Parent and Provider Decision-Making for Infants with HIE

by

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Duke University

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C. Michael Cotten

Dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy in Nursing in the Graduate School of Duke University

2012
ABSTRACT

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Abstract

Hypoxic ischemic encephalopathy (HIE) is a serious birth complication of full-term infants; 40-60% of affected infants die by 2 years or have severe disabilities. Infants with HIE often have a normal gestation and parents anticipate a healthy birth. HIE can be managed aggressively with moderate hypothermia < 6 hours of life, cardiopulmonary support, and seizure management. Experimental interventions such as moderate hypothermia > 6 hours of life and umbilical cord stem cell transplant are also available. Additional decision-making for these infants may include long-term developmental therapy, nutritional support, and respiratory support. However, who makes these decisions, what factors influence decision-making and the long-term impact of decision-making on parents and health care providers remains unknown. Therefore, the purpose of this study was to explore parental and health care provider decision-making for infants with HIE.

A longitudinal case study design was used to study 11 cases of infants with HIE. Each case included the infant, the parent, and the infant’s providers. Infant medical record data, interviews and questionnaires were used to collect data from infant birth through 6 months of age. Content analysis was used to analyze the interviews. Descriptive statistics were used with the questionnaires. Visualization techniques were used to search for patterns and trends in the assembled data.
All infants required resuscitation and their treatment plans included aggressive care or aggressive and experimental care. The level of parental participation varied with in the first week of life depending on whether the infant was enrolled in experimental interventions plus aggressive care or only aggressive care. Parental hopefulness was lower in parents of infants who received experimental interventions, but the infants receiving experimental interventions were less critically ill than infants who received aggressive care only. Parental stress was also lower among parents of infants who received experimental interventions over the first 2 months of life.

Parents were concerned about the short and long-term impact of HIE, few parents understood that even though their infant had appropriate developmental outcomes at 6-months that did mean that neurological damage occurred. However in one case of an infant, the neurological development became central to the parental decision-making for the infant. Parents became less hopeful as diagnostic examinations continued find more complex conditions that were individually not problematic for the parents, but when the complexity of the infant’s illnesses continued to unfold, parents feared that too many complications existed for their daughter to have an acceptable quality of life. Yet, when parents broached the topic of transitioning from aggressive care to palliative care with providers, they were told that withholding/withdrawing treatment was not appropriate for the infant. Not discussing withholding or withdrawing treatment ultimately created conflict between parents and providers due to
differences in opinions about the predicted neurological outcomes for the infant. The conflict led to distrust and parents regretted most decisions they made for their infant.

Parental and provider decision-making is complex and many of the decisions within the 6-month trajectory were made within the first 6 hours of birth. Parents felt that the decision-making was appropriate in most cases, but the extent of the infant’s injury remains unknown. How parents will evaluate the decision-making when the infant begins to miss developmental milestones is unknown. Results from this dissertation suggest that decision-making is a trajectory and decisions are not made in isolation. Implications for practice include discussing and educating parents during the first 6 months and later about developmental milestones and the importance of continuing therapy, even when the infant appears normal. Providers can also acknowledge to parents, up front, that the extent of the neurological injury is unknown and different providers may have different opinions about the long-term effects. By acknowledging these differences, providers can begin discussing the treatment options with parents and educating them about the specific needs of their infant.
Dedication

This dissertation is dedicated to Sherry K. and Phillip B. Melton. Aunt She She, you are the reason I became a nurse and you are my inspiration. Uncle Dip words cannot describe what you mean to me. You were one of the kindest and most gentle individuals I have ever known. I miss you both so much and owe much of my success to you.
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good times and funny moments. Finally to my four legged babies, Dooley and Pacer, every day you smiled and wagged no matter what and that is priceless.
1. Introduction

Hypoxic ischemic encephalopathy (HIE) is one of the most serious birth complications affecting full term infants (Schiariti et al., 2008) occurring in 1.5 to 2.5 per 1000 live births (Graham, Ruis, Hartman, Northington, & Fox, 2008; Kurinczuk, White-Koning, & Badawi, 2010). HIE results in brain injury from a hypoxic-ischemic event during the prenatal, intrapartum or postnatal period preventing adequate blood flow to the infant’s brain (Long & Brandon, 2007). Infants with HIE experience associated morbidities with 40-83% either dying by age 2 years or having severe disabilities (Jacobs et al., 2011; Kwon et al., 2011; Simbruner, Mittal, Rohlmann, & Muche, 2010). The long-term neurological consequences of HIE include mental retardation, epilepsy, and cerebral palsy (Simbruner et al., 2010).

The incidence of HIE has not declined with advances in obstetric care (i.e., fetal monitoring) aimed at preventing the hypoxic-ischemic event (Kumar & Paterson-Brown, 2010); thus much of the current neonatal research focuses on preventing further brain injury after the hypoxic-ischemic event (Shankaran, 2009). In the past, treatment options were limited to standard medical treatment (e.g., antiepileptic medications, respiratory support) (Hoehn et al., 2008; Vannucci & Perlman, 1997). Currently, experimental interventions (e.g., moderate hypothermia, phenobarbital administration, umbilical cord transplant) are available to potentially ameliorate the consequences of HIE (Jacobs et al., 2011; Pimentel-Coelho, Rosado-de-Castro, da Fonseca, & Mendez-Otero, 2012); however,
long-term outcomes in school-aged children remain largely unknown. Even with the advent of experiment interventions, many infants will require therapy through early childhood because of severe disabilities (Jacobs et al., 2011; Shankaran, Pappas, et al., 2012; Simbruner et al., 2010).

Parents and health care providers are often surprised when faced with an unexpected complication at delivery due to HIE. The evolving pathology of HIE and limited research on long-term outcomes of newer treatments leave parents and providers making decisions about which life-sustaining treatments to initiate and whether experimental interventions will benefit the infant (Allen & Brandon, 2011; Cotten & Shankaran, 2010). Diagnostic evaluation tools are available such as the Sarnet neonatal encephalopathy staging criteria (Sarnat & Sarnat, 1976) and neuroimaging, but these tools are unable to precisely predict infant outcomes and responses to treatments (White et al., 2012). Yet parents and providers must determine how to proceed with treatment for these infants immediately following birth and throughout the infant’s life with limited knowledge of how the brain injury will impact the infant’s long-term outcomes. The decisions immediately following birth and throughout the first 6 months of life of infants with HIE remain unknown. Additionally it is unclear how decisions are made for these infants and who is involved in decision making across the illness trajectory. Therefore, the purpose of this dissertation was to explore the trajectory of parental and provider decision-making for infants with HIE.
1.1 Hypoxic Ischemic Encephalopathy

1.1.1 Etiology

HIE is a disorder characterized by clinical manifestations, which indicate brain dysfunction (Kurinczuk et al., 2010). Although the exact cause is not always identified (Shankaran, 2009), antecedents to hypoxia or asphyxia include cord prolapse, uterine rupture, abruptio placenta, placenta previa, maternal hypotension, breech presentation, or shoulder dystonia (Madan, Hamrick, & Ferriero, 2005; Okereafor et al., 2008). The manifestations of perinatal HIE are abnormal fetal heart rate tracings, poor umbilical cord gases (pH < 7.0 or base deficit ≥ 12 mmol/L), low Apgar scores (de Vries & Cowan, 2009), presence of meconium stained fluid (Kumar & Paterson-Brown, 2010), or the need for respiratory support within the first several minutes of postnatal life (Cotten & Shankaran, 2010). Most underlying pathologic events of HIE are a result of impaired cerebral blood flow (Shalak & Perlman, 2004) and oxygen delivery to the brain (Cotten & Shankaran, 2010).

1.1.2 Pathophysiology

The pathologic events of HIE occur in two phases: primary energy failure and secondary energy failure (Cotten & Shankaran, 2010). Primary energy failure is defined as the initial impairment of cerebral blood flow (Shalak & Perlman, 2004). Impaired cerebral blood flow leads to a decrease in oxygen and glucose, which are critical for energy production in the brain. Most of the effects of the primary energy failure lead to
impaired cellular integrity (Cotten & Shankaran, 2010) through disruption of the cytoskeleton and membrane injury (Volpe, 2008). The extent of primary energy failure contributes to further injury in the secondary energy failure phase (Cotten & Shankaran, 2010). Once blood flow is restored, there is a brief period of recovery (Shalak & Perlman, 2004). This latent phase is considered the optimal timing for therapeutic interventions (Cotten & Shankaran, 2010).

The secondary energy failure phase occurs 6 to 48 hours after the initial injury (Shalak & Perlman, 2004; Vannucci & Perlman, 1997). This phase is characterized by decreased amounts of high-energy phosphate levels and normal pH (Lorek et al., 1994). The exact mechanisms of secondary energy failure remain unclear (Cotten & Shankaran, 2010) but appear to be related to oxidative stress, excitotoxicity, and inflammation (Fatemi, Wilson, & Johnston, 2009; Ferriero, 2004; Shalak & Perlman, 2004).

Oxidative stress is caused by the overproduction of free radicals causing damage to neuronal cell membranes leading to necrosis or apoptosis (Buonocore, Perrone, & Bracci, 2001). Excitotoxicity causes cellular death from overstimulation of glutamate receptors (Johnston, Trescher, Ishida, & Nakajima, 2001). Glutamate is used by a variety of neuronal pathways including hearing, vision, somatosensory function, learning and memory (Fatemi et al., 2009), which are all important pathways during the development of infants. Because the neonatal brain is changing at a rapid pace, predicting the short and long-term effect of cognitive and motor function on the brain is difficult. Differences
in the maturational stage of the brain development affect how the perinatal brain responds to the overstimulation of glutamate receptors (Fatemi et al., 2009; Ferriero, 2004). Inflammation is also thought to be important to the development of the brain injury, but the exact mechanism remains unknown (Ferriero, 2004). Therefore, the impact of maturational development, excitotoxicity, oxidative stress, and inflammation changes makes it more difficult to predict how the brain will respond to the secondary energy failure.

The primary and secondary energy failures can lead to two types of cell death: necrosis and apoptosis (Shalak & Perlman, 2004). Necrosis occurs in conditions of very severe hypoxia and ischemia (Johnston et al., 2009), causing cells to swell and rupture, thus allowing the release of cellular contents and resultant inflammation (Volpe, 2008). If the insult is less severe, the cells may recover or progress to apoptosis, programmed cell death (Johnston et al., 2009). The apoptosis can occur days following the initial injury (Fatemi et al., 2009). Both necrosis and apoptosis can lead to decreased brain function. The primary and secondary energy failures are the underlying causes of the clinical manifestations observed in infants with HIE.

In addition to changes in the brain, systemic consequences are also associated with HIE. Multi-system organ dysfunction can occur because of the lack of oxygen and blood flow to the other organs (Shah, Beyene, To, Ohlsson, & Perlman, 2006; Shalak & Perlman, 2004; Vannucci, 2000). Specific systemic complications include acute tubular
necrosis, hepatic necrosis, cardiomyopathy, necrotizing enterocolitis, persistent fetal circulation, inappropriate secretion of antidiuretic hormone (ADH), and adrenal insufficiency (Vannucci, 2000). So in addition to pathophysiologic changes in the brain, these infants can also experience systemic effects leading to a poorer prognosis for the infant and increasing the challenge of treating and predicting the long-term outcomes of infant with HIE.

1.1.3 Assessing the Severity of HIE

The assessment of the severity of HIE consists of physical assessments, biochemical assessments (e.g., umbilical cord gases), and neuroimaging over the first several hours to weeks of neonatal life. The American College of Obstetricians and Gynecologist and American Academy of Pediatrics (2003) suggest the following criteria to diagnosis moderate or severe HIE in the term infant: metabolic acidosis with an umbilical cord pH of < 7 or a base deficit ≥ 12 mmol/L; early onset of encephalopathy; multi-system organ dysfunction; and exclusion of other etiologies. The clinical symptoms of neonatal encephalopathy evolve over hours to days following the insult (Ferriero, 2004). Providers often use the Sarnat staging criteria (1976) or an adapted version to describe of the severity of encephalopathy within the first several postnatal days of life (Cotten & Shankaran, 2010). See Table 1 for Neonatal Encephalopathy Staging.

Infants with Stage 1 or mild encephalopathy appear hyperalert with relatively
few clinical symptoms other than a weak suck reflex that could be related to a variety of other clinical conditions. Moderate encephalopathy (Stage 2) presents as an infant with lethargy, decreased spontaneous activity, and hypotonia. In stark contrast, infants with Stage 3 or severe encephalopathy, often exhibit a decreased level of consciousness and periodic breathing with apnea or bradycardia in the first several hours, along with flaccid muscle tone (Vannucci, 2000). Another sign of HIE is seizures during the first postnatal Day of Life (Yager, Armstrong, & Black, 2009). The longer the neurological exam remains abnormal the more likely the infant is to have developmental delays (Murray et al., 2010).

**Table 1: Sarnat Stages of Neonatal Encephalopathy**

<table>
<thead>
<tr>
<th>Assessment</th>
<th>Stage 1</th>
<th>Stage 2</th>
<th>Stage 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mental status</td>
<td>Hyperalert</td>
<td>Lethargic</td>
<td>Stuporous</td>
</tr>
<tr>
<td>Suck reflex</td>
<td>Weak or absent</td>
<td>Weak or absent</td>
<td>Absent</td>
</tr>
<tr>
<td>Moro reflex</td>
<td>Strong</td>
<td>Weak</td>
<td>Absent</td>
</tr>
<tr>
<td>Muscle tone</td>
<td>Normal</td>
<td>Hypotonia</td>
<td>Flaccid</td>
</tr>
<tr>
<td>Autonomic function</td>
<td>Generalized</td>
<td>Generalized</td>
<td>Absent</td>
</tr>
<tr>
<td>Pupils</td>
<td>Mydriasis</td>
<td>Miosis</td>
<td>Variable</td>
</tr>
<tr>
<td>Seizures</td>
<td>None</td>
<td>Common</td>
<td>Variable</td>
</tr>
<tr>
<td>EEG</td>
<td>Normal (awake)</td>
<td>Early: low-voltage</td>
<td>Early: periodic pattern with isopotential phases Late: isopotential</td>
</tr>
<tr>
<td>Duration</td>
<td>&lt; 24 hours</td>
<td>2-14 days</td>
<td>Hours to weeks</td>
</tr>
</tbody>
</table>

Adapted from (Cotten & Shankaran, 2010)

In addition to a thorough neurologic and physical examination, biochemical assessments of the infant’s umbilical cord blood can help providers determine the
probability of an infant having a poor outcome. A meta-analysis by Malin, Morris, and Khan (2010) found that low umbilical cord pH (pH < 7.2) was strongly associated with neonatal mortality and morbidity (e.g., HIE, seizures, IVH, PVL). Low umbilical cord arterial pH was more strongly associated with HIE than with seizures, IVH, or PVL. The low umbilical cord pH was also associated with CP (Malin et al., 2010). An initial assessment of umbilical cord pH may help providers identify the presence of a problem and plan careful monitoring and evaluation.

In order to determine location and extent of injury (Chau et al., 2009), neuroimaging of the infant with HIE is recommended between Day of Life 2 and Day of Life 8 (Ment et al., 2002). The use of magnetic resonance imaging (MRI) has emerged as a potential tool for providing prognostic information. MRI within the first several days of postnatal life can be helpful in determining the pattern of injury. Identifying the specific pattern of injury the infant sustained may help the providers determine the frequency of outpatient monitoring.

Parents and health care providers must make decisions about resuscitation, experimental interventions, and medical management for the infant with HIE within hours of delivery. While more extensive diagnostic and prognostic indicators of the extent of the hypoxic ischemic insult are available over a period of hours and days following delivery, the initial diagnostic information available is limited to the initial physical examination, biochemical evaluation of umbilical cord gases, and possibly
aEEG (White et al., 2012). In addition to the limitations in diagnostic and prognostic evaluations for making immediate treatment decisions, the pathophysiologic changes in the brain are also occurring over hours to weeks, leaving parents and providers to make treatment decisions that impact the quantity and quality of life for the infant with little knowledge about the severity of the insult.

### 1.1.4 Managing HIE

Depending on the severity of HIE, five types of treatment decisions must be made across the trajectory of illness for infants with HIE: initiation of life-sustaining treatment, initiation of experimental interventions, escalation of treatment, maintenance of treatment, and withholding/withdrawing support. This classification of decisions types across the trajectory of illness was developed from analysis of pilot data for an ongoing study about how parents and providers make decisions for infants with complex life-threatening conditions (extreme prematurity, complex congenital heart disease, and genetic disorders requiring stem cell transplant) (2008-2013, 1R01-NR010548, Docherty, PI). Decisions about the initiation of life-sustaining treatments include the extent of cardiopulmonary resuscitation, stabilization, and the types of treatments the infants need to support vital functions and prevent further injury. Experimental interventions are available for infants with HIE at some academic medical centers that may prevent secondary energy failure (Zhu et al., 2009) or potentially repair areas of the brain injured during primary injury failure (Pimentel-Coelho et al., 2012).
Thus decisions about transferring an infant to a center offering these experimental interventions, and which of these treatments is most appropriate for the infant, must be made. For infants who survive the initial weeks of life, parents and providers must make decisions about the types of technology they need to support respiratory and nutrition status and when to start physical, speech, and occupational therapy. Infants with severe HIE who required extensive resuscitation and had a high base deficit noted on the umbilical cord blood gas may need to transition to palliative care options rather than exposing them to the potential side effects of experimental interventions (Azzopardi, 2010). In some cases of severe, extensive injury, treatment for the infants may be transitioned from aggressive care to palliative care. For infants with HIE, the first 6 months of the life are the most intensive and complex for deciding which treatments need to be initiated for the infant. Additionally, at 6 months of age, infants can be evaluated for major developmental delays (Robertson & Perlman, 2006)

1.1.4.1 Initiation of Life-Sustaining Treatment

Initiation of life-sustaining treatments for infants with HIE aim to prevent further damage from hypoxia and ischemia by restoring oxygen and nutrients and potentially ameliorating injury from the secondary energy failure. The initiation of life-sustaining treatments for infants with HIE usually begins at birth because most infants require respiratory and cardiac support when transitioning from intrauterine to extrauterine life (Jacobs et al., 2011). The initial treatment is guided by the neonatal resuscitation protocol
(NRP), which recommends cardiopulmonary resuscitation for at least 10 minutes if an infant does not have an adequate heart rate (Kattwinkel et al., 2010). If HIE is suspected, the overhead heater should be turned off in the delivery room (Thoresen, 2008) because elevated temperatures increase the odds of death or disability in infants with HIE (Laptook et al., 2008). Infants who survive initial resuscitation are then transferred to the intensive care unit for further management and evaluation (Azzopardi, 2010).

Once in the intensive care unit, the extent of life-sustaining treatments needs to be determined including correction of acidosis, cardiopulmonary management, fluid balance and metabolic management, and monitoring for multi-organ failure and seizure activity (Azzopardi, 2010) and whether the infant will benefit from transferring to a facility that offers moderate hypothermia treatment for infants < 6 hours of age (known as moderate hypothermia treatment throughout the dissertation). The use of moderate hypothermia treatment as an initial life-sustaining treatment is common practice in Level IV neonatal intensive care units (NICU) in the United States (Kapetanakis, Azzopardi, Wyatt, & Robertson, 2009; Lang, Hartman, Hintz, & Colby, 2007). Moderate hypothermia treatment is thought to be effective because it reduces free radicals and glutamate levels, decreases oxygen demand, and decreases apoptosis (Roka & Azzopardi, 2010). Evidence of decreased mortality and improved neurological outcomes from moderate hypothermia treatment have only been found when infants are < 6 hours of age at treatment initiation (Shah, 2010; Shankaran, Pappas, et al., 2012). See Table 2 for
general criteria used to determine which infants may benefit from moderate hypothermia treatment.

**Table 2: Moderate Hypothermia Treatment Criteria**

<table>
<thead>
<tr>
<th>Moderate Hypothermia Treatment Criteria</th>
</tr>
</thead>
<tbody>
<tr>
<td>- &gt; 36 weeks gestational age and &gt; 1800 grams at birth</td>
</tr>
<tr>
<td>- pH &lt; 7.0 or base deficit &gt; 16mEq/L or</td>
</tr>
<tr>
<td>- History of acute perinatal event and either 10 minute Apgar scores &lt; 5 or need for continued ventilation</td>
</tr>
<tr>
<td>- Clinically determined to have moderate or severe HIE</td>
</tr>
</tbody>
</table>

(Shankaran et al., 2005)

Initiation of moderate hypothermia treatment at < 6 hours of life appears to decrease death or major disabilities by 2-years of age (Eicher et al., 2005; Gluckman et al., 2005; Jacobs et al., 2011; Shankaran, 2009; Simbruner et al., 2010). However, infants with HIE who receive moderate hypothermia treatment continue to have mortality rates of 25-38% and severe disability rates of 21-35% (Jacobs et al., 2011; Shankaran, Pappas, et al., 2012; Simbruner et al., 2010). However recently, one of the first randomized clinical trials (National Institute of Child Health and Human Development) comparing whole body moderate hypothermia (n = 97) and standard care (n = 93) in infants with HIE initiated < 6 hours of age found no significant difference in the primary outcome of death or IQ < 70 in the same children at 6-7 years, even when severity of HIE was
considered (Shankaran, Pappas, et al., 2012). Additional large clinical trials that used selective head cooling with mild systemic hypothermia (CoolCap) or whole body hypothermia (TOBY) have not published outcomes of their participants at 6-7 years of age, at this time. Whether moderate hypothermia or selective head cooling treatment improves childhood outcomes of infants with HIE remains uncertain.

Moderate hypothermia treatment is delivered through either selective head or whole body cooling of the infant (Shah, 2010). The treatment involves decreasing the infant’s body temperature to between 33°C (Eicher et al., 2005) and 36.5°C (Gunn, Gluckman, & Gunn, 1998). Infants are generally cooled for 48 to 72 hours and then rewarmed slowly to prevent complications (e.g., hypotension) (Long & Brandon, 2007). The induced systemic hypothermia can lead to systemic side effects including dysrhythmias, hypotension, coagulopathy, thrombocytopenia, renal failure, hepatic issues, infection, and pulmonary hypertension (Shah, 2010).

Despite the acceptance of moderate hypothermia as common practice in level IV neonatal intensive care units, evidence as to what is the best treatment protocol remains unknown. The first challenge of using moderate hypothermia treatment is how to administer the treatment. There are no studies that compared selective head cooling to whole body cooling; therefore, the comparative effectiveness remains unknown. Additionally, the optimal hypothermic temperature that slows the progression of injury and limits the systematic complications of hypothermia has not been explored in infants.
The length of time the infant should receive moderate hypothermia therapy is also not known. Therefore, many aspects of therapy delivery remain unknown. The second limitation is there is only 1 trial that has data about the effects of the hypothermia on outcomes of children at 6-7 years of age (Shankaran, Pappas, et al., 2012). The ability to generalize long-term developmental outcomes based on a single trial is limited. Without these data, providers do not know if the treatment will result in better long-term outcomes for infants than standard treatment.

1.1.4.2 Initiation of Experimental Interventions

Experiment interventions are available that may slow the progression of the secondary energy failure within the brain (Allen & Brandon, 2011). The two experimental interventions available for the sample of infants with HIE at the site of this study include the experimental intervention of moderate hypothermia initiated $> 6$ hours of age and autologous umbilical cord stem cell transplant. The randomized clinical trial using the experimental intervention of moderate hypothermia enrolls infants with HIE who are greater than 6 hours of age when the experimental intervention is initiated. The decision to transfer an infant becomes more complex and possibly creates more uncertainty because parents and providers do not know if the infant will be randomized to the experimental intervention (moderate hypothermia) or the control group.
Autologous umbilical cord blood transplant is the second experimental intervention offered to infants. Animal model data supports that stem cell transplantation following a hypoxic ischemic injury to reduce dying neurons in basal ganglia but not in the cerebral cortex, inhibit apoptosis in the basal ganglia, decrease the size of the cerebral lesion, and reduce microglial activation leading to decreased inflammation (Pimentel-Coelho et al., 2010; van Velthoven, Kavelaars, van Bel, & Heijnen, 2010a, 2010b). A phase I clinical trial the umbilical cord blood study evaluates the safety of blood reinfusion in infants with HIE (Cotten, 2009). The decision to harvest the umbilical cord blood is made by parents prior to or around the time of delivery. The stem cells from the umbilical cord blood is infused within the first couple of days of life and the infant receives 2-3 additional infusions within the first 14 days of life. Decisions to implement either experimental intervention require transfer to the academic medical center.

1.1.4.3 Escalation of Treatment

For infants with moderate or severe HIE, parents and health care providers may need to consider advanced technologies to provide respiratory and nutrition support by escalating treatments provided. The exact treatments these infants need remain unknown. Placement of a gastrostomy tube may be needed if the infant is unable to achieve adequate oral intake despite therapy. The treatment is considered an escalation because the first line therapy failed to help the infant maintain adequate nutrition. Some
infants with HIE may require long-term ventilator support. The infant may initially fail extubation attempts and require a more permanent solution through placement of a tracheostomy. Placement of a tracheostomy tube may also be discussed if the infant has been intubated and on a mechanical ventilator for several weeks without demonstrating substantial improvement in respiratory status. Exactly how and when treatment is escalated is unknown in infants with HIE.

1.1.4.4 Maintenance of Treatment

Regardless of whether the infant receives experiment therapy, the severity of the evolving brain injury (mild, moderate, or severe) determines the types of treatments and therapies infants need to reach optimal physical and developmental outcomes. Infants with mild HIE who are feeding without difficulty and exhibit no hypotonia can be discharged and receive the same follow-up as other infants without brain injuries. If the infant exhibited hypotonia, the infant will most likely need to be followed in a developmental clinic for continued assessment and referral to services when necessary (Robertson & Perlman, 2006). Parents and providers making treatment decisions for infants with mild HIE will generally not make additional decision-making about HIE because the mild injury most likely will not effect physical and cognitive functioning.

Follow-up in a neurodevelopmental clinic is advised to ensure early intervention for infants with moderate and severe HIE. Assessment in the neurodevelopmental clinic generally occurs at 4-8 months, 12-24 months, 3-5 years, and 8-10 years of age. The
scheduling coincides with time points when impairments can be detected including severe motor or sensory loss (12 months), low developmental quotient (24 months), fine and gross motor function (2-4 years), abnormalities in cognitive function (4-7 years), and learning disabilities (7-9 years) (Robertson & Perlman, 2006). Referrals to specialists (e.g., physical, speech, and occupational therapists) can be made during these appointments to ensure that these infants receive early intervention.

1.1.4.5 Withdrawing/Withholding Life-Sustaining Treatment

For some infants with severe, extensive hypoxic-ischemic injury, parents and health care providers must decide whether to transition from curative treatment to symptom-focused palliative care treatment. This transition can occur within a couple days of birth or following hypothermia treatment (Azzopardi, 2010). A single indicator does not exist to allow the provider to be certain the infant will have severe disability or die. Infants who remain in the severe encephalopathic stage for several days, have extremely low or absent voltage on the EEG (Azzopardi, 2010), or a basal-ganglia-thalamus pattern on MRI (de Vries & Jongmans, 2010) generally have a poor prognosis. Sarkar et al. (2010) found that following hypothermia treatment, infants with Apgar scores of 0 at 10 minutes were significantly more likely to die from HIE than infants with Apgar scores > 0 at 10 minutes of life. At best, health care providers may be able to use the degree of encephalopathy, clinical indicators, and neuroimaging as prognostic
indicators to predict the impact of HIE on short and long-term outcomes to aid parents in making decisions about palliative care treatment (Sarkar et al., 2010)

1.1.5 Long-term Outcomes of HIE

The long-term outcomes for infants with HIE are dependent upon the pathophysiologic effects of primary and secondary energy failures. The excitotoxicity during the secondary energy failure phase can lead to hearing, vision, somatosensory function, learning and memory deficits (Fatemi et al., 2009). Seizures are a result of the damage from injury and infants with HIE who have seizures experience poorer motor and cognitive outcomes than those who do not experience seizures (Glass et al., 2009; Pisani et al., 2009). The evolving injury from HIE appears to be more related to apoptosis than necrosis (Ferriero, 2004). The reason for this is because apoptosis is a normal part of critical pathway development of the brain, but if apoptosis is altered, the development of the brain is also impaired (Cotten & Shankaran, 2010). When these pathways are disrupted, a broad range of neurologic dysfunction may be observed. Excitotoxicity, seizures, and apoptosis can lead to long-term disabilities. Determining the areas of the brain injury and the severity of the injury may allow parents and providers to better predict long-term outcomes.

1.2 Aspects of HIE that may Impact Parent and Provider Decision-Making

Most mothers of infants with HIE deliver a full term infant without congenital anomalies, and have had expectations of a healthy infant. Most mothers have a history
of normal prenatal ultrasounds, normal gestation (38-42 weeks), and anticipation of an uncomplicated delivery of a healthy infant with a normal birth weight. Within seconds to minutes following delivery parents and health care providers must make decisions about the initiation of life-sustaining treatment (e.g., extent of cardiopulmonary resuscitation) and experiment interventions for infants with HIE.

Parents experience intense stress and shock after giving birth and learning that their ‘normal’ infant has suffered brain damage (Rosenthal, Biesecker, & Biesecker, 2001; Wilson & Miles, 2001). Research shows parents have difficulty processing information under such stress (Alderson, Hawthorne, & Killen, 2006; Kavanaugh, Savage, Kilpatrick, Kimura, & Hershberger, 2005; Pector, 2004; Yee & Ross, 2006). Despite the evidence of stress and shock parents must make decisions about initiation of life-sustaining and experimental interventions within 6-24 hours of birth as this is a critical window of time in which to potentially prevent further organ damage and neurological injury (Hoehn et al., 2008; Shalak & Perlman, 2004; Shankaran, 2009; Stola & Perlman, 2008). Providers must assist parents in making decisions about initiating life-sustaining treatments, especially the extent of treatments available and possibly the need to escalate treatments. Providers must ascertain, with limited prognostic information, whether the infant may benefit from experiment interventions. Very little is known about how parents and providers decide to initiate life-sustaining and experiment interventions, when and to what extent treatments are escalated, how therapy for infants is maintained over the first
6 months of life, and when transitioning from aggressive care to palliative care is better for infants with HIE. How parents and providers approach decision-making for infants with HIE with limited knowledge of infant outcomes remains unknown.

There is some evidence regarding influences on parent and provider decision-making for infants with other life-threatening conditions. For example, factors such as severity of illness (Balkan et al., 2010) and the relationship between parents and providers (Grobman, Kavanaugh, Moro, DeRegnier, & Savage, 2010), including trust (Woodgate & Yanofsky, 2010) influence decision-making. The quality of the exchange of information affects the relationship between parents and providers and can determine the type of treatments pursued (Alderson et al., 2006; Pector, 2004; Quine & Rutter, 1994; Wocial, 2000).

Evidence from decision-making about other life-threatening conditions suggest parents need providers to present information across multiple meetings with compassion and the provision of hope (Charchuk & Simpson, 2005; Payot, Gendron, Lefebvre, & Doucet, 2007; Wocial, 2000). Parents need the information presented in a clear comprehensible manner. Explaining a condition such as a complex brain injury can be challenging as parents are often distressed and providers have limited ways to show direct evidence of the brain injury. The direct evidence of abnormalities such as symptoms and physical examination findings as seen in other conditions is lacking for infants with HIE (Bellali & Papadatou, 2007; McHaffie, Lyon, & Fowlie, 2001; Wocial,
In the absence of obvious evidence of an abnormality, parents have difficulty accepting information (Bellali & Papadatou, 2007). For example, parents of extremely premature infants can see the fragility and acuity through the very small size of the infant; often a parent cannot "see" a brain injury. The injury takes time to develop and while infants with HIE do have clinical signs and symptoms; parents often see these visible signs as normal infant fatigue and sleepiness. For parents of infants with HIE limited information exists about how to explain the hypoxic-ischemic injury and the optimal time for discussing the extent of the injury.

Providers need information about the severity of HIE to determine what options to offer to parents. However, providers are limited in their ability to predict the physiologic and developmental outcomes for infants with HIE because HIE evolves over hours to days (Cotten & Shankaran, 2010; Ferriero, 2004). While the results of experiment interventions appear to have short-term benefits, the outcomes of school-aged children did not suggest moderate hypothermia decreased mortality or IQ scores < 70 compared to those who did not receive a hypothermia intervention (Shankaran, Pappas, et al., 2012). The uncertainty of how the injury impacts outcomes makes advising and providing parents with information difficult for providers. Exactly how providers deal with these uncertainties, communicate the information, and provide recommendations to parents is not known.
Because of the sequelae of HIE, including severe cognitive dysfunction, epilepsy, and cerebral palsy (CP), infants with HIE who survive the acute phase generally require long-term care (Hoehn et al., 2008; Long & Brandon, 2007; Okereafor et al., 2008). Additionally they may be dependent on technology because of challenges related to nutrition and ventilation (Volpe, 2008). Parents need information on how to care for infants dependent on technology including how to access services such as home health care and obtain the supplies needed. The infant may also have motor and developmental delays (Volpe, 2008). To treat these delays, speech, physical, and occupational therapists may be involved in the care of the infant. How parents and providers effectively access these services and continue to support the infant with HIE remains unknown. Because of the infant’s profound needs, parents and providers must determine how and when to access services, and how to interact with each other (Minnes & Steiner, 2009; Rentinck, Ketelaar, Jongmans, & Gorter, 2006).

In addition to understanding the severity of the illness and potential impact on infant outcomes, evidence suggests that other factors influence parent and provider decision-making for infants with complex conditions. These factors include stress and anxiety (Alderson et al., 2006; Kavanaugh et al., 2005; Pector, 2004; Yee & Ross, 2006), hope (Boss, Hutton, Sulpar, West, & Donohue, 2008; Kavanaugh et al., 2005), race (Blackhall et al., 1999; Hallenbeck & Goldstein, 1999; Kagawa-Singer & Blackhall, 2001; Miles, Burchinal, Holditch-Davis, Brunssen, & Wilson, 2002; Moseley et al., 2004), and
religiosity and spirituality (Haward, Murphy, & Lorenz, 2008; Plotz, van Heerde, Kneyber, & Markhorst, 2008; Sprung et al., 2007). However, how each of these factors impacts parents and providers across the acute trajectory for infants with HIE remains unknown. Additionally, how these factors change over time as parents and providers make decisions about initiation of life-sustaining treatment, initiation of experimental interventions, escalation of treatment, maintenance of treatments, and possibly withdrawing/withholding care remains unknown.

1.3 Theoretical Foundation

The advent of advanced technology and complex interventions to treat infants, who would have previously died, has increased survival, but has also created more treatment options for parents and HCPs to choose from for critically ill infants. The availability of immediate and long-term treatments for infants with HIE may have created a complex decision-making trajectory for parents and HCPs. As with other trajectories, the individuals involved in making the decisions, their previous experiences, and personal characteristics may effect how the decision-making trajectory progresses (Magnusson & Cairns, 1996). The initial decision and infant’s response to continued resuscitation (e.g., chest compressions) and immediate stabilization (e.g., respiratory support) affects the next decisions. Figure 1 depicts the predicted decision-making trajectory for infants with HIE within the first week after birth. The infant’s
response to each treatment chosen will impact the decisions available as the infant progresses through the illness trajectory.

Figure 1: Predicted Decision-Making Trajectory for First Week of Life
Figure 2: Predicted Decision-Making Trajectory after First Week of Life

Figure 2 represents the possible decisions that may be made once the infant is initially stabilized. The main decisions after initial week of life will reflect the extent of the damage from HIE and the infant’s response to previous treatment decisions. The infant responses required prior to discharge are adequate nutrition and ventilation, which can be accomplished through different methods, if necessary. After discharge infants will require follow-up in developmental clinics initially. Some infants may also need physical therapy depending on the severity or area of brain injury. Some infants may continue to remain unstable or have severely impaired cognitive function and the decision may be made to transition from aggressive care to palliative care. However,
who makes these decisions and what factors influence how the decisions are made remains unknown.

A case study methodology was chosen to allow for the complexity and context of the decision-making to be explored through multiple sources and types of data (D. J. Edwards, F. M. Dattilio, & D. B. Bromley, 2004). This approach will allow for in-depth exploration of whom, how, and why decisions are made for infants with HIE. Additionally, decision-making for infants with HIE develops over time, the use of multiple types and sources of data is needed to fully capture the context of decision-making trajectory (D. J. Edwards et al., 2004). Examining the convergence of multiple sources of data related to decision-making by parents and health care providers will allow for the examination of processes used to make decisions (Gerring, 2007a).

1.4 Potential Implications

An exploration of the parent and health care provider decision-making trajectory will provide critical information about the types of influences that impact how and when decisions are made for infants with HIE. This study will generate key hypotheses that can be tested in future studies. Eventually these studies will lead to the development of interventions to optimize decision-making for infants with HIE. For example, previous research suggests that parents and providers disagree about the neurological status of infant prior to withdrawing/withholding treatments (Garros, Rosychuk, & Cox, 2003). The disagreement may create communication breakdowns by
providers not being as hopeful as parents or viewing the quality of the infant’s life as minimal. The contrast of parental and provider appraisals of neurological outcomes can lead to conflict and parents advocating against treatments suggested by the providers (Garros et al., 2003). Understanding how and why decisions and treatments are initiated from birth through the first 6 months of life may allow for identification of when in the decision-making trajectory becomes disconnected between parents and providers.

Additionally, identifying what types of treatments are needed and if or when provider treatment recommendations are not understood by parents or not implemented due to conflicting views of what is best for the infant. Ensuring infants are provided with needed therapies may prevent parents and health care providers from regretting their decisions, and possibly reducing the $51.2 billion financial costs for individuals with neurologic disabilities (CDC, 2004).

1.5 Purpose Statement and Specific Aims

Therefore, the overall goal of this dissertation was to understand how parents and health care providers make decisions across the immediate (first 48 hours) and short-term trajectory (next 6 months) for infants with HIE. To accomplish this goal, there are four aims for this study. Each chapter of the dissertation is devoted to a single aim. The first aim was to conduct an integrated literature review to describe the empirical evidence related to influences on parent decision making for infants and children with life-threatening illnesses. Thus Chapter II presents a systematic review of the empirical
evidence to determine themes, critique research designs, and offer further recommendations for research. The second aim was to examine the ethical issues inherent in interviewing parents about sensitive topics (i.e., life-threatening illness and palliative care). Since the study of decision-making for infants with HIE involved collecting intensive, in-depth data with parents during a very difficult and stressful time of life, understanding the impact of the study was important. Therefore, in Chapter III data from an ongoing longitudinal study about parent and health care provider decision-making was analyzed using the lens of the ethical principles of autonomy and nonmaleficence to explore the impact on parents of data collection regarding sensitive topics.

The third aim was to explore the trajectory of parent and health care provider decision making for infants with HIE, across the immediate trajectory (first 72 hours) and short term trajectory (next 6 months). The exploration of the trajectory of decision-making, using a multiple case study design, allowed for examination of how, why, and when parents and health care providers make decisions for infants with HIE. A case study design was chosen because it allowed for description, explanation, and illumination of the complex, real-life phenomena. This design enabled critical factors and interrelationships of parent and provider decision-making for infants with HIE to be explored (Gerring, 2004; Yin, 2009). Chapter IV presents an in-depth case study of the immediate trajectory (first 48 hours) and short-term trajectory (next 5 months) for one
infant with HIE. The fourth aim was to explore the similarities and differences across decision-making trajectories of parents and providers for infants with HIE. Chapter V is a study of 10 cases of decision-making for infants with HIE over the first 6 months of life. An exploration of the trajectory of illness and its impact of the decision making trajectory is executed through an across case comparison. A complete summary of the dissertation results are also presented with implications for nursing practice and research detailed to further improve the treatment of infants with HIE.
2. Parental Decision-Making for Infants and Children with Complex Life-Threatening Conditions: An Integrated Literature Review

2.1 Introduction

Many infants and children with complex life-threatening conditions (CLTC) who earlier would have died in the first months of life are now living long enough to receive highly complex treatments. Children with complex life threatening conditions generally require extensive or multiple hospitalizations within the 1st year following diagnosis and likely result in chronic conditions, if death does not occur within the first, such as extreme prematurity, complex cardiac anomalies, and metabolic conditions (Brandon, Docherty, & Thorpe, 2007). These conditions require complicated life-sustaining treatments (e.g., heart transplant), require extended hospitalization, have high rates of mortality within 1 year following diagnosis, and for those infants who survive high rates of chronic illness (Feudtner, Feinstein, Satchell, Zhao, & Kang, 2007). Advances in technology and treatment modalities have vastly improved the survival rates for infants and children (hereafter referred to as children) who would have previously died from complex life threatening conditions (Morris, 2009; Ortenstrand et al., 2010). These technological and treatment advances bring challenges for parents and health care providers (HCPs) who must make the difficult decisions when caring for children with CLTCs about whether to initiate and proceed with curative models of care, when to escalate curative care, how to maintain the treatments, and if or when to transition to
palliative (Docherty, Miles, & Brandon, 2007). The decision-making process for parents and HCPs is a trajectory that begins at diagnosis and continues throughout the child’s life with each decision effecting subsequent decisions and impacting the child’s future.

The decision-making trajectory for children with CLTCs may begin prenatally when fetal diagnostic and imaging studies give parents and HCPs information about a possible CLTC that may elicit making a choice between whether to terminate or continue a pregnancy (Rempel, Cender, Lynam, Sandor, & Farquharson, 2004). For others, the decision-making process does not begin until birth or at the time of a later diagnosis when it must be decided whether to initiate curative care or proceed to palliative care (Grobman et al., 2010). For children who struggle to survive the initial stage of the illness, parents and HCP must determine the extent to which they are willing to escalate life-sustaining treatments (Bluebond-Langner, Belasco, Goldman, & Belasco, 2007) and engage in experimental interventions (Woodgate & Yanofsky, 2010). If they survive, some children require technological interventions and support (e.g., home ventilation) to sustain life (McNamara, Dickinson, & Byrnes, 2009). Other children do not respond well to initial treatments, the CLTC progresses and parents and HCP are faced with choosing when and how to withdraw or withhold support (Michelson et al., 2009). All of these types of decisions are challenging for parents and HCPs because some of the choices inflict pain and/or shorten the duration of the child’s life (Sharman, Meert, & Sarnaik, 2005), and other choices have a profound impact on parental and health care
system resources (Michelson et al., 2009). While all of these decisions are complex and dependent upon previous experiences and infant responses, they also require parent and HCP to work together to determine the optimal treatment for the child. However, little research exists that has explored the dynamic of decision-making of parents and providers across a child’s illness trajectory.

The development of knowledge about parent and HCP decision-making is important because parental conflict and parental regret about decision-making could lead to psychological distress, decreased physical health, and decreased quality of life of the parent (Brehaut et al., 2003; Korenromp et al., 2005). Parental regret of decisions led to psychological stress (pathological grief, post-traumatic stress symptoms) at more than 2 years after the decision was made (Korenromp et al., 2005). The psychological stress, particularly following the death of a child, increased parent mortality (Harper, O’Connor, & O’Carroll, 2011; Li, Precht, Mortensen, & Olsen, 2003), mental illness (Li, Laursen, Precht, Olsen, & Mortensen, 2005), and morbidity (Olsen, Li, & Precht, 2005).

While most decisions along the illness trajectory for infants and children with life-threatening conditions involved an integrated decision-making process between parents and HCPs, the research to date has focused on either parents or HCPs with few studies exploring the interactions between parents and HCPs within the context of decision-making about the same child. The aim of this integrated literature review is to explore possible factors that affect parental decision-making and how these factors play
Parents of children with life-threatening illnesses are faced with a range of choices that occur during the illness trajectory and are dependent upon diagnosis and stage of the disease. Thus an orienting framework that categorizes decisions based upon their location along the trajectory of illness was used in this review. The decision categories were developed in an ongoing study of decision-making and illness trajectories of infants and children with life-threatening illnesses (2008-2013, 1R01-NR010548, Docherty, PI). The categories allow factors impacting parental decision-making at particular points along the illness trajectory to be compared across various diagnoses for a more comprehensive understanding about the decision-making process.

See Table 3 for definitions of treatment decision types and examples of each treatment decision type. The examples of decisions are dependent upon the child’s previous illness course. For example, a child born with a congenital heart defect may require placement of a gastrostomy tube prior to being discharged from the hospital initially because he was unable to feed appropriately despite speech therapy. Escalating to the placement of a gastrostomy tube is necessary for the child because the primary treatment was not effective. However, placement of a gastrostomy tube in a child diagnosed 4 years ago
with cystic fibrosis (CF) may be considered maintenance treatment because the child needs to gastrostomy tube to maintain adequate nutrition because he simply cannot eat enough to maintain adequate nutrition. The specific examples of decisions may vary depending on the situation of the child and how the illness trajectory has progressed.
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<th>Type of Treatment Decision</th>
<th>Definition</th>
<th>Examples of decisions</th>
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| Aggressive Care            | Care focused on curing disease or illness | • Proceed with pregnancy following a prenatal diagnosis of a severe chromosomal or genetic abnormality  
• Proceed with cardiac surgery for a major congenital malformation |
| Initiation treatment       | Decisions about the initial treatment plan for the infant or child based on the illness or condition | |
| Experimental interventions | Decisions about treatments and medications that are not considered standard treatment for an illness or condition | • Participation in experimental trials (e.g. stem cell transplant for a condition not yet standard of care) |
| Escalation of treatment    | Decisions about addition of medical treatments (e.g., cardiopulmonary resuscitation and medications) and technologies (e.g., intubation and placement on mechanical ventilator, additional chemotherapy agents) in an attempt to treat a worsening illness or condition | • Change from a conventional ventilator to a high frequency oscillator |
| Maintenance of treatment   | Decisions about whether and how to maintain the level of care for an infant or child who has received initial and/or escalation of treatment | • Gastrostomy tube feedings  
• Humidification for ventilators  
• Home ventilators |
| Palliative care            | Decisions about non-curative treatments that focus on relief of suffering | |
| Withdrawing/withholding treatment | Decisions about how much treatment to provide for a child | • Withdrawal of ventilator  
• Do not resuscitate orders |
| End-of-life                | Decisions about specific details of circumstances surrounding death | • Physician-assisted death  
• Location of death |
2.2 Methods

PubMed, Cumulative Index of Nursing and Allied Health Literature (CINAHL), and PsycINFO were searched using the combined key terms ‘parents and decision-making’ to obtain English language publications from 2000 to May 2012. The time frame was chosen because the success in treatment for infants and children with life-threatening illnesses has improved substantially in the past 10 years (Bell, 2007). In addition, the level of involvement of parents in the decision-making process has changed due to the influence of family centered care and the endorsement of involving individuals in their health care (Kon, 2010; Malusky, 2005; van den Brink-Muinen et al., 2006).

The search strategy generated 2509 articles. The inclusion criteria were studies of factors impacting parental decision-making for infants and children with life-threatening illnesses. Infants and children were defined as those < 12 years of age, including fetuses. The exclusion criteria were studies of decisions about non-life-threatening illnesses, decisions about organ donation, consenting process with experimental interventions, legal reports and rulings on decision-making, child participation in decisions, and non-data based articles. After reviewing the abstracts articles that met inclusion criteria were extracted for full review. Excluded articles did not discuss factors impacting decision-making, explored decisions about non-life-
threatening illnesses or conditions (e.g., vaccinations, infant feeding), were non-data based, or only described parental response to the child’s life-threatening illness.

### 2.3 Results

Using the orienting framework of decision categories, synthesis of the findings from the 43 articles was first categorized by type of decision. Then within each decision category findings were organized by factors impacting decision-making. The factors were grouped into themes by each type of decision. The themes included information needs, seriousness of illness, no other treatment options, child’s best interests, religiosity and spirituality, parental characteristics and past experiences, and emotional support. The themes of “child’s best interests” and “support” did not appear to vary across decision type and thus the results were collapsed into one section called additional influences.

#### 2.3.1 Initiation of Life-Sustaining Treatment

Initiation decisions are critical because the type or level of treatment to initiate for an infant with a CLTC impacts the full trajectory of the child’s illness course and influences future decisions parents make for their child (Menahem & Grimwade, 2003). This section explores what factors influenced parental decision-making when deciding whether to initiate life-sustaining treatment.
2.3.1.1 Information Needs

Parents needed information about their child’s illness or condition (Grobman et al., 2010; Moro et al., 2011) and treatment options (Hinds et al., 2000). However, being in a state of emotional shock after receiving the diagnosis of a life-threatening illness (Boss et al., 2008; Lan, Mu, & Hsieh, 2007; Payot et al., 2007; Vandvik & Forde, 2000) limited understanding and comprehension of the illness and treatment options (Boss et al., 2008; Chaplin, Schweitzer, & Perkoulidis, 2005; Grobman et al., 2010; Partridge et al., 2005). Thus it was important to know what types of information parents needed and how to effectively communicate this relevant information.

The information parents needed when deciding to initiate life-sustaining treatments included the seriousness of the illness or condition (Balkan et al., 2010; Chaplin et al., 2005; Lam et al., 2009), whether there were any structural or chromosomal abnormalities (Ahmed et al., 2008; Rempel et al., 2004; Zyblewski et al., 2009), how the illness impacted the child’s quality of life (Ahmed et al., 2008), and whether the child would be ‘normal’ (Ahmed et al., 2008). Information about the benefits and adverse effects of specific treatments (Hinds et al., 2000) with opportunities to ask and have questions answered was also necessary (Kavanaugh, Moro, & Savage, 2010). Parents sought and/or relied on the HCPs’ knowledge and opinion about what treatment options were best for the child (Partridge et al., 2005; Rempel et al., 2004) and what scientific evidence supported the efficacy of the treatment (Ellinger & Rempel, 2010;
Rempel et al., 2004). Having written and electronic materials and information from organizations specific to the child’s illness and treatment options were also viewed as helpful (Chaplin et al., 2005; Grobman et al., 2010; Redlinger-Grosse, Bernhardt, Berg, Muenke, & Biesecker, 2002).

In addition to needing information, parents reported that the way the information was presented also impacted their decision-making. Information presented multiple times in a clear, honest manner with limited jargon was helpful (Grobman et al., 2010). Parents needed to feel that HCPs were compassionate and hopeful as these behaviors demonstrated the HCPs respected their child as an individual, instead of a ‘protocol’ (Boss et al., 2008; Redlinger-Grosse et al., 2002). Objective and neutral communication from HCPs left parents feeling that HCPs had little hope of a positive outcome (Payot et al., 2007; Rempel et al., 2004). The lack of hopeful communication leads to a strained relationship between the parents and HCPs because parents are still hoping for their child to be cured of the illness (Edwards et al., 2008), while they thought providers had ‘given up’ (Boss et al., 2008). Opposite assessments of hope can create mistrust between parents and providers, which leaves parents to advocate for their child by protecting against the perceived negative recommendations of HCPs (Boss et al., 2008).

Whether or not parents received enough information to make a decision varied. For some parents who chose to terminate a pregnancy of a fetus with a prenatal
diagnosis of congenital heart disease, receiving enough information allowed them to make the decision and felt they had a good or excellent understanding of the anomaly (Menahem & Grimwade, 2003). However, for parents that continued the pregnancy, 20% reported that they received inadequate information about the anomaly and more than 50% reported having at most only an adequate understanding of the anomaly (Menahem & Grimwade, 2003).

2.3.1.2 Seriousness of Illness

The seriousness of the illness and predicted long-term outcome of the child influenced parental decisions to initiate life-sustaining treatments. Parents considered the extent of congenital anomalies and the presence of chromosomal abnormalities when deciding whether to terminate or continue a pregnancy (Chenni et al., 2012; Menahem & Grimwade, 2003; Rauch, Smulian, DePrince, Ananth, & Marcella, 2005; Zyblewski et al., 2009). The severity of the heart defect (Chenni et al., 2012) and the presence of a chromosomal abnormality were associated with proceeding or terminating the pregnancy when a heart defect was identified (Menahem & Grimwade, 2003; Zyblewski et al., 2009). One study found that the presence of multiple anomalies rather than a single anomaly led parents to terminate a pregnancy (Rauch et al., 2005). Multiple anomalies would likely increase the morbidity for infants (Rauch et al., 2005). However, other parents reported that the HCPs’ prediction about the seriousness or negative prognosis for the child did not affect their decision (Boss et al., 2008; Partridge et al.,
2005) and parents could not consider pursuing any initial treatment that was not curative regardless of the condition (Ellinger & Rempel, 2010; Vandvik & Forde, 2000).

An important aspect of the seriousness of illness was the extent to which the condition would affect the child’s quality of life if life-sustaining treatment were initiated (Ahmed et al., 2008; Lam et al., 2009). Poor quality of life was defined as suffering, limitation of both physical and emotional well-being, and not having a ‘normal’ life (Ahmed et al., 2008). Suffering was described as physical and emotional pain. The physical and emotional pain the child may endure also affected decisions about initiation of aggressive treatment (Ahmed et al., 2008; Moro et al., 2011). Physical pain could come from the treatments the child endured (Hinds et al., 2000) while the emotional pain was related to the child not being able to ‘enjoy’ life because of the seriousness of the condition (Ahmed et al., 2008). Emotional pain could come from the child having visible signs of a condition. Parents felt that society would treat the child poorly and the child would suffer emotionally (Ahmed et al., 2008). The suffering the child could endure led parents to terminate a pregnancy, even if religious beliefs prohibited it (Ahmed, Atkin, Hewison, & Green, 2006; Ahmed et al., 2008; Menahem & Grimwade, 2003; Vandvik & Forde, 2000).

Knowing the infant’s life would be shortened (Rauch et al., 2005) or that the infant had no chance of survival (Chaplin et al., 2005; Menahem & Grimwade, 2003) influenced parents’ decision to terminate pregnancy. Any amount of time that they
could spend with their infant was valued, while others felt that it was more difficult to cope with the infant’s death after birth than when terminating the pregnancy (Ahmed et al., 2008).

Long-term outcomes of the CLTCs were also discussed with parents during the decision-making process (Boss et al., 2008; Partridge et al., 2005). During these conversations, parental perception of normality as a factor that impacted their decision-making (Ahmed et al., 2008). A normal life was described qualitatively as the infant could be happy and interact with the environment; the child could cope with the condition, and would be able to lead an independent life (Ahmed et al., 2008; Chaplin et al., 2005; Lam et al., 2009).

2.3.1.3 No Other Option

Parents chose to initiate life-sustaining treatment because they felt they had no other options. Parents described that even when other options were available, they were seen as not acceptable (Hinds et al., 2000; Vandvik & Forde, 2000) because the other option would result in certain death of their child (Vandvik & Forde, 2000).

2.3.1.4 Religious Beliefs

Religious beliefs prohibited parents from choosing termination of pregnancy (Ahmed et al., 2006; Chaplin et al., 2005; Ellinger & Rempel, 2010; Redlinger-Grosse et al., 2002); particularly Muslims and Christians. However, parents felt that even if their religion dictated a certain decision, the parent would consider what they felt was best
for their child (Ahmed et al., 2006) because religious leaders did not understand the illness (Ahmed et al., 2006). Other parents relied on their personal belief system about the sanctity of life and accepting their child diagnosed with a severe congenital defect (Redlinger-Grosse et al., 2002). Religion and spirituality provided a sense of a possible miracle for the child (Boss et al., 2008).

2.3.1.5 Parental Characteristics

Researchers studied how maternal characteristics (e.g., education level, age) influenced decision-making. Mothers of premature infants who had secondary education levels were more likely than mothers with primary education to attempt save an infant at ‘all costs’ (Lam et al., 2009). Several other variables were explored in four studies (Chenni et al., 2012; Lam et al., 2009; Rauch et al., 2005; Zyblewski et al., 2009) with mixed results on whether certain demographic characteristics influenced decision-making. Factors that remained inconclusive were maternal age, maternal gravida, maternal parity, race/ethnicity, and gender of the fetus (Chenni et al., 2012; Lam et al., 2009; Rauch et al., 2005; Zyblewski et al., 2009).

2.3.2 Initiation of Experimental Interventions

The decision as to whether to pursue experimental interventions for children with CLTC was often a last resort to treat or ameliorate the effects of an incurable condition.
2.3.2.1 Information Needs

Three studies (Culbert & Davis, 2005; Snowdon, Elbourne, & Garcia, 2006; Woodgate & Yanofsky, 2010) addressed factors impacting parental decision-making about experimental interventions. Deciding to initiate experimental interventions was emotionally difficult for parents who experienced fear, shock, and anxiety when the child was diagnosed with a CLTC and experimental interventions were offered (Snowdon et al., 2006; Woodgate & Yanofsky, 2010). The emotional distress led to difficulty integrating information about the diagnosis and participation in clinical trials (Woodgate & Yanofsky, 2010). Two studies evaluated parents’ preferred timing of information delivery. To understand when to present information to parents about the potential for enrollment in clinical trials on neonatal resuscitation, Culbert and Davis (2005) used hypothetical scenarios with parents in their second trimester, parents with infants who required admission to the neonatal intensive care unit, and parents with infants who experienced no complications. Results suggested that parents wanted verbal and written information about the treatment including the benefits and risks delivered by HCPs they trusted (Culbert & Davis, 2005). Parents wanted to be able to ask questions about the clinical trial and have them answered honestly (Culbert & Davis, 2005). When experimental interventions could be discussed prior to delivery of an infant, parents preferred the information be presented during prenatal visits and be offered consent for participation prior to delivery of the infant (Culbert & Davis, 2005).
However, if situations arose during delivery where the infant required resuscitation, parents were somewhat comfortable with receiving information during resuscitation and deciding about participation in an experimental interventions (Culbert & Davis, 2005). Others also found that approaching parents during labor resulted in parents consenting to participation because they trusted the HCPs (Snowdon et al., 2006). Some mothers in labor reported they were too overwhelmed to understand the information presented and did not feel they took enough time to make a decision (Snowdon et al., 2006).

As the seriousness of the child’s illness progressed parents needed to determine whether to participate in an experimental interventions (Snowdon et al., 2006). Parents who decided to participate in an experimental interventions wanted information on how the experimental interventions could impact the child’s level of pain and suffering (Culbert & Davis, 2005; Snowdon et al., 2006). Information about whether or not the experimental interventions required additional invasive procedures or monitoring impacted parents decision regarding participation (Woodgate & Yanofsky, 2010). Knowledge about how the experimental interventions could impact the child’s quality of life during the study period also influenced parental decision-making (Woodgate & Yanofsky, 2010).
2.3.2.2 No Other Options

When HCPs approached parents about experimental interventions, many parents felt this was the only option left (Snowdon et al., 2006; Woodgate & Yanofsky, 2010). Parents reported that the experimental interventions might be the only way to save their child’s life or improve the condition (Snowdon et al., 2006; Woodgate & Yanofsky, 2010). The experimental interventions offered some hope of a solution to help the child (Woodgate & Yanofsky, 2010).

2.3.3 Escalation of Treatment

As the severity of the child’s illness increases or becomes more complex, parents are often asked to consider further treatment options to escalate care or increase the amount of sustaining measures. Two studies (Bluebond-Langner et al., 2007; Hinds et al., 2000) explored decisions about escalating treatment. The two main factors that influenced decision-making were information needs and the illness severity.

2.3.3.1 Information Needs

When choosing to escalate treatment, parents considered treatment options available and information about scientific evidence supporting these options. Parents of children with recurrent cancer, in which standard treatments failed, wanted information about other available treatments (e.g., cancer-directed therapies, symptom-focused treatments) (Bluebond-Langner et al., 2007). Parents requested second opinions, searched the Internet, and asked HCPs about the treatment options (Bluebond-Langner
et al., 2007). The scientific evidence supporting the treatments was judged on what seemed “logical, sound, and promising” to parents (Hinds et al., 2000, p. 1238).

2.3.3.2 Seriousness of Illness

As their child’s illness severity increased, parents chose to escalate care in order to have more time with their child (Bluebond-Langner et al., 2007). Parents felt that they should continue to pursue curative efforts as long as there was a chance the child could survive and all treatment options were explored (Hinds et al., 2000). Parents often searched for curative treatments even when the infant’s death was imminent (Bluebond-Langner et al., 2007). In fact, Bluebond-Langner et al. (2007) found that parents of children with cancer never initiated the discussion about the discontinuation of curative therapies. Not trying additional curative treatments was unacceptable to some parents based on their personal convictions (Hinds et al., 2000). It is unclear how parents of children with CLTCs other than cancer make decisions about escalating treatment based on the seriousness of the child’s illness.

2.3.4 Maintenance of Treatments

The maintenance of treatments included decisions that are necessary to help the child maintain their chronic medical conditions resulting from the life-threatening illnesses. Three studies (Brotherton, Abbott, Hurley, & Aggett, 2007; Guerriere, McKeever, Llewellyn-Thomas, & Berall, 2003; McNamara et al., 2009) examined maintenance of treatment.
2.3.4.1 Information Needs

Parents of children who required technological advancements to sustain life needed information on the type of technology, the side effects of the treatments, the impact the treatment would have on the child, and the benefits of the treatments (Guerriere et al., 2003; McNamara et al., 2009) to make decisions. Parents used the content of the information to make decisions and did not focus on how the information was presented (Guerriere et al., 2003). At least 60% of parents reported receiving enough information to make decisions about maintenance of treatment (Brotherton et al., 2007; Guerriere et al., 2003).

2.3.4.2 Seriousness of Illness

Once the severity of the child’s illness stabilized, parents were faced with decisions about maintaining the high level of care needed for their child’s survival. Parents of children with high illness severity based decisions on the child’s quality of life. They described quality of life as related to suffering of the child (Guerriere et al., 2003) or how comfortable the treatment was for the child (McNamara et al., 2009). When parents considered additional surgical interventions for their child who had already undergone surgery for other reasons, parents were concerned that another surgical treatment would cause the infant to suffer more (Guerriere et al., 2003). Parents wanted to ensure the treatment option chosen was most comfortable for the child (McNamara et al., 2009). Parents also reported that the placement of a gastrostomy tube would deprive
the child of the pleasure of eating (Guerriere et al., 2003). Other parents felt that their child had an acceptable quality of life despite the dependence on technology (Brotherton et al., 2007). The discomfort the infant experienced influenced which treatments parents chose (McNamara et al., 2009).

### 2.3.5 Withdrawing/Withholding Treatment

#### 2.3.5.1 Information Needs

Parents needed clear, accurate information about the child’s condition and the treatment options available to participate in decisions about withdrawing/withholding treatment (Hinds et al., 2009; McHaffie, Lyon, & Hume, 2001). Parents welcomed having all information presented to them about the disease, prognosis, and treatment options (Einarsdottir, 2009; McHaffie, Lyon, & Hume, 2001), even if the HCPs held differing opinions (Einarsdottir, 2009). In addition, parents wanted information specific and unique to their child (Sharman et al., 2005). Once parents decided to proceed with withdrawal or withholding technological support for their child, they still wanted information about the type of care that would be provided to their child while waiting for death (Sharman et al., 2005). Dussel et al. (2009) found that parents who were given information about treatment during the period before their child’s death were more likely to plan the location of death for their child than parents who were not given this information. Parents continued to need information from the initial discussions about end-of-life care until after the child died. Parents sought information from other HCPs,
the Internet, other parents, and disease-specific organizations about the child’s illness and treatment options (Sharman et al., 2005).

Parents also continued to want the information conveyed in an empathic and caring manner when faced with decisions about withholding/withdrawing treatments (Brinchmann, Rorde, & Nortvedt, 2002) and were interested in knowing the HCP’s opinion about what was the best treatment option for their child (Sharman et al., 2005). In addition, they appreciated when the information provided in the formal and informal meetings with the HCPs was communicated to all members of the health care team because if the information was not passed along, parents were left having to communicate the information multiple times to other HCPs (Hinds et al., 2009). Parents also wanted the HCPs’ approval when they sought additional information about treatment options from other sources (e.g., other HCPs, Internet) (Hinds et al., 2009).

2.3.5.2 Seriousness of Illness

Parents of children with a high likelihood of imminent death continued to want their child to survive the disease or condition (Brinchmann et al., 2002), and wanted to know whether or not their child had any chance of survival (Meyer, Burns, Griffith, & Truog, 2002). In order to make a decision about withdrawing or withholding treatment, parents needed to believe that their child could not survive (Hinds et al., 2000). Knowing that their child had a poor prognosis and was progressively getting worse helped parents make decisions about end-of-life care (Dussel et al., 2009; McHaffie, Lyon, &
Hume, 2001). Also seeing physical signs (e.g., infant looks sick) of clinical deterioration helped parents understand the seriousness of the illness (McHaffie, Lyon, & Hume, 2001).

Parents were also concerned with how the illness affected the child’s quality of life. Meyer et al. (2002) found that quality of life was very important to 82% of parents who chose to withdrawal support from their child. Quality of life was described in terms of suffering (Carnevale et al., 2011; Hinds et al., 2009; Michelson et al., 2009), neurologic functioning, and technological dependence (Michelson et al., 2009). Suffering was described as pain (Carnevale et al., 2011; Meyer et al., 2002; Michelson et al., 2009) and fear (Hinds et al., 2000). To evaluate pain and suffering parents used their own assessment of pain and suffering of the child rather than of the HCPs (Sharman et al., 2005), and parents advocated against treatments that caused the child to suffer (Hinds et al., 2000; Hinds et al., 2009).

In addition to concerns about pain and suffering, quality of life concerns also extended to neurologic functioning of the infant. Parental assessment of the neurological state was dependent upon whether the infant was aware of his/her surrounding and if he/she was able to communicate and interact with the world (Einarsdottir, 2009; Michelson et al., 2009). If the infant was neurologically devastated, parents would consider end-of-life care (Michelson et al., 2009). However, some parents questioned whether it was their decision to determine if certain characteristics made life not worth
living (Einarsdottir, 2009). Technological dependence was also viewed as possibly causing the child to suffer (Michelson et al., 2009).

For other parents quality of life of the child was not an influence. Despite receiving information about survival, suffering, and future prognosis (Einarsdottir, 2009), some parents did not consider withdrawal or withholding support a viable option and wanted all measures taken to sustain their child’s life (Carnevale et al., 2011; Michelson et al., 2009). Parents felt that the survival of their child demonstrated that people could live despite severe disabilities (Sharman et al., 2005).

2.3.5.3 No Other Treatment Options

Parents often proceeded to end-of-life care decisions when they lost hope in the infant surviving (Michelson et al., 2009) or because of a lack of viable treatments with the potential to cure the illness or disease. Parents needed to know that all treatments were exhausted and they did everything they could for their infant (Hinds et al., 2000). This meant that there were no effective treatments left. When all options were exhausted, parents acquiesced to withdrawal or withholding life-sustaining treatments (Michelson et al., 2009).

2.3.5.4 Religiosity and Spirituality

Parents relied on religiosity and spirituality as a source of guidance in decisions about withholding or withdrawing treatments (Meyer et al., 2002; Michelson et al., 2009). While some parents relied upon God or faith to guide their decision-making
(Sharman et al., 2005); others felt that the decision was up to God and not one to be made by humans (Michelson et al., 2009; Roy, Aladangady, Costeloe, & Larcher, 2004; Sharman et al., 2005). The degree of religiosity a parent reported influenced their decision-making. Very religious parents were less likely to plan the location of their child’s death than parents who were somewhat or not religious at all (Dussel et al., 2009), possibly because very religious parents continued to pray for miracles and awaited divine intervention (Michelson et al., 2009; Sharman et al., 2005).

Parents also participated in religious and spiritual rituals for guidance in the decision-making process. Einarsdottir (2009) reported that some parents in Iceland relied upon old dreams and interactions with mediums for information about how to proceed with end-of-life decisions and to ask the medium to help/support the HCPs caring for their infant. Some parents chose an emergent baptism when their infant or child was close to death, while others felt that having a baptism in the intensive care unit was an act of surrendering to death (Einarsdottir, 2009). If the child’s condition improved, parents interpreted this act as a miracle (Einarsdottir, 2009). Others prayed for miracles or divine intervention (Sharman et al., 2005), but would consider withholding or withdrawing support if ‘enough’ time had passed and no miraculous recovery occurred (Michelson et al., 2009).
2.3.5.5 Parental Characteristics

Parental race and ethnicity may impact the types of recommendations parents received and whether parents chose to accept the recommendations regarding end-of-life care (Moseley et al., 2004; Roy et al., 2004). In a small study of the impact of race on parental acceptance of HCP recommendations, a non-statistically significant difference was found between African Americans who accepted the recommendation to withhold treatment 62% of the time compared to white parents who accepted the recommendation 80% (Moseley et al., 2004). In another study, Black African and Jewish parents were less likely to agree to withdrawal support for their critically ill children than White, Indian, and Afro-Caribbean parents (Roy et al., 2004). Specifically why ethnicity affected the acceptance of recommendations to withdrawal support was not further explained because data were collected with surveys.

Some parents were concerned about their ability to care for their infant, if he or she survived the hospitalization. Parents were concerned about how they would actually provide care for their infant both logistically and financially. Parents were worried about how to financially support the infant and also how to logistically provide the intensive care they would need at home (Sharman et al., 2005). Other parents felt that finances were not at all part of their decision to withdraw support from their infant or child (Meyer et al., 2002).
Parents’ previous experiences with death of a family member also affected their
decision-making for their child. Parents used previous experiences with deaths of family
members to justify and understand how their infant was feeling while being supported
by technology (Sharman et al., 2005). Parents also explained that they compared the
physical appearance of their family member who died with their infant to determine if
they thought the infant was also going to die (Sharman et al., 2005). Parents who
experienced a previous loss were more likely to plan the location of death for their infant
than parents who did not have a previous experience with loss (Dussel et al., 2009). This
previous experience with a death of a family member may provide parents with an
understanding of what occurs around the time of death and what decisions are needed
during this time.

2.3.6 Additional Influences

2.3.6.1 Child’s Best Interests

Having the child’s best interests in mind was critical to decision-making of
parents. This theme was described as an influence during initiation of life-sustaining
treatment, experimental interventions, and withholding/withdrawing treatment
decisions. Since parents described the influence similarly across treatment decisions, the
results are presented together. Parents did not describe this theme for escalation or
maintenance of treatment decisions, but limited research exists.
Parents relied on HCPs to have their child’s best interest in mind and thus valued the opinion of the HCP (Culbert & Davis, 2005; Woodgate & Yanofsky, 2010). Parents felt maintaining and preserving the dignity of their child was in the child’s best interests (Hinds et al., 2000; Hinds et al., 2009), which was determined by parental knowledge of their child and the illness (Boss et al., 2008; Hinds et al., 2000; Michelson et al., 2009), their experience with their child (Lan et al., 2007), and their child’s preference (Hinds et al., 2000). Considering what they would want if they were in the same situation (Sharman et al., 2005). Ensuring parents were included as experts in knowing their child was important when including parents in the decision-making process.

2.3.6.2 Support

Support was critically important to all decision types for parents. Parents described sources of support similarly across the different types of treatment decisions. Support for parents was provided by other family members (Boss et al., 2008; Lan et al., 2007) and families with similar experiences (Lan et al., 2007), and HCPs (Brotherton et al., 2007; Guerriere et al., 2003; Kavanaugh et al., 2010; Woodgate & Yanofsky, 2010). Emotional support from HCPs was demonstrated by HCPs listening, being kind and comforting, maintaining hope, providing spiritual support (Kavanaugh et al., 2010), and acknowledging the difficulty and uncertainty associated with making decisions (Guerriere et al., 2003; Redlinger-Grosse et al., 2002). Parents felt the support of HCPs
when the provider spent time with them and their infant even once the decision was made (Payot et al., 2007; Woodgate & Yanofsky, 2010).

2.4 Discussion

Researchers have described how parents, both mothers and fathers, make decisions for children with complex life-threatening illnesses. Parental decisions span the trajectory of the child’s illness to include initiation of life-sustaining treatment, initiation of experimental interventions, escalation of treatment, maintenance of treatments, and finally withdrawing/withholding treatments. This range of decisions was described in research conducted with children with cancer, extreme prematurity, congenital heart disease, neurological injuries and diseases, and chromosomal and genetic abnormalities.

Parental decision-making for children with life-threatening illnesses is impacted by a range of factors including the type and content of information provided to them as well as the information they sought, the seriousness of the child’s illness, whether other treatment options exist, what is best for the child, religiosity and spirituality, parental factors, and support. Parents need information to make decisions across the illness and treatment trajectory. Yet they did not always understand or receive enough information to make decisions (Brotherton et al., 2007; Guerriere et al., 2003; Menahem & Grimwade, 2003). Different approaches to explaining complex information could be explored through hypothetical scenarios with parents or comparing what is different between
parents who report understanding and those who do not understand. Additionally, the
types and amounts of information parents need when making decisions about
experimental interventions and escalating and maintaining treatments needs further
description.

The development and maintenance of parental trust in the health care team is a
critical area that few studies have focused on. While communication of hopefulness by
HCPs increased parental trust (Boss et al., 2008), it is unclear how much of the
information must be hopeful. Additionally, researchers do not know how a trusting
relationship between parents and HCPs develops over time. The communication of
hopefulness has been studied as it relates to establishing a trusting relationship at the
initiation of life-sustaining treatments, but has not studied in escalation of treatment
decisions nor withdraw and withholding of treatments. Further understanding of how a
trusting relationship develops and its impact across the child’s illness trajectory is
needed to understand how to improve parent and HCPs relationships.

Parents described not having any other treatment options as impacting their
decision-making in initiation of life-sustaining treatments (Hinds et al., 2000; Vandvik &
Forde, 2000), experimental interventions (Snowdon et al., 2006; Woodgate & Yanofsky,
2010), and withdrawing or withholding treatment decisions (Hinds et al., 2000;
Michelson et al., 2009). However, this theme was not described in decisions to escalate or
maintain treatment. Escalating treatment may be viewed as an option that is better than
the current treatment and thus this theme was not described, further studies are needed
to determine how parents view escalation of treatment decisions. Maintenance of
treatment decisions often have multiple choices, so parents may feel they have more
options to offer their child and are not forced into a treatment because it is the only
treatment.

Research is needed to identify and clarify the concept of “the best for the child”
across the full illness trajectory. Helping parents explore what is best for their child
when making decisions about initiation of life-sustaining treatment and experimental
interventions and reevaluating how their ideas about what is in the best interest of the
child changes throughout the child’s illness may aid parents in making decisions they
perceive as ‘good’, thus decreasing conflict and regret.

Religiosity, religious preferences, personal belief systems, and spirituality
influenced parental decision-making about initiating life-sustaining treatments (Ahmed
et al., 2006; Chaplin et al., 2005) and end-of-life care (Meyer et al., 2002; Michelson et al.,
2009), but how these impacted other types of decisions remains unknown. Further
exploration about how religiosity and spirituality directly affects parental decision-
making is needed. By understanding the parents’ specific needs, interventions can be
developed to provide parents with the support and guidance.

Parental characteristics impacted initiation of life-sustaining treatment (Lam et
al., 2009) and withdrawing or withholding treatment decisions (Moseley et al., 2004; Roy
et al., 2004), but were not described as impacted experimental, escalation, or maintenance treatment decisions. Therefore, further research to identify if parental characteristics influence these types of decisions is needed. Additionally, how and why certain demographic factors influence parental decision-making is needed. By understanding how and why demographic factors influence decision-making, researchers can develop interventions targeted at different groups to better support and meet their needs.

With the technical ability to sustain children’s lives by providing long-term respiratory support through mechanical ventilation and nutrition through placement of gastrotomy tubes and enteral feeding, a consideration of how parents cope and care for these children is important. Limited information exists about how parents cope with decisions to provide or not provide advanced technology support for children. The psychological long-term outcomes of parents caring for technology dependent children and teenagers remain limited because many of these technologies did not exist in the past.

The strengths of the empirical research reviewed are that initiation of life-sustaining treatments and end-of-life care decisions have received significant attention. Researchers have explored how many different factors impact decision-making and have used multiple different research designs and data collection methods to explore the decision-making process. These initial studies lay the foundation for future research and
have provided insight into parental decision-making during times of crisis. However, there are still many questions that need to be addressed.

The first gap is that many of the research studies look at parents and HCPs separately without taking into account the individual child and how the parents and HCP interact to make decisions for an individual child. The lack of knowledge about how decisions are made for an individual child is due partly to the research designs employed in most of the studies. Only four studies (Brotherton et al., 2007; Grobman et al., 2010; Kavanaugh et al., 2010; Payot et al., 2007) included parents and HCPs caring for the same children in the sample. All of the other studies included only parents or included both parent and HCPs but not those caring for the same child. By not examining how parents and HCPs make decisions for a single infant, researchers are unable to understand how differences in perceptions about communication of information and severity of illness impact the decision-making process. Studies must begin to include both parents and HCPs so that researchers can evaluate how decisions are made for individual children with life-threatening illnesses to understand the dynamics between parents and parent-provider relationships.

The second gap in research to date is related to the diagnostic categories of children included in the research samples. The majority of studies focused on one homogenous diagnostic group of premature infants, children with complex congenital heart disease, or children with cancer. Thus comparisons across other child illness
categories cannot be made. Also, comparison across studies is difficult. Therefore, researchers do not know if the parents of different groups of children need the same type of information to make decisions or need different information. Parents of children with neurologic conditions may need information explained differently or may use other factors to make decisions for these children, but this remains unknown.

Most studies also used cross-sectional and/or retrospective research designs. Thus the third gap in the empirical knowledge is that researchers have limited understanding of how factors change over time for parents. The weakness of using a cross-sectional or retrospective designs in decision-making is there is limited ability to understand how and when parent influences change over the course of the child’s illness. Only four studies (Feudtner et al., 2010; Grobman et al., 2010; Kavanaugh et al., 2010; Payot et al., 2007) examined decision-making using a prospective longitudinal design. Therefore researchers have limited understanding about how trust and mistrust develop across the child’s illness course. Additionally, how individual parents use different influences as the child’s illness progresses also remains unknown. Thus when feasible researchers should consider using a prospective, longitudinal design to better understand when and how influences change across the decision-making trajectory for parents.

While the current empirical evidence about influences on parental decision-making for children with life-threatening illnesses has limitations, the knowledge gained
through the studies is valuable. Researchers can now move forward with an understanding of what influences parent decision-making broadly and begin to explore whether or not the previously identified influences exists within all decision types. Additionally, researchers know that some of the influences most likely change over time and identification of when these influences change and why they change can be examined. Finally, researchers must also focus on understanding decisions about escalation and maintenance of treatment because these decisions can lead to parent-HCP conflict and impact the parents’ psychological and physical status and the child’s quality and duration of life.

This integrated literature review included only articles published in English. Additional empirical evidence about parent and health care provider decision-making for infants with life-threatening illnesses could exists in other languages. In addition, only articles from the last 12 years were reviewed. The authors felt this was appropriate because of the recent push to include parents in the decision-making process and the scientific advances in neonatal and pediatric critical care.

### 2.5 Conclusions

The purpose of understanding the influences on parent decision-making for children with complex life-threatening illnesses is to improve the short-term functioning and long-term outcomes of parents and families who are thrust into these very difficult situations. Examples of improvements in short-term functioning include decreasing
parental-health care team conflict and improving parent quality of life during the trajectory of the child’s illness. Examples of positive long-term outcomes include decreasing parental regret and improving family functioning. Multiple influences impact how parents make decisions with no single influence identified as the sole reason for the decision. A combination of these influences leads parents to an eventual decision about treatments. Through identification and evaluation of the different factors that impact parental decision-making, researchers can develop interventions to support parents forced to make difficult, challenging, life-changing decisions for children with life-threatening conditions.
3. An Ethical Analysis of Longitudinal In-Depth Interviews with Parents’ of Infants with Life-Threatening Illness

Nurses play a unique role in caring for people experiencing serious illness. Through their constant presence, nurses bear witness to suffering while attempting to minimize its’ impact by providing physical care as well as listening to the feelings and concerns of patients and families. Often the content of these discussions include intimate and sometimes sensitive topics (Ferrell & Coyle, 2008; Kavanaugh et al., 2010). A sensitive topic is a deeply personal experience that, when exposed, can be emotionally distressing (Lee & Renzetti, 1990).

As a means of investigating the complex needs of patients and families, nurse researchers have increasingly focused their research on sensitive topics that can involve the discussion of dying such as palliative care (Shipman et al., 2008), end-of-life care (Emanuel, Fairclough, Wolfe, & Emanuel, 2004; Shipman et al., 2008), bereavement (Emanuel et al., 2004; McCreight, 2008; Williams, Woodby, Bailey, & Burgio, 2008), and life-threatening illnesses (Lowes & Gill, 2006; Sherman et al., 2005). Other researchers are interested in understanding the needs of individuals who face social stigma from interpersonal violence (Campbell & Adams, 2009; Hlavka, Kruttschnitt, & Carbone-Lopez, 2007), HIV and AIDS (Sandelowski, Barroso, & Voils, 2009) and mental illness (Kidd & Finlayson, 2006). In order to fully capture the depth and meaning of sensitive health and illness experiences, investigators often use in-depth interviews as a central
source of data collection. Yet, questions arise as to whether these interviews are an added burden or even increase distress for participants in difficult situations (Decker, Naugle, Carter-Visscher, Bell, & Seifert, 2011; Newman, Risch, & Kassam-Adams, 2006). More specifically, investigators and clinicians have raised concerns about the balance between the societal benefit of the research being conducted and the burden that interview methods place on patients and families who are already under distress from an acute or chronic health crisis at the time of the interview (A. M. Beck & Konnert, 2007; McGrath, 2003).

Guidance is available on methodological challenges such as how to recruit participants for studies on sensitive topics (Campbell & Adams, 2009; Chiang, Keatinge, & Williams, 2001), how to design questions to elicit information about sensitive topics (Williams et al., 2008), and how to train the research team to conduct the interviews (Jansen, Watts, Ellsberg, Heise, & Garcia-Moreno, 2004). Yet, there is limited empirical evidence regarding the impact of intensive, longitudinal interviews on participants throughout the study period. Insufficient empirical data about these potential burdens can make assessment of the balance between benefit and burden difficult and lead institutional research ethics review boards (IRBs), funding agencies, and clinicians to be cautious about supporting research that uses in-depth interview methods on sensitive topics with vulnerable populations (Chiang et al., 2001; Corbin & Morse, 2003; Emanuel et al., 2004; Lee & Renzetti, 1990; McIlfatrick, Sullivan, & McKenna, 2006).
Two key ethical principles may be helpful in guiding researchers who are conducting research on sensitive topics. These principles—respect for persons and beneficence—were first elaborated upon in the Belmont report which focused on protecting participants in research from both physical harm and emotional distress (DHEW, 1979). If in-depth interviews increase distress, they may violate the ethical principle of beneficence. Yet, if in an effort to protect potentially vulnerable participants from the burden of interviews about sensitive topics they are not given the opportunity to choose whether to participate in the study, the principle of respect for person may be violated. Participation of different genders, financial backgrounds, ethnicities, races, and cultural backgrounds are necessary to advance the understanding of phenomena. This is particularly important because study participation may have benefits such as feeling good about making a contribution to the research problem or lessen burden as a result of telling one’s story (Campbell & Adams, 2009). Striking a balance in the design of research protocols is critical such that it afford individuals the opportunity to participate in studies without causing them harm.

Research on the experiences of parents of critically ill infants is likely to be particularly sensitive as parents are often distressed, anxious, and shocked after learning their infant has a critical illness (Clark & Miles, 1999; Kavanaugh, Moro, Savage, Reyes, & Wydra, 2009). Parents are then faced with decisions along the infant’s trajectory of illness and treatment such as about how to proceed with medical care, the special
individualized care needs, and the possibility of their infant dying (McCreight, 2008). In-depth interviews are a useful method to explore the experience of these parents. These interviews have the potential to increase distress, particularly if the research involves longitudinal designs (Lazovski et al., 2009; Lyons et al., 2004; Siden et al., 2010) because of repeated encounters with emotive discussions and potential burden of participation in the study. Conversely, there is some evidence that parents may benefit from such studies (Emanuel et al., 2004; Lowes & Gill, 2006).

While there is some empirical literature on how the experience of participating in research on sensitive topics impacts parents, few studies have included the parent perspective. One of the main reasons for examining the parental experiences of participation in research is to understand how parents respond to the strategies employed by research teams to protect their ethical rights of respect for persons and beneficence. Therefore, the primary aim of this analysis was to understand the parental responses to strategies implemented in the study to address the principles of respect for persons and beneficence and describe strategies initiated in response to parental experiences throughout the study period.

3.1 Background

3.1.1 Respect for Persons

The Belmont report (DHEW, 1979) describes the principle of respect for persons as fundamental to ethical human subject research. Respect for persons includes the
opportunity of study participation to an individual who meets eligibility criteria, as well as respecting each participant as being capable of making an informed decision regarding research study participation (Beauchamp & Childress, 2008). Providing access to individuals who meet study criteria can be difficult because participants can be recruited using different methods. For example, individuals for bereavement studies could be recruited using support groups or churches, obituaries, public health records, and hospital databases (Stroebe, Stroebe, & Schut, 2003). However, each of these potential recruitment sources may exclude groups of bereaved individuals and deny them access to a study. Health care clinicians and investigators may impact on this aspect of respect of persons by denying study access to individuals they assess as vulnerable. This can occur when a clinician denies access to the individuals in an effort to protect the individual from potential pain or distress that could be brought on by study participation. Allowing all eligible individuals in the study population the opportunity to participate upholds the ethical principle of respect for persons.

When given the opportunity to approach individuals, respect for persons requires full disclosure about a study through an informed consent process. Investigators must ensure that participants are fully informed and understand the purpose, procedures to be followed, along with benefits and risks of participation before being asked to make a decision about whether or not to participate (Allmark & Spedding, 2007; Thomas, 2005). While individuals are given information about the
purpose of a study, re-informing them throughout the process is important since every question cannot be explicated during the initial informed consent process and participants may not be fully aware of the exact procedures involved (Allmark et al., 2009).

On the other hand, respect for persons requires allowing individuals to make choices about participation without coercion. Many participants are recruited from hospital settings and may feel obligated to participate so their future care is not jeopardized (Meert et al., 2008). Individuals in bereavement research may also feel pressure to participate in the study if the research is being conducted by the clinicians or the hospital that cared for their family member prior to death (Williams et al., 2008). Individuals who agree to participate should be reminded that they can withdrawal from the study or choose to skip a data collection point in longitudinal studies (McGrath, 2003). Participants have reported that knowing they did not have to participate, that they could withdrawal from the study, and that they did not have to participate in parts of the data collection process helped them feel that they had control over their relationship with the investigator (McGrath, 2003).

In addition, respect for persons involves ensuring participants have control over their participation in the research process. If possible, participants should be given control over the time of day and date they participate in a study and the location of the data collection (Williams et al., 2008).
and date demonstrates respect of the participant’s time and daily schedule. Careful consideration of the location of the data collection is important because some areas of the hospital or the hospital in general may bring back memories or strong emotional responses that could lead to emotional distress (Williams et al., 2008). Ensuring the study is designed and implemented with respect for persons including providing eligible individuals access to the study, fully disclosing the research study to the individuals, minimizing coercion, and allowing the participant control over the data collection process is essential to respecting the ethical rights of research participants.

3.1.2 Beneficence

The second research ethics principal that is instructive in guiding investigators using in-depth interviewing is beneficence, which involves minimizing physical harm, emotional distress, and participant burden (McIlfatrick et al., 2006). It can also encompass acknowledging the contribution of participants to the study aims. Issues of physical harm in research are often conceptualized as harm from study components such as medications or surgical interventions, but physical harm can occur in other ways (Lazovski et al., 2009). Physical harm may also occur when the research study places participants in vulnerable life situations (Logan, Walker, Shannon, & Cole, 2008). For example, participation in research on intimate partner violence may pose a safety issue for the participant (the person who experienced violence) because it could intensify negative feelings of the partner leading to further violence (Logan et al., 2008). Therefore
possible physical harms in research about sensitive topics are dependent upon the topic and participants and thus require careful assessment by the investigator.

Emotional distress can occur because participants are asked to relive or explain an experience that was distressing (Hlavka et al., 2007; Newman & Kaloupek, 2004; Templeton, 1993). Methods involving in-depth interviews may be perceived as intrusive (McGrath, 2003) because participants may disclose more information than intended (Corbin & Morse, 2003) and in-depth interviews may include intimate questions. However, in a qualitative study to explore the experience of the treatment trajectory by parents of children with cancer, parents indicated that the interviews were not intrusive (McGrath, 2003).

There has been a substantial amount of work on the impact of sensitive topics research on participants who are adult with chronic or terminal illnesses and their families (Emanuel et al., 2004; Lowes & Gill, 2006; Pessin et al., 2008; Pierce, Steiner, Pitzen, & Grothaus, 2009; Shipman et al., 2008; Takesaka, Crowley, & Casarett, 2004) with one or two data collection points (K. M. Dyregrov, 2004; Emanuel et al., 2004; Griffin, Resick, Waldrop, & Mechanic, 2003; Michelson, Koogler, Skipton, Sullivan, & Frader, 2006; Pessin et al., 2008; Pierce et al., 2009; Shipman et al., 2008; Takesaka et al., 2004). The results from adults with chronic or terminal illnesses and their families who participate in research with limited data collection points suggest that most participants in sensitive topics research do not experience distress (Emanuel et al., 2004; Griffin et al.,
However, after consenting, some participants have reported feeling nervous, anxious, or uncertain about how the interview would transpire (K. M. Dyregrov, 2004; Kari Madeleine Dyregrov et al., 2011; Lowes & Gill, 2006). During emotive interviews, some participants express intense emotions, but none of the participants regretted taking part in the study (Lowes & Gill, 2006).

Evidence also suggests that individuals who participate in research on sensitive topics may find the interviews helpful (Emanuel et al., 2004; Lowes & Gill, 2006; McGrath, 2003; Shipman et al., 2008) because it meant someone cared about the person and topic (Shipman et al., 2008). Interviews also allowed participants time to process information and cope with illness crises (K. M. Dyregrov, 2004; McGrath, 2003; Shipman et al., 2008). In contrast, other researchers found that participation in interviews about end-of-life care or decision-making ranged from ‘a little’ painful to distressing to participants (K. M. Dyregrov, 2004; Takesaka et al., 2004), but when asked participants did not regret participating in the interviews (K. M. Dyregrov, 2004).

Minimizing burden is another component of beneficence. Data collection methods, such as interviews or multiple surveys, can be burdensome to participants (Emanuel et al., 2004; Lazovski et al., 2009), especially when required at multiple time points. Interviews generally take 30 minutes to 90 minutes (Lowes & Gill, 2006; Michelson et al., 2006), depending on the purpose of the interview and the participants’ responses. Data suggests that the interview may feel burdensome to some participants.
and others report ambivalence towards the interview (Kari Madeleine Dyregrov et al., 2011; Pessin et al., 2008; Pierce et al., 2009). In some cases, participation in interviews were completed in conjunction of caring for a family member at home (Pierce et al., 2009) or interfered with other activities the participants wanted to do (Pessin et al., 2008), which could be perceived as an additional burden. However, the majority of participants reported they would participate in a similar interview if offered again (Michelson et al., 2006; Pessin et al., 2008) indicating the interviews were not burdensome to most participants.

In summary, maintaining respect of persons and beneficence is the upmost responsibility of researchers to protect the participants and protect the integrity of the findings. Therefore ethical principles must be built into the design and procedures for a study, particularly recruitment processes, data collection methods, and supervision and training of the research team. Using the two guiding principles, of respect of persons and beneficence, parents’ responses to participation in a longitudinal study involving intensive interviews was explored. Parental responses to strategies the research team used to address these important ethical principles were identified.

### 3.2 Methods

Data for this analysis was collected for an ongoing case-based, longitudinal, mixed method study designed to understand the trajectory of decision-making about life-sustaining treatment for infants with complex life-threatening conditions (Decision-
making for Infants with Life-Threatening Conditions, NR010548, PIs Docherty and Brandon). Each case involved a critically ill infant, at least one parent, and at least three health care providers (e.g., nurse, physician, nurse practitioner, surgeon, or social worker). Parents and health care clinicians participated in narrative interviews and self-report questionnaires for up to one year starting at enrollment, which generally occurred during hospitalization. Data for this analysis were collected between November 2007 and November 2011.

3.2.1 Participants

The setting for this study was a southeastern tertiary academic medical center, which includes a children’s hospital. The hospital offers high-risk specialty care for infants and children ranging from extreme prematurity to surgical care along with solid-organ and bone marrow transplants.

A purposive sampling technique was used to recruit parents and providers of infants with complex life-threatening conditions including complex congenital heart disease, extreme prematurity, or metabolic diseases requiring hematopoietic stem cell transplant. Parents of infants within each diagnostic category were sampled to include case variations on race and socioeconomic statuses (SES) and those with a prenatal diagnosis. Parents were excluded if they were less than 18 years of age, non-English speaking, or if they were not the biological parents. Parents were interviewed
individually and were able to participate if the other parent did not consent to participation.

Forty-nine parents of 29 infants with complex congenital heart disease (n = 9), extreme prematurity (n = 11), and metabolic disease requiring stem cell transplant (n = 9) were part of this analysis. Four parents of three infants (twins, singleton) withdrew from the study prior to completing any data collection forms or interviews. Twenty-five mothers and 20 fathers participated with the majority of parents being married (71%), Caucasian (51%) with a yearly income of $51,000 to $75,000. The average age of the parents was 30 years (range 21-46) with an average education level of 14 years (range 7-20). A total of 364 interviews from parents were included in this analysis.

3.2.2 Data Collection Methods

The data collection techniques and time points were selected, in an attempt, to capture episodes of decision making along the infant’s illness and treatment trajectory (e.g., birth, following surgical procedure). Narrative-style interviews and self-report questionnaires were utilized to collect data regarding the parent’s experience with the illness and decision making trajectory. The main focus of the interview questions centered on (a) how the infant and the parent had been doing since the last contact, (b) types of decisions made, (c) what or who influenced the decisions, and d) relationships with their family and the health care team. To better understand the parental experience of participating in this longitudinal study the following questions were utilized: (a) “Can
you tell us about your experience of being in the study?”; (b) “Did you feel that the
interviews were difficult (e.g., scheduling, time commitment, emotion, tough
questions)?”, and (c) “Were the interviews helpful in anyway?” These questions were
asked every 2-3 months and at the final interview after participating in the study for the
year.

Parents were interviewed within 1 week of birth/diagnosis or stem cell
transplant, following each life-threatening event or significant change in treatment, at
discharge from the hospital, and monthly for 1 year. Parents of infants who died within
the study period were also interviewed about the bereavement process and their
retrospective assessment of the decision-making trajectory for their infant at 6-weeks
and 6-months following the infant’s death. A life-threatening event was defined as an
acute, unexpected event in which the health status of the infant suddenly changed,
requiring a change in treatment, such as emergency procedures, life-sustaining
treatments, or cardiopulmonary resuscitation. A significant change in treatment was
defined as a decision to intensify curative treatment, to shift from curative to symptom-
focused palliative care, or to withhold or withdraw treatment. Following the completion
of narrative interviews with the parents, interviewers recorded field notes detailing the
setting, timing, and the parent’s physical and emotional state during the interview,
within one hour of contact with the parent.
An example of the data collection time points for parents of an infant with complex congenital heart disease portrays the intensive data collection used in this ongoing study. For an infant who survived 30 days, the parents of an infant with hypoplastic left heart syndrome (HLHS) participated in an interview within 1 week of birth, an interview within 1 week following the Stage I cardiac palliation (around 1-2 weeks of life) because the surgical procedure was a life-threatening event (LTE), an interview following an exploratory laparotomy due to suspected necrotizing enterocolitis, and 2 subsequent interviews at 6-weeks and 6-months following the infant’s death. Since the overall aim of the larger study is to understand the infant’s illness trajectory and the decisions parents make for the infants, data collection time points were selected to be close in time to infant illness events. However, the intensive design and content of the study could have a high potential for burden on the parents.

3.2.3 Ethical Considerations

The medical center institutional review board approved this study. During the informed consent process, parents were told that information provided during the interviews would not be shared with the other parent or any of the health care clinicians. Parents were compensated $20 for their time after completing each interview. Interviewers were not involved in the direct care of any infant whose parents participated in this study.
3.2.4 Data Analysis

Digitally recorded interviews and field notes were transcribed and then reviewed for accuracy. The names of the participants were changed to pseudonyms.

A content analysis procedure was used to analyze the data to identify themes and related patterns (Graneheim & Lundman, 2004; Sandelowski, 1995). Coding focused on identifying data related to the impact of the interviews on the parents, parental response to interview participation, and parental perception of burden. Interviews were reviewed and coded independently by KA and DB. The analysts compared results of the individual analysis among all analysts (KA, DB, SD) to identify the core themes. The core themes were then organized using the ethical principles of respect for persons and beneficence as a framework to understand how research participants responded to research strategies and how providing ethical protections may impact study outcomes.

3.3 Results

The results of the study are presented by ethical principle (respect for persons and beneficence). Within each ethical principle, core themes related to the principle are presented including which research strategies supported the core themes and parental response to the employed research strategy. See Table 4 and 5 for strategies utilized for respect for persons and beneficence and the responses to each strategy.
3.3.1 Respect for Persons

The ethical principle of respect for persons was comprised of four core themes: access to study, full disclosure, minimization coercion, and control over the data collection process. Access to study participation was protected by ensuring parents were approached and given the opportunity to choose whether or not to participate in the study. The research team was trained by the investigators prior to the start of the study about the procedure for parent recruitment and consent, which was IRB approved. Investigators also presented the study, discussed their role in recruitment, and answered questions from health care clinicians on the units prior to initiation of the study. To identify eligible participants, the research team screened admissions to the units 2-3 times per week. At times, health care clinicians attempted to protect the parents from being approached for study participation who they thought of as vulnerable because of family troubles, stresses related to the infant’s illness, or other issues. These clinicians tried to protect the parents by voicing their concerns to the research team prior to recruitment of the case, discussing the study with parents to ensure they knew their participation was optional, and explaining that parents were having a difficult time during a crisis period when the research team attempted to approach parents throughout the illness trajectory. However, we found that many of these parents, whom the health care clinicians thought would not participate, did in fact consent to participation and continued through the study. One parent noticed a nurse trying to
protect his wife from the study: “A nurse came up and told Janice that she didn’t have to do that [study]”. Even after reminding the parent she did not have to participate, both parents consented and completed the study.

Through the consent process, the purpose and data collection methods were fully disclosed to the parents, including the approximate length of each data collection and the length of the entire study. While only one parent was needed per case, study personnel attempted to contact both parents, even if the first parent approached thought the other parent would not consent. After the research team described the study to some parents, they chose not to consent to participation because they thought the study would be too burdensome and some reported not wanting to participate in any studies. The research team also attempted to maintain respect for persons through re-addressing consent at different points of the interview and throughout the study period. For example, the research team asked parents if it was okay to proceed with discussing the death of their infant during bereavement interviews. By using this technique we were attempting to continue to inform the parent of the next topic, so that if the parent did not want to discuss this topic, he/she could refuse. A member of the research team began a 6-week bereavement interview with: “Peter, I really appreciate you taking the time to talk with me this morning. I just want to preface this interview with anything that I ask that you’re not comfortable answering like in the past when we’ve talked…let me know.”
To minimize coercion to participate in the study and ongoing data collection, the research team with a clinical practice did not provide patient care to infants enrolled in the study. Parents reported, “it’s just been nice to actually talk to someone completely unrelated to the NICU”. Parents did not appear to feel obligated to participate as 3 of the 27 consented cases withdrew. Some parents also discussed the importance of the members of the research team not offering advice or recommendations throughout data collection.

Throughout the study, the research team was mindful of the principle of respect for persons by giving parents control over the option of completing the interviews at a time and place (or over the telephone) convenient for them and allowing parents to opt out of an interview. The mother of an infant with a metabolic disease explained during the final 12-month interview that “if there was a moment that it wasn’t appropriate or something like that we wouldn’t talk”. Another mother of an infant who died of a complex congenital heart condition, who chose not to participate in the 6-week bereavement interview, but did complete the 6-month bereavement interview described not scheduling the 6-week interview because she was not ready to discuss the infant’s death.

You [referring to the interviewer] had a really hard time setting this one up with me because I knew what we were going to talk about and I know what it’s like going back into that time, so that’s hard. But it’s not necessarily you guys or the study itself; it’s the circumstances on having to relate that’s hard.
Other parents did not feel the need to justify the reasons for not wanting to participate at certain points of the study. One father explained, “I didn’t feel like I had to justify to you why we weren’t, we didn’t want to participate that day. I mean you understood, I didn’t have to convey that to you or anything like that.” Even though the research team attempted to schedule interviews based on the parent’s schedule, parents still reported scheduling of the interviews as one of the most difficult parts of the study because they could not predict events within their infant’s illness trajectory, the times their other children would be napping, or when they would be able to come to the hospital.
<table>
<thead>
<tr>
<th>Theme</th>
<th>Study Strategy</th>
<th>Responses to Strategy</th>
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<tbody>
<tr>
<td>Recruitment</td>
<td>• Study personnel trained by principal investigators prior to initiation of study and throughout study period about rationale for approaching eligible participants</td>
<td>• Health care providers attempted to protect potentially vulnerable participants</td>
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<td></td>
<td></td>
<td>• 74% of parents (n = 51) approached chose to participate in the study, despite their infant(s) being critically ill and self-reported high levels of parental stress</td>
</tr>
<tr>
<td>Consent Process</td>
<td>• Full disclosure of study purposes and data collection procedures prior to consent and throughout study period</td>
<td>• 66% (n = 27) of cases approached consented to participate; however, 2 of the 27 cases withdrew following consent and 1 withdrew after completing 1 month of data collection</td>
</tr>
<tr>
<td>Minimization of Coercion</td>
<td>• Study personnel did not provide patient care to infant’s enrolled in the study</td>
<td>• Participants appreciated that the study personnel had health care knowledge and also specific information about their infant, but were not involved in the infant’s care. Participants also acknowledged that study personnel did not offer advice or recommendations because that was what happened in conversations with others (e.g., friends, family).</td>
</tr>
<tr>
<td>Control over data collection process</td>
<td>• Participants chose where and when to complete the data collection</td>
<td>• Participants chose to complete data collections at temporary apartments, their homes, conference rooms, infant’s bedside, and over the telephone</td>
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<tr>
<td></td>
<td>• Participants could chose to not participate in any data collection time point</td>
<td>• Participants completed data collection in the morning, during clinical appointments, and during the evening</td>
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<td>• Most participants missed at least 1 data collection.</td>
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<td>• Some participants provided</td>
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3.3.2 Beneficence

Beneficence was comprised of four core themes: minimization of physical harm, minimization of emotional distress, minimization of burden, acknowledgement of parent and infant contributions. Several protocols were utilized throughout the study period to minimize physical and psychological harm to participants. The first protocol developed was parental use of a cellular phone to complete an interview while driving. Study personnel found that some parents were completing the potentially emotive interviews while driving to and from work or the hospital because they were alone and the environment was quiet. Thus, a protocol was developed that study personnel requested and reminded the parent to not schedule or complete the interview, when they were driving or doing any dangerous activity. The second protocol was developed to guide study personnel in how to manage extraneous situations that were unrelated to the study, but was brought to the attention of a study member by a parent. For example, a member of the study was contacted by one of the study participants who was sick and had no family in the area. The parent requested medical help from study personnel. Thus the protocol developed was broad since several participants were in the area.
without family and could face similar situations and included notifying the on-call principal investigator, notifying the infant’s social worker if appropriate, and contacting emergency responders if necessary.

Two additional protocols were utilized for developed to protect against emotional distress. A third protocol for suspected suicidal ideation was used as a guide for study personnel to follow if potential physical harm was suspected or identified. The study itself was not thought to lead to suicidal ideation; however, the critical time period within the parent’s life could potentially lead to the co-occurrence of thoughts and/or acts self-inflict physical harm. The protocol for suspected suicidal ideation was initiated on one parent, but the parent was not deemed suicidal. The fourth protocol added during the study period was utilized to provide the research team with necessary resources (e.g., domestic abuse hotline or crisis center phone number) for participants who encountered domestic violence or crisis related situations. Finally, investigators were available to the research team at any time for consultation.

In addition to study protocols to minimize psychological distress and physical harm, the research team was trained on interview techniques. The interview techniques described how to ask potentially sensitive, emotive questions without creating undue distress. For example during bereavement interviews, interviewers sought permission before proceeding with questions about the final episode(s) that precipitated the infant’s death. Once a question was asked, the research team allowed the parent to fully describe
his/her experience before probing about particular details of an event. The interview encouraged the parent to continue the story through non-verbal cues (e.g., nodding) and subtle interjections (e.g., yes, please continue). The research team also was aware that some parents might become fatigued, tearful, or possibly emotionally distraught. The research team was taught to offer to stop the data collection point and initiate the crisis protocol or suicidal screening protocol, if necessary. The research team was trained prior to study initiation and throughout the study period. Principal investigators conducted at least one mock interview with the research team members, as well. Completed parent interviews were reviewed during weekly team meetings to provide feedback to the members of the research team and also assess the response of parents to participation in the interviews. If a particular interview was complex or difficult, it was reviewed by one of the investigators immediately to ensure that the issues were handled adequately and to give both constructive and positive feedback to the research team.

Most parents were described by the interviewers as calm, cooperative, pleasant and friendly, engaged, and relaxed during the data collection point. The father of an infant with complex congenital heart disease described his experience with the study personnel as:

You’ve [interviewer] been very easy to work with, so it’s not like it’s been uncomfortable. But you’ve made us relax through the whole process and that might have been different if it would have been someone else that we didn’t feel comfortable with but as far as you all that we work with didn’t make us feel uncomfortable at any point in time.
A few parents became tearful during the interview when discussing the implications of the life-threatening events or changes in the infant’s medical status. Also some parents experienced tiredness or fatigue during interviews, but when the research team offered to stop or reschedule the interview, parents chose to continue. At times, some interviews were concluded before all the planned questions were asked due to the parent’s emotional state. For example, an interview of a mother who completed the study with her infant was ended early based on the following interaction:

Participant: I’m getting to the point where I don’t want to talk anymore. Not just for interviews, just to anyone…

Interviewer: It sounds like you’re really withdrawing [emotionally]. [Silence] Well let’s call this a wrap okay?

In addition to reviewing parental interview transcripts to evaluate interviewer technique, the research team also examined the parental responses to the interview questions to discern whether the questions were emotionally charged or evoked unexpected responses. Generally, the interview questions did not evoke unexpected responses; however, the ordering of the interview questions was altered in an attempt to initially describe more of the information on how the infant’s illness trajectory had changed since the previous interview and proceed to more sensitive topic areas such as how the parent’s hope has changed for the infant, how the parent was coping with changes, and possible impending decisions that may be necessary based on the infant’s current medical status. Despite the intensive discussions and content of the interviews (e.g., surgery, decreasing parental hope, and increased conflict), most parents responded
“no” when asked about whether the interviews were difficult at the end of monthly interviews and at the completion of the study. Some of the parents who reported feeling that the interview questions were difficult explained the difficulty was related to their infant’s medical situation, the emotive content of the interviews, or their personal struggle of sharing their feelings and emotions with others or trying to suppress their emotions. The father of an infant who required a heart transplant reflected on his experience with the first few study interviews at his final 12-month interview:

It’s just basically—because you don’t want to re-live it because it’s like ‘okay I can’t believe this is happening, I’ve got to talk about it, what if everything don’t go right with the surgery, or it’s not the right heart?’

Three strategies were employed to minimize burden. First, the data collection time points were set to occur no more than 7 days after the previous data collection and generally about 30 days apart. For example, the mother of an infant with a complex congenital heart disease would often approach the interviewer in the hospital between scheduled data collection points to update the research team on the status of her infant and to inform the research team when she thought an interview should be scheduled. Other parents also sent the research team electronic mail and text messages, called the office telephone, and left voicemails to ensure their primary interviewer was aware of how the infant was progressing. Second, interviews were designed to last 30-45 minutes. On average, the interviews last 35 minutes (range 2-164 minutes) per data collection point. Third, to the extent possible, the same member of the research team interviewed the parent throughout the study period to develop rapport and maintain
the relationship. Core members of the research team were also introduced to the parents by the primary interviewer, in case another member of the team needed to conduct an interview. The mother of an infant with complex congenital heart disease described her experience, “I guess it’s been good to like continually talk to someone who met me during this and has like the perspective of the hospital and the doctors and you can like kind of see that side of everything.”

Overall, participation in the research study also did not prove to be too burdensome for parents, despite the fact that their infant often remained critically ill in the hospital or was discharged with chronic illnesses or died as a result of their illness. Finally when parents were directly asked if the study was burdensome, most parents reported the study was not too burdensome or time consuming. The father of an infant following a bone marrow transplant explained the interviews for the first 6 months of the study were not burdensome:

Interviewer: Was this or has it been a burden to have me call you and ask you to do an interview?

Participant: No, no, I mean if I can I can, if I can’t I just can’t, it’s just a lot of times pretty busy but as soon as we get time I’m up for it.

Participation in the interviews did impact the parents in this study. Parents reported that the interviews affected them in three ways: provided an outlet to discuss thoughts and feeling, provided an opportunity to reflect on past events and gain perspective, and were a way of coping with their infant’s life-threatening illness. Parents felt that the interviews provided an appropriate place and time to discuss their feelings
and thoughts about different issues they were facing. Some of the discussions centered on conflict with clinicians or conflict with the other parent. Parents reported that the interviewer allowed them to express their thoughts and feelings without attempting to change the subject and without offering advice. A mother of an infant with a complex congenital heart disease stated: “I think it’s good too that you call just to talk without trying to like fix a problem or change the subject.” The mother of an infant born prematurely described the interviews as:

They’re fine and then for one, even if some things have crossed my mind or whatever, you give me a chance to just speak on them. Somebody’s actually sitting there listening and I think it’s good, to tell you the truth.

Parents reported that many of the conversations they had with their family and friends were superficial and failed to delve into the essence of their experience and how emotionally difficult it was being a parent of a critically ill infant. The interviews allowed parents to explain the details of the infant’s medical condition and how decisions were made for the infant. Through participation parents had an opportunity to reflect on past events and gain perspective. The father of a premature infant explained:

I think it’s good, you know especially in our case just to get in and kind of talk about this stuff, sort of kind of helps put things into perspective and get it out. I think it’s good to come here and talk and kind of, let’s me get some-work some things out that or, come to different conclusions and kind of piece it together in my head and figure things out and I guess another form of you know, release as opposed to like working out which sometimes I don’t get a chance to do… And
it’s been good, and I can talk about how pissed off Sandy [wife] makes me at times and I have something to say she’s not going to get mad at me! [laughs]

The interviews were also seen as a way to cope with the infant’s life-threatening illness and the overall situation. The father of an infant who died following a bone marrow transplant explained this at the 6-week bereavement interview:

Uh…it puts your mind at ease, it lets you get your stuff of your chest instead of holding it in. I mean, if I wouldn’t have went through this to talk through all this stuff I don’t know how I’d be when I came home.

Most parents viewed the interviews as helpful and even reported that they were beneficial. Since the study provided an outlet for parents to discuss thoughts and feelings, the study may have decreased parental stress, anxiety, and post-traumatic stress symptoms. The ability for parents discuss their thoughts and feelings provides insight into what influences the parent’s thoughts and potentially the processes utilized to make decisions. Parents did not report that participation in the study impacted the way they made decisions for their infants. Parents felt that by participating in the interviews they were helping future parents and infants in similar situation. The mother of an infant who required a bone marrow transplant explained, “I mean I don’t think so, I mean it’s definitely hard to talk about it sometimes but that’s the whole point [laugh]. So you all could get you know ways to help other families, so.”

Acknowledgement of the parent’s time and participation in the study helps to demonstrate our appreciation towards their contribution and respect for their willingness to share their time with us. At the end of each interview, study personnel
expressed our gratitude to the parents. Study personnel also sent parent’s birthday cards with a small gift for the infant in recognition of the infant reaching a milestone. For parents of infants who died, study personnel mailed a packet with local bereavement resources and a photo album with photos of the infant included, if possible. A mother of an infant who died following a bone marrow transplant explained that the photo album was something she enjoyed, “when they took pictures and that kind of thing, definitely love the photo book that you guys made of Logan”.


Table 5: Beneficence Strategies

<table>
<thead>
<tr>
<th>Theme</th>
<th>Study Strategy</th>
<th>Responses to Strategy</th>
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| Minimization of physical harm | • Suicide screening protocol  
• Domestic violence and crisis protocol  
• Cell phone protocol  
• Extraneous situations protocol | • No suicidal participants during study period  
• Domestic violence protocol necessary in 1 case  
• Participants were requested to schedule interviews during times when they were not driving or performing dangerous activities. If study personnel became aware that participant was driving, participants were rescheduled.  
• Participant referred to emergency department for illness  
• Suicidal screening protocol utilized during study period  
• Principal investigators also regularly available to study personnel for debriefing to aid staff in managing the intense, emotional responses experienced related to the study.  
• Study personnel improved skill of transitioning participants between emotive topics.  
• Most parents reported that interview questions were not difficult.  
• Participants who thought questions were difficult explained that the difficulty arose due to their infant’s situation, the content of the interview, and that they often try to suppress their emotions. |
<table>
<thead>
<tr>
<th>Minimization of Burden</th>
<th>Acknowledgement of parent participation</th>
</tr>
</thead>
<tbody>
<tr>
<td>• Data collection points set to occur no more than 7 days after the previous data collection and generally 30 days apart.</td>
<td>• Study personnel expressed appreciation at the end of parent interviews for the willingness to share their experience and fit the research study into their schedule.</td>
</tr>
<tr>
<td>• Interviews were designed to last 30-45 minutes</td>
<td>• Birthday cards and token gifts provided to parents to celebrate the milestone.</td>
</tr>
<tr>
<td>• Same study personnel conducted most of the data collection with a single participant, when possible.</td>
<td>• Packets mailed to parents following the infant’s death that included local resources for bereavement support.</td>
</tr>
<tr>
<td>• Between data collection points, some participants contacted study personnel to inform them of changes in the infant’s status and that an interview may be necessary.</td>
<td>• Parents expressed gratitude for cards and joy over celebrating their infant’s birthday. Some parents invited study personnel to the infant’s birthday party.</td>
</tr>
<tr>
<td>• Interviews lasted on average 35 minutes per data collection point</td>
<td>• Most parents reported already receiving bereavement information from the hospital.</td>
</tr>
<tr>
<td>• Most participants reported feeling comfortable and at ease with study personnel. Many commented on the rapport they had developed with the interviewer.</td>
<td>• Parents reported cherishing the photos in the photo albums and thanked the study personnel.</td>
</tr>
<tr>
<td>• When participants were directly asked about whether the study was burdensome, most participants did not report that the study was too burdensome or time consuming.</td>
<td></td>
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</table>

Most parents thanked interviewers for taking time to speak with them and appreciated the flex schedule of the interviewer.

Parents expressed gratitude for cards and joy over celebrating their infant’s birthday. Some parents invited study personnel to the infant’s birthday party.

Most parents reported already receiving bereavement information from the hospital.

Parents reported cherishing the photos in the photo albums and thanked the study personnel.
3.4 Discussion

The purpose of this analysis was to describe how the principles of respect for persons and beneficence were addressed throughout a longitudinal study of parental decision-making for infants with complex life-threatening conditions. The principle of respect for persons was addressed throughout the study by giving participants the option to participate in the study, even when members of the health care team did not think the parents would consent. The research team also attempted to promote respect for persons of the parents by re-consenting participants throughout the interviews. Finally, interviewers were flexible with the time and place the interviews took place. For the most part, parents wanted to meet in a quiet room on the unit, in their infant’s room, or at a location near the hospital during the week. There were some parents that needed to participate via telephone or on the weekends because of their family and work obligations. At times, parents did not feel that they could participate in an interview during a scheduled time and informed the interviewer. Therefore, having flexibility in the research design allowed participation when convenient for the parent and promoted respect for persons.

Implementing strategies to promote beneficence within the study, which was a priority, appeared effective in reducing undue physical or psychological distress. Concern that parents will experience undue physical or psychological distress by participating in research on sensitive topics was not supported by our results. Parents
were tearful when telling their story, but this was expected while discussing the possible
death or actual death of their infant. However, these parents did not appear to
experience undue distress during single interviews or during their time in the study.

Our findings that participation in interviews was beneficial to some participants
are analogous to other findings by researchers conducting studies on sensitive topics
(Emanuel et al., 2004; McGrath, 2003; Michelson et al., 2006). Demonstrating caring
behaviors towards individuals can be perceived as beneficial (Kavanaugh, Moro,
Savage, & Mehendale, 2006). However, participants involved in research on sensitive
topics did not generally feel participation in interviews changed the way they
approached the situation. Michelson et al. (2006) directly asked parents if participating
in the interviews changed their minds or their thoughts about end-of-life decision-
making and found that 79% of the parents did not think participation in the interview
changed their minds. This lends evidence to support that listening to the parent’s
experience does not change their decision-making process. Therefore, we were still able
to capture the experience of making decisions without intervening on the way the
parents make decisions.

A limitation of this analysis is that the parents were asked how the study
impacted them by the interviewer who conducted the interview. Thus, by not having
another independent interviewer complete the questions about the impact of the
participation in the interview, parents may have not expressed any negative thoughts
about the process for fear of hurting the interviewer’s feelings. However, the parents may have felt more comfortable with the interviewer they knew allowing them to provide honest and complete answers. Parents may have expressed different emotions after the interview was completed and they were not with the interviewer. The third limitation was parents who might have thought that interviews would be distressing or burdensome may not have participated in the study. Thus, the findings are limited to parents who did decide to participate. Information about parents who did not participate was not available and further research is needed in this area. Likewise, parents who did not speak English were not included and thus we do not know how parents from different cultural backgrounds would have experienced the interviews.

3.5 Conclusions

This study provides additional evidence to IRBs, funding agencies, and clinicians regarding the response and impact on participants of research studies exploring sensitive topics. This longitudinal study design incorporated specific strategies to promote respect for persons, reduce participant burden and minimize emotion distress when identified. These strategies may have contributed to the positive experience of participants. Individuals are willing to participate in research on sensitive topics, even during stressful times in their lives. It also offers empirical evidence of the impact of prospective longitudinal research. The interviews allow participants an outlet to discuss thoughts and feeling, a way to cope with life-threatening illness, and an opportunity to
reflect on past events and gain perspective. Participants do not experience undue
distress when participating in interviews and completing self-report questionnaires
about sensitive topics. In the future, it may be helpful for researchers to report the
impact and effects of being interviewed on participants within their research study
results to expand empirical evidence.
4. Expected and Unexpected Life-Threatening Conditions: A Single Case Study

Infants who would have previously died of complex life threatening conditions (CLTC) are now surviving due to advances in treatments and technology (Tripp & McGregor, 2006). However, there are still several diagnostic categories that experience high morbidity and mortality despite advances, including infants with complex cardiac anomalies, extremely premature infants (Brandon et al., 2007), and infants with hypoxic ischemic encephalopathy (HIE) (White et al., 2012). Infants with complex life-threatening conditions require parents and health care providers (HCP) to make crucial treatment decisions that affect the infant’s duration and quality of life. Understanding the infant illness trajectory and decision-making trajectory of parents and health care providers (HCP) is vital to determine how to better support parental and provider needs in making decisions. Providing the support parents and providers’ need may improve the overall quality of the infant’s life, decrease parental and provider conflict, and prevent guilt and regret over decisions made for an infant.

Decision-making for infants with life-threatening conditions involves determining which life-sustaining treatments to implement during the immediate postnatal period. These treatments can include continuation of pregnancy (Balkan et al., 2010; Chenni et al., 2012), resuscitation at delivery (Moro et al., 2011), implementation of cardiac surgery (Ellinger & Rempel, 2010), or initiation of other primary treatments.
specific to the infant’s diagnosis. If primary treatments fail, parents and providers must decide when or how to escalate treatment for the infant. Once infants are medically stabilized and are not requiring initiation or escalation of life-sustaining treatments, parents and providers determine how to maintain treatments for the infant to optimize health and well-being (McNamara et al., 2009). For infants for whom life-sustaining treatments have not been effective, parents and providers must make decisions about when and how to transition to a symptom-focused treatment plan that focuses on quality of life. Thus parents and providers caring for infants with CLTC make different types of treatment decisions across the infant’s trajectory of illness.

A critical opportunity on the decision-making trajectory for infants with CLTC is the time prior to delivery during which parents and providers of some CLTC groups can discuss the prognosis and potential treatment options (Payot, Gendron, Lefebvre, & Doucet, 2007). The time allows providers to understand to what extent parents want to be involved in the initiation of life-sustaining treatment decisions (Partridge et al., 2005) and what influences parental decision-making. The communication that occurs between parents and providers may impact the initial decisions to initiate life-sustaining treatment and continue throughout the infant’s illness course (Moro et al., 2011). There is some available research on decision-making for infants with a prenatally diagnosed life-threatening condition (e.g., complex congenital heart disease, holoprosencephaly) or extremely premature infants and how life-sustaining treatment decisions are initiated or
when to withdraw/withhold treatments (Moro et al., 2011; Redlinger-Grosse et al., 2002; Rempel et al., 2004).

Parents and providers of infants with prenatally diagnosed diseases may begin the decision-making trajectory for the infant by determining whether to continue or terminate the pregnancy (Ahmed et al., 2006; Balkan et al., 2010; Chaplin et al., 2005; Chenni et al., 2012; Rauch et al., 2005; Zyblewski et al., 2009). The decision of whether to continue to pregnancy after a prenatal diagnosis may be influenced by the predicted severity of the disease (Balkan et al., 2010; Chaplin et al., 2005; Chenni et al., 2012; Rauch et al., 2005; Zyblewski et al., 2009) and the resources required to care for the infant (Balkan et al., 2010; Rempel et al., 2004). Parents also consider infant pain and suffering (Ahmed et al., 2006; Menahem & Grimwade, 2003) and likelihood of death during infancy (Menahem & Grimwade, 2003; Rauch et al., 2005) when making pregnancy termination decisions. Once the decision to proceed with the pregnancy is made, parents and providers must then decide to what extent life-sustaining treatments (e.g., cardiac surgery, intubation) should be implemented at birth or within the immediate postnatal period. Once parents decide to continue the pregnancy, the parents in conjunction with providers need to determine the extent of life-sustaining treatments to implement for the infant upon delivery and within the first weeks of life. Determining the extent of resuscitation parents desire for their infant prior to delivery allows providers to make immediate treatment decisions with the knowledge of how the parents view initiation of
life-sustaining treatments based on the circumstances (Boss et al., 2008; Grobman et al., 2010; Moro et al., 2011). Providers may also be able to ascertain whether a belief system, personal values and other factors impact a specific parent’s decision-making to ensure the parent’s wishes are respected (Grobman et al., 2010; Moro et al., 2011).

Alternatively, some infants are born with CLTC for which there is no advance warning. Parents who just heard their infant has a life-threatening illness experience emotional shock (Boss et al., 2008; Lan et al., 2007) and difficulty understanding information (Boss et al., 2008; Chaplin et al., 2005; Partridge et al., 2005). The way providers communicate with parents during this distressing time can lead to a trusting relationship (Boss et al., 2008). Parents report that when information is conveyed honestly, compassionately, and with hope, they trust providers more than when they are not given all of the information, are treated like a treatment protocol, and provider’s express no hope for their infant (Boss et al., 2008; Grobman et al., 2010; Moro et al., 2011). The initial communication between parents and providers may influence whether parents trust provider recommendations about the initiation of life-sustaining treatment (Chaplin et al., 2005; Lan et al., 2007).

When full-term infants have a hypoxic-ischemic event at birth, parents and providers face an unexpected complication and the delivery of a very sick infant who was expected to be a ‘normal, healthy’ infant. Providers usually recognize the infant is severely deprived of oxygen only moments before an intervention is initiated to deliver
the infant under emergent circumstances. The emergent situation does not allow time for parents and providers to discuss their wishes regarding life-sustaining treatments. Additionally, exactly how long the infant was deprived of oxygen and the extent of the injury is not known at delivery because the consequences of hypoxic ischemic encephalopathy (HIE) continue to evolve over the first several days of life (Shankaran, Laptook, et al., 2012; Volpe, 2008). This leaves parents and providers with no time to discuss which life-sustaining treatments the parents would like implemented and with limited information about the prognosis of the infant. How parents understand the complex neurological injury, when the infant appears to be a ‘normal, healthy’ full-term infant with no complications throughout the gestational period remains unknown.

Limited research exists about how parents participate and make treatment decisions with providers when an infant is born with an unexpected complex life-threatening condition such as HIE. Yet the early decision-making required to initiate life-sustaining treatments for infants with unexpected complex life-threatening conditions impacts the subsequent decisions for parents and providers because without initiation of life-sustaining treatments, most of the infants die within days or weeks of birth. How the relationship between parents and providers develops during this critical time remains unknown. Therefore, the purpose of this longitudinal, single case study was to explore how parents and providers make decisions for an infant with a known congenital heart defect, who experienced HIE at birth.
Using unique case sampling (Yin, 2009), a single case was chosen from the larger study on decision-making for infants with HIE. Unique case sampling is used when the phenomenon is rarely encountered and understanding the complexities of the case may be useful because patterns that have not been previously described can be identified with in-depth exploration of relationships (Yin, 2009). This case displayed an important phenomenon of parents and providers making critical, emergent decisions that resulted from both expected (congenital heart defect) and unexpected (HIE) infant diagnoses. The emergent decisions related to the unexpected complications were impacted by decisions parents made regarding the prenatal condition of the infant.

4.1 Method

A prospective, longitudinal case study design with qualitative and quantitative data collection and analysis techniques was used to examine the immediate (first 72 hours) and long-term decision-making trajectory (next 6 months) for one infant with a prenatally diagnosed congenital heart defect and an unexpected birth diagnosis of hypoxic ischemic encephalopathy (HIE). Case study design was chosen to allow for the context and complexity of the decision-making trajectory to be explored through multiple sources and types of data (D. J. Edwards et al., 2004). Examining over time the convergence of multiple sources of data related to decision-making of parents and providers allowed for the emergence of patterns and possible causal pathways (Gerring, 2007b).
4.1.1 Sample and Setting

The single case study presented here was chosen from a larger multiple case study of 11 cases of decision making for infants with HIE. In the larger study, a purposive sampling technique was used to select the 11 cases (Sandelowski, 2000). Purposive sampling technique involved choosing cases that varied on potential variables that may impact decision-making for infants: birth hospital, race/ethnicity, and socioeconomic status. The phenomenon of interest in the larger study was decision-making for infants with HIE. To be considered a case the following inclusion criteria were set: (a) infant diagnosed with HIE and enrolled in moderate hypothermia therapy with no major congenital abnormalities that would impact neurological development, (b) at least 2 health care providers had to be willing to participate in the study, and (c) the parent(s) had to speak English.

4.1.2 Measures

Three classes of data were collected to explore how parents and providers made decisions for Alexandria (a pseudonym given to the infant in the case) throughout her life and following her death. The first was medical chart data. This infant illness data was collected daily to develop a portrait of her trajectory of illness and to describe changes in the severity of her illness. The second class of data was narrative interview data. Interviews were conducted with parents within 4 days of enrollment in the study, after any life-threatening event or change in treatment (e.g., cardiac surgery,
tracheotomy placement), monthly until the infant’s death, and 8-weeks after her death. Interviews were conducted with providers within 7 days of enrollment in the study, after any life-threatening event, and within 8 weeks of Alexandria’s death. The third class of data collected was self-report data to describe parents’ level of anxiety and stress across Alexandria’s illness course. Parents and providers also rated the infant’s level of discomfort and their hope for the infant across the illness course. Self-report instruments and hope and discomfort ratings were completed each time an interview was completed.

4.1.2.1 Infant Illness Severity

Illness and treatment characteristics were collected daily from the infant’s medical record. The technology dependence scale (TDS) was utilized to quantify the infant’s severity of illness and technological dependence. This scale was developed by Docherty, Brandon, and Miles across three longitudinal studies of parental caregiving of medically fragile children. The scale includes 12 items, with higher scores indicating more technology dependence and increased severity of illness. Items include care environment, invasive lines, nutrition, monitors, blood draws, respiratory assistance, skin care, specialized oral care, external drains/catheters, colloid administration, mobility, and medications. The initial development and content validity of the instrument was established by focus groups of nurses and parents. Previous studies have shown that TDS significantly decreased over time in samples of premature infants.
and children with cancer. Interclass correlations range from 0.8 to 0.98, in previous samples (Docherty, Brandon, Allen, Silva, & Miles, 2011).

4.1.2.2 Self Report Data

4.1.2.2.1 Parental Stressor Scale: Infant Hospitalization

The Parental Stressor Scale: Infant Hospitalization (PSS:IH) was used to assess parent perceptions of their illness-related stress during the infant’s hospitalization (Miles & Brunssen, 2003). The 28-item scale consisted of 3 subscales: parental role alteration, infant appearance/behavior, and sights and sounds. Each item was rated from 1 (not at all stressful) to 5 (extremely stressful); higher scores indicated more stress. Cronbach’s alphas for the total scale range from 0.85 to 0.91 (Miles & Brunssen, 2003).

4.1.2.2.2 Parental or Provider Perception of Infant Discomfort

A 0 to 100 numeric rating scale was used to assess participant perception of the infant’s level of discomfort. Participants were asked to complete this rating scale at a point during each interview when describing how they felt the infant was doing from a medical standpoint. Thus in addition to providing a quantification of the level of discomfort they felt the infant was in, the rating scale also served as an elicitation tool to encourage parents to talk more about the infant’s suffering. The rating scale has four anchors (no discomfort, some discomfort, moderate discomfort, severe discomfort), evenly spaced across the horizontal line. Participants were asked to mark the line with an X and date their rating. The same scale, with prior ratings, was presented to the participant at each data collection time point so that participants could describe how
their view of infant discomfort changed as the infant’s illness course progressed. The discomfort rating scale was developed by Drs. Docherty and Brandon (2008-2013, 1R01-NR010548, Docherty, PI) for an ongoing study exploring parental and provider decision-making for infants and children with complex life-threatening conditions.

### 4.1.2.2.3 Parental or Provider Perception of Hope

A 0 to 100 numeric rating scale was used to assess participant’s level of hope for the infant. Participants were asked to complete this hope rating scale at a point during each interview when describing how they felt the infant was doing from a medical standpoint. Thus in addition to providing a quantification of their level of hope, the rating scale also served as an elicitation tool to encourage participants to talk more about their hopefulness for the infant. The rating scale had five anchors (no hope, hopeful, somewhat hope, very hopeful, extremely hope), evenly spaced across the horizontal line. Participants were asked to mark the line with an X and date their rating. The same scale, with prior ratings, was presented to the participants at each data collection time point so participants could describe how their view of hope changed as the infant’s illness course progressed. The hope rating scale was developed by Drs. Docherty and Brandon (2008-2013, 1R01-NR010548, Docherty, PI) for an ongoing study exploring parental and provider decision-making for infants and children with complex life-threatening conditions.
4.1.2.3 Parent Interviews

Narrative-style interviews were conducted with each parent individually at study entry, monthly, 1 week following any life-threatening event/significant change in treatment, and 8 weeks following the death of the infant. Narrative style interviews allowed parents to tell the story of the infant’s illness course and decision-making trajectory for the infant and allowed them to discuss important and relevant factors and situations related to their experience (Sandelowski, 1991). Interviews focused on the parent’s account of how the infant’s illness course was progressing and the decisions that had been made for the infant. Additional foci included how decisions were made, factors that influenced the decisions, and the parent-provider relationship. See Table 6 for sample interview questions. Interviews were digitally recorded.

4.1.2.4 Health Care Provider Interviews

Narrative-style interviews were conducted with providers (3 physicians and 3 nurses) at study entry, following any life-threatening event/significant change in treatment, and death. Interviews were digitally recorded. The provider interviews focused on the provider’s story of treating the infant across the illness trajectory and the decisions made for her. Additional foci included the provider’s perception of parental understanding of the infant’s condition, the way decisions were made, the provider’s feelings about the decisions, and factors that influenced the provider’s decisions. See Table 6 for sample interview questions. Providers were interviewed individually.
### Table 6: Sample Narrative Interview Questions

<table>
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<th>Parent Interviews</th>
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| **Study Entry**    | • Tell me the story about how you and your infant came to be here in the ICU.  
|                    | • How are treatment decisions made for your infant?  
|                    | • What decisions have you made for your infant?  
|                    | • Who or what influenced your decisions?  
| **Post Life-threatening Event** | • Tell me what happened to your infant on [day of event].  
|                    | • How was your infant’s treatment plan altered in response to this event?  
|                    | • How were decisions made for your infant related to this event?  
|                    | • What decisions have you made for your infant?  
|                    | • Who or what influenced your decisions?  
|                    | • How was information about the event communicated to you?  
| **Monthly**        | • Tell me what has been going on over the last month with your infant.  
|                    | • Who has been involved in making treatment and care decisions for your infant over the last month?  
|                    | • What decisions have you made for your infant over the last month?  
|                    | • When you think back over what has happened since your infant was born, how do you feel about the specific decisions you have made?  
|                    | • How is your relationship with the providers?  
| **Bereavement**    | • Tell me about how have you been doing the last several weeks?  
|                    | • Tell me what happened when your infant died.  
|                    | • Who was involved in your infant’s care when he/she died?  
|                    | • When you think back about her life, what decisions were critical and why?  

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<th>Provider Interviews</th>
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| **Study Entry**     | • Tell me about how you met this infant and his parents.  
|                     | • What decisions have been made for this infant?  
|                     | • Who was involved in the decisions?  
|                     | • What or who influenced the decisions?  

Post Life-threatening Event
  • Tell me what happened this week to this infant.
  • Did you anticipate this event?
  • How is the infant progressing now?
  • What decisions were made for this infant?
  • What or who influenced the decisions?

Bereavement
  • Tell me about what happened leading to this infant’s death.
  • What do you think about this infant’s illness course and treatment trajectory?
  • What were the critical decisions and how do you view them now?

4.1.3 Data Collection Procedures

All data were collected by the first author. Self-report and interview data were collected in a private location in the hospital. The parental bereavement interviews were conducted in the parents’ home. At each data collection time point, parents completed the self-report data and then participated in the narrative interview. Self-report data took approximately 15 minutes to complete and the narrative interviews lasted an average of 70 minutes. A total of 11 interviews were conducted with the parents (6 with mother and 5 with father). Providers completed the self-report instruments and then participated in narrative interviews that lasted an average of 30 minutes. A total of 10 interviews were conducted with providers (6 with physicians and 4 with nurses). The data for this case study included 21 narrative interviews with participants.

The digitally recorded interviews were transcribed verbatim (Bloomberg & Volpe, 2008). Each written transcript was checked to ensure congruence between the written transcript and the digitally recorded interview. Each participant was given a
pseudonym to ensure confidentiality. The list of participants was kept locked in a file cabinet separate from the transcribed interviews.

Approval for the study was obtained from the Institutional Review Board for Research Involving Human Subjects. Once consent was obtained from the parents for their participation in the study they were asked if we could approach their infant’s health care providers for study participation. Health care providers were then consented. Data collection started the day after the infant was born and continued for the 172 days of her life and through 8 weeks after her death.

4.1.4 Data Analysis

The overall goal of the analysis was to explore the decision-making trajectory for Alexandria and to describe how parents and health care providers made decisions and what factors influenced these decisions. The over-arching method of analysis for this case study was exploration of displays of data trajectories and the visual search for patterns or trends in the trajectories to gain a deep understanding of the patterns of decision-making. In order to construct these trajectories, several steps occurred to transform the different classes of data into visually comparable units.

In the first step of data transformation, daily infant illness and treatment data from the medical chart reviews was used to classify each day of the infant’s life into one of four treatment categories: initiation of treatment, maintenance of treatment, escalation of treatment, or withdrawing/withholding treatment. The characterization of each day of
the infant’s life into one of these categories allowed visual display of the types of
treatment the infant needed throughout her illness course. This also allowed for the
visual display of the infant’s illness trajectory using these categories as a context in
which all of the illness and decision events occurred. The visual display was used to
understand how the treatment categories interacted with other attributes such as
parental hope and parental stress and themes from the interview data. Table 7 provides
the definitions for each decision type and examples from Alexandria’s illness course.
Table 7: Treatment Categories

<table>
<thead>
<tr>
<th>Treatment Categories</th>
<th>Definition</th>
<th>Examples</th>
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<tbody>
<tr>
<td>Curative Care</td>
<td>Care focused on curing disease or illness</td>
<td>• Cardiopulmonary resuscitation at delivery</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Respiratory support at or near delivery</td>
</tr>
<tr>
<td>Initiation of treatment</td>
<td>Decisions about the treatment plan based on the initial diagnoses or new diagnoses</td>
<td>• Moderate hypothermia</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Cardiac surgery</td>
</tr>
<tr>
<td>Escalation of treatment</td>
<td>Decisions about addition of medical treatments and technologies in an attempt to treat the illness or condition</td>
<td>• Use of Octreotide for reoccurrence of pleural effusions</td>
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<tr>
<td></td>
<td></td>
<td>• Tracheotomy tube placement</td>
</tr>
<tr>
<td>Maintenance of treatment</td>
<td>Decisions about whether and how to maintain the level of care</td>
<td>• Gastrostomy tube feedings</td>
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<td></td>
<td></td>
<td>• Management of ventilator setting</td>
</tr>
<tr>
<td>Palliative Care</td>
<td>Care focusing on noncurative treatments to relieve suffering</td>
<td></td>
</tr>
<tr>
<td>Withdrawing/withholding</td>
<td>Decisions about withdrawing or withholding life-sustaining treatment</td>
<td>• Withdrawal of mechanical ventilation</td>
</tr>
<tr>
<td>treatment</td>
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Conventional content analysis was used to analyze the narrative interview data, which allowed for identification of themes and related patterns (Hsieh & Shannon, 2005). The data were approached systematically, extracting the main storyline to identify key events in the infant’s illness course and the associated decision-making trajectory.

Then individuals involved in specific decisions were identified (Sandelowski, 1995). Factors that impacted decision-making were then grouped together under the broad
categories such as ‘influences’, ‘communication’, ‘hope’, and ‘conflict’. Definitions for categories were developed from the data and refined as the analysis progressed (Hsieh & Shannon, 2005). Once data were separated into broad categories, subcategories emerged allowing similar ideas to be clustered together. Each subcategory was also defined and clarified as the analysis continued. Data analysis was concurrent with data collection to inform future data collection.

Next, the self-report data was analyzed using descriptive statistics and graphing techniques. This data provided additional descriptors about possible influences on the decision-making trajectory. Scores from each instrument and subscales of the PSS:IH were graphed to identify patterns. The PSS:IH subscales were evaluated to identify if any subscale had more variability over time and further described data collected from interviews. Once total scores for instruments and the subscales were calculated, the scores were normalized on a scale of 0 to 1 to allow for visualization of all scales simultaneously without losing the variability of each scale.

The last step in the analysis was to develop the decision-making trajectory displays to search for patterns and describe the relationship between decisions that were made and the influential factors. The background of each display was the infant illness trajectory with the treatment categories and discrete events (e.g., cardiac surgery) that altered the illness course. The decision-making trajectory was constructed from the medical chart data and interview data to include decisions by parents and providers. A
decision was defined as a choice between two or more options. The decision-making trajectory was visually represented by short textual descriptions chronologically as the decision was made.

Single variables (e.g., hope, discomfort, and infant illness severity) were then superimposed on the illness trajectory and decision-making trajectory for each participant to isolate factors that may have impacted decision-making of the participant. After displaying each variable by participant, a single variable was compared between participants over time. Each visual display was searched for patterns including areas with similar or discrepant participant views, specifically at times where decisions were made.

4.2 Results

4.2.1 Alexandria’s Illness Trajectory

The case included an infant, parents, three attending physicians, and three primary care nurses. The infant, Alexandria (pseudonym), was born at 36-weeks gestation with a prenatally diagnosed congenital heart defect. She experienced a hypoxic-ischemic event during delivery. Alexandria’s mother and father were an unmarried couple and had been cohabitating for the previous 4 months prior to delivery. The mother had a child (2-years-old) and the father had three children (all adults) from prior relationships. The parents reported Christianity as their religious preference. The mother had a master’s degree and worked in social services. The father
had a college degree and owned an information technology company with reported annual family income between $26,000 and $50,000. Three attending physicians and three primary registered nurses who cared for the infant participated in the case. The attending physicians included the primary attending physician in the intensive care unit, a pulmonologist, and a cardiothoracic surgeon. All of the providers were Caucasian, the five providers had children, and five providers reported Christianity as their religious preference. Data collection spanned from birth through Alexandria’s death at 5 months of age, and continued through 8 weeks post death.

Alexandria’s entrance into the world was expected to require some medical interventions because of her prenatally diagnosed congenital heart defect. However, the majority of the complications she experienced during the perinatal period were unexpected both in that she had great difficulty transitioning to neonatal life and the intensive interventions she required to stabilize her physiologic status. Alexandria required cardiopulmonary resuscitation during the delivery period due to fetal distress. She experienced a hypoxic-ischemic event during the perinatal period and underwent moderate hypothermia therapy for 72 hours. The cranial ultrasound on the day she was born did not reveal any acute intracranial bleeds. But the extent of the hypoxic-ischemic injury could not be determined early in the hospitalization.

Following the initial stabilization, Alexandria required high frequency ventilation and nitric oxide until her heart was surgically repaired at DOL 6. Cardiac
surgery proceeded as planned with no complications. Alexandria was diagnosed with partial DiGeorge syndrome in her second week of life, which weakened her immune system and she had multiple episodes of sepsis. She was unable to be weaned from the ventilator and diagnostic examination revealed a new diagnosis of severe bronchotracheomalacia. Tracheostomy and gastrostomy procedures were performed to facilitate long-term ventilation and nutrition. The gastrostomy tube dislodged within 72 hours of the initial surgery and Alexandria required an emergency laparotomy. Around two months of age, Alexandria continued to have pleural effusions that failed to resolve without chest tubes. In total, she was diagnosed with four primary complex illnesses: congenital heart defect, HIE, bronchotracheomalacia, and DiGeorge syndrome. To illustrate the complexity and intensity of Alexandria’s first 2 months of life, each day of treatment was categorized by the primary treatments she required to meet the overall goals of therapy. The daily treatment categories (initiation of life-sustaining treatment, escalation of treatment, maintenance of treatment, or withdrawal of treatment) are displayed as the background colors in Figure 3. A quantification of Alexandria’s severity of illness using the TDS over time is displayed over the first 2 months of life. Two peaks of increased severity of illness were demonstrated during the period around her cardiac surgery and placement of the tracheostomy and gastrostomy tube and need for the emergency laparotomy. Throughout her fourth and fifth months of life, Alexandria began having multiple episodes per day of desaturations, bradycardia, and hypoxia. In
her final week of life, the episodes increased and Alexandria was slower to respond to curative treatments. Alexandria died from respiratory failure at Day of Life 172.

Alexandria lived 172 days in the intensive care unit through the support of mechanical ventilation and other life-sustaining treatments.

Figure 3: Infant Illness Severity over First 2 Months of Life

4.2.2 Decision-Making Trajectory for Alexandria

Daily decisions about the treatments needed to remain hemodynamically stable and sustain life were made for Alexandria. Figure 4 illustrates the major treatment decisions made for Alexandria throughout her 172 days of life. The treatment decisions are superimposed on the daily treatment categories representing the treatments she required to meet the overall goals of therapy. Alexandria experienced several life-threatening events with 19% (n = 32) of the intensive care focused on escalating treatments (pink columns). Her decision-making trajectory is marked by 77% (n = 132) of her the treatment decisions focused on how to maintain her level of stability (blue
columns). These decisions generally focused on adjustments to the ventilator settings and trying to get Alexandria to tolerate the full volume of tube feeds to meet or exceed her caloric needs.

Figure 4: Major Treatment Decisions Associated with Illness Severity

4.2.3 Trajectory of Parental Experience: Stress, Infant Discomfort, and Hope

Throughout Alexandria’s life, parental stress and parents’ view of infant discomfort and hope varied over time for both mother and father. Figure 5 (Trajectory of Mother’s Experience) and Figure 6 (Trajectory of Father’s Experience) display each of the three normalized parental experience attributes and the decisions that coincided on the illness trajectory.
Figure 5: Trajectory of Mother's Experience: Stress, Infant Discomfort, and Hope

Initial displays of PSS: IH subscales and total score were examined visually in the context of the findings from the interview data. For the mother, the infant behavior and
appearance subscale of the PSS:IH demonstrated the most variability and converged with themes from the interview data into a pattern of high stress associated with the behavior and appearance of Alexandria across her illness trajectory. Maternal stress was the lowest when Alexandria was born, but increased as her illness trajectory progressed with the highest stress about behavior and appearance during the month prior to her death. The mother’s view of Alexandria’s discomfort was the lowest at birth and reached the highest level around the time she underwent the tracheostomy, gastrostomy, and emergent laparotomy. The mother’s hope for Alexandria was highest at birth and during periods of her illness trajectory that consisted mostly of maintenance of treatment. The mother’s hope was lowest when the tracheostomy, gastrostomy, and emergent laparotomy were performed.

Mother’s attributes of infant discomfort, hope, and stress appear to vary based on her overall view of how Alexandria would be able to interact and communicate with others in the future. Throughout her infant’s illness course, when asked about her decisions about continuation of curative treatment, the mother focused on the future and her infant’s ability to function in society and lead a happy life. When asked about the point in her daughter’s treatment that she would consider withdrawal of therapies, the mother replied:

If she was going to be on a ventilator forever, never learn to read or write or communicate in a meaningful way. I think that’s probably the biggest part is if she’s never able to communicate. Then I don’t think you could have a good quality of life without communicating. So if she’s for whatever reason, if it’s brain damage, if it’s just the DiGeorge is so severe or if for some reason she’s
never able to hold a conversation with somebody just because she can’t articulate her thought or whatever, I think that’s probably what scares me the most if that’s going to keep her from having all these social relationships and sometimes I wonder if people who have trouble communicating are thinking and feeling things that they just can’t get out and that’s got to just frustrate the hell out of them and make them feel absolutely miserable all the time and I don’t want that for her.

The mother’s hope decreased as her view of the infant’s discomfort increased and coincided with an increasing maternal stress level. The infant’s technology dependence and severity of illness also increased along with this pattern of increasing discomfort and decreasing hope. The mother’s hope that her daughter would lead a ‘normal, healthy’ life faded over time as the infant’s illness trajectory continued with limited improvement in her overall condition, continued dependence on technology, and few possible treatment options available for the severe bronchotracheomalacia. The mother explained that her stress was related to the uncertainty that Alexandria was not getting the stimulation and interactions she needed to develop in a healthy way. After 3 months in the ICU, the mother’s primary concern was development.

I guess our biggest concerns now are some of the just physical therapy aspects. Just whether she’s being set up and played with and whether they’re worried about her circuit so she has a chance to move the other arm. Usually her left arm gets pinned under it pretty often and she’s showing a lot of stiffness in the left arm, so we’re worried about that. Just interaction in general. Especially since she’s still under precautions it seems like they just kind of do what they have to do and scoot out of there so she doesn’t get a lot of play time, interaction time or people just talking to her or playing with her.

The mother’s highest level of hope occurred during a period at 3-months of age as Alexandria’s illness trajectory began to stabilize. During the stabilization period, the providers and parents were discussing plans to move Alexandria to a step-down unit.
and beginning to coordinate the services she would need when transitioning home with her parents.

The father’s stress varied throughout Alexandria’s illness course reaching a peak with the placement of the tracheotomy and gastrotomy tubes. When the PSS:IH subscales and total scores were graphed, the father exhibited the highest stress related to parental role attainment, which is displayed in Figure 6. The father’s stress levels related to his parental role attainment were a prominent discussion during the interviews. At birth, the father faced many unexpected events upon arrival of his daughter, as he was unable to protect his daughter from a difficult transition from intrauterine to extrauterine life. The father described his thoughts about not knowing more about Alexandria’s condition: “This is not the road that we thought we’d be on, I think this could have been easier on Natalie if I had more knowledge, a better understanding on all the technical stuff that goes on with Alexandria.” The father had his lowest level of parental role alteration stress following the cardiac surgery. The low level of stress coincided with the father reporting that he felt he did something to help his daughter by researching and finding the best cardiothoracic surgeon and hospital for her, prior to her birth. His stress was highest and hope was the lowest following placement of the tracheostomy and gastrostomy tube surgery. The high stress level was attributed to his feelings of failing to advocate strongly enough that Alexandria not have the surgical
procedures (tracheostomy and gastrostomy tube) because he did not believe his infant was stable enough to tolerate a non-emergent invasive procedure.

She had not been in my opinion she had not been doing well. This goes back to we just wanted her to be more stable before they start the next set of shit on her. She had her feeds turned off, turned back on twice I think it was prior, within four days of going into the surgery. They did a study on her the day before, concerns because her abdomen was already distended or swollen whatever fluid, she had diarrhea, she had fever, three different fevers over the course of four days prior to the surgery.

While the father’s hope was highest when his daughter was born, his hope quickly decreased reaching its lowest level after his daughter underwent the placement of the tracheotomy and gastrostomy tube at day 37. The father’s hope slowly increased over the months following the placement of the tracheostomy and gastrostomy tube, but never reached the level of hope he had at Alexandria’s birth. The father focused on his ability to provide and protect both the infant and the infant’s mother. The father’s view of the infant’s discomfort level remained relatively low and constant, even as the infant’s condition worsened over time. High parental stress and low levels of hope associated with the decision to proceed with the tracheostomy and gastrostomy tube procedures appeared to lead up to the development of conflict between the parents and providers.

4.2.4 Conflict Related to Mismatched Hope

4.2.4.1 Parents

As demonstrated on the decision-making trajectory, prior to the diagnosis of bronchotracheomalacia, parents’ only experience with treatment decisions was related to the decision to proceed with cardiac surgery. And this decision was made prior to the
birth of Alexandria. The parents reported no involvement in the decisions to initiate life-sustaining treatment through resuscitation of Alexandria at birth and initiation of moderate hypothermia therapy that occurred immediately after delivery. The physician caring for Alexandria during this period reported that parents were not involved in the decision for moderate hypothermia therapy because “it wasn’t a research protocol at the time. I think we informed them that was what we were doing. But it wasn’t a protocol that we would typically ask permission for.”

The father explained that despite not knowing the extent of the neurological injury, the parents felt that the original prenatal plan to proceed with cardiac surgery did not need to change.

We still are unaware of neurological issues, but nothing they are doing today or tomorrow [cardiac surgery] is going to have any impact on where that’s already going to be. That’s already developed, she’s already been depleted of oxygen, she’s already had these things happen…We’re aware of the possibility of having to make some tough decisions, I guess fortunately we have not had to make any yet.

The parents did not disagree with the decision to initiate life-sustaining treatments through cardiopulmonary resuscitation at birth or moderate hypothermia therapy during Alexandria’s first week of life. In fact, parents had the highest level of hopeful before Alexandria had cardiac surgery during her first week of life. Yet when the father reflected on the decision to initiate cardiopulmonary resuscitation after Alexandria was diagnosed with bronchotracheomalacia, he disagreed with the decision to initiate cardiopulmonary resuscitation because he believed that ‘it crossed the line’ of
what was an acceptable level of treatment for his daughter, which was when the parents
had the lowest level of hope.

That line’s been crossed by reviving her, but it was not a decision that we made. And
that decision, if we were to have made it was dependent on if we knew how
severe she was and really how sick she was at the time. Now I’m at a point to
where I feel that line has been crossed, as far as, bringing her further than what
maybe we should have. And the reason for that is because deciding factors now
on how to handle the situation are all unknown questions: ‘will she suffer brain
damage?’ we don’t know, ‘how long will she be on the vent?’ we don’t know.
Well we did know at delivery that if they did not aggressively treat her heart and
respiratory system she would have died. That was a for-sure, black and white
situation. There was no question. There was no guessing.

The decision to escalate treatment by choosing to place a tracheotomy and
gastrostomy tube was seen by parents as a decision to continue her life without evidence
that the procedures would improve her overall happiness and well-being. The decisions
could, in fact, prolong the quantity of her life, but not improve the quality of her life. The
mother explained, “we debated the trach and whether we were I guess putting her
through too much for how her life was going to be afterwards”.

The decision was not about life and death now, it was about whether it was just
and fair to possibly prolong their daughter’s life without knowledge of her mental
capacity to communicate and perform tasks on her own and form meaningful bonds
with other individuals. Figure 7 demonstrates how hope scores varied between
participants, specifically at the time the tracheostomy and gastrostomy were performed.
Figure 7: Parental and Provider's View of Hope

When the parents were deciding whether to proceed with the placement of the tracheostomy and gastrostomy tube, their hope for the infant decreased and their view of the amount of discomfort the infant increased. The parents reached their lowest level of hope and highest level of perceived infant discomfort during this time period. The parents described this point in time of choosing for their infant to be dependent on technology as a decision to travel down a specific path. Her father stated, “We needed to decide where we were as her parents, are willing to take her or put her through”. The parents’ decreased hope was related to feeling as though their daughter would never recover to an acceptable level of functioning and would be dependent on medical technology that would impact her ability to interact with the others and the environment.
An interesting contrast in the focus of parental hope occurred when comparing the focus of hope surrounding the period of her cardiac surgery and the focus of hope surrounding the period of her tracheotomy and gastronomy tube placement. Table 8 contrasts how parental hope changed by highlighting descriptively how a hope for a normal life and recovery without needing ‘radical’ treatments after cardiac surgery became hope for her to having a ‘somewhat’ normal life and giving her whatever she needed to be happy.

Day of Life 32 seemed to become a transition point on the decision-making trajectory during which parents began to deeply question whether or not they should continue implementing additional life-support measures that would be a permanent fixture in their daughter’s life for at least the first several years of her life and possibly forever. They sought information from many sources to understand the possible complexity of the infant’s illness in both short- and long-term scenarios. The parents discussed palliative care and withdrawal of support for their daughter with each other and with “trusted” members of the health care team only.

I do not feel that when or if this decision needs to be made, I do believe that Natalie and I will be able to find that middle ground. I don’t think that our differences are so great that we will not come to an agreement without major complications if that makes sense. Decision on whether or not to bring in hospice, and take her off life support. And just let her go, stop sticking stuff in her and cutting her open doing all these things because man is so damn smart and we’ve got all this figured out and we can fix anything, and it’s coming with a great cost...we’ve decided not to have her go down for the surgery [pleurodesis] yesterday. She’s going to need the surgery if we choose to keep her on life support, the ventilator. But if we don’t, there’s no sense in putting her through that again, so we’re getting down to a 94, 48 hour, maybe even 24 hour time
period of Natalie and I, hopefully talking to the right people and getting enough information for us to make a decision as far as what direction we go with Alexandria.
### Table 8: Participant Descriptive Changes in Hope Between Cardiac Surgery and Tracheotomy and Gastronomy

<table>
<thead>
<tr>
<th>Participant</th>
<th>Cardiac Surgery</th>
<th>Tracheotomy and Gastronomy</th>
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<tbody>
<tr>
<td>Father</td>
<td>The obvious is to live...I want her to get to a point, as quickly as possible and as easily as possible to be comfortable and start growing and go through the normal process of being a baby to a toddler and grow up. I want her to get through the surgery and I want her to recover quickly...I want her to maintain stability so they do not have to do additional radical things to keep her going through recovery.</td>
<td>Every parent’s hope. Long, healthy and happy life...We’ve just got to give her everything she needs to have a happy life, but ultimately she’s the one that has to find that happiness right?</td>
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<tr>
<td>Mother</td>
<td>Just overall health like coming out of it, being able to function. Not needing machines to keep her alive. Just be a normal kid.</td>
<td>I just hope that she would continue to get stronger and come home soon and be off the ventilator in a year and maybe have a little trouble learning to write her letters but otherwise not so much difficulty and maybe it’s a little tougher for her to know how to socialize and not have all the social skills but still be able to make friends and have relationships and go to the prom and still at least have a somewhat normal life.</td>
</tr>
<tr>
<td>Attending Physician</td>
<td>I think there's some hope that she'll be able to be a meaningful member of society and a positive participant in her family life...I think if there was no hope at all then we shouldn't be doing what we're doing.</td>
<td>My hope for her right now is that the initial hypoxic event won’t amount to anything. I’ve seen kids with much rockier births that they may have some cerebral palsy or some degree of dysfunction but you wouldn't know it... My hope is that any injury from that would be mild. Right now we can’t really know because she’s remained sedated on the ventilator.</td>
</tr>
<tr>
<td>Surgeon</td>
<td>She's got DiGeorge syndrome and that has some implications for higher likelihood of neurologic or developmental delay neurologic</td>
<td>I'm still hopeful that she will stop draining fluid out of her chest, be weaned from the ventilator, and eventually get her trach</td>
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difficulties, but I’m hopeful that she won’t have those things and I am hopeful she will have long intervals between subsequent cardiac operations she’s going to require to have her valve changed and so forth. Depending on how much or how little she was injured by the combination of the traumatic birth and the operation itself a pretty reasonable quality of life.

Primary Nurse

My hope for her would be to get her surgery done...then for her to get extubated and to do well and go home. Considering her actual diagnosis that’s very hard to basically quantify. You really don’t know. One patient can do super great and go home and within a week be awesome and then some can just not do that at all.

She’s been awake and kind of looking around but just be a little bit more interactive with her family and sitting up in the chair and doing baby stuff and you can do that now with a trach. For her to just hit those milestones like she should. She’s going to hit them a little bit later but to go home with her family and to be doing well and not have any issues.

In addition to the decision to add to her technological dependence it was at this point in the illness trajectory during which the parents began struggling with how their daughter’s hypoxic-ischemic event at birth and the multiple episodes of hypoxia she had experienced since her initial transition to the extrauterine environment would impact her cognitive development. The decision of whether to proceed with the placement of the tracheotomy and gastronomy tubes and add to her technological dependence or transition to palliative care was seen as a no win decision as described by the mother:

If we choose to throw every procedure possible at her and maybe one day bring her home and when she’s 15 and all she does is cry and watch cartoons all day because she can’t communicate in a more meaningful way we’re probably going to make a different decision now, but we had no way to know that. But I don’t want to decide it was too much for her and in 15 years wonder what kind of 15-year-old she would be. What if she had been okay, what if she was going to get
better and we quit on her and we just didn’t give her a chance. So no matter what I decide I’m going to worry I made the wrong decision.

These difficult discussions regarding the balance between continuing with aggressive, curative focused care and symptom focused palliative care were initially kept between the parents. However, at 48 days of life parents raised the issue during a family meeting with the pediatric quality of life team, the cardiothoracic surgeon, and the primary physician whom they felt they could trust. These team members were individuals they felt could be trusted to give them a balanced assessment of what Alexandria was facing and the implications of such a decision on the family. While some providers appeared to recognize that the parents could be weighing the options of palliative care or withdrawal of support this was the first point on the illness trajectory that topic of an alternate, symptom focused, pathway for Alexandria was discussed between parents and the health care team. A physician explained why the parents were not approached by the providers about withdrawal of support:

I think that I have not broached the issue because I haven’t felt it appropriate from my perspective and that’s caregiver bias, but I think that if they haven’t verbalized it to other people I’d be surprised although there’s a tremendous amount of guilt that goes with that for parents. So they may not have but it’s certainly something that when they’re by themselves or when they’re together and alone from the outside world they’ve either thought about or discussed it.

The providers waited for the parents to bring up the issue of transitioning to palliative care. The mother explained how the discussion about palliative care unfolded:

We were questioning whether to move forward with her care or just kind of make her comfortable and we decided to move forward with care because of a lot of things. I think largely because of the reaction that we got when we told everyone that we were considering not moving forward and everyone seemed to
be really shocked that we would consider that at this point since she’s I guess she was so close to recovering with the exception with this one little problem, they thought they could treat pretty simply.

When the parents did approach the topic of palliative care, the providers told them that they did not think Alexandria’s illness trajectory would result in a poor quality of life and that she had a good chance at leading a happy normal life. A physician explained after the tracheotomy and gastrostomy tube placement that he was Still hopeful that she will stop draining fluid out of her chest, be weaned from the ventilator, eventually get her trach decannulated and have her own life that doesn’t involve a trach and is going to involve some more cardiac surgery but go to kindergarten and grow up and do the other things that kids do.

The parents ultimately decided to proceed with the tracheotomy tube and gastrostomy tube based on their morals and logic that without the procedures the infant had no chance of being discharged home to be with her family. The dislodgement of the gastrostomy tube following the placement of the tracheostomy and gastrostomy tube with resultant emergency laparotomy was the ‘tipping point’ in the parental decision-making trajectory. The parents now questioned whether proceeding with the initial tracheostomy and gatrostomy surgery was just because they were not sure Alexandria would benefit from the surgery in regard to long-term happiness and quality of life.

4.2.4.2 Providers

While parents struggled immensely with the decision to proceed with the tracheostomy tube and gastrostomy tube, some of the providers did not think there was any grey area in the decision. As the parents struggled with whether they had done too
much to the infant, they asked an attending physician if placement of the tracheostomy tube and gastrostomy tube were appropriate, he stated, “I don’t think we’ve crossed the line where we’re doing too much.” The surgeon and nurse were in agreement that proceeding with placement of the tracheotomy and gastrostomy tube was an appropriate decision for the infant because neurologically she appeared intact and she was responding to the other treatments. The providers’ reported the interventions as an improvement of Alexandria’s ability to interact and engage more with her parents and the environment. The procedures were also expected to aid in her strength and growth so that she could be discharged home. However, the attending physician was more cautious with his hope because of his experience with children with severe bronchotracheomalacia and the long-term consequences and complications of mechanical ventilation. If Alexandria was going to be able to go home with her parents, the providers’ felt that tracheostomy tube placement with mechanical ventilation and gastrostomy tube was necessary. The nurse remained hopeful that the infant would be able to go home with her family with few complications. Table 8 highlights how provider hope changed between cardiac surgery and the tracheotomy and gastrostomy tube placement. Providers continued to maintain an overall positive outlook on the infant having a few difficulties, but being able to developmentally progress.

The cardiothoracic surgeon and attending physician reported that palliative care through withdrawal of the ventilator was not appropriate at the time of the placement of
the tracheostomy and gastrostomy tubes point in the infant’s illness trajectory. A physician caring for the infant explained that

She’s tolerated all the other interventions reasonably well from a neurologic standpoint and she seems to be doing what I would expect a 1-month-old baby who’s been as sick as she’s been in fact, she’s quite alert and interactive or at least alert and I think to make the decision now not to go forward with something that we had anticipated was likely to be required wouldn’t make a lot of sense at this point.

Another provider explained the reasoning behind that decision was that the infant did not have, in his view, a condition that was “immediately lethal or lethal in any reasonable time frame”. The providers thought of the placement of the tracheostomy and gastrostomy tube as a ‘normal’ progression for the infant to continue along her illness course. The decision did not appear to create any conflict between providers.

4.2.5 A Paradox Over Doing More

By day 32 the interviews with parents and providers presented a paradox in their views on how Alexandria was progressing in her course of illness. While the parents were uneasy about the aggressive nature of the focus of her care given limited prognostic information on her likelihood of recovery, they eventually acquiesced to placement of the tracheotomy and gastrostomy tube placement, but it was not without reservations. Following a meeting between parents and the general surgeon responsible for inserting the gastrostomy tube, the father described his feeling that there was lack of understanding by the general surgeon of the significance of the surgical procedure to this family and the father felt very negative after this meeting. While gastrostomy tube
placement is a common procedure for chronically critically ill children, for parents this
decision was not just about their daughter’s need for safe nutrition. This decision was
complex and required them to resolve themselves to ‘doing more to’ their daughter with
limited information about her neurological status and how she would develop over the
coming years.

Yet, when the general surgeon discussed the technical aspects of the gastrostomy
tube placement with the parents, she failed to address the implications that this common
procedure had for the parents. The father described his discussion with the general
surgeon about placement of a gastrostomy tube, as:

The whole attitude I got from her was I think she used a “walk in the park”, a
“piece of cake” a couple times. And I looked at her I said um ‘yeah but this one’s
different, that’s my daughter’ and I was very serious, and I was having a bad day
already and was nervous about the surgery.

The attempt by the general surgeon to calm the parents’ fears and apprehensions
about the upcoming surgical procedures left the parents feeling as though their
daughter’s life had been minimized and that their concerns were invalid and
unimportant. Despite the parents understanding of the complexity of the first several
weeks of their daughter’s life, the general surgeon did not acknowledge the moral and
emotional struggles the parents were facing with this decision. On the day that these
surgical procedures were to occur Alexandria’s father felt that her condition had been
too unstable over the past couple of days to warrant a non-emergent procedure and he
asked to have the surgery postponed. He expressed these concerns to one of the
providers, but he was not given the opportunity to decline the procedures at this point. The father was told that there were “questions over whether I had the right to give consent”. Thus, the surgery proceeded as scheduled because the mother signed consent for the procedures before she had spoken with the father between the time he was okay with the surgery and the change in his thoughts about the impending surgery.

Initially, Alexandria did not experience any complications related to the surgery. However within 72 hours, she demonstrated signs and symptoms of sepsis and possible dislodgement of the gastrostomy tube. The same surgeon who reassured the parents that a gastronomy tube placement was routine, common, and posed little risk to their daughter, was now telling the parents that their daughter must return to the operating room for repair and replacement of the gastronomy and that her septicemia required close monitoring. The parents did not assign blame to the surgeon for the gastrostomy tube dislodging, instead the father felt that he was the one responsible for the complication, “I didn’t want her to go into that surgery and I should have stood up for my daughter but I didn’t, um yeah there’s some guilt there.”

4.2.6 Parents Become Watchful, Alert, and Vigilant

Between Trajectory Days 70 and 90, parents and providers were intensively involved in making maintenance treatment decisions for Alexandria. This epoch of time appeared to demonstrate a great deal of parent and health care provider conflict. Following the emergency laparotomy surgery, Alexandria’s parents developed a pattern
of response to her illness and treatment trajectory that can be characterized as constant and intense monitoring for any signs or symptoms that the medical team may be overly aggressive with their treatments. Here the Father describes is experience with recognizing a problem just prior to the emergency laparotomy:

Monday and Tuesday, Natalie and I together with each other went to the nurse and the doctors and asked about her belly being, she looks like she’s retaining fluid, yeah that’s common after surgery. That’s normal, that’s normal, that’s normal.

The father reported having guilt after not advocating ‘enough’ for his daughter in the 48 hours before her emergency laparotomy, even though he knew something was wrong:

A friend of mine told me it’s not my job. It is my job because I’m her father and that’s where the guilt comes from not doing a good enough job, being a good enough advocate for her up here. Come to find out Tuesday they eventually did a contrast study on her, realized that the tube had come out of her belly.

It is at this point in time when Alexandria’s father expressed his need to be more involved in the decision-making related to maintaining the treatments for his daughter’s care. He felt that his assessments and fears regarding how his daughter was tolerating the maintenance treatments were not taken seriously. He explained:

I will literally ask and tell them that we would feel better if, this specific thing was done. Like at first reducing her feeds and then ultimately taking her off the feeds to give her belly a break because of the distention and firmness that she is dealing with.

The father’s feeling that his concerns were not taken seriously continued through the remainder of his daughter’s life. At 56 days on the trajectory, due to perceived conflict, the medical team assigned a consistent intensive care attending physician to
care for Alexandria in order to provide continuity in the plan of care and meet weekly
with the parents to discuss major changes in the plan of care, answer questions on a
regular basis, and discuss the plan of care for the following week. Yet even with the
weekly meetings with the primary intensive care physician and planning Alexandria’s
care weekly, the father felt that he was not included in the decision-making.

Having a family meeting and thinking of a game plan or set of goals was made
for that week and within 24 hours later having a new or a different fellow come
on and decide that they wanted to just choose a different direction that wasn’t
talked about, not knowing that we had already had a discussion on that and start
making changes that were not a list goals that we had already set forth for that
following week.

The mother also exhibited constant monitoring behavior in the care of her
daughter through searching for any sign of physical or psychological discomfort. The
mother discussed her concerns about the level of discomfort her daughter felt between
tube feedings:

We just don’t think it [feral bag] works as well as just actually popping a tube off
and putting a syringe with gauze in it for half an hour or so. It just doesn’t seem
to be as effective. It’s not like we can look at it and see whether it’s working so
much as she seems to be more comfortable when they put the syringe on every
couple hours. Because the bag stays on all the time and she gets kind of fussy
with gas, but when they do the syringe every few hours she seems to be pretty
fine with the gas, so we just prefer it.

However, the mother reported discussing her concerns and recommendations
with one of the providers and when she came back the next day, the message had not
been relayed and her daughter had been uncomfortable. The mother thought a lot about
the psychosocial needs of the infant and how the infant rarely had pleasant experiences,
“never gets loved”, and “she just gets poked”. The mother was concerned about the
quality of life their daughter would experience and not how many days or years she could live within the hospital. While the mother searched for an understanding of the long-term outcome if their daughter survived, few providers discussed the implications.

The mother explained her concerns:

Sometimes I don’t know if we’re blowing this so out of proportion and we’re thinking that her life is going to be so bad. There are plenty of kids that have it worse and they still have happy lives and no one up here has really offered an opinion of how they think it’s going to be for her. Except Dr. A told us when we talked to him about ligation surgery it kind of got brought up that he thinks there definitely is a point that you’re just putting a child through more than it’s going to be worth to them later in life and he doesn’t think Alexandria is at that point. He thinks she still has a very good chance at a happy life.

The differences in the interpretation of how to best care for the Alexandria led to a breakdown of communication between the parents and providers. One of the primary nurses explained:

The providers, a lot of time, were very accommodating but frustrated also. Because they wanted to be able to take care of Alexandria first and foremost and I think there at the end it was kind of a repetitive story, as far as, what had happened in an event…He (father) wanted to know everything and you know sometimes you can’t recap everything. I mean you condense it and when you condense things sometimes things get missed. Not intentionally, but you know. So I think he got up-to-date information but it was sometimes delayed.

As the communication breakdown continued the parents began to lose trust in anything the providers tried to talk to them about. The perceived lack of communication and the parents feeling that their simple requests were not honored progressed to parents questioning whether they could trust providers on decisions about what was best for their daughter. The father questioned the motives behind why providers were adamant about replacing the central line at DOL 100 that she had since DOL 6.
I think that they forced this surgery to try to get her out of the ICU quicker and they have not said this to me yet but I believe that they are going to tell me that she will not be able to get up on full feeds and she will be coming home on some type of TPN. We asked them a few days ago when this line was first brought up and we were heavily assured that they believe she will go home on full feeds but I believe that the placement of this line and not wanting to wait a few more days to put it in is a push to get it in because they already know she’s not going to get to full feeds.

The mother questioned whether she could trust the medical team to make a decision about palliative care when the time came that interventions truly became futile because the team’s job was “to keep people alive”.

4.2.7 Decision to Transition to Palliative Care

On Day of Life 172, Alexandria’s parents decided to withdrawal her life-support. It was the first time that the parents were approached by the providers to consider a DNR order. Providers were unable to maintain Alexandria’s oxygen saturation, despite increased respiratory support and medications to improve physiologic compliance of the lungs. During an emergent event when providers were unable to stabilize Alexandria, the parents chose to withdraw support, the mother explained how they decided to withdraw life-support:

Just seeing her that way she was in an emergency situation like that just out of nowhere I think it really signaled hey she’s not going to be okay and I couldn’t stand there for hours and hours while they tried, only to have the same result. I think we were both tired of watching her suffer, and watching more and more things needing to be done just to keep her alive, it’s too much, she was miserable and it wasn’t getting her anywhere.
4.2.8 Regret Impacts Future Decisions

The parents learned they were pregnant with another child several weeks before Alexandria died. Their experiences with Alexandria’s illness trajectory and their decision making process impacted their thoughts and feelings about this new pregnancy. The mother explained her view during an interview 8-weeks after Alexandria’s death:

I called Megan [social worker] last week to see if she knows if we would need a lawyer if we wanted to make some kind of DNR or do not intubate for the pregnancy now. So far nothing is wrong so thank God everything looks fine. But I don’t think we can go through that again if this baby is born and there is something wrong enough and they need to intubate. I don’t want to go there. I don’t want to play that game of well we think it will be fine, cause obviously we had no idea, but I regret a lot of the choices that we made and wish we had stopped this a long time ago because all we did was just torture her, she was miserable. It wasn’t a recovery, it was just an experiment and she didn’t get better. I understand that they didn’t know either and that some babies goes through 6 months in the hospital and then by the time they are 10 they’re fine, but that didn’t happen. I feel like we had 6 months of mistakes or longer if you count knowing we I was 20 weeks pregnant.

The father continued to view the decision to resuscitate Alexandria at delivery to be the most important decision the parents did not get to make. He too regretted continuing aggressive care for an extended period for Alexandria.

DNR before this baby is delivered. I compromised myself. I have always felt I had that line drawn to where we intervene too much medically. What pisses me off this most of this whole thing is that we weren’t given the opportunity to make the decision and one that was most critical to make and that is when she was born. I don’t think the heart surgery was crossing the line, no matter how her delivery went. That was a reasonable procedure. Given all of her complications that became an unreasonable procedure, but we brought her that far to where I felt that we now owed it to her that chance. We have crossed a line where do you draw that new line at and that is the thing that I personally have struggled with, I never expected to have to draw a second line. I think that is harder than drawing the first line.
4.3 Discussion

A prenatal diagnosis of a congenital heart defect prepared Alexandria’s parents and health care providers to expect the birth of an infant who would require some respiratory support to bridge her through to necessary cardiac surgery. But undiagnosed bronchotracheomalacia resulted in her respiratory arrest with subsequent cardiac arrest requiring cardiopulmonary resuscitation at birth and the initiation of multiple life-sustaining treatments. While the parents had prepared prenatally for their daughter to undergo cardiac surgery, they did not expect Alexandria to have additional complex health problems. Throughout Alexandria’s life her parents remained concerned about how the undiagnosed bronchotracheomalacia, impaired oxygenation, and resultant HIE would impact their infant’s neurological status.

The parents focused on how the unknown neurological status of Alexandria would impact her quality of life. They described a good quality of life as being able to interact with others, communicating and forming relationships, and being happy. This description is consistent with previous explanations parents have given about assessing quality of life (Ahmed et al., 2008; Hinds et al., 2000; Hinds et al., 2009). Alexandria’s parents questioned to what extent life-sustaining support was appropriate and at what point, the care was futile because her quality of life would be poor. Questioning the futility of treatments based on the neurological status and whether the infant will suffer from developmental disabilities has been identified previously as influencing parental
decisions (Einarsdottir, 2009; Vermeulen, 2004); however, many parents want all treatment options exhausted before withdrawing/withholding support even if severe disabilities are a likely long-term complication (Carnevale et al., 2011; Moro et al., 2011).

Parental experiences with stress, hope, and infant discomfort appeared to vary as their view of Alexandria’s quality of life changed across her illness trajectory. Parental view of hope for Alexandria was not aligned with the providers’ hope for her. Most providers were more hopeful for the infant than the parents. The discrepancy led to conflict about how much to escalate Alexandria’s treatments, while balancing the parents’ overall objective of ensuring their daughter would have a good quality of life. The parents were considering transitioning from curative care to palliative care after learning the seriousness of the bronchotracheomalacia. The parents questioned whether a good quality of life could be attained for Alexandria given the combination of all the complications she had endured within a short period of life. However, when the parents attempted to broach the topic with providers, they were given the impression that their assessments of how their daughter’s illnesses impacted her quality of life were incorrect. This disagreement led to conflict between parents and providers, specifically around the decision to escalate care by placing a tracheostomy and gastrostomy tube. This decision became the tipping point in the parental decision-making trajectory, in which parents refused to allow their daughter to be ‘put through’ more invasive procedures. The parents regretted the decision and refused to continue providing invasive treatments.
Differences between parent and provider hope is not uncommon and has been reported to lead to impaired relationships between parents and providers (Grobman et al., 2010). Neurological status of the infant is usually the underlying problem that leads to conflict between parents and providers (Verhagen et al., 2009). However, the parents generally view their infant’s future neurological outcomes as minimally impacted; whereas, providers predict a more grim prognosis of long-term outcomes for the infant (Verhagen et al., 2009).

After acquiescing to escalate support for their daughter and the subsequent emergency laparotomy, the parents became alert, watchful and vigilant about the day-to-day treatment of her illnesses. When Alexandria suffered the complication with the gastrostomy tube dislodging, the parents decided that they were not going to allow Alexandria to endure any more pain and suffering from procedures and treatments. The parents attempted to control Alexandria’s treatments through being involved in each minor decision and requests for frequent medical updates. However, the parental request of increased communication between the parents and providers on a daily basis were not honored as often as the parents wanted.

During the latter period of their daughter’s illness course, the parents viewed the lack of follow through with their requests as a sign of poor communication and a lack of respect for their input about the treatment of their daughter. This point on the decision-making trajectory appeared to be the start of major conflict between the parents and
health care providers. Poor communication spiraled into parents not trusting the providers. The lack of trust led to more conflict and parents questioning the care being provided. Overall, these parents were very concerned with how each decision to continue treatment for their daughter would impact her quality of life. While each individual illness was not seen as detrimental to Alexandria’s quality of life, parents felt that the summative effect of all of the chosen treatments would result in a developmentally compromised infant with a poor quality of life. Ultimately, the parents withdrew life-support but not until, in their perception, late in the trajectory, and at the point when providers thought it was an appropriate decision. By this time, the parents felt guilt and regret about the extent of medical treatment and suffering their daughter endured.

4.4 Implications for Future Research

Decision-making for expected and unexpected complications at birth remains difficult with no ‘right’ or ‘wrong’ choices. Parents and HCPs must collaborate with each other to provide the best treatments for individual infants. How to make decisions about the best treatments remains unknown. Continuing to explore how decisions are made over a trajectory of the infant’s illness may provide information on how to aid parents and HCP in making decisions. Determining when to discuss infant quality of life or how long to continue providing curative care for the infant is necessary. Future research may focus on how parents change their view of infant quality of life throughout the illness.
trajectory. Longitudinal, mixed methods designs may help to identify when views change or perhaps become a more prominent influence in decision-making.

Specifically focusing on how parents and HCPs communicate with each other throughout the entire trajectory may allow researchers to identify key areas or key events that likely create conflict within the parent-parent or parent-HCP relationship. Resolving conflicts quickly may help decrease parental mistrust of HCPs and decrease parental regret. Future research should focus on times when conflict is known to occur and try to identify illness trajectories or characteristics of infants, which conflict is common in and study the entire illness course from birth or diagnosis until treatment decisions are concluded and short or long-term outcomes are available.

4.5 Implications for Clinical Practice

Prenatal diagnosis of complex life-threatening conditions allows both parents and providers time to discuss how to proceed with treatment for the unborn infant. However, the prenatal decision-making may create a false sense of security in terms of expectations at delivery and the first several weeks of the infant’s life. Providers may assume that parents want all available treatment options exhausted at delivery if a complication occurs so that the prenatal decision to initiate life-sustaining treatment of the diagnosed condition is continued. Yet, some parents have ideas about an acceptable quality of life for their infant or the extent of invasive medical treatments they view as appropriate that is in conflict with the views of the health care team. When the injuries
or treatments are ‘too much’, but there is no recourse to go back, parents and providers must work together to understand how to renegotiate quality of life and extent of medical treatment that are now deemed acceptable proceeding forward in the infant illness trajectory. Prior to delivery, providers and parents need to discuss ‘what if’ situations and attempt to clearly delineate to what extent life-sustaining treatments are appropriate and continue to reevaluate these views as the infant’s illness trajectory continues. Nurses can help facilitate these discussions with the parents through asking them what they want for their infant and parental understanding of how treatments impact the long-term outcomes. Nurses are critical in communicating how the infant is progressing and what treatments have been provided on a daily basis. Identifying how and when parents want to have information communicated to them may also help decrease communication breakdowns between parents and providers. Providers also need to acknowledge that within the health care team, differences in opinions on how the infant is progressing may occur. Providers also ask when parents have differences in the way they want their infant treated. Maintaining open communication between parents and providers appears to be essential to decreasing conflict and ultimately parental regret about decisions.
5. Trajectories of Decision-Making for Infants with Hypoxic Ischemic Encephalopathy

Hypoxic ischemic encephalopathy (HIE) is a brain injury that occurs because of a hypoxic or asphyxia event during the prenatal, intrapartum or postnatal periods that prevents adequate blood flow to the infant’s brain (Volpe, 2008). Hypoxic ischemic encephalopathy (HIE) is one of the most serious complications of full term birth (Schiariti et al., 2008), occurring in up to 2.5 per 1000 live births (Graham et al., 2008; Kurinczuk et al., 2010; Martinez-Biarge, Madero, Gonzalez, Quero, & Garcia-Alix, 2012). Infants with HIE experience associated morbidities with 40-83% of these infants either dying by age 2 or having severe disabilities (Jacobs et al., 2011; Kwon et al., 2011; Simbruner et al., 2010). The long-term neurological consequences of HIE include hearing impairment, seizures, bilateral blindness, severe cognitive disabilities, and cerebral palsy (Shankaran, Pappas, et al., 2012; Simbruner et al., 2010). Thus much of the current neonatal research on HIE focuses on preventing further brain injury or repairing areas of the brain that are damaged (Jacobs et al., 2011; Shankaran, 2009; Shankaran, Pappas, et al., 2012).

Infants with HIE transition into the extrauterine environment in a compromised physiologic state (Volpe, 2008). Failure to successfully transition from the intrauterine to the extrauterine environment can be a result of fetal or maternal complications (Martinez-Biarge et al., 2012). The negative impact of oxygen and nutrient deprivation to
vital organs must be corrected within minutes to hours to prevent a cascade of deleterious effects (Cotten & Shankaran, 2010). If the deleterious effects are interrupted through medical treatments, the infant may not suffer additional consequences of the hypoxic-ischemic event (Shankaran, Laptook, et al., 2012). However, the injury that has already occurred, specifically to the brain, may be permanent. Determining which treatments to provide to infants with begins immediately because adequate oxygen and nutrition must be restored to prevent further cellular injury and death (Volpe, 2008).

Critical decisions must be made within minutes to several hours following the birth including the length of resuscitation, type of respiratory support, the level of care the infant needs (i.e., need to transfer to a NICU that has different technologies and therapies available), and experimental interventions (see Figure 8 for predicted decision-making trajectory over the first week of life).
Resuscitation of full-term infants for at least 10 minutes is recommended by the American Heart Association (AHA) (Kattwinkel et al., 2010); therefore, the decision to initiate resuscitation is not generally optional, but continuing past the recommended 10 minutes is left to the discretion of HCP and parents. In the past, treatment options were limited to standard medical treatment (e.g., antiepileptic medications, respiratory support) (Hoehn et al., 2008; Vannucci & Perlman, 1997).

Recently, Level IV NICUs began implementing moderate hypothermia treatment, as common practice, for infants who demonstrate signs of HIE within 6 hours of birth (Kapetanakis et al., 2009; Lang et al., 2007). Some infants with HIE are
transferred to NICUs that offer moderate hypothermia treatment (Jacobs et al., 2011). Even infants with HIE who receive moderate hypothermia treatment continue to have mortality rates of 25-38% and severe disability rates of 21-35% (Jacobs et al., 2011; Shankaran, Pappas, et al., 2012; Simbruner et al., 2010). One of the early randomized clinical trials (National Institute of Child Health and Human Development) comparing whole body moderate hypothermia (n = 97) and standard care (n = 93) in infants with HIE initiated < 6 hours of age found no significant difference in the primary outcome of death or IQ < 70 in the same children at 6-7 years, even when severity of HIE was considered (Shankaran, Pappas, et al., 2012). Additional large clinical trials that used selective head cooling with mild systemic hypothermia (CoolCap) or whole body hypothermia (TOBY) have not published outcomes of their participants at 6-7 years of age, at this time. Whether moderate hypothermia or selective head cooling treatment improves childhood outcomes of infants with HIE remains uncertain. All of the decisions about the length of resuscitation, type of respiratory support, the level of care the infant needs, and the use of moderate hypothermia treatment must be made within the first 6 hours of life by parents and HCPs.

Decision-making about enrolling in experimental interventions usually need to be made within the first 24-72 hours of life to potentially obtain optimal outcomes (Allen & Brandon, 2011). Experimental interventions are available for infants with HIE at some academic medical centers that may prevent secondary energy failure (Zhu et al., 2009) or
potentially repair areas of the brain injured during primary injury failure (Pimentel-Coelho et al., 2012). The two experimental interventions available for the sample of infant with HIE at this academic medical center were experimental moderate hypothermia intervention initiated after the first 6 hours of age and autologous umbilical cord stem cell transplant. Experimental moderate hypothermia intervention enrolls infants with HIE who cannot receive the intervention within 6 hours of life. Autologous umbilical cord stem cell transplant is a safety trial with the main objective to determine whether umbilical cord blood can be harvested and administered in conjunction with other treatments available for infants with HIE without side effects.

For infants who survive the initial weeks of life, parents and HCP must decide the types of technology they need to support respiratory and nutrition status and when to start physical, speech, and occupational therapy (see Figure 9 for predicted decision-making trajectory after the first week of life). Infants with severe HIE who required extensive resuscitation and had a high base deficit noted on the umbilical cord blood gas may benefit more from supportive treatment options rather than exposing them to the potential side effects of experimental interventions (Azzopardi, 2010). In some cases of severe, extensive injury, infants may be transitioned to palliative care treatments.

Predicting which infant will respond to each treatment and how the treatments may affect the short and long-term outcomes for these infants are uncertain. Limited data exist about outcomes among school-aged children who received moderate
hypothermia (Shah, 2010), short-term outcomes other experimental treatments (Allen & Brandon, 2011; Pazos et al., 2012; Sarkar et al., 2012; Zhou et al., 2010), and the long-term outcomes of advanced treatments (e.g., surgical procedures, high frequency ventilators) available to support the underlying cause of the hypoxic-ischemic event and subsequent complications. How parents and HCP determine, which treatments may be most beneficial also remains unknown.

Figure 9: Predicted Decision-Making after Initial Week of Life

The advent of advanced technology and complex interventions to treat infants, who would have previously died, has increased survival, but has also created more treatment options for parents and HCPs to choose from for critically ill infants. The
availability of immediate and long-term treatments for infants with HIE may have created a complex decision-making trajectory for parents and HCPs. As with other trajectories, the individuals involved in making the decisions, their previous experiences, and personal characteristics may effect how the decision-making trajectory progresses (Magnusson & Cairns, 1996). The initial decision and infant’s response to continued resuscitation (e.g., chest compressions) and immediate stabilization (e.g., respiratory support) affects the next decisions. As seen in Figure 8, the treatment decision and the infant’s response to each treatment chosen will impact the decisions available as the infant progresses through the illness trajectory.

The main decisions after initial week of life will reflect the extent of the damage from HIE and the infant’s response to previous treatment decisions. The infant responses required prior to discharge are adequate nutrition and ventilation, which can be accomplished through different methods, if necessary. After discharge infants will require follow-up in developmental clinics initially (Robertson & Perlman, 2006). Some infants may also need physical therapy depending on the severity or area of brain injury. Some infants may continue to remain unstable or have severely impaired cognitive function and the decision may be made to transition from aggressive care to palliative care.

The predicted extensive parental and HCP decision-making trajectory for infants with HIE is complex. The initial decisions must be made within 6 hours of birth for an
infant who experience an unpredicted complication (Martinez-Biarge et al., 2012).

Parents and HCPs must made immediate decisions that will affect the infant’s illness course and the long-term outcomes. However, without warning of an impending complication, parents and HCPs have little time to discuss treatment options and how parents are included in this decision-making remains unknown. Additionally, what factors influence how the decisions are made also remains unknown.

5.1 Purpose and Research Questions

Understanding the parental and HCP decision-making trajectory may lead to the development of interventions to optimize decision-making for infants with HIE. Previous research suggests that parents and HCPs disagree about the neurological status of infant prior to withdrawing/withholding treatments (Garros et al., 2003). The disagreement may create communication breakdowns by HCPs not being as hopeful as parents or viewing the quality of the infant’s life as minimal. The contrast of parental and provider appraisals of neurological outcomes can lead to conflict and parents advocating against treatments suggested by the providers (Garros et al., 2003).

Understanding how and why decisions and treatments are initiated from birth through the first 6 months of life may allow for identification of when in the decision-making trajectory becomes disconnected between parents and providers. Therefore, the aim of the study was to explore parental and HCP decision-making trajectories for infants with HIE from birth through 6 months of age.
Research Questions

1. What are the key decisions parents and healthcare providers make when caring for infants with HIE?

2. Who is involved in decision-making for infants with HIE over time?

3. What factors influence parents and health care providers decision-making for infants with HIE and how do these influences affect decision-making?

5.2 Methods

A longitudinal, prospective, multiple case study design was used to study the parent and HCP decision-making trajectories for infants with HIE. Case study design was chosen to allow for the context and complexity of decision-making to be explored through multiple sources and types of data (D. Edwards, F. Dattilio, & D. Bromley, 2004). This approach allowed for an in-depth exploration of how and why decisions were made for infants with HIE. In order to explore how parents and HCPs make decisions for infants with HIE over time, the case study design enabled the examination of multiple types and sources of data (D. Edwards et al., 2004). Exploring the convergence of multiple sources of data related to decision-making of parents and HCP allowed for the search for patterns (Gerring, 2007b).

5.2.1 Setting

The setting for the study was a tertiary academic medical center in the Southeastern United States. Infants in the study were cared for in a Level IV neonatal
intensive care unit (NICU), pediatric intensive care unit, and step-down areas. Infants with HIE received follow-up care between 5-7 months of age at the outpatient special infant care clinic (SICC).

5.2.2 Sample

A purposive sampling technique was used to select the 11 cases that were enrolled in the study (Bloomberg & Volpe, 2008; Sandelowski, 2000a). To be considered a case the following inclusion criteria were set: (a) infant diagnosed with HIE and enrolled in moderate hypothermia treatment with no major congenital abnormalities that would impact neurological development, (b) at least 2 HCP willing to participate in the study, and (c) the parent(s) had to speak English. If two parents consented, data collection methods were conducted separately with each parent. The HCP for each case included at least two of the following: physician, nurse practitioner, and primary registered nurse.

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<th>5.2.2.1 Participant Demographics</th>
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<td>Ten of the 11 cases studied are included in this analysis. The findings from one case were analyzed and reported in a separate paper because the infant had congenital abnormalities and a genetic syndrome, which led to different types of decisions and a more complex trajectory. In total 10 infants, 17 parents, and 33 HCPs (15 physicians and 18 nurses) were studied across the 10 cases. Table 10 provides a broad description of the illness course for each infant. Infants were between 34 and 41 weeks gestation at delivery. All of the infants required respiratory or cardiopulmonary resuscitation at</td>
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delivery with 8 of the 10 infants receiving mechanical ventilation at least the first Day of Life. Nine of the infants were transferred in from an outlying facility to receive moderate hypothermia treatment that was not available at the birth hospital. Seven of the 10 infants were diagnosed with moderate HIE and the other 3 infants had severe HIE. The infants were hospitalized between 12 and 64 (mean 29 days) days of life. All infants were discharged home in the care of their parents. All infants survived through the 6-month study period.

Nine mothers and 8 fathers participated with a mean age of 29 years (range 20-39). Eight parents were Caucasian not Hispanic, seven were African-American, and two were Caucasian with Hispanic ethnicity. The majority of parents (n=10) were not married and 11 parents had more than 1 child prior to the birth of their new infant. Eleven of the 17 parents had a high school education or less and 2 parents had some college and 4 parents had a college education. The majority of parents (n = 9) were unemployed with 8 parents reporting an annual household income level of $15,000 to $25,000 and 6 reporting less than $15,000 in annual household income. Of the 15 physicians who participated, 10 were male, 10 were Caucasian not Hispanic and 4 were Caucasian, Hispanic and 1 was Asian. All nurses were female, 12 were Caucasian, 5 were African-American and 1 was American Indian.

Table 9: Characteristics of Infant Illness Courses

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<tr>
<th>Case</th>
<th>Description of infant illness course</th>
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2 Transferred from outlying facility after intubation and 10 minutes of cardiopulmonary resuscitation. Admitted to hospital with severe HIE, hypotension, respiratory distress syndrome, rule out sepsis, thrombocytopenia, metabolic acidosis, and DIC. Started moderate hypothermia treatment at < 6 hours of life. Seizures throughout first 16 DOL requiring continuous EEG monitoring. Gastrostomy and Nissan at DOL 55. Discharged home at DOL 64. Required in-home occupational and speech therapy at least once a week.

3 Transferred from outlying facility for seizure activity at 8 hours of life. Admitted to hospital with moderate HIE, seizures, and rule out sepsis, and coagulopathy. Enrolled in an experimental moderate hypothermia intervention because > 6 hours of life at symptom onset. Multiple seizures throughout the first 5 DOL requiring continuous EEG monitoring. Infant discharged home at DOL 13. Readmitted for rule out sepsis at DOL 35.

4 Transferred from outlying facility with poor Apgar scores and initial pH < 7.0, meeting guidelines for treatment with moderate hypothermia treatment at < 6 hours of life. Admitted to hospital with moderate HIE and rule out sepsis. Enrolled in experimental umbilical cord transplant study. Required intubation at DOL 4 through DOL 8 for respiratory distress. Discharged home at DOL 15.

5 Transferred from outlying facility after intubation and 2 minutes of cardiopulmonary resuscitation for moderate hypothermia treatment < 6 hours of life. Admitted to hospital with severe HIE, rule out sepsis, coagulopathy, possible seizures, and right pleural effusion. Multiple seizures throughout first 6 DOL requiring continuous EEG monitoring. An infection and feeding difficulties complicated the hospital course. Discharged home at DOL 42. Required in-home physical therapy.
6 Transferred from outlying facility after requiring respiratory support in the delivery room, low Apgar scores, and seizure activity. Admitted to hospital with moderate HIE, rule out persistent pulmonary hypertension, respiratory insufficiency, metabolic acidosis, rule out sepsis, rule out HSV, rule out MRSA, and clinical and electrograph seizures. Started moderate hypothermia treatment at < 6 hours of life. Multiple seizures repeated throughout first 3 DOL requiring continuous EEG monitoring. Discharged home at DOL 14.

7 Transferred from outlying facility after requiring intubation and cardiopulmonary resuscitation for 12 minutes for moderate hypothermia treatment at < 6 hours of life. Admitted to hospital with severe HIE, persistent pulmonary hypertension, possible seizures, liver dysfunction, renal insufficiency, cardiomegaly, and persistent metabolic acidosis. Gastrostomy and Nissan at DOL 31. Discharged home at DOL 42. Required in-home occupational and speech therapy.

8 Transferred from outlying facility after requiring intubation and cardiopulmonary resuscitation for 1 minute for treatment with moderate hypothermia treatment < 6 hours of life. Admitted to hospital with moderate HIE, meconium-aspiration, persistent pulmonary hypertension, rule out sepsis, acidosis, DIC, and fetal distress. Hospital course complicated by respiratory failure and severe meconium-aspiration requiring ECMO from DOL 2 through 24. Gastrostomy and Nissan at DOL 52. Discharged home at DOL 62. Required in-home physical therapy.

9 Transferred from outlying facility after requiring intubation and having no spontaneous movement for first 12 minutes of life for moderate hypothermia treatment at < 6 hours of life. Admitted to hospital with severe HIE, seizures, r/o persistent pulmonary hypertension, r/o sepsis, transitory neonatal electrolyte disturbance, term pregnancy acidosis, chronic hypokalemia, and neonatal hypoglycemia. Discharged home at DOL 14.

10 Admitted to hospital with moderate HIE after requiring intubation at birth for moderate hypothermia treatment at < 6 hours of life. Enrolled in experimental umbilical cord transplant. Discharged home at DOL 12.
5.2.3 Measures

To study parent and HCP decision-making trajectories for infants with HIE, multiple sources and types of data were collected including infant chart reviews and narrative interviews with parents and HCP. The purpose of obtaining information about the infant’s illness was to determine how changes in the infant’s illness course impacted parent and HCP decision-making. The narrative interviews with parents and HCP allowed for identification of influences on decision-making including parental understanding of the injury and the relationships between parents and the parent-HCP relationship. The parent self-report instruments provided information about parental anxiety, stress, and posttraumatic stress symptoms, which may impact decision-making. Parents and providers also rated infant discomfort and their overall hope for the infant with HIE.

5.2.3.1 Infant Illness and Treatment Characteristics

Illness and treatment characteristics were collected daily from the infant’s medical record while the infant was hospitalized and then at each visit for follow-up care. Following discharge, information about infant health status was collected in the monthly parent interviews.

The technology dependence scale (TDS) was utilized to quantify the infant’s severity of illness and technological dependence. This scale was developed by Docherty, Brandon, and Miles across three longitudinal studies of parental caregiving of medically
fragile children. The scale included 12 items, with higher scores indicating more technology dependence and increased severity of illness. Items include care environment, invasive lines, nutrition, monitors, blood draws, respiratory assistance, skin care, specialized oral care, external drains/catheters, colloid administration, mobility, and medications. The initial development and content validity of the instrument was established by focus groups of nurses and parents. Previous studies have shown that TDS significantly decreased over time in samples of premature infants and children with cancer. Interclass correlations range from 0.8 to 0.98, in previous samples.

5.2.3.2 Parent Demographics

Parents completed a demographic questionnaire upon enrollment with information on parental age, race, occupation, marital status, religious preference, education level, income, number of people living in household, number of children, distance to the hospital from home, previous intensive care experience, and previous experiences with life-threatening illnesses.

5.2.3.3 Parent Interviews

Narrative interviews were conducted at study entry, monthly, 1 week following any life-threatening event/significant change in treatment, and 2 weeks following hospital discharge. Interviews were digitally recorded. Data collection measures continued until the infant was 6 months of age. Narrative interviews were used to
encourage the participants to tell their story about making decisions for their infant. It also gave the participant some control over the direction of the interview (Sandelowski, 1991). The two main foci of the monthly interviews were how the infant was progressing through the illness course and the parental experience of making decisions for the infant with HIE. Parents were interviewed individually. Interviews lasted about 15 minutes (range 4-54 minutes). See Table 10 for sample interview questions posed to parents over the 6-month study period.
### Table 10: Sample Narrative Interview Questions

<table>
<thead>
<tr>
<th>Parent Interviews</th>
<th>HCP Interviews</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Study Entry</strong></td>
<td><strong>Study Entry</strong></td>
</tr>
<tr>
<td>• Tell me the story about how you and your infant came to be here in the ICU.</td>
<td></td>
</tr>
<tr>
<td>• How are treatment decisions made for your infant?</td>
<td></td>
</tr>
<tr>
<td>• What decisions have you made for your infant?</td>
<td></td>
</tr>
<tr>
<td>• Who or what influenced your decisions?</td>
<td></td>
</tr>
<tr>
<td><strong>Post Life-threatening Event</strong></td>
<td><strong>Post Life-threatening Event</strong></td>
</tr>
<tr>
<td>• Tell me what happened to your infant on [day of event].</td>
<td></td>
</tr>
<tr>
<td>• How was your infant’s treatment plan altered in response to the event?</td>
<td></td>
</tr>
<tr>
<td>• How were decisions made for your infant related to this event?</td>
<td></td>
</tr>
<tr>
<td>• What decisions have you made for your infant?</td>
<td></td>
</tr>
<tr>
<td>• Who or what influenced your decisions?</td>
<td></td>
</tr>
<tr>
<td><strong>Discharge</strong></td>
<td><strong>Discharge</strong></td>
</tr>
<tr>
<td>• What are your thoughts about your infant’s hospitalization?</td>
<td></td>
</tr>
<tr>
<td>• What decisions did you make for your infant throughout the hospitalization?</td>
<td></td>
</tr>
<tr>
<td><strong>Monthly</strong></td>
<td><strong>Monthly</strong></td>
</tr>
<tr>
<td>• Tell me what has been going on over the last month with your infant.</td>
<td></td>
</tr>
<tr>
<td>• Who has been involved in making treatment and care decisions for your infant in the last month?</td>
<td></td>
</tr>
<tr>
<td>• What decisions have you made for your infant over the last month?</td>
<td></td>
</tr>
<tr>
<td>• When you think back over what has happened since your infant was born, how do you feel about the specific decisions you have made?</td>
<td></td>
</tr>
<tr>
<td>• How is your relationship with the providers?</td>
<td></td>
</tr>
</tbody>
</table>
How was the discharge of the infant and parents managed?

5.2.3.4 Parent Self-Report Instruments

5.2.3.4.1 Parental Stressor Scale: Infant Hospitalization

Parental Stressor Scale: Infant Hospitalization (PSS:IH): was used to assess parent perceptions of illness-related stress during the infant’s hospitalization (Miles & Brunssen, 2003). This scale was only administered while the infant was hospitalized. Parents rated their stress related to parental role alteration, infant appearance/behavior, and sights and sounds in the hospital on a 28 item scale (Miles & Brunssen, 2003). Each item is rated from 1 (not at all stressful) to 5 (extremely stressful); higher scores indicate more stress. The content validity of the PSS:IH was established through literature review and interviews conducted with parents of medically fragile infants (Miles & Brunssen, 2003). Cronbach’s alphas for the total scale range from 0.85 to 0.91 (Miles & Brunssen, 2003).

5.2.3.4.2 Impact of Events Scale-Revised

The 22-item Impact of Events Scale-Revised (IES-R) was used to assess current parental distress in response to a specific life event (Weiss & Marmar, 1997). Items reflect posttraumatic stress symptoms including intrusion, avoidance and hyperarousal (J. G. Beck et al., 2008). Respondents rate each item on a scale from 0 (not at all) to 4 (extremely) for the past 7 days. Scores range from 0 to 88. Criterion, content, and construct validity have been established (Weiss & Marmar, 1997). Internal consistencies for the total scale are 0.95 to 0.96; for the intrusion scale 0.90 to 0.94; for the avoidance
scale 0.86 to 0.87; and for the hyperarousal scale 0.85 to 0.91 (J. G. Beck et al., 2008; Creamer, Bell, & Failla, 2003).

5.2.3.5 Parental and Provider Self-Report

5.2.3.5.1 Parental or HCP View of Infant Discomfort

A 0 to 100 numeric rating scale was used to assess participant perception of the infant’s level of discomfort. Participants were asked to complete this rating scale at a point during each interview when describing how they felt the infant was doing from a medical standpoint. Thus in addition to providing a quantification of the level of discomfort they felt their infant was in, the rating scale also served as an elicitation tool to encourage parents to talk more about their infant’s suffering. The rating scale has four anchors (no discomfort, some discomfort, moderate discomfort, severe discomfort), evenly spaced across the horizontal line. Participants were asked to mark the line with an X and date their rating. The same scale, with prior ratings, was presented to the participant at each data collection time point so that participants could describe how their view of infant discomfort changed as the infant’s illness course progressed. The discomfort rating scale was developed by Drs. Docherty and Brandon (2008-2013, 1R01-NR010548, Docherty, PI) for an ongoing study exploring parental and provider decision-making for infants and children with complex life-threatening conditions.

5.2.3.5.1 Parental or HCP View of Hope

A 0 to 100 numeric rating scale was used to assess participant’s level of hope for the infant. Participants were asked to complete this hope rating scale at a point during
each interview when describing how they felt the infant was doing from a medical standpoint. Thus in addition to providing a quantification of their level of hope, the rating scale also served as an elicitation tool to encourage participants to talk more about their hopefulness for the infant. The rating scale had five anchors (no hope, hopeful, somewhat hope, very hopeful, extremely hope), evenly spaced across the horizontal line. Participants were asked to mark the line with an X and date their rating. The same scale, with prior ratings, was presented to the participants at each data collection time point so participants could describe how their view of hope changed as the infant’s illness course progressed. The hope rating scale was developed by Drs. Docherty and Brandon (2008-2013, 1R01-NR010548, Docherty, PI) for an ongoing study exploring parental and provider decision-making for infants and children with complex life-threatening conditions.

5.2.3.6 HCP Interviews

Narrative interviews were conducted with HCPs (physician and nurse) at study entry, following any life-threatening event/significant change in treatment, and following hospital discharge. Interviews were digitally recorded. Interviews focused the HCP’s story of treating the infant and the decisions made for the infant with HIE. Additional foci included the HCP’s perception of parental understanding of the infant’s condition, the way decisions were made, the HCP’s feelings about the decisions, and factors that influenced the HCP’s decisions. HCP were interviewed individually.
Interviews lasted an average of 15 minutes (range 5-37 minutes). See Table 10 for sample interview questions posed to HCP.

5.2.4 Data Collection Procedures

All data were collected by the first author. New admissions of infants to the NICU were screened 3 times per week. Data collection began within 1 week of study enrollment (enrolled within 1 week of life) and data collection measures continued monthly until the infant was 6 months of age (see Table 11 for specific data collection time points). Interview data were collected at a private location chosen by the parents and HCPs or via telephone.
Table 11: Data Collection Time Points

<table>
<thead>
<tr>
<th></th>
<th>Infant Inpatient</th>
<th>Outpatient</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>SE</td>
<td>LTE</td>
</tr>
<tr>
<td>Parent Interviews</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>HCP Interviews</td>
<td>X</td>
<td>X</td>
</tr>
<tr>
<td>Demographic Data (parent, HCP, infant)*</td>
<td>X</td>
<td></td>
</tr>
<tr>
<td>Infant medical record review</td>
<td>3x/week</td>
<td></td>
</tr>
<tr>
<td>Technology Dependence Scale</td>
<td>Weekly</td>
<td></td>
</tr>
<tr>
<td>Infant Status Checks</td>
<td>Daily</td>
<td></td>
</tr>
</tbody>
</table>

SE = study entry; LTE = life threatening event; * HCP demographic data was obtained from each HCP being interviewed

5.2.5 Data Preparation

The digitally recorded interviews were transcribed verbatim. Each written transcript was reviewed to ensure congruence with the digitally recorded interview. Each participant was given a pseudonym to ensure confidentiality. The key linking of the pseudonym with the participants was kept locked in a file cabinet separate from the transcribed interviews.

5.2.6 Data Analysis

The overall goal of the analysis was to explore how parent and HCP decision-making trajectories develop over time. The over-arching method of analysis for this multiple case study was comparison of displays of data trajectories and the visual search for patterns or trends in the trajectories to gain a deep understanding of similar and variant patterns of decision-making over the first 6 months of life for infants with HIE.
Cases were each examined as a separate case study and then compared across cases of infants with HIE to gain a better understanding of the typical infant illness trajectories and decision-making trajectories of parents and HCP (Stake, 2006).

In the first step of data transformation, daily infant illness and treatment data from the medical chart reviews was used to classify each day of the infant’s life into one of five treatment categories: initiation of treatment, experimental interventions, maintenance of treatment, escalation of treatment, or withdrawing/withholding treatment. The characterization of each day of the infant’s life into one of these categories allowed visual display of the types of treatment the infant needed throughout her illness course. This also allowed for the visual display of the infant’s illness trajectory using these categories as a context in which all of the illness and decision events occurred. The visual display was used to understand how the treatment categories interacted with other attributes such as themes from the interview data. Table 12 provides the definitions for each decision type and examples from the illness trajectory of infants with HIE.

Conventional content analysis was used to analyze the narrative interview data, which allowed for identification of themes and related patterns (Hsieh & Shannon, 2005). The data were approached systematically, extracting the main storyline to identify key events in the infant’s illness course and the associated decision-making trajectory. Then individuals involved in specific decisions were identified (Sandelowski, 1995). Factors that impacted decision-making were then grouped together under the broad categories such as ‘influences’, ‘communication’, ‘hope’, and ‘conflict’. Definitions for categories were developed from the data and refined as the analysis progressed (Hsieh & Shannon, 2005). Once data were separated into broad categories, subcategories emerged allowing similar ideas to be clustered.
together. Each subcategory was also defined and clarified as the analysis continued. Data analysis was concurrent with data collection to inform future data collection.
**Table 12: Treatment Categories**

<table>
<thead>
<tr>
<th>Treatment Categories</th>
<th>Definition</th>
<th>Examples</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Curative Care</strong></td>
<td>Care focused on curing disease or illness</td>
<td></td>
</tr>
<tr>
<td>Initiation of treatment</td>
<td>Decisions about the treatment plan based on the initial diagnoses or new diagnoses</td>
<td>• Cardiopulmonary resuscitation at delivery</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Respiratory support at or near delivery</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Moderate hypothermia &lt; 6 hours</td>
</tr>
<tr>
<td>Experimental intervention</td>
<td>Decisions about devices or medications for clinical trials</td>
<td>• Moderate hypothermia &gt; 6 hours</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Umbilical cord transplant</td>
</tr>
<tr>
<td>Escalation of treatment</td>
<td>Decisions about addition of medical treatments and technologies in an attempt to treat the illness or condition</td>
<td>• ECMO</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Gastrostomy tube placement</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Addition of more than monotherapy for seizure management</td>
</tr>
<tr>
<td>Maintenance of treatment</td>
<td>Decisions about whether and how to maintain the level of care</td>
<td>• Gastrostomy tube feedings</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Management of ventilator setting</td>
</tr>
<tr>
<td><strong>Palliative Care</strong></td>
<td>Care focusing on noncurative treatments to relieve suffering</td>
<td></td>
</tr>
<tr>
<td>Withdrawing/withholding treatment</td>
<td>Decisions about withdrawing or withholding life-sustaining treatment</td>
<td>• Withdrawal of mechanical ventilation</td>
</tr>
</tbody>
</table>

The self-report instruments provided additional information about influences in the decision-making trajectory. Scores from each instrument were graphed to identify patterns. Then descriptive statistical analyses were conducted for each instrument to identify patterns over time.
The last step in the analysis was to develop the decision-making trajectory displays to search for patterns and describe the relationship between decisions that were made and the influential factors. The background of each display was the infant illness trajectory with the treatment categories and discrete events (e.g., gastrostomy tube surgery) that altered the illness course. The decision-making trajectory was constructed from the medical chart data and interview data to include decisions by parents and HCPs. A decision was defined as a choice between two or more options. The decision-making trajectory was visually represented by short textual descriptions chronologically as the decision was made.

Visual trajectories of infant illness and decision-making data from each case were explored to identify common patterns shared across cases (Stake, 2006). Infant illness trajectories from each single case were displayed together on a plain board and then clustered into groups based on length of hospitalization and severity of HIE for infant illness trajectories. Decision-making trajectories from each single case were grouped together on demographic characteristics (e.g., all fathers, all mothers) and other combinations with the goal of determining how the trajectories were similar and different. Typical sample infant illness trajectories were identified and typical sample parent and HCP decision-making trajectories were also identified during the comparisons across each single case. The themes and subthemes from single cases were also searched to identify how themes and subthemes changed over the decision-making
trajectory for parents and HCP. Definitions of themes and subthemes between single cases were compared to determine if participants explained a similar concept, but were impacted differently by the concept. Themes and subthemes were then displayed over the sample infant illness trajectories and sample decision-making trajectories to determine how decision-making trajectories develop over the course of the illness trajectory for infants with HIE.

5.3 Results

The findings are presented beginning with a description of the infants’ illness trajectories that were commonly observed over the first 6 months of life. Then parent and HCP decision-making about treatments for infants with HIE across the illness trajectory were explored and the predicted decision-making trajectory was revised. Next the individuals who were involved in the decision-making process are presented. Finally, the factors that influenced the key decisions were explained by treatment type.

5.3.1 Infant Illness Trajectories

Across the 10 cases in this study, two distinct types of infant illness trajectories were observed over the first 6-month of life of infants with HIE. These typologies included infants (n = 4) who experienced complex illness complications and infants (n = 6) who experienced few illness complications during the initial 6-month period. See Table 10 for the illness trajectory of infants with complex illness complications and see Table 11 for the illness trajectory of infants with few complications.
The infants in both typologies required respiratory or cardiopulmonary resuscitation at birth, but the infants with complex illness complications required hospitalization greater than 40 days of life compared to infants with few illness complications who were hospitalized for 12-14 days. Infants with complex illness
complications were generally hospitalized 42-62 days. The majority of infants (n = 7) had seizure activity during the hospitalization period. The first 6 months of follow-up for infants appeared to be dependent on the complications identified during the hospitalization period.

A typical illness trajectory of infants with complex illness complications throughout the hospitalization period is displayed in Figure 10. Three of the four infants had severe HIE upon admission to the hospital. These infants had cardiopulmonary arrest at delivery, demonstrated signs of multisystem organ damage on admission to the hospital, experienced clinical or electrographic seizure activity, and required intensive medical treatments for hemodynamic stability over the first 1-2 weeks of life. Once infants were stable and were able to tolerate additional examination, delayed motor skills and difficulty establishing a nutritive pattern to obtain adequate nutrition by mouth were common. Follow-up for infants in this typology consisted of 5 to 9 visits to specialty clinics, in addition to primary care pediatrician visits, during the first 6 months of life.

Figure 11 displays a typical infant with few illness complications from birth to hospital discharge. Most of these infants were admitted with moderate HIE. They were usually stabilized within the first several days of hospitalization; several had seizure activity (n = 4), and required few days of respiratory support. Two infants received experimental moderate hypothermia intervention at > 6 hours of life and two infants
received umbilical cord blood transplants. Some infants displayed mildly, delayed motor skills during the hospitalization. The infants were able to quickly tolerate feedings by mouth and were discharged home within 2 weeks of life. Follow-up in specialty clinics during the first 6 months of life was between 2 to 4 visits.

5.3.2 Decision-Making Trajectories

All infants had a normal 9-month fetal gestational period and parents were encouraged by ultrasound reports of a healthy developing fetus. However, at birth the infants experienced unexpected complication that might affect their long-term development. The major treatment decisions made over the course of the 6-month study period were aimed at initial stabilization within the first week of life and the long-term treatments necessary to optimize long-term outcomes by preventing complications and treating comorbidities.

The decision-making trajectory was dependent upon the infant’s illness trajectory. Infants with complex illness complications required more treatments than infants with few illness complications. The underlying decision-making trajectory was dependent upon infant complications. Figure 12 describes the decisions that were made within the first week of the infant’s illness trajectory. Aggressive care included cardiovascular support, supplemental oxygen, supplemental nutrition, and moderate hypothermia treatment at < 6 hours of age. Experimental care included the delivery of experimental interventions: moderate hypothermia > 6 hours of age or umbilical cord
stem cell transplant. Successful was defined as survival or achievement of necessary goals. Figure 13 depicts the decisions points in the management of long-term treatments. These figures represent the overall decision-making trajectory identified by combining both parental and HCP decisions.

Figure 12: Observed Decision-Making Trajectory within First Week of Life
5.3.2.1 Parental and HCP differences in Participation in Decision-Making Trajectories

The parental and HCP participation in the decision-making trajectory for infants with HIE was distinctly different. HCPs reported involvement in all treatment decisions across the first week of life and the long-term treatments; expect for the length of neonatal resuscitation when the infants were born at an outlying hospital. However, parental participation in the decision-making trajectory in the first week of life week only occurred when experimental care was an option. Parents reported collaborating with HCPs after the infant was stabilized and the long-term treatments were implemented.
5.3.2.2 Decision-Making in the First Week of Life

All infants experienced physiologic instability at birth. Parents reported no participation in decision-making, even when the infant had no heart rate after 10 minutes of resuscitation. Parents were not consulted at any point during intubation and CPR, they were informed after the event. The father of an infant with severe HIE explained his lack of participation in the decision for resuscitation over 10 minutes:

They had just started and they just put me in a room and I sat there for a good hour before they even came in and said anything; so nobody consulted me on anything, they just came and updated me after it happened, so they were just telling me what was going on.

Once the infants were successfully resuscitated, transfer to the Level IV NICU at the study hospital or transfer to the in-house NICU of the study hospital was initiated for intensive monitoring and treatment. Parents of infants who received aggressive care reported no involvement in the decision to transfer their infant to the study hospital. Mothers ‘barely’ remembered being ‘told’ by birth hospital HCP that their infant was being transferred to the study hospital for aggressive treatments. Parents did report that they thought it was possible to decline the offer to transfer the infant to the study hospital, but they felt HCPs made the decisions. HCP initiated aggressive care including seizure medication, antibiotics, vasopressor medications, and moderate hypothermia treatment. HCPs reported basing their decisions about aggressive care on protocols and hospital policies. HCPs explained parental participation in decisions about medications administered “we haven’t asked if we can give them a specific medicine, but have
discussed it with them [parents].” Thus the majority of the decisions involved within the first week of life were made by the HCPs when aggressive care was provided.

When aggressive care and experimental care were options, parents and HCPs were involved in the decision-making. Four of the infants received experimental interventions with two receiving moderate hypothermia > 6 hours of age and two receiving umbilical cord stem cell transplant. These parents reported both involvement to transfer the infant to the study hospital and the decision to proceed with experimental interventions. The mother of an infant with moderate HIE explained her involvement in the decision-making to transfer the infant and participate in the experimental moderate hypothermia intervention > 6 hours of age:

I was originally at [birth hospital]. They induced my labor at 39 weeks. She was born with the cord wrapped around her neck tightly. And she also ended up having seizures. I was approached about doing the cooling study for her. Unfortunately they couldn’t do it at [birth hospital]...because they don’t go beyond 6 hours for the cooling...so I was asked if I wanted to transfer to [study hospital] and we decided that it would be a good study.

Though HCPs were concerned that parents may not understand the implications of participating in clinical research due to emotional shock they were experiencing:

A challenge as a clinician and a researcher and a parent to feel you is giving them so much information because you want them to know it all at once. You need to do the study to find out if the intervention helps. It’s hard to know where to allow them an out and say ‘yeah okay you can’t cope with this. I can see that you’re overwhelmed’ rather than being more insistent that they stay and they really listen.
5.3.2.3 Decision-Making after the First Week of Life

Parents and HCPs reported participating in decision-making after the first week of life. Some infants who experienced more severe complications from the hypoxic-ischemic event needed additional medical support, such as speech therapy and placement of a gastrostomy tube. Parents, in conjunction with HCP, participated in decisions about how to reach milestones required for discharge from the hospital including adequate ventilation and nutrition. Infants were given what parents and HCP agreed was an adequate amount of time to demonstrate progress towards set goals (e.g., full feeds). If the infant was not progressing, the decision to escalate treatment and place a gastrostomy tube was necessary. A physician described the reason behind his recommendation to escalation treatment for an infant with severe HIE:

It was clear the prognosis was going to be so slow. In the short term, he needed a feeding tube. The other issue was that he had terrible gastroesophageal reflux (GER) for a term baby. We were feeding him in ways we usually feed tiny preemies, which means we were running the food in very slowly over a couple of hours and it was impossible to really advance the volume and calories to a point where he could really grow and so it was pretty clear needed a gastrostomy tube, to keep his food down and grow.

Parents and HCPs also collaboratively made decisions about treatments such as therapies (e.g., physical, speech), follow-up clinic visits, and daily medication administration. Parents chose to keep appointments with specialty clinics and therapy because they thought that examination by HCP was essential to identifying potential problems quickly. Parents of infants, who were eligible for therapies based on the severity or potential disabilities, were eager to have their infant start care as soon as
possible. A mother of an infant with severe HIE explained: “I’ve decided to get her the physical and occupational therapy. I want the best for her and I want them to, if she was to have delays, I want them to be able to see it and help her get past those delay.”

5.3.2.4 Parental Differences in Attributes

Overall, the parental level of participation in the decision-making trajectory was divergent only when participation in aggressive care was compared to aggressive care and experimental care. Meaning that parents of infants with HIE who received aggressive care did not report being involved in the decision to initiate aggressive care. Parents of infants with HIE who received aggressive and experimental care reported being involved in the decision-making. To better understand the affect of parental participation in decision-making within the first week of life for infants receiving aggressive care compared to infants receiving aggressive and experimental care, parental characteristics were compared between the groups over the first 2 months of life.
Figure 14: Infant Illness Severity and Parental Hope by Group

Infants who received aggressive care and experimental care (known as experimental care) all had moderate HIE and had a lower illness severity and dependence on technology measured by the TDS compared infants who received aggressive care. However, as shown in Figure 14, parents of infants in the experimental care had lower hope than parents in the aggressive care from birth to 1 month. Parents of infants in the experimental group were less hopeful, despite their infants having lower severity of illness scores. Figure 15, also demonstrates that parents of infants who received experimental care also reported less stress than parents of infants who received aggressive care through the first 2 months of life.
5.4 Discussion

The predicted decision-making trajectory for infants with HIE was not supported in this study. The decision-making trajectory for infants with HIE was dependent upon the extent to which life-sustaining treatments were initiated during the first week of life; as well as, the infant’s response to these treatments. Parents were not involved in decision-making about neonatal resuscitation. HCP made the decisions and were guided by professional guidelines and hospital protocols (Kattwinkel et al., 2010). The guidelines and protocols appear to leave few actual ‘decisions’ for HCP and the inclusion of parents in the decision-making. In fact, throughout the first week of life the decision-making trajectory appear to offer fewer options and less opportunity for parents and HCPs to transition from aggressive care to palliative care. This finding could be related to infants with extensive, severe HIE not being transferred to the study hospital. However, four infants in the sample did have complex illness complications.
from the hypoxic-ischemic event. Parents were part of decisions about when to place gastrostomy tubes, how to participate in therapy, and administration of daily medications.

Most parents of infants who received aggressive care reported no participation in decision-making in the first week of life. Only parents who made decisions about experimental care were involved in the decision-making in the first week of life. Parents’ of infants receiving experimental care may have viewed themselves as more involved because they were required to sign consent for their infant to participate in the clinical trials. The participation in decision-making for experimental care appeared to decrease parental stress while the infant was hospitalized (PSS:IH) and the first 2 months of life (IES-R). The decreased parental stress could be that parents were given more information about HIE and the overall prognosis because HCPs had to spend time discussing the why the infant qualified for the clinical study and the risks and benefits of the study. The additional information may also explain why parents were less hopeful throughout the first month of the infant’s life, even though the infant’s were less illness compared to infants who received aggressive care only. However, further exploration as to why participation in experimental care led parents to view they participated in care is necessary. Signing the consent forms and being directly asked by HCPs could have led parent to be more involved, but what led them to be more involved remains unknown.
After the first week of life, most parents and HCP were simply left with making decisions in response to how the infant responded to the initial life-sustaining treatments. Some infants were unable to sustain adequate nutrition by mouth despite work with speech therapy. Thus the gastrostomy tube was needed because speech therapy was unable to help the infant feed enough by mouth. The escalation in care was more related to how long to wait for placement of the gastrostomy tube. Not placing a gastrostomy tube in an infant who is awake and maintaining respiration without assistance is not generally acceptable practice. Previous research showed that parents were involved in the decision-making for placement of a gastrostomy tube for their infant (Brotherton et al., 2007). Parents reported being influenced by infant suffering, wanting to make the best choice, and trust in HCP (Guerriere et al., 2003). Parents of infants with HIE did not report trust as a factor influencing their decision to proceed with placement of a gastrostomy tube or infant suffering. Parents from this study were focused on the infant’s need for the gastrostomy tube based on the infant’s response to feeding.

Decision-making about how to maintain treatments was clustered around hospital discharge. Determining whether infants needed therapy was generally identified before the infant was discharged from the hospital. Follow-up care was also arranged prior to discharge. Thus most of the parental and HCP decision-making for
infants with HIE from birth to 6 months of age is confined to the initial hospitalization period with intensive decision-making in the first week of life.

5.4.1 Future Implications for Research

Parent and HCP decision-making for infants with HIE must be explored as a trajectory that begins with birth. Each decision made by HCPs during delivery and the first several hours of life impacted the extent of treatments needed to escalate and/or maintain treatment for these infants. Particularly, parents of infants with a prolonged illness severity typology are forever dependent upon the extent to which life-sustaining treatments were initiated. How no parental involvement in decisions to initiate life-sustaining treatments for these infants impacts long-term treatment decisions remains unknown. With rates of death and severe disability still 25-35% for infants by age 2 years treated with moderate hypothermia (Jacobs et al., 2011; Shankaran, Pappas, et al., 2012; Simbruner et al., 2010), how parents and HCP make decisions about escalating treatments during subsequent critical care admissions and determine how to best treat the individual infants remains unknown. Further exploration of infant illness trajectories after the first 6 months of life and decision-making trajectories is necessary to understand how parents and HCP negotiate the decision-making roles, the relationship between parents and HCP, and what impacts future decisions for parents and HCP.
5.4.2 Implications for Practice

Parents of infants with HIE have little to no part in making decisions about initiating life-sustaining treatments. In addition to the shock parents experience with an unexpected complication at birth, infants with minimal illness severity within the course of the hospitalization are discharged home with parents within 2 weeks of life. The parents and HCP have limited time to discuss what HIE is, how it happened, treatments provided, and the complex long-term outcomes associated with HIE. Many of the parents of infants with minimal illness severity were cautious about the treatments and care the infants received within the first 1-2 months after discharge, but quickly began to think of their infant as ‘normal’ and stopped participation in physical therapy exercises and regimented adherence to medication administration. Teaching parents how to identify if their infants are progressing developmentally appropriate ways and how to determine when additional health care is needed is critical to optimizing long-term infant development. Early identification of developmental delays can help infants obtain optimal outcomes.
6. Conclusion

Decision-making for infants with complex life-threatening conditions (CLTC) begins at diagnosis and continues throughout the infant’s life because of associated chronic complications. The decision-making trajectory for parents and HCP can begin prenatal or postnatal depending on the condition with short periods of intensive decision-making about initiating life-sustaining or experimental interventions and long periods of treatments being maintained to promote optimal infant outcomes. Intermixed with these are more discrete periods of time in which treatments are escalated due to an acute change in the condition. The points, in which, treatments are escalated may occur gradually over time with parents slowly altering their view of the condition or may occur suddenly without warning. How each of decision point is managed will impact how future decisions are made and developmental outcomes of children.

Most decisions have been explored as single points within an infant’s illness trajectory. However, the infant’s illness trajectory evolves over time, along with the types of treatments necessary, and the experiences of the parents and providers. Without exploring the infant illness trajectory as a whole taking into account the previous decisions, influences on the decisions, and primary decision maker, researchers cannot understand how initial decisions impact all subsequent decisions within the parent and provider decision-making trajectories.
To conduct research that explores critical times within an infant’s illness and emotive situations for parents to better understand parental decision-making and determine how to best guide parents through the decisions, prospective longitudinal designs are necessary. How the ethical principles of respect for persons and beneficence of conducting research on parents who are very vulnerable and under intense stress are concerns of researchers, clinicians, IRBs, and other funding agencies when attempting to conduct research on sensitive topics. To better understand how parents responded to participation in interviews during highly emotive times, analysis of interview data from an ongoing study on parents and provider decision-making for infants with CTLC was conducted. Forty-nine parents of 29 infants with complex congenital heart disease (n = 9), extreme prematurity (n = 11), and metabolic disease requiring stem cell transplant (n = 9) participated in the study and completed a total of 364 interviews. Findings from this study suggest that research team members with health care backgrounds, intensive training and implementation of several protocols (e.g., suicide screening) can ethically conduct interviews during highly emotive times without harming the participants. In fact, parents found participation in the study to be helpful. The results from this analysis provided direction and guidance about how to design and implement a study while protecting and respecting parents during a highly emotive period when their full-term infant sustained a hypoxic-ischemic event at birth. To understand how the decision-making trajectory for infants with HIE unfolds, close attention to periods when decision-
making could occur was built into the exploratory design of this study. Because of the intensive data collection that was proposed for this study, knowing how to respect individuals and protect them was critical for ensuring ethical treatment of the participants.

Viewing parent and HCPs decision-making as a trajectory that develops over time is based on the theoretical perspective of developmental science (Magnusson & Cairns, 1996). In the case of infants with HIE, immediate decisions are needed. HCPs may have to make the decisions without input from the parents on what they want. HCPs may use their clinical experience to determine at that point in time what is ‘best’ for the infant. These immediate decisions are usually associated with the initiation of life-sustaining treatments (e.g., endotracheal intubation) and proceeding with aggressive care or aggressive care and experimental care. Finding from this study suggested that decision-making trajectory for the majority of infants with HIE could be viewed as decision-making within the first week of life and decision-making after the first week of life. The decision-making within the first week of life was more linear than predicted, Figure 16 demonstrates the decision-making trajectory observed during the first week of life. Parents were not involved in the decision to resuscitate the infant or how long to continue resuscitation. Parental input was minimal unless the infant was eligible for experimental interventions. If infants qualified for experimental interventions, parents reported participating in the decision to transfer the infant to a Level IV NICU for
potential participation in the experimental intervention.

Figure 16: Observed Decision-Making Trajectory within First Week of Life

Once the infant was stabilized after the first week of life, the decision-making process involved both parents and HCPs. Figure 17 depicts the decisions that the majority of the parents and HCPs encountered. Interestingly, most decision-making occurred prior to hospital discharge and few decisions were made once the infant was discharged unless a minor complication arose the decision-making trajectory remained unchanged.
The final aim of the dissertation was to explore the trajectory of parent and health care provider decision making for infants with HIE, across the immediate trajectory (first week of life) and short term trajectory (next 6 months). Eleven cases of infants with HIE were recruited from the intensive care units at Duke University Hospital. A total of 11 infants, 19 parents, and 39 HCP were enrolled in this prospective, longitudinal, case study. The overall findings suggest that parent and HCP decision-making begins at birth and continues at least through the first 6 months of the infant’s life. The first decisions about the extent of life-sustaining treatments to initiate affect the infant and their parents forever, especially infants with prolonged illness severity. HCP decision-making trajectory for a typical infant with complex complications began at birth and continued through the first 6 months of the infant’s life. However, despite the
extensive impact that the initiation of life-sustaining treatments can have for these infants, parents were not part of the decision-making initially. Parents were only involved in the decisions about when to escalate and how to maintain the treatments for infants with complex complications. These decisions often did not allow the parents to determine if the quality of life was acceptable for their infant at that point because the infant was stabilized and parents simply needed to decide when to place a gastrostomy tube and how to optimally provide necessary follow-up treatments and therapy.

A single case of an infant with both an expected congenital heart defect and HIE provided insight into the potential negative effect with parents are not involved in the decisions to initiate life-sustaining treatments. The parents were concerned about how the lack of oxygen during the hypoxic event would impact the infant’s long-term neurological status. The uncertain neurological status and divergent views of hope for the infant having a ‘normal’ life created conflict between parents and providers. This conflict led to parents mistrusting HCP and feeling that only they could make decisions that were best for their infant. Both parents became vigilant and on high alert at any sign of pain, discomfort, or change in the infant’s status or treatments. Ultimately, the relationship between parents and HCP diminished. When the parents were finally allowed to withdraw life-sustaining treatments, they did immediately. Unfortunately, at 8 weeks following the death of their daughter, the parents still blamed themselves for making ‘bad’ decisions and allowing their daughter to suffer for 5 months. Parents
regretted not being involved in the first decisions to initiate life-sustaining treatment for their daughter who died that they were going to seek legal council to ensure any future infant they delivered would never have the drastic life-sustaining treatments initiated.

Overall, the parental decision-making trajectory for infants with HIE and no additional complications was limited to decisions to participate in experimental treatments and how long to wait for placement of a gastrostomy tube. Parental participation in decisions to participate in experimental treatments appeared to decrease stress over the first 2 months, even though parents were less hopeful. The reason for this relationship is unknown, but warrants further investigation. The parents may have a better understanding of HIE initially because HCPs spend time explaining the injury and the affect the experimental intervention may have and the associated risk. The other decisions about therapy and medication administration were made by HCPs in conjunction with parents; however, parents of infants without complex complications often did not understand the need for continuing therapy and the importance of not discontinuing seizure medications. The main barrier to this appeared to be lack of understanding that the extent of the injury to the brain may take longer than the first several months to manifest all complications. Continued follow-up with these parents and infants is necessary to learn if the initial limited involvement in decision-making has a negative impact on the infants or parents. In the single case of Alexandria, the parents quickly realized how the hypoxic-ischemic event could impact the neurological status of
the infant in the long-term. This became a source of conflict and parents regretted not being involved in the decision to resuscitate the infant. The conflicting view of the neurological impact HIE could have, led parents to mistrust of HCPs. While differences in predicted neurological outcomes were known to cause conflict about withdrawing support, usually the HCPs were less hopeful about the potential for recovery. By evaluating the decision-making trajectory over time, factors leading to conflict and changing hope were identified, along with the negative affect the decision-making had on the parents.
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Biography

Kimberly Allen was born in Augusta, GA on April 16, 1983. She attended University of South Carolina-Aiken earning an associate (May 2003) and bachelor degree (May 2005). She continued her education at Emory University earning a master degree in nursing in August 2007.

As a graduate student at Duke University, she received two grants from the National Institutes of Health, an individual national research service award (NRSA) as a predoctoral fellow and a clinical and translational science award (CTSA). She also received grant funding through the American Nursing Foundation. She also published three articles in peer-reviewed journals:


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