs (1)
	Decision Making			• Prayer/Trust in God (4) • Clergy support (1)	

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Table 8: Mixed Methods Integration of Coping, Decision Making, and Parenting Findings

Survey Data	Interview Data
Decision Making Findings	
1. Prenatal decision making engagement (52%) was lower than Postnatal decision making engagement (86%), although not statistically significant (Fisher's Exact p=0.07)	**Interviews clarified that mothers in both the prenatal and postnatal groups were engaged in decision making, but at different time points.** **Mothers in the prenatal group were engaged in decision making before birth, choosing to whom and to what facility they would entrust the care of their infant at birth, and then relinquished decision making to the health care team once in the NICU**. • *Um, as far as the decision for surgery, I think that it was premade before we got here—because we did understand um, what we were up against and had about 15 to 18 weeks to make the decision. Um, so we spent that time educating ourselves and researching the defect that we're up against. And decided on who was the best. And then after we made that decision, have that confidence level in that surgeon, you know, we were taking their lead. We already decided that this was the right team for the job, and the right facility for the job. So whatever they recommended, really, we just—our only decision was we're gonna go with whatever they recommend. (Participant 1004-Prenatal)* **Mothers in the postnatal group described being engaged in decision making in the NICU, at the time they learned of their infant's congenital diagnosis.** • Interviewer: So it's really important for you to stay in the loop with the medical team and understand not only what's going on with your daughter, but know what the plan is? Mother: Absolutely, and that's been the hardest part with the specialist, that we have not been kept in the loop… I just gave birth to this child almost five days ago, and we don't even know what's going on… (Participant 1007-Postnatal)
General Coping Findings	
1. Mothers in the quantitative sample most frequently identified positive forms of coping, rather than negative forms of coping	**Mothers in the qualitative sample named examples of reframing, planning, acceptance, religion, active coping, emotional support and instrumental support, all positive forms of coping.**

Survey Data	Interview Data
	• *So I didn't plan on breastfeeding with him. Um, I said I would never do that. I just felt really weird towards it. [Laughter] Um, but after talking to the doctors, he's gonna have, like, a really hard time eating. And, um, the breast milk—well, how it is, like, his, um, like, his body will take to the breast milk way easier and better, um, than formula. So right away I was like, "Oh, yeah, I'm—I'm breastfeeding." And, um, now that, you know, I had to watch him lay here like this, and I—I feel so helpless because, like, again, I can't—with my other baby, I could do everything. And I can't do anything with him. Um, I actually, like, feel good breast—like, it makes me—And I just keep up on it really good. And, um, it makes me, like—it just makes me feel so good because they said it's like medicine for him.—it's, like, the one thing that I have that I can do for him, you know? (Participant 1023-Prenatal)*
2. In the quantitative sample, mothers in the prenatal group utilized more acceptance coping (7.04±1.15) than mothers in the postnatal group (6.00±1.04), (U=78; p=0.01).	**In the qualitative sample, mothers in the prenatal group indicated they had time to consider the probable treatment option(s) for their infant before delivering, which seemed to contribute to their acceptance of the infant's treatment.** • *Um, our other—you know, the other place we were originally gonna get treatment said a lot of—we won't know until he get—till the baby gets here. You know, we'll have to decide then. You'll just have to wait. They said it here, too. But they said we won't know until the baby gets here, but if it's this then we do this. And if it's this, then they do that. They had plans. They had experience. So by the time he was born, you know, they came in, they talked to us so much, they—it wasn't even like—it was like do you have questions? (Participant 1004-Prenatal)* • *And, um, you know, surgery's always a big word. Um, so—and we had just kind of—we were really prepared for that, cuz they told us that, a couple months ago, that that's what she probably needs. (Participant 1006-Prenatal)*
3. In the quantitative sample, mothers' acceptance coping was significantly correlated with # of other children (0.334; p=<0.05) and reframing (0.522; p=<0.01)	**Mothers in the qualitative sample provided examples of acceptance through reframing.** • *I was like blamin' myself. I don't know what was goin' on. Cuz—maybe I hadn't drunk enough milk or maybe I hadn't done this—and done that. But, um, at the end of the day, I just really had to learn that, you know, it was nothin' I could really do to prevent the situation, so—I was chosen. [Laughter] I had to just, you know, be strong and get my testimony out of this one. (Participant 1008)*
4. In the quantitative sample, mothers'	**Mothers in the qualitative sample described their perception of their NICU infant's size when**

Survey Data	Interview Data
perception of their infant's illness severity was significantly correlated with infant's birth weight (r=-0.347; p=0.035)	**discussing the baby's illness.** • *We also had to worry about him being small, him tryin' to eat, and all that stuff that come with the preemie life that—that took center stage for a little bit. (Participant 1029)*
Religious Coping Findings	
1. Mothers in the quantitative sample identified more positive (19.7 ±6.5) than negative forms (9.5 ±3.5) of religious coping & 92% of mothers reported religion as somewhat or very important	**Two mothers in the qualitative sample described questioning or feeling tested by God, but all other examples of spiritual coping were positive.** • *I feel like, I mean, when things go wrong—um, you know, there—uh, there was a phrase that my mom always said. She said that um, God won't give you more than you can handle. And so, when things start to go wrong, I'm like [sigh], there has to be a reason. Like, you know, I know that I'm strong enough to get through this. Like this—like the situation with having to be here in the NICU. Like, if we couldn't do it, then we wouldn't have been given this. (Participant 1026)*
2. Negative religious coping was significantly correlated with self blame (0.608, p=<0.05) and depression (0.549, p=<0.01) in the quantitative sample	**Mothers in the qualitative sample who used negative spiritual coping described negative spiritual coping in terms of questioning or feeling tested by God.** • *I mean, sometimes I do question it. I'm like, "Really, dude? You can't give me a break?" I mean, come on. Four kids in the NICU and then all of this on top of it? I'm like, [sighs]. I'm like, "Okay, whatever." [Laughter] I'm like, "You're testing me, but—Yeah. And I'm like, "I'm doin' it, but, I mean—" And I know they said—they say He only gives you as much as you can handle. I'm like, "Dude, you must think I'm, like, really strong, but no. But, like, you're testing me. That's what you're doin'." [Laughter] (Participant 1029)*
3. Positive religious coping was significantly correlated with denial (0.337, p=<0.05) and substance use (0.326, p=<0.05)	**One mother in the qualitative sample described the role of spiritual coping in her addiction recovery process.** • *Um, I'm a recovering heroin addict. From being a recovering addict and going through, like, the Twelve Steps, Um, you know, they—God's in there a lot. And, um, I started goin' to church a lot and stuff like that. And, um, ever since I've, like, welcomed God into my life and been praying and, um, all that, it definitely, like, helps. Um, so I don't know. I just—I believe in God. I feel like prayers help, and I pray for my son a lot. (Participant 1023)*

Survey Data	Interview Data
Depression Findings	
1. Depression was significantly correlated with instrumental support (0.364, p=<0.05), denial (0.359, p=<0.05), self blame (0.802, p=<0.01), venting (0.330, p<0.05), and negative religious coping (0.549, p=<0.01) in the quantitative sample	**One mother in the qualitative sample described her experience with depression and denial following the diagnosis of her infant in utero.** • *Participant: [T]hey confirmed that it was CDH, because they could see that she had something wrong with her diaphragm. And that organs that originally in your abdominal area were in her chest cavity. And that it wasn't a heart defect. So then I broke into tears when they told me that she had a 50/50 chance of surviving. And it was extremely hard. I couldn't even make it out of that room without bawling my eyes out and almost falling to my knees.* *Interviewer: That sounds very hard. [pause] What did you do after that?* *Participant: Tried to cope. Tried to deal with all of it. First I was in denial, and then I went to a really high depression stage. I've always had some sort of issue with depression…I cried a lot. Listened to a lot of music because music's always been a big coping skill for me. I would read a lot of books, books have always been my thing. And that's about it. Other than playing, unfortunately, very violent videogames…Up until probably 2:00 or 3:00 in the morning every night. But I guess I was trying to get myself to not sleep, cuz I had a lot of nightmares after the first week of knowing it was something that I was very, very frightened of. I was afraid I was gonna lose her. (Participant 1017)*
Parenting Findings	
1. Mothers in the quantitative sample identified the top 2 parenting values as "Putting Child's needs above my own when making medical decisions" and "Focusing on my child's health"	**Mothers in the qualitative sample offered examples of what it meant to put their child's needs above their own when making medical decisions and focusing on their child's health.** • *I think it was more of um, you know, in the beginning, doing so much of the educating, so—cuz we've been confident here as far as like um, thinking we're in the right place and knowing everything. But I think just being a good parent to me was the fact that we—I will—I would never sound like I'm throwing this up anyway, or like putting it up there, like we have uprooted our whole lives cuz we felt like, you know—45 minutes away was not good enough. Like we wanted, you know, we made major life changes and decisions [to come to this facility].*

Survey Data	Interview Data
	(Participant 1004)
2. In the quantitative sample, mothers in the postnatal group ranked "focusing on my child's quality of life" 3rd, whereas mothers in the prenatal group ranked this value 6th.	**In the qualitative sample, mothers in the postnatal group described choosing treatments that would allow infants to get better and live with fewer devices/technology.** • *Interviewer: Yeah. So you had a choice of either doing the jaw distraction surgery or letting her go to another hospital where they would do the trach? And you thought the trach was better? Participant: Right. She [i.e. my daughter who died] had to have, um, a breathing tube and it wouldn't go all the way down to her stomach or somethin', so they was thinkin' about doing a trach and I don't think it was- I don't- I don't like the trach. My son [i.e. who died] had the trach. I don't like it. The other hospital just goin' give her a trach and they can do the surgery to make it better, so when they told me they can fix it in less than a week, I was okay. Somethin' goin' on instead of just lettin' her lay there with the stuff all in her. (Participant 1024-Postnatal*)

CHAPTER FIVE: Discussion

Introduction

Congenital anomalies account for 1 in 5 infant deaths in the U.S. and contribute substantially to childhood disability (Canfield et al., 2014). Inpatient care in the neonatal intensive care unit is often necessary for infants born with a congenital diagnosis (Russo & Elixhauser, 2007). This is the first known study to explore mothers' coping, parenting, and decision-making response when addressing their infant's congenital anomaly treatment in the NICU. Based on previous studies, we sought to explore whether the timing of receiving a congenital diagnosis (prenatal vs. postnatal), and other infant or parent factors, influence mothers' coping, parenting, and decision making for their infant who required treatment for a congenital anomaly in the NICU context. We utilized a cross-sectional, sequential explanatory mixed methods design, guided by the Roy Adaptation Model.

A total sample of 37 mothers of NICU infants with a congenital anomaly participated in the study. A purposive sample of 20 mothers was interviewed for the qualitative phase of the study. The three specific aims of the study were:

Quantitative Aim: 1. To estimate differences and associations of coping factors (infant illness severity perception; general coping; religious coping), parenting values (i.e. "good parent" attributes), and decision-making engagement for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth.

Qualitative Aim: 2. Explore perceptions of barriers and facilitators related to coping, parenting, and decision making for mothers of NICU infants with a congenital anomaly.

Mixed Aim: 3. To describe and compare the processes and barriers/facilitators of coping, parenting, and decision making for mothers of NICU infants diagnosed with a congenital anomaly before versus after birth.

By examining key variables related to coping, parenting, and decision making, we have identified several key findings.

Summary of Findings

Decision making engagement for prenatal vs. postnatal groups

The findings of this research illuminate the decision-making processes of mothers who learned of their infant's diagnosis prenatally vs. postnatally. Quantitative findings suggested that mothers who received a prenatal diagnosis were less engaged in decision making than mothers who received a postnatal diagnosis. However, our demographic findings revealed that the postnatal diagnosis group had significantly higher education and significantly more mothers identifying a religious affiliation. These demographic variables were positively correlated with decision-making engagement, so this correlation could confound our quantitative decision-making findings. The qualitative interviews further clarified our decision-making findings. Many mothers who received a prenatal diagnosis were engaged in decision making before their infant's birth, seeking specialized facilities and providers who could adequately address their infant's health care needs. Deciding on an expert provider at a reputable hospital and establishing a trusted relationship with this provider led many mothers to feel comfortable deferring subsequent decisions related to their infant's congenital diagnosis treatment once NICU admission occurred. On the other hand, mothers who received a postnatal diagnosis were more

actively involved in decision making in the NICU, when they first learned of the congenital diagnosis, infrequently relinquishing decision-making to the health care team.

Mothers reported utilizing more positive than negative coping mechanisms

Overall, mothers in our sample reported utilizing more positive than negative coping techniques, including emotional support, acceptance, active coping, planning, instrumental support, religion, and reframing. Additionally, the mean depression scores reported by mothers in our sample (9.35 ±4.6) were well below the cutoff (>13) used to indicate depressive symptomatology on the Edinburgh Postnatal Depression Scale. Mothers in the prenatal group reported utilizing more acceptance coping (7.04 ±1.15), compared to mothers in the postnatal diagnosis group (6.00 ±1.04), (U=78; p=0.01). Mothers also reported utilizing more positive religious coping (19.7 ±6.5) compared to negative religious coping (9.5 ±3.5) and 92% of mothers rated religion as somewhat or very important. Some mothers did report utilizing negative religious coping, namely questioning or feeling tested by God. Negative religious coping was significantly correlated with both self blame (r_s=0.608, p=<0.05) and depression (r_s=0.549, p=<0.01). However, overall these findings suggest that most mothers were unhindered by their coping processes in their decision making for their infant.

Prioritization of parenting values related to health

Our sample prioritized "putting my child's needs above my own when making medical decisions" and "focusing on my child's health" as the top two "good parenting" values when making decisions for their infant with a congenital anomaly. These values represent mothers' perception that illness management is a key parenting responsibility and are consistent with findings in other pediatric studies measuring good parent

attributes in parents of children with critical illnesses (Feudtner et al., 2015; Mooney-Doyle et al., 2018; October et al., 2014). Mothers prioritized "keeping a realistic outlook" least, among the parenting value options. Since mothers in this study described receiving a great deal of new information about treatment and prognosis, in addition to managing many emotions, they may not have been able to determine what a "realistic" outlook was for their infant.

Strengths & Limitations

This study did have several limitations. The utilization of convenience sampling to obtain participants from a single NICU limits the generalizability of study findings. In addition, the cross-sectional design did not allow for causal examination of relationships between study variables, nor how study variables may have changed over time. Because this study was a beginning effort in exploring the influence of congenital anomaly diagnosis timing on parents' adaptation to a NICU admission for the purposes of treating a congenital anomaly, we limited our participants to mothers, who were generally available for recruitment. Therefore, we did not include data from fathers. Comparisons between mothers' and fathers' coping, parenting, and decision-making regarding congenitally-related treatments performed in the NICU is important and warranted in the future. We limited recruitment to English-speaking parents, which means we did not capture data from some ethnic minorities, who have higher rates of certain congenital anomalies (Canfield et al., 2014). The use of self-report measures introduced the potential for self-report bias and social desirability bias. Additionally use of non-probability sampling introduced self-selection bias. Finally, in order to obtain a reasonable sample size, we collected data on several types of anomalies, not one specific

type (i.e. cardiac), which prevents us from drawing disease-specific conclusions or recommendations from our findings.

Despite these limitations, this study had several strengths. First, while our sample size was small, reporting our data is important because of the limited existing studies, to date, focused on the NICU congenital anomaly population. We have provided an important contribution to the understanding of parent adaptation to a NICU admission, as it was occurring, for the purposes of an infant's congenital anomaly treatment. We utilized a mixed methods design and the Roy Adaptation Model guided variable selection. The quantitative findings, though exploratory, highlighted important comparisons in coping, parenting, and decision making for mothers who received a prenatal versus postnatal congenital diagnosis for their infant. Qualitative interviews further explained quantitative findings, clarifying mothers' coping, parenting, and decision making following real-life decision making instances in the NICU.

Practice Implications

The results of this study have implications for providers in the NICU who manage the care of infants with congenital anomalies after birth. Understanding the trends in NICU decision-making engagement for mothers who have received a prenatal vs. postnatal diagnosis offers providers insight into how to approach communication based on this contextual factor. Interventions that enhance parent-provider communication, such as the parental shared decision-making preferences (PSDM) instrument that researchers at Children's Hospital of Philadelphia are developing, holds great promise for assessing decision-making barriers and identifying facilitators for individual parents (Children's Hospital of Philadelphia Research Institute., 2018). In addition, the stress of a

NICU admission can cause negative changes to a parent's affect, which in turn decreases the parent's ability to engage and process information during decision making for their child (Estrada, Isen, & Young, 1997; Verkuil, Brosschot, Meerman, & Thayer, 2012). Use of clear communication that is reinforced several times is helpful when discussing congenital anomaly treatments in the NICU (Drotar, Baskiewicz, Irvin, Kennell, & Klaus, 1975).

Emotions of anger, sadness, and anxiety in parents of infants with congenital diagnoses have long been documented, especially immediately following diagnosis (Drotar et al., 1975). Some of the mothers in our study reported their experience with these emotions. It's important for providers to understand that these emotions are not pathological, but a normal process of parental grieving. Such an understanding can guide providers' in offering their support to parents during particularly difficult times. Emotional and instrumental support by nurses was particularly helpful to mothers in this study. Other forms of provider support may include connecting parents to support groups (Drotar et al., 1975). Parents in our study verbalized interest in support groups, and previous qualitative studies confirm that parent-to-parent peer support can facilitate a shared social identity, mutual learning from one another, and personal growth. Parent-to-parent peer support also offers parents an opportunity to support others (Shilling et al., 2013). Our findings also revealed that mothers were relieved after birth when anomalies were less severe than originally predicted, which may have influenced which mothers decided to participate. Regardless, providers' focus of conversations around infants' normal physical attributes has been shown to facilitate positive coping for parents of infants with a congenital anomaly (Drotar et al., 1975). Finally, nurses may be

instrumental in identifying mothers' negative religious coping, which we found to contribute to negative coping. In the acute care setting, nurses spend the most time at the bedside with families and through conversation can identify when mothers report interpreting a stressor as punishment from God. Nurses' awareness of how to respond when negative religious coping is identified is important and may include making referrals to spiritual care specialists, such as hospital chaplains.

Because mothers in this study who received a prenatal diagnosis for their infant reported that having time to research both the congenital anomaly and potential providers or treatment facilities was helpful, a prenatal consultation with a neonatologist can be beneficial for providing parents knowledge, individualized care, and time to prepare for the future (Miquel-Verges et al., 2009). Prenatal counseling, both genetic and non-genetic, may also be beneficial, offering parents informational resources and support group contacts, both of which may reduce anxiety (Marokakis, Kasparian, & Kennedy, 2016).

While the concepts of religion and spirituality have been discussed in relation to congenital anomalies, in terms of influencing mothers' continuation or termination of pregnancy, our study revealed that many mothers utilize religious or spiritual beliefs to guide decision making or cope with congenital anomaly treatments upon their infant's admission into the NICU. Mothers are frequently asked about their religious affiliation upon hospital admission, but other spiritual assessment questions are infrequently addressed in the inpatient setting, perhaps because the Joint Commission: Accreditation, Health Care Certification (JCAHO) does not stipulate what should be included in a spiritual assessment (The Joint Commission, 2018). Our findings highlight the

importance of thorough spiritual assessment of mothers by providers in the NICU to guide provider-parent communication, health care decision making, and parent support.

Many NICUs that are capable of treating infants with a congenital anomaly are urban, academic medical centers, which draw patients from a broad catchment area. Consequently, many infants receiving inpatient care for a congenital anomaly and their families live some distance from the treating hospital. In our study, nearly 25% of all infants and their families lived out of the state and 50% lived 60 miles or more from where they were receiving treatment. Interventions that facilitate parents' connection to their infant, especially when treatments require lengthy hospital stays during which parental visitation might be limited, are beneficial. Use of technologies, such as videoconferencing, videophones, and commercially available modalities, such as Skype, show promise in terms of keeping parents connected during their child's hospitalization (Epstein et al., 2017). However, along with the use of these interventions come concerns about patient privacy and data security, which require input from the research ethics and regulatory realms (Epstein et al., 2017).

Policy Implications

Given that the cause of more than 70% of congenital anomalies remains undiscovered, the commitment of Congressional leaders at both the federal and state levels is needed in order to support research to determine genetic and non-genetic origins of congenital conditions. For those congenital anomalies that cannot currently be prevented, consistency in health insurance coverage policies for antepartum fetal surveillance is imperative. In addition, continuation of health care coverage policies that

allow infants with congenital anomalies to obtain health insurance despite having a pre-existing condition are necessary for optimizing child health outcomes.

Research Implications

This study points to several opportunities for future research, which can inform our understanding of parents who are faced with NICU treatment decision making for an infant with a congenital anomaly. While our study focused on mothers, there is a need for research capturing fathers' experiences. Recruitment of fathers in NICU studies is historically low, as little as 6%-37% (Moro, Kavanaugh, Okuno-Jones, & Vankleef, 2006; Ward, 2005). Given the high-risk nature of the congenital anomaly population, recruitment of fathers may be challenging, but is important so that we have an understanding of the coping, parenting, and decision-making response of both parents.

Research that explores study variables in specific congenital anomaly populations and racial/ethnic minority populations is also needed. Congenital anomalies disproportionately affect certain racial/ethnic minorities. For example, Hispanics have higher rates of anencephaly, encephalocele, and anotia/microtia (Canfield et al., 2014). There is a gap in the literature regarding these minority groups. In addition, understanding the needs of parents of specific congenital anomaly groups (i.e. cardiac, digestive, etc.) is important so that we can design interventions to meet their unique needs. Multisite studies are likely necessary in order to capture large enough samples to explore coping, parenting, and decision-making variables quantitatively. While this dissertation obtained IRB approval at 3 total recruitment sites, study team members successfully recruited participants at only one study site. The two study sites where no participants were recruited are historically non-research intensive institutions, which may

mean staff at these institutions was less familiar or experienced with recruitment and/or the research process. Utilizing creative and varied recruitment strategies in future studies may yield samples that allow more in-depth quantitative analyses.

The role of health literacy in parent-provider communication about congenital anomalies and potential treatments is another important area of research. Low health literacy has been shown to inhibit shared decision making with providers, especially if individuals have difficult with written clinical materials, such as informed consents or information sheets that provide disease or treatment-specific explanations (N. Cox, Bowmer, & Ring, 2011). While health literacy has been explored in some pediatric outpatient contexts, health literacy is underexplored in inpatient pediatric settings (Keim-Malpass, Letzkus, & Kennedy, 2015).

Finally, the NICU is only one setting where parents of children with congenital anomalies face treatment decision making. Infants who survive beyond the NICU with a congenital anomaly face potential life-long disability. Parents of these surviving infants potentially face multiple treatment decision points as the infant develops. Further research that explores parents' coping, parenting and decision making at other decision points is justified in order to intervene with appropriate interventions at critical points to maximize both child and parent health outcomes.

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APPENDIX A: Study Instruments

Participant ID _____ Interviewer Initials_____ Date _____

Decision Checklist

Background: Parents who have an infant with a condition that develops before birth are frequently involved in making decisions with the health care team after their child is admitted into the neonatal intensive care unit (NICU). Below is a list of common medical and treatment decisions for infants who have conditions that develop before birth.

Directions: Please place a check mark (✔) in front of the decision(s) you have been involved in, or have helped to make, since your child was admitted into the NICU.

Place a (✔) next to decisions you have made or helped to make	**Decision**
	Giving infant medications (i.e. intermittent medications, such as antibiotics)
	Giving infant medications (i.e. continuous medications, such as vasopressors/nitric oxide)
	Placing infant on oxygen
	Placing infant on ventilator
	Placing a central line
	Giving infant blood/blood-like products
	Laboratory testing
	Radiology testing
	Evaluation by a specialist
	Surgery (Type _____)
	Other (Name_____)
	I have let/asked someone else to make decisions for my infant (i.e. the health care team/family member)

[Note: If parents indicate they have made more than one decision, they will be asked to pick the one they think is most important to discuss.]

Participant ID_____ **Interviewer ID**_____ **Date**_____

Control Preferences Scale-Pediatrics

Regarding the decision you have made for your infant, as identified on the Decision Checklist, how would you describe the way you made that decision?

1. I made the final decision about which treatment my child would receive.

2. I made the final decision about my child's treatment after seriously considering the health provider's opinion.

3. I shared the responsibility for the decision about the best treatment for my child with my infant's healthcare provider.

4. My infant's health care provider made the final decision about which treatment was best for my child, but after seriously considering my opinion.

5. I left the final decision about all of my child's treatments to the health care provider.

Participant ID _____ Interviewer Initials_____ Date _____

Brief Illness Perception Questionnaire Items

After reading each question please circle the number corresponding to your answer

1) How much will your/the baby's illness or condition affect his or her life?

No affect: **1 2 3 4 5 6 7 8 9 10** :*Severely affect*

2) How long do you think your/the baby's illness will continue?

Very short time: **1 2 3 4 5 6 7 8 9 10** :*Forever*

3) How much control do you feel you have over your baby's illness?

Absolutely no control: **1 2 3 4 5 6 7 8 9 10** :*Extreme amount of Control*

4) How much do you think your/the baby's treatment can help his/her illness?

Not helpful: **1 2 3 4 5 6 7 8 9 10** :*Extremely helpful*

5) How sick is your/the baby?

Not sick at all: **1 2 3 4 5 6 7 8 9 10** :*Extremely sick*

6) How concerned are you about your/the baby's illness?

Not at all concerned: **1 2 3 4 5 6 7 8 9 10** : *Extremely concerned*

7) How well do you feel you understand your baby's illness?

Don't understand at all: **1 2 3 4 5 6 7 8 9 10** :*Understand very clearly*

8) How much does your baby's illness affect you emotionally?

Not at all affected emotionally: **1 2 3 4 5 6 7 8 9 10** :*Extremely affected emotionally*

9) How much do you think your/the baby's condition is a serious condition?

Not serious: **1 2 3 4 5 6 7 8 9 10** :*Extremely serious*

Participant ID _____ Interviewer Initials_____ Date _____

Brief COPE

After reading each of the 28 coping behaviors and thoughts, please rate your response based on how often each statement is true for yourself.

1= *I haven't been doing this at all*

2= *I've been doing this a little bit*

3= *I've been doing this a medium amount*

4= *I've been doing this a lot*

_____ 1. I've been turning to work or other activities to take my mind off things.
_____ 2. I've been concentrating my efforts on doing something about the situation I'm in.
_____ 3. I've been saying to myself, "This isn't real."
_____ 4. I've been using alcohol or other drugs to make myself feel better.
_____ 5. I've been getting emotional support from others.
_____ 6. I've been giving up trying to deal with it.
_____ 7. I've been taking action to try to make the situation better.
_____ 8. I've been refusing to believe that it has happened.
_____ 9. I've been saying things to let my unpleasant feelings escape.
_____ 10. I've been getting help and advice from other people.
_____ 11. I've been using alcohol or other drugs to help me get through it.
_____ 12. I've been trying to see it in a different light, to make it seem more positive.
_____ 13. I've been criticizing myself.
_____ 14. I've been trying to come up with a strategy about what to do.
_____ 15. I've been getting comfort and understanding from someone.
_____ 16. I've been giving up the attempt to cope.
_____ 17. I've been looking for something good in what is happening.
_____ 18. I've been making jokes about it.
_____ 19. I've been doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping.
_____ 20. I've been accepting the reality of the fact that it has happened.
_____ 21. I've been expressing my negative feelings.
_____ 22. I've been trying to find comfort in my religion or spiritual beliefs.
_____ 23. I've been trying to get advice or help from other people about what to do.
_____ 24. I've been learning to live with it.
_____ 25. I've been thinking hard about what steps to take.
_____ 26. I've been blaming myself for things that happened.
_____ 27. I've been praying or meditating.
_____ 28. I've been making fun of the situation.

Participant ID _____ Interviewer Initials_____ Date _____

The Brief RCOPE: Positive and Negative Coping Subscale Items.

The following items also deal with ways you coped with your infant's NICU admission. Please tell me how much or how frequently you did what the item says, not whether it worked or not.

*The phrase "higher power" and "power greater than myself" refer to a belief in a religious/spiritual entity.

1= *Not at all*

2= *I did this a little bit*

3= *I did this a medium amount*

4= *I did this a lot*

Positive Religious Coping Subscale Items

_____ **1.** Looked for a stronger connection with God.
_____ **2.** Sought God's love and care.
_____ **3.** Sought help from God in letting go of my anger.
_____ **4.** Tried to put my plans into action together with God.
_____ **5.** Tried to see how God might be trying to strengthen me in this situation.
_____ **6.** Asked forgiveness for my sins.
_____ **7.** Focused on religion to stop worrying about my problems.

Negative Religious Coping Subscale Items

_____ **8.** Wondered whether God had abandoned me.
_____ **9.** Felt punished by God for my lack of devotion.
_____ **10.** Wondered what I did for God to punish me.
_____ **11.** Questioned God's love for me.
_____ **12.** Wondered whether my church had abandoned me.
_____ **13.** Decided the devil made this happen.
_____ **14.** Questioned the power of God.

Edinburgh Postnatal Depression Scale[1] (EPDS)

Name: _____ Address: _____

Your Date of Birth: _____ _____

Baby's Date of Birth: _____ Phone: _____

As you are pregnant or have recently had a baby, we would like to know how you are feeling. Please check the answer that comes closest to how you have felt **IN THE PAST 7 DAYS**, not just how you feel today.

Here is an example, already completed.

I have felt happy:
- ☐ Yes, all the time
- ☒ Yes, most of the time
- ☐ No, not very often
- ☐ No, not at all

This would mean: "I have felt happy most of the time" during the past week. Please complete the other questions in the same way.

In the past 7 days:

1. I have been able to laugh and see the funny side of things
   - ☐ As much as I always could
   - ☐ Not quite so much now
   - ☐ Definitely not so much now
   - ☐ Not at all

2. I have looked forward with enjoyment to things
   - ☐ As much as I ever did
   - ☐ Rather less than I used to
   - ☐ Definitely less than I used to
   - ☐ Hardly at all

*3. I have blamed myself unnecessarily when things went wrong
   - ☐ Yes, most of the time
   - ☐ Yes, some of the time
   - ☐ Not very often
   - ☐ No, never

4. I have been anxious or worried for no good reason
   - ☐ No, not at all
   - ☐ Hardly ever
   - ☐ Yes, sometimes
   - ☐ Yes, very often

*5 I have felt scared or panicky for no very good reason
   - ☐ Yes, quite a lot
   - ☐ Yes, sometimes
   - ☐ No, not much
   - ☐ No, not at all

*6. Things have been getting on top of me
   - ☐ Yes, most of the time I haven't been able to cope at all
   - ☐ Yes, sometimes I haven't been coping as well as usual
   - ☐ No, most of the time I have coped quite well
   - ☐ No, I have been coping as well as ever

*7 I have been so unhappy that I have had difficulty sleeping
   - ☐ Yes, most of the time
   - ☐ Yes, sometimes
   - ☐ Not very often
   - ☐ No, not at all

*8 I have felt sad or miserable
   - ☐ Yes, most of the time
   - ☐ Yes, quite often
   - ☐ Not very often
   - ☐ No, not at all

*9 I have been so unhappy that I have been crying
   - ☐ Yes, most of the time
   - ☐ Yes, quite often
   - ☐ Only occasionally
   - ☐ No, never

*10 The thought of harming myself has occurred to me
   - ☐ Yes, quite often
   - ☐ Sometimes
   - ☐ Hardly ever
   - ☐ Never

Administered/Reviewed by_____ Date _____

[1]Source: Cox, J.L., Holden, J.M., and Sagovsky, R. 1987. Detection of postnatal depression: Development of the 10-item Edinburgh Postnatal Depression Scale. *British Journal of Psychiatry* 150:782-786 .

[2]Source: K. L. Wisner, B. L. Parry, C. M. Piontek, Postpartum Depression N Engl J Med vol. 347, No 3, July 18, 2002, 194-199

Participant ID _____ Interviewer ID _____ Date _____

GOOD PARENT RANKING EXERCISE

Parents have shared with us different things that they feel are very important for them to do to be good parents for their ill child, but parents have different views about which of these things are more important or less important.

The following pages show several different sets of 4 important things for a parent to do to be a good parent for their ill child. Thinking about <u>today</u>, please read each set on its own, and rank the items in the set from 1 to 4 in order of importance to you, with

- 1 being the most important in that set
- 2 is 2nd most important
- 3 is 3rd, and
- 4 is 4th in importance.

Here is a completed **SAMPLE SET:**

	Most Important	2nd	3rd	4th
Keeping a positive outlook	○	○	○	⊗
Staying at my child's side	○	⊗	○	○
Focusing on my child's comfort	○	○	⊗	○
Making sure that my child feels loved	⊗	○	○	○

	Most Important	2nd	3rd	4th
Keeping a positive outlook	O	O	O	O
Staying at my child's side	O	O	O	O
Focusing on my child's comfort	O	O	O	O
Making sure that my child feels loved	O	O	O	O

	Most Important	2nd	3rd	4th
Focusing on my child's spiritual well-being	O	O	O	O
Keeping a positive outlook	O	O	O	O
Focusing on my child's health	O	O	O	O
Advocating for my child with medical staff	O	O	O	O

	Most Important	2nd	3rd	4th
Making sure that my child feels loved	O	O	O	O
Keeping a realistic outlook	O	O	O	O
Keeping a positive outlook	O	O	O	O
Focusing on my child's quality of life	O	O	O	O

	Most Important	2nd	3rd	4th
Focusing on my child's quality of life	O	O	O	O
Focusing on my child's spiritual well-being	O	O	O	O
Making informed medical care decisions	O	O	O	O
Focusing on my child's comfort	O	O	O	O

	Most Important	2nd	3rd	4th
Advocating for my child with medical staff	O	O	O	O
Making informed medical care decisions	O	O	O	O
Making sure that my child feels loved	O	O	O	O
Focusing on my child having as long a life as possible	O	O	O	O

	Most Important	2nd	3rd	4th
Staying at my child's side	O	O	O	O
Making sure that my child feels loved	O	O	O	O
Focusing on my child's spiritual well-being	O	O	O	O
Focusing on my child having as long a life as possible	O	O	O	O

	Most Important	2nd	3rd	4th
Focusing on my child having as long a life as possible	O	O	O	O
Focusing on my child's quality of life	O	O	O	O
Focusing on my child's comfort	O	O	O	O
Putting my child's needs above my own needs when making medical care decisions	O	O	O	O

	Most Important	2nd	3rd	4th
Focusing on my child's comfort	O	O	O	O
Focusing on my child's health	O	O	O	O
Advocating for my child with medical staff	O	O	O	O
Keeping a realistic outlook	O	O	O	O

	Most Important	2nd	3rd	4th
Focusing on my child's health	O	O	O	O
Focusing on my child having as long a life as possible	O	O	O	O
Keeping a realistic outlook	O	O	O	O
Focusing on my child's spiritual well-being	O	O	O	O

	Most Important	2nd	3rd	4th
Making informed medical care decisions	O	O	O	O
Putting my child's needs above my own needs when making medical care decisions	O	O	O	O
Keeping a positive outlook	O	O	O	O
Focusing on my child's health	O	O	O	O

	Most Important	2nd	3rd	4th
Keeping a realistic outlook	O	O	O	O
Making sure that my child feels loved	O	O	O	O
Staying at my child's side	O	O	O	O
Making informed medical care decisions	O	O	O	O

	Most Important	2nd	3rd	4th
Putting my child's needs above my own needs when making medical care decisions	O	O	O	O
Advocating for my child with medical staff	O	O	O	O
Focusing on my child's quality of life	O	O	O	O
Staying at my child's side	O	O	O	O

Participant ID _____ Interviewer ID _____ Date _____

Here is a list of all twelve items that were in the sets that you just worked with. Please tell us if there is anything else that is important to you to be a good parent to your ill child that was not included in this list. There is space at the bottom to write your thoughts.

Focusing on my child's comfort

Focusing on my child's quality of life

Making informed medical care decisions

Keeping a realistic outlook

Focusing on my child's health

Advocating for my child with medical staff

Staying at my child's side

Keeping a positive outlook

Putting my child's needs above my own needs when making medical care decisions

Making sure my child feels loved

Focusing on my child having as long a life as possible

Focusing on my child's spiritual well-being

Please write any additional items that are important to you to be a good parent to your ill child:

_____

_____

_____

Participant ID _____ Interviewer Initials_____ Date _____

Demographic Survey

Parent Demographics:

Please describe yourself:

1) Your sex: ____Male _____Female

2) How old are you? ____ Years

3) With which race group do you identify yourself?
- a) ____African American/Black
- b)____ White
- c) ____ Asian
- d) ____ Native Hawaiian/Other Pacific Islander
- e) ____ American Indian/Alaskan Native
- f) ____ Multiracial (Please check all that apply)
- g) ____ Other _____

3) With which ethnic group do you identify yourself?
- a) ____ Hispanic
- b) ____ Non-Hispanic
- c) ____ Other

4) Relationship status:
- a) ____Married
- b) ____ Widowed
- c) ____ Separated
- d) ____ Divorced
- e) ____ Single/Never Married
- f) ____ Living with significant other/partner
- g) ____ Other

5) Which of the following best describes your employment status at this time?
- a) ____ Full-time employee
- b) ____ Part-time employee
- c) ____ Student
- d) ____ Homemaker
- e) ____ Disabled
- f) ____ Unemployed
- g) ____ Other _____

6) Which of these ranges matches your total household income from the past year?
- a) ____ Less than $15,000
- b) ____ $15,001-$30,000
- c) ____ $30,001-$45,000
- d) ____ $45,001-$60,000
- e) ____ $60,001-$75,000
- f) ____ Over $75,000
- g) ____ Don't know
- h) ____ Decline to answer

7) Which of these describes your highest level of education?
- a) ____ Less than High School
- b) ____ High School or GED
- c) ____ Some college
- d) ____ College graduate
- e) ____ Some graduate school
- f) ____ Completed graduate school

8) How many other children do you have? ____

9) Have any of your other children ever been hospitalized before?
- a) ____Yes
- b) ____ No

10) Do you consider yourself one of the following?
- a) ____ Catholic
- b) ____ Protestant
- c) ____ Jewish
- d) ____ Muslim
- e) ____ None
- e) ____ Other _____

11) Before this child's birth, how often did you attend religious services
- a) ____ Never
- b) ____ Once or twice a year
- c) ____ Once a month
- d) ____ Once a week
- e) ____ More than once a week
- f) ____ Not applicable

12) How important is religion to you?
- a) ____ Not important
- b) ____Somewhat important
- c) ____ Very important

13) Your infant was born with a condition that developed before birth.

Did you know about this condition before your child was born, or did you learn of the condition after your child was born?

a) ____ I learned of my child's condition before he/she was born
b) ____ I learned of my child's condition after he/she was born

14) What type of health insurance do you currently have?
a) ____ Private _____
b) ____ Medicaid
c) ____ Other _____
d) ____ None

Participant ID _____ Interviewer Initials_____ Date _____

Contact Information:

Parent Name: _____

Parent Address:_____

_____

Home Phone: _____ Cell Phone_____

Work Phone _____

Email: _____

Preferred contact method (circle): Email / Text / Phone

Preferred time of day: Mornings, Lunchtime, Evening

Emergency Contact: _____

Address (if different):_____ _____

_____

Phone: _____

Email_____

Emergency contact preferred contact method (circle): email / text / phone

Record Review Form

Patient Name _____

Patient Date of Birth _____

Patient Gender _____

Patient Medical Record Number _____

Study ID Number _____

Patient Address _____
_____
_____

Phone Number _____

Email _____

Health Insurance Information _____

Type of Congenital Anomaly _____

Birth weight (grams) _____

Gestational Age (weeks/days) _____

Gestational status (single or multiple gestation) _____

Current age (days, corrected GA) _____

Date of Admission _____

Date of medical record review _____

STUDY LOG PAGE

☐ **CONSENT SIGNED/DATED (BY SUBJECT AND RESEARCHER)**

☐ **CONSENT COPY TO SUBJECT**

☐ **CONSENT PLACED IN CHART**

☐ **PARKING STICKER GIVEN**

☐ **GIFT CARD GIVEN (QUAN)**

☐ **GIFT CARD GIVEN (QUAL)**

☐ **SURVEYS COMPLETE**

☐ **INTERVIEW COMPLETE**

L1_1_____STUDY ID NUMBER

L1_2 _____MEDICAL RECORD NUMBER

L_1_3 _____SUBJECT NAME (**name**)

L_1_4 _____ ADDRESS (**address**)

_____

_____

L_1_5 _____ PHONE WHERE WE MAY CALL YOU (PHONE)

L_1_5A _____ ALTERNATE PHONE – OKAY TO USE? (**Alternate phone**)

L_1_5B _____ E-MAIL ADDRESS PROVIDED BY PARTICIPANT (**e-mail**)

L_1_6 _____EMERGENCY CONTACT (NAME) & _____PHONE (#)

L_1_7 _____DATE OF SURVEY COMPLETION

L_1_7A _____ TIME NEEDED TO COMPLETE SURVEYS

L_1_8_____OUTCOME 1 = enrolled 2 = not enrolled

L_1_9 _____REASON not enrolling 1 = N/A pt was enrolled, 2 = declined, 3 = other _____(specify)

L_1_10 _____REASON DECLINED 1 = N/A subject completed surveys/was interviewed 2 = too tired, 3 = not comfortable talking about NICU decision making, 4 = no time, 5 = no interest, 6 = no reason, 7 = Other_____(Please specify)

L_1_11A _____METHOD 1 = subject completed survey in NICU 2 = subject to return survey via phone

L_1_12 _____DATE QUALITATIVE INTERVIEW COMPLETED

L_1_13_____OUTCOME (**t1outcome**) 1 = interviewed 2 = not interviewed

L_1_14 _____REASON not Interviewed (**t1notint**) 1 = N/A pt was interviewed, 2 = declined, 3 = not reached, 4 = other _____(specify)

L_1_15 _____REASON DECLINED (**t1noreason**) 1 = N/A pt was interviewed 2 = too tired, 3 = not comfortable talking about donor decision, 4 = no time, 5 = no interest, 6 = no reason, 7 = Other _____(Please specify)

L_1_16_____ TIME NEEDED TO COMPLETE INTERVIEW (minutes)

Appendix B: Qualitative Interview Guide

Qualitative Interview Guide

Background: The following qualitative interview guide was subject to pilot testing on a minimum of 3 parents and then used to interview NICU parents who learn of their infant's congenital diagnosis before vs. after birth.

1) Please share with me what it's like to be a mother/father in the NICU? How is being a mother/father to your other children different than being a mother/father in the NICU?

(Note similarities/differences)

2a) You noted, on the Decision Checklist, that you made [Name specific decision made around congenital anomaly] for your NICU infant.

1. Who has helped you make this decision?
    i. Family?
        1.Partner/Spouse
    ii. Friends?
        1. Members of your community
    iii. Clergy?
    iv. The health care team?
2. How have these individuals helped you make decisions? What did the person do that helped?
3. Are there other ways that you felt helped in making this decision?
    i. How you felt inside?

ii. How you're coping/handling things?

4. What hinders you from making decisions?
    i. Did your perception of how ill your infant was, hinder your decision making?
    ii. Did other things hinder your decision making?

2b) How is making decisions in other areas of your life the same or different from how you make decisions in the NICU?

(Note similarities/differences)

3) In previous situations when parents have been faced with making a difficult decision, such as the one you made for your baby, parents have said they make their decision to benefit their child in some way. These parents have described their decision making as 'doing what a "good parent would do" or "deciding as a 'good parent'". If you could describe to the nurses/doctors/health care team here in the NICU what your definition of being a "good parent" for your child is at this point in your child's life (i.e. around making the decision related to the congenital anomaly) how would you describe what 'being a "good parent" means to you?
    a. Do you have a story about being a "good parent" to your child here in the NICU? (Use probing questions to get at the meaning of being a "good parent" for the parent being interview.)
    b. Is there anything else we haven't talked about that is important in being a good mother/father to your child?
    c. What helped you achieve being a good parent? What's not so helpful?
        i. Prompts
        1. Family?
            a. Partner/Spouse

        2. Friends?

            a. Members of your community
        3. Clergy?
    4. The health care team?

5. How you're feeling inside?
6. How you're coping with stress? (general/religious coping)

4) Some parents like you who have had a baby in the NICU have mentioned that spirituality or religion is important to them. I'm wondering how spirituality or religion might be important for you during this time that your baby is in the NICU?
    a. What faith tradition is the parent part of (if any)?
    b. Is prayer important? If so, what types of things does the parent pray for?
        1 Baby's/Other's Health issues?
        2 Family issues?
        3 Finance/Other issues?
    c. If parent mentions "miracles," what would a miracle look like in this situation, for their baby in the NICU?
    d. How does spirituality and/or religion play a role in decision making here in the NICU? (Ex: Does spirituality/religion help you as you make decisions in the NICU? If so, how?) How does spirituality/religion play a role in being a parent in the NICU? (i.e. Does religion/spirituality play a role in being a parent in the NICU? If so, how?)

5) Are there other specific strategies that you use to manage having a child in the NICU?
    a. Probe for potential coping methods (Ex: Humor, seeking emotional support, seeking practical support, etc.)
    b. How do parents apply these other strategies for managing the experience of having a child in the NICU?

6) What else is important in this situation in the NICU for you as a parent making decisions for your infant that we haven't talked about? Or What do you think is important for other parents who have infants in the NICU to know?

CURRICULUM VITAE

Part I

PERSONAL DATA

Melissa Kurtz Uveges
mkurtz4@jhu.edu

EDUCATION

2013-Present, PhD Student, Johns Hopkins University School of Nursing, Baltimore, MD
2011- Master of Science in Nursing (Policy, Leadership, and Management Specialty), Yale University School of Nursing, New Haven, CT
2011- Master of Arts in Religion (Ethics Concentration), Yale Divinity School, New Haven, CT
2006- Master of Arts, Biblical Studies, Reformed Theological Seminary, Orlando, FL
2000- Bachelor of Science in Nursing, University of Florida School of Nursing, Gainesville, FL
1996- Bachelor of Science in Chemistry, Berry College, Rome, GA

CURRENT LICENSE AND CERTIFICATION

2013-Present, Registered Nurse, State of Maryland Board of Nursing, #R206066
2000-Present, Neonatal Resuscitation Program Certification
2000-Present, Registered Nurse, Florida Board of Nursing, #9169607

PROFESSIONAL EXPERIENCE

2016-Present *Research Coordinator*, Patient Navigation and Clinical Trial Participation: A Randomized Controlled Trial Design, PI: Jennifer Wenzel, PhD, RN, Johns Hopkins IRB Approval #: NA_00072282
2013-present *Per diem nurse clinician*, Neonatal ICU Johns Hopkins Hospital Intrastaff, Baltimore, MD
2015-2017 *Research Assistance*, Comparing two parent training programs for at-risk families: A randomized trial, PI: Deborah Gross, DNSc, RN, Johns Hopkins IRB Approval #: NA_00039328
2011-2013 *Bioethics Fellow* Montefiore Medical Center, Bronx, NY
2008-2011 *Registered Nurse*, Neonatal ICU Yale-New Haven Hospital, New Haven, CT
2002-2008 *Registered Nurse*, Neonatal ICU Florida Hospital for Children, Orlando, FL
2000-2002 *Registered Nurse*, Neonatal ICU Shands Hospital, University of Florida, Gainesville, FL

HONORS AND AWARDS

2017-18 Philanthropic Educational Organization Scholar Award ($15,000)
2016-2018 American Academy of Nursing, Jonas Policy Scholar (Breastfeeding)
2016 March of Dimes Graduate Nursing Scholarship ($5,000)
2016 Miriam M. Powell Scholarship, Nurses Educational Funds, Inc. (4,000)
2014-Present Member of The Johns Hopkins Hospital Ethics Committee and Consultation Service
2014-16 Elected to the American Society for Bioethics & Humanities Nursing Affinity Group Advisory Council
2014-16 Ethics Representative, Doctoral Student Organization, Johns Hopkins University School of Nursing
2014 Professional Development Award, Johns Hopkins School of Nursing ($2,000)
2014 Isabel Hampton Robb Scholarship Recipient, Nurses Educational Funds, Inc. ($10,000)
2013 Outstanding Young Alumnus Award, University of Florida
2011 Charles King, Jr., Memorial Scholars Prize, Yale School of Nursing ($1,000)
2011 Nightingale Award for Excellence in Nursing, V.N.A. of South Central Connecticut
2009 Susan C. Clarke Scholarship Recipient, Yale Divinity School
2009 Featured in Yale Nursing Matters Magazine, Yale School of Nursing
2000 Outstanding Clinical Practice Award, University of Florida School of Nursing
2000 Sigma Theta Tau Nursing Honor Society Induction
2000 Presidential Recognition for Outstanding Achievement and Contributions to the University of Florida

RESEARCH FUNDING

2016 Southern Nursing Research Society (SNRS) Dissertation Award ($5,000)
2016 Sigma Theta Tau Nu Beta Research Award ($1,500)
2016-18 Heilbrunn Family Nurse Scholar Award ($24,000)
2016 STTI/Rosemary Berkel Crisp Research Award ($5,000)
2016 ENRS/Council for the Advancement of Science (CANS) Dissertation Award ($5,000)
2014-2015 NIH-funded TL 1 Pre-doctoral Clinical Research Training Awardee (#5TL1TR001078-02)

SCHOLARSHIP

Journal Publications

2017 Gross, D., Belcher, H.M.E., Budhathoki, C., Ofonedu, M. E., Dutrow, D., **Uveges**, **M.K**., & Slade, E. Reducing preschool behavior problems in an urban mental health clinic serving low-income families: A pragmatic, randomized, non-inferiority comparative effectiveness trial. *Journal of the American Academy of Child and Adolescent Psychiatry. (In Review).*

2017 Gross, D., Belcher, H.M.E., Budhathoki, C., Ofonedu, M.E., **Uveges, M**.K. Does Parent Training Format Affect Treatment Engagement? A Randomized Study of Families at Social Risk. *Journal of Child and Family Studies. (Accepted for publication November 27, 2017).*

2017 Davidson P.M., Rushton, C.H., **Kurtz, M**., Wise, B. , Jackson, D., Broome, M. A social ecological framework: A model for addressing ethical practice in nursing. *Journal of Clinical Nursing*. (*Nov 9, 2017) [Epub ahead of print, PMID: 29119653]*.

2017 Kim, K., Heinze, K., Xu, J., **Kurtz, M**., Park, H., Foradori, M., Nolan, M. Theories of health care decision making at the end of life: A meta-ethnography. *Western Journal of Nursing Research, (Aug 1, 2017) [Epub ahead of print, PMID: 28816094]*.

2017 **Kurtz, M.** & Hurst, A. Religion, spirituality, and pediatric patients and families: Research, assessment, and care. *Children's Project on Palliative/Hospice Services (ChiPPS) E-Journal, (2017) 48, 30-36.*

2016 **Kurtz M.J**., Starbird L.E. Interprofessional clinical ethics education: The promise of crossdisciplinary problem-based learning. *AMA Journal of Ethics*. 2016; 18:917-924. http://journalofethics.amaassn.org/2016/09/nlit1-1609.html.

2016 Hamilton, J.B., Worthy, V. C., **Kurtz, M.J**., Cudjoe, J, & Johnstone, P.A. Using religious songs as an integrative and complementary therapy for the management of psychological symptoms among African American cancer survivors. *Cancer Nursing* (EPub ahead of print, 2016 Feb 8).

2016 Xu, J., Kim, K., **Kurtz, M**., Nolan, M. Mentored peer reviewing for PhD faculty and students. *Nursing Education Today 37* (2016), 1-2.

2015 Rushton, C.H., Caldwell, M., **Kurtz, M**. Moral distress: A catalyst in building moral resilience. *American Journal of Nursing*, 116(7), 40-49.

2014 Zhang, X., **Kurtz, M**., Lee, S., Liu, H. Early intervention for preterm infants and their mothers: A systematic review. *Journal of Perinatal and Neonatal Nursing*. Advance online publication (Nov 18 2014).

2014 & 2017 (forthcoming in November) **Kurtz, M.** & Powell, T. "Physician-assisted suicide, physician aid in dying, and euthanasia." In *The Encyclopedia of Elder Care: The Comprehensive Resource on Geriatric Health and Social Care, 3rd ed. & 4th ed.(forthcoming),* edited by E.A. Capezuti, M.L. Malone, P.R. Katz, and M.D. Mezey, 578-82. New York: Springer Publishing Company, 2014.

2012 **Kurtz, M. J.** 'God can still save my granddaughter:' The role of spirituality in ethical decision- making for a critically ill neonate. *Progress in Palliative Care 20*, no.2 (2012), 86-93.

2010 **Kurtz, M**, Black-Golde, P, Berlinger, N. Ethical considerations in CYP2D6 genotype testing for codeine-prescribed breastfeeding mothers. *Clinical Pharmacology and Therapeutics 88*, no.6 (2010), 760-762.

2009 **Kurtz, M.** More than a village: Meeting the health care needs of multiples. *Hastings Center Report* 39, no.3 (2009), 25-26.

1997 Breton GW, **Kurtz MJ**, Kurtz SL. Acetylation of unsymmetrical diols in the presence of Al2O3. *Tetrahedron Letters* 38, no.22 (1997), 3825-3828.

Educational Pamphlets

2015 Rushton, C.H. & Kurtz. Moral distress and you: Supporting ethical practice and moral resilience in nursing. American Nurses Association Publication (2015).

Book Chapters

2016 Kurtz, M. & Rushton, C.H. The impact of ethical conflict and dilemmas on nurses. In J. Erickson & K. Payne (Eds.), *Ethics in oncology nursing*, Pittsburgh: Oncology Nursing Society (2016).

CONFERENCE PRESENTATIONS

International

Kurtz, M., Nolan, M., Hamilton, J. The role of religion and spirituality in parent decision making for seriously ill children. Poster Presentation, Sigma Theta Tau 2017 International Nursing Research Congress. Dublin, Ireland, July 29, 2017.

National

Rushton, C.H., Epstein, E., Turner, M., & **Kurtz, M**. Nurses fulfilling their ethical mandate: Living the Code of Ethics. Oral Presentation. The American Society for Bioethics and Humanities Annual Conference, Houston, TX, October 22, 2015.

Kurtz, M. The Role of 'The Good Parent,' Religiosity, and Spirituality in NICU Parent Decision Making. Association for Clinical and Translational Science Annual Conference. Poster Presentation. Washington, D.C., April 17, 2015.

Kurtz, M. Too Close for Words: Ethical Issues that Arise when Certified Healthcare Interpreters Share the Patient's or Family's Cultural or Ethnic Beliefs. Oral Presentation. The American Society for Bioethics and Humanities Annual Conference, Washington D.C., October 19, 2012.

Kurtz, M. Caring For Angela: Spiritual and Cultural Factors in Ethical Decision-Making for a Critically Ill Neonate. Oral Presentation. The American Society for Bioethics and Humanities Annual Conference, San Diego, CA, October 22, 2010.

Regional

Kurtz, M. Moral Distress and the Nursing Profession. Oral Presentation. Catskill Hudson Area Health Education 2012 Nursing Summit, Columbia-Greene Community College, Hudson, NY, May 30, 2012.

Kurtz, M. Emerging Technologies in Healthcare. Oral Presentation. Foundations of Professional Practice Nursing Conference, Dominican College, Orangeburg, NY, May 2009.

Local

Weck, K., **Kurtz, M**., Budhathoki, C., & Gross, D. Does parental attachment style influence child behavior? Poster Presentation. Johns Hopkins School of Nursing 2017 Research Honors Day, Baltimore, MD, April 26, 2017.

Kurtz, M. Nurse Clinicians: What keeps us up at night and how inter-professional collaboration can help. Oral Presentation. A Discussion on the New Hastings Center Guidelines for Decisions on LifeSustaining Treatment and Care Near the End-of-Life, The Program for Biomedical Ethics, Yale University School of Medicine, October 7, 2013.

Kurtz, M. Bioethics and the Anesthesiologist. Oral Presentation. Presentation to the Anesthesiology House Staff at Montefiore Medical Center, Bronx, NY, July 26, 2012.

Kurtz, M. Ethical Considerations in Pediatric and Disability Care. Oral Presentation. Yale Interdisciplinary Center for Bioethics, pediatric and disability seminar guest speaker, New Haven, CT, June 20, 2012.

Kurtz, M. Bioethics and the Risk Manager. Oral Presentation. CNA HealthPro Risk Control, Teleconference Presentation, May 21, 2012.

Kurtz, M. Ethical Considerations When Caring for Children and Families. Oral Presentation. Yale- New Haven Hospital, New Haven, CT, February 16, 2011.

Kurtz, M. Issues of Moral Distress in the NICU. Oral Presentation. Yale Interdisciplinary Center for Bioethics Working Group, New Haven, CT, June 2009.

EDITORIAL ACTIVITIES

2011-Present Editorial Advisory Board Member, Medical Ethics Advisor Newsletter, an AHC Media Publication.
2016-Present Co-Editor of the Ethics column for *AACN Advanced Critical Care Journal*

PROFESSIONAL ACTIVITIES

2016-Present, The Council for the Advancement of Nursing Science (CANS)
2015-Present, Southern Nursing Research Society (SNRS)

2015-Present, American Association of Critical-Care Nurses (AACN)
2015- Present, American Nurses Association (ANA)
2014- Present Member, Association of Women's Health, Obstetric & Neonatal Nurses (AWHONN)
2014- Present Member, Eastern Nursing Research Society (ENRS)
2007- Present Member, American Society for Bioethics and Humanities (ASBH)
2000-Present Member, Sigma Theta Tau International (STTI)

Peer Review Activities
2016 Ad Hoc Journal Article Review for *Circulation*
2015 Ad Hoc Journal Article Review for *The Hastings Center Report*
2013-2014 Ad Hoc Journal Article Review with Dr. Marie Nolan for *Journal of Palliative Medicine*

Curriculum Vitae
Part II

EDUCATIONAL ACTIVITIES
Fall 2015, Spring 2016, Fall 2016, Fall 2017, **Professionalism Nursing Ethics Seminar** (NR120.501), Instructor, portion of 2 credit course, Masters level, 30 students each term.
Summer 2016, **Philosophical, Theoretical, and Ethical Basis for Nursing** (NR 110.500), CoInstructor, 3 credits, Masters level, 7 students.
Fall 2015, **Nursing for Adult Health I** (NR 110.315), Teaching Assistant, 2 credits, BSN level, 120 students.
Summer 2015, **Bioethics Consultation** (Albert Einstein College of Medicine), Co-Instructor, Masters level, 9 students.
Spring 2015, **Philosophical, Theoretical, and Ethical Basis for Nursing** (NR 110.500), Teaching Assistant, 3 credits, Masters level, 37 students.
Fall 2014, **Philosophical, Theoretical, and Ethical Basis for Nursing** (NR 110.500), Teaching Assistant, 3 credits, Masters level, 32 students.
Summer 2014, **Applications of Research to Practice** (NR110.503), Teaching Assistant, 3 credits, Masters level, 35 students.
Spring 2014, **Applications of Research to Practice** (NR110.503), Teaching Assistant, 3 credits, Masters level, 32 students.
2012-2013, **Biomedical Ethics II** (Albert Einstein College of Medicine), Seminar leader, medical student level, 15 students.