PROJECT POPPYSEED: A MIXED-METHODS EXPLORATION OF PARENTS’ LIVED EXPERIENCES WITH ANENCEPHALY

By

SHANDEIGH N. BERRY

A dissertation submitted in partial fulfillment of the requirements for the degree of

DOCTOR OF PHILOSOPHY

WASHINGTON STATE UNIVERSITY
College of Nursing

MAY 2020
To the Faculty of Washington State University:

The members of the Committee appointed to examine the dissertation of SHANDEIGH N. BERRY find it satisfactory and recommend that it be accepted.

Gail Oneal, Ph.D., Chair

Andra Davis, Ph.D.

Lonnie Nelson, Ph.D.

Billie Severtsen, Ph.D.
ACKNOWLEDGMENT

I would like to extend my deepest gratitude to all who make this achievement possible. To Dr. Oneal, whose patient guidance grounded me, to Dr. Severtsen, who taught me so much about philosophy and thinking deeply. To Dr. Davis, whose transparent mentorship inspires me, and to Dr. Nelson, who showed me tools to emotionally navigating the exploration of perinatal bereavement. I would also like to thank Dr. Haberman, my most ardent cheer leader. Thank you to Dr. Fritz, who has gone above and beyond in being a boss, a mentor, a guide, and a friend. My gratitude is extended to those behind the scenes as well. To Electra Enslow, the most attentive librarian, and Sarah Wilson and Rychelle Wagner, for keeping track of the regulations and requirements. And thank you to the unsung heroes of the IT world. Mike Lynch, you made my journey along the road of hybrid learning possible.

I would like to thank those not of WSU who supported me along my way. Jake, my patient and loving partner, thank you for sticking by my side. Thank you for not giving up on me, thank you for having deep, never ending faith in my brain, my abilities, and my potential. Crystal, you’re the best. Never will I ever have another friend like you. Grandpa Ric, you’re one in a million, you’re good people. It is a privilege to have each of you in my life.

I would like to thank little Ayden, whose brief time with us inspired so many. I am grateful to my sister and her husband, for showing me the power of intentionality. I would also like to thank the families who came forward to participate in this study. I was absolutely humbled by the overwhelming response of those who have had a pregnancy complicated by anencephaly. It is an honor to listen to each story, and I am amazed and encouraged by the strength and resilience of each of you. I am grateful to have had this experience. It is
encouraging to know that from profound heartbreak can come strength, resilience, compassion and love.

To those I have mentioned, to those I have not, thank you for your part in my journey.

This research was supported by a Sigma Theta Tau research grant, the Carl M. Hansen Foundation, the Linbald Foundation, and the Karen Krinshaw award.
Objective: One in four women who experience perinatal loss develop intense grief. Adverse outcomes of intense grief include depression, anxiety, posttraumatic stress disorder, substance abuse, suicidal ideation, obesity, hypertension, and diabetes. Congenital anomalies such as anencephaly are the leading cause of perinatal mortality in the United States. The purpose of this study was to identify the impact of common experiential themes on perinatal grief intensity by exploring parents’ lived experiences with a pregnancy complicated by anencephaly.

Methods: This study used a convergent parallel mixed methods design. Twenty women and four men between 18-59 years old with a history of an anencephalic pregnancy participated. Participants completed one open-ended, individual interview, a demographic questionnaire and the Perinatal Grief Intensity Scale. Heidegger’s philosophy of an ontological death guided the analysis of interview transcripts in a Hermeneutic Circle. Qualitative findings were synthesized with overall and subscale Perinatal Grief Intensity Scale scores using a Pearson’s correlation coefficient.
Results: Overall, 75% of parents (men=50%, women=80%) scored intense grief. Total and subscale scores indicating specific care practices associated with decreased grief included control over care ($r=-.475$, $p=.019$), providing parents with treatment options ($r=-.484$, $p=0.17$), and supportive obstetric providers ($r=-.426$, $p=.038$), specialists ($r=-.437$, $p=.033$) and hospital staff ($r=-.445$, $p=.029$). Qualitative patterns included overwhelming trauma, patient-centeredness as critical, stigmatizing perinatal loss, embracing personhood, and reframing reality.

Conclusion: Congenital anomalies are commonly diagnosed during routine care yet transform the pregnancy experience. Receiving a diagnosis of anencephaly in utero is a traumatic experience, impacting parents for years after the completion of the pregnancy. Parents in this study were three times as likely to develop intense grief following their experience with anencephaly, indicating a critical need to improve screening practices, adopt a supportive, patient-centered approach which places parents in control over care decisions, and implement follow up care. It is critical for healthcare providers to recognize the traumatic and transformative experience of a pregnancy complicated by anencephaly on parents.
# TABLE OF CONTENTS

<table>
<thead>
<tr>
<th>ACKNOWLEDGMENTS</th>
<th>iii</th>
</tr>
</thead>
<tbody>
<tr>
<td>ABSTRACT</td>
<td>v</td>
</tr>
<tr>
<td>LIST OF TABLES</td>
<td>xi</td>
</tr>
</tbody>
</table>

## CHAPTERS

<table>
<thead>
<tr>
<th>CHAPTER I: INTRODUCTION</th>
<th>1</th>
</tr>
</thead>
<tbody>
<tr>
<td>Background and Significance</td>
<td>2</td>
</tr>
<tr>
<td>Problem Statement</td>
<td>5</td>
</tr>
<tr>
<td>Purpose Statement</td>
<td>6</td>
</tr>
<tr>
<td>Research Question</td>
<td>7</td>
</tr>
<tr>
<td>Specific Aims</td>
<td>7</td>
</tr>
<tr>
<td>Definition of Terms</td>
<td>8</td>
</tr>
<tr>
<td>Summary</td>
<td>9</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>CHAPTER II: LITERATURE REVIEW</th>
<th>11</th>
</tr>
</thead>
<tbody>
<tr>
<td>Anencephaly</td>
<td>11</td>
</tr>
<tr>
<td>Perinatal Loss, Grief and Bereavement</td>
<td>13</td>
</tr>
<tr>
<td>Perinatal Loss</td>
<td>14</td>
</tr>
<tr>
<td>Perinatal Grief</td>
<td>14</td>
</tr>
<tr>
<td>Perinatal Bereavement</td>
<td>16</td>
</tr>
</tbody>
</table>

<p>| Perinatal Grief Theory                                                           | 18  |
| Perinatal Grief Psychometrics                                                    | 20  |</p>
<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Perinatal Grief Intensity Scale</td>
<td>21</td>
</tr>
<tr>
<td>History and Development</td>
<td>22</td>
</tr>
<tr>
<td>Psychometrics (Reliability)</td>
<td>23</td>
</tr>
<tr>
<td>Perinatal (Validity)</td>
<td>23</td>
</tr>
<tr>
<td>Parental Experience with Fetal Anomaly Diagnoses</td>
<td>25</td>
</tr>
<tr>
<td>Perinatal Palliative Care</td>
<td>28</td>
</tr>
<tr>
<td>Gaps in the Knowledge</td>
<td>30</td>
</tr>
<tr>
<td>Summary</td>
<td>31</td>
</tr>
<tr>
<td>CHAPTER III: METHODOLOGY</td>
<td>33</td>
</tr>
<tr>
<td>Research Design</td>
<td>33</td>
</tr>
<tr>
<td>Qualitative Method: Hermeneutic Interpretive Phenomenology</td>
<td>36</td>
</tr>
<tr>
<td>Philosophical Foundation</td>
<td>37</td>
</tr>
<tr>
<td>Heideggerian Terminology</td>
<td>39</td>
</tr>
<tr>
<td>Heidegger on Death</td>
<td>40</td>
</tr>
<tr>
<td>Philosophical Foundation</td>
<td>37</td>
</tr>
<tr>
<td>Philosophical Interview Underpinnings</td>
<td>41</td>
</tr>
<tr>
<td>Application of Heideggerian Philosophy to Nursing Research</td>
<td>41</td>
</tr>
<tr>
<td>Appropriateness for this Research</td>
<td>37</td>
</tr>
<tr>
<td>Quantitative Method: Perinatal Grief Intensity Scale</td>
<td>45</td>
</tr>
<tr>
<td>Method</td>
<td>46</td>
</tr>
<tr>
<td>Disclosure of Researcher Bias</td>
<td>46</td>
</tr>
<tr>
<td>Sample and Recruitment</td>
<td>46</td>
</tr>
<tr>
<td>Data Collection</td>
<td>48</td>
</tr>
<tr>
<td>Section</td>
<td>Page</td>
</tr>
<tr>
<td>--------------------------------------------------------------</td>
<td>------</td>
</tr>
<tr>
<td>Interview Protocol</td>
<td>48</td>
</tr>
<tr>
<td>Data Analysis</td>
<td>49</td>
</tr>
<tr>
<td>Interview Transcripts</td>
<td>49</td>
</tr>
<tr>
<td>Perinatal Grief Intensity Scale</td>
<td>50</td>
</tr>
<tr>
<td>Demographic Questionnaire</td>
<td>51</td>
</tr>
<tr>
<td>Synthesized Findings</td>
<td>51</td>
</tr>
<tr>
<td>Evaluation of Rigor and Trustworthiness</td>
<td>52</td>
</tr>
<tr>
<td>Human Subjects Protection</td>
<td>53</td>
</tr>
<tr>
<td>Pilot Study</td>
<td>54</td>
</tr>
<tr>
<td>Summary</td>
<td>55</td>
</tr>
<tr>
<td><strong>CHAPTER IV: RESULTS</strong></td>
<td>56</td>
</tr>
<tr>
<td>Results</td>
<td>56</td>
</tr>
<tr>
<td>Respondents</td>
<td>57</td>
</tr>
<tr>
<td>Participant Demographics</td>
<td>58</td>
</tr>
<tr>
<td>Data Collection</td>
<td>60</td>
</tr>
<tr>
<td>Qualitative Findings</td>
<td>60</td>
</tr>
<tr>
<td>Overwhelming Trauma</td>
<td>61</td>
</tr>
<tr>
<td>Patient-Centeredness as Critical</td>
<td>56</td>
</tr>
<tr>
<td>Stigmatizing Perinatal Loss</td>
<td>71</td>
</tr>
<tr>
<td>Embracing Parenthood</td>
<td>75</td>
</tr>
<tr>
<td>Reframing Reality</td>
<td>79</td>
</tr>
<tr>
<td>Perinatal Grief Intensity Scale</td>
<td>83</td>
</tr>
<tr>
<td>Data Synthesis</td>
<td>84</td>
</tr>
</tbody>
</table>
LIST OF TABLES

TABLE 4.1: CHARACTERISTICS OF STUDY PARTICIPANTS ........................................58
TABLE 4.2: QUALITATIVE SUBTHEMES WITH VARIABLE INDICATORS .....................89
TABLE 5.1: PREGNANCY & LABOR CHARACTERISTICS ........................................98
Dedication

To those taken from us by anencephaly. You are loved.
CHAPTER I

Introduction

Perinatal loss, defined as the death of a fetus or neonate during the perinatal period (conception to the 28th day of life), affects 15%-25% of pregnancies in the United States (US) each year (Johnson & Langford, 2015). Congenital anomalies, the leading cause of infant death in the US, are responsible for approximately 20% of the annual infant mortality (Cole et al., 2017). Anencephaly, a uniformly lethal congenital anomaly in which the neural tube does not fully form by the 28th day after conception, affects 1 in 4,600 births in the US annually (CDC, 2017). Due to the discrepancies in reporting requirements and practices, it is difficult to accurately estimate the annual perinatal mortality in the US (Hoyert & Gregory, 2016). Furthermore, despite declining by 30% from 1990-2011, perinatal mortality plateaued from 2011-2013, and continued to remain unchanged between 2014-2016 (Gregory, Drake, & Martin, 2018). Currently, the prevalence of anencephaly in Washington state is nearly five times the national average, at 9.5 per 10,000 live births (Barron, 2016). Resulting perinatal loss places parents at risk for developing intense grief, which may persist for the remainder of parent’s lives (Hutti et al., 2017; L. J. Toedter, Lasker, & Alhadeff, 1988). Multiple studies have linked perinatal grief to long lasting adverse physical psychosocial outcomes, including obesity, hypertension, diabetes, cancer, depression, anxiety, obsessive compulsive disorder (OCD), posttraumatic stress disorder (PTSD), guilt, shame, substance abuse, marital conflict, disrupted parenting, and suicidal ideation (Calderon-margalit, Friedlander, & Deutsch, 2007; Due, Chiarolli, & Riggs, 2017; Hutti et al., 2018; Samuelsson, Radestad, & Segesten, 2001). Approximately 25% of women experiencing perinatal loss develop adverse psychosocial
outcomes, which, according to Healthy People 2020, is one of the leading causes of disability in the United States (Cole et al., 2017; Hutti et al., 2017; U.S. Department of Health and Human Services, Healthy People 2020, 2018).

Despite the nationally expanding patient population, uncertainty remains concerning how to appropriately care for the psychosocial needs of parents experiencing perinatal loss (Lisy et al., 2016). With the increasing prevalence of anencephalic pregnancies in the US and long-term impact of adverse psychosocial symptoms resulting from complicated grief following perinatal loss – such as depression, anxiety, and substance abuse – it is imperative to identify key experiential elements contributing to complicated grief. Therefore, the aim of this study was to explore the impact of the lived experience of a pregnancy complicated by anencephaly on parents. A mixed methods approach was utilized to accomplish the aim of the study. Using hermeneutic interpretive phenomenological informed by Heidegger’s philosophy of an ontological death, important and meaningful themes were identified, providing valuable insight into the unique needs of this population. These themes may inform individualized interventions to facilitate healthy grieving and subsequently promote healing. Additionally, grief intensity following perinatal loss was measured using the Perinatal Grief Intensity Scale (PGIS), identifying the intensity of the grieving response not only to each individual’s experience, but to common themes and patterns emerging from the collective experiences of the participants.

**Background and Significance**

Historically, the diagnosis of an anencephalic pregnancy did not occur until birth, at which time physical deformities could be observed. Today, however, anencephaly can be diagnosed as early as 11 weeks gestational age through ultrasound diagnostic technology (Bonhotal, 2015; Hall et al., 2016). Advancement of ultrasound diagnostic imaging may
contribute to complicated grief surrounding anencephaly, primarily relating to parental misconceptions of the purpose of the procedure. While ultrasound imaging is intended for assessing amniotic fluid volume and placental location, verifying gestational age, and screening for anomalies and multiple gestation, many parents believe the sole purpose to be determining the sex of the fetus (Greiner & Conklin, 2015). Furthermore, as many as 75% of congenital anomalies detected via ultrasound occur in otherwise healthy pregnancies (Carlsson & Mattsson, 2018; Greiner & Conklin, 2015; Xafis, Watkins, & Wilkinson, 2016). Communication of a fetal anomaly diagnosis meets the American Psychiatric Association (APA) criterion for trauma (Aite et al., 2011), and may develop into long-term symptoms of PTSD (Cole et al., 2017; Hutti et al., 2018; Johnson & Langford, 2015). In one study, nearly 90% of mothers and over 80% of fathers receiving a diagnosis of a congenital anomaly experienced the communication of the diagnosis as a traumatic event (Aite et al., 2011). Furthermore, the communicative stance, method of information delivery, and language providers use to inform parents of a life-limiting congenital anomaly detected in utero largely influences the extent of the emotional trauma parents experience (Berry & Colorafi, 2019). Parents, excited to discover the sex of the fetus, are rarely prepared to receive a diagnosis of a uniformly lethal congenital anomaly. During a single routine appointment, future dreams and possibilities are arrested upon receiving a terminal diagnosis (Côté-Arsenault & Denney-Koelsch, 2011a). Parents must face treatment decisions such as termination versus carrying the pregnancy to term, which place strain on parents and providers alike. Thus, the unique needs of this patient population require further exploration to guide and inform patient-centered care.

Literature illustrates a strong connection between patient-centered care in both general and acute practice settings with positive outcomes, including improved health status, increased
efficacy of care, and enhanced recovery (Weston & Jordan, 2016). As one of six core values established by the National Academy of Medicine (formerly named Institute of Medicine), patient-centered care is a lexicon of health policy (Epstein & Street, 2011; Weston & Jordan, 2016). Moving beyond disease etiology, patient-centered care seeks to understand the patient’s perspective of the illness, the impact on their daily life, and their expectations of care. Building provider-patient relationships which guide treatment decisions consistent with the patient’s values decreases anxiety and depression, while increasing well-being and quality of life (Epstein et al., 2010). Literature further suggests a patient’s perception of care influences biological processes, impacting healing and recovery (Weston & Jordan, 2016). Inversely, failure to provide patient-centered care can result in poor health status, increased utilization of healthcare services, and increased healthcare costs (Epstein & Street, 2011; Weston & Jordan, 2016). Therefore, it is necessary to translate concepts of patient-centered care to the care of parents experiencing perinatal loss.

There remains a significant gap in nursing literature exploring the experience and provision of bereavement resources to parents suffering from perinatal loss, irrespective of the etiology (Lisy et al., 2016). While knowledge concerning perinatal loss and grief has evolved significantly over the past 50 years, ambiguity remains for best practice regarding communication of the diagnosis of a uniformly lethal congenital anomaly to the parents (Berry & Colorafi, 2019). Such gaps in knowledge may exist due to the scant literature available investigating anencephaly, which has focused primarily on disease etiology. Furthermore, perinatal grief and bereavement research is centered predominantly around miscarriage (Czeizel & Dudas, 1992; Czeizel et al., 2013; Gong et al., 2016; Moore, 2010; Petrini et al., 2008; Prasoon et al., 2016; Prue, Hamner, & Flores, 2010), even though anencephaly rarely results in
spontaneous miscarriage, and over half of anencephalic neonates are born alive (Machado, Martinez, & Barini, 2012). What remains to be explored is the effect of perinatal grief experienced by parents receiving the terminal diagnosis of anencephaly. Therefore, it is critical to understand and incorporate patient perspectives derived from the experience of those enduring perinatal loss.

By gaining a better understanding of the meaning parents apply to their lived experience of an anencephalic pregnancy, important concepts in preventing symptoms of long-term adverse psychosocial outcomes may be uncovered. Exploring the impact of the experience of those who have received a diagnosis of a pregnancy complicated by anencephaly will offer unique perspectives, providing crucial insight necessary to fill this knowledge gap.

**Problem Statement**

As the prevalence of anencephaly rises, so too does the frequency of nursing encounters with parents carrying an anencephalic fetus. Nursing literature exploring perinatal loss indicates interactions between parents and healthcare professionals “can have significant and lasting impacts, either positive or negative, on parents” (Lisy et al., 2016, p. 7). While qualitative studies have explored common themes surrounding various congenital anomalies, to date, no studies have explored the experience of a pregnancy complicated by anencephaly from both the mother and fathers’ perspective through the lens of an ontological death. Furthermore, no guidelines or bereavement theories have been established regarding the care for parents experiencing perinatal loss (Brownlee & Oikonen, 2004; Fenstermacher & Hupcey, 2013; Peters et al., 2015). With so little known about a parent’s experience of anencephaly, healthcare professionals are frequently ill-equipped to effectively care for this patient population (Côté-Arsenault & Denney-Koelsch, 2011a).
Comparatively little research has focused on anencephaly, and there is a significant gap in the literature exploring the experience, perception, and difficulties, and psychological effects of perinatal loss from the perspective of the fathers (Carlsson & Mattsson, 2018). Thus far, exploration of the paternal role has been limited, focusing primarily on the father in a support role to the expectant mother. Lastly, while a few studies have explored the experiences of parents who have had a pregnancy affected by a uniformly lethal congenital anomaly, no studies have investigated common themes corresponding to intense grieving reactions. To provide holistic, patient-centered care, it is crucial to understand elements which potentiate intense grieving reactions. From the knowledge gained through exploring the lived experience of an anencephalic pregnancy, a holistic approach to appropriate care may be constructed, guiding healthcare professionals caring for those experiencing perinatal loss.

**Purpose Statement**

The purpose of this mixed methods study was to explore the impact of the lived experience of an anencephalic pregnancy on parents. Hermeneutic interpretive phenomenological guided by Heidegger’s philosophy of an ontological death guided qualitative data analysis. Quantitative data was gathered using the PGIS and a demographic questionnaire. Selection of this approach assisted in understanding the meaning parents applied to their experience, how they reconstructed meaning and identity in the “world collapse” following perinatal loss, as well as in identifying key themes which correlated to intense grieving reactions, thus informing patient-centered, individualized care. The long-term goal of this research is further study the identified important concepts and practices able to decrease adverse symptoms of psychosocial outcomes associated with intense, complicated perinatal grief. Such concepts may guide the development of future interventions and inform patient-centered care. The
primary goal of this study was to explore the impact of the lived experiences of an anencephalic pregnancy on parents. This was accomplished by using the PGIS to measure the intensity of grieving reactions surrounding perinatal loss and synthesizing the results with hermeneutically analyzed interview transcripts. The secondary goal was to raise important questions for future perinatal grief and bereavement research. Ultimately, a collaborative effort between participant and researcher may aid in the prevention of symptoms of adverse psychosocial outcomes by integrating patient perspectives into healthcare practice.

**Research Question**

Hermeneutic interpretive phenomenology explores the meaning of being within a lived experience. Heidegger postulated that following ontological death – brought about by an event which directly contrasts with an individual’s understanding of how the world operates, what is meaningful in the world, and what ‘should happen’ in light of these preconceived notions which are embedded in each individual’s culture – a new identity and understanding of what is personally meaningful emerges (Aho, 2016; Heidegger, 1927). This philosophy guided the exploration and interpretation of a parent’s experience of having an anencephalic pregnancy. Therefore, the research question for this study was: What is the impact of the lived experience of an anencephalic pregnancy on parents?

**Specific Aims**

1. Determine the primary lived experiential components of parents with a history of a pregnancy complicated by anencephaly using hermeneutic interpretive phenomenology.

2. Characterize residual perinatal grief using the Perinatal Grief Intensity Scale.
3. Identify key shared experiences linked with grief intensity to identify common themes associated with poor psychosocial outcomes, such as symptoms of depression, posttraumatic stress disorder (PTSD), and anxiety.

4. Identify opportunities for change in current practice which could improve the care of bereaved parents experiencing a pregnancy complicated by anencephaly.

Definition of Terms

- **Bereavement**: The state of having experiences a loss. This includes grief and mourning; that is, the inner feelings and outward reactions of the survivor.

- **Bereavement Period**: Referring to the time it takes to mourn, grieve, and adjust to a world without the physical, psychological, and social presence of the deceased. This is a highly individualized timeframe (Corless, 2015; Limbo & Davies, 2015).

- **End-of-Life Care**: One aspect of palliative care which supports a peaceful, dignified death for the infant and the provision of support to the family and healthcare providers (Aatlin et al., 2015).

- **Grief**: The emotional response to a loss. This is a personalized feeling and response which an individual experiences to real, perceived, or anticipated loss (Limbo & Davies, 2015).

- **Loss**: The experienced absence of a possession or person. The value of the loss is determined by and unique to the individual experiencing the loss (Limbo & Davies, 2015).

- **Mourning**: The outward, social expression of a loss. Cultural norms, customs, and practices, including rituals and traditions, dictate how one outwardly expresses a loss (Corless, 2015).
- **Neonatal Period**: The timeframe between birth up to 28 days of life (Tektas & Çam, 2017).

- **Perinatal Loss**: Death occurring at any time within the perinatal period, including spontaneous, therapeutic and elective abortions, stillbirths, and neonatal death (Brownlee & Oikonen, 2004; Tektas & Çam, 2017).

- **Perinatal Palliative Care**: An interdisciplinary specialty focused on improving a patient’s quality of life and may be offered concurrently with curative care to treat symptoms, minimize suffering, and offer improved quality of life (Aatlin et al., 2015).

- **Perinatal Period**: The timeframe between conception and birth (Tektas & Çam, 2017).

**Summary**

Anencephaly is a uniformly lethal congenital anomaly which significantly impacts parents for the remainder of their lives. With the increasing prevalence of anencephaly, healthcare professionals maintain responsibility for a patient population in which little guidance is provided through care standards. Interactions between healthcare professionals and parents experiencing perinatal loss frequently have lifelong implications, and can positively or negatively impact a parent’s experience. Both short- and long-term physical and psychosocial health and wellness are a primary concern for parents experiencing perinatal loss. Patient-centered care has been demonstrated to lead to positive patient outcomes, and interaction with healthcare professionals is highly influential on patient experience. Understanding the meaning parents apply to their situation surrounding perinatal loss provides valuable insight that can inform future guidelines for patient care. This mixed methods, phenomenological study is conducted to understand the impact of a pregnancy complicated by anencephaly, and thus to provide evidence for best practice surrounding perinatal loss. This study ultimately has the
potential of translating to the care of parents experiencing perinatal loss resulting from a wide range of pathologies.
CHAPTER II
LITERATURE REVIEW

The domains of knowledge framing this study include anencephaly, perinatal loss, grief and bereavement, parental experience with fetal anomaly diagnoses, and perinatal palliative care. Chapter two explores each of these domains in turn, providing evidence for the need to study parental experience concerning perinatal loss due to uniformly lethal congenital anomalies, specifically, anencephaly. Subsequently, gaps in the knowledge surrounding this phenomenon, which present opportunities for future research, are identified.

Anencephaly

Anencephaly is a uniformly lethal neural tube defect (NTD) in which the anterior neural tube does not fully form by the 28th day after conception (Hall et al., 2016; Jaquier, Klein, & Boltshauser, 2006; Moore, 2010). The cerebral hemispheres and cranial cap are either absent or severely reduced (Chapman, 2013). Associated facial deformities include bulging eyes, prominent ears and a shortened neck. Cranial structures superior to the orbits are absent to varying degrees. These structural deformities can be detected with ultrasound diagnostic technology as early as 11 weeks gestational age (Moore, 2010). All other organs and structures remain unaffected in pregnancies complicated solely by anencephaly (Cameron & Moran, 2006; Hall et al., 2016; Moore, 2010; Raineri, 2017). Serum alpha-fetoprotein screening, a fetal protein biomarker that can indicate neural-tube defects, is also used in early detection of NTDs, and demonstrates nearly 100% diagnostic accuracy (Jaquier et al., 2006; Moore, 2010).
Risk factors associated with anencephaly include folic acid deficiencies, prior pregnancies complicated by NTDs, genetics, obesity, diabetes, certain pharmacological agents such as diet pills, and dieting. Anencephalic pregnancies have also been linked with exposure to fumonisins, a toxin found in corn mold. This toxin negatively impacts neural tube development in early pregnancy, but is otherwise harmless to children and adults (Barron, 2016; Czeizel et al. 2013; Gong et al., 2016; Petrini et al., 2008; Prue, Hamner, & Flores, 2010). Other suspected risk factors include pesticides and ethnicity. Hispanic populations have largely been affected in clustered outbreaks of anencephaly, both in Central Washington in 2012 and Brownsville, Texas in 1991 (Barron, 2016). Additionally, the prevalence of anencephaly in Mexico is 15.23 per 10,000 births, substantially higher than the national United States (US), at 2.1 per 10,000 (Barron, 2016).

In the late 1980s and early 1990s, multiple randomized control trials noted prenatal folic acid supplementation decreased the risk of NTDs, including anencephaly, by 70-90% (Czeizel et al., 2013; Gong et al., 2016; Moore, 2010; Prue et al., 2010; Williams et al., 2002). In response, the Food and Drug Administration (FDA) recommended women of child bearing age consume a daily dose of 0.4mg of folic acid, and mandated folic acid supplementation of grain products in 1992 (Barron, 2016; Gong et al., 2016; Prue et al., 2010). Despite an initial reduction of NTDs following the initiative, the prevalence of anencephaly has increased over the last 15 years (Barron, 2016; Center for Disease Control and Prevention, 2017; Gong et al., 2016; Report, 2010; Williams, Kennedy, & Boss, 2015). Over 3,000 pregnancies are affected by anencephaly annually in the US. In Washington State, the prevalence of anencephaly is five times the national average (Barron, 2016; Center for Disease Control and Prevention, 2017; Washington State Department of Health, 2015).
With the increasing prevalence of parents carrying an anencephalic pregnancy, healthcare providers encounter an influx of parents requiring unique and specialized care. The complex emotional and psychosocial elements associated with terminal pregnancies necessitates a multidisciplinary approach among physicians, nurses, psychologists, social workers, and spiritual care specialists (Chapman, 2013; Fenstermacher & Hupcey, 2013; Lalor, Devane, & Begley, 2007). The grief parents experience during the bereavement period preceding and following perinatal loss does not follow traditional grief theories (Brownlee & Oikonen, 2004; Fenstermacher & Hupcey, 2013). Furthermore, no perinatal grief theory has been established to date, further complicating the provision of care (Brownlee & Oikonen, 2004; Fenstermacher & Hupcey, 2013). To better understand the complexities surrounding perinatal loss due to anencephaly, it is necessary to examine scientific understanding of perinatal grief and bereavement.

**Perinatal Loss, Grief and Bereavement**

Perinatal grief and bereavement following pregnancy loss are frequently used interchangeably in the literature. According to a principal-based concept analysis by Fenstermacher et al. (2013), perinatal grief and bereavement lack explicit definition in scientific literature, contributing to the ambiguity of the phenomenon. This further complicates advancement in both scientific knowledge and clinical practice surrounding perinatal loss, as both require a clear definition to distinguish the boundaries of perinatal grief and bereavement. With no clear theoretical definition, nursing practice and research depend on implied meaning dispersed throughout the literature for developing care plans and reporting outcomes measures for perinatal bereavement. Ultimately, the theoretical and clinical definition of this phenomenon
not only impacts the direction of future research, but the planning, implementation, and evaluation of care interventions as well (Fenstermacher & Hupcey, 2013; Hutti & Limbo, 2019).

**Perinatal loss.** Though the constructs of perinatal grief and bereavement remain undefined, significant advancements have been made in the past few decade concerning perinatal loss. As defined in chapter one, perinatal loss refers to unintended or involuntary pregnancy loss occurring at any time within the perinatal period, including spontaneous, therapeutic and elective abortions, stillbirths, and neonatal death (Brownlee & Oikonen, 2004; Fenstermacher & Hupcey, 2013; Robinson, Baker & Nackerund, 1999). Perinatal loss can be further categorized as “early loss (<20 weeks), stillbirth (>20 weeks gestation) or neonatal loss (newborn through 28 days of life)” (Fenstermacher & Hupcey, 2013, p. 2390). Additional losses are experienced with a terminal pregnancy, including the loss of future hopes and expectations, loss of a healthy pregnancy, and loss of the future parenting role (Côté-Arsenault & Denney-Koelsch, 2011a).

**Perinatal grief.** Grief is a highly personalized emotional response to a loss which may be real, perceived, or anticipated (Limbo & Davies, 2015). Grief is expressed through mourning, the outward, social expression of a loss. Cultural norms, customs, and practices, including rituals and traditions, dictate how one outwardly expresses a loss (Corless, 2015). Historically, perinatal grief was not acknowledged as a psychological construct or a meaningful loss until 1970, when Kennel et al. recognized perinatal grief as its own distinct entity, sparking a proliferation of research (Fenstermacher & Hupcey, 2013; Kennell et al., 1970; Toedter et al., 1988). In 1984, the Perinatal Loss Project began, investigating factors influencing the length and intensity of the grieving reaction following the loss of a neonate (Toedter et al., 1988). Investigative efforts found nearly 25% of women who experience perinatal loss develop adverse psychosocial outcomes, including posttraumatic stress disorder, depression, anxiety, substance abuse, and
suicidal ideation (Johnson & Langford, 2015; Ryninks et al., 2014; Trinidad & Wick 2017). In a cohort study investigating psychosocial outcomes in women following miscarriage, women in the miscarriage cohort were 2.5 times more likely to develop a major depressive disorder, and 5.2 times more likely to develop a minor depressive disorder (Klier, Geller, & Ritsher, 2002). Neugebauer et al. (1997) found women with previously diagnosed depressive disorders are 54% more likely to develop a major depressive disorder following perinatal loss (Klier et al., 2002). Additionally, in a study examining coping, posttraumatic growth, and perinatal grief intensity in individuals experiencing perinatal loss, intense grieving reactions were negatively correlated with adaptive coping and posttraumatic growth, illustrating the significant impact of intense grief on negative psychosocial symptoms and outcomes (Lafarge et al., 2017; 2013). While it is readily recognized perinatal loss may lead to adverse psychosocial symptoms, very few women receive routine follow-up care or psychosocial support (Kersting & Wagner, 2012).

Gender plays a significant role in grief experience (Capitulo, 2005; Carlsson & Mattsson, 2018). However, very few studies have focused on men’s experience surrounding perinatal grief, particularly in the US (Carlsson & Mattsson, 2018). In a Swedish study, investigators found expectant fathers distance themselves from their emotional reactions and set their personal needs aside, focusing on supporting their partner (Carlsson & Mattsson, 2018). As a result, men are often at risk of not receiving the psychosocial support they may need. Furthermore, differing grief responses and expressions between men and women may marginalize individual needs, introducing both personal and relational strain (Capitulo, 2005). Consequently, many families struggle with preserving the intimate partner relationships throughout the complexities of perinatal loss (Fenstermacher & Hupcey, 2013). Though bereavement and psychosocial support is available for both men and women, such resources are commonly underutilized (Kavanaugh et
Overall, men and women experience and express grief differently, and despite this knowledge, limited research is available concerning the psychosocial consequences of perinatal loss on male partners (Capitulo, 2005; Carlsson & Mattsson, 2018).

Ethnocultural influences on perinatal grief must also be considered. While grief literature acknowledges cultural differences in bereavement, very few studies have included ethnically diverse participants, particularly in regards to perinatal grief (Montigny, Verdon, & Mcgrath, 2015; Whitaker, Kavanaugh, & Klima, 2010). Various cultures hold differing beliefs on grief practices, expression, rituals, and gender roles. Western cultures value autonomy and individuality, while Eastern cultures honor family and duty (Montigny et al., 2015). LatinX and Hispanic populations, according to one study, express grief much differently compared to Caucasian populations (Whitaker et al., 2010). Furthermore, LatinX women have one and a half times greater risk of undergoing perinatal loss, drawing into sharp contrast the need to identify culturally appropriate care practices and interventions for LatinX families (Whitaker et al., 2010). The US is home to a diverse and culturally complex population, therefore, it is of great import to expand research efforts to include evidence for culturally competent, individualized and holistic care.

**Perinatal bereavement.** The conventional bereavement period refers to a interval of mourning, grieving, and adjusting to a world without the physical, psychological, and social presence of the deceased, and is a highly individualized timeframe (Corless, 2015; Limbo & Davies, 2015). Perinatal bereavement, however, challenges traditional understanding of grief and bereavement models and theories (Brownlee & Oikonen, 2004), and is deeply influenced by culture (Corless, 2015). While the literature does agree the perinatal bereavement period begins
immediately following perinatal loss, consensus has yet to be reached concerning the conceptual boundaries of time (Fenstermacher & Hupcey, 2013). Various timeframes have been posited, suggesting the perinatal bereavement period spans four to six months, one to two years, or beyond a year (Fenstermacher & Hupcey, 2013; Johnson & Langford, 2015; Kavanaugh et al., 2015; Klier et al., 2002). Other researchers argue the bereavement period does not have an end point, but persists for a lifetime (Capitulo, 2005). As a result, perinatal bereavement is beginning to be seen as a journey rather than an event (Fenstermacher & Hupcey, 2013). Furthermore, ethnocultural norms and practices influence ongoing grief, particularly with ritualistic or meaningful dates, such as Mother’s Day or Father’s Day, the day the pregnancy was announced, the anniversary of the death, and religious or culturally significant holidays (Montigny et al., 2015).

Despite the absence of an articulate definition and timeframe, perinatal bereavement is characterized by a complex emotional reaction influenced by situational, internal and external factors (Fenstermacher & Hupcey, 2013). Situational factors influencing perinatal bereavement include the nature in which the death occurred, number of living children, prior perinatal loss, stressful life situations, and intimate partner relationship status (Côté-Arsenault & Denney-Koelsch, 2011; Fenstermacher & Hupcey, 2013; Hutt et al., 2015; Kavanaugh et al., 2017; Kersting & Wagner, 2012). Internal factors include perceived reality of the pregnancy, level of attachment, gender, and personality, while external factors involve culture, bereavement support, and interactions with family, friends, and the healthcare team (Côté-Arsenault & Denney-Koelsch, 2011a; Fenstermacher & Hupcey, 2013; Hutti, M. et al., 2015; Hutti et al., 2018; Kavanaugh et al., 2015). Perinatal bereavement is both highly personalized and deeply complex. Individual expressions of mourning will vary from person to person, influenced by culture,
gender, religion, and tradition. Emotions experienced during the bereavement period commonly consist of profound sadness, tearfulness, denial, anger, feeling alone, shock, and guilt (Brownlee & Oikonen, 2004; Côté-Arsenault & Denney-Koelsch, 2011; Hutti et al., 2015; Lalor et al., 2007). While there are no defined boundaries during the bereavement period, bereavement support such as perinatal palliative care, creating memories, rituals, and counseling may decrease the intensity and duration of the bereavement period (Chapman, 2013; Cole et al., 2017; Fenstermacher & Hupcey, 2013).

**Perinatal Grief Theory**

Traditional grief and bereavement theories demonstrate limited transferability to perinatal grief and bereavement practice models (Brownlee & Oikonen, 2004). The Freudian Grief Work Theory, the Kulber-Ross Stage Theory, and Stroebe & Shul’s Dual Process Model, having been briefly entertained in perinatal bereavement research, were quickly recognized as inappropriate guides for practice models (Brownlee & Oikonen, 2004; Hamilton, 2016). Furthermore, while many grief theories encourage detachment from the deceased, such as Kubler-Ross’s Five Stages of Grief and Parke’s Theory of Bereavement, parents experiencing perinatal loss demonstrate more positive outcomes when focusing on memories rather than severing emotional bonds (Capitulo, 2005; Whitaker, Kavanaugh, & Klima, 2010). Concepts and care interventions have been guided by auxiliary theories, including Swanson’s Middle Range Theory of Caring (Swanson, 1991), Attachment Theory (Bowlby, 1969), and Maternal Attachment Theory (Rubin, 1975). Hutti et al. (2017) have also constructed a perinatal grief intensity theoretical framework to predict grief intensity based on the perceived reality of the pregnancy, the mother’s ability to confront others, and the congruence between maternal desire and eventual outcome. The framework posits the meaning women attribute to their pregnancy experience is predictive of
grief intensity. Those who perceive their pregnancy as real will have greater intensity of grief than those who do not. Congruency between the mother’s desires during her pregnancy experience and the events which transpire are also predictive of lower grief intensity. For example, a woman who wishes to carry her pregnancy to term and is offered treatment choices which are then respected will have a less intense grieving reaction than a woman who is not offered treatment choices and feels she has no control concerning the healthcare she receives. Finally, women who are able to confront those they perceive as insensitive or unsupportive are less likely to develop intense grieving reactions. Women who are unable to confront others are less able to make decisions to increase the congruency of her pregnancy experience, and often feel isolated and alone (Hutti et al., 2017).

Robinson et al. (1999) posited gestational age was positively correlated to grief intensity following perinatal loss, using Bowlby’s Attachment Theory. However, Peppers & Knapp (1980) argued gestational age is not a significant predictor of grief intensity based on their study of 65 women experiencing either miscarriage, stillbirth or neonatal loss (Klier et al., 2002). These findings were consistent with findings by Kennel et al. (1970), as well as later researchers (Hutti, et al., 2015; Klier et al., 2002; Toedter et al., 1988). However, Attachment Theory is not without credence. With advancements in ultrasound diagnostic imaging, parents, particularly fathers, are able to see the fetus prior to birth, thus facilitating a relationship and sense of reality early on in the pregnancy (Klier et al., 2002). Further investigation on the role of antenatal ultrasound imaging on attachment warrants investigation.

Swanson’s Middle Range Theory of Caring, containing five caring process interventions, is becoming widely applied to nursing care of parents experiencing perinatal loss (Fenstermacher & Hupcey, 2013). The five caring processes include Knowing, Being with, Doing for, Enabling
and Maintaining belief (Swanson, 1991). These concepts align with emerging themes explored by qualitative researchers, providing guidance for nurses attempting to provide appropriate, holistic care for this patient population (Côté-Arsenault & Denney-Koelsch, 2011a; Lalor et al., 2007).

**Perinatal grief psychometrics:** As researchers endeavored to close the gap in knowledge concerning bereavement studies featuring perinatal loss, the need for a tool to measure perinatal grief quickly became evident (Potvin, Lasker, & Toedter, 1989; Toedter et al., 1988). Using the Texas Grief Inventory as a foundation, constructs were extracted from the 58 item Likert-type scale. However, the incompatibility between generalized constructs of grief and measuring grief relating to perinatal loss quickly became apparent. Recent studies have demonstrated key differences between grieving the loss of an adult loved one and that of a neonate (Brownlee & Oikonen, 2004). Furthermore, though the Texas Grief Inventory maintained face validity, reliability and validity has yet to be established (Potvin et al., 1989; Toedter et al., 1988). Eventually, over 22 studies were conducted in four different countries, and collaborative effort between researchers resulted in the development of the widely utilized Perinatal Grief Intensity Scale (PGIS) (Hutti et al., 2017; Hutti et al., 2013; Toedter, Lasker, & Janssen, 2001).

In addition to the PGIS, several other scales have been developed over the last few decades in an attempt to measure perinatal grief and bereavement. The Perinatal Bereavement Scale, developed by Theut et al. (1989) sought to determine whether or not parents who had experienced late perinatal loss displayed unresolved grief during subsequent pregnancies. Ritsher & Neugebauer (2002) developed the Perinatal Bereavement Grief Scale in an effort to measure perinatal grief, hypothesizing yearning was a key construct in measuring grief and mourning, essentially providing an epistemological base for prognostic and intervention studies. Other
researchers have utilized an assortment of psychometric tools to better understand perinatal grief and bereavement. Lafarge et al. (2017) for example, combined the Short Perinatal Grief Scale, the Posttraumatic Growth Inventory, and the Brief COPE scale to assess posttraumatic growth following perinatal loss. Other scales utilized during the investigation of perinatal grief include the Beck Depression Inventory, the Center for Epidemiological Studies Depression Scale, the Clinical Self-Rating Scale, the General Health Questionnaire, and the Hospital Anxiety and Depression Scale (Klier et al., 2002). Despite the vast assortment of psychometric tools utilized in assessing perinatal grief and bereavement, the issue of perinatal grief and bereavement not having been consistently defined in the literature remains, risking incongruously synthesizing study outcomes (Fenstermacher & Hupcey, 2013).

Perinatal Grief Intensity Scale

Based on the Perinatal Grief Scale (33 items), the PGIS (14 items) was developed to provide clinicians with a screening tool which would not overly burden grieving parents (Hutti et al., 2017). The PGIS is a reliable and valid measurement tool used to assess the intensity of grieving reactions in individuals experiencing perinatal loss (Hutti et al., 2018). It is not time limited, can be administered at any point following perinatal loss, and is accurately able to predict individuals at risk for developing chronic, complicated grief (Hutti et al., 2017). The purpose of the tool is to screen for women at high risk of developing intense grieving reactions, predict future grief intensity, and identifying those in need of additional grief interventions and follow-up care. The subscales of the scale are based on the perinatal grief intensity theoretical framework.

The PGIS is a 14 item Likert-type scale ranging from 1 (strongly disagree) to 4 (strongly agree), with three subscales, Reality (6 items), Congruence (4 items), and Confront others (4 items) (Hutti et al., 2017). International testing of the scale (Australia, Canada, England, New
Zealand, and South Africa) demonstrated good internal consistency reliability with Cronbach’s alpha ≥0.70 on all sub and total scales between the PGS and PGIS. Concurrent validity was established by examining the association between the PGS and PGIS at Time 1 and Time 2. A significant relationship on at both times between all total and subscales with the exception of Active Grief was demonstrated (r = 0.54, p < 0.001 at T1; r = 0.40, p < 0.001 at T2). No significant relationship was demonstrated between Active Grief and any of the PGIS subscales (Reality, Congruence, Confront Others). The constructs of each of the PGIS subscales supported the original 3-factor structure of the PGS through confirmatory factor analysis, and Chi-squared testing confirmed the hypothesis that the PGIS is non-inferior to the PGS (χ² = 509.2, p < 0.001).

The optimal cutoff for the PGIS is 3.535, with 57.7% of the variance explained by the three factors, and sufficient sensitivity (61.3%) and specificity (84.4%) in predicting grief (Hutti et al., 2017).

**History and development.** Kennell et al. (1970) began examining variables associated with perinatal grief by extracting constructs from bereavement and grief theories. Using a Likert-type scale derived from the Texas Grief Inventory, Kennell et al. (1970) asked participants to rate their grief experience on six key aspects associate with mourning (sadness, loss of appetite, inability to sleep, increased irritability, preoccupation with the lost infant, and inability to return to normal activities). Guilt was subsequently added to the constructs measured by Benfield & colleagues in 1978. Concerned by the concept of the ‘route’ by which an individual may arrive at a particular grief score, in 1980 Peppers & Knapp added nine constructs to Kennell’s original six, which included difficulty in concentration, anger, guilt, failure to accept reality, time confusion, exhaustion, lack of strength, depression, and repetitive dreams of the lost child (Toedter et al., 1988, p.436).
Eventually, collaborative effort between researchers in the 1980s and 1990s resulted in the development of a PGS, which later developed into the PGIS. The PGS consisted of 104 Likert-type items ranging from 1 (strongly agree) to 5 (strongly disagree), using subscales assessing a) Active Grief b) Difficulty Coping, and c) Despair. Recognizing the potential for participant burden, a PGIS was later developed, using subscales from the perinatal grief intensity theoretical framework (Reality, Congruence, Confront Others) (Hutti et al., 2017; Potvin et al., 1989).

**Psychometrics (reliability).** Test-retest reliability of the PGS was examined by Potvin et al. (1989) by interviewing 112 women at the time of perinatal loss and one year later. Eleven subsequent studies have further investigated the test-retest reliability of the PGS with resulting Cronbach’s alpha ranging from 0.70 to 0.96 at a significance level of $p<0.001$ (L. Toedter et al., 2001). The coefficient for the total PGS score ranged from .92 to .96, with the average subscale coefficients for Active Grief ($a=0.92$), Difficulty Coping ($a=0.89$), and Despair ($a=0.88$) demonstrating high reliability (Hutti et al., 2018). Using the PGS as a gold standard, Hutti et al. (2017) established reliability and validity of the PGIS, and were able to determine an optimal cutoff score (sensitivity of 61.3%, specificity of 84.4%). Using a Receiver Operating Characteristic (ROC) curve and traditional epidemiological measures such as sensitivity, specificity, positive and negative predictive value, and positive and negative likelihood ratio facilitated the identification of an optimal cutoff score for the PGIS (Hutti et al., 2018). The internal consistency reliability between the two scales demonstrated good consistency using Cronbach’s alpha (>0.70) on all subscale scores and the total score (Hutti et al., 2017).

**Psychometrics (validity).** Establishing validity of a new construct presents unique complexities. Toedter et al. (1989) approached this problem using construct validation suggested
by Cronbach and Meehl in 1955 (Potvin et al., 1989). This method is indicated for constructs such as perinatal grief which do not have previously established, criterion specific processes by which to compare individual measures and study outcomes. However, the differing opinions in perinatal grief literature at the time presented additional complications. This resulted in difficulty predicting behaviors relating to the construct of perinatal grief identified in the literature. For example, it was debated in the 1980s whether or not having living children affected perinatal grief intensity (Potvin et al., 1989). Nonetheless, Toedter & Colleagues were able to extract variables from the literature upon which to build the foundations of construct validity. The internal consistency of the resulting PGS produced a standardized alpha coefficient of 0.97. Additionally, multiple regression and correlation analysis supported the construct validity of the PGS (Potvin et al., 1989). Convergent validity was later established through consistent findings in 22 international studies (Hutti et al., 2017; L. Toedter et al., 2001). In 2015, the construct and convergent validity were further established by Hutti & Colleagues by combining the Pregnancy Outcomes Questionnaire (POQ) to assess pregnancy related anxiety (α=0.84), the Impact of Event Scale (IOE) to assess previous traumatic stressors (α=0.78-0.81), and the Center for Epidemiologic Studies Depression Scale (CES-D) to assess depressive symptoms experienced in the past week (α=0.93) with the PGIS. Additional studies in 2018 showed high sensitivity (97.9%) in predicting depression with a specificity of 29.6%. Prediction of future anxiety also demonstrated high sensitivity (95.2%) and moderate specificity (56.2%) in relation to perinatal grief intensity (Hutti et al., 2018).

Factorial validity of the PGIS was investigated to identify the ability of the PGS subscales (Active Grief, Difficulty Coping, and Despair) to accurately represent the subscales of the PGIS (Reality, Confront Others, and Congruence). No significant difference was
demonstrated between the subscales, with confirmatory factor analysis supporting the representation of the newly proposed three factor structure (Hutti et al., 2017). Additionally, all testing conducted concerning the validity of the PGS and PGIS were focused on determining convergent validity. Constructs regarding discriminant validity have yet to be established.

The PGIS is becoming more widely used in clinical settings to assess current grief levels surrounding perinatal loss, as well as to predict the future impact of perinatal grief on bereaved individuals (Hutti et al., 2017). While several studies have identified experiential themes associated with carrying a pregnancy affected by a fetal anomaly (Côté-Arsenault & Denney-Koelsch, 2011a), the impact of various thematic experiences on grieving the loss of a neonate has yet to be explored.

**Parental Experience with Fetal Anomaly Diagnoses**

As scientific inquiry has progressed over the decades, a methodological shift from quantitative to qualitative research has occurred as researchers investigate phenomena surrounding perinatal loss. To date, no studies have examined the experience of both mothers and fathers who have had a pregnancy complicated by anencephaly. However, a number of studies have explored the experience of parents carrying a fetus affected by life-limiting congenital anomalies (Carlsson et al., 2016; Côté-Arsenault & Denney-Koelsch, 2011; Lalor et al., 2007). Several notable themes and concepts have emerged, such as continuity of care, communication with healthcare staff, making memories, social disconnect, impeded decision-making, and grieving multiple losses (Côté-Arsenault & Denney-Koelsch, 2011; Greiner & Conklin, 2015; Lalor et al., 2007). Many of these experiences have been identified by parents as causing emotional distress (Côté-Arsenault & Denney-Koelsch, 2011a).
Fragmented healthcare was reported to cause significant distress and anxiety by a number of parents experiencing a pregnancy complicated by a congenital anomaly (Côté-Arsenault & Denney-Koelsch, 2011a). While anencephaly can be definitively diagnosed via ultrasound technology, other fetal anomalies, such as trisomy 13 and 18, require additional confirmatory testing (Bhide et al., 2013; Côté-Arsenault & Denney-Koelsch, 2011; Hall et al., 2016). As a result, parents come into contact with a number of healthcare professionals during diagnostic procedures over the course of several weeks. Interaction with a large variety of healthcare professionals and associated uncertainties common in fetal anomaly diagnoses invite opportunity for relaying inconsistent information to parents (Côté-Arsenault & Denney-Koelsch, 2011a; Lalor et al., 2007). Parents have reported severe emotional distress from receiving varied information, as well as from a state of limbo in which no one seemed to have answers (Côté-Arsenault & Denney-Koelsch, 2011a; Lalor et al., 2007). Such experiences can be highly distressing for parents, increasing the risk of developing intense, complicated grief (Greiner & Conklin, 2015; Hutti et al., 2017; Lalor et al., 2007). Additionally, parents wishing to carry a non-viable pregnancy to term frequently interact with a number of healthcare professionals during routine antenatal appointments. Limiting the number of healthcare professionals parents interact with on the treatment team has been associated with positive outcomes and higher reports of parental satisfaction (Greiner & Conklin, 2015; Lalor et al., 2007).

Communication during the discussion of a fetal anomaly diagnosis, treatment options, and end-of-life preferences is a central aspect associated with intense perinatal grief (Berry & Colorafi, 2019; Greiner & Conklin, 2015; Lalor et al., 2007). The centrality of communication in this area is dramatic, as negative consequences of insensitivity toward emotionally vulnerable parents can result in life-long adverse psychosocial symptoms (Greiner & Conklin, 2015; Lalor
et al., 2007; Wilkinson et al., 2016). Parents who do not fully understand the implications of the anomaly diagnosis or their treatment options are frequently unprepared to make treatment decisions concerning the fetus. This lack of understanding may stem from healthcare professionals being ill equipped to communicate with and care for parents with a pregnancy complicated by a fetal anomaly, as little training is provided to healthcare staff concerning the communication of bad news during pregnancy (Berry & Colorafi, 2019; Xafis et al., 2016).

As perinatal bereavement research has grown, it is no longer common place to immediately remove the neonate from the delivery room (McNamara et al., 2013). While study findings have mixed results, many parents have reported finding solace in holding the neonate (Carlsson et al., 2016; Carlsson & Mattsson, 2018; Lisy et al., 2016; Peters et al., 2015; Üstündağ-Budak et al., 2015). Spending time with the neonate, introducing family members, taking photographs, and other memory making activities have been reported as providing healing and closure (Côté-Arsenault & Denney-Koelsch, 2011a; Fenstermacher & Hupcey, 2013; Lisy et al., 2016). Many parents strive to find meaning in their experience, and do so by creating memories (Fenstermacher & Hupcey, 2013; Kersting & Wagner, 2012).

Parents experience grief not only over the loss of the fetus, but also over the loss of personal hopes and dreams of future parenting (Côté-Arsenault & Denney-Koelsch, 2011a). Many parents fantasize over the future of the unborn child, entertaining thoughts of sports, graduations, college, and marriage. Upon learning the fetus will not survive, the idealistic healthy baby with a bright future is shattered, leaving parents feeling lost and alone (Côté-Arsenault & Denney-Koelsch, 2011; Marc-Aurele & Nelesen, 2013). Parents also often feel socially isolated from their friends and family, perceiving those around them as wishing to ignore perinatal loss (Côté-Arsenault & Denney-Koelsch, 2011; Marc-Aurele & Nelesen, 2013).
Feelings of being utterly alone develop when parents reach out to friends, family and healthcare professionals for support without finding the support and understanding they seek (Côté-Arsenault & Denney-Koelsch, 2011). As perinatal loss is uncommon, individuals in the support role, such as friends and family, are often at a loss for how to interact with grieving parents. Frequently, co-workers and acquaintances wish to operate as if the loss never occurred (Côté-Arsenault & Denney-Koelsch, 2011a).

Care interventions and planning have emerged based on qualitative findings investigating parental experience surrounding perinatal loss. As a result, care models have transitioned from a protective model in which the death was denied or unacknowledged, to an open and supportive model, validating and recognizing perinatal loss (Capitulo, 2005). Several recurrent themes described by parents as being helpful include acknowledging the baby as real, participating in ceremonies and rituals, symbolism, and creating memories (Capitulo, 2005; Côté-Arsenault & Denney-Koelsch, 2011). Perinatal palliative care programs have subsequently been developed based on these findings, with the purpose of supporting mothers and families experiencing perinatal loss.

**Perinatal Palliative Care**

In 1997, the San Diego Hospice and Institute for Palliative Medicine established a perinatal palliative care (PnPC) program consisting of a hospice nurse, social worker, chaplain, and physician (Leong, Marc-Aurele & Nelesen, 2013). Years later, Hoeldtke and Calhoun recognized the lack of guidance informing clinicians caring for women with life-limiting perinatal diagnoses, formally proposing a model for perinatal hospice in 2001 (Cole et al., 2017). This proposal advocated for the development of a multidisciplinary approach to provide family-centered, compassionate care to expectant mothers choosing to carry a non-viable pregnancy to
term (Cole et al., 2017; Kavanaugh et al., 2015). Interdisciplinary teams included representation from maternal/fetal medicine, nursing, social work, and spiritual care. Core components of the proposal included collaboratively developing individualized birth plans addressing the emotional, psychosocial, familial, and spiritual needs of each woman and her family (Cole et al., 2017; Kavanaugh et al., 2017; Stenekes et al., 2014; Wool & Northam, 2011).

Throughout the PnPC process, parents are guided through their experience by a multidisciplinary team, which provides education on the condition of the fetus and assists in creating an individualized birth plan, incorporating elements of making memories, participating in spiritual or religious rituals, and making funeral arrangements (Cole et al., 2017). Making memories includes activities such as making hand and foot prints of their neonate at birth, taking pictures, and collecting memorabilia for scrapbooks as desired. Baptism or naming rituals are suggested for parental consideration. Often parents are devastated by grief and do not consider meaningful activities or intentional memory making. Social workers or spiritual care professionals work with families to incorporate these meaningful activities which may be vital in the healing process. In developing a birth plan, healthcare professionals may assist women and their partners not only in identifying their values and wishes through clear communication and shared decision-making, but in simultaneously acknowledging and celebrating the life of their unborn child (Cole et al., 2017; Kavanaugh et al., 2015). The resulting holistic approach to providing care is accomplished through a therapeutic incorporation of emotional as well as physical needs, and bereavement counseling is provided in both the antenatal and postpartum periods (Chapman, 2013; Kavanaugh et al., 2017).

Over 200 PnPC programs have since been developed throughout the US (Leong, Marc-Aurele & Nelesen, 2013). However, despite demonstrating positive outcomes, only 11% of
eligible women are referred to these programs (Marc-Aurele, Hull, Jones, & Pretorius, 2017). Several studies have investigated barriers associated with PnPC referrals (Cross, 2014; Inati et al., 2018; Wool, 2015; Wool & Northam, 2011). One of these studies developed an instrument to measure reasons preventing physicians and nurses from making PnPC referrals (Wool & Northam, 2011). Several barriers identified included a lack of awareness of PnPC programs among nurses and providers, a lack of time for healthcare professionals to provide counseling to patients, limited staff available for PnPC services, and societal misconceptions (Wool, 2015).

Furthermore, PnPC programs primarily exist only in large, urban hospitals. Smaller hospitals and towns, with fewer recourses, often do not have these programs and may be unaware of their existence (Stenekes et al., 2014). Additionally, limited reimbursement for PnPC programs introduces financial strain and impedes sustainability of these programs (Limbo et al., 2017).

**Gaps in the Knowledge**

Currently, studies featuring perinatal loss focus primarily on middle-class white women. Gender differences concerning the experience and expression of perinatal grief are acknowledged, yet male partners are severely underrepresented in scientific literature (Carlsson & Mattsson, 2018). In addition to gender, culture and ethnicity play a significant role in the grief response, necessitating further inquiry in scientific research (Fenstermacher & Hupcey, 2013). Whitaker et al. (2010) call for expanding perinatal bereavement research into LatinX communities, thus expanding the knowledge base across a more diverse population.

Furthermore, the majority of PnPC programs and research is conducted at major university hospitals or in large cities. Rural populations are in need of exploration, and perinatal grief and bereavement research has yet to expand into lower socioeconomic populations (Brownlee & Oikonen, 2004; Fenstermacher & Hupcey, 2013; Whitaker et al., 2010).
The examination of parental experience has uncovered key elements which distinctly place perinatal grief apart from traditional understandings of grief. New concepts and losses are introduced with perinatal loss, and the bereavement process does not follow traditional grief models. To date, there are no perinatal bereavement theories to guide practice. Generic grief theories are ineffective in explaining perinatal grief, and do not consider important aspects of the human experience, such as gender, age, culture and socioeconomic status (Brownlee & Oikonen, 2004). Furthermore, a theoretical definition of perinatal grief and bereavement remains unestablished, the absence of which “is particularly concerning in light of the scientific studies reporting outcomes of the measurement of perinatal bereavement.” (Fenstermacher & Hupcey, 2013).

Parents undergoing perinatal loss experience a unique, highly individualized bereavement response. The highly individualized nature of the complex bereavement reaction challenges researchers and clinicians alike. Though research efforts to close the knowledge gap concerning perinatal grief have expanded over the last few decades, studies examining the psychosocial consequences following perinatal loss remain sparse (Klier et al., 2002). While many important themes surrounding perinatal loss have contributed to the developing knowledge base, what remains to be explored is the impact of experiential themes, gender and culture on perinatal grief intensity.

Summary

Anencephaly is a uniformly lethal congenital anomaly. Receiving a diagnosis of a pregnancy complicated by anencephaly is a traumatic experience for parents. One quarter of women experiencing perinatal loss develop adverse psychosocial symptoms such as depression, anxiety, substance abuse, and suicidal ideation. Perinatal palliative care programs have been
developed to provide parents with resources and guide them through the bereavement period. However, despite the positive outcomes of these programs, referral rates remain as low as 11%. Furthermore, the majority of perinatal grief and bereavement research focuses on educated, Caucasian women, creating a need to explore the needs of fathers and ethnically diverse populations.

While perinatal grief and bereavement literature has identified common themes associated with perinatal loss, the impact these themes have on the intensity of the grieving reaction requires exploration. Hermeneutic interpretive phenomenology is ideal in exploring the essence and meaning of the lived experience of human phenomena, and will facilitate the identification of meaningful themes and patterns as experienced by parents who have lost a fetus or neonate to anencephaly. Utilizing the PGIS to measure grief intensity, the impact of experiential components surrounding an anencephalic pregnancy can be identified through measuring the grief intensity of each individual. By identifying key elements which are associated with intense grieving reactions, current practice may be informed and evaluated to provide holistic, individualized care to parents experiencing perinatal grief related to anencephaly.
CHAPTER III
METHODOLOGY

Perinatal loss is a global, complex and multifaceted phenomenon, introducing unique challenges to nursing practice and research (B. Chapman, 2013; Fenstermacher & Hupcey, 2013). The diagnosis of an anencephalic pregnancy transforms the pregnancy experience, requiring sensitive, unbiased care to facilitate the healing process (Wool, 2013b). Over the past five decades, recognition and research featuring perinatal grief has deepened nursing understanding of the psychosocial consequences of complicated grief, symptoms of which may persist for the remainder of the individual’s life (Hutti et al., 2018; Kennell et al., 1970).

However, to successfully achieve positive nursing outcomes which facilitate healing surrounding perinatal loss, it is necessary to understand the impact of various experiential components of perinatal loss on subsequent grief intensity. To investigate the impact of the lived experience of an anencephalic pregnancy on perinatal grief, this study utilized a mixed methods research design. Participants were invited to share their experiences through individual interviews, after which quantitative data was collected through a demographic questionnaire and the Perinatal Grief Intensity Scale (PGIS). The resulting comparison of qualitative experiential themes and grief intensity scores provide insight to the impact of an anencephalic pregnancy on parents. Washington State University (WSU) institutional review board (IRB) approved this study.

Research Design

Mixed methods research draws upon the paradigms and strengths of both quantitative and qualitative methods, recognizing the epistemological and ontological influence of the natural world as well as the reality of human phenomena (Östlund, Kidd, Wengström, & Rowa-Dewar, 2011). Established relatively recently, mixed methods research is a methodology in which
researchers utilize both quantitative and qualitative methods of data collection and analysis (Creswell, 2014; Doorenbos, 2015). Originating in specialty fields such as education, sociology and health sciences, mixed methods design has since been adopted by nurse researchers (Vandermause, 2011; Walker, 2011; Wilson, 2014). A mixed methods approach provides a strong design solution when significant limitations are introduced by using only quantitative or qualitative methods, and is ideal for newly developing areas of research.

A convergent parallel mixed methods approach with an interactive level of interaction was utilized for this study, in which quantitative and qualitative data was collected concurrently, then separately analyzed. Each method held an equal weight in priority (Cresswell, 2006). An assumption of this approach is that each method is able to gather different information which will ultimately synthesize results more completely than using only one method. Researchers feel this method is particularly useful when exploring psychological traits (Creswell, 2014; Mckim, 2017; Östlund et al., 2011). Qualitative data collection may include interview, observation and records, while quantitative data is generally collected using a psychometric tool. One limitation of a mixed methods approach is the differing requirements of sample size between qualitative and quantitative data. There are several solutions in addressing the issue of sample size, including weighing the rigor of the qualitative data to equal the quantitative sample size, increasing the qualitative sample size, or simply to not consider the differing sample size an issue (Tariq & Woodman, 2010). Interpretive Phenomenology typically uses a small sample size as it seeks to isolate themes, patterns and meanings from the lived experience (Cohen et al., 2000). Determination of an appropriate sample size is achieved by identifying sample sizes in the literature of similar studies, clinical experience, and determination of the intensity of contact necessary to gather sufficient data (Cohen et al., 2000). Sample sizes from perinatal loss studies
range from eight (Côté-Arsenault & Denney-Koelsch, 2011a) to 27 participants (Lafarge et al., 2017). Ideally, it was anticipated that up to 12 parent dyads would participate in this study, with the potential of 24 individual participants in total. However, as reflected in previous studies, fewer participants are sufficient to explore experiential phenomena surrounding perinatal loss (Côté-Arsenault & Denney-Koelsch, 2011a).

In this study, qualitative data was collected using individual unstructured participant interviews which were audio recorded. Qualitative data was gathered with a demographic questionnaire and the PGIS (Hutti, M. et al., 2017), which measures the intensity of grief reactions following perinatal loss. Both types of data were collected from all participants. The purpose of gathering PGIS scores in addition to conducting participant interviews was to identify common experiences which either facilitate or potentiate the grief response. This was best done by measuring the grief intensity using the PGIS and comparing results with the experiential themes emerging from the interview transcripts. Therefore, a mixed methods approach was carried out to thoroughly explore the impact of the lived experience of an anencephalic pregnancy on parents.

Hermeneutic interpretive phenomenology guided data collection and analysis of the qualitative data (Geanellos, 2000; Van Manen, 2014). Individual, unstructured, face-to-face interviews were conducted by the principle investigator (PI). All interviews began with the leading question, “please tell me, in as much detail as you are comfortable, about your experience of having a baby with anencephaly.” Prompts and clarifying questions were used to explore and enrich understanding of the meaning individuals apply to their lived experience. Interviews were audio recorded and subsequently transcribed by a professional transcriptionist. Upon completion of the participant interviews, quantitative data was collected by immediately
administering the PGIS and a demographic questionnaire through WSU Qualtrics (Hutti et al., 2017). The demographic questionnaire was developed by the PI by extracting questions from the literature featuring similar studies (Hutti et al., 2018; Prue, Hamner, & Flores, 2010; Roeske & Garlough, 2014), assessing the number of pregnancies and living children before and after the anencephalic pregnancy, relationship status, religion, annual household income, population density, educational level, ethnicity, and gender. The PGIS was scored, and all scores and demographic information were coded to the corresponding interview transcript. All data individually collected from parent dyads was coded to reflect each parental unit. For example, the interview transcript, PGIS results, and demographic questionnaire were all labeled with the pseudonym chosen by each participant, and parental dyads were identified by placing a false last name on all information (i.e., “John Smith” and “Amy Smith”).

The data interpretation phase began with hermeneutic interpretive phenomenological analysis of the interview transcripts by a team of individuals trained in the requisite method. Grief intensity levels were measured and assessed according to PGIS scores (Hutti et al., 2017). Scores greater than 3.535 indicate an intense grieving reaction, while scores less than 3.535 represent normal grieving reactions (Hutti et al., 2017). Data was synthesized using a Pearson’s correlation coefficient in SPSS25. Synthesizing qualitative and quantitative findings facilitated the identification of recurrent themes which contribute to normal or intense grieving reactions. These findings would not be possible through the use of a single methodology. Therefore, to adequately and thoroughly investigate key themes which contribute to intense grieving reactions, it was necessary to utilize a mixed methods approach.

**Qualitative Method: Hermeneutic Interpretive Phenomenology**
Hermeneutic interpretive phenomenology is a philosophy designed to elicit the lived experience of an individual. Developed by Martin Heidegger, this philosophy is concerned with understanding the meaning individuals apply to their everyday life experiences (Lopez & Willis, 2004; MacKey, 2005; Sokolowski, 1999). Rather than establishing theory, phenomenology provides insight into reality, allowing for a deeper understanding of experiential phenomena (Shosha, 2012). This philosophy is ideal for exploring areas in which very little is known on the subject matter, “making manifest that which is normally hidden in the human experience” (Lopez & Willis, 2004, p.728). As the parental experience of an anencephalic pregnancy had yet to be explored, hermeneutic interpretive phenomenology was the idyllic method to develop and enrich nursing knowledge concerning the care of individuals experiencing an anencephalic pregnancy.

**Philosophical foundation.** Phenomenology was founded by the German philosopher Edmund Husserl (1859-1938), in the turn of the twentieth century, coinciding with World Wars I & II (Sokolowski, 1999). Husserl, tiring of the circular philosophical questions of absolute truth, embraced the subjective nature of humanity and delved into the study of the human experience. Intrigued by the work of psychologist Brentano, Husserl argued philosophy need not consist of casual explanation, but could entail rich, deep description. Husserl maintained influences from empiricism, particularly relating to Hume and Mills (Sokolowski, 1999). Husserl’s work would become known as descriptive phenomenology. Distinguishing thoughts of this philosophical paradigm involve a rich description of major concepts, and of discovering the true nature, or essence, of the phenomena under investigation (Shosha, 2012). Descriptive phenomenology is essentially an epistemological standpoint by which to explain human experiences (MacKey, 2005). Rigor of this methodology is achieved by “bracketing” researcher bias, background, and
prior understanding of a concept. This approach to bracketing is known as transcendental subjectivity, an attempt to avoid influence on the object of study. Husserl also argued for radical autonomy, stating culture, society, and politics are not influential in how one experiences the world (Lopez & Willis, 2004).

Expanding on the work of Husserl, Martin Heidegger (1889-1976), modified the philosophical assumptions of phenomenology (Shosha, 2012). Rather than being satisfied with the current thought guiding descriptive phenomenology, Heidegger challenged many of Husserl’s assumption, including universal essences, bracketing, and describing. Arguing the idea of situated freedom, Heidegger believed in the influence of personal history, culture and contexts on one’s life experiences. Furthermore, Heidegger believed the phenomena of human experience warranted interpretation, discovering the meaning applied to the situation. Heidegger coined the term lifeworld to signify the influence of reality on an individual’s experience. Furthering this exploration, being-in-the-world became central to interpretive phenomenology, suggesting the human experience is more than subjective content, but rather a narrative of everyday life experience and meaning (Lopez, & Willis, 2004).

Max van Manan, an educator and phenomenologist, argued it is in the researcher’s favor to explore a phenomenon in which they have personal experience (Bynum & Varpio, 2017). Heidegger believed interpretation already exists, and what remained to be explored was the expression of the experience. Some scholars reject interpretive phenomenology based on Heidegger’s political affiliation with the National Socialism movement, an affiliation he was never recorded having had denounced (MacKey, 2005). Others have made attempts to remove portions of his philosophical teachings which appear to be influenced by Nazi propaganda. However, such attempts have been fruitless, as the philosophy collapses when observed only in
part. The very nature of interpretive phenomenology, in its reflection of time and space, mandates acknowledgement of one’s *being-in-the-world* as a central component of existence. Heidegger’s life and experiences are no exception, nor are his philosophical developments under his National Socialist affiliation. As a result, those choosing to explore phenomena using Heidegger’s philosophy must consider his political affiliation which, while regrettable, must nonetheless be acknowledged. Despite Heidegger’s questionable political choices and demonstrations of arguably poor character, his philosophy has been highly influential in scientific research (Bakewell, 2016; MacKey, 2005). The attempt to separate his philosophy from his *lifeworld* experiences would be philosophically inconsistent and inappropriate (MacKey, 2005).

**Heideggerian terminology.** Heidegger’s phenomenology entails four major concepts: *being-in-the-world*, *fore-structure*, time, and space (MacKey, 2005). *Being-in-the-world* acknowledges the person as being more than an object or viewer of the world. The idea of inseparability from the world is essentially the “framework of an all-encompassing world,” allowing individuals to wonder about their own existence (MacKey, 2005, p. 181). To capture the ability to question one’s being or existence, Heidegger coined the term *dasein*. *Dasein* is the idea that one exists authentically, having awareness of not only one’s being, but the potential for not being, in which the researcher can look for the meaning embedded in everyday life (MacKey, 2005; Shosha, 2012). Furthermore, Heideggerian phenomenology is not satisfied with exploring what can be known, but rather seeks to uncover the *meaning* of *Being* (MacKey, 2005). Fore-structure refers to the *a priori* awareness, or knowledge, of the phenomena under study. With prior understanding of the background surrounding the phenomena of interest, researchers are able to anticipate meaning prior to data interpretation (Benner, 1985; Mackey, 2005; Plager, 1994).
While it is argued that introducing researcher perceptions into data collection and analysis results in bias, Heidegger argues it is impossible to rid oneself of *a priori* knowledge and experiences, thus the concept of *fore-structure* acknowledges and embraces such backgrounds and understandings (Lopez & Willis, 2004). Furthermore, de Witt & Ploeg (2006) suggest acknowledging *fore-structure* is consistent with the rigor of interpretive phenomenology through exhibiting openness and balanced integration, which will be further explored below.

*Time* is argued as an essential construct to understanding meaning and experience. Without being grounded in time, the fundamental structures of the human experience do not exist. The temporal nature of time is an essential component of interpretive phenomenology which researchers must take into consideration. Finally, the concept of *space* is central to being-in-the-world. Space refers not to the geographic location of the person, but how they position themselves and what they perceive as important in their lives. Objects, concepts, or persons of great import will be brought to the foreground of their spatial world, while that which is of lesser meaning will be pushed into the background (MacKey, 2005).

**Heidegger on death:** In his sentinel work, *Being and Time* (1927/1962), Heidegger refers to three types of death: ‘perishing’ (verenden), ‘demising’ (ableben), and ‘dying’ (sterben). ‘Perishing’ refers to a physiological death, while ‘demising’ denotes the anxiety which accompanies the self-awareness of one’s inescapable demise. Heidegger spends the majority of his efforts on ‘dying’ (sterben). Dying, rather than a physiological absences of life, refers to a breakdown or collapse of meaning itself, or an ontological death (Aho, 2016). Thus Heidegger explores dying from the perspective of a death of one’s understanding of the underlying structure of how the world is ‘supposed’ to operate. An ontological death often accompanies great personal loss, be it wealth, physical health, or a loved one, and has begun influencing an
examination of the illness narrative (Frank, 1991). An ontological death commonly results in a loss of identity, for the significance and meaning of previously understood roles and practices in which self-identity was understood is lost. Furthermore, Heidegger maintains this event of ‘dying’ may occur more than once in an individual’s life, and that experiencing the loss of identity through an ontological death provides one with the opportunity to redefine ‘self,’ or a “discursive framework that allows the individual to make sense of and give meaning to their particular experience and to their identities as a whole” (Aho, 2016, p. 56).

**Philosophical interview underpinnings.** Hermeneutic interpretive phenomenology requires unique consideration of the interview structure. Consistent with interpretive phenomenological philosophy, interviews were unstructured in order to fully elicit the lived experience of the research participants (de Witt & Ploeg, 2006). Each interview began with an opening question (“please tell me, in as much detail as you are comfortable, about your experience of having a baby with anencephaly”), followed by prompts and clarifying questions as necessary. Prompts were used to gain an in-depth response or clarify participant statements, thus endeavoring to encourage a full revelation of the participant’s experience without biasing responses with directive questioning. The goal of this approach was to see what revealed itself in each participant experience.

**Application of Heideggerian philosophy to nursing research.** Heideggerian phenomenology has been extensively utilized in the social sciences to understand phenomena associated with the human experience. Seeking to explore the meaning individuals apply to their lived experience, phenomenology is particularly appropriate in the exploration of a patient’s experience of the health continuum, and has been utilized by a number of nurse researchers. In *Phenomenology of Practice*, Max van Manen states phenomenology “serves to foster and
strengthen an embodied ontology, epistemology, and axiology of thoughtful and tactful action,” offering unique insight capable of enriching nursing knowledge and practice (van Manen, 2014, p.15). However, the application of phenomenology as a research methodology by nurse scientists has been widely criticized, as the primary focus has been placed on the experience of an individual rather than the essence of the phenomena (Dowling, 2007). Nevertheless, the use of phenomenological philosophy in nursing research has transformed practice regarding patient centered nursing interventions and care.

Numerous examples can be found illustrating the use of Heideggarian interpretive phenomenology in nursing research. To summarize the application of this methodology, two specific examples will be provided, one about the experience of addiction and one about postpartum active duty Air Force women and their experiences with fitness exams after delivery.

In the first example, hermeneutic interpretive phenomenology was used to explore the lived experience of “Gabriel,” a 55 year old woman regarding methamphetamine use (Vandermause, 2011). The purpose of the study was to learn about methamphetamine addiction and recovery, with the aim of uncovering the meaning of this experience, generating an understanding of the complexities introduced by this phenomenon (Vandermause, 2011). The overarching pattern uncovered in this study was Being Wholesome, which contained two underlying themes: Using/Recovering as Caring and Connecting and Using/Recovering as Losing. Vandermause (2011) uncovers patterns and themes able to guide practice in a meaningful way, stating, “without understanding the salient experiences of caring and connecting amidst suffering and loss during periods of both addiction and recovery, prevention and treatment efforts will be insufficient” (p.314). She connects her findings with current literature, which likens addictive behavior to chronic illness, containing phases of acute
exacerbation (relapse) and remission (recovery). However, based on her findings, rather than current practice treating addiction as a chronic illness, offering assistance during an exacerbation of the condition, individuals are often “greeted with distain or disgust” (p.313). Gabriel illustrates this point by commenting on the perceived chasm between herself and healthcare professionals, the “us and them” (p. 314). Gabriel shared during the interview feeling like a cracker, which, though slowly disintegrating away, remains distinctly human, searching for wholeness. She recounts feelings of rejection and betrayal as well as triumph and love. Unexpectedly, these emotions are experienced both while using methamphetamines as well as during the recovery process. The paradox of experiencing loss during recovery, as uncovered by this research study, necessitates reevaluating current thought on the meaning of recovery from drug addiction, as these findings challenge current thinking of the dichotomous relationship between addiction and recovery (Vandermause, 2011).

In the second example, Armitage (2013) explored the lived experiences of active duty women training for the U. S. Air Force (USAF) fitness exam postpartum using interpretive phenomenology. Based on the phenomenological interpretation of the interview transcripts, two overarching themes were uncovered during this study: Striving to Perform under Pressure through Profound Life Transitions of Childbirth and Seeking Understanding from Others. Despite the variety of differences in the study sample, these distinct themes were shared between all participants. The study illuminated the profound stress and anxiety experienced by these airmen as they prepared to take the required fitness assessment, as well as the strain of transitioning into the role of motherhood. In the theme Seeking Understanding From Others, women felt no one, including spouses and close friends, understood the difficulties they were going through. Despite seeking support and understanding from family, spouses, peers,
healthcare professionals, and work centers, the women in this study did not feel they received the support and guidance they required to be successful in the many transitions associated with childbirth in the USAF. Furthermore, participants felt the policies and protocols of the USAF were unrealistic, and were unable to receive support from the institution as a whole.

Armitage (2013), linking her findings with current literature surrounding transitioning into motherhood, postpartum recovery and physiology, and fitness assessment requirements of the USAF, demonstrated an unrealistic rigidity in policy which profoundly impacted postpartum airmen. These findings ultimately contributed to USAF policy change regarding the timeframe in which active duty airman must take the fitness assessment after childbirth.

**Appropriateness for this research.** In order to provide individualized, appropriate care to parents experiencing perinatal loss, it is imperative to understand the unique needs of this population. Neimeyer and Sands (2011) suggest that the construction of meaning surrounding the death of a loved one is an integral part of the grieving process (Hamilton, 2016). Furthermore, perinatal grief has been found to not follow traditional grief models (Brownlee & Oikonen, 2004). It becomes necessary to understand the meaning parents apply to their loss to better understand how to best provide care.

Phenomenology is essentially the study of phenomena as experienced by humans (Sokolowski, 1999). As such, phenomenological inquiry has become increasingly popular among nurse researchers since the 1970s (MacKey, 2005). Interpretive phenomenology seeks to understand the meaning individuals apply to their lived experience (Walker, 2011). The intent of this philosophy is to stimulate new questions from formerly unexamined ways of thinking, thus uncovering that which was previously hidden or overlooked, ultimately achieving new understandings from complex phenomena (Vandermause, 2011). Furthermore, interpretive
phenomenology endeavors to understand first-hand accounts, thus illuminating human experiences (MacKey, 2005). Lastly, Heidegger recognized the influential role of cultural, social and gender implications, insisting existence cannot be separated from the world (Dowling, 2007; MacKey, 2005). This philosophy was chosen to guide this research as the overall purpose was to understand the meaning parents apply to their lived experience of having a pregnancy complicated by anencephaly.

**Quantitative Method: Perinatal Grief Intensity Scale**

Quantitative research reflects post-positivist paradigms, testing objective hypotheses and examining relationships between variables (Creswell, 2014). The variables used in a quantitative approach are objective, measurable, and able to be analyzed with statistical calculations. These inquiries test theory, protect against bias, and control alternative variables. Results must be generalizable and replicable to lend credence to the findings (Portney & Watkins, 2000). Using the scientific method, the post-positivist paradigm carefully examines objective realities through observation and measurement, following empiricist thought (Meleis, 2012). The post-positivist paradigm which guides this approach is informed by philosophers such as Comte, Mill, Newton, and Locke (Creswell, 2014). The goal of quantitative research is to shape knowledge through providing relevant, true statements, demonstrating cause and effect relationships, and testing theory. Clinical research, randomized control trials, and scientific experiments have revolutionized modern thought in western medicine.

The quantitative data collected during this mixed methods study will include the PGIS and a demographic questionnaire. The PGIS is a 14 item Likert-type scale with answers ranging from 1 (*strongly disagree*) to 4 (*strongly agree*) and three subscales, Reality (6 items), Congruence (4 items), and Confront others (4 items) (Hutti et al., 2017). The scale was
developed as a tool for clinicians to screen for women at high risk of developing intense grief following perinatal loss, in order to allocate resources and provide follow-up care. The PI developed a demographic questionnaire by extracting items from previous perinatal loss studies, assessing gender, ethnicity, socioeconomic status, household income, number of living children, and marital status. Both the PGIS and the demographic questionnaire were administered upon completion of the interview process. Collecting the quantitative data provided valuable information needed to answer the research questions concerning the impact an anencephalic pregnancy has on parents, providing important knowledge to inform and evaluate current practice.

**Method**

**Disclosure of Researcher Bias**

In contrast to Husserl’s phenomenological philosophy, Heidegger believed the researcher cannot separate prior experience and knowledge from personal interpretation of meaning (Bynum & Varpio, 2017; Sokolowski, 1999). It is necessary to disclose personal preunderstandings or presuppositions of a phenomenon (Spence, 2017). The purpose of this research arose from personal experience of the PI from close interaction with parents experiencing an anencephalic pregnancy.

**Sample and Recruitment**

Voluntary, purposive sampling was conducted online using a recruitment website detailing the research project. The host of anencephaly.info distributed the website link to parents on her Listserv. Recipients of the link proceeded to share the information on additional anencephaly support Facebook sites. The goal was to enroll up to 12 parent dyads (24 individuals). While a sample size of 24 is exceptionally large for a phenomenological study, parent dyads, though individually unique, essentially account for one experience, as they share
the same event. Individuals over the age of 18 years who have had a pregnancy complicated by anencephaly and are fluent in reading and speaking English were eligible for the study. Individuals who were currently pregnant were not eligible to participate. Though participants preferably included parental dyads, parents’ wishes to participate without their partner were not be excluded. To fully explore the lived experience of these parents, inclusion criteria encompassed anencephalic pregnancies which were both terminated and carried to term. There were no exclusions based on the amount of time which had elapsed since the completion of the anencephalic pregnancy. To protect the identity of the participants, each individual was asked to choose a pseudonym to be used during the study.

In contrast to most qualitative research methods which seek saturation of thematic findings, hermeneutic interpretive phenomenology requires neither saturation nor a specific sample size. The concept of saturation, in which no new themes or content emerge from transcript analysis, is inconsistent with phenomenological philosophy (van Manen, 2014). The complexities and depth of phenomenological meaning can never be fully explored; therefore, to strive for thematic saturation using phenomenological philosophy is impossible (van Manen, Higgins, & van der Riet, 2016). Instead, data collection ceases when group consensus among the analysis team is reached concerning the depth and reliability of the data in identifying the meaning the participants apply to their lived experience. When this transpires, researchers can agree the phenomenon has been appropriately investigated; typically eight to twelve interviews are considered necessary using methods consistent with phenomenology (Creswell, 2014). As this study utilized a mixed methods approach, and though quantitative methods require large sample sizes, the goal of this study was not to produce statistical significance. Therefore, the
small sample size was sufficient to produce a general association on the impact of experiential components surrounding an anencephalic pregnancy and perinatal grief intensity.

**Data Collection**

Informed consents were obtained prior to data collection and identifying information was secured in a private locked research office belonging to a member of the dissertation committee, in a WSU research office. Interview transcripts, demographic information, and PGIS forms were de-identified with encryption codes linking appropriate participant information. All online and e-mail correspondence was conducted through secured WSU e-mail servers. Data was stored on the secured online WSU College of Nursing intranet platform behind firewalls.

Individual open-ended interviews were conducted by the PI. All interviews began with a leading question (i.e., the research question), followed with prompts such as, “can you explain further,” or “can you give an example,” as necessary to more fully convey their lived experience (Vandermause, 2011). Each interview lasted until the participant has had enough time to thoroughly explore their story of this phenomenon, which ranged from 12 to 90 minutes. Interviews were audio recorded and professionally transcribed. Upon completion of the interview, participants completed the PGIS and demographic questionnaire using WSUQualitrics software program. Information was de-identified and coded to the corresponding interview transcripts. Parent dyads had codes indicating their relationship to further aid in data analysis.

**Interview Protocol**

To gain a detailed and contextualized meaning of the participant’s experiences, 24 individual, unstructured interviews were conducted. Each participant chose a pseudonym for the interviews. Every interview began with the opening question: “Please tell me, in as much detail as you are comfortable, about your experience of having a baby with anencephaly,” followed with
clarifying questions as necessary. Prompts and clarifying questions were open-ended, non-evaluative, non-judgmental, and consistent with hermeneutic interpretive phenomenological interviewing technique (de Witt & Ploeg, 2006). The interviews explored and clarified participant accounts of their lived experience, and included any combination of the following: “tell me more about…” “what do you mean by…” “what is an example of…” All interviews were conducted by the PI and audio recorded. Participants were informed that their participation was voluntary and that they could stop the interview at any time without repercussion. Though nearly all of the participants cried during their interview, none of the participant became overtly emotionally distressed. Should this have occurred, the interview would have been stopped and the established crisis management plan would be enacted, which included a) normalizing the reaction; 2) assessing for suicidal ideation; and 3) assisting the individual in choosing from a list of local service referrals. Mental health resources were made available to all participants in the form of a list of mental health services in their local area. They would have been assisted in choosing and connecting to an appropriate mental health service as necessary.

After each interview, field notes were taken and a post-interview memo was completed. All data was stored on a secure, WSU College of Nursing password-protected online intranet storage platform, which is maintained by a university-employed custodian.

**Data Analysis**

**Interview Transcripts.** Upon interview completion, transcripts were analyzed and interpreted using a team approach and following hermeneutic interpretive phenomenology processes. The interpretation team consisted of the PI and experts in obstetric and neonatal nursing, psychology, trauma, palliative care, mixed methods, and interpretive phenomenology. All analysis team members were trained in the requisite method.
Interview transcripts were analyzed for the emergence of common themes, patterns, and meanings using the hermeneutic circle (Fitzpatrick & Kazer, 2012; Vandermause, 2011). Several steps are involved in this analytical approach:

1. Transcripts were carefully read start to finish
2. Transcripts were re-read line by line
3. Notes were recorded of concepts, patterns and themes which stood out
4. Notes were reviewed. Observations were made of general categories containing
   a. Frequency with which ideas appear
   b. Position of ideas and concepts within the text
   c. Style of responses (works used, fluidity of speech, pauses)
5. Preliminary list of emerging patterns, themes and meanings were drafted
6. Transcripts were reviewed while reflecting on findings
7. A detailed summary of the transcript was drafted to convey emerging themes, patterns and meanings, including any combination of personal impressions, interpretive commentary, and/or a reiterating of the account, each justified by supporting quotes (with line references).

Written summaries were shared in weekly online analysis meetings by each team member and discussed in turn. Interpretations were collectively analyzed, and commonly emerging themes, patterns and meanings were recorded by the PI. With each subsequent analysis meeting, similarities and differences between participant accounts were discussed and analyzed. Through collaborative effort, final consensus was reached concerning interpretive findings.

**Perinatal grief intensity scale.** The PGIS, which measures the intensity of the grieving response, is indicated for all perinatal loss, and is not limited by the passage of time following
the loss. Score calculation is complex, however, the authors of the scale have developed an electronic application available online which calculates the scores automatically. If researchers are unable to utilize the electronic application, an Excel spreadsheet which has been set up to weigh individual items is also available (Hutti et al., 2017). All scores were calculated, interpreted, and coded to the corresponding interview transcripts using the pseudonym chosen by each participant. Results greater than 3.535 are indicative of intense grieving reactions and predictive of a need for follow up care (Hutti et al., 2017).

**Demographic questionnaire.** The demographic questionnaire was created by the PI. Items were extracted from the literature based on similar studies (Hutti et al., 2017; Johnson & Langford, 2015; Tseng et al., 2017), and asked questions relating to age, gender, ethnicity, income, and time elapsed since the anencephalic pregnancy diagnosis. To fully investigate the impact of an anencephalic pregnancy on the participants, it was necessary to obtain contextual and historical aspects of the experience. Furthermore, qualitative research investigating perinatal loss has focused primarily on white, educated, upper class women. This study included a diverse ethnic sampling and include the male gender. Identifying ethnicity, gender and socioeconomic status will expand the horizon of understanding through the exploration of the meaning participants attribute to their experience.

**Synthesizing Findings**

Upon completion of individually analyzing the qualitative and quantitative data, findings were synthesized using Person’s correlation in SPSS25. Subthemes from the overall emerging interpretive patterns were assigned a variable indicator, indicating whether the participant experienced a specific subtheme (1), did not experience the subtheme (-1) or did not mention the subtheme (0). For example, being provided with treatment options regarding decision-making
surrounding the anencephalic pregnancy was a subtheme in an overarching pattern. Participants who were given options from which they felt confident to make an informed decision were labeled as being present (1) in the experience. Participants who were told their only choice was to terminate the pregnancy were noted to have an absence of that particular theme (-1). Several participants did not mention one or more subthemes during their interview (0).

Thematic findings and demographic characteristics were then synthesized with the corresponding individual overall and subscale PGIS score using a Person’s correlation in SPSS25. Results were interpreted, identifying demographic characteristics and thematic findings associated with decreased grief intensity scores. Thematic and demographic characteristics which were associated with decreased grief intensity scores were further analyzed to identify elements which correlated to each theme or demographic characteristic.

**Evaluation of Rigor and Trustworthiness**

Methods to generically evaluate the rigor of qualitative studies prove to be inappropriate for evaluating the rigor of interpretive phenomenological studies. Out of necessity and based on the unique nature of the philosophy, assessment of rigor must be tailored to the method of interpretive phenomenology itself (de Witt & Ploeg, 2006). Traditional terminology of assessing qualitative rigor, such as trustworthiness, confirmability, and theoretical sampling techniques, are inappropriate (de Witt & Ploeg, 2006). Trustworthiness is achieved in reaching a single, empirical truth in the research findings. Interpretive phenomenology, however, does not reach a single truth, but rather multiple truths based on multiple, subjective interpretations. Additionally, interpretive phenomenology does not seek to remove researcher bias, but rather embraces and acknowledges the bias and foreknowledge of the interpretive research team (de Witt & Ploeg, 2006; Lopez & Willis, 2004; Reiners, G., 2012; Sokolowski, 1999). Sampling criteria are also specific to this
methodology, focusing on the participant’s willingness to share their experience of the phenomena in question, rather than utilizing standardized, criteria driven inclusion methods (de Witt & Ploeg, 2006). Such a sampling technique inarguably introduces sampling bias when evaluated by generalized standards for qualitative rigor. However, to adequately answer the research question using interpretive phenomenology, such sampling methods are necessary.

Based on the inconsistencies and inappropriateness of generalized qualitative criteria for assessing rigor, de Witt & Ploeg (2006) proposed a framework for assessing the rigor of interpretive phenomenology. The concept of balanced integration, which mandates the incorporation of the philosophical underpinnings of phenomenology, such as Heidegger’s dasein, lifeworld, and being-in-the-world, addresses the ‘frequent misrepresentation of and/or superficial inclusion of philosophy’ commonly found in nursing studies utilizing interpretive phenomenology (De Witt & Ploeg, 2006, p.224). Openness refers to the transparency of the decision making process throughout the study. Concreteness uses real life examples to connect the reader to the phenomena, illustrating the usefulness of the concepts in nursing practice. Resonance and actualization are the ‘epiphany’ moments, as described by van Manan, in which the reader is able to grasp the concept. A deep, resonating understanding of the phenomena occurs for the reader both at the point of reading the study results, and continues to be realized and interpreted with the passing of time (de Witt & Ploeg, 2006).

Human Subjects Protection

Institutional Review Board (IRB) approval was granted through WSU prior to beginning the pilot study, and informed consent was obtained from all participants before beginning the interviews. It was determined participants were at minimal risk for harm; however, due to the sensitive and emotional nature of the interview question, potential remained for participants to experience anxiety and/or emotional retraumatization while sharing their experience.
Furthermore, sensitive issues such as suicidal ideation could potentially surface during the interview, necessitating ending the interview, initiating the pre-established crisis management plan, and reporting the incident to the office of human subjects research protection. Statements regarding mandatory reporting regulations were included in the informed consent and reviewed with each participant prior to obtaining a signed consent.

Participant confidentiality was maintained throughout. Informed consents were the only document containing the participant’s name. Participants chose a pseudonym to be used throughout the interview. Identifiable information was kept in a locked cabinet in a designated WSU research office. All information shared between members of the research team was de-identified and shared only through university approved and maintained, password protected intranet storage platforms behind firewalls. The full-scale study obtained an addendum for further IRB approval from Washington State University, and followed the same procedures.

**Pilot Study**

A pilot study was carried out with the support of Drs. Severtsen and Oneal for the purpose of testing the appropriateness of the guiding philosophy, methodological design and interviewing process in answering the research question (see appendix A). Washington State University IRB approval was obtained for this pilot study. A total of three interviews were conducted using the same opening question: “*can you please tell me about your experience of having a baby with anencephaly?*” Participants included a married couple (man and woman), each of whom was interviewed separately, and one married woman whose husband declined to participate in the study, stating he was intrigued by the idea but unwilling to be interviewed. It was determined by the research team the study design and process were appropriate in answering the research question. Therefore, this study followed the same procedures and methods as the pilot study.
Summary

Perinatal loss resulting from anencephaly is a devastating, transformative event for parents. Intense grieving responses can have a lifelong impact on parental psychosocial wellbeing. In order for nurses to understand the impact of perinatal loss and how to provide sensitive, unbiased, and compassionate care, it is necessary to identify key elements which potentiate or facilitate the grieving process. Through a mixed methods approach, hermeneutic interpretive phenomenological exploration of parental experience surrounding an anencephalic pregnancy, meaningful shared experiences were explored and identified, providing valuable insight into patient needs. Identification of grief intensity associated with common meanings and experiences expands and informs nursing practice and perinatal palliative care programs. Demographic data augments current understanding of perinatal loss in a more diverse population by comparing participant characteristics to meaningful experiences. Overall, this mixed methods approach informs nursing practice and research by providing valuable insight to the impact of the lived experience of an anencephalic pregnancy on parents.
CHAPTER IV
RESULTS

The purpose of this mixed methods study was to explore the impact of the lived experience of an anencephalic pregnancy on parents. The specific aims of the study were as follow: i) interpret the primary lived experiences of parents regarding their history of a pregnancy complicated by anencephaly using hermeneutic interpretive phenomenology; ii) characterize residual perinatal grief using the Perinatal Grief Intensity Scale; iii) identify key shared experiences linked with grief intensity to identify common themes associated with poor psychosocial outcomes, such as symptoms of depression, posttraumatic stress disorder, and anxiety; and iv) identify opportunities for change in current healthcare practice which could improve the care of bereaved parents experiencing a pregnancy complicated by anencephaly.

Using a convergent parallel design, qualitative and quantitative data were gathered concurrently and analyzed separately. Qualitative data, gathered through individual, open ended interviews, were interpreted using interpretive phenomenology viewed through Heidegger’s lens of an ontological death. Heidegger’s philosophy of an ontological death is a unique approach to understanding how persons reestablish the meaning of Being in the wake of total world collapse. This approach was used to identify how parents who have experienced perinatal loss related to anencephaly reconstruct their lifeworld, thus identifying what was important to parents during this critical time of change. Exploring how parents rearranged and prioritized space, and interacted with and experienced time, was noted with the intention of educating healthcare professionals how to better understand the needs of parents experiencing perinatal loss. Quantitative data, in the form of the Perinatal Grief Intensity Scale (PGIS) and demographic questionnaire, were analyzed using
descriptive statistics and frequencies in SPSS25. Upon completing the separate analysis of both data sets, qualitative and quantitative data were synthesized using a Pearson’s correlation coefficient and quantization of experiential components from thematic findings occurring in the interviews (Sandelowski, 2001). Qualitative subthemes of overall interpretive patterns were labeled with variable indicators (discussed below) to calculate the correlation coefficient, thus identifying demographic characteristics and experiential components associated with more and less intense grieving reactions, as measured with the PGIS.

Respondents

Recruitment materials were distributed by the host of anencephaly.info to individuals who had previously experienced an anencephalic pregnancy. Recipients of recruitment materials subsequently voluntarily forwarded the study information to additional online anencephaly support groups such as Facebook. Seventy-six individuals sought to participate in this study in response to recruitment materials posted on anencephaly.info (76%), Facebook support groups (9%), and word of mouth (15%). The ages of the 76 respondents ranged from 16-62 years, the time which had elapsed since completion of the anencephalic pregnancies ranged from three weeks to 40 years, 74 received a diagnosis of anencephaly in utero, and 72 carried their pregnancy to term. Two women, whose pregnancies occurred 40 years ago, were not aware their child had anencephaly until delivery. One hundred percent of respondents were female, six stated their male partners also wished to participate, and four men volunteered to participate.

The majority of the respondents (88%) were from the United States, with additional responses from Australia (4%), United Kingdom (3%), Canada (3%), Switzerland (1%) and the Philippines (1%). Six individuals were ineligible to participate due to a current pregnancy (n=4) or being under 18 years of age (n=2). Due to the overwhelming response to recruitment
materials, respondents were enrolled in the study on a first come, first served basis. Three participants were delayed in confirming the interview appointment, and so were unable to participate. One woman declined to participate after confirming the interview appointment, stating she did not feel ready to talk about her loss, which had occurred only three weeks prior. Twenty-four participants, in keeping with IRB approval, were enrolled, interviewed and administered the PGIS and demographic questionnaire.

**Participants’ Demographics**

A total of 24 individuals, both male (n=4) and female (n=20), between the ages of 18-59 participated in this study. Participants were Caucasian (75%) and LatinX (25%). Four married couples participated and were interviewed individually, and the remaining 16 participants’ partners did not wish to participate. The majority of participants reported a Christian (58.3%) or Catholic (25.0%) affiliation. Other reported religious affiliations included Atheism (4.2%) and Judaism (4.2%), with one participant wishing not to answer. The educational level of the participants ranged from a high school degree (16.7%) to a doctoral degree (4.2%), and a collective annual income less than $25,000 (4.2%) to greater than $100,000 (16.7%), with the majority of participants earning $26,000-$49,000 (33.3%) and $50,000-$100,000 (41.7%) annually. Fifty percent of participants self-reported living in a suburban area during their anencephalic pregnancy, 33.3% lived in rural areas, and 12.5% lived in an urban area. Demographic characteristics, including number of pregnancies and living children before and after the pregnancy complicated by anencephaly, and marital status are shown in Table 4.1.

| Table 4.1 Characteristics of Study Participants |
|----------|---------|----------|
| Sex      | Frequency | %        |
| Male     | 4         | 83.3     |
| Female   | 20        | 16.7     |

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<thead>
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<th>Age</th>
<th>Frequency</th>
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<tr>
<td>Age Range</td>
<td>Frequency</td>
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<td>-----------</td>
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<td>12.5</td>
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<tbody>
<tr>
<td></td>
<td>Frequency</td>
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<td>6</td>
</tr>
<tr>
<td>1</td>
<td>10</td>
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<td>3</td>
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<td>&gt;5</td>
<td>2</td>
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<tr>
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<td>8</td>
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<tr>
<td>1</td>
<td>12</td>
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<tr>
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<td>2</td>
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<td>3</td>
<td>1</td>
</tr>
<tr>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>&gt;5</td>
<td>1</td>
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Data Collection

Data collection took place in October, 2019. Participants chose the interview modality, which included face-to-face (n=3), and by telephone (n=19), or Zoom (n=2). Face-to-face interviews were held in a location of the participant’s choosing, which included a coffee shop (n=1), the home of the participant (n=1) and the home of the researcher (n=1). All interviews were audio recorded and transcribed verbatim by a professional transcriptionist. Each interview lasted between 12 and 90 minutes in length, beginning with the opening question, “can you please tell me, in as much detail as you are comfortable, about your experience of having a baby with anencephaly?” All participants were interviewed only once as the details of each experience were richly conveyed. Upon completion of each interview, all participants invited the PI to contact them with any follow up questions. The PGIS and demographic questionnaire were distributed through WSU Qualtrics after completion of the interview and 100% of respondents completed both questionnaires. A personal research journal, which included reflective notes after each interview and analysis meeting, was kept to maintain an audit trail and contribute to reflexivity. The interpretive team agreed the content captured in the 24 interviews was sufficient to fully explore the phenomenon of having a pregnancy complicated by anencephaly.

Qualitative Findings

Twenty three of 24 participants provided in depth and rich accounts of their experience of having a pregnancy complicated by anencephaly. One participant, a Latino male, was eager to participate in the study, yet unlike the other participants, required frequent prompting. All but two participants began their story with the day they received the diagnosis of anencephaly, or “D Day,” as it has become to be known in the “anencephaly community.” The other two participants began their story by sharing the number of failed pregnancies they experienced prior to their
pregnancy complicated by anencephaly. Through the lens of Heidegger’s ontological death, emerging themes which arose from these stories as parents endeavored to rebuild their understanding of their *lifeworld* included *overwhelming trauma, patient-centeredness as critical, stigmatizing perinatal loss, embracing parenthood*, and *reframing reality*.

**Overwhelming trauma.** Throughout each narrative account, the magnitude of the trauma associated with the diagnosis of an anencephalic pregnancy outstripped each individuals’ previously established coping mechanisms. Participants, upon hearing the terminal nature of their pregnancy, found themselves in a state of profound shock. All participants described feeling surreal, and struggled to comprehend or process any information following the diagnosis. Several participants reported time standing still, while others felt like a third party looking in on the situation.

> I just could not believe that that was happening, as far as being inside the womb. That’s the safest place for them at that time. You know? So I just didn’t understand a lot of things. (Miriam)

> Initially, we were just in such shock, we really didn’t quite understand. You know, the doctor told us your child would not survive. We just went into such shock. (Dan)

> I really was in that P.T.S.D. state where I was shut off and could not take in anything. (Rebecca)

> I had just seen a heartbeat, and the arms and legs kicking, and breathing, I just didn’t, it took a long time for me to understand, because according to the ultrasound, she looked perfect. (Amy)

> She kinda proceeded to explain that it was a terminal diagnosis. Once you find out as a parent, at least for me, I know, and I suspect for my wife, you don’t really hear anything else. She did go on to explain what it was. Um, but, it kinda goes in one ear and out the other. You really don’t, at least, I did not really retain much other than “it was terminal.” (Michael)

All participants immediately consulted the internet, attempting to learn what anencephaly was and if it was possible to for the diagnosis to be made in error. As participants began to
accept and understand the diagnosis and the initial shock wore off, parents were faced with the need to make decisions regarding the course of their pregnancy and treatment (discussed further in “Patient-centeredness as critical”).

As time progressed, the demands of the experience continued to render previously established coping mechanisms as insufficient to negotiating day to day life. Participants exhibited hypersensitivity and reactivity to social triggers. The predominant source of reactivity was prompted by triggers emerging from interacting and conversing with other individuals. Interactions with others, whether family, friends, religious community members, coworkers, medical professionals or complete strangers, were highly influential in contributing to the intensity of the experience. Participants encountered both those whose comments were intended to be helpful, and those whose comments were described as cruel, regardless of the intention behind the words.

I did also sever a lot of ties with people when I saw the way they reacted towards me, and the cold comments that were made. (Sophia)

A lot of people mean well, and they just say a lot of stupid, and very hurtful things, even though they’re meaning well…stop trying to be helpful, ‘cause you’re not good at it. Just accept that that person is miserable. (Kim)

Don’t say, ‘it’s time for your grieving to be over.’ Or ‘you’ll have this baby and then you’ll have another baby.’ ‘It’ll be okay. You’ll still be able to have more babies.’ Like that is not helpful, I guess. (Rebecca)

The majority of parents continue to bear deep wounds from the words of others. Sophia reported that were it not for the insensitivity of others around her, she may have recovered quite differently. Kim struggles with members of her religious community, who would tell her that her son wasn’t going to live because she didn’t pray hard enough, or that her son wasn’t going to heaven. Dan was deeply wounded when others told him his son was not a person because he had
anencephaly. Participants relayed example upon example of heartbreaking responses they received from friends, family and community members, many of which continue to haunt them to this day.

In the overwhelming trauma of perinatal loss, participants experienced what could be considered acute and chronic emotional responses. Acute emotional responses were characterized by overwhelming emotions which interfered with and were disruptive to daily functioning.

And it’s difficult because you’re worried about real-life, and yet there’s this – this other thing happening. And I just wanted to enjoy this time with my son [in utero], and, you know, it was – it was a conflict. (Delia)

It was just overwhelming. It was all very – very overwhelming. I experienced a lot of trauma. I was very disassociated. Yeah, I just got bad. I was disassociated from like everything. Like I was going through the motions, but I was in haze. I wasn’t fully present. (Rebecca)

[I was] coming home to just, cry without sound. I remember being so broken that no sound would come out. Tears would just run down my face. (Forget-me-not)

And I just remember, the first few days were a rollercoaster of emotion. And one day, I tried to go to work. I got halfway there and I burst into tears and had to go home. You know, one minute you’re completely fine, and the next minute you would just break down in tears. (Elaine)

And then I just – she kept me off work that whole next week. And I don’t think I ever left my bed one time. (Michelle)

All participants reported profound sadness and grief which interfered with their ability to function in their given roles as parent, coworker, and/or spouse. Several participants reported not being able to get out of bed, others entered a dissociated state. Every participant, male and female, reported crying more than they had in their entire life. The strong emotional reactions continued to persist for years after the birth of their child as parent moved into what could be called a chronic state of grief.
But you do – you feel – you never, ever go a day without thinking about him. Like every day – every day I think about him. (Elaine)

And that whole first year after was just miserable. And like I fell into a pretty deep depression because of all of this. And it took a toll on – on my marriage. After losing that baby, it just never was the same. (Michelle)

Ever since I had (name reference) and I went through that process, I developed a social anxiety that I’d never had in my life. Um, and I couldn’t do things...I just psychologically was going through so much trauma. (Sophia)

Honestly, even now [10 years late], there’s days where I have to tell myself you have to get out of bed. You have to – you have to keep living. You have to keep going. Um, because there’s still days where I think it isn’t so right to keep living when your child is not. So there’s still days like that. (Delia)

I’m looking at a calendar on my wall right now that is August, 2013. I still cannot bring myself to bring this calendar off my wall. I glance at it every day, I don’t know that I look at it every day. But I know that it’s there. (Forget-me-not)

You can’t go through that and not come out with some sort of PTSD I suppose. (Dan)

The severity of the grieving reactions strongly impacted participants’ lives beyond the birth and death of their child. Michelle and Audrey quit their jobs, no longer able to function in their professional role, experiencing difficulty readjusting after their loss and the way their colleagues treated them. Elaine and Rebecca report remembering very little of the first year following their loss. Sophia gained 100 pounds and for filed bankruptcy. All participants continued to be affected by social triggers (fully discussed in “Reframing Reality”). Being around pregnant women or other babies was extremely difficult, and many participants would avoid social situations in which they knew they would encounter such social triggers. Two women reported contemplating suicide, at which point they recognized their need for professional help. All participants save two cried during their interview.

Learning one’s pregnancy is complicated by anencephaly is a traumatic and overwhelming experience. Parents struggled to function, and their experience required elevated
methods of coping. All parents, regardless of their coping capabilities, and regardless of the time which has passed since the event, continue to grieve the loss of their child to this day.

**Patient-centeredness as critical.** Patient-centered care respects patients’ values and needs, allowing patient preference to guide decision-making and care delivery (Tzelepis, Sanson-Fisher, Zucca, & Fradgley, 2015). The narrative accounts of all parents in the study were characterized by a presence or absence of patient-centeredness throughout their prenatal care. All but two participants began their story by describing the day they received a diagnosis of anencephaly, illustrating the lasting impact of those moments.

We had zero thought in our mind that any, that she wouldn’t be healthy, that anything would be wrong with her. Looking back, it’s heart wrenching, we were just excited to find out the gender. (Michael)

And then – then you – and they use words like abnormality and – and words you can’t really remember. And then, they tell you that your – that your – the baby’s not developing appropriately, and – and everything just stands still. I can’t even remember. (Tim)

It’s some stranger who, you know, basically ripped out your whole, you know, ruined your life, and, she wouldn’t know me if she saw me walking down the street. (Delia)

Eight parents (33%) were provided with treatment options, while the remaining 16 (67%) were told they must terminate their pregnancy. Several parents felt the information conveyed to them by their obstetrician (OB) was intended to coerce them into terminating their pregnancy, while others felt they were given neither information nor treatment options. The physician caring for Dan and Rebecca scheduled an induction of labor immediately following their son’s diagnosis, without their knowledge or authorization, insisting they go to the hospital to resolve the ‘non-viable pregnancy.’ Chloe didn’t realize she could continue her pregnancy until she arrived at her termination appointment. Lucero and Michelle elected to end their pregnancies after being told their lives would be in danger should they continue their pregnancy. The
experience of learning of the terminal nature of their pregnancy was imprinted in the minds of every parent.

Very curiously she immediately said, “Well we can go ahead and induce, let’s go get you admitted to the hospital and we can induce right now.” And Rebecca and I are not medical doctors, but we know enough to know that if you induce at 20 weeks your child is not going to survive. So at that point you are aborting your child. And we didn’t have any idea what we were going to do at that point, but we knew that wasn’t going to be it. (Dan)

Doctors had kind of pushed me to end my pregnancy…she gave me the option to terminate. (Miriam)

And a few minutes later, the doctor came back in and she was like, “Okay, I contacted your OB and she has you set up for a D&C Friday at the hospital.” She was like – and she told us what time to be there. And I was kinda like – we weren’t even really given a choice. (Michelle)

But they really push [to] terminate on the mother. I felt like termination was – my doctor basically told me, “Well, this was a mistake, and if you’d like to try again then we suggest that you terminate. But you want to carry to term, then you have to wait longer [to become pregnant again].” (Chloe)

We did feel like we fought the healthcare system the whole way…my doctors and medical professionals were -- I felt handled the situation very poorly at the time of diagnosis. (Rebecca)

Parents were frustrated with the lack of information provided by their OB, with only six of the 24 participants (including two married couples) feeling they received sufficient education regarding their child’s diagnosis to make informed decisions. All participants relied on the internet and online anencephaly support groups for their information, including a family practice physician who participated in this study.

I was still searching for answers. I was doing research on the internet. I knew he was going to die, but I wanted to connect with other people that went through what I went through. (Sophia)

We did ask like, “Can you give us more information on this?” So she went and printed out, um, some – some stuff off the computer and gave it to us. (Rebecca)
It was difficult in that when he was diagnosed, it was a very confusing time. I didn’t know anything about the condition, and if you don’t do much beyond the Google search, you’re, you know, not going to get a lot of information that’s representative of – of what it really means to continue a pregnancy in that situation. (Delia)

So that’s one thing that I would change in the doctor’s office was that they just give more knowledge to the mothers expecting with a fatal diagnosis. (Chloe)

And that’s the biggest thing is just education for even medical professionals. ‘Cause like I said, the nurse in the hospital didn’t even understand, and she’s in the medical field. (Michelle)

The dichotomy of patient-centeredness versus a lack thereof continued to be characterized by a presence or absence of a perceived emotional sterility and medicalization of the anencephaly by medical professionals. Sophia described her prenatal care as a cold experience. Chloe, Christiana and Laura felt their presence was merely tolerated, and were constantly reminded of the “non-viability” of their pregnancy. Dan was frustrated with feeling the specialists overseeing his wife’s care thought they were “idiots.” The moment Maria’s pregnancy was diagnosed with anencephaly, her “baby” immediately became a “fetus.”

She used the word, um – I can’t remember it off the top of my head. You know, the real medical term. You know, it isn’t used to induce emotion of any sort, is what I would describe it as. (Rebecca)

And they – that doctor thoroughly recommended that we have an abortion – or – I’m sorry – a selective reduction. (Elaine)

His words were just so harsh. (Sophia)

My doctor called my son an unviable fetus. He even made the point of telling me that if he had a viable fetus in a delivery room when I’m delivering, that other baby will take credit over me and my baby. (Chloe)

No one treated her [his daughter] like she was an entity, like she mattered. (Mac)

Parents were further frustrated with the lack of information they received from their providers regarding the nature of an anencephalic pregnancy, much of which directly contrasted with their personal experience, and many parents felt the information provided by their OB of the
risks associated with anencephaly were merely scare tactics. Chloe was told her son would be
deaf and only feel pain. Sophia was told if her neonate survived birth, he would not have any
sensations. However, parents, whose neonates were carried to term and briefly survived, reported
that those babies were able to cry, swallow, and eliminate. They reacted to sound and were
comforted by touch. Sarah, a family practice physician, was surprised by the normalcy of her
son, who lived for 28 days. She described him as behaving like any other newborn. He would cry
when he was hungry or needed changed, and was comforted when held. Forget-me-not’s
daughter’s feet were ticklish. Mac and Samantha’s daughter would turn her head towards their
voices, and enjoyed being snuggled. Elaine’s son suckled, enjoyed electronic dance music, and
was comforted by touch.

Despite the overwhelmingly negative experiences of some parents, others had positive
interactions with their providers. Tim and Forget-me-not, after transferring to another facility
following a negative and cold experience, felt supported and listened to by their physician.
Miriam and Elaine’s providers researched anencephaly in an attempt to provide them with the
care they required. Miriam was very moved by the compassion of her provider, who told her he
admired her for carrying her pregnancy to term, and prayed with her during her prenatal visits.
Experiencing patient-centeredness and being provided with education and options was extremely
important to participants.

I’m glad that I was given the choice. And for me, the choice was to carry to term.
(Elaine)

We were eternally grateful that we did not feel any pressure to either option, she did not
present it in a, ‘you should really get an abortion,’ ‘you should really go to full term,’
there was really a non-existent pressure, she presented the options that were there and let
us decide. (Michael)
I think that there’s a lot of work that can be done there, more like a person-centered approach, instead of a, ‘there’s no reason for you to carry this baby because it will not survive after they’re born.’ Yes, that’s – that’s a big takeaway obviously. (Rebecca)

Many participants felt they were no longer a priority to their care team following the diagnosis of anencephaly. Parents, wishing to make good decisions for their anencephalic child, began researching anencephaly on their own. Despite telling their providers how they would like their care to continue, many were ignored, and their desires and concerns disregarded.

It was frustrating for me that she wasn’t doing, with what I assumed would happen. (Kim)

The doctors just ignored me. I begged them to induce me when I was about thirty-eight or thirty-nine weeks pregnant because I was in so much pain and I was so humungous and I couldn’t breathe. And they – and then they told me that they weren’t going to. That the baby – it would be hard to get the baby out because he was missing parts of his head, and they said I had to wait it out. And they made me wait forty-two weeks. (Sophia)

My doctor wouldn’t even give me a C-section. I told him a C-section would better my chances – better chance that my baby will live and I’d get to meet him – and my doctor refused to do a C-section. (Chloe)

I just wasn’t a priority. (Laura)

Following weeks of continuing a pregnancy which was treated as ‘non-viable’ and low priority, all but six parents were relieved by the nursing care they received from the labor and delivery (L&D) nurses in the hospital setting. Seventy five percent of parents received patient-centered, compassionate nursing care. The L&D staff made exceptions for the parents, extending visiting hours and allow additional family members to visit after the birth of their child. Parents were roomed at the end of the hallway, away from healthy neonates. Birth plans were acknowledged and incorporated, parents were asked about their preferences, and the nursing staff treated their babies with respect, “as if they were any other baby.” Furthermore, for the first time
during the entire experience, parents were given control over their care, and asked what they wanted to happen throughout the delivery process.

We really felt the people at the hospital, which were interacting with us, were absolutely incredible. They did things like gave us a printout of her heartbeat, and did another ultrasound as we were getting admitted. (Amy)

I had a nurse, he actually took me out through the back so I didn’t, you know, have to experience seeing mothers leave with their children or anything. So that just – he actually took me in the back of the hospital when I went home. (Miriam)

She already told her staff, you know, expect this. And they had already made little memory boxes, and a couple people had made blankets or they had relatives that made blankets for him. And they were very welcoming and ready for this. You know? And they were trying to make it a positive experience. (Elaine)

We wrote up a birth plan, and we included our letter in the birth plan...But, you know, handed her the birth plan and the letter. And she came back a little while later and she said, “Your story made me cry.” And then after that, it was like she was our friend. (Delia)

They extended visiting hours for our family, and accommodated over 50 people in the waiting room. They gave us conjoining rooms, so our family could stay in one room. (Forget-me-not)

So while I was in ICU, my whole family was able to come up and meet him. (Elaine)

The nurse was beyond amazing. She came to my room and she was like, “I’m usually pretty good with death, but this is like really different than anything I’ve ever handled before.” She was like, “If I start getting emotional, I’m going to excuse myself from the room.” And she was like, “But I just want to tell you that I feel for you.” (Audrey)

The stark absence of patient-centeredness many participants experienced shattered their expectations of the medical system, dissolving any notion of a patient/provider relationship. The lack of compassion and empathy throughout prenatal care and communication potentiated parental grief. The majority of parents were not treated as a priority and their desires and preferences were disregarded. Once participants were admitted to the hospital to deliver their baby, the “harsh” treatment they had experienced was sharply contrasted by the compassionate
and empathetic nursing care in the L&D ward. However, though it was not discussed by the participants, it was noted by the research team that only one woman received follow up care.

**Stigmatizing perinatal loss.** Each narrative account was impregnated by a stigmatization of perinatal loss throughout the participants’ experiences. Parents were acutely aware that talking about their loss made others uncomfortable and unsure of how to act around grieving parents. Many participants lamented the obvious avoidant behavior of friends, family, coworkers and acquaintances they encountered in an everyday setting, such as a grocery store or at church. Such avoidant behavior demonstrated to parents an unwillingness to listen to parents talk about their experiences.

Nobody wants to talk to you ‘cause they don’t know how to deal with it. (Michelle)

I needed to grieve and I needed to talk about what was going on, but there were certain people in my life that wouldn’t allow me to. (Sophia)

I honestly have never discussed it with anyone. This is the first time. I’ve never really been able to talk about it. (Miriam)

I came to realize later that many, many, many pregnancies do not go well, but it’s just that people never discuss it. (Dan)

I wish people would have talked about her more. Or not avoided the topic, the diagnosis. Or us, for that matter. (Forget-me-not)

People don’t want to talk about it. It’s nice to have people acknowledge your child and to ask questions. (Elaine)

The silence and unacknowledged loss caused parents to feel isolated and alone. Consequently, parents learned to keep their grief to themselves to avoid making others uncomfortable.

I think we did feel very alone, like we weren’t allowed to talk about it in front of family and friends. Like whenever we’d bring it up, they’d get angry or change the subject. (Sophia)

I wish people knew how lonely it was, not just after she was gone, but through the whole pregnancy. We like to talk about her, that’s how she’s a part of the family. But people don't want to talk about it, 'cause it's awkward, but that's really hard. (Mac)
And it was very isolating because people just kinda tiptoed. They never wanted to – to talk about anything. So people would just avoid me, and it was – that was really, really difficult. (Michelle)

The importance to each parent of being able to talk about their experience with someone quickly became apparent during each interview. Forget-me-not expressed her gratitude for the opportunity to share her story multiple times during her interview. Elizabeth described the interview as “therapeutic,” as she is commonly “not allowed” to share her story with others. Mac shared after his interview that he had never told his story start to finish before in the 12 years following his daughter’s passing. Miriam told her story for the first time in nine years after the passing of her daughter. Talking about their experience and sharing their story was reported to by parents as more than simply therapeutic; speaking of their children was a way for the parents to acknowledge their brief existence in this world and of keeping their child’s memory alive.

And so for me, (name reference)’s story and for me telling everybody about (name reference) is my way of keeping him alive. (Chloe)

I did like talking about it. I did like sharing ‘cause it did help. (Audrey)

And right away it felt like a weight had been lifted because, you know, other people were carrying this with us. (Delia)

The ensuing isolation was only one aspect of the disenfranchisement and stigmatization brought about by silence. A societal misconception of finality was also attached with the stigma of perinatal loss, characterized by assumptions of an “appropriate” timeframe for the bereavement period. Parents felt pressed to fit into the expected grief trajectory, that is, to suddenly no longer be grieving after the completion of the pregnancy.

Everyone wants it to be like a book, with a beginning and end, when all of the sudden you’re done dealing with your, you know, sadness and your grief, and all that, but it’s really not, at least in my experience. (Michael)
I felt like I was expected to go on as if nothing had happened. I started sleeping a lot, it became too hard to face the day. (Christiana)

When everybody went home and everything is when the true hell began. After that, it was super – probably four years of complete hell for me and my husband. (Sophia)

After my rainbow baby I mentioned to a friend that I wondered what (name reference) would think of his little sister, and she looked at me and said, “you’re still on that?” Like I wasn’t supposed to feel anything about my baby I lost. (Laura)

Society also imposed gender “appropriate” behavioral norms. Kim watched as her husband struggled to fulfill his role as a man and provider of the family, returning to work shortly after the loss of their son. She states he was expected by his employer to “suck it up and work.” The men in this study expressed their desire to focus on the needs of their grieving partner. They were expected, by themselves and others, to be strong and supportive.

So he was struggling with how to be a man, go to work, you know, but also deal with all the things that losing a child presents you with. (Kim)

I just wanted to support my wife, but I had my own grief. (Mac)

After she was born it was, you know, notifying people, consoling them, not consoling them. People avoiding you. Picking up the pieces. Being strong. (Tim)

The stigma associated with perinatal loss further resulted in societal judgement of parents for their decisions. Others, whether family, friends, and members of their social community, held strong opinions regarding the anencephalic pregnancy and grief trajectory, which they frequently expressed to the grieving parents. Dan was told by several individuals that he and his wife were selfish to carry their pregnancy to term, and that his son was not a person if he had anencephaly. Laura had to continuously defend her decision to carry her pregnancy to term as others expressed their lack of understanding for her desire not to terminate. Michelle and Lucero experienced rudeness and harsh comments from friends, family, and coworkers who disagreed with their treatment decisions not to continue the pregnancy. Rebecca struggled to understand why those in
her social circle felt comfortable telling her she made a selfish choice. Samantha had to put boundaries into place in response to the cascade of insensitive comments by family and friends.

Every parent continues to bear emotional scars at the judgmental hands of others.

They looked at us like we were nuts for having a funeral. Like we were nuts for carrying a child to term knowing that he is most certainly going to die during birth. And you know, they say it’s selfish to try and have him born alive just so we might hear him cry for a moment, or just have him take one breath. (Dan)

People would come up to me and say, ‘I don’t really understand your choice of why you’re doing this right now.’ Which they meant continuing the pregnancy. (Laura)

She stopped dead in her tracks and came in my office and told me, ‘How dare you [terminate the pregnancy].’ She was like – she was like, ‘You had no right to do that.’ She was like, ‘That was not God’s plan.’ And I just sat there. I – I didn’t know what to say. (Michelle)

Even the nurses there. She was like, she understood, but I guess with her beliefs she thought it was wrong to do it. (Lucero)

Perinatal loss is not only silent and commonly unrecognized by society, but is unacknowledged by governing authorities. Stillborn neonates are not issued a birth certificate, have no social security number, and consequently have no legal documentation that they ever existed.

We don't even have a birth certificate for her. (Maria)

I found out that stillborns aren't considered people, so she doesn't have a death certificate, or a social security number. It's like she never existed. (Amy)

Termination of a pregnancy complicated by a fetal anomaly is not recognized by society as a traumatic event, consequently, medical leave is not issued, and Michelle was expected to return to work the week after her termination. Paternity leave, if granted, may be as little as one week. Kim’s husband was not granted extended time off of work to grieve the loss of his son. Michael, however was provided a month off work, which he found very beneficial in his attempt to heal after the loss of his daughter.
He was working at a place at the time where they only gave him a week off. So he had a week, and then he had to go back to work. (Kim)

She kept me off work that next week. And I don’t think I ever left my bed one time. (Michelle)

I had three months off from work, which was crucial. I know a lot of women aren’t able to have that opportunity. (Amy)

Parents found solace through storytelling as a means to keep the memory of their child alive. However, they were frequently met with avoidant behavior and clear social queues that it was not socially appropriate to discuss their loss with others. Parental grief was disenfranchised not only by silence, but also by an expected timeframe of the bereavement period, and the associated lack of understanding of the profound impact of losing a child they never met had on their lives. Fathers functioned in the roles expected by others, which was that of the strong man who supported his grieving partner. Parents were constantly questioned about their decisions regarding their pregnancy, whether they decided to carry or terminate. Frequently, parents were labeled as selfish regardless of their treatment decision. Parental grief was thus disenfranchised by the lack of acknowledgement and silence of some, and potentiated by the judgement of others, judgement which commonly stemmed from personal beliefs and ignorance about the condition of anencephaly. The stigma, silence, and judgement impregnating perinatal loss was very isolating to parents and marginalized their grief.

**Embracing personhood.** Despite the pregnancy being deemed medically ‘non-viable,’ every participant in this study experienced their pregnancy – and child within – as real. Many parents had already picked out names for their child, and even fantasized about personality traits and characteristics. Elaine reported her son liked electronic dance music, and every time she felt he was not moving enough, she would put headphones to her abdomen so he would start moving inside the womb. After birth, many parents recognized physical characteristics similar to family
members, such as noses and hair color. Parents marveled at their child’s hands and feet. When participants shared their recollection of the moment they met their child after birth, they used words such as “perfect” and “beautiful” to describe their neonates. Every parent struggled with the competing emotions of wanting to meet their child after birth and never wanting the pregnancy to end. Amy cried as she shared that she knew the moment she said hello, she would also have to say goodbye. The reality and love each parent felt for their anencephalic child echoed through each story as parents described the moment they met their son or daughter.

He was just the most beautiful thing. And it’s silly now that I worried what he would look like because he was just the most beautiful thing I’d ever seen. (Delia)

So once I had the baby, I held him for a very long time. He looked just like my other son, (name reference). He had black hair, though. (Sophia)

She was everything I hoped for. Perfect, beautiful. (Forget-me-not)

She was perfect in every other way. Her little body was just perfect. You know, she didn’t look any different than any of my other kids, but, you know, she wasn’t animated. (Tim)

Parents attempted to find meaning throughout their pregnancy experience in an effort to make sense of the situation. Towards this endeavor, parents commonly gave their child a role. Forget-me-not shared she felt blessed to carry her daughter to term, and felt her daughter’s story could be a part of her helping the adolescent children she counseled. Several parents chose to donate their child’s organs to spare other parents the pain of loss they themselves were going through.

Thinking, you know, you’re in this with me, you’re doing this right alongside me. I felt honored that she would be a help to people, and she could be a part of that process. I think that’s part of how I was able to get through. (Forget-me-not)

We had looked into organ donation, and found an opportunity for our daughter to participate in whole body research. (Amy)
I was very proactive. I fought for three weeks on end to get my son’s organs to somewhere where they needed to be. (Chloe)

I think the thing that got us through it mostly was knowing that we were going to donate and spare someone else the burden of feeling that loss. Or, you know, saving another kid’s life. If ours can’t live, let’s please give someone else – let him give someone else his life to continue on. (Elaine)

Parents overwhelmingly expressed a wish for their pregnancy to be treated as any other pregnancy, that is, to be treated as if their child was going to live. They desired to continue routine prenatal care, such as receiving RhoGAM injections as necessary, and to be treated with respect and compassion.

And I wish he [the doctor] just understood that I loved my baby as much as the next mom loved her baby…because to the doctors, our babies don’t have any brain or brain function, but our babies have a heart. And they are very, very viable and they are loved. (Chloe)

She's not less because she's not going to live as long. I wish people could have understood that. (Mac)

They talked about how beautiful she was, and brought her little hats, and blankets…they treated her just like any other baby. (Sandy)

When we found out she was a girl, she was like, have you thought of a name? She treated her like a person, and I really appreciated that. (Gianna)

But the hospital staff, they treated my son with respect. (Elaine)

Parents reported being frustrated and dishearten when routine care was withheld, or when they were not considered a priority. Rebecca noted the language her doctor chose to use was meant to remove any sort of emotion from the situation. Chloe was informed by her doctor that he would not administer care during her labor should a “viable” labor coincide with hers, and when she was in labor he informed her he was tired and left the facility. Several families had to wait over five hours to begin a planned induction or C-section.
Participants actively parented their child throughout their pregnancy. They shared that the awareness of the limited time with their child caused them to be deliberate and intentional with their choices, actions, and decisions.

And so at that point we were only going to be parents to our child for nine months, and that was probably going to be it, and so those weekly appointments ended up becoming very important, because that’s how we could connect with our child. (Dan)

You only have a certain amount of time to be parents, and we were going to do the best we could during that time. (Mac)

They said it wasn't worth putting my life in danger, but that's not what it was for me. For me, it was giving my daughter as much life as possible. (Christiana)

Intentionally making memories echoed throughout the narrative accounts. Parents carried out parent/child activities such as going to the park or beach, traveling, baking cookies, and watching children’s films in the theater while their child remained in the womb.

Because we chose to go full term we were able to create memories with her. We did a day at the zoo and we delivered cookies on Valentine’s Day, and suddenly everything with her, with every day her heart was beating, was just so precious, and so meaningful that we started living with so much intention, and so much gratitude. (Amy)

I did whatever I could while we were pregnant to create those memories. (Rebecca)

We took him -- everywhere that we would want to take him when he was born, we took him when he was inside me. And those memories will keep me going forever. (Chloe)

Many parents used positive association and symbolism to establish a place in their family and maintain a connection with the child they would lose.

We chose to use the poppy as a symbol of everything good that came from everything we experienced, the experiences of her, her memory and everything she taught us. So ah, poppies, every time we see poppies now, we think of love and kindness and choosing joy. And that has been really powerful. (Amy)

We would collect them [rocks] and bring them to the cemetery. So she, it would just symbolize, [that] she got to be a part of those experiences with us. (Forget-me-not)
Despite the reality of the pregnancy and child to the parents, society recognized neither the reality of the pregnancy nor the profound depth of the subsequent loss. Making memories and actively parenting their child brought solace and positive associations to those who were able to carry their pregnancy to term. In contrast, Michelle, who was unable to carry her pregnancy to term, strongly believes having been able to make those memories and have time with her child would have eased her pain, potentially allowing her to heal from the profound emotional turmoil she continues to experience nine years after her terminated pregnancy. Regardless of the choice to carry to term or terminate the pregnancy, each participant unmistakably experienced their pregnancy and as real, and their anencephalic child as a person.

**Reframing reality.** Parents in this study were required to reframe their new reality as they struggled to exist and function in the wake of perinatal loss. Parents’ sense of self dissolved, introducing the need to understand their new identity, particularly their identity as a parent. Parents without living children questioned their identity as a parent, and all parents were unsure how to answer questions such as, “do you have any children,” or “how many children do you have.”

> For me it was about understanding that this was her journey just like my journey was to me to my parents. And she wasn’t meant to live forever. She was meant to live for those five hours. And, you know, my job was just to make sure that, ah – that I carried her to -- you know, to the altar until – you know, where God took her home. (Tim)

> You will always be a mother, that’s what I tell women. (Chloe)

Many parents experienced difficulty reestablishing intimacy and relationships with spouses and children. Miriam continues to struggle nine years after her loss to hug and kiss her living daughter. Michelle and Gianna are no longer married to the fathers of their anencephalic children. Tim and Forget-me-not no longer attend church, despite its large role in their life before
losing their daughter. Many social relationships were dissolved as parents struggled with the lack of understanding they received from others.

It made me want to avoid certain situations, just because I knew people would, and I think people genuinely feel bad for that situation, but you know, aren’t always capable of responding in a way that would be helpful, or like, comforting. (Kim)

I started to sever relationships with people that would say snotty things to me. (Sophia)

However, many parents reported growing closer to their partner during this time. Sophia stated feeling nothing could shake her marriage after having gone through an anencephalic pregnancy together. Amy and Michael grew closer together as they coped through a nightly ritual of talking about their day while Amy was pregnant with their daughter. Dan and Rebecca, despite the posttraumatic stress and disassociated state in which they existed for years after the loss of their son, have reestablished normalcy in their marriage and lives. As others sought to establish balance and normalcy, many became intentional about reframing their experience by making memories, creating positive associations, and helping others in similar situations.

You know where this is going to end, with your child dying. So we kind of made the decision, my wife and I to try and give us some sense of purpose, to basically experience all of these things, and live very intentionally with the time that we had with our daughter. (Michael)

We’re gonna donate his tissue and organs. I’m gonna try to prevent any other parents from losing a child. Because it’s – it’s a horrible thing to feel that you know that your child isn’t going to live. But you want to spare someone that pain. (Elaine)

But I try not to get mad. I try not to feel all the negative emotions. If you don’t, it will eat you alive. (Chloe)

But we centered around joy. (Forget-me-not)

And so, you know, if my story in any way will help any other parents to, not make it better, but to at least know that, you know, um, there are, in the years – it will always hurt -- it will always hurt, but not with the same intensity that it did. (Miriam)
Parents struggled with the many emotions they faced throughout their experience and had difficulty understanding how to reenter society after the birth and death of their child. Audrey and Michelle quit their jobs after coworkers continued to expressed judgement rather than empathy and understanding that the women were unable to move beyond their loss in a timeframe consistent with societal expectations. Michelle and Lucero expressed extreme emotional difficulty in returning to work promptly after the loss of their children, as neither were granted maternity leave. Women who were granted maternity leave were grateful to have an extended mourning and adjustment period, though the eventual return to work was very difficult. Of the fathers in this study, only Michael was granted paternity leave. Additionally, social triggers were a significant barrier to functioning in daily life following the completion of the pregnancy.

We were eating dinner in a restaurant and someone was celebrating a birthday, and I just broke down and started sobbing. (Gianna)

That kind of triggered – it kind of set me back again. I started going through a lot of the anger and the grief and the horrible stuff that I’d gone through. (Sophia)

I feel like I definitely felt a huge triggers for months after that. Like I can remember being, um, a huge issue with shopping. I don’t know why. Um, that was a trigger. One time I was at the store trying on clothes, and I like totally freaked out in the store. And there was a mom in the – the dressing room next to me nursing her baby, which was just so hard. (Rebecca)

Milestones were also difficult for parents, especially as society commonly recognized neither the parents’ grief, nor the meaningful markers associated with their loss. Parents who lost a twin to anencephaly stated that every milestone their surviving twin achieved was a reminder of the child they lost. Michelle, Lucero, and Maria, all of whom were unable to carry their pregnancies to term, struggled as the due date came and passed. Holidays, birthdays, and anniversaries of “D-day” are accompanied with varying levels of sadness and grief regardless of
the time which has passed since the loss of each child. Though time may cause friends and
family to overlook or forget parents’ grief, parents continued to experience milestones as painful
reminders of the child they do not have.

The milestones are [the] hardest because you watch your five-year-old go to kindergarten
and she’s going alone, not with her brother. (Elaine)

We celebrate his birthday all the way through to the day he passed every year. (Delia)

The due date was the hardest, which was the first really hard day when the due date came
and went and you’re like, well, I’m supposed to have a baby right now, and I don’t.
(Michelle)

It's been six years, and I'm still not okay. Every year, her birthday is really hard, it's really
emotional. (Christiana)

That’s not saying I didn’t cry every day, or think about him every day, or monthly
anniversary, the fifth of every month, thinking, well if you were here, this and this. And
the holidays were really hard. (Kim)

Perinatal death interrupted parents’ understanding of how the world ‘ought’ to be. Parents
in this study subscribed to the underlying societal assumptions that pregnancies are surrounded
by happiness and celebration, parents will die before their children, and ‘bad things’ only happen
to ‘bad people.’ Many negative emotions were introduced with the lifeworld collapse following
the terminal diagnosis of anencephaly. Some of the many emotions parents struggled to process
as they attempted to reconcile with their new reality included guilt, anger and unfairness.

Don’t feel guilty for how you feel and don’t feel guilty for your decisions. (Laura)

I was growing this baby and something happened in my body that killed my baby and
didn’t help that baby grow and be perfect and healthy. And it’s really hard. It makes you
feel like a complete failure. (Michelle)

I remember driving to our appointment with the specialist and it just didn’t seem fair that
the rest of the world was going on with their day. (Forget-me-not)

People want to compare other people’s grief. Which is unfair because, you know, people
would come and say, ‘Well, you know, she’s better off,’ or, ‘At least you didn’t have a
room prepared for her,’ or, um, ‘At least you weren’t attached to her and had her for three or four years and she passed away.’ (Laura)

I would get so mad at god, asking him why he took my baby when I would be such a good parent, I would have loved her so much. (Christiana)

Me and my husband started to resent every pregnant woman that we saw and we became people that would – we would say, ‘Why are they getting to have a healthy baby and not us?’ And – and, you know, it was just horrible. (Sophia)

In addition to the many difficulties in reframing the new reality, parents struggled to learn how to live with the long-term nature of their grief. Rebecca stated she was in a disassociated state for about a year after the death of her child. Ten years later, Dan cried heavily throughout the interview. Sophia developed severe social anxiety and required years of pharmaceutical intervention. Samantha, Sophia, Maria, and Rebecca have very little memory of the year following the loss of their child. Michael shared that he will always carry his grief with him. Chloe stated that the grief never goes away, but the griever becomes stronger, better able to live with the grief. The intensity and long-term nature of grief directly contrasted with both parental and societal expectations and assumptions. Nonetheless, all parents but Michelle reported being a stronger and better person for having gone through their experience. Those who were able to carry their pregnancy to term steadfastly held to their decisions, stating they would not change it if they could. Undergoing perinatal loss established a new benchmark for pain and suffering, and parents reported feeling confident they would be able to survive anything life could throw their way in the future.

**Perinatal Grief Intensity Scale**

After analyzing pilot study findings, it was determined by the research team that while interpretive phenomenology was appropriate to explore parents’ the lived experience of a pregnancy complicated by anencephaly, it was not sufficient to fully to explore the impact of
experiencing specific phenomena associated with an anencephalic pregnancy. The research method was revised to a mixed methods approach and included the administration of the PGIS after conclusion of each participant interview.

The PGIS is a valid and reliable screening tool for clinicians to assess the risk of women experiencing perinatal loss of developing intense grief (Hutti et al., 2017). The use of this psychometric tool facilitates the identification of common experiential components and themes which may place parents at an increased risk for developing intense grief and the associated adverse psychosocial outcomes, such as depression, anxiety, posttraumatic stress disorder, substance abuse, and suicidal ideation. The scale is comprised of three subscales, which measure the reality of the pregnancy, the congruence between maternal desires and reality which transpires in the overall pregnancy experience, and the ability of the woman to confront others perceived as being insensitive. While this tool has only been validated in female Caucasian populations, it is recommended for all persons experiencing perinatal grief regardless of gender or ethnicity (Hutti et al., 2018).

An overall score of greater than 3.535 indicates intense grief. In total, 18 participants (75%) experienced intense grieving reactions (male=2 [50%], female =16 [80%]). Of the subscales, 100% of participants experienced the pregnancy as real, 54.2% reported congruency throughout the pregnancy experience, and 33% felt able to confront others they perceived as being insensitive.

**Data Synthesis**

Correlation coefficients of PGIS scores were separately calculated with demographic variables and qualitative themes in SPSS25. Variables indicator were used to label the presence, absence or omission of interpretive themes (see table 4.2). Though the sample size (n=24) was
not large enough to generalize statistically significant relationships, some general associations can be described from the correlation coefficients. Demographic and thematic characteristics associated with intense grieving reactions are discussed separately below.

**Correlation (demographic data).** No demographic characteristics were significantly associated with total grief intensity scores; however, total PGIS scores were weakly negatively correlated with population density (i.e., rural, suburban, urban) (−.219) and weakly positively correlated with the relationship status (.270), and educational level (.245) at the time of the anencephalic pregnancy. The associations between grief intensity scores and demographic data suggests that of the variables examined, age and educational level of the parent may negatively influence the intensity of the grieving reaction, while the population density may be protective.

Of those with a score indicating an intense grieving reaction, 33% did not have a college degree, and of those, 50% reported having only a high school diploma. Similarly, 39% of those with intense grief scores lived in a rural setting at the time of their anencephalic pregnancy and reported having to drive two or more hours to weekly prenatal appointments. Of the six participants whose PGIS score did not indicate an intense grieving reaction, only one participant lived in a rural setting, while the others lived in suburban (n=4) and urban (n=1) areas. This suggests that living in an urbanized setting with increased access to healthcare services may lessen grief intensity experienced with perinatal loss. Gender, ethnicity, current relationship status, and the number of pregnancies prior to anencephaly did not appear to be strongly related to grief intensity. Of note, the amount of time which has elapsed since the pregnancy and subsequent loss was not associated with decreased grief.

When comparing demographic characteristics with PGIS subscales, Reality was noted to be moderately positively correlated with education (.415, p=.043), Congruence was moderately
positively correlated with income (.573, \( p=0.003 \)), and Confront Others was moderately negatively correlated with being in a relationship at the time of the anencephalic pregnancy (-.414, \( p=0.044 \)). Additional non-significant associations included a moderate correlations between Congruence and age (-.403, \( p=0.051 \)), population density (.342) and time which has passed since the completion of the pregnancy (-.383).

**Correlation (qualitative data).** Patterns, themes and subthemes which emerged from the hermeneutic circle were assigned a variable indicator as being present (“1”), absent (“-1”), or omitted (“0”) in each participant’s interview. The presence, absence, or omission of each qualitative theme was compared to the respective grief intensity score using a Pearson correlation coefficient in SPSS25.

Grief intensity was significantly negatively correlated with parents feeling they had control over their care (-.475, \( p=0.019 \)). Variables significantly associated with a sense of control over their care included being provided with options regarding pregnancy and treatment decisions (.507, \( p=0.011 \)), supportive specialists (.475, \( p=0.019 \)), OBs (.642, \( p=0.001 \)), clinic (.483, \( p=0.017 \)) and hospital staff (.577, \( p=0.003 \)), and patient-centered care (.753, \( p<0.001 \)). Non-significant but meaningful findings of grief intensity included weakly negative associations with bereavement support (-.270), patient-centeredness (-.235) and follow up care (-.221), indicating these practices are associated with less intense grief surrounding perinatal loss resulting from anencephaly.

Interestingly, intense grieving reactions were weakly positively correlated to hospice care (.300) and receiving education (.310) at the appointment during which the diagnosis of anencephaly was communicated, indicating these variables were not associated with lower grief intensity. While education was positively correlated with providing options (.462) and supportive
clinic staff (.642), all participants sought information from the internet and online support groups, suggesting the education provided at the time of the anencephalic diagnosis was not received as positive by parents, or was not sufficient for parents to make an informed decision. The communicative stances of healthcare providers was frequently described as “cold” and “detached” by the majority of patients, with only five (20.8%) participants (including one married couple) reporting positive and compassionate communication at the time they received a diagnosis of anencephaly.

Of the subthemes, Reality was moderately positively correlated with supportive obstetric providers (.426, \(p=.038\)), control over care (.712, \(<.001\)) and community support (.534, \(p=.007\)). Congruence was moderately positively correlated with being provided options (.484, \(p=0.17\)), and supportive obstetric specialists (437, \(p=033\)), hospital staff (.445, \(p=.029\)), family (.443, \(p=.030\)) and community (.569, \(p=.004\)). Confront Others did not show any significant associations with the qualitative themes. Non-significant associations moderately associated with Reality included perinatal palliative care referrals (.383) and familial support (.331).

**Summary**

Heidegger wrote of an ontological death, in which an event transpires to cause a total collapse of meaning; a collapse of an individual’s *lifeworld*. The meaning which previously allowed an individual to understand and make sense of the world is suddenly absent. Without it, “one’s way of being or identity slip away because the familiar world—that is the basis of which I understand who I am – has collapsed into meaninglessness” (Aho, 2016, p. 56). Interpreting parents’ narratives regarding their experience with anencephaly through the lens of Heidegger’s philosophy of an ontological death allows a deeper understanding of what it means to live with perinatal death related to anencephaly. Observing that which transpires following ontological
death in the wake of perinatal loss facilitates an understanding of the subsequent discursive framework individuals construct to once again give meaning to their experience, and ultimately, their identity (Aho, 2016).

Receiving an intrauterine diagnosis of anencephaly is a traumatic experience in which parents find themselves in a state of ontological death. The silence surrounding perinatal loss from anencephaly results in stigmatization and societal expectations of parental grief, failing to acknowledge the reality and personhood of the anencephalic child. This further potentiates parents’ grief, leading to isolation and disenfranchisement. After the completion of the pregnancy, parents struggled with new intensities of grief as they attempted to reframe the new reality in which they found themselves and recreate their identity in the wake of total world collapse. It was further found that parents experiencing perinatal loss required a compassionate, patient-centered approach to care, in which they are able to maintain control over their treatment and care decisions. The observed correlations supported these qualitative interpretations, as control over care was significantly associated with lower grief scores.

Each parent, despite the intensity of their PGIS score, continues to grieve the loss of their child, be it three months or more than twelve years later. Perinatal loss presents an ontological death through the collapse of previously understood or taken for granted meanings of what it to be pregnant. Examining the experience of parents who have had a pregnancy complicated by anencephaly through the lens of an ontological death – and more importantly, how parents reconstruct meaning from this experience – may offer invaluable insight to healthcare professionals caring for those experiencing perinatal loss.
<table>
<thead>
<tr>
<th>Table 4.2: Qualitative Subthemes with Variable Indicators</th>
</tr>
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<tr>
<td>Pseudonym</td>
</tr>
<tr>
<td>-----------</td>
</tr>
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<td>Samantha Evans</td>
</tr>
<tr>
<td>MacEvans</td>
</tr>
<tr>
<td>Forget-me-not Jones</td>
</tr>
<tr>
<td>Tim Jones</td>
</tr>
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</tbody>
</table>
CHAPTER V

Discussion

This study was a mixed methods approach to exploring the impact of the lived experience of a pregnancy complicated by anencephaly on parents. Twenty-four parents participated in individual, open-ended interviews, after which they completed the Perinatal Grief Intensity Scale (PGIS) and a demographic questionnaire. Interview transcripts were interpreted in a Hermeneutic Circle using interpretive phenomenology and viewed through the lens of Heidegger’s ontological death. Each member of the interdisciplinary interpretive team had backgrounds in nursing and psychology, bringing expertise in labor and delivery (L&D) and neonatal intensive care, palliative care, trauma recovery and resiliency, interpretive phenomenology, and mixed methods research. Subthemes of the overall patterns (refer to table 4.2) which emerged from the Hermeneutic Circle were labeled with a variable indicator. Associations were assessed between PGIS scores, qualitative subthemes and demographic characteristics using a Pearson’s correlation coefficient in SPSS25.

Findings from this study suggest the diagnosis of anencephaly is a traumatic event which overwhelms parents’ previously established coping skills. Throughout the perinatal period, parents needed a patient-centered approach from supportive and empathetic care providers. All parents in this study embraced the personhood of their anencephalic child, focusing on living with intention through actively parenting their child in utero and making memories. Despite the reality of the pregnancy and anencephalic child to each parent, parents were met with a societal silence which stigmatized their grief and experience. Reengaging in society and roles such as parent, spouse, employee, and friend, was difficult for many parents, who struggled with how to
reestablish intimacy and relationships with others. Parents found themselves grieving deeply for their child, struggling with how to process guilt, blame, sorrow, and unfairness. Many women talked about social triggers which reintroduced trauma, such as seeing others with healthy babies. Such encounters could be so devastating that women would reorganize their routines and lives to avoid the possibility of triggering intense and disruptive emotional responses. Parents rarely received the bereavement support they needed, and only one woman was provided follow up care after her loss.

This study shared many similarities with previous perinatal grief and bereavement studies. The reality of the pregnancy, feeling validated when healthcare providers are respectful to both the mother and the developing fetus, and the desire to not be treated as a “non-viable” pregnancy, are consistent with prior research (Côté-Arsenault & Denney-Koelsch, 2011b; O’Connell, Meaney, & O’Donoghue, 2019). Experiencing silence and disenfranchisement throughout the pregnancy and loss experience has been described (Kelley & Trinidad, 2012; Rowlands & Lee, 2010; St John, Cooke, & Goopy, 2006). The trauma of receiving a life-limiting diagnosis in utero has also been previously identified (Aite et al., 2011).

Perinatal loss research has expanded to include fathers’ experiences (Armstrong, 1999; McCreight, 2004; Samuelsson et al., 2001). However, “recent research on the impact of pregnancy loss on male partners has usually identified fathers within the context of a supporting role for the mothers, while grief is assumed to be a predominantly maternal domain” (McCreight, 2004, p. 328). Despite this perception, 50% of the fathers in this study experienced an intense grieving reaction. Additionally, Mac’s overall PGIS score (4.02) was the highest of all participants, and Dan developed posttraumatic stress disorder (PTSD) after the birth of his son.
While many of the findings from this study are in agreement with perinatal loss literature, several new findings emerged. To date, this is the first study to measure and report perinatal grief intensity of fathers. Additionally, this is the first study to measure residual postnatal grief in comparison to common experiential themes. Thus, the synthesized findings provide new insight to perinatal loss, grief, and bereavement. Placing parents in control over their care was associated with less overall intense grief, while patient education immediately following the diagnosis of anencephaly was associated with more intense grief. Interestingly, decreased grief intensity was not associated with the passage of time. Fifty percent of the parents whose PGIS score indicated intense grief experienced anencephaly five or more years prior to enrolling in the study. Lastly, 75% (male = 50%; female = 80%) of the parents in this study experienced intense grief. Previous perinatal loss research, which reports that 25% of women who experience perinatal loss experience intense grief, does not report statistics of male perinatal grief (Hutti et al., 2017). While these results are not generalizable, it is worth mentioning the substantially increased percentage of intense grief in the study population as compared to general perinatal loss.

Findings from this study illuminated several key areas which require further investigation. Concepts such as communication, grief theory and language, and perinatal palliative care standards require a deeper examination in comparison to the narrative experiences shared by the study participants. As the majority of healthcare professionals interacting with and caring for the parents in this study appeared to be unfamiliar with anencephaly, it is necessary to understand why this gap in knowledge exists. Of note, key aspects of the participants’ experiences directly contrasted with information provided from the physicians overseeing their care. The characteristics and physiological processes assumed to be known about an anencephalic fetus, such as the fetus will be deaf and only feel pain, were inconsistent with
parents’ experiences. The live neonates responded to noise, suckled, reacted to physical stimuli, and were comforted by touch. Additionally, several women were told their lives would be in danger should they continue the pregnancy, and others were informed they would not be able to become pregnant for several years if they did not terminate the anencephalic pregnancy. However, 35% of the women who carried their pregnancy to term had no complications. Complications which did arise included gestational diabetes, polyhydramnios, shoulder dystocia, and failure for labor to progress. Two women experienced extraneous complications (postpartum hemorrhage requiring an emergency hysterectomy and an amniotic emboli resulting in disseminated intravascular coagulation and a myocardial infarction) which cannot be attributed specifically to anencephaly. Educational curriculum in nursing and medical pre-licensure training may be the first introduction healthcare professionals encounter regarding perinatal loss. Thus, it is necessary to evaluate the content provided in such training curricula, whether they are pre-licensure, residency, fellowship, certification, or continuing education courses. Each of these aspects will be discussed in turn throughout this chapter, as well as implications for practice, policy and future research.

**Pregnancy & Birth Characteristics Summary**

Of the 20 women in this study, 17 pregnancies were carried to full term, one miscarried at 25 weeks, one pregnancy was terminated at 12 weeks, and one woman elected to have a hysterectomy at 16 weeks related to complications with placenta previa from prior cesareans. The primary pregnancy complications experienced by the women in this study were polyhydramnios (20%), gestational diabetes (10%) and failure for labor to progress (15%). Other complications included postpartum hemorrhage which required a blood transfusion and surgical intervention (n=1) and an amniotic emboli with subsequent myocardial infarction and
disseminated intravascular coagulopathy, requiring an emergent caesarian and hysterectomy (n=1). Seven women (35%) reported no abnormal symptoms throughout their pregnancy or labor. Pregnancy characteristic are outlined in table 5.2.

Ten of the 18 (56%) pregnancies carried to term resulted in a live birth. Of those, nine neonates lived for more than one hour, and four lived for more than one day. Contrary to the medical assumptions of the care providers, the neonates who survived birth demonstrated distinct characteristics common in healthy neonates. All live neonates responded to noise, demonstrated startle reflexes with loud noises, and turned towards parents’ voice. Several neonates were able to suckle and were bottle fed. Neonates who lived long enough to be discharged home had functioning urinary and gastrointestinal organs, as evidenced by soiled diapers. Parents also reported the “normalcy” of their anencephalic neonate regarding communicating needs (i.e., crying). One woman reported her son liked to be held, and several participants observed comforting gestures, such as touch, rocking, cradling and feeding to be effective in calming their neonate. Sarah, a family practice physician, had 28 days with her son and was amazed by the normalcy of her neonate, which directly challenged her medical understanding of the neonatal characteristics of anencephaly. According to parents’ experiences reported in this study, current scientific understanding of anencephaly is challenged, requiring further investigation.

Communication

Pediatric specialists at Stanford University of Medicine have begun researching the contextual influence of language when delivering bad news to parents of critically ill pediatric patients. In a news story featured in Hospice and Palliative Nurses Association, pediatric fellow Dr. Mills stated, “The way we communicate, the words we choose, the tone we use and the emotion we’re
<table>
<thead>
<tr>
<th>Pseudonym</th>
<th>Age</th>
<th>Time Since Loss</th>
<th>Gestational Age at Diagnosis</th>
<th>Complications</th>
<th>Length of Labor</th>
<th>Delivery Method</th>
<th>Delivery Outcome</th>
<th>Age of Deceased</th>
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<tbody>
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<td>Dolores</td>
<td>30-39</td>
<td>4 years</td>
<td>20 weeks</td>
<td>Miscarriage 25 weeks</td>
<td>25 hours</td>
<td>Vaginal</td>
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<td>Sandy</td>
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<td>9 years</td>
<td>17 Weeks</td>
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<td>C-Section</td>
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<td>Forget-me-not Jones*</td>
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<td>6 years</td>
<td>20 weeks</td>
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<td>n/a</td>
<td>Scheduled C-Section</td>
<td>Live</td>
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<tr>
<td>Gianna</td>
<td>18-29</td>
<td>2 years</td>
<td>12 Weeks</td>
<td>Gestational Diabetes, Polyhydramnios, amniotic fluid leak</td>
<td>7 hours</td>
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<td></td>
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<td>Live</td>
<td>6.5 hours</td>
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<tr>
<td>Elaine</td>
<td>30-39</td>
<td>5 years</td>
<td>14 weeks</td>
<td>Amniotic Embolism, MI, DIC, Emergency Hysterectomy</td>
<td>n/a</td>
<td>Emergency Hysterectomy</td>
<td>Live</td>
<td>29.5 hours</td>
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<tr>
<td>Christiana</td>
<td>40-49</td>
<td>6 years</td>
<td>12 Weeks</td>
<td>Breech</td>
<td>n/a</td>
<td>C-Section</td>
<td>Live</td>
<td>1 hr 22 min</td>
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<tr>
<td>Marie</td>
<td>40-49</td>
<td>5 years</td>
<td>19 Weeks</td>
<td>Shoulder Dystocia</td>
<td>Pushed 6 hours</td>
<td>Vaginal</td>
<td>Stillborn</td>
<td></td>
</tr>
<tr>
<td>Cloe</td>
<td>18-29</td>
<td>3 months</td>
<td>13 Weeks</td>
<td>Failure to Progress</td>
<td>48 hours</td>
<td>C-Section</td>
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<td>Weeks</td>
<td>Conditions</td>
<td>Outcome</td>
<td>Duration</td>
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<td>Samantha</td>
<td>12</td>
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<td>Twin - hospitalized for premature labor; polyhydramnios</td>
<td>Live</td>
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<td>Evans*</td>
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<td>40wk 5d</td>
<td>Postpartum Hemorrhage</td>
<td>Vaginal</td>
<td>Stillborn</td>
<td></td>
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<td>9</td>
<td>12</td>
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<td>Termination (D&amp;C)</td>
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<td>Lucero</td>
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<td>C-Section</td>
<td>Live</td>
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<td>Rebecca</td>
<td>30-39</td>
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<tr>
<td>Johnson*</td>
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<tr>
<td>Miriam</td>
<td>9</td>
<td>20</td>
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<td>Vaginal</td>
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<td></td>
<td></td>
<td></td>
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<td>Sarah</td>
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<td>Vaginal</td>
<td>Live</td>
<td>28 days</td>
<td></td>
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<td>Amy Smith*</td>
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<td>20</td>
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<td>Stillborn</td>
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<td>Kim</td>
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<td>20</td>
<td>None</td>
<td>Vaginal</td>
<td>Live</td>
<td>9 Hours</td>
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</table>

*Indicates participants’ partners also participated
able to get across can completely shape the family’s perspective and their experience. That is, to me, as powerful as anything else we do, because this is an event, a time in the parent’s life, that’s going to be remembered forever” (Hannon, 2020). Drs. Magnus and Batten have found that when physicians attempt to use layman’s terms when communicating important diagnostic and prognostic information to parents, parents and physicians hold differing underlying assumptions of words and their meanings. A breakdown in communication follows, in which the parent does not interpret the physician’s words with the meaning the physician intended. Dr. Magnus suggests that such conversations should be considered “high-stake,” and treated with the utmost priority and respect. Stanford researchers are currently investigating innovative approaches to increase experience and expertise in high-stakes communication in pediatric fellowship programs.

Communication has been identified as integral to the quality of healthcare provided to patients (Lewis & Kitchen, 2010). Unsurprisingly, the quality of communication between parents and healthcare professionals influenced the perinatal care experience of the participants in this research. In particular, parents spent a long time describing ‘D-Day,’ the day they received the diagnosis of anencephaly. This moment in time is deeply engrained in each parents’ memory, and the interaction, communication, and attitude of the diagnosing physician dramatically influenced the severity of their emotional experience. Interestingly, parents who received education regarding anencephaly at the initial diagnostic appointment had higher grief intensity scores than parents who did not receive education. Such findings suggest that the method of education, information being relayed, or communicative stance adopted by the diagnosing physician are not appropriate for communicating a lethal congenital anomaly diagnosed in utero to parents. Studies have yet to explore the impact of the initial diagnostic communication of a
lethal fetal anomaly on parents, though it has been hypothesized that the communicative stance of providers is largely influential on parental experience (Berry & Colorafi, 2019). Indeed parents who did not have a PGIS score indicating intense grief all reported compassionate and respectful communication with the diagnosis of anencephaly, while those whose PGIS scores indicate intense grief all reported poor communication. It is not unreasonable to consider the communication of a lethal or life-limiting congenital anomaly diagnosis as a high-stakes conversation which requires compassion, sensitivity, and expertise.

Grief Language and Theory

Prior research indicates perinatal grief is different from all other forms of grief, and thus requires a different approach to treatment and care (Black, Wright, & Limbo, 2016). As a result, perinatal palliative care programs have been developed to better suit the needs of parents experiencing perinatal loss, as traditional palliative care services are not appropriate to meet the needs of grieving parents (Cole et al., 2017; Kavanaugh et al., 2015). Likewise, traditional grief theories have also been identified as inappropriate to describe, treat, and care for grieving parents. Researchers suggest it may not be appropriate for a single theory to explain the complex and multifaceted experience of perinatal loss, grief, and bereavement. Therefore, no single perinatal bereavement theory has been developed. Rather, excerpts from multiple theories have been drawn upon to guide the care and research of grieving parents. It is necessary to evaluate the appropriateness of these theories and the feasibility of a multi-theory approach to perinatal bereavement. Additionally, it is necessary to evaluate the appropriateness of the language of grief used to describe, treat, and care for parents experiencing perinatal loss.

Grief language. As in other pathologies, terms such as “acute” and “chronic” are used to describe the timeframe and associated characteristics of grief. Pathological grief is typically
considered a chronic condition in which the adverse and intense emotions associated with acute
grief persist beyond six months after loss (American Psychiatric Association, 2013).
Approximately 12% of bereaved individuals in the general population (outside of perinatal loss)
experience persistent and disruptive symptoms of grief. Pathologic grief does not naturally
resolve with the passage of time, that is, beyond six months after the loss of a loved one
(Flemming, 2020). However, the majority of parents in this study, despite the intensity of the
grief response indicated by the PGIS scores, experienced intense grief and difficulty functioning
in their roles for at least one year. Many experienced difficulties beyond a year after the birth and
death of their child. Furthermore, as anencephaly can be diagnosed as early as 11 weeks
gestational age, parents who chose to carry their child to term experienced complex emotions
associated with anticipatory grief for the remainder of the pregnancy. After the completion of the
pregnancy, the grief experience changed and commonly intensified following the birth and death
of the child. If measuring the timeframe of grief from the initial diagnostic appointment, many
parents in this study experienced intense and complex grief for at least the following two years.
Furthermore, all parents in this study reported deeply missing and grieving the loss of their child
up to the day of the interview, which ranged from three months to over twelve years after the
birth and death or termination of their child.

Much remains to be explored regarding the phenomenon of healthy grieving periods
surrounding perinatal loss. According to the PGIS scores, 75% of parents in this study continue
to experience intense grief, and of those, 94% experienced this grief for more than one year
following the death of their child. Finding from this study suggest that parents experiencing
perinatal loss resulting from anencephaly are at an extremely high risk of developing chronic
grief. The timeframe in which parents were able to reestablish balance in their life and function
in their daily roles varied greatly. Yet, intense emotions and even triggers continued to persist beyond six months after the loss of their child. The overall timeframe would, by traditional Western grief language, be considered pathologic or ‘chronic’ grief. This leads to several questions: Should grief be considered pathological if a parent is unable to recover from the loss of their child in less than six months? Is the term ‘chronic’ appropriate to describe or label the grief many parents continue to experience? Is ‘chronic’ grief abnormal or an expected norm in perinatal loss? When does this ‘chronic’ grieving become unhealthy? What efforts are in place to recognize healthy versus unhealthy grief responses, how commonly are they used, and are they based on sound evidence? What interventions are in place to recognize chronic grief and help ease parents’ transition into ‘normal life’ after perinatal loss, and how commonly are they implemented? At what point should parents feel ‘normal’ again, and is it pathological if parents’ grief never goes away?

**Grief theories.** Theories are especially useful in guiding complex care and explaining human experiences, such as perinatal bereavement. A conceptual definition of perinatal bereavement has only recently been introduced in the literature, the epistemological foundation of the construct remains ambiguous, and no perinatal bereavement theory has been established (Fenstermacher & Hupcey, 2013). Researchers in the field of perinatal bereavement maintain that no single theory is appropriate to guide the care of parents experiencing perinatal death, but rather care should be informed by multiple theories. Several theories have been identified as appropriate to more meaningfully inform research of perinatal grief and bereavement, including attachment theory (Bolby, 1980), psychodynamic theory (Freud, 1917, 1957), cognitive stress theory (Lazarus & Folkman, 1984) and interpersonal theory (Weissman, Markowitz, & Klerman,
2000). Feminist theory is also widely used to inform research exploring perinatal loss (Black et al., 2016).

Bowlby’s (1980) theory of attachment explains the interrelationships between attachment, affection, bonding, separation and loss in human relationships. However, Bowlby’s theory was developed to explain the emotional attachment between children and their mothers, providing a scientific explanation for behavioral disturbances in children. Subsequent researchers expanded on Bowlby’s attachment theory to include prenatal attachment, hypothesizing that maternal attachment begins to form prior to birth (Brandon, Pitts, Denton, Stringer, & Evans, 2009). Attachment theory describes different attachment patterns individuals develop, as well as patterns exhibited with loss, and is therefore considered to be a helpful framework for understanding behavioral patterns associated with perinatal loss (Black et al., 2016). Despite never having physically met the fetus, both the mothers and fathers in this study developed strong bonds and attachments with their anencephalic child in the womb. Parents were intentional about parenting their children in the womb, creating memories, and finding meaning. However, as the attachment and bonds between parents and their unborn child is not a tangible construct which others can experience and observe, others have difficulty understanding the depths of the grief which occurs at the death of the anencephalic child. Consequently, parents endured insensitive comments and judgments from family and friends who misunderstood the depth of the parents’ grief. Many parents learned to hide their grief from others, and felt isolated and alone. Thus, attachment theory is appropriate to inform practice as it illuminates the intrauterine parental bond. However, the majority of research has focused on maternal intrauterine attachment of healthy pregnancies. Further work is indicated to better understand
prenatal attachment in fathers and in pregnancies complicated with a life-limiting congenital anomaly.

Leon (1992) expanded on the original psychodynamic theory, positing four psychodynamic frameworks with which to understand pregnancy loss: developmental, conflictual, object-oriented, and narcissistic. The developmental framework depicts perinatal loss as an interruption to the normal developmental stages in adulthood and parenthood. Similarly, the conflictual framework posits the negative impact on the female psyche when the drive to reproduce is disrupted. The object-oriented framework presupposes a need to detach from the deceased neonate, and the narcissistic framework predicts that feelings of self-blame or a loss of self-esteem may arise in the mother who ‘failed’ to have a successful pregnancy. Three of the four frameworks resonate with the findings of this study. However, it may not be appropriate to encourage parents to sever attachment to their deceased neonate as the object-oriented framework suggests. Parents in this study actively sought to keep the memory of their child alive, many of whom continue to perform ritualistic and symbolic activities decades later, such as celebrating birthdays or other milestones. Contrary to the object-oriented framework, parents seemed to find solace in continuing the bond with their spirit child.

The interpersonal theory (IPT) seeks to understand prolonged depression which may arise from interpersonal struggles throughout the mourning period following the loss of a loved one, and has been found to be helpful in treating post-partum depressions (Weissman, Markowitz, & Klerman, 2000). Subscribers to IPT endeavor to assist the bereaved individual through the mourning period by reestablishing relationships and interests which are meant to take the place of that which was lost. The IPT can also be extended to explain gender differences in grieving processes and intensities, and recognizes men are also impacted by perinatal loss.
Lazarus and Folkman’s cognitive stress theory (1984) observes the cognitive processing and restructuring individuals undergo in response to the stress, discomfort and pain of loss. Such emotional processes are commonly influenced by underlying assumptions such as ‘bad things only happen to bad people,’ or ‘pregnancies are full of happiness.’ Later work by Lazarus (2000) recognized three common responses to stress, namely that of an individual’s assessment, response, and subsequent emotions in response to an event. Individuals who are able to develop positive or helpful coping strategies are postulated to be better equipped in the recovery process over time. The majority of parents in this study struggled with feelings of unfairness, unable to understand why their pregnancy was allowed to end in their neonate’s death. Many also struggles with understanding how to respond to the stress of perinatal loss, particularly as the trauma of the event outstripped previously established coping skills. Thus, the cognitive stress theory may be helpful in framing research questions when exploring grief related to perinatal loss.

Several aspects of the theories listed above do not align with the experiences or needs of parents in this study. Contrary to Leon’s object-oriented framework of the psychodynamic theory, expecting or encouraging parents to sever their attachment with their deceased child may not be appropriate. Parents in this study religiously celebrate milestones, most commonly birthdays. Parents also reported displaying photographs, scrapbooks, and symbolic mementoes, such as butterflies, rainbows, and poppies around their home to keep the memory of their child alive. Parents also reported wanting to talk about their deceased child with others, which served as another form of keeping their memory alive. Miriam, who did not speak of her loss for nine years after it occurred, participated with the intention of sharing her story. She strongly believed her silence has stunted her recovery, and conveyed her gratitude and relief at the conclusion of her interview for being able to openly share her experience. The IPT encourages replacing the
lost child with a new relationship. However, parents who bore healthy children following the anencephalic pregnancy repeatedly stated no one can ever replace the child they lost, and were frustrated with friends and family members who expected this replacement process to occur. With the lifelong impact of perinatal loss on parents, it is vital to ensure the theoretical and conceptual foundations guiding the care of bereaved parents are appropriate. Though each of the theories discussed above has its own merits, the question remains, is it practical to utilize a complex, multi-theoretical approach to perinatal bereavement care? Familiarity with each of these theories and the necessary corresponding interventions requires clinicians to be theoretical experts in grief and bereavement on top of all other care responsibilities. Furthermore, as perinatal bereavement care education and training is often lacking, even non-existent, it may not be practical to expect healthcare professionals to be familiar with a multitude of theoretical approaches. While the theories may appropriately serve as theoretical frameworks for perinatal loss, grief and bereavement research, there remains a need for a theoretical framework guiding the care of bereaved parents experiencing perinatal loss.

Each of the theories described above have been borrowed and adapted to conceptualize perinatal loss. However, while these theories seek to explain cognitive processing and behavioral patterns, and understand why parents grieve the loss of someone they never knew, they do not seek to explain the needs of parents throughout their loss experience. What remains to be explored is what parents need while grounded in their emotional experiences throughout the complex, multifaceted, unfamiliar and world-shattering experience with perinatal loss. A complex, multi-theoretical approach caring for parents throughout the journey of perinatal loss may be inappropriate. Thus, it may be necessary to develop a perinatal bereavement theory using
the theories above as conceptual guiding frameworks, seeking not to explain or describe what happens when a parent experiences perinatal loss, but rather what a parent needs.

**Perinatal Palliative Care Standards**

In the last five years, specialty organizations such as the American College of Obstetricians and Gynecologists (ACOG) and National Association of Neonatal Nurses (NANN) have developed perinatal palliative care (PnPC) standards to guide healthcare professionals in providing appropriate End-of-Life (EoL) care to parents experiencing life-limiting pregnancy or neonatal complications (ACOG, 2019; NANN, 2015). Offering PnPC services to parents whose fetus or neonate has been diagnosed with a life-limiting condition is considered an essential element to holistic care (Rana Limbo et al., 2017). Both the ACOG and NANN recommend referring parents to PnPC resources upon diagnosing a life-limiting fetal anomaly. Though the vast majority of study participants (88%) did not receive PnPC resources, most nurses caring for participants in the L&D wards exemplified compassionate, patient-centered care consisted with NANN recommendations. Nurses endeavored to create memories for parents, acknowledged and treated the neonates as a real person, and allowed unlimited family members to visit the ward. While not all participants had positive experiences, 75% of participants expressed gratitude towards the compassion shown to them by the L&D nurses.

The NANN’s position statement recommends “NICUs should have ready access to all components of palliative and end-of-life care either through their own programs or community organizations” (NANN, 2015, p.1). However, participants who were treated at facilities with NICUs did not receive PnPC referrals or resources. Additionally, participants living in rural areas neither receive nor had access to PnPC or bereavement resources. Such ready access to PnPC services may not be a plausible reality due to limited financial and personnel resources in
smaller facilities or rural locations. Nonetheless, the need for PnPC resources and services are equally as necessary in rural communities as in high population density regions, especially since 39% of participants whose PGIS scores indicated intense grief lived in rural communities. Implementing perinatal palliative telehealth programs may begin addressing this gap in care coverage, and is worth exploring.

It is unclear if PnPC or bereavement resources are available for individuals who opt for termination due to a fetal anomaly (TFA). Michelle, despite the nine years which have passed since her terminated pregnancy, continues to experience depression, intense grief, and regret. Sarah, a family practice provider, stated her experience of having a son with anencephaly gave her a new found understanding for the grief parents must feel after a TFA, which she now applies in her medical practice. Michelle’s grief and Sarah’s recognition of the strong emotions associated with TFA are consistent with previously conducted research (Lafarge, Mitchell, & Fox, 2013). Whether or not parents who chose TFA are candidates for PnPC services, follow up care and bereavement resources may be highly beneficial in the journey towards healing.

The ACOG (2019) concludes their position statement on PnPC recommendations with: “Once a life-limiting condition is suspected antenatally, the tenets of informed consent require that pregnant patients be given information of sufficient depth and breadth to make an informed, voluntary choice for their care” (p.88). They elaborate further, stating physicians should support families in shared-decision making and avoid coercion or undue influence on treatment choices. However, one of the largest causes of distress for parents during their experience with perinatal loss was the communicative stance of the diagnosing physician and the lack of information and options provided upon diagnosis. All participants whose PGIS score did not indicate an intense grieving reaction described the providers who informed them of the anencephalic diagnosis as
kind and compassionate, and did not feel pressured to make one decision over another. As mentioned above, the diagnostic communication of a lethal congenital anomaly should be considered a high-stakes conversation and be engaged in with the utmost of respect and compassion.

Despite recent implementation of PnPC care standards, parents with recent losses did not benefit from PnPC resources. Though Chloe’s loss occurred three months prior to her interview, her care provider did not inform her she had any choice other than to terminate her pregnancy, and when she decided to carry her son to term he informed her he would no longer provide her with care. Tim, Forget-me-not, and Delia had perhaps the most positive experiences with their care providers, and their losses occurred between six and 10 years ago. The absence of care guided by ACOG or NANN PnPC recommendations brings into question the success of disseminating knowledge and evidence-based practice among care professionals. Furthermore, established guidelines are appropriate not only for PnPC specialists, but for all healthcare professionals routinely interacting with and caring for parents experiencing perinatal loss. As the majority of prenatal care takes place in obstetric (OB) clinics, it may be necessary to implement annual specialty training featuring PnPC recommendations and education to better prepare clinic staff to care for parents receiving prenatal care for terminal pregnancy conditions.

The development of PnPC programs has largely improved the care of parents grieving the impending loss of a fetus or neonate (Rana Limbo et al., 2017). Based on the narrative accounts in this study, PnPC recommendations by both ACOG and NANN would have been beneficial to parents as they traversed the unfamiliar journey of perinatal loss. Unfortunately referral rates to such programs and resources remains as low as 11% (Leong Marc-Aurele & Nelesen, 2013). Findings from this study were similar, as only one married couple and two other women were
referred to perinatal palliative care services (12%). Even parents who transferred to larger treatment centers in neighboring cities were not referred to PnPC. Interestingly, none of the parents who were referred to PnPC services received follow up care, despite that being a recommendation of both National Coalition for Hospice and Palliative Care and PnPC recommendations (ACOG, 2019; NANN, 2015; NCHPC, 2018).

Investigative efforts over the last decade have attempted to explore reasons for barriers to PnPC (Wool, 2013a, 2013b, 2015; Wool & Northam, 2011). However, what has yet to be explored is the appropriateness of and adherence to perinatal loss care standards, and the potential benefits of the adoption of, familiarity with, and adherence to such standards in the non-palliative healthcare setting. This is particularly important as the large majority of care is occurring in this setting, without the resources, guidance, and expertise of a multidisciplinary PnPC team. Additional opportunities for improving PnPC in non-PnPC settings include nursing and medicine curriculum, residency programs, and perinatal fellowships. Furthermore, parents in this study encountered and interacted with a variety of professionals in the obstetric setting beyond nurses and physicians, including receptionists, laboratory and diagnostic imaging technicians, medical assistants, midwives, and doulas. As the majority of care parents received in this study was provided in obstetric settings and clinics, it may be necessary to evaluate care education, standards and practices relating to perinatal loss in these care environments.

Perinatal Grief Intensity Scale

Inclusion of the PGIS in this study offered unique insight into the impact of a pregnancy complicated by anencephaly on parents. A retrospective screening of individuals at risk for intense grief following perinatal loss permitted the research team to observe participants experienced prolonged adverse psychosocial and emotional responses and those that did not.
Based on the narrative accounts of parents, all those whose PGIS score indicated an intense grieving reaction did indeed experience prolonged and intense adverse emotional and psychosocial symptoms, primarily depression, anxiety, PTSD, dissolution of relationships, and suicidal ideation. However, two women whose PGIS score did not indicate they were at risk for an intense grieving response experienced adverse psychosocial outcomes. Miriam (PGIS score = 3.44), a Latina woman whose loss experience occurred nine years prior to her interview, continues to struggle with intimacy with both her husband, from whom she was separated for an extended period after the birth and death of her anencephalic daughter, and her living daughter. She stated in her interview that to this day, hugging her living daughter is such a struggle that she is rarely affectionate towards her. She also stated that she had never spoken of her experience with anencephaly to anyone before her interview, including her husband. Sophia (PGIS score = 3.46), a Caucasian woman whose experience occurred 10 years ago, gained 100 pounds and developed severe social anxiety after the birth and death of her son. She became unable to leave her home and deeply struggled interacting with friends and family. After several years of isolating herself to her home in fear of experiencing social triggers, she sought professional help and was prescribed antidepressant and antianxiety medication. After nearly five years of pharmaceutical intervention, Sophia was able to return to work and leave her home again.

The PGIS was originally validated primarily on women experiencing miscarriage. Though only two (8%) of the study participants whose PGIS score contradicted their narrative account, it is reasonable to question whether or not congenital anomalies introduce an unknown indicator for intense grief. As part of the demographic questionnaire administered in this study, participants were asked of there were any items on the PGIS which did not make sense, seemed inappropriate, or were missing, to which all participants answered ‘no’ or left blank.
Additionally, participants were not instructed to answer the PGIS questions according to how they felt at the time of their anencephalic pregnancy or how they felt currently. Six participants were later contacted and asked how they answered the questions (past versus present), and if their answers would change if they answered according to one timeframe or another. Only one woman, who answered how she felt at the time of her anencephalic pregnancy, said she would have changed her answer according to a timeframe. Having later become a nurse, she stated she would have changed her answer regarding the nursing care she received with the knowledge and experience she had recently gained in her nursing practice.

The PGIS is, however, a very useful clinical screening tool, the use of which is highly beneficial in identifying persons at risk for intense grieving reactions following perinatal loss. Increased use of the tool in clinical settings such as diagnostic imaging, emergency departments, and prenatal care clinics would enhance healthcare professionals’ ability to identify parents at risk for intense grief who would benefit from immediate intervention and allocation of bereavement resources. Further research regarding the validity and reliability of the PGIS in predicting men at risk for intense grief, as well as those of a more ethnically and socioeconomically diverse backgrounds is needed.

**Strengths and Limitations**

This study has many strengths. Heidegger’s philosophy of an ontological death was uniquely appropriate for exploring the meaning of parents’ lived experience of an anencephalic pregnancy. A strong, multidisciplinary team with backgrounds in nursing and psychology, both male and female, consisted of experts in L&D and neonatal intensive care, palliative care, trauma recovery and resiliency, interpretive phenomenology and mixed methods research. The study sample included a diverse array of ethnic, geographical, educational, and socioeconomic
backgrounds from both the mothers’ and fathers’ perspectives. Furthermore, the inclusion of experiences which happened from three months to over twelve years prior allowed a cross-sectional retrospective analysis of loss experiences, changes in perinatal practice over time, and both long- and short-term parental responses to grief. Using a mixed methods approach further allowed the research team to assess the impact of common experiential components parents experienced throughout the perinatal loss journey, which would not have been identified using interpretive phenomenology alone.

There were also several limitations in this study. The PGIS was validated in populations consisting of predominantly educated, middle class Caucasian females experiencing miscarriage. However, in this study it was used to measure grief intensity in a population which consisted of male, female, Caucasian and LatinX participants from diverse socioeconomic and educational backgrounds. Additionally, while the sample size was large for an interpretive phenomenological study, it was too small to detect all but the most robust correlations. It did, however, introduce new knowledge and generated hypotheses, particularly regarding the impact of the communicative stance of providers relaying the initial diagnosis of anencephaly. Lastly, anencephaly.info, the primary online site from which participants were recruited, is dedicated to parents who chose to carry their anencephalic pregnancy to term. Recruitment materials were shared with other online anencephaly support groups by members of anencephaly.info, and two participants who opted not to carry their pregnancy to term were a part of this study. Furthermore, parents who voluntarily seek community support demonstrate engagement in their recovery, and may not be an accurate representation of all parents experiencing perinatal loss. While this is not contraindicated for the interpretive phenomenological method chosen in this study, it merits note as parents who are not as engaged in their recovery may possess different
thematic experiences than those identified from this self-selected study population. Interestingly, 100% of individuals requesting to take part in this study in response to recruitment materials were women. The four men who did participate were specifically asked to participate by either the PI or their partner. One woman stated her husband thought the idea of the study was interesting, but was deeply opposed to participating. Of the 76 women who responded, six male partners expressed interest in participating, though only four volunteered to enroll in the study.

**Implications for Practice**

The findings in this study provide further evidence upon which to build best practice recommendations for parents experiencing perinatal loss related to anencephaly (see table 5.4). Parents experiencing a terminal pregnancy, whether they choose to carry to term or terminate, have the right to receive prenatal care consistent with care that is provided to a healthy pregnancy. Such care includes patient-centeredness, shared-decision making, bereavement support, implementing practice policies and procedures, and follow up care. Seven practice recommendations are presented based on the findings of this study.

Perhaps the greatest barrier to patient-centered care that parents experienced in this study were the attitudes of healthcare professionals. Therefore, the first recommendation is that of increasing awareness of the *reality* of the terminal pregnancy parents may experience. Stated another way, it is important for healthcare professionals to understand that many parents consider an anencephalic fetus to be a son or daughter rather than a ‘non-viable product’ resulting from a malformation of the neural tube in the early stages of development. This is particularly important in the clinic setting, where the majority of prenatal care is provided. Furthermore, according to the synthesized findings from the qualitative themes and PGIS scores, *control over care* was shown to have the strongest association with lower grief intensity. Without question, parents in
this study would have benefitted from patient-centered, compassionate care, over which they were placed in control. Therefore, placing parents in control over their care and treatment decisions in the clinic setting is the second recommendation based on findings from this study. Aspects associated with control over care include patient-centeredness, education, shared-decision making and supportive healthcare professionals. Each of these aspects are consistent with practice recommendations of ACOG and NANN for PnPC practice standards. Sadly, very few of the parents in this study received perinatal care consistent with these recommendations, likely due to the scant PnPC referrals. It may therefore be necessary to integrate the ACOG and NANN PnPC care standards into the routine prenatal care setting. For the parents in this study, healthcare professionals in the prenatal clinic setting were the most frequently encountered healthcare professionals throughout the journey of carrying a pregnancy with a life-limiting condition. Healthcare professionals who adopt an attitude of patient-centeredness which respects the reality of the pregnancy are necessary to provide compassionate, patient-centered care.

All but five parents, including one married couple, were treated by a provider who had never encountered a patient with a pregnancy complicated by anencephaly in the practice setting. Though anencephaly is thought to be a rare birth defect, it affects 4,600 birth in the US annually (CDC, 2017). Thus, it is critical to equip healthcare professionals across the care spectrum with the skills and knowledge necessary to provide holistic, patient-centered and compassionate care to parents experiencing perinatal loss related to anencephaly. The third recommendation for practice is an increase in specialty education and training of healthcare professionals regarding parental needs associated with perinatal loss. Training and education should extend beyond PnPC specialty teams to emergency healthcare services, diagnostic imaging, perioperative services, and obstetric care settings. Moreover, the size of the healthcare establishment is not an
appropriate indicator of the likelihood of encountering parents with an anencephalic pregnancy, thus even small and rural care settings require specialty training and education. It may be necessary to implement annual training courses discussing perinatal loss and associated resources, with heavy emphasis in residency and fellowship programs, whether or not palliative and end-of-life care is the focus of the residency or fellowship training. Furthermore, training and education should be based on high quality evidence-based recommendations. Embedded within the training and education courses should be an emphasis on communication, high-stakes conversations, and the communicative stance adopted by healthcare professionals, as well as available PnPC services and bereavement support. Several programs have been previously established to provide specialty training to healthcare professionals, such as Resolve Through Sharing, and End-of-Life Nursing Curriculum Consortium. Both training curricula focus on providing culturally sensitive, compassionate care to parents experiencing loss.

The level of distress parents in this study experienced with a poorly communicated diagnosis of anencephaly may have set the tone for their pregnancy and recovery experience, potentially even influencing the intensity of the grief experienced. Therefore, the fourth recommendation is that healthcare professionals place extreme emphasis on the critical importance of communication. Furthermore, the diagnostic communication of a lethal or life-limiting intrauterine condition should be considered a high-stakes conversation. Relaying such information is a life-altering event, and parents receiving such a diagnosis require specialized care. Communication which is clear, concise, and not coercive, as recommended by ACOG, should be implemented in such scenarios. As communication is vitally important to high quality care, healthcare professionals should also be provided with the training and mentorship necessary to facilitate high quality communication.
Fifth, it is necessary for each healthcare facility to review and evaluate the presence and appropriateness of practice policies and procedures addressing the care of parents experiencing perinatal loss. Practices such as assisting families in making memories, disposing of remains, and providing follow up care influence both the care experience from the perspective of the parents as well as their recovery process. Like any other chronic condition requiring extensive follow up care, perinatal loss requires equal attention. In addition to the existence and appropriateness of such policies and procedures is the departments in which they are implemented. As mentioned numerous times, perinatal loss is not strictly associated with L&D wards, but may be encountered in emergency and urgent care services, perioperative services, and diagnostic imaging, among others. Thus, it is necessary to ensure all departments which may encounter perinatal loss are prepared and equipped with policies and procedures which are appropriate to provide patient-centered and compassionate care.

The sixth recommendation is to ensure follow up care is provided to all men and women who experience perinatal loss. Of the 24 participants who took place in this study, only one woman, including the 12% of participants who were referred to PnPC services, received follow up care. The remainder of the participants were left to their own devices, seeking to understand how to reframe their new reality and reenter society in the midst of the world-shattering ontological death of perinatal loss. Understanding how to recover after perinatal loss relating to anencephaly is a complex and multifaceted endeavor requiring specialized care. It is necessary to implement programs which actively seek to provide follow up bereavement care to parents. Waiting for parents to seek medical attention is not only unrealistic, but irresponsible on the part of the healthcare professional care team. All healthcare facilities which may come into contact
with persons experiencing perinatal loss need not only a plan for implementing appropriate care to the parents experiencing perinatal loss, but also of actively providing follow up care.

It is a testament to the advancement of modern medicine that society is no longer acutely aware of perinatal, neonatal or pediatric death as a common occurrence. Healthcare professionals in most settings consider perinatal and neonatal death to be a rare event, and commonly encounter such events with great infrequency (Black et al., 2016). Unfortunately, from such triumphs in perinatology and neonatology emerges a lack of awareness, knowledge, confidence, and competency caring for families experiencing perinatal loss. Therefore, the seventh and final recommendation is to increase awareness, both medically and societally, of perinatal loss and the associated adverse psychosocial outcomes. While celebration of the rarity of perinatal loss is warranted, there remains the necessity of awareness, preparation and competency of providing compassionate, patient-centered care to parents who experience perinatal loss relating to anencephaly.

Policy Change

The loss, grief and bereavement resulting from perinatal death are distinctly unique from other forms of loss and grief, and are considered the most traumatic event a parent could experience (Black et al., 2016). As demonstrated through the narrative accounts of parents in this study, perinatal loss, grief and bereavement are rarely recognized and poorly understood by society, both medical and otherwise. Consequently, societal silence regarding perinatal loss disenfranchises parents’ grief through a failure to acknowledge the loss (Kelley & Trinidad, 2012; McCreight, 2008; Rosenberg, 2012; St John et al., 2006). It is necessary to increase awareness of the long-term negative impacts associated with perinatal loss. Legislation in Washington State (RCW 43.70.442), requires licensed healthcare professionals to complete a
suicide recognition and prevention training course to keep an active license (WDOH, 2016). In light of the long-term adverse psychosocial outcomes associated with perinatal loss, it is not unreasonable to require healthcare professionals to complete a specialty training course focusing on perinatal loss. Doing so would increase awareness and knowledge of the unique, multidisciplinary needs of parents experiencing perinatal loss, and be a future referential resource when healthcare professionals come into contact with parents experiencing perinatal loss.

According to the Lunt Group, LLT (2018) *Employee Law Book*, there are no federal regulations mandating employers to provide either paid or unpaid bereavement leave to employees. In 2014, Oregon was the first and only state in the US which has passed a law requiring employers to provide employees with up to 14 days of unpaid bereavement leave (ORS 659A.159). Specifications include the loss of a “child,” but do not use terminology which would indicate that miscarriage or stillbirth are included, nor is the inclusion of the father. Based on the findings from this study, both mothers and fathers are deeply impacted and commonly unable to function after the birth and death of their anencephalic child. The one week allotted to Michelle after the passing of her child was insufficient for her to recover enough to reenter society. Kim’s husband, also given one week, experienced extreme difficulty returning to work. It is necessary to federally recognize the trauma associated with perinatal loss, specifically related to anencephaly, mandating employers to grant extended bereavement leave. Included in bereavement leave for perinatal loss should be the allocation of bereavement resources and support which includes counseling.

Several parents desired to donate the organs of their anencephalic child, but were met with resistance. Each organization has specific regulations and requirements regarding eligibility
criteria for organ and tissue donation. One such requirement is the weight of the neonate. However, neonates with anencephaly are not commonly impacted by additional congenital anomalies, and have intact organ systems apart from the cerebral hemispheres. Furthermore, the absence of posterior and lateral cranial structures significantly impacts the weight of the neonate, causing many viable organs to be ineligible for donation. It is necessary to evaluate current protocols regarding the donation of anencephalic organs and tissue, revising the protocols as necessary according to scientific findings.

**Future Research**

Not only is it necessary for healthcare professionals to be familiar with anencephaly, the associated unique care needs of the parents, and recommendations for evidence-based practice, it is also necessary that the state of the science regarding anencephaly accurately reflects the physiological nature of the pregnancy, fetus and neonate, as well as the experience of the parents. Much remains to be explored concerning both anencephaly and the needs of parents experiencing a pregnancy complicated by anencephaly. Throughout this chapter, seven aspects of the perinatal loss experience requiring further research have been identified (see table 5.5).

First, the information regarding anencephaly and an anencephalic pregnancy conveyed to the majority of parents in this study, namely the adverse pregnancy related complications and characteristics of the anencephalic neonate, was found by parents to be inconsistent with their own personal experience. Parents frequently felt the diagnosing physician was using scare tactics to coerce them into terminating the anencephalic pregnancy. While this cannot be confirmed with any certainty, it is important to assure the scientific information healthcare professionals’ reference is based on sound scientific research rather than myth, speculation and anecdotal evidence. If indeed the information conveyed to the parents in this story was based on medical
training and education, it is necessary to reevaluate our understanding of the physiological processes of a pregnancy complicated by anencephaly and the characteristics of an anencephalic neonate.

Second, perinatal loss research has focused predominantly on mothers, assuming fathers, functioning in the support role, are not as affected by the perinatal loss experience (McCreight, 2004). The fathers in this study, however, were deeply impacted by the anencephalic pregnancy of their partner, many of whom struggled or continue to struggle with intense grief years after the birth and death of their child. It is necessary to further explore fathers’ experiences with perinatal loss relating to anencephaly and other lethal congenital anomalies. Identifying the needs of fathers will then allow the development of appropriate grief interventions, support, and resources. Any support and resources which are currently available require evaluation to ensure the needs of fathers are appropriately met. Additionally, as perinatal loss has traditionally been considered a predominantly maternal concern, theoretical frameworks exploring perinatal loss have also focused on the maternal experience. It is therefore necessary to develop a theoretical framework which seeks to describe, explain, and guide the care fathers may require throughout their journey of perinatal loss.

Third, while efforts have been underway to identify grief theories which are appropriate to guide the care and research of persons experiencing perinatal loss, the appropriateness of such theories requires evaluation. Perinatal loss is a complex, multifaceted phenomenon, which takes form in many different circumstances (i.e., miscarriage, stillbirth, life-limiting congenital anomalies, neonatal complications, obstetric complications, etc.), and thus requires attentive examination. The feasibility of a complex, multi-theoretical approach to care requires evaluation, as the theoretical approach to care is only as effective as it is appropriate and realistic. Clearly
there are no simple answers, however, the need for a theoretical framework is evident. Further research is indicated regarding the appropriateness of a multi-theoretical approach versus developing a perinatal bereavement theory to inform patient care.

Fourth, along with the question of perinatal bereavement theory comes the question of the appropriateness of terms commonly used in the grief language. The ‘language’ of grief, as previously discussed, borrows the term ‘chronic’ from pathology science to explain adverse symptoms associated with grief which are present for a prolonged period of time. As seen in this research study, ‘chronic’ may not be appropriate to identify healthy versus unhealthy grief. Perhaps new terminology is required to help explain the grief associated with perinatal bereavement, which in turn assists in identifying complex and intense grief versus a healthy recovery, thus identifying those in need of additional bereavement services and allocating resources as appropriate.

Fifth, high-level communication is one of the unique characteristics which differentiates humans in the animal kingdom. Understanding the meaning behind each syllabic utterance allows humans as a species to move forward and advance civilization. However, a breakdown in communication leads to various misunderstandings and potential unintended consequence. Misunderstandings in communication surrounding perinatal loss, specifically the diagnostic communication of a lethal or life-limiting congenital anomaly, may have life-long adverse consequences of unpredictable intensities. Therefore, the diagnostic communication of a lethal or life-limiting congenital anomaly should be considered a high-stakes conversation. Healthcare professionals who may be responsible for such communication require specialty training and resources. It is necessary to recognize both the critical importance of such conversations as well as the need of training for professionals whom will take part in such interactions. Further
research regarding the curriculum, training, and state of the science regarding high-stakes conversations and perinatal loss is necessary to ensure healthcare professionals are equipped with the tools, knowledge and skills necessary to compassionately communicate sensitive and life-altering information to expectant parents.

Sixth, healthcare professionals cannot be expected to innately know how to best communicate with and care for parents experiencing perinatal loss. The care of such a unique patient population requires evidence-based specialty education and experience. Unfortunately, a review of nursing psychology text books revealed that 100 % of texts contained at least one myth about grief, and 78% contained up to four myths about grief (Black et al., 2016). If training and education is based on myth rather than evidence, healthcare professionals are at a disadvantage when it comes to providing the best care possible for bereaved parents. Therefore, it is necessary to evaluate not only the education and training healthcare professionals receive regarding perinatal loss, but the state of the science upon which perinatal loss curriculum is based. Such curriculum includes, but is not limited to, nursing and medical pre-licensure curriculum, continuing education curriculum, and specialty certification.

Lastly, policy regarding organ donation practices are also in need of evaluation. Several parents in this study mentioned seeking organ donation as a way of making meaning in their loss experience, finding solace, and preventing other parents from experiencing the grief of losing a child. However, many parents were met with resistance from either healthcare professionals overseeing their care, or institutional regulations preventing the donation of their child’s organs. It is necessary to further investigate barriers limiting the precious resource of healthy tissue, not only to provide solace for grieving parents, but to potentially spare the lives of other neonates.
Experiencing a pregnancy complicated by anencephaly is a life altering event for parents. Clearly there are ample opportunities for research in the area of perinatal loss, grief and bereavement, and those mentioned above are among the primary prospects to further explore phenomena surrounding perinatal loss related to anencephaly. Such a life altering event requires the utmost attention of healthcare professionals and researchers alike, and presents complex challenges to the healthcare professions.

**Conclusion**

Parents experiencing perinatal loss due to anencephaly find themselves experiencing an ontological death, in which their understanding of how the world operates is shattered. Such loss is a life altering, traumatic event which commonly outstrips their ability to cope. The attempt to reestablish meaning is a life-long journey, one which requires professional support and care. Eighty percent of the women and 50% of the men in this study continue to experience intense grief, far exceeding the one in four women reported in the literature to have intense grieving reactions following perinatal loss. These findings suggest the perinatal grief intensity of the study participants was severely under recognized – particularly in father figures – and consequently under treated. Seven (29%) participants, less than one third of those who participated in this study, were offered bereavement support, and only 12% of parents were referred to PnPC service. Follow up care, though critical, was provided to only one woman.

Though anencephaly is not the most common of healthcare ailments, it is one of high priority. Parents experiencing a pregnancy complicated by anencephaly require compassionate, patient-centered care. It is necessary to increase screening practices, thus identifying parents who are in need of additional bereavement support and resources throughout their perinatal loss experience. The pain of loss, strength, resolve, and love echoes through the narrative accounts
shared by every parent in this study. It is through these narratives we can endeavor to better understand the impact of an anencephalic pregnancy on parents and the resulting needs they face throughout their journey towards recovery in the wake of perinatal loss.
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Theme 1: Perinatal Loss as a Journey

- Shock
  - Powerful emotions
  - Denial
  - Grief
  - Guilt
  - Marital strain

- Activation
  - Seeking information
  - Decision making
  - Finding joy
  - Positive association
  - Making memories
  - Living intentionally
  - Embracing the moments
  - Learning self-care

- Reframing
  - Reentering society
  - Process of moving on
  - Processing emotions
  - Learning to cope
  - Long-term experience
**Theme 2:** Struggling with Societal Expectations

**Theme 3:** Communication with Providers and Others as Challenging
APPENDIX B
Research Consent

WASHINGTON STATE UNIVERSITY
College of Nursing
Spokane, WA
Research Study Consent Form

Study Title: Piloting Project PoppySeed: Experiencing Anencephaly, A Parent’s Perspective

Researchers:
Shandeigh Berry, MSN, RN, CNOR
Principal Investigator
PhD Candidate
Olympia, WA 98513
(360) 880-1097
shandeigh.berry@wsu.edu

Billie Severtsen, PhD, RN
Co-Investigator
Associate Professor
SNRS 222BPO BOX 1495
509-324-7286
severt@wsu.edu

Gail Oneal, PhD, RN
Analysis Team
Assistant Professor
SNRS 345PO BOX 1495
509-324-7263
goneal@wsu.edu

Lonnie Nelson, PhD, RN
Methods Expert, Analysis Team
Assistant Professor
1100 Olive Way Ste 1200
You are being asked to take part in a research study carried out by Drs. Billie Severtsen and PhD candidate Shandeigh Berry. This form explains the research study and your part in it if you decide to join the study. Please read the form carefully, taking as much time as you need. Ask the researcher to explain anything you don’t understand. You can decide not to join the study. If you join the study, you can change your mind later or quit at any time. There will be no penalty or loss of services or benefits if you decide to not take part in the study or quit later. This study has been approved for human subject participation by the Washington State University Institutional Review Board.

What is this study about?

This research study is being done to better understand parent’s lived experience when they learn their baby has anencephaly. Very little is known about this experience. Gaining a better understanding of what parents go through during this difficult time will help doctors and nurses know how to provide more appropriate, sensitive, and patient-centered care. You are being asked to take part because you have lost a baby to anencephaly. This study entails an interview which will take about one hour. However, we wish to know your story as you wish to share it. If the interview takes more or less time than one hour, we will prolong or cut short your time as you tell your story. It may take more than one interview for you to tell us your story, if that is the case, we will set up a second or third interview. You will also be asked to complete the Perinatal Grief Intensity Scale (14 questions) and a demographic questionnaire (17 questions).

You cannot take part in this study if you are under 18 years of age, cannot read, write or communicate in English, or are currently pregnant.

What will I be asked to do if I am in this study?

If you take part in the study, you will be asked to tell us about the time you found out your baby had anencephaly.
• **Face to face interview.** During your interview, you and the interviewer will be the only persons in the room. You will be asked to tell the interviewer about the time you found out your baby had anencephaly. The interviewer may ask you “clarifying questions,” which means asking a question to assure they understand what you are saying.

• The length of time each interview will take may vary. The typical time each interview will take is likely one hour. However, we wish to understand your entire story, and interviews may take more or less than one hour. It may also take more than one interview to tell your story.

• During the interview, you will pick a name for the interviewer call you (a pseudonym) for the remainder of the study. This is to protect your identity. If you would rather we not refer to you by another name, we will use non-descriptive terms such as he/she, or they.

• You may decline to share certain information if you wish. If you become distressed at any point during the interview, or if you wish the interview to be over, we will stop the interview. There will be no penalty or consequence to not finishing the study.

• Each interview will be audio recorded by the interviewer and later transcribed by a professional, university approved transcriptionist. All your identifying information will be removed from the audio recording prior to being transcribed.

• **Perinatal Grief Intensity Scale (PGIS):** The PGIS is a 14 item questionnaire which helps healthcare professionals such as doctors and nurses better recognize those who are likely to have intense grief after an early or late pregnancy loss or death of an infant.

• The length of time this will take to complete is approximately 15 minutes. However, this time approximation may vary for each person.

• **Demographic Questionnaire:** This is a 17 item questionnaire to better help the research team understand your gender, age, race, and other related characteristics. These questions are important to answer as we attempt to better understand how to help parents who are experiencing a pregnancy complicated by anencephaly.

• The length of time to complete this questionnaire is approximately 20 minutes, however, the time may vary from person to person.

**Are there any benefits to me if I am in this study?**

There is no direct benefit to you from being in this study. However, a potential benefit to you for taking part in this study is finding solace and meaning in your experience of having a baby with anencephaly by helping those in the future who may experience the same situation.

If you take part in this study, you may help others in the future who have a baby with anencephaly. The information you provide will help us improve practice in the healthcare system.

**Are there any risks to me if I am in this study?**

A potential risk from taking part in this study includes experiencing emotional discomfort or distress as you tell your story. It is also possible that your identity will become known from being in our study. Furthermore, all members of the research team are mandated suicide reporters,
which means if we suspect you may be at risk for suicide, we will be required to alert the proper authorities.

To minimize the potential risk to you while participating in our study, we have the following plans in place:

- **Emotional discomfort or distress**
  - If at any time you do not wish to continue in the study, you may withdraw without consequence or penalty.
  - We can stop the interview at any time and finish another day if you wish to do so.
  - We will provide you with a list of local mental health resources in your community. We will also help you make an appointment with whichever resource you choose or recommend one which will be appropriate for you. These resources will be provided at no cost to you.

- **Re-Identification – Your identity becoming known**
  - We remove all identifying information from the audio recording before it is transcribed. This means that there is nothing linking your interview to your identity.
  - Your identity will not be made known during this study.
  - All electronic identifying information will be encrypted and kept on a password protected computer with a firewall system and limited access.
  - Consent forms will be kept in a locked cabinet in a researcher office requiring key card for access.
  - Should your identity become known, or be suspected of becoming known, you will be contacted by Shandeigh Berry. The research team will make every effort to protect your identity.

- **Risk of suicide**
  - Should Shandeigh Berry, the interviewer, suspect you are at risk for suicide at any time during the interview, the interview will be stopped immediately. Shandeigh Berry, a Registered Nurse with suicide training and a mandated reporter, will care for you and remain with you until you have been transferred to the appropriate care.
  - You will be provided with a list of mental health resources in your community available to you free of charge. Shandeigh Berry will assist you in choosing the resource you feel is most appropriate. Assistance will be provided in making appointments with the resources as necessary.

**Will my information be kept private?**

The data for this study will be kept confidential to the extent allowed by federal and state law. No published results will identify you, and your name will not be associated with the findings. Under certain circumstances, information that identifies you may be released for internal and external reviews of this project.
o All identifying information will be removed from your data and replaced with an encryption code. A master key list with identifying information will be kept in a separate, limited access, locked location. Access to the master key will be restricted. All information stored on a computer will be password protected behind a firewall system.

o To protect your privacy, your identifying information will not be shared. The research team will meet in private rooms when discussing your data. During this study, you will have no interactions with other participants.

o All members of the research team are mandated by the state to report suspicion of suicide. Should any of the researchers suspect you are at risk for suicide during the study, we will be required to report this. If during the interview risk of suicide is suspected, the interview will be stopped and you will be provided with the necessary resources. Our number one concern is your safety.

o Your data will be stored behind firewall systems on a password protected computer. Consent forms will be stored in a locked cabinet in a limited access personal researcher office requiring a key card for access.

o Your data will be available to the researcher team, which includes four university employed research faculty, Drs. Oneal, Severtsen, and PhD student Shandeigh Berry, and members of the Institutional Review Board (IRB).

Each interview will be audio recorded. Audio recordings of the interviews are necessary for the study for data analysis. The audio recordings will be transcribed by a professional transcriptionist. Your identifying information will be removed from the audio recordings prior to transcription.

The results of this study may be published or presented at professional meetings, but the identities of all research participants will remain anonymous.

The data for this study will be kept for five years after completion of the study.

**Are there any costs or payments for being in this study?**

There will be no costs to you for taking part in this study.

You will not receive money or any other form of compensation for taking part in this study.

**Who can I talk to if I have questions?**

If you have questions about this study or the information in this form, please contact the researcher Shandeigh Berry: (360) 880-1097, shandeigh.berry@wsu.edu. If you have questions about your rights as a research participant, or would like to report a concern or complaint about
What are my rights as a research study volunteer?

Your participation in this research study is completely voluntary. You may choose not to be a part of this study. There will be no penalty to you if you choose not to take part. You may choose not to answer specific questions or to stop participating at any time.

What does my signature on this consent form mean?

Your signature on this form means that:

- You understand the information given to you in this form
- You have been able to ask the researcher questions and state any concerns
- The researcher has responded to your questions and concerns
- You believe you understand the research study and the potential benefits and risks that are involved.

☐ I grant permission to be audio recorded by WSU PI Shandeigh Berry of Washington State University.

☐ I grant permission to the research team to use my de-identified data (interview transcripts) for secondary analysis in future studies.

☐ I grant permission to WSU PI Shandeigh Berry to contact me in the future concerning participation in additional studies investigating perinatal grief, anencephaly, or associated topics.

Statement of Consent

I give my voluntary consent to take part in this study. I will be given a copy of this consent document for my records.

__________________________________ ______________________
Signature of Participant Date
Printed Name of Participant

Statement of Person Obtaining Informed Consent

I have carefully explained to the person taking part in the study what he or she can expect.

I certify that when this person signs this form, to the best of my knowledge, he or she understands the purpose, procedures, potential benefits, and potential risks of participation.

I also certify that he or she:

- Speaks the language used to explain this research
- Reads well enough to understand this form or, if not, this person is able to hear and understand when the form is read to him or her
- Does not have any problems that could make it hard to understand what it means to take part in this research.

__________________________________  ________________________
Signature of Person Obtaining Consent  Date

__________________________________  ________________________
Printed Name of Person Obtaining Consent  Role in the Research Study
APPENDIX C

Demographic Questionnaire

Name:

Your name will be removed from this survey to protect your identify. All efforts to preserve your anonymity are observed and enforced.

1. What was your assigned sex at birth?
   a. Male
   b. Female

2. What gender do you identify as today?
   a. Male
   b. Female
   c. Transgender
   d. Two-Spirited
   e. Questioning
   f. Agender
   g. Bigender
   h. Gender Variant
   i. Pangender

3. Which category below includes your age?
   a. 18-29
   b. 30-39
   c. 40-49
   d. 50-59
   e. 60-69
   f. Greater than 70

4. Which best describes your race?
   a. White
   b. Hispanic or Latino
   c. Black or African American
   d. Native American or American Indian
   e. Asian / Pacific Islander
   f. Other

5. Which best describes your religious preference?
   a. Atheist
   b. Agnostic
   c. Buddhist
   d. Catholic
   e. Christian
   f. Hindu
   g. Jewish
6. What is the highest level of school you have completed or the highest degree you have received?
   a. Some High School
   b. High School Graduate
   c. Some College
   d. Associate Degree
   e. Bachelor’s Degree
   f. Master’s Degree
   g. Doctorate Degree

7. How much total combined income did your household earn the year you or your partner experienced a pregnancy complicated by anencephaly?
   a. Less than $25,000
   b. $26,000-$49,000
   c. $50,000 - $100,000
   d. Greater than $100,000

8. Which best describes the location you lived at the time you or your partner experienced your pregnancy complicated by anencephaly?
   a. Rural
   b. Suburban
   c. Urban

9. Which category best describes your current relationship status?
   a. Single
   b. In a relationship
   c. Married
   d. Separated
   e. Divorced
   f. Widowed

10. Which category best describes your relationship status at the time you or your partner experienced a pregnancy complicated by anencephaly?
    a. Single
    b. In a relationship
    c. Married
    d. Separated
    e. Divorced
    f. Widowed

11. How much time has passed since you or your partner experienced a pregnancy complicated by anencephaly?
    a. Less than 6 months
b. 7 months – 1 year  
c. 2-3 years  
d. 4-5 years  
e. 6-10 years  
f. Greater than 10 years  

12. Which number best describes the number of pregnancies you or your partner experienced before experiencing a pregnancy complicated by anencephaly?
   a. 0  
   b. 1  
   c. 2  
   d. 3  
   e. 4  
   f. 5  
   g. Greater than 5  

13. Which number best describes the number of living children you had at the time of your or your partner’s pregnancy complicated by anencephaly?
   a. 0  
   b. 1  
   c. 2  
   d. 3  
   e. 4  
   f. 5  
   g. Greater than 5  

14. Which number best describes the number of living children you have currently?
   a. 0  
   b. 1  
   c. 2  
   d. 3  
   e. 3  
   f. 4  
   g. 5  
   h. Greater than 5  

15. Were there any questions on the Perinatal Grief Intensity Scale that seems inappropriate or did not make sense to you?  

16. Were there any questions on the demographic questionnaire that seemed inappropriate or did not make sense to you?  

17. Are there any questions you feel should have been asked that were not?
APPENDIX D

Perinatal Grief Intensity Scale

This survey helps health care providers (HCPs) to better recognize those who are likely to have intense grief after an early or late pregnancy loss or death of an infant.

Your name will be removed from this survey to protect your identify. All efforts to preserve your anonymity are observed and enforced.

Name:

1. The pregnancy did not seem real to me
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
2. I did not think of the baby as a person
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
3. I did not think of the baby as having a specific personality
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
4. I felt I had lost my son or daughter, not just my pregnancy
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
5. Both my pregnancy and the baby seemed real to me
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
6. It seemed more like the loss of a pregnancy than the loss of a baby
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
7. In the first hours and days after my loss, if people said or did things that made me feel bad, I was able to ask them to stop
   a. Strongly Disagree
   b. Disagree
c. Agree
d. Strongly Agree
8. In the first hours and days after my loss, if something happened that I did not like, I was usually able to resolve the problem
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
9. In the later weeks after my loss, if people said or did things that made me feel bad, I was able to ask them to stop
   a. Strongly Disagree
   b. Disagree
   c. Agree
   d. Strongly Agree
10. In later weeks after my loss, if something happened that I did not like, I was usually able to resolve the problem
    a. Strongly Disagree
    b. Disagree
    c. Agree
    d. Strongly Agree
11. During and after my perinatal loss, I was satisfied with the way my loss experience was unfolded, given that I had to go through it
    a. Strongly Disagree
    b. Disagree
    c. Agree
    d. Strongly Agree
12. During and after my perinatal loss, I was satisfied with my interactions with my family
    a. Strongly Disagree
    b. Disagree
    c. Agree
    d. Strongly Agree
13. During and after my perinatal loss, I was satisfied with my interactions with my friends
    a. Strongly Disagree
    b. Disagree
    c. Agree
    d. Strongly Agree
14. During and after my perinatal loss, I was satisfied with my interactions with my nurses
    a. Strongly Disagree
    b. Disagree
    c. Agree
    d. Strongly Agree