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ANTICIPATORY GRIEF IN PARENTS WHOSE CHILD HAS A NON-MALIGNANT LIFE-
LIMITING CONDITION

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Approval of Thesis

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was my first experience of anticipatory grief, and reflecting on the experience of losing her is what inspired me for the topic of my thesis. I miss her beyond words, but I am comforted by knowing that she'd be proud of me and this work.

Abstract

Parents who have a child with a non-malignant life-limiting condition (NMLLC) universally experience anticipatory grief (AG), which is grief prior to death. While there are numerous studies on post-death grief, research on AG in parents who have lost a children with a NMLLC is scarce. This qualitative study used an interpretive description approach and semi-structured interviews were conducted with parents who have a child with a NMLLC recruited through a children's hospice. The data illuminates the complex challenges experienced by these parents. Participants learned to navigate uncertainty while grieving ongoing losses. They experienced varying degrees of sadness, but also found joy in creating memories. Maintaining their parental role empowered them and helped them cope with grief. Parents often mentioned the importance of empathy and transparency when communicating with health care providers. The benefits of holistic care of the whole family by a specialized pediatric palliative care team were also highlighted.

Keywords: anticipatory grief, grief, interpretive description, life-limiting condition, pediatric palliative care

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List of Accronyms

AG : Anticipatory grief

HCP : Health care provider

ID : Interpretive description

LLC : Life-limiting condition

NMLLC : Non-malignant life-limiting condition

Chapter 1. Introduction to the Study

Introduction

Although the prevalence of children with life-limiting conditions (LLC) in Canada is unknown, a large number of families anticipate and experience the death of their child due to these conditions (Siden, 2018). The time between a child's diagnosis of LLC and their death is represents a rollercoaster of emotions for parents; it is a period filled with grief, uncertainty, and fear. My research explores the experience of anticipatory grief (AG) in parents who have a child who died from a non-malignant life-limiting condition (NMLLC).

In North America, children with cancer comprises 30–40% of the cases seen by pediatric palliative care clinical teams, but a broad range of other LLC affecting the brain, the muscles, the heart and lungs, warrant health care professionals' attention (Siden, 2018). In British Columbia, Canuck Place Children's Hospice provides interdisciplinary pediatric palliative care services to over 800 families, of which approximately 85% have a child with a NMLLC (Canuck Place Children's Hospice, 2019). Given that NMLCC are often rare diseases for which little is known, parents live a life filled with uncertainties, with death as the only certain outcome. Due to the rarity of their child's diagnosis, parents are often met with confusion from health care providers and from family and friends. This chapter provides background information on the experience of caring for a child with a NMLLC and presents a brief chronology of the concept of AG. Then, the research problem as well as the purpose of the study and research questions will be stated. I will conclude by providing an overview of my personal and professional stance and by defining key terms used throughout my thesis.

Background

Caring for a child with a non-malignant life-limiting condition

Parents who have a child with a NMLLC carry a significant burden; they experience intense emotions, and live in uncertainty with death as the only certain outcome for their child (Siden & Steele, 2015). Childhood death is out of step with life's expected journey (Meiring, 2011), but for children with a NMLLC, death is the normal, expected outcome. Parents live knowing that the future they had wished for their child no longer exists (Higgs et al., 2016; Lucca & Petean, 2016; Yang et al., 2016). Some parents think about the eventual loss of their child every day (Verberne et al., 2019). The loss of the child they had wished for, combined with other losses such as loss of physical abilities and loss of being able to go to school and to participate in activities with their peers, causes immense grief (Higgs et al., 2016; Lucca & Petean, 2016; B.-H. Yang et al., 2016). Parents also grieve the loss of their own goals, for example career goals, due to caregiving demands related to their child's condition (Verberne et al., 2019).

Parents of children who have a NMLLC assume many roles in addition to the "typical" parental responsibilities. These roles extend across multiple domains: physical, emotional, social, and spiritual (Caicedo, 2014). Parents' feeling of being worth to their child's well-being helps them cope with the demands of caregiving, however, their caregiver responsibilities (personal hygiene, feeding, medications, managing symptoms, etc.) become more difficult and harder to maintain as their child's disease progresses (Verberne et al., 2019). Parents spend on average more than one-third of their day every day providing care for their child at home and need to use an average of five medical devices (Lazzarin et al., 2018). Decision-making about their child's

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treatment and specialists, and advance care planning are also tasks that add to the burden of care by parents (Brotherton & Abbott, 2012; Hellmann et al., 2013).

Parents directly influence their child's wellness and health outcomes, so attention to their well-being is very important when caring for their child (Jones & Wolfe, 2014; Weaver et al., 2019). These parents are more likely than parents of healthy children to experience symptoms of depression, fatigue, sleeping issues (Koch & Jones, 2018). They also suffer from social isolation due to the caregiving demands, frequent hospitalizations or frequent appointments with specialists, and lack of respite care (Collins et al., 2016).

Experiences of family members prior to the death of a loved one can have long lasting effects even after death (Davies et al., 2013). Gaining insight into the experience of grief in parents of children with NMLLC will not only help HCP better support parents prior to the death of their child, but it will also create a lasting impact on their bereavement journey.

Anticipatory Grief – a brief chronology

Anticipatory grief is a concept that was first coined in 1944 by Erich Lindemann to describe the emotions a person experiences prior to losing a loved one (Lindemann, 1944). He observed that wives of men in the armed forces had already completed their “grief work” during the period where they were separated from their husband and anticipating their potential death. When the husbands returned from war, their wives had already emancipated themselves and had moved on to focusing on new relationships (Lindemann, 1944). Lindemann (1944) concluded that his patients/study participants were so concerned with their adjustment in the event that their father, son, or husband would die that they went through all the phases of grief. Initially, AG was considered “grief work” that occurred prior to an actual loss, and during this “grief work”, a person would slowly and gradually detach their bonds with the dying person (Lindemann, 1944).

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Lindemann's (1944) studies led to the assumption that AG, i.e., grief prior to death, was the same as bereavement grief.

The 1950s-1960s gave rise to many advances in the field of grief and AG. Many studies on "forewarning of loss" were conducted with parents of terminally ill children, showing the adaptive potential of anticipatory mourning (Rando, 2000). The 1950s and 1960s also witnessed the birth of the hospice movement which was founded by Cecily Saunders in 1967 (Richmond, 2005). The hospice movement created a space for providing holistic care to dying patients and their families. The 1960s provided our first encounter with Kubler Ross' five stages of grief: denial, anger, bargaining, depression and acceptance (Kübler-Ross, 1969). In her theory, she drew our attention to our avoidance of death and dying and to the importance of hope and of learning from the dying patients.

In the early 1970s, Fulton and Fulton (1971) qualified AG as having a "two-edged effect". These authors noted that AG "possesses capacity to enhance our lives and secure our well-being, while possessing at the same time the power to undermine our fragile existence and rupture tenuous social bonds" (Fulton & Fulton, 1971, p.99). Fulton and Fulton thought that experiencing AG could result in a lack of emotional response at the time of death, which could precipitate judgement by others. Futterman et al. (1972) may have been the first to differentiate AG from anticipatory mourning, the latter being a "set of processes that are directly related to the awareness of the impending loss, to its emotional impact, and to the adaptive mechanisms whereby emotional attachment to the dying is relinquished over time" (p.251). In their study of parental anticipatory mourning, they proposed five interdependent and continually interacting processes of anticipatory mourning: acknowledgement (acknowledging that the child's death is inevitable), grieving (experiencing emotions resulting from the anticipated loss), reconciliation

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(developing a sense of confidence in the worth of the child's life), detachment (detachment of emotional connection with the child) and memorialization (developing a fixed mental representation of the child that will endure after death). In 1974, the first book devoted to AG was published (Schoenberg, 1974). In one of the chapters, Aldrich (1974) highlighted that while pre- and post-death grief shared similarities, there were important differences worth noting, such as the idea that AG has a definite endpoint (death), and that AG can also be experienced by the person who is dying, as opposed to conventional death which can only be experienced by those surviving the death of a person. In 1976, Lebow posited a set of adaptational tasks for anticipatory mourning, which represent areas that could be targeted by interventions from professionals: a) remaining involved with the patient, b) remaining separate from the patient, c) adapting suitably to role changes, d) bearing the affects of grief, e) coming to some terms with the reality of the impending loss and f) saying goodbye (Patinadan et al., 2020).

In 1986, the second book focusing on AG was published by Therese A. Rando (Rando, 1986). Rando recognized the complexity and multidimensionality of AG and offered the following definition:

Anticipatory grief is the phenomenon encompassing the processes of mourning, coping, interaction, planning and psychosocial reorganization that are stimulated and begun in part in response to the awareness of the impending loss of a loved one and the recognition of associated losses in the past, present and future. It is seldom explicitly recognized, but the truly therapeutic experience of anticipatory grief mandates a delicate balance among the mutually conflicting demands of simultaneously holding onto, letting go of, and drawing closer to the dying patients (Rando 1986, p.24).

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The late 1980s and 1990s represent an era of refinement and operationalization of the concept of AG. The Anticipatory Grief Inventory was developed in 1991 by Levy and his colleagues (Levy, 1991). Many researchers also elucidated various other concepts related to AG, for example *disenfranchised grief*, which is defined as “(…)grief that persons experience when they incur a loss that is not or cannot be openly acknowledged, publicly mourned, or socially supported”(Doka, 1989, p.4). In 1994, Rolland contributed to AG knowledge by developing the Family Systems-Illness Model (Rolland, 1994). The Family Systems-Illness Model views family relationships as a potential resource in families where a member is suffering from a chronic illness and it emphasizes the possibilities for resilience and growth (Rolland & Walsh, 2006). Rolland discusses anticipated loss experienced by families dealing with a chronic of life-threatening illness, suggesting that anticipatory loss can be as challenging and painful as the actual death of a loved one (Rolland, 1990).

Since Lindemann’s (1944) studies, the concept of AG has evolved but is still believed to be a misnomer by some researchers. The term *anticipatory grief* leads us to believe that the grieving is for losses that will occur in the future; however, it is also used to describe grief that encompasses emotions that are also a result of past and present losses (Fulton, 2003; Moon, 2016; Rando, 1986). In 2000, Therese Rando, who had to this date used the term “anticipatory grief”, changed the concept’s terminology to *anticipatory mourning*. Rando (2000) defines anticipatory mourning as “the phenomenon encompassing the process of mourning, coping, interaction, planning and psychological reorganization that are stimulated and begun in part in response to the impending loss of a loved one and the recognition of associated losses in the past, present and future” (p.24). Rando (2000) describes six dimensions of anticipatory mourning that helps with personal readjustments and accommodate the loss of a loved one: 1) perspective, 2)

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time focus, 3) influencing factors, 4) major sources of adaptational demands, 5) generic operations, and 6) contextual levels. The first three describe the experience while the last three are experiential dimensions that constitute areas suitable for interventions (appendix A).

In the current literature, *anticipatory grief* and *anticipatory mourning* are often used interchangeably. Other terms to describe the experience of grieving before the death of a loved one are also used, for example, *anticipatory loss* and *pre-death grief*. For the purpose of this proposal, *anticipatory grief* will be used, although other terms may be used in the literature review to reflect the preferred terminology of different authors.

Researcher's Personal and Professional Contexts

My interest in exploring the topic of AG in parents who have a child with a life-limiting condition stems from my personal experience and my professional experience working as a research assistant for many years in the field of pediatric palliative care, and in more recent years, from my work as a pediatric critical care and pediatric palliative care registered nurse.

As a nurse in pediatric critical care and palliative care, I witness immense grief and suffering, but also hope and joy, as parents live a life constantly overshadowed by the thought that their child's death is inevitable. As a parent once told me shortly after their infant died, "yes, there were happy moments, but I never had the chance to know him without cancer; cancer was always in the background". While talking about death and grief is never comfortable, because of my experience in pediatric palliative care research, and then through my experience as a nurse, I feel like I am able to walk alongside these parents in their grief, in their hope and happiness and everywhere in between. I always wonder how much grief is hiding behind these parents' smiles? How do they manage to find the strength to keep going? How can they still hope, when there is only one horrible possible outcome at the end of it all? Is it faith? Is it a

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good support network? As a nurse, how can I help them carry the weight on their shoulder? I also wonder why post-death grief is increasingly getting more attention, while grief prior to death is not. Why do scholars continue to measure grief with numbers? How can numbers help me, as a nurse, to better care for families? I knew at the very start of my master's degree that I wanted to do a qualitative study that would uncover more of the experience of parents who care for a child with a life limiting condition, and I decided to focus on AG.

I do not have a child diagnosed with a serious illness, so while my professional experience sets the stage for my inquiry, it is important that I locate myself within the topic of AG. When I started my graduate studies, someone precious in my life died. No matter how much I thought I knew about grief, I was not prepared for all the emotions that I experienced and that I still, at times, experience today. Seven months passed between diagnosis and death, and I often reflect on how those monthly trips across the country to help her actually helped me. I reflect on the hopes I had that maybe she would beat the odds, or maybe she had been misdiagnosed. It was my heart speaking and winning over my head who knew all too well that someone simply cannot survive this disease. I hoped until the very end, even on her last day. I do not remember what I was hoping for, but I definitely had hope. But I also felt grief. I grieved for seven long, yet too short months, knowing she would die some time in a soon future. Many times, I reminded myself that I was lucky to get more time with her, although no amount of time would ever be enough.

Grieving the loss of a person who has not died yet, while trying to keep oneself afloat, is challenging. As I was experiencing AG, I often thought of others who are experiencing a similar situation; were my reactions normal? I wondered how it was for families that I care for every day at work, as they grieve the future loss of their child, as they grieve the loss of an idealized child. These questions and reflections influenced my decision to study AG in parents who have a child

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with a NMLLC. I hope that through my inquiry, we can come to a better understanding of the grief these parents experience as they care for a child who will inevitably die and that we can find better ways to support them.

Statement of Problem

To date, the experience of AG in parents who have a child with a NMLLC has been given little attention in the scientific literature. Interpersonal, intrapersonal and health-care related factors that potentially impact AG are yet to be fully uncovered. In-depth exploration of AG in parents who have a child with a NMLLC can make a valuable contribution to the understanding of AG and can improve care and support for the parents whose child will die.

Purpose of the Research Study and Research Questions

The purpose of this study is to explore the experience of AG in parents who have a child with a NMLLC. Using an interpretive description (ID) methodological approach informed by principles of naturalistic inquiry, the study was to answer the following research questions:

- How do bereaved parents of children with a NMLLC describe their experience of AG?
- How do parents describe factors (interpersonal, intrapersonal, health care related) that may have influenced their AG experience?

Definition of Terms

Caregivers: Individuals, often parents, who provide care to a child (Spicer et al., 2015).

Child/Children: Infants, children and youth aged 0–19 years (Spicer et al., 2015).

Grief: Emotional reaction or response to the loss of a loved one through death (Weiss, 2008).

Anticipatory grief: Grief that is experienced prior to the death of a loved one and that is a result of past, present and future losses (Rando, 2000).

Life-Limiting Condition (LLC): Sometimes called life-threatening condition. LLC are conditions for which there is a likelihood of death before adulthood (Spicer et al., 2015).

Together for Short Lives, a UK charity for children with life-threatening and LLC, provides a helpful categorization of these conditions based on their trajectory (Figure 1) (Together for Short Lives, n.d.):

Figure 1

Categories of Life-Threatening and Life-Limiting Diseases

Category 1	<p>Life-threatening conditions for which curative treatment may be feasible but can fail.</p> <p>Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services.</p> <p><i>Examples: cancer, irreversible organ failures of heart, liver, kidney.</i></p>
Category 2	<p>Conditions where premature death is inevitable.</p> <p>There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities.</p> <p><i>Examples: cystic fibrosis, Duchenne muscular dystrophy.</i></p>
Category 3	<p>Progressive conditions without curative treatment options.</p> <p>Treatment is exclusively palliative and may commonly extend over many years.</p> <p><i>Examples: Batten disease, mucopolysaccharidoses.</i></p>
Category 4	<p>Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health complications and likelihood of premature death.</p> <p><i>Examples: severe cerebral palsy, multiple disabilities such as following brain or spinal cord injury, complex health care needs, high risk of an unpredictable life-threatening event or episode.</i></p>

Pediatric Palliative Care: A holistic and multidisciplinary approach to care of children with LLC and their families, from the onset of first symptoms, through the disease trajectory and during bereavement. The focus of pediatric palliative care is on comfort, not on cure, although

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comfort does not exclude cure. Pediatric palliative care includes symptoms management, respite care, coordination of services, end-of-life care, and bereavement support (Spicer et al., 2015).

Summary

Caring for children who have a NMLLC comes with a significant burden for parents. By examining the experience of AG in this population, I hope that health care providers increasingly acknowledge AG and its impact on the well-being of the whole family, which can then lead to improved care. The next chapter presents a review of the literature on AG.

Chapter 2. Literature Review

Introduction

Understanding the experience of AG in parents is necessary to provide adequate support to families. To date, most studies on AG have focused on AG in caregivers of adult patients with dementia, cancer or other terminal illnesses, with only a handful of studies that have looked at parental AG in a pediatric context. The goal of this literature review was to understand the nature of current knowledge around my proposed topic and to identify knowledge gaps in the literature in order to drive my research questions and research design. The thoughtful methods directing this review resemble a narrative review (Green et al., 2006). The primary purpose of a narrative review is to provide a comprehensive background for understanding existing knowledge and highlighting the significance of the proposed new research (Paré & Kitsiou, 2017). A narrative review takes a less formal approach than systematic reviews in that it does not always employ search and reporting methods that are as rigorous (Jahan et al., 2016). Nevertheless, “they are helpful in presenting abroad perspective on a topic” (Green et al., 2006, p.103).

This chapter will first describe my search strategy. Then, challenges with AG terminology, characteristics of the included studies as well as key themes that were identified in the literature will be discussed. Lastly, I will summarize the findings and identify research gaps.

Search Strategy

Searches were carried out to identify the current knowledge of AG in parents. CINAHL, PubMed, Pubmed Central and PsycINFO were searched with the following key words: parent, mother, father, anticipatory grief, anticipatory loss, anticipatory mourning, pre-death grief and pre-death mourning. The terms were combined using Boolean operators and truncation, and the searches were limited to articles in English or in French. A search through Google Scholar was

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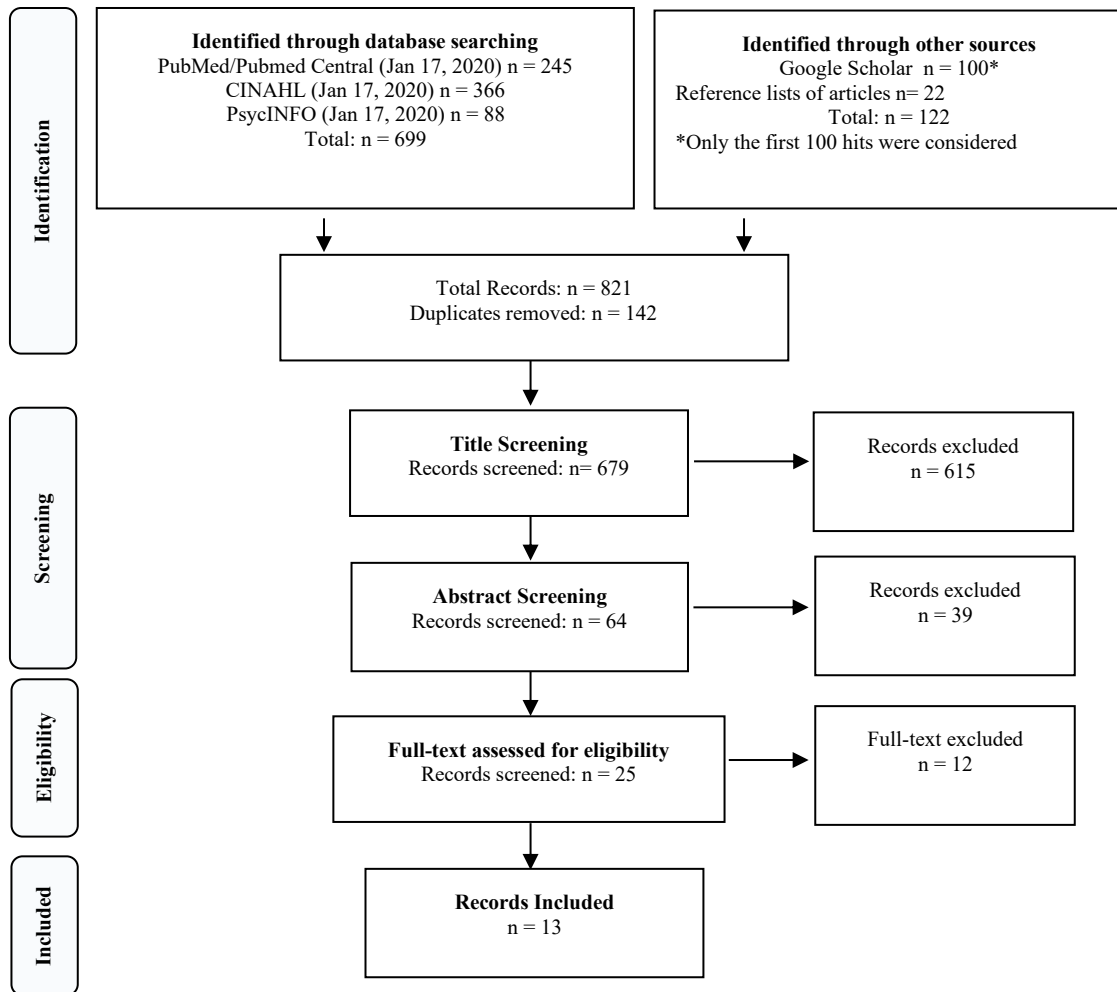
also performed with the same key words. Considering the number of hits (>5000) and the time that would be required to screen each one, the decision was made to screen the first 100 hits only (sorted by relevance). It is unlikely that further screening past the 100th hit would yield more articles relevant to this review (Haddaway et al., 2015). The reference lists of publications were also scrutinized to identify other key publications. This literature took place in January 2020. Due to the paucity of research on AG in parents and to capture a maximum of articles, no search limits were placed on the year and location of publications.

Studies published in peer-reviewed journals and that focused on AG in parents in a pediatric context were considered for this literature review, while studies looking at AG in a spouse, AG in parents of adult children or AG experienced in children were excluded. In addition, grey literature such as dissertations, reports, opinion pieces, and documents published from various organizations were excluded from this study.

The title and abstract of all records identified through the various searches were screened and those which fit the inclusion criteria were retained. A full-text review of the articles that were retained during the title and abstract screening were then assessed for inclusion (Figure 2). In total, 13 peer-reviewed articles were included in this review.

Figure 2

Review Flow Diagram



Results

Anticipatory Grief Terminology

All the articles included in this review provide a theoretical or empirical definition of grief that parents experience prior to the death of their child, with most authors using the term AG to describe parents’ responses to past, present and anticipated losses in relations to their child. Most authors use Rando’s definition of AG which is, “the phenomenon encompassing the process of mourning, coping, interaction, planning and psychological reorganization that are

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stimulated and begun in part in response to the impending loss of a loved one and the recognition of associated losses in the past, present and future” (Rando, 1986, p.24). Other terms such as chronic sorrow (Batchelor & Duke, 2019), anticipatory loss (Yang et al., 2016) and anticipatory mourning (Rini & Loriz, 2007) are also used in the included studies to describe similar experiences. A common feature of these experiences is that grief occurs as a result of the irrevocable loss of an idealized child or of the irrevocable future loss of a child. In this literature review, both AG and anticipatory mourning will be used to reflect the terms used in the different studies.

Study Characteristics

Both qualitative (n=6) and quantitative (n=7) studies were included in this review. In this section, these studies will be discussed in terms of their study design, including study population and study location.

Qualitative Studies

Six of the studies included in this review used qualitative methods to examine parental AG (Batchelor & Duke, 2019; Dighe et al., 2008; Higgs et al., 2016; Lucca & Petean, 2016; Rini & Loriz, 2007; Yang et al., 2016). All of these studies are cross-sectional and use semi-structured interviews, either face-to-face or via phone. Most authors do not specify their qualitative approach, except Yang et al. (2016) and Batchelor and Duke (2019) who specified that their study methodology was based on hermeneutic phenomenology principles.

Quantitative Studies

Seven studies in this review were quantitative and two different multidimensional tools were used to assess the emotional, social, and relational aspects of living and caring for a sick child who will die in a foreseeable future (Al-Gamal, 2013; Al-Gamal E et al., 2009; Al-Gamal

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& Long, 2010, 2014; Benfield, 1976; Valizadeh et al., 2013; Zamanzadeh et al., 2013). Authors either used a modified version of the Marwit-Meuser Caregiver Grief Inventory (MM-CGI) (MM-CGI Childhood Cancer and MM-CGI Cerebral Palsy) or an AG scale that includes seven questions. Items on the AG scale measure feeling of sadness, anger, sleeping difficulty, loss of appetite, mental preoccupation, irritability and guilt, while the MM-CGI comprises 50 items measuring physical, emotional, cognitive and social responses that are thought to be related to AG. The MM-CGI contains 3 subscales: personal sacrifice burden, heartfelt sadness and longing, and worry and isolation. Two of the seven quantitative studies focused on the development and testing of the modified versions of the MM-CGI (Al-Gamal et al., 2009; Al-Gamal & Long, 2014)

Study Population and Study Geographical Location

Studies either focused on parents of children with a specific disease (spinal muscular atrophy, Duchenne muscular dystrophy, cancer, cerebral palsy, chronic illness) or at parents in a specific context (newborns in neonatal intensive care units, children who died while hospitalized). Researchers studied mothers only (n=1), fathers only (n=3) or both parents (n=9). Three studies were conducted in the United States (Batchelor & Duke, 2019; Benfield et al., 1976; Rini & Loriz, 2007), while the others were conducted outside of North America (Australia, India, Brazil, Taiwan, Iran and Jordan) (Al-Gamal, 2013; Al-Gamal et al., 2009; Al-Gamal & Long, 2010, 2014; Dighe et al., 2008; Higgs et al., 2016; Lucca & Petean, 2016; Valizadeh et al., 2013; Yang et al., 2016; Zamanzadeh et al., 2013).

Key Themes from the Literature

This section presents the major themes discussed in the literature: characteristics of AG, factors impacting AG and impact of AG on bereavement.

Anticipatory Grief as a Result of Anticipatory Loss

Anticipatory grief occurs as a result of the anticipated loss of a child and/or the loss of an idealized child. The process of loss and associated grief starts when symptoms are first perceived and it worsens due to the physical and emotional overload that accumulate as a result of the child's disease progression (Higgs et al., 2016; Lucca & Petean, 2016). Parents grieve as they witness their child's health progressively decline and as they continue to anticipate future losses such as the loss of their child's ability to walk and eat orally (Lucca & Petean, 2016; Yang et al., 2016). In addition to losses directly related to the physical and/or cognitive abilities of the child with a LLC, AG may be compounded by grief related to other aspects of living with and caring for a child with a serious illness, e.g. witnessing the loss of a the child's relationships with their siblings, or loss of reproductive freedom in the case of hereditary diseases (Higgs et al., 2016).

Responses of Anticipatory Grief

Based on the tools used in quantitative studies (MM-CGI and anticipatory grief scale), we can infer that multiple authors agree that sadness, feeling depressed, anger, irritability, preoccupation, worrying, feeling isolated, as well as physical symptoms such as loss of appetite and difficulty sleeping are common responses of AG in parents. Similar characteristics to the ones mentioned previously were reported in qualitative studies. Parents expressed feeling sad and depressed and felt isolated (Batchelor & Duke, 2019; Dighe et al., 2008; Yang et al., 2016). Isolation was described by parents as losing social connection due to loss of work and career as a result of becoming the caregiver of a child with complex needs, but also due to lack of understanding by other people of the reality of caring for their child (Batchelor & Duke, 2019). Many parents also expressed feeling helpless because they are not in control of the situation and they cannot change the fate of their child (Lucca & Petean, 2016; Yang et al., 2016).

Factors Impacting Anticipatory Grief

Gender. Four studies addressed the differences between mothers and fathers in their experience of AG (Al-Gamal, 2013; Al-Gamal & Long, 2010; Benefield et al., 1976; Valizadeh et al., 2013). Three of these studies did not report any statistical differences between mothers and fathers in overall AG scores (Al-Gamal, 2013; Al-Gamal & Long, 2010; Valizadeh et al., 2013). Al-Gamal (2013) and Al-Gamal and Long (2010) showed that both mothers and fathers' total AG and individual subscale scores were not statistically different. Similarly, in Valizadeh et al.'s study (2013), mothers and fathers' total AG scores were not significantly different; however, mothers expressed more sadness than fathers, and fathers expressed more anger than mothers. Psychological symptoms (e.g. sadness, irritability, etc.) were reported more often than physical symptoms (e.g. sleep disturbances, loss of appetite) in both mothers and fathers and the AG responses that were the most reported were "feelings of sadness", "preoccupation thinking or dreaming about baby" and "feeling of anger".

Benfield et al.'s (1976) study results are contradictory. In their study of 101 mother-father pairs, the mean maternal AG scores significantly exceeded the paternal mean scores. Looking at the items individually, significantly more mothers reported feelings of sadness, loss of appetite and feelings of guilt. There was also a significant difference between mothers and fathers on the following items: "feeling depressed", "being in disbelief" and "crying and praying for their child".

Interestingly, most qualitative studies that included both mothers and fathers do not consider gender differences, other than Rini and Loriz (2007), who state that due to their study sample size (n = 11 (2 fathers)), the influence of gender or parental role on anticipatory mourning could not be determined.

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Uncertainty. Parents who care for a child with a serious illness live in uncertainty. They fear that any day could be the day their child dies (Lucca & Petean, 2016; Yang et al., 2016). Feeling anxious and depressed were emotions commonly reported in Yang et al.'s (2016) study as a result of the uncertainty related their child's condition and inability to predict the future. This is consistent with results from Zamanzadeh et al.'s (2013) study of parents of neonates in the neonatal intensive care unit, in which grief responses were also heightened due to uncertain outcomes of the disease.

Age of the Child, Illness Severity and Duration of Illness. Findings on the relationship between AG and the age of the child, duration of illness and illness severity are contradictory. Rini and Loriz's (2007) study showed that the neither the duration of illness nor the amount of time the child was hospitalized before death influence how parents described their experience. In their study with fathers of children who have Duchenne muscular dystrophy, Lucca and Petean (2016) reported that "anticipatory mourning worsens with physical and emotional overload accruing from the child's condition" (p.3087). Similarly, in a study of parents with children who have cerebral palsy, results showed that the older the child was, the more parents reported personal sacrifice burden (Al-Gamal, 2013). The authors hypothesized that this could be attributed to physical strain on parents related to older children requiring, for example, more heavy lifting and assistance with bathing. Al-Gamal and Long's (2010) study of parents of children with cancer showed different results. No correlation was found between AG scores and the child's age. Furthermore, parents of children newly diagnosed with cancer experienced more AG responses than parents of children who had been diagnosed 6-12 months earlier, with most parents of newly diagnosed children expressing difficulty accepting what was happening, while less than half of the parents in the other group felt this way.

Support and Coping. Coping strategies, including having a good support system, were identified as helpful by families who have a child with a LLC (Batchelor & Duke, 2019; Benefield et al., 1976; Higgs et al., 2016; Lucca & Petean, 2016; Yang et al., 2016). Families often emphasize the positive role support groups, family members and neighbours play in supporting them through their journey (Dighe et al., 2008; Higgs et al., 2016; Batchelor & Duke, 2019). In many studies, parents also expressed spiritual components as a source of strength and coping (Batchelor & Duke, 2019; Benefield et al., 1976; Lucca & Petean, 2016; Yang et al., 2016). Religion and spirituality can act as protective factors and help parents cope with uncertainties and find a purpose in their experience, helping them transform their grief into the strength to live life to the fullest (Yang et al., 2016).

Relationships With Health Care Providers. Attitudes and actions of health care providers at the end-of-life may have a profound effect on parents. Honesty, compassion and consideration of parent's needs are seen by parents as having positive effects on their experience at the end-of-life of the child, while the opposite showed to negatively affect the anticipatory mourning process (Rini & Loriz, 2007). Furthermore, studies show that parents sometimes feel misunderstood by health care providers who disregard their expertise in their child's care (Batchelor & Duke, 2019; Yang et al., 2016). In Yang et al.'s (2016) study, parents felt that they had more experience in caring for their child than health care providers, and this contributed to parents feeling depressed, sad and frustrated. For parents, receiving clear, sufficient and timely information from empathetic health care providers at the time of diagnosis and during the disease trajectory including at the time when death is imminent supported informed and shared decision-making (Lucca & Petean, 2016; Rini & Loriz, 2007). Being able to participate in shared

decision-making with the child's health care team, and being able to focus on common, achievable goals were especially important.

The Role of Sociocultural Contexts. Some authors mention the role of sociocultural influences in parents' experience of living and caring for a child with a LLC, which can affect the AG process. Yang et al (2016) states the "Chinese culture embraces the concept of destiny, and some parents believe that when their children have a rare disorder, the children are not favoured by their grandparents" (p.2655). Parents felt that grandparents were ashamed of their grandchild with spinal muscular atrophy, which added to the burden and grief. Similarly, a study of Indian families who care for a child with cancer, a number of parents felt that their family and their child experienced social stigmatization (Dighe et al., 2008). They also mentioned that family members were the main source of support, which, as per the authors, reflect Indian cultural values. Controversies regarding mother or father roles and what is expected from the different family roles are also discussed (Batchelor & Duke, 2019; Lucca & Petean, 2016). How mothering and fathering is socially portrayed impact how parents who have a child with a LLC cope.

Anticipatory Grief and Bereavement

One study explored parents' perspective on the relationships between anticipatory mourning and bereavement (Rini & Loriz, 2007). In this study, parents who were aware of their child's imminent death mentioned that having the time to grieve prior to their child's death helped to facilitate bereavement. Parents who were unaware of their child's imminent death expressed that they felt unprepared. They stated that their bereavement would have been positively impacted if they had the time to begin the tasks associated with anticipating and preparing for their child's death.

Discussion of the Review of the Literature

This literature review presents results from a literature synthesis of thirteen research studies on AG experienced by parents of children up to 19 years of age. While AG terminology continues to be ambiguous and various terms are being used interchangeably in the literature, the consensus seems to be that AG occurs as a result of the irrevocable loss of an idealized child or the irrevocable future loss of a child. AG and anticipatory mourning have many similarities and appear to be a form of response to anticipatory loss, while chronic sorrow is a different concept that shares conceptual boundaries with AG and anticipatory mourning.

Different methodologies were used to study AG in parents. Because the concept of AG remains vague and poorly operationalized, using a qualitative design allows the exploration of the subjective experiences of parents. In the seven quantitative studies included in this review, two different AG measurement tools were used, one consisting of 50 items, the other consisting of seven items. Measuring AG has been an ongoing and unresolved issue in the literature. Nielsen et al. (2016) found that in 14 studies, six instruments that were substantially different were used to measure AG, and no more than three studies used the same tool. The lack of clarity about the AG conceptual definition makes the operationalization of AG challenging.

The existing literature on AG in parents varies in terms of study populations. Studies of parents whose children have various diseases were included in this review which made comparison of findings difficult; there are great differences between caring for a child who is fully independent and caring for a child who depends on their caregiver for all activities of daily living. These differences could impact the intensity of grief and may explain why findings on the relationships between AG and the age of the child or time since diagnosis are contradictory.

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The studies included in this review were conducted in various geographical locations. Lehto and Stein (2009) argue that the process of AG is strongly influenced by social and cultural representations of death and dying. While grief is universal, important differences exist across cultures in how grief is expressed and understood (Rosenblatt, 2008). Interestingly, the majority of studies on parental AG in the pediatric context were conducted in countries outside of North America. In Western culture, there is a pervasive fear of death and some current death and grief discourse reinforce the idea that the expressions and feelings of grief should be kept private. While Western societies appear to become increasingly open to conversations about death, denial of grieving is still favored (Tradii & Robert, 2017; Zimmermann, 2007). Death-denying can be seen as a way to preserve and pursue happiness (Ariès, 1975). Nevertheless, there is sufficient data to suggest that AG is a multidimensional concept experienced across various cultures. Different cultures share commonalities but also show differences in how AG is expressed, felt and understood. Regardless of the study locations, parents often report feeling depressed and experiencing sadness, anger, isolation and feeling worried as a consequence of having a child with a serious condition. The literature addresses gender differences and shows that both mothers and fathers experience AG at similar intensity levels, although they may express their grief differently. Mothers and fathers are expected and encouraged to grieve in gendered ways, and these reactions are expected to align with societal expectations of mothering and fathering and men and women (Macdonald et al., 2010). Whether emotional reactions are considered appropriate or disproportionate is often based on mainstream norms.

Various other factors appear to play a role in parents' experience of AG, notably uncertainty, support and coping and relationships with health care providers. Living with a child who has a LLC involves adjusting to continuous uncertainty, which can cause/exacerbate AG.

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The literature shows that uncertainty is associated with self-reported psychological outcomes for parents (Szulczewski et al., 2017). Parents will often turn to friends and families, as well as find strength in spirituality and religiosity to cope with uncertainties and with physical and emotional demands resulting from their child's diagnosis. The meaning of a disease is permeated by the parent's belief and value system and by spirituality and/or religion, with some parents thinking they have been 'chosen' (Lucca & Petean, 2016).

In addition to having a good support network and positive coping mechanisms, parents feel supported when they have good and trusting relationships with health care providers (Davies et al., 2017). Parents who have a child with a LLC have no control over their child's symptoms and eventual death, so having their voices heard by health care professionals and being able to share their knowledge about their child and participate in decision-making is empowering. Optimal parent-provider relationships are those where health care providers communicate effectively and with compassion, support emotions, acknowledge uncertainty and promote shared decision-making (Hauer & Wolfe, 2014; Jones & Wolfe, 2014), while insensitive parent-provider interactions may have a negative influence on how parents cope with their child's illness and may affect bereavement (Contro et al., 2004; Davies et al., 2007; Macdonald et al., 2005).

Only one study addressed the role of anticipatory mourning in bereavement. Findings from Rini and Loriz (2007) study of parents of children who died from various conditions while hospitalized showed that parents felt that their bereavement would have been impacted positively if they had had the opportunity to experience anticipatory mourning. Contradictory results were found in a literature review on AG in caregivers during the end-of-life of an adult loved one

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(Nielsen et al., 2016). Nielsen et al. (2016) concluded that grief prior to death could be considered as a risk factor for poor bereavement outcomes.

Gaps in the Literature

Based on the review of the literature, gaps in the knowledge of AG in parents were identified. First, the concept of AG is operationally vague and the literature mainly focuses on parents of neonates, and parents of children with neuromuscular conditions, cerebral palsy or cancer. A deeper understanding of the concept of AG, especially in parents who have children with different pediatric LLC, is needed. This literature review also showed that factors that have a potential impact on AG, such as coping, parent/health care provider relationships and sociocultural contexts, deserved to be further investigated.

These research gaps guided the development of the following research questions:

- How do bereaved parents of children with a NMLLC describe their experience of AG?
- How do parents describe factors (interpersonal, intrapersonal, health care related) that may have influenced their AG experience?

Summary

The goal of this review was to examine the existing literature on AG in parents of children and young adults up to 19 years of age. Similar to grief after losing a child, parental AG is a ubiquitous experience that remains poorly studied. The findings from the literature confirm the multidimensionality of AG, but because of the different study methodologies used, different populations studied and different study locations, the comparison and generalizability/transferability of results is limited. Further studies are needed to gain a better understanding of the phenomenon of AG. By doing an in-depth exploration of the experience of

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AG in parents who have a child with a NMLLC, this research makes a significant contribution to the limited existing knowledge base. Capturing the whole picture of AG in parents provides opportunities for the development and implementation of therapeutic interventions.

Chapter 3. Theoretical Scaffolding

Introduction

The literature review presented in Chapter 2 revealed that parents who have a child with a NMLLC continuously adapt to and cope with past, present and future losses. The theories and concepts of attachment, ambiguous loss and chronic sorrow are especially relevant to the experience of AG in parents and therefore may provide a useful lens through which I can examine AG. This section will briefly examine John Bowlby's Attachment Theory, the Middle Range Theory of Chronic Sorrow and the concept of Ambiguous Loss.

Attachment Theory

The importance of attachment between parents and children is well documented (Ding et al., 2014; Feeney, 2000; Wittkowski et al., 2020). Attachment theory emerged from John Bowlby's early studies of children who were deprived of maternal care (Bowlby, 1952). He concluded that "the prolonged deprivation of the young child of maternal care may have grave and far-reaching effects on his character and so on the whole of his future life" (Bowlby, 1952, p.46). Bowlby's attachment theory suggests that in their first year of life, infants develop a bond with their caregivers that serves a purpose of protection and self-preservation. According to Bowlby (1973), attachment behaviors are biologically rooted and promote proximity, affection and connectedness to another person. Bowlby (1973) believed that attachment behaviors are most readily manifested in situations that are stressful or threatening. For example, when a child is in a distressing situation, they may seek comfort by crying, and will return to a calm state once they are comforted by a figure of attachment. Early relationships and attachment experiences create a mental script that Bowlby referred to as an Internal Working Model (Bretherton & Munholland, 2008). An internal working model consists of internal representations of the self

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and of others and of the self in relation to attachment figures. It influences a wide range of psychological functions, and it guides interactions with attachment figures (Bretherton & Munholland, 2008).

The nature of the relationship between infants and their caregivers has an impact on attachment in relationships later in life (Bretherton & Munholland, 2008). As per Bowlby (1973), internal working models of self and others act as a template and guide cognitive processes and attachment behaviors in all subsequent relationships. Habitual response patterns in attachment relationships are referred to as attachment styles (Cobb & Davila, 2009). Attachment styles affect how an individual will interpret their social world, and how they will express and try to meet their attachment needs (Cobb & Davila, 2009). Individuals who have a secure attachment style tend to show more resiliency when experiencing grief than individuals with less secure attachment styles (Uren & Wastell, 2002). The degree of attachment security, influenced by a person's attachment style, was shown to be a predictor of grief acuity in bereaved parents who experienced stillbirth or neonatal death (Uren & Wastell, 2002). The sensory deprivation a parent feels in their inability to fulfill their caregiving role and responsibilities after their child died may be a contributing factor to the intensification of grief (Schatz, 1986, as cited in Uren & Wastell, 2002). It is apparent from attachment theory that the attachment style and the nature of the bond between a parent and child who has a LLC may be affected prior to death.

Attachment theory also suggests that stress alters family function (Bowlby, 1982). I therefore hypothesized that the stress and burden of care experienced by parents who have a child with a NMLLC may jeopardize attachment bonds. In addition, studies on attachment have shown that caregivers possess an instinctive caregiving system that aim to protect the physical and psychological wellbeing of another person (Shear et al., 2007) and caregiving may provide a

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sense of wellbeing for the caregiver as well (Deci et al., 2006). Examining AG from the perspective of attachment theory may partly explain the impact of AG on the wellbeing of parents.

Ambiguous Loss

The term “ambiguous loss” was coined by Pauline Boss in the 1970s to describe the loss experienced by families of soldiers missing in action (Boss, 2007). Ambiguous loss encompasses the grief that is experienced by individuals when “a person is both physically present and psychologically unavailable, or psychologically present, yet physically unavailable” (Boss, 1999, p.11). It is a loss that is incomplete, unclear and often uncertain (Boss, 1999, 2007).

The main difference between ordinary loss and ambiguous loss is that ambiguous loss does not allow for the grief process to get resolved (Boss, 2007).

Since its initial conceptualization, the concept of ambiguous loss has been studied in various contexts, including situations where children are separated from their parents (Luster et al., 2009) and families dealing with chronic illness (Boss & Couden, 2002; O’Brien, 2007). Based on this increasing application of the concept, ambiguous loss offers a solid framework on which to construct a deeper understanding of the experience and the impact of AG in the context of caring for a child with a LLC.

Parents who have a child with a chronic disease face a situation where the healthy child they once had is now changed (O’Brien, 2007). Parents may perceived the child as being physically present but psychologically unavailable, or vice-versa, thereby experiencing ambiguous loss. There is a long and painful grieving process associated with ambiguous loss (Dupuis, 2002). It also results in confusion, uncertainty (Faber et al., 2008), and ambivalence (Lee & Whiting, 2007). In referencing ambiguous loss, Boss (2006) noted that when an

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individual is not in control of an aspect of their life, oftentimes they have difficulties in coming to terms with that aspect. Boss (2006) recommends remaining flexible and choosing to accept the ambiguity and uncertainty, rather than focusing on gaining full control or surrendering completely and she suggests that this acceptance and tolerance for ambiguity can strengthen resiliency.

The ways with which parents who have a child with a LLC cope with ambiguous loss could impact their experience of AG. The concept of ambiguous loss therefore offers a good theoretical lens through which AG and the complex dynamics inherent to caring for a child with a LLC can be explored.

Chronic Sorrow

Chronic sorrow is described as an expected and normal “set of pervasive, profound, continuing, and recurring grief responses resulting from a loss or absence of crucial aspects of oneself (self-loss) or another living person (other-loss) to whom there is a deep attachment” (Roos, 2017, p.25). The concept was first described in 1962 by Simon Olshanky, a clinical psychologist who in the 1950s studied parents of children with a diagnosed mental disability (Batchelor & Duke, 2019). Chronic sorrow was the overarching term used to describe the long-term grief and episodic sadness experienced by these parents. Since then, the concept was adapted by Burke et al. (1992) and became incorporated as a core essence of a middle-range nursing theory developed by the Nursing Consortium for Research on Chronic Sorrow in the 1990s (Burke et al., 1992). The Nursing Consortium for Research on Chronic Sorrow characterized the ‘chronic’ part of chronic sorrow as “pervasive, permanent, periodic, and potentially progressive” (Eakes et al., 1998, p.180). Antecedents that have been widely identified

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in the literature as possible causes of chronic sorrow include adversity, loss, sadness, disparity, and recurring or repeated loss experiences (Eakes et al., 1998; Teel, 1991).

According to the middle range theory of chronic sorrow, the chronic sorrow cycle begins when one becomes aware of the onset of a non-finite loss (Harris & Winokuer, 2020). This non-finite loss may be a single, isolated occurrence, or it may be part of a series of ongoing losses (Harris & Winokuer, 2020). The loss creates a gap between what a person expected from a situation and what actually ensued as a result of the loss. The middle range theory of chronic sorrow has been useful to explore grief and loss experiences in multiple settings and populations, including in parents of a child who has a chronic condition (Batchelor & Duke, 2019; Coughlin & Sethares, 2017; Eakes et al., 1998). Parents who have a child with a chronic condition face many losses that are continually redefined as the child's condition progresses, and the perpetual layering of losses subsequently requires them to continually adapt (Batchelor & Duke, 2019; Eakes et al., 1998; Northington, 2000; Roos, 2017). Once a loss has been experienced, the cycle of chronic sorrow can be triggered again by subsequent losses such as progression of the disease or step-back of milestones their child had already achieved (Eakes et al., 1998).

Chronic sorrow is often exacerbated by accumulating stressors and by the complexity of illness (Melnik et al., 2001). Within the middle range theory of chronic sorrow developed by the NCRCS, internal (personal) and external (interpersonal) coping strategies are used to address chronic sorrow and are part of the caregiver adaptation to their situation (Eakes et al., 1998; Olwit et al., 2018). Because of the conceptual boundaries shared between chronic sorrow and AG, I anticipate that the coping mechanisms suggested by the middle range theory of chronic sorrow overlap with mechanisms for coping with AG.

Summary

In this chapter I have discussed three major theories and concepts which have scaffolded this study. Attachment, ambiguous loss, chronic sorrow are concepts that are closely intertwined with AG. These concepts and theories provide perspectives that guided an in-depth and informed understanding of the phenomenon of AG in parents who have a child with a LLC.

Chapter 4. Methodology

Introduction

This chapter provides an overview of the research methodology used to answer the research questions guiding this study which are: 1) how do bereaved parents of children with a NMLLC describe their experience of AG? and 2) how do parents describe factors (interpersonal, intrapersonal, health care related) that may have influenced their AG experience? First, I will give an overview of the study's research paradigm, methodological framework, philosophical underpinnings and theoretical scaffolding. Then, I will discuss research methods including strategies to enhance quality and trustworthiness as well as ethical considerations of doing this kind of complicated emotional qualitative study.

Interpretivist/Constructivist Research Paradigm

A strong research design requires a research paradigm that aligns with the researcher's beliefs about the nature of reality (Mills et al., 2006). This study has been informed and grounded by an interpretivist/constructivist paradigm. The interpretivist/constructivist paradigm posits that "knowledge is relative to particular circumstances - historical, temporal, cultural, subjective - and exists in multiple forms as representations of reality (interpretations by individuals)" (Benoliel, 1996, p.407). There are multiple realities that are socially constructed, and subjective meanings are formed through social interactions and inherently embedded in historical and social contexts (Creswell, 2017). The researcher and the researched interact and influence each other; the knower and the known cannot be separated (Thorne et al., 2004).

Interpretive Description Methodological Framework

To gain a better understanding of AG and of the subjective, experiential perspectives of parents who have child who died from a NMLLC, this study used an interpretive description (ID)

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methodological framework. ID guides sound empirical research within the nursing discipline for which traditional qualitative research methodologies are sometimes ill-fitted (Thorne, 2016). ID was developed as a methodological framework specific to disciplinary knowledge. Within nursing, ID encourages nurse researchers to answer questions derived from applied practice in order to generate findings that are pertinent to the discipline. ID is a flexible, non-categorical approach. There are various ways in which all the dimensions of research can be enacted as long as the decisions made throughout the research process stay consistent with ID logic (Thorne, 2016). ID studies may use a combination of techniques used in grounded theory, or ethnography to collect and analyse data. While research design can vary between studies using ID, each tool is chosen in a specific way to inform and support guiding principles of integrity, logic and validity.

The epistemological and ontological assumptions of ID, which will be discussed in the next section, align with my own personal assumptions about the fundamental nature of reality and knowledge generation. Within constructivist theory, each person makes sense of experiences through a specific sociocultural lens. Consequently, the meanings ascribed to objects, emotions, or phenomena are influenced by socialization. In nursing clinical practice and nursing research, interactions between patients and nurses or between researchers and participants are mediated by the meanings ascribed to these interactions by all people involved. In a qualitative study, data collection and interpretation are part of the social contexts in which the interviews took place and reflect the interaction of the participants and the researcher in the creation and analysis of data. This constructivist viewpoint suits the study of complex phenomena like AG. The outcomes of my ID study will build on existing clinical knowledge of AG in parents so nurses can improve holistic care of parents who have a child who has a NMLLC.

Philosophical Underpinnings and Assumptions

Ontological and Epistemological Assumptions

The philosophical underpinnings of ID align with a relativist ontology and subjectivist epistemology. Relativist ontology is based on the belief that reality is a finite subjective experience (Denzin & Lincoln, 2011). The ID researcher acknowledges that within specific contexts, ‘truth’ is relative and multiple realities exist. From a subjectivist epistemological perspective, knowledge is “always filtered through the lenses of language, gender, social class, race, and ethnicity” (Denzin & Lincoln, 2011, p. 21).

Guiding Principles of Interpretive Description

Key axioms of naturalistic inquiry provide the philosophical underpinnings for ID (Lincoln & Guba, 1985). ID studies are conducted in a naturalistic context and ID researchers respect the ethical rights of participants, acknowledging power differential and that decisions made throughout the research process can impact participants (Thorne, 2016; Thorne et al., 2004). ID also pays attention to the value of subjective and experiential knowledge, recognizing the expert perspective that people have of their own experience. ID is attentive to the spatial, temporal and cultural context of the phenomenon of interest and recognizes that there are multiple realities that may or may not be shared (Thorne, 2016). Another important guiding principle of ID is that it capitalizes on human commonalities and individual variances, recognizing the importance of framing findings so that it can illuminate diversities (Thompson Burdine et al., 2020). ID acknowledges that the researcher and the researched relationally influence one another, and this inadvertently impacts research outcomes. Therefore, the knowledge generated from ID research can be considered, to some extent, co-produced (Thorne et al., 2004).

Research Methods

Sample

Purposive sampling was used to recruit bereaved parents with a child who died from a NMLLC. Qualitative methodologies are not prescriptive about sample size parameters, and ID in particular supports a sample that provides enough data to inform a study in order to inform an inductive generation of knowledge. A small sample size does not uncover everything there is to know about AG, but it still add to the grief literacy and it increases AG awareness and highlights the need for AG in parents to be better acknowledged.

Purposive sampling ensured a good variation between the various NMLLC the deceased child was affected with, therefore offering a better representation of the experience of AG across various NMLLC diagnoses. Thorne (2016) warns us that true representation cannot be achieved, but rather, the researcher must think of representation as reflecting “a certain kind of perspective built from an auditable set of angles of vision whose nature and boundaries we can explicitly acknowledge and address” (p.97-98).

Eligibility criteria. Parents were eligible to participate in the study if they spoke English, if they had a child with a NMLLC, and if their child had died 12 months to 5 years prior to recruitment. One or both parents were invited participate. Parents of children who I directly cared for in the end-of-life period were excluded.

Recruitment and Enrolment. Eligible parents were identified by the researcher with the help of the Canuck Place Children’s Hospice counseling team manager. Potential participants then received an email from the Canuck Place Children’s Hospice receptionist with an invitation to participate in the study and the study letter of information (Appendix B). Potential participants were asked to contact the researcher via email if interested. A phone call was then set up with

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each parents who were interested to describe the study in more details and go over the consent form. A date and time for the interview was scheduled with each participants. Implied consent was granted from the participants by the overt action of signing into an invitation only and password protected private online Zoom meeting room. Consent was reaffirmed verbally at the start of each interview.

Data Collection

Data was collected via semi-structured interviews (Appendix C) via Zoom and the interviews took place between August 2022 and December 2022. Due to the rich nature of the subjective AG experience of participants, phenomenological-type interviews were appropriate as the data-collection tool. While allowing for a free-flowing conversation, semi-structured interviews ensure that key topics are covered during the interview, therefore providing consistency in data collection. Interview questions were be open-ended and served as a guide to facilitate the dialogue. I used active listening techniques, such as using prompts to promote open responses and encourage participants to reveal more. However, during the interviews value-laden prompts (e.g., “good”, “I agree”, “I understand”) were avoided as much as possible in order to avoid steering conversations or preferencing certain types of responses (Thorne, 2016).

The interview were scheduled at a time that was most convenient to participants. Each interview lasted between 75 and 90 minutes. Consent was reaffirmed at the start of the interview and the participants were informed that they might decide to end the interview at any time if they felt uncomfortable with any of the questions asked. Interviews were video and audio-recorded to enable transcription accuracy (Polit & Beck, 2013). With participants’ permission, I took notes during the interviews while ensuring that my primary focus remained on participants’ unfolding

narratives (Thorne, 2016). Each interview was transcribed via NVivo Transcription, and I reviewed each transcript for accuracy.

Demographic information. Demographic information was collected at the beginning of the interview to help situate the experience of participants within their context. Demographic information included gender, age of the parent, relationship to the child, date of birth of the child, year of onset of symptoms/diagnosis, child's diagnosis, year of child's death and location of death. A description of the demographic data collected can be found in Appendix D.

Reflexivity

Reflexivity enhances and strengthens the credibility of qualitative studies. It is a mechanism through which the researchers are transparent about the assumptions they bring to the research (Guillemin & Gillam, 2016). ID acknowledges that there is a use for a priori knowledge and clinical observations, and that these often act as catalysts for nursing inquiry (Thorne, 2016).

Preconceptions, theoretical allegiances, clinical opinions, prior knowledge, decision audit trail, and evolving interpretations of the data are only a few examples of what can be found in the reflexive notes of an ID researcher. Journaling was an important element in supporting my inductive and iterative analytic process (Thorne et al., 2004). Field notes were written in a reflexive journal immediately after each interview to document my initial thoughts, impressions, and analytic ideas.

Situating myself within the research role

I am aware that my professional role as a nurse may have influenced my ability to effectively conduct research interviews (Thorne, 2016). Despite my professional experience as a pediatric palliative and pediatric critical care nurse working with the same population as the one I interviewed, I considered myself a "curious learner" by acknowledging the value of what I was

learning from my interactions and conversations with the participants. (Thorne, 2016, p. 140).

Despite the influence my clinical nursing role could have on the interview process, I felt obliged to disclose my professional role to the participants. This information was disclosed in both the letter of information and consent, and again at the start of the interview. I acknowledged that this may have influenced the information participants have shared with me.

Data Analysis

In ID, inductive data analysis occurs concurrently with data collection and “requires that we learn to see beyond the obvious, rigorously deconstructing what we think we see, testing hunches as to how it might fit together in new ways, and taking some ownership over the potential meaning and impact of the outcomes that we will eventually render as findings” (Thorne 2016, p. 156). ID does not provide a step-by-step process for data analysis. The focus in the initial stages of the analysis is on using broad-based codes to allow the researcher to group bits of data that might (or might not) be thematically related, and examine these bits of data as a whole and also in relation to each other, so that the researcher can form a good understanding of the whole data set (Thorne, 2016). The goal is to identify and group data that has similar attributes, and to compare and contrast groupings that have different properties.

Data analysis began after the first interview. To familiarize myself with each transcript, I read each transcript multiple times (Thorne 2016). While reading each transcript, I also watched the video recording to fully engage with the interview simultaneously (Thorne, 2016). This allowed me to “develop a “feel” for who participants are, what their storylines represent, and what the many aspects of their accounts could represent.” (Thorne, 2016, p.167). As I read the transcripts, I highlighted passages that appeared to share thematic similarities and assigned

broad-base codes to these data bits (Thorne, 2016). Passages containing powerful quotes were also flagged for potential use during the process of writing findings.

Data analysis oscillated between analyzing individual transcripts and looking for similarities and differences between the different transcripts, and from data analysis back to data collection (Thorne, 2016). These iterations are driven by constant comparison (Thorne, 2016; Thorne et al., 2004). While most often associated with grounded theory, principles of constant comparative analysis may be borrowed by ID researchers (Thorne, 2016). This method of analyzing data involves comparing parts of the data, looking for similarities and differences across and between participants. It supports consistent and plausible interpretations that are close to the data (Thompson Burdine et al., 2020). Using constant comparison allows both the researcher and the study participants to "co-construct" a coherent narrative that can support clinical practice (Teodoro et al., 2018).

In ID, findings do not consist of simply reporting patterns found in the data. It is rather an exploration of what the pieces of data mean, individually as well as in relation to other data pieces and what the foundations of these relationships may be (Thorne, 2016). Throughout my analysis, I continuously asked myself the following questions: "What are the data telling me? What is it I want to know? What is the dialectical relationship between what the data are telling me and what I want to know?" (Srivastava & Hopwood, 2009, p.79). I considered the multiple ways pieces of data could fit together into themes and what are the implications of each configurations were. This process involved re-grouping the data according to my evolving interpretations. By critically looking at my data from multiple angles and trying multiple configurations of the data, I avoided premature closure, which occur when the researcher has not completed an in-depth data analysis and stops at the level of superficial findings (Connelly &

Peltzer, 2016). In addition, in the initial stages of data analysis, I did not give too much weight to words and ideas that occurred more frequently in the data set. I also considered and questioned the importance and the meaning of words/ideas that did not occur as frequently as other ideas in my data, as when words or ideas are seen very infrequently, it does not mean that they are not meaningful (Thorne, 2016). To ensure I did not steer away from the goal of my inquiry, I kept nursing practice in mind. A researcher's disciplinary scaffolding can be extremely useful to ensure that they stay focused and on the right track, however, they must remain flexible and open to interpretations that are not totally consistent with typical reasoning of their discipline (Thorne, 2016). Every nursing researchers using ID should strive to find a good balance between being fully open to new interpretations and "staying true" to the nursing discipline. I also acknowledge that because of the nature of the researcher-researched relationships, the researcher becomes part of the story (Thorne, 2016), so the interactions between myself and the research participants inadvertently influenced the data. I used reflexivity to help me "stay out of the way" of my research as much as possible (Sandelowski & Barroso, 2002).

It is important to note that throughout the process of data collection and analysis, I met several times with my supervisors to discuss data collection and my evolving analysis.

Quality and Trustworthiness

In establishing quality and trustworthiness of qualitative research, including in ID studies, Lincoln and Guba (1985) created the following criteria: credibility (validity of the conclusions that are drawn from the data), dependability (extent to which similar findings would be obtained if the study were repeated), confirmability (degree of objectivity of the researcher in data collection and reporting) and transferability (how well the study conclusions can be applied to other similar settings). In this study, various strategies were used strengthen the credibility,

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transferability, dependability and confirmability of my findings: prolonged engagement and follow-up interviews with participants, regular meetings with my supervisors to discuss my research process, purposive sampling approach, keeping an audit-trail of my analytical decisions as well as reflexive journaling.

There are additional “quality attributes” that ID must possess (Thorne, 2016). ID is expected to show:

- Epistemological integrity: Assumptions researcher makes about the nature of knowledge must align with the research design. Epistemological integrity is demonstrated by the alignment of my research questions with the stated epistemological standpoint and with the meaningful interpretations of my data.
- Representative credibility: The theoretical claims must be consistent with the study sample. I demonstrated representative credibility by avoiding claims that my findings are generalizable/transferable to all parents who have a child with a NMLLC. Credibility will also be strengthened by having prolonged engagement with participants (75-90 minutes interviews). Member-checking was not employed in this study, as in ID studies, conventional member-checking is generally not recommended. It may lead to false confidence in interpretations if participants confirms findings or can potentially steer one away from valuable analytic interpretations if participants disagree with the findings (Thorne & Darbyshire, 2005).
- Analytic logic: Researchers must show evidence that decisions made throughout the research process followed a logical path (Morse, 1994). Analytic logic was shown through keeping an audit-trail of all the decisions I made about my research process.

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- Interpretive authority: The researcher must ensure that their interpretations are trustworthy; their findings must reveal unbiased “truths” (Thorne, 2016). To support interpretive authority, I disclosed my own values, beliefs and biases throughout the research process. Keeping a reflexive journal, as well as writing field notes enhanced interpretive authority.

In addition to the factors mentioned above, Thorne (2016) suggests five additional criteria against which ID research ought to be judged:

- Moral defensibility: There needs to be convincing claims about the value of the knowledge the researchers will generate. Moral defensibility speaks to how we use knowledge in our society. Researchers must consider all the possible use of their research findings, especially when studying vulnerable populations (Thorne, 2016). The benefits to the study populations must outweigh the risks that the study findings may bring upon them, e.g. worsening stigma.
- Disciplinary relevance: Criteria for judging quality and trustworthiness of qualitative research are closely related to the particular discipline in which the study is conducted (Morrow, 2005). ID aims to generate knowledge within the applied disciplines, so it must generate knowledge that will expand disciplinary knowledge. This study generated knowledge and awareness to the experience of parents who have a child with a NMLLC. With this knowledge and awareness, nurses, especially those less familiar with working with this population and with pediatric palliative care principles, will be better equipped to support parents.
- Pragmatic obligation: Pragmatic obligation “reflects the inherent tension within practice realities, in which respect for the uniqueness of individuals creates sympathy

for an idealist epistemology at the same time that the moral mandate of practice discipline requires usable general knowledge” (Thorne, 2016, p.237). In other words, Thorne (2016) suggests that the researcher doing ID must have the moral responsibility and awareness that claims based on study findings may be taken as hard evidence used to inform clinical practice whether or not these findings are sufficiently developed to be applied to practice.

- Contextual awareness: ID researchers must develop awareness that their position within their discipline as well as their social and historical context influence their own perspectives. While techniques such a reflexive practice can mitigate the influence the researcher’s contexts have on their own perspectives, there may still be invisible shared assumptions between the researchers and the researched, and between the researchers and others in the field (Thorne, 2016).
- Probable truth: Searching for absolute truths may seem ludicrous, as it is nearly, if not completely, impossible to say with total confidence that study findings are entirely true (Denzin, 2009). ID researchers accept that there is value in revealing probable truths.

Ethical Considerations

The consideration of ethics in ID research is essential and the basic ethical principles that apply to all types of research involving human participants, such as “respect for persons”, “concern for welfare” and “justice” (Government of Canada - Panel on Research Ethics, 2014), also apply to ID studies.

Procedural Ethics

Ethics approval from the Athabasca University research ethics board (REB) was requested as well as approval from the Canuck Place Children’s Hospice in Vancouver, BC

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where participants were recruited. Parents who agreed to participate were asked to verbally consent after reading the informed consent form that outlined the study procedures. The consent form stated that participation was entirely voluntary and that participants could decide to withdraw from the study at any time. Throughout the study, I ensured that the data collected remain confidential; participant number were assigned to participants to protect their identity.

Relational Ethics

All relationships are moral in health care contexts (Austin, 2008), such that “ethical considerations occur in every situation, every encounter, and with every patient” (Bergum, 2013, p. 127). AG is a topic laden with emotions, making this research ethically challenging. These emotions can be positive and promote catharsis in participants, or negative, potentially causing distress (Silverio et al., 2022). It was therefore morally and ethically necessary to reflect on the risks versus benefits of research on participants. I provided a list of free online resources that parents could access if they were to experience intense negative emotions due to the sensitive nature of my research topic.

Due to the nature of qualitative inquiry, I, as the instrument of inquiry, was mindful of my influence on participants (Tracy, 2010). Being a nurse at the children’s hospice where participants were recruited, I remained aware of my dual role as both a nurse and a researcher and that an imbalance of power may exist in the relationship between myself and the participants, resulting in a threat to the ethics principle of “justice”. My nurse researcher role can be confusing for parents since I may have cared for their child at some point in their illness trajectory, and this definitely adds a layer of complexity to the researcher-researched relationship (González, 2000). I disclosed my clinical role to the participants as well as any potential conflicts of interest to the university REB and to the children’s hospice research committee.

Limitations

There are limitations to my research design. Similarities in the experience of bereaved parents may be highly influenced by context, i.e., all the participants were recipients of pediatric palliative care services by a highly skilled team of interdisciplinary health care and allied health professionals. These families also had access to bereavement counseling from Canuck Place Children's Hospice. Differences could be attributed to the differences between the various NMLLC, however, heterogeneity in the conditions the children were affected with is something I sought in my sample. In addition, as previously mentioned, my professional role as a nurse had the potential to influence my ability to effectively conduct research interviews (Thorne, 2016). I acknowledged that this may have influenced the information participants shared with me.

Summary

This qualitative study was scaffolded using ID and was designed to allow a deep exploration of the experience of AG in parents who have a child with a NMLLC. In this chapter, I presented the theoretical premises of ID and provided details of my research methods. I also conveyed my positionality in relation to the research, and discussed ethical considerations and study limitations.

Chapter 5. Results

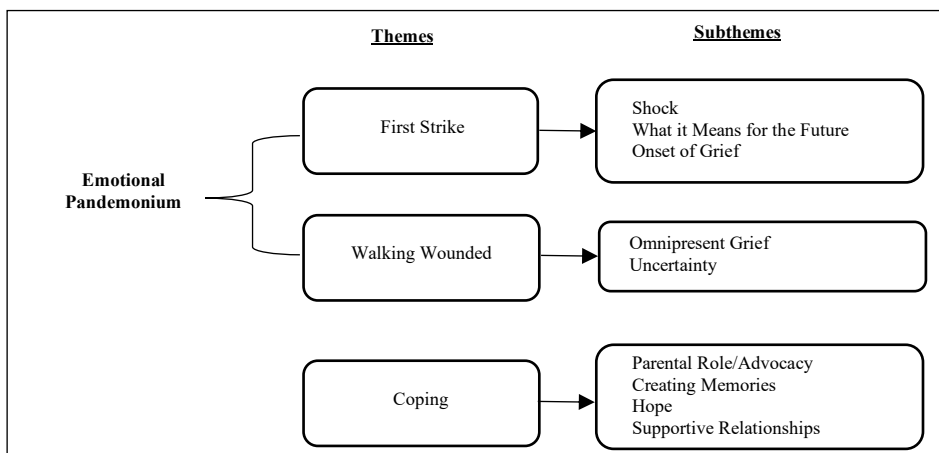
Introduction

This research was conceived to explore the anticipatory grief experience of four parents as they navigated the uncertainties of living with and caring for a child experiencing NMLLC and anticipated to die. This project sought to increase knowledge and understanding of parents' experience of AG through looking for significant moments in their journey, from the onset of their child's symptoms, to death and bereavement. Results from the in-depth analysis of the interviews provided insight into parents' grieving process prior to the death of their child and are supported by what has been previously understood in the existing literature on anticipatory grief in parents (see literature review in Chapter 2). This study also adds further weight on the importance to attend to the social, emotional and physical well-being of parents.

Findings were categorized into three main themes ("First Strike", "Walking Wounded", and "Coping"), the first two together forming the umbrella theme "Emotional Pandemonium". Each main themes is divided in a few subthemes, as illustrated in Figure 3 below. In this chapter, I will present an analysis of the interviews and give examples of participants' narrative to support my interpretation of the data.

Figure 3

Themes and Subthemes



Participants

Four parents (biological n=2, adoptive n=2) participated between August 2021 and December 2021. As mentioned in the previous chapter, participants were given a participant number (P1, P2, P3, P4) and will be referred as such in this chapter in order to protect their identity and maintain confidentiality. The sample characteristics are shown in table 1. The range of time between the death of the child and the interview varied between 18 months to 5 years.

Table 1

Study Sample Characteristics

Participant	Gender	Age range (years)	Relation to child	Child’s diagnosis	Age of child at onset of sympto	Age of child at death	Location of death
P1	F	30-39	mother	Severe cardiomyopathy	1 day	7m	hospital
P2	F	40-49	mother	Mitochondrial EPC	7 months	1y, 2m, 26d	hospice
P3	M	>60	adoptive father	Spastic Cerebral Palsy	birth	12y, 4m, 14d	hospice
P4	F	>60	adoptive mother	Spastic Cerebral Palsy	birth	12y, 4m, 14d	hospice

Emotional Pandemonium

Pandemonium is a “situation in which there is a lot of noise and confusion because people are excited, angry, or frightened” (Cambridge University Press, n.d.). In this study, research participants reflected on their grief experience and grieving process as being full of unknowns, often receiving a lot of information from different sources, especially at the time of initial diagnosis. Initially, they found themselves in a state of emotional chaos, of loss of control, and experiencing a lot of confusion and uncertainty about what is and what will be. For

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participants, this initial state of shock and emotional chaos eventually gave space to the ebbs and flows of grief that was then compounded by ongoing losses. I refer to the breadth and depth of emotions experienced by participants throughout their grief journey as “emotional pandemonium”. Under the umbrella concept of emotional pandemonium are the two first themes: theme 1) First Strike and theme 2) Walking Wounded.

Theme 1: First Strike

It is not surprising that the onset of symptoms and/or initial diagnosis was a very significant moment in participants’ lives. The “first strike” experienced by research participants reflects a period of time during which their life as they knew it would be turned upside down. From the moment they receive the news of the diagnosis, or from the onset of first symptoms, their lives and the life of their child would never be the same. The initial shock triggers a period of questioning regarding what this new diagnosis meant for the future and how it would affect their life, their child’s life, the lives of siblings and the lives of others around them. It also represents the onset of grief, a grief that will be ongoing throughout the child’s life and after their death. This theme contains the following subthemes: shock, what it means for the future, and onset of grief.

Shock

Being given the bad news instilled an emotional chaos in parents as they experienced the shock precipitated by new information. P2 states that this shock was combined with a lack of understanding the details of the situation: “But shock and simply not understanding certainly the finite medical and genetic knowledge, that's not my field.” P2 used an analogy to describe how she felt when she first found out about her child’s diagnosis:

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Certainly on that day when the words come out of the doctor's mouth (...) that your child is going to live a short life, if I can compare it to a visual image, is the sound that we hear when we're in the water. There's a muffled sound. There's nothing around (...) you feel the pressure (...) and you feel sound rather than hearing it, (...) sort of a dissociation from space and time in reality.

Similarly, P3 describes the news of the diagnosis and remembers feeling like being punched really hard:

Physically struck (...) that's the way I felt. I felt like somebody had punched me. I was like stunned, absolutely stunned by the news and the prognosis.

Later in the interview, when talking about his child's diagnosis, P3 re-emphasized that finding out the diagnosis was "like a real solid punch." The planned life of participants would be altered in ways they could not yet comprehend. P1 and P3's analogies were an efficient way to capture the complex set of emotions experienced when finding out that their child had a life-limiting condition. The analogies show that there was a period of initial shock and loss of senses. The units of meaning (e.g. under water, solid punch) depict a picture that provides a small window into parents' lived experiences.

Interestingly, despite being initially in shock, three of the four participants cohesively recounted the journey from pregnancy to the delivery of the diagnosis with a high level of detail. For example, P1 gave a very detailed and emotional recount of how time unfolded in the diagnostic process and its incremental impact:

Before Birth – So I had a really healthy pregnancy and everything was fine until the very last visit to my obstetrician, she did recognize a little bit of a heart murmur, but she said that everything was totally, totally fine.

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Birth – When she came out she wasn't able to feed, she couldn't latch (...) So they said, “Oh we're just going to do some preliminary tests and we'll take her to the ICU.” But once again everything seemed OK (...) nobody seemed too concerned.

The first few days of life – The next morning, the pediatrician said (...) I think your daughter has a syndrome called Noonan. Her heart condition can be quite severe. We're going to send her to Children's Hospital. And then she said, you know, don't look up Noonan (. ...) When I got to Children's, the pediatrician on call had actually fought with the cardiologist (. ...) she was saying, “I need somebody to read these results.” (. ...). And she kept trying to advocate (. ...) When I got airlifted with her [participant’s child] and we were in the NICU at the Children's Hospital (...) the [doctor] made fairly cutting remark. He was getting the low down to all the to all the students that were there. And he said “she displays with a moderate heart defect heart or moderate thickening of the heart (...) And then he turned and he said (...) “I'm surprised you didn't wait until Monday {the be airlifted to Children’s Hospital] (. ...) We spent 10 days, they had the geneticists come out into the room (...) they said that they didn't actually think she had Noonan, but that they would take the test and the whole sequencing genome, that would take six weeks . But from everything that they could see, she was a handsome little girl.

As this participant recounted the story of her daughter’s birth, tears in her eyes, it was clear that the events surrounding the delivery of the diagnosis have had a long-lasting impact on her. For all the study participants, despite the confusion and initial shock of finding out about their child’s diagnosis, the details of the delivery of the diagnosis and the moments that followed have been etched deeply in participants’ memories. It highlights the significance of empathetic communication from HCPs when having these difficult and sensitive conversations with parents.

What It Means For The Future

Having a child who has life-limiting diagnosis drastically alters parents' view of their whole family's future, as illustrated by a quote from P2: "And then (...) slowly, slowly, just realizing what that could mean. So, in the hours that followed, the days that followed, what did it mean for me? What did it mean for him, for his brother, for his father?"

For this mother, the diagnosis was the first step of an unknown journey; what the diagnosis meant as they had to keep moving forward was unknown. All the participants also recognized that the illness of their child would impact their entire social circle. P2 wondered how she would share the news with her community:

And an understanding as well that we had to inform people around us, grandparents , cousins, (...) daycare, right? (. ...) as you look at the circles around a family or a person, the community gets involved in how you would have to live through that or share that experience or that knowledge with everyone. That was the varying degrees of how impactful that was.

Participants had the feeling of being adrift. NMLLC are often rare, therefore illness trajectories are highly unpredictable. Parents were faced with the challenging task of navigating an uncharted territory. P3 reflected on the difficulties of not knowing what to do next:

How do we navigate this? How do we negotiate the future? Be it three days, three weeks, three months, three years? (. ...) And at that point, there were no answers (. ...) we were kind of set adrift.

Onset of Grief

For all participants, the diagnosis or first symptoms triggered an overwhelming wave of painful emotions that marked the beginning of what would be a long grieving process, as

illustrated by P1 who mentioned that her grief began when she was told that her 2-day old daughter might have a syndrome.

And I think (...) on the second day of her life, having the pediatrician say “I think she has Noonan” (...) that very much started the grief process because it had been a healthy pregnancy. I think it was when it looked like she might have a syndrome. It was grieving the life that I had imagined for my little girl.

Similarly, P4 recalls that her grief started at the moment they found out something was wrong with their child: “I think right from the start we grieved because we knew that we had lost a grandchild that we should have had.” Despite their child still being alive, P4 spoke of having already lost her child the moment they received news of a poor prognosis. Her narrative suggests that she was grieving both the psychological and the physical loss of her child, even though he was still physically present. The anticipated losses and the anticipated psychological and physical limitations that this child would experience were being grieved from the very beginning of the illness trajectory.

Theme 2: Walking Wounded

The complexity of the child’s diagnosis was an emotionally challenging period filled with uncertainties that go beyond their concerns for their child, and resulted in parents swimming through the emotional pandemonium that was described earlier in this chapter. Parents were “walking wounded”. Throughout their child’s illness trajectory, parents’ emotional, psychological and physical well-being were impacted. This theme is divided into the following subthemes: omnipresent grief, and uncertainty.

Omnipresent Grief

As mentioned in the previous theme, when parents receive the news that their child has a NMLLC, they are in shock and it is at that moment their grief begins. Some of the feelings associated with the initial shock appear to eventually subside to give space to a new normal, however, it was clear that for all participants, grief was omnipresent throughout their child's life. Their imagined hopes, life and dreams for their child were clouded by grief. While grief may have changed in nature and intensity throughout the child's illness trajectory, it never fully left the participants. This was often attributed to the ongoing losses that had a compounding effect on grief, as illustrated by this poignant quote from P2:

It's a little grief every time something happens. Which amounts to just grief building on itself, letting go when you realize, OK, my child is going to pass away before I do. And then (...) he can't see anymore. He can't sit anymore. He can't eat anymore. (. ...) Those are all losses that accumulate upon themselves. And so when the very the real end comes when his life ends, it feels like he has been dying this whole time. So I have been grieving this whole time.

Interestingly, in reflecting on her grief experience, P2 also questioned the validity of the term anticipatory grief:

The grief was constant (. ...) I thought it was interesting that your study is called anticipatory grief because the moment it happens is no longer anticipatory (. ...) And what I mean by that is that, it's not his passing that causes grief, it's everything. It's every loss leading up to his passing. And then it's another version of grief.

This mother's experience represents grief as a continuum, that changes in nature and intensity, happens in the past, present and future and holds an ubiquitous presence. This is also

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reflected in other participants' narratives. P1's experience illustrates this continuum of grief, where she started to grieve the life imagined for her daughter when she received the news of a dim prognosis, followed by grieving the life that she pictured having with her daughter, and then after death, grieving her daughter and everything that she did not have the opportunity to experience or become:

The grief of the life that I imagined for my child started to occur, and then after all of that already passed, you know, two or three months in and it was just ok. So what I thought would be my life isn't. And now it was grieving the life that I thought, (...) and then after she passed, grieving who she was and what we had, the life that she could have had.

After the onset of grief, omnipresent grief became the new normal for these parents, a new normal they had to try to adjust to:

P3: And it's not like every day was a party. But, you know, we celebrated birthday parties (...) Once he started going to school, we did all those activities (...) not with the idea that, oh my gosh, we should squeeze all this stuff in before he dies. It was that he's a kid and he needs to do this stuff.

All the participants tried to maintain some sense of normalcy, for example, by having their child participate in activities that healthy children participate in. They also tried to maintain a "normal" family life for the siblings:

P2: So having to continue living through our grief (...) we had to normalize that part of it (...) How did I do it? I just lived it very openly. There was no pretending it didn't exist and everything I did for [child] or for [sibling] or myself or my husband, it [child's illness] would always just be taken into consideration. We simply worked with it. There was no pretending that it wasn't there.

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Participants described different reactions to grief being their new normal. Two participants mentioned not letting grief “get in the way”, and that this was how they could keep going day after day:

P4: I think in small ways we were grieving throughout his whole journey through life, we did grieve in moments, but we didn't allow it to take over totally, because we couldn't have gone on, I don't think, if we had .

Similarly, P3 recalls the omnipresence of grief and how they did not let it rule every aspect of their daily life: “It wasn't like we spent all our time thinking, oh my gosh, he's going to pass away (. ...) I didn't spend a lot of time dwelling on it. It was it was just there, and I knew it was there.”

In contrast, the other two participants described more constant outward physical/emotional manifestations of their grief. P2 stated that she lived her grief openly, mentioning that she would cry regularly: “I'm not one to push down emotions (...) I fully embrace a situation as difficult as it might be (. ...) I fully embrace my sadness, totally (. ...) I would cry regularly, not all the time, rather regularly.”

P1 described feeling that her body was constantly in a “fight mode”, as she was “either fighting with the nurses or it was fighting with trying to control the environment to keep [child] safe.”

A prevalent message across all participants when describing their grief experience is that throughout their child's trajectory, they continuously grieved the “life imagined” for their child. They described how they grieved the milestones they would never see their child reach. P4 recalled that what she grieved for her child were “just the little things, everyday things that he should have been able to do like run and climb.” Participants indeed grieved expected childhood

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activities like playing sports, but most also mentioned bigger life milestones such as graduation, marriage, etc., like P2 who acknowledged the loss of not seeing her son growing up to live a full life:

I think my son's name is beautiful (. ...) But I never thought that I that I would see it on a grave as opposed to a diploma (. ...) I would not see him ride a bicycle (. ...) I would not see him graduate. I would not see him live his life (. ...) all those milestones.

Participants grieved every aspects of their child's life that they could have otherwise accomplished if they were not given their life-limiting diagnosis. The emotions that transcended the parents' stories suggest deeply rooted grief.

Participants' narratives described losses across all aspects of their lives and not only directly related to their child. For example, P4 expressed the loss of social connections, and of careers:

You do get isolated and you lose your friends (. ...) we both gave up our careers (. ...) So we grieved that too (...) but we were prepared to give it up for [child] because, well, what do you do? He deserved it, right? We needed to.

P2 mentioned the grief associated with losing what should have been good memories of a vacation meant to celebrate their older son's recovery from an injury. Unfortunately, it is during this trip that her younger son displayed his first seizure:

That was my first trip to Hawaii, my husband's first trip to Hawaii, [child], of course , first trip to Hawaii and [sibling]'s first trip. We will never look at Hawaii the same way. So even the grief of "that was supposed to be just a nice vacation."

Uncertainty

In addition to adjusting to living and caring for a child who has a NMLLC, the unclear prognosis, unclear trajectory, and the complexity of navigating the medical system introduced a lot of uncertainties in the parents' lives. Uncertainty was a prevalent theme throughout all of the participants' narratives, and this uncertainty permeated various aspects of their life. Often, this uncertainty was related to the diagnosis and prognosis, and anticipating the next health crises. For example, two of the four participants felt compelled to have their cell phone on at all time in case they would receive an emergency call. P1, whose daughter needed a heart transplant, had to be reachable at all time: "We couldn't be without our phone (. ...) I couldn't go to a yoga class without leaving my phone with my husband. And then I would drive home and I would think to myself "would they call"?"

There was also uncertainty about symptoms and what disease progression would look like. P2 recalled being told the changes she should expect to see, but feeling unprepared to see what those changes actually looked like:

There could be (...) all sorts of things that could happen to him. He would eat less and drink less (. ...) I was told by his neurologist that his vision would likely go (. ...) I understand what that means (. ...) But when I saw him, and I realized that he could not see me, there's nothing that prepares you for that. I can't imagine that there is something that prepares anyone for that (. ...) It's every step of the way (...) I knew it was going to happen, but I didn't know what it would look like."

The uncertainty of when death would occur also permeated participants' lives. P4 said that her husband and her were always fearful that their son was going to die:

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It was always a fear that he was going to die. And that that could happen almost any time because [child] had so many close calls throughout his life. So that was our fear (...) the uncertainty of it all.

Theme 3: Coping

Trying to cope with grief and continue living life required parents to balance their needs and the care of their ill child with other family demands. Coping helped parents navigate the emotional pandemonium described through themes 1 and 2. They identified the following factors (subthemes) as necessary for them to maintain some stability and cope with the uncertainties: parental role/advocacy, creating memories, hope, and supportive relationships.

Parental Role/Advocacy

One consistent message across participants was the need for their role as parent to be valued and respected. They wanted to feel connected to their child, emotionally and physically, and they wanted to be given the space to be a parent. Parents also wanted to be acknowledged as the parents of their child, wanted others to recognize that they knew their child best, and wanted to be involved in decision making.

All the participants reflected on moments where they felt connected to their child, whether it was through exchanged smiles, or through a finger being held. P2 recalled that so many of her son's care was provided by HCPs. Holding her son, or even changing diapers was important to her, because it made her feel that she could still do something for her son and be a good mom; she said that it was her way of showing her love for her son, of feeling connected:

It made me realize how the mundane things were important, because then the ability to hold him in my arms could be the most I would get from him. And that it'd be the most that he would get out of me.

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It was evident that participants often had to process complex information and use that information to make difficult decisions. While parents agreed that HCPs had the medical knowledge, parents felt they knew their child best and wanted to be involved in decision-making. The following example recounts the experience that P1 had as her child was having a cardiac event that ultimately led to her death. According to the participant, the team thought it would be preferable to forgo shocks as a resuscitative measure, but the participant and her husband were willing to try everything and to this day, they still think to this was the best decision for them:

When it did happen, they asked whether we want them to try the CPR shocks. And we said yes. And they said, “are you sure you're prepared for all of that? Really?” They were quick trying to discourage it. And it's like, well no, she's going to die this other way, if there's two options and one is definitely death, and the other is probably death, but it'd be a bit harder, let's go for that one.

Another participant (P2) highlighted the impact of being involved in decision-making: “There is so much I couldn't do, but as much as I could understand, “what if we try this medication and what if we try this procedure?”(. ...) The knowledge empowered me.” As one could expect, not being recognized as an important voice in their child care was, as P4 mentioned, “devastating”:

One intensive care nurse who had been off for years and had come back to work told us, “oh no, I think he's just having a tantrum” (. ...) he was vocalizing and arching and we were saying, “No, he's in pain, and this is why he's doing this. We know how to read him.

Creating Memories

Participants often recalled doing “normal” things, like going to the aquarium, going to a park, or even simply going outside, and it is clear that they cherish those memories. Being able to

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do “normal” things and having fun with their child created long lasting memories and seemed to help them cope with grief on a day-to-day basis.

P2: If there was a milestone that he could reach, he did turn one, (...) I wanted to take him to Stanley Park, for example, so I took him to Stanley Park. It's not much, right ? We did things, I guess, with more purpose (. ...) So although I didn't get to do a million things with him, those little things (...) became very important, very meaningful.

Similarly, another participant (P3) emphasized the importance of making fun memories:

You know, as long as he was going to be there, it was going to be a party as much as I could make it (. ...) We just didn't think about it. It was always there, obviously. We knew that if it was going to be a short time, then it ought to be a good time.

This participant also mentioned that their son was involved in various school activities, such as popcorn sales, basketball games, and sport days. The pride in the father’s voice as he recounted these experience bears witness to the impact of seeing his son participate in activities with other children.

Hope

Living with uncertainty in the face of a NMLLC is challenging, and sustaining hope was a mean of coping for participants. This hope was not necessarily hope for a cure; sometimes it was hope for more time, or hope was aimed at finding comfort in a good day where their child is not experiencing a lot of symptoms. Hope could be cultivated and focused on their child reaching a milestones, and acknowledging that despite challenging circumstances, joy could still be found.

For example, P2 said:

I would always hope that it would last as long as possible. That was my hope, really , and that's the limit of that hope. If you know there's an end to something, well make it last,

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make it last today, make it last as much as possible, make it worthwhile. And that's where my hope rested.

P4 reflected on whether hope was actually helpful to them or not, and she came to the conclusion that it would have been difficult to cope without hope:

He would fool us, you know, because he would get back up and you tended to get optimistic again and think: "Oh, he's going to be good. Everything's going to be fine."

And that in itself was not necessarily good for us, (...) but then again, maybe it was good because I don't know if we could have gone on if we didn't have hope, right?

However, for one participant, hope added to the difficulties of navigating life with a medically complex child. P1 was hoping that her child would survive to be old enough to get a heart transplant. She referred to her hope as a "sickening place", because in her gut, she knew chances were very slim that her daughter would get a heart in time. She compared this hope to the concept of "doublethink":

It's when your body holds two things and one, you know isn't true, but you're believing it.

And then you're being told to believe it too. The other one is kind of that pit in your stomach, instinctual. And that was the really sickening part.

Supportive Relationships

Participants identified that external support, such as support from HCPs, helped parents. This support fostered hope and helped mitigate the impact of grief on parents' day-to-day life.

Parents were asked if there were things HCPs had done well and things they could have done better to support them in their grief. Similar to the subtheme of parental role/advocacy, parents appreciated when HCPs gave them the opportunity to be involved as much as possible in

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their child's care. They felt supported when they could communicate with HCPs without fear of being judged and/or dismissed and when information was empathically conveyed to them.

Parents wanted honest information that was not "sugarcoated". Honesty made it easier to anticipate what was ahead, and to feel more prepared. P2 emphasized that having the time and space to sit with the information helped her make an informed decision about her child:

Being informed of what was happening, given information, given the time and space to express myself, being taken seriously. And there was never any fog, so I knew where to go, what I wanted to do, what I thought we should do for [child]. So, I give a lot of importance to that, the space, the time that we were given.

Another participant remembered the importance of good relationships with HCPs and how suboptimal communication would at times lead to conflicts:

P3: Convincing medical experts that we knew stuff (...) and (...) that we did know something about his condition and that we probably knew a lot more about how to care for him than perhaps they did. They had the science, but we had the practice (...) so it ended up that sometimes we came in conflict.

Empathy and humanness promoted supportive connections between HCPs and parents.

P1 described a very impactful interaction in the final hours of her child's life where a doctor had asked her if she could step out of her daughter's room to get an update on her condition:

[Child] was holding my finger and looking into my eyes. And I said "you can tell me right here , I don't really want to leave her at this moment." She was quite good (...) they were doing everything to make sure she [her child] was able to see me, and I really appreciated that.

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In one participant's narrative (P3), a doctor demonstrated supportive communication by the simple act of taking off their mask to have a difficult conversation. Listening to this parent recounting this specific moment, it was clear that the act of taking off their mask conveyed the doctor's empathy, humanness and respect.

The majority of the participants emphasized the positive impact of receiving services from a specialized pediatric palliative care team and having access to in-house hospice care. Participants mentioned the caring nature of pediatric palliative care nurses, as well as their expertise in caring for their children. Having a place where they felt supported as individuals and as a family was very important:

P3: I can tell you that when we came to [children's hospice], when we came up the driveway, we just relaxed. We just knew we were in a safe place where you didn't have to explain things to people (...) I think having people who knew what we knew and who understood as we understood without always having to explain stuff all the time is such a relief.

Interestingly, most participants did not emphasize friends and family as significant support. One participant did mention that the home caregivers for their son became helpful friends, but most participants highlighted the lack of time and/or energy to connect with others outside their family units:

P2: A few friends were very close and very present. Most were completely out of our lives. And certainly, it's not the time to make new friends, right? I honestly did not care. Such a diagnosis, such an experience, crystallises values. And certainly, everything else is dismissed immediately. And I was comfortable with that.

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One participant (P1) also remembered the hospital organizing coffee nights for parents whose child was hospitalized. However, she differentiated between parents whose child was in a very stable condition and who might feel comfortable stepping away from their child for some time, to parents, like herself, whose child had a life-limiting condition and who might die suddenly from disease complications. She said that “somehow having a community of mothers [in a similar situation as hers], it would be helpful.”

This participant also mentioned the existence of Facebook groups, but that all her attention was directed at her daughter, so she felt like she did not have time to really engage in those groups.

Summary of Findings

A thorough analysis of the interview transcripts revealed three main themes: theme 1) First Strike, theme 2) Walking Wounded, and theme 3) Coping. The first two themes fell under “Emotional Pandemonium”, representing a state of turmoil experienced by parents as they learned that their child would live a short life. Parents described their experience of living with a grief that started as soon as their child showed their first symptoms, and a grief that was omnipresent and permeated every aspect of their lives. Parents’ narratives indicated many factors that helped them cope with their grief on a day-to-day basis, emphasizing the importance of being valued as parents and included in their child’s care team, and the value in having opportunities to make long-lasting memories with their child. The theme of coping was discussed as being separate from the themes that fell under emotional pandemonium (theme 1: First Strike, and theme 2: Walking Wounded). However, it is important to note that theme 1 (First Strike), theme 2 (Walking Wounded) and theme 3 (Coping) are not mutually exclusive. Together they

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provide an holistic view of the experience of AG in the participants. The next chapter will present a thoughtful discussion of the results and of their relevance for nursing practice.

Chapter 6. Discussion

Introduction

This research explored anticipatory grief (AG) in parents who have a child who died from a NMLLC. The richness of the data I obtained through interviewing parents provided rich insights into their experience of AG while caring for their child. The first two themes, “First Strike” and “Walking Wounded”, describe the experience of AG, while the last theme (coping) describes factors that had an impact on the experience of AG. AG was experienced by all participants, however, it is the nature of the child’s illness and the features unique to each family and each participant that created similar and unique experiences of AG. This discussion will focus on three main points. First, I will compare and contrast my study results to the literature review on AG presented in Chapter 2 and to other current literature. Second, I will integrate my findings more explicitly with my theoretical scaffolding to show the connections between AG and other concepts. Finally, I discuss implications for practice, research and education, as well as strengths and limitations of this study.

Uncertainty and Ongoing Losses

The participants in this study vividly recounted the complexities of their AG experience by sharing detailed stories of their pregnancy, birth, encounters with health care providers, life with their child, and the death of their child. These rich conversations were emotionally charged and highlighted the importance of addressing their unique parental journey and the significance of giving parents space to be heard.

Throughout the interviews, it was evident that the news of a NMLLC diagnosis turned the participants’ lives upside down, and that the initial shock started the grieving process. Parents did not know how much time their child had, but they knew that time together would be

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short. They grieved the past and what was before the diagnosis; they grieved the present and their current situation, and they grieved for the future and the life they had imagined for their child. Similarly, Rando (2000) outlines time focus as one of the six dimensions of AG and defines anticipatory mourning a response to all the losses (past, present and future) encountered in the course of an illness.. In fact, the participants were quite cognizant of their past, present and future losses. The interviews revealed that there is continuity to grief, and that after the death of their child, parents continued to grieve everything that the child did not have the opportunity to see, participate in, and become.

Participants experienced ongoing losses which also meant that grief was omnipresent. The losses occurred across various aspects of participants' lives, for example, loss of social connections, loss of income, and loss of imagined family life. Their whole life and their grief were interwoven. The successive losses participants were subjected to caused "another layer of grief that adds on to another layer"(P2). This confirms results from previous studies where parents of children with neuromuscular conditions continuously anticipated future losses as their child's health declined (Lucca & Petean, 2016; Yang et al., 2016). As also found in other studies (Pelentsov et al., 2016; Smith et al., 2015), this study showed that in addition to facing ongoing losses, participants' lives were also filled with uncertainties. There was no way to know when the next medical crisis would be, or if their child would live another day.

A meta-analysis on caregiver and youth uncertainty in pediatric chronic illness showed that caregiver's uncertainty about their child's illness increased caregiver anxiety and depression (Szulczewski et al., 2017). Similarly, in the qualitative studies reviewed prior to conducting this study, participants expressed feeling sad and depressed (Batchelor & Duke, 2019; Dighe et al., 2008; Yang et al., 2016). While this study did not investigate the psychological functioning of

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parents, the interview data certainly suggests the presence of some anxiety and occasional distress in participants as they navigated the uncertain futures for their child, themselves and their family. While participants universally experienced omnipresent grief, the way they lived their grief on a daily basis differed (e.g., body in “fight mode” the whole time, versus not letting grief take over their everyday life). The NMLLC that affected P1 and P2 versus P3 and P4 were quite different. P1 and P2’s children had a shorter life expectancy (P1’s child prognosis was dependent on a heart transplant), so this could potentially explain the difference in the way parents experienced AG in their day-to-day life.

Coping

When a child is diagnosed with a NMLLC, family functioning and ability to manage daily life activities are substantially impacted. The ability to cope via various mechanisms can help mitigate grief and the impact that a life-limiting diagnosis impose on parents.

Advocacy and the Importance of Good Communication. Advocacy is a significant aspect of caregiving for parents. Other scholars have noted that (a) advocacy allows parents to feel more in control despite ongoing uncertainty and stressors (Carter, 2014); (b) it supports positive coping and health outcomes for parents and children (Ellenwood & Jenkins, 2007); and (c) it helps parents to construct of a “new normal” (Hess et al., 2006). In this study, participants wanted be given the space to fulfill their role as parents and advocate, and be recognized for their expertise in caring for their child. Their involvement in decision-making for their child was extremely important and they felt empowered when their opinion was valued. Similar results were found in other studies (Davies et al., 2017; Miller & Nelson, 2012; Rafferty & Sullivan, 2017). However, while participants were aware of the importance of their role in their child’s care, they also acknowledged their limitations as parents. As one parent (P3) mentioned “ they

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[HCPs] had the science, we had the practice.” Honest and empathetic communication with health care providers was key in supporting parents in their grief journey and it helped them feel comfortable advocating for their child and navigate life according to the limitations imposed by their child’s diagnosis. It played a vital role in aiding parents’ interactions with HCPs.

Similarly, literature suggests that consideration for parent’s needs was seen as having a positive impact on parents’ experience at the end-of-life of the child, while the opposite showed to negatively affect the AG process (Rini & Loriz, 2007). Findings from my study as well as other scholars have shown that receiving clear, sufficient and timely information from empathetic health care providers throughout the disease trajectory including at the time of death supported parents to collaborate with HCPs in making informed and shared decisions (Lucca & Petean, 2016; Rini & Loriz, 2007). Communication that is effective and compassionate can foster trusting relationships, provide anticipatory guidance in the face of uncertainty, provide emotional support, and support informed decision-making (Hauer & Wolfe, 2014; Koch & Jones, 2018).

Impact of Receiving Pediatric Palliative Care Services. Navigating the complexities of health-care systems is a challenge for all families, especially those caring for a child with a LLC (Charlton et al., 2017; Son et al., 2018; Whiting, 2014). A specialized health care team who supports families through their complex journey is extremely important. For example, a lack of awareness and knowledge about their child’s condition by some HCPs adds to the parents’ uncertainty during the early stages of the disease (Pearce et al., 2021). In this study, participants appreciated the expertise of a pediatric palliative care team, which typically have more expertise in caring for children with rare LLC than HCPs with less experience caring for children who are medically complex. Pediatric palliative care teams care for the whole family and provide support with decision-making, communication, psychosocial support, pain and symptom

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management, end-of-life care and bereavement support (Widger et al., 2016). Pediatric palliative care teams are also specifically trained in having difficult conversations, which is so important for parents caring for a child with a NMLLC who will receive an incredible amount of information and difficult news. Further, parents are asked to make decisions, sometimes time-sensitive, about their child's life based on the information they receive from their HCPs (Koch & Jones, 2018). In this study, families who received pediatric palliative care services from a specialized pediatric palliative care team were provided with the information they needed to adjust to their new and changing normal and to make decisions based on the comprehensive and supportive information they received. Previous studies concur that with the involvement of a pediatric palliative care team, symptom control is improved for the child and families feel more supported in making decisions and are more satisfied with their child's care (Mack & Wolfe, 2006; Marcus et al., 2020; Osenga et al., 2016).

Support Network. In contrast to previous studies on AG (Batchelor & Duke, 2019; Dighe et al., 2008; Higgs et al., 2016), parents in this study did not highlight support from friends and family as a significant help in coping with their grief. While this does not mean that friends and family were not important to them, their influence on well-being was not emphasized. Most parents indicated that they had very little time to engage with friends due to the demands of caring for their child and their desire to spend as much time as possible with their child. Based on this study's results, it appears that parents could still benefit from connecting with parents in similar situations, as long as these opportunities to connect are convenient. For example, there is an overwhelming number of specific diseases group on social media where parents can turn for support, and parents can decide to engage with these groups as much or as little as they wish, and at any time. In addition, as one study participant mentioned, when their child was admitted at the

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children's hospice, they would meet other families who shared similar experiences. A mutual understanding of each other's reality can make parents feel less alone in their journey and give them a sense of belonging (Dunbar & Carter, 2021). It is evident that children hospices are a great opportunity to facilitate supportive connections between families.

Creating Memories. Parents often recalled that everyday activities, such as going to the park and participating in school activities, were the most meaningful and brought them joy. The International Children's Palliative Care Network, a global organisation for children's pediatric palliative care that aims to improve care and quality of life for children with LLC and their families, published a charter of rights for children with LLC (International Children's Palliative Care Network, 2022). The Charter states that, wherever possible, a child with a LLC should be provided with opportunities to play, access leisure opportunities, interact with siblings and friends and participate in normal childhood activities. Those activities are not only beneficial for children with LLC, but also have a positive impact on parents. Creating memories provide healing for the family throughout the illness trajectory (van Breemen, 2009). Parents in this study described that making memories of their medically complex child interacting with other children or with their siblings helped them cope with their AG. The current literature on AG does not comment on the benefits of making family memories or participating in recreational activities together. This highlights the importance of further research.

Hope, Religion and Spirituality. Hope is a "deliberate, life-sustaining, dynamic, cyclical process" of uttermost importance for parents who care for a child who is medically complex (Bally et al., 2014, p. 363). In this study, participants spoke of hope in the context of having a good day, one more day, or one more year. A study on parents who have a child with cancer revealed that hope in can alleviate parents' psychological suffering and help them adapt

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to the reality of having a child with cancer (Eche et al., 2022). Similarly, the well-being of parents of children who have a LLC was found to be positively impacted by hope (Bally et al., 2018). In my study, hope for more time was often one of the factors that helped parents navigate their new normal and mitigate their feelings of grief.

Hope and religion have often been studied together, and scholars found that religious and spiritual beliefs positively affect levels of hope (Afrooz et al., 2014; Bally et al., 2014; Shamsalinia et al., 2016). For parents caring for a child with a LLC, spirituality can be a source of hope, strength and coping (Batchelor & Duke, 2019; Benefield et al., 1976; Lucca & Petean, 2016; Yang et al., 2016). Furthermore, religion and spirituality help parents cope with uncertainties, helping them transform their uncertainties and grief into strengths (Yang et al., 2016). In this study, when asked what helped them cope with AG, parents did not mention religion or spirituality as a significant factor. This result must be interpreted with caution due to the size and characteristics of the sample.

Anticipatory Grief and Attachment Theory

In chapter 3, I mentioned that stress and burden of care experienced by parents who have a child with a NMLLC could potentially jeopardize attachment bonds. While study questions were not specifically designed to examine this, the way with which participants recalled their experience indicates that the participants' bond with their child might actually be stronger despite the burden of care. Parents' experiences evoked memories of moments where emotional and physical connection to their child was of uttermost importance. Recalling these moments crystalizes the strong nature of the bond between parents and their child. Findings from studies on caregiver of medically complex child and well-being varies. While multiple studies emphasized the negative effects of caregiving (Toledano-Toledano et al., 2020; Yang et al.,

2022), a study by Deci et al. (2006) on attachment theory showed that caregiving may promote wellbeing in caregivers. My study results concur with the findings from Deci et al. (2006); despite the challenges of caring for a their medically complex child, parents felt they had purpose and felt that they could be a good parent when they were given the space to be involved in their child care and decision-making. None of the parents mentioned burden of care.

Anticipatory Grief and Ambiguous Loss

Ambiguous loss encompasses the grief that is experienced by individuals when “a person is both physically present and psychologically unavailable, or psychologically present, yet physically unavailable” (Boss, 1999, p.11). It is a loss that is incomplete, unclear and often uncertain (Boss, 1999, 2007). Studies on AG in parents often describe ongoing losses that are incomplete, for example, the child is still there, but parents witness physical or psychological losses in their child. Interestingly, despite describing losses that are ambiguous, the literature on AG in parents who have a child with a NMLLC rarely identify those losses as such. Parents in this study experienced ambiguous losses as they coped with the uncertainties and ambiguities, and with the loss of their “imagined” child. Most parents were flexible and chose to accept the uncertainties and ambiguities, as they knew they could not be fixed. This aligns with Boss’s (2006) recommendation of remaining flexible rather than focusing on gaining full control or surrendering completely as this acceptance and tolerance for ambiguity can strengthen resiliency. While one parent mentioned the negative impact of living with uncertainties on her mental health, overall, parents cope with ambiguous loss through embracing their child and defining their role as their child’s advocate, allowing them to better manage their day-to-day life despite experiencing AG.

Anticipatory Grief and Chronic Sorrow

Chronic sorrow is described as periodic recurrence of continuing, profound, pervasive and recurring feelings of grief associated with non-finite losses (Roos, 2017). The loss creates a gap between what a person expected from a situation and what actually ensued as a result of the loss. Parents experience chronic sorrow when there is a gap between the imagined child and the reality of a child with a chronic condition (Bettle & Latimer, 2009; Eakes et al., 1998; Waite-Jones & Madill, 2008). Parents in this study experienced chronic sorrow regardless of the diagnosis and life expectancy of the child, leading me to think that it is the chronic nature of the NMLLC that triggers chronic sorrow.

Study participants talked about experiencing loss after loss and this had a compounding effect on grief. The cycle of chronic sorrows (e.g. sadness, feelings of loss, disappointment, and fear) is triggered again by every loss along the disease trajectory (Eakes et al., 1998). In the middle range theory of chronic sorrow, management methods are described as personal coping strategies (internal) and to interventions provided by healthcare professionals (external) (Eakes et al., 1998). Parents in this study described both internal and external coping strategies that increased comfort and helped them adapt continuously to new normals. AG, in the context of a chronic, life-limiting illness, is complex and resolution is often elusive because of the constant reminders of what has already been lost, and of what will be lost in the future.

While chronic sorrow occurs in the context of a loss that has happened, it is possible, based on this study's findings, that anticipatory losses also trigger the cycle of chronic sorrow. The way participants described their experience of grief (e.g. the "layered" ongoing grief and ongoing losses) highlights the blurred conceptual boundaries between chronic sorrow and AG in

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parents who have a child with a LLC, thereby emphasizing the value of looking at AG through the lens of chronic sorrow.

Implications for Practice, Research, and Education

Implications for Practice

Children who have a NMLLC are often connected to multiple health care professionals, and at every point of care, health care providers have the opportunity to provide family centered care. Parents in this study really emphasized the impact of communication on their experience. HCPs must listen to parents and recognise their expertise in caring for their child. Building communication skills and learning how to listen with intent is imperative when caring for families who have a child who is medically complex. In addition, while caring holistically for the whole family is one of the main principles of pediatric palliative care, pediatric palliative care principles are not always upheld in settings outside pediatric palliative care settings. All health care professionals have a role in guiding parents through uncertainties, and HCPs should not hesitate or defer connecting with pediatric palliative care teams for guidance on having difficult conversation or for advice regarding interventions to help alleviate parents' grief and promote positive coping mechanisms. In locations with access of pediatric palliative care services, involvement of the pediatric palliative care team early in the disease trajectory would be beneficial, as it allows for improved symptom management, better parental adjustment to their new reality, support in the home, and better preparation for the end-of-life period (Kaye et al., 2016; Vadeboncoeur & McHardy, 2018). In order to facilitate early integration of pediatric palliative care, it is important for HCPs to understand that pediatric palliative care is not synonymous with "giving up" on trying to find curative treatments; palliative and curative are not mutually exclusive. Nurses who identify patients and families who could benefit from

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pediatric palliative care should advocate for their patients/families to have early access to such services.

The child's health care team and/or the parents' HCPs should have tools to assess parents' AG and provide interventions to promote their well-being, keeping in mind that parents may not be able to utilize coping mechanisms that were working for them prior to receiving their child's diagnosis. Nurses typically spend more time with families, so they are well-situated to assess parents, provide support and connect them with the appropriate resources. While nurses are not necessarily trained in grief therapy, some methods, such as narrative approach, can be learned and be integrated in nursing practice. Narrative therapy within the context of grief is a process of reflexive storytelling of loss or traumatic experiences (Nelson et al., 2022).

Storytelling and sharing emotions has long been recognized as playing a critical role in addressing grief (Byng-Hall, 2004; Moules & Amundson, 1997). Hedtke (2014) states that "narratives contain feelings and emotions but are also much more than them. Narratives are ways of organising feelings, thoughts and actions into meaningful units of living." (p.15) Narrative therapy approaches can be used within the context of nurse-patient relationships to explore stories that give meaning to parents' experience (Aloi, 2009). Nurses can intentionally walk alongside parents without necessarily trying to "fix" or assume they know what is their best course of action to cope with their AG. While literature on narrative therapy in the context of grief mainly focuses on grief after death, storytelling can without a doubt perform a cathartic function for parents experiencing AG.

Implications for Research

Most of the research on AG in parents focuses on children with oncological disease, cerebral palsy or neuromuscular conditions. Research focusing on rare genetic conditions with

unique trajectories is needed. In addition, further research could aim at determining if cultural factors influence the experience of AG in parents and the impact on coping.

This study highlights the positive impact on parents of creating memories with their child. Research in this area is scarce, therefore this topic should be further explored. In addition, guidelines for AG assessment and therapeutic interventions that can be applied within the time-constraint of health care providers should be developed and their effectiveness assessed.

Finally, methods such as photovoice or other photographic methods could be considered. Some parents may not be comfortable sharing their experience of grief and other emotionally-charged experiences through words and face-to-face interactions, especially fathers (Macdonald et al., 2010). These methods have been found to empower research participants (Budig et al., 2018). Participants in pediatric palliative care research are mainly females (Macdonald et al., 2010), therefore research methods such as photo-elicitation or photovoice could be used as an attempt to recruit more fathers and close the gender gap in pediatric palliative care research.

Implications for Education

As healthcare and technology continues to evolve, it can be anticipated that children with NMLLC will survive longer (Fraser et al., 2021). HCPs in settings other than pediatric palliative care should be educated on palliative care approaches with a focus on communication and holistic care for the whole family. This education should be included in curricula for all health professionals. As findings from this study suggest, effective, compassionate and honest communication played a critical role in parent's experience, including in their coping with grief. Education on serious illness conversation across health care settings, such as training on how to use the *Serious Illness Conversation Guide-Pediatric* (van Breemen et al., 2020) would be highly valuable.

Contributions

This study contributes to the body of literature on AG in parents who have a child with a NMLLC. In the past, researchers seldom specifically explored AG in parents, and when they did, studies have often focused on parents of children with cancer or cerebral palsy. Furthermore, a number of these studies were quantitative in nature. This study, through rich descriptions obtained via interviews, shines a light on the experience of AG in parents of children with diverse NMLLC and provides a diving board for further exploration of the topic. It calls attention to the role of nursing in identifying AG and in helping parents navigate and cope with the unknowns.

This study showed that AG in parents who have a child with a NMLLC is very complex and multidimensional, and I believe that their experience of AG is not entirely captured in well-known grief models. The operational definition of AG is also vague. While the goal of this study was not to provide an updated conceptual definition of AG, the study participants' narratives allowed for a deeper understanding of the concept, notably its non-linear nature and the ambivalence between experiencing multiple losses, adjusting to new normals and hope. AG starts at the onset of the first symptoms or at the time of diagnosis and continues throughout the disease trajectory. There are, however, periods during which parents adapt to new normal. The mental processes required to adapt to these new normal require further attention. These periods of adaptation are then often followed by waves of more intense grief caused by, for example, a child experiencing new symptoms. While the existing grief models may still reflect some aspects of the experience of AG in parents, they don't fully capture the experience of grief prior to death. This study therefore highlights the importance of looking at AG in parents who have a child with a NMLLC through current grief theories as well as other theories/models, such as ambiguous

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loss, chronic sorrow and attachment. This could help develop a model of AG that provides a more accurate picture of the experience of parents.

This study also generated important knowledge not often discussed in AG literature and that represents areas for interventions that may facilitate coping with AG, such the importance of access to pediatric palliative care, providing recreational opportunities for families, etc. In fact, the heuristic value of developing a more accurate and expansive conceptualization of AG in parents who have a child with a NMLLC lies in the potential for development and implementation of holistic and creative interventions to support parents through their unique journeys.

Study Strengths and Limitations

Strengths

The criteria to establish quality and trustworthiness in qualitative research includes credibility (validity of the conclusions that are drawn from the data), dependability (extent to which similar findings would be obtained if the study were repeated), confirmability (degree of objectivity of the researcher in data collection and reporting) and transferability (how well the study conclusions can be applied to other similar settings) (Lincoln & Guba, 1985). Within this study, these qualitative research indicators were addressed through various measures.

While transferability of this study may be limited due to its small sample size, the characteristics of the study participants and methods were well described, therefore, the study findings may be used to understand the experience of AG in parents who have a child with a NMLLC in different settings. Interviews lasting between 75-90 minutes provided rich data, integrating multiple quotes from participants and holding regular meetings with my supervisors helped establish study credibility while a thoughtful selection and detailed description of study

methods in Chapter 4 supported dependability. Credibility and dependability were further enhanced by keeping an audit-trail of my analytical decisions (appendix E) as well as by writing field notes and keeping a reflexive journal. Lastly, confirmability was addressed through grounding the study in participants' experience. While a limit to confirmability lies on the assumptions that I brought to my research as a pediatric palliative care nurse, practicing reflexivity and considering my positionality from early on in the research process through journaling and writing field notes after each interviews allowed me to express my initial impressions and question my assumptions, therefore supporting confirmability and contextual awareness.

Limitations

Study design

The study included four participants, therefore it might limit transferability of the results. Participant recruitment was an unanticipated challenge. Consequently, I revised the recruitment strategy to include special interest online groups, however, I did not receive any responses from the groups I reached out to. It is also important to note that within the context of this being a master's thesis, considerations of time bordered the study and the length of the recruitment period was limited to approximately 12 months. In addition, the study sample comprised parents who had received pediatric palliative care services, which included access to counseling. Therefore, their experience might not reflect the experiences of parents who have no access to such services.

I did not conduct follow-up interviews with participants to ask additional questions and to allow participants to reflect on their first interviews. However, in interpretive description studies, conventional member-checking is not generally recommended; it may lead to false

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confidence in interpretations if participants confirm findings, or it can potentially steer the researcher away from valuable analytic interpretations if participants disagree with the findings (Thorne & Darbyshire, 2005).

Nurse-researcher Role

Because of my role as nurse in pediatric critical care and pediatric palliative care and familiarity with caring for families who experience AG, I may have had unintentional bias based on my previous knowledge and assumptions. This could have resulted in a biased interpretation of the study data. I addressed this limitation throughout the research process by first identifying my preconceptions and assumptions at the beginning of the study, and by revisiting these assumptions throughout the research process, from the interviews to the writing of the results. In addition, at the beginning of each interview, I disclosed my nursing role in pediatric critical care and pediatric palliative care to the participants. I intentionally remained curious and asked for more details or clarifications when I felt participants omitted to share certain details based on their awareness of my role or on an assumed mutual understanding of their experience. It is also important to note that while “closeness does not make bias and loss of perspective inevitable; distance is no guarantee of objectivity” (Patton, 2002, p.49).

Reflections on Conducting Sensitive Research

I think it is important to share my reflections on being a novice qualitative researcher studying a sensitive topic. As a nurse in pediatric palliative care and pediatric critical care, I witness grief on a regular basis at work. While this might have prepared me to conduct an in-depth exploration into the topic of AG, there were challenges I did not expect to encounter throughout my study.

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After the first interview, I remember feeling emotionally drained. When I work at the bedside, while I support parents in their grief, my main role remains caring for their child, especially in the pediatric intensive care unit. My natural instinct as a nurse is to try to “fix” things. By offering support to parents at the bedside, I can hopefully help them navigate their emotions, at least for a brief moment. My main role when conducting research interviews was to listen without trying to “fix”. Listening to stories about some of the most difficult times in parents’ lives often made me feel powerless. While listening may seem like a simple task, it is far from trivial. To sit in the presence of such intense emotions, without focusing on trying to “fix”, was more challenging than anticipated. Conducting the interviews via an online platform was an additional challenge. I found it more difficult to convey empathy and compassion through body language via an online platform.

The challenges with processing the information shared with me continued beyond the interviews. After each interview, I needed time to process the emotionally charged stories that were shared with me. Furthermore, to immerse myself in the data, I watched the video recordings, listened to the audio and read the transcripts of each interview multiple times. While these research tasks seem purely technical, every encounter with the data was a reminder of parents’ grief and suffering. I am immensely passionate about the topic of AG and pediatric palliative care, but at times, I found myself not wanting to engage with the data for a period of time. I wonder if the impact of the COVID-19 pandemic played a role in finding this work more emotionally difficult than I had anticipated. At the height of the pandemic, everyone was constantly reminded of grief and death. I was conducting a study on grief while being surrounded by grief and death at work and in my personal life due to the pandemic. This likely resulted in conducting this study being more challenging than expected.

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While these challenges should not deter novice researchers from studying sensitive topics, their impacts are worth taking into consideration. I would encourage every novice researchers undertaking research on a topic that can elicit strong emotions to have a solid plan in place to cope with the emotional challenges their studies can present.

Chapter 7. Conclusion

This thesis has presented a research project which sought to explore and better illuminate the anticipatory grief experience of parents who have a child who died from a NMLLC. The shared stories of parents describe a grief that encompasses all aspects of their lives, from the loss of an imagined child to the loss of careers and social connections. This study suggested that parents experience multiples losses, including ambiguous losses, which have not been fully identified within the literature in previous studies on AG. Because of the ongoing losses and the omnipresence of grief in their lives, parents have to continuously adjust to new normals and adapt their day-to-day life based on the limitations imposed by their child's conditions. Various coping mechanisms were described, most importantly the importance of maintaining parental control, the value in having honest, compassionate communication with health care teams and the long-lasting impact of creating memories with their child. This study showed that AG in parents who have a child with a NMLLC would benefit from being examined through the lens of current grief models as well as other models/theories with overlapping concepts, such as ambiguous loss and chronic sorrow. Overall, this study provides enhanced knowledge about AG in parents of children who have a NMLLC and improves our understanding of the nuanced ways in which parents experience complex emotions surrounding ongoing losses and grief.

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Appendix A

Rando's Six Dimensions of Anticipatory Mourning (Rando, 2000, p. 52)

1. Perspective: patient, intimate, concerned other, caregiver
2. Time Focus: past, present, future
3. Influencing Factors: psychological factors, social factors, physiological factors
4. Major Sources of adaptational demands: loss, trauma
5. Generic Operations: grief and mourning, coping, interaction, psychological reorganization,
planning, balancing conflicting demands, facilitating an appropriate death
6. Contextual levels: intrapsychic-level processes, interpersonal processes (with the life-
threatened or dying person), systemic-level processes

Appendix B

Study Information Letter and Consent Form



Anticipatory grief in parents who have a child with a non-malignant life-limiting condition

Letter of information

Principal Investigator: Ms. Joanie Maynard, BSN RN, MN Student
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Project supervisors:

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Purpose of the Study

As part of my Master in Nursing degree, I am conducting a study to understand parents' experiences of anticipatory grief, which is grief that is experienced prior to the death of a loved one. This research topic has been informed by my work as a nurse at Canuck Place Children's Hospice and in the pediatric intensive care unit at BC Children's Hospital.

This research will address gaps in knowledge on the topic of anticipatory grief as experienced by parents of a child with a life-limiting condition. The overarching goal is to provide education and context to health care providers so they can better support parents who are navigating life with a child who lives with a life-limiting condition.

What will happen during the study?

As a parent of a child who lived with a child with a life-limiting condition, you are invited to share your experience during a one-to-one interview via video conference. You may be asked to participate in a second interview to provide further information or clarifications. However, you are not obligated and can decline the second interview. During the interview, I will ask you questions that are related to you and child, and to your experience of grief in the period prior to the death of your child. The first interview is expected to last 1-1.5 hours, while the second

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interview should last approximately 30 minutes. The interview(s) will be audio recorded. You will also be invited to offer feedback on your participation via an online questionnaire that will be sent to you a few weeks after your interview.

Are there any risks to the study?

This research addresses your experiences prior to the death of your child and you may feel increased emotional stress or distress as a result. You can decide to skip an interview question that you feel is too difficult to answer and/or you can choose to end the interview at any time and for any reason. A list of support resources will also be provided to you. You also have access to bereavement support at Canuck Place.

Are there any benefits to participating in this study?

You may appreciate having the opportunity to talk about your child and your experiences of caring for a child with a life-limiting condition. You may also benefit from knowing that your participation could help other families in pediatric palliative care in the future.

Privacy and Confidentiality

Your privacy and confidentiality will be respected. Any identifying information will be removed and kept separately from the data. All documents will be identified only by participant number. The results of this study, including direct quotes, may be reported in academic and professional journals and presentation(s), however, any potential identifying information in direct quotes will be removed/modified.

Paper documents will be kept in a locked filing cabinet and all documents that are kept on a computer will be encrypted and password protected. The data will be stored securely for five years, after which all study files and documents will be destroyed. The recordings of the interviews will be transcribed with no identifying information. You will not be identified by name in any reports, publications or presentations about the study. The data will be accessible only to Ms. Joanie Maynard (principal investigator), and Dr. Karen Cook and Dr. Jennifer Stephens (project supervisors).

What if I change my mind about being in the study?

Your participation in the study is voluntary, and you can withdraw from the study at any time. If you withdraw, there are no consequences to you, and your anonymous data will remain in the study. You do not have to answer all of the questions to remain in the study.

How will I find out what was learned in this study?

A report of the key findings of this study will be shared with participants who indicate that they are interested in receiving it.

Has ethical approval been secured?

The Athabasca University Research Ethics Board and the Canuck Place Children's Hospice Research Committee have reviewed and approved this study. If you have any comments or concerns regarding your treatment as a participant in this study, please contact:

Athabasca University Office of Research Ethics by telephone at 1-780-675-6718 or by email at .

Questions about the study:

If you have questions, or need more information about the study, please contact:

Ms. Joanie Maynard, BSN RN

Master in Nursing Student, Faculty of Health Disciplines, Athabasca University

Email address: jmaynard@cw.bc.ca Phone: 604 349 1233



Informed Consent Form

Project Name: Anticipatory grief in parents who have a child with a non-malignant life-limiting condition

Principal Investigator: Ms. Joanie Maynard, BSN RN, MN Student
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The time between a child's diagnosis of life-limiting condition and their death is often a period filled with grief and uncertainty. As a parent who has lost a child who had a life-limiting condition, you have been invited to participate in this study to share your experience. While I am conducting this study as a requirement to complete my Master of Nursing degree, I strongly believe that the knowledge gained from you sharing your experience is invaluable to health care providers and will help to improve support for families in the future. Your participation in this study is entirely your choice. Before you decide, it is important for you to read this consent form carefully so you fully understand what the research involves. This consent form will tell you about the study and why the research is being done. It will also tell you what will happen during the study and the possible benefits and risks of taking part.

What is the purpose of this research?

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The purpose of this study is to learn more about the grief you have experienced in the period prior to the death of your child. The grief experience of parents as they live knowing that the death of their child is inevitable is relatively unknown. I expect that this study will improve health care providers' understanding of what parents who care for a child with a life-limiting condition experience and that in turn, this will improve their practices.

What is involved in taking part in this study?

As a parent of a child who has a life-limiting condition (biological, step, adoptive, or legal guardian) will be invited to participate in one or two interviews via video conference. The interviews will consist of questions that are related to your experience of grief in the period prior to the death of your child. The first interview is expected to last 1-1.5 hours and the second one approximately 30 minutes. Interviews will be audio recorded. You will also have the opportunity to provide feedback on your participation via an online questionnaire that will be sent to you a few weeks after your interview.

What are the risks of taking part in this study?

This research study deals with a sensitive topic and you may experience increased emotional stress as a result. You can decide to skip an interview question that you feel is too difficult to answer and/or you can choose to end the interview at any time and for any reason. I will also give you a list of resources to help support you if needed. You can choose to leave the study at any time. If you tell us that you are thinking about hurting yourself or others, a health care provider will be notified.

What are the benefits of taking part in this study?

While there is no direct benefit to you for taking part in this study, you may find it helpful to talk about your child and experiences. Some parents appreciate having the opportunity to participate in a study that will help other families who may be facing a similar situation in the future.

How will my privacy and confidentiality be maintained?

Your privacy and confidentiality will be respected. Any identifying information will be removed and kept separately from the data. All documents will be identified only by participant number. The results of this study, including direct quotes, may be reported in academic and professional journals and presentation(s). Any potential identifying information in direct quotes will be removed/modified.

Paper documents will be kept in a locked filing cabinet and all documents that are kept on a computer will be encrypted and password protected. The recordings of the interviews will be transcribed with no identifying information. You will not be identified by name in any reports, publications or presentations about the study. All documents related to the study will be destroyed 5 years after the study is complete.

What if I decide to stop taking part in the study?

Your participation is voluntary and you may withdraw at any time. Any data collected up to the point of your withdrawal from the study will be kept for data analysis purposes, under strict provisions of confidentiality. You can let us know any way you like (for example by phone or email) or by telling us directly if you want to stop taking part in the study. If you decide to

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withdraw from the study, there will be no negative consequences on the services that you and your family receive from health care professionals.

Will I be reimbursed for taking part in the study?

As a token of appreciation for your time and willingness to share your experience, you will receive a \$25 e-gift card to a store of your choice.

Who do I contact if I have questions about the study?

If you have any questions or want more information about this study, you may contact Joanie Maynard, principal investigator, by phone at 604-349-1233 or via email at jmaynard@cw.bc.ca.

Who do I contact if I have concerns about our rights as research subjects?

This study has been reviewed by the Athabasca University Research Ethics Board. If you have any comments or concerns regarding your treatment as a participant in this study, please contact the Office of Research Ethics at 1-800-788-9041, ext. 6718 or by email to rebsec@athabascau.ca.

How do I get a copy of the results of the study?

You may ask for a copy of the final reports from this study.

I would like to receive a copy of the final results for the study. If so, please provide an email address where you would like the results sent: _____

Can I take part in other research?

Please indicate your preference about being contacted for other research studies in the future that you might be able to take part in:

I am willing to be contacted about other research that I might be able to take part in. If so, please provide an email address to contact you: _____

I do NOT want to be contacted about other research.

CONSENT

I read the consent form and understand what the study involves. Any questions I had about the study have been answered. I will keep a signed and dated copy of this form for my records.

My signature below confirms that:

- I agree to participate in this study;
- I understand the expectations and requirements of my participation in the research;
- I understand the provisions around confidentiality and anonymity;
- I understand that my participation is voluntary, and that I am free to withdraw at any time with no negative consequences;
- I am aware that I may contact the principal investigator, project supervisors or the Office of Research Ethics if I have any questions, concerns or complaints about the research procedures.

Name: _____

Date: _____

Signature: _____

By initialing the statements below,

_____ I am granting permission for the researcher to audio record the interview(s) .

_____ I acknowledge that the researcher may use specific quotations of mine, without identifying me.

Appendix C

Guiding Questions for Semi-Structured Interview

- Could you start by telling me how/when you found out about your child's diagnosis ?
- How did you feel in the immediate period after you found out about your child's diagnosis?
 - o And how did you feel in the weeks/hours (depending on circumstances of death) before he/she died?
 - o And how do you feel now?
- How would you describe your experience of having a child with a LLC?
- What main fears and anxieties related to your child's conditions did you experience?
- Even if you knew that your child would most likely not survive to adulthood, did you feel like you were somewhat prepared for an eventual decline in your child's health?
- Do you feel that you had already started to grieve the death of your child even if you didn't know exactly how much time he/she has left?
 - o Any other losses that you were grieving as their disease was progressing?
- How did you cope with knowing you would lose your child due to their diagnosis?
 - o What were your sources of strength during that time?
 - o Do you have any thoughts about what was not helpful in your experience?
- How would you describe the support and care you and/or your family received from your or your child's health care team (family physicians, specialists) during their disease trajectory, especially during the few months leading up to their death?

AG IN PARENTS WHOSE CHILD HAS A NMLLC

- Do you have any thoughts about what would be helpful in terms of supporting families like yours who have a child with a LLC, as they have to live with the knowledge that they will lose their child?
- Anything else you would like to share with me?

Appendix D

Demographic Questionnaire

Questions about you

Gender:

- Female
- Male
- Other
- Prefer not to answer

Relationship to child:

- Biological parent
- Adoptive parent
- Step parent
- Legal Guardia

Date of birth (month/year): _____

To which ethnic or cultural groups do you (or the majority of your ancestors) primarily belong?

What is your religious affiliation (if any): _____

How important is religion/spirituality to you?

- Very important
- Of medium importance
- Of little importance
- Not at all important

Questions about your child

** Questions about your child are referring to your child who has a life-limiting condition

What is your child's date of birth? _____

What is your child's diagnosis and when were they diagnosed (month/year)?

When did you notice something might be wrong with your child (month/year)? _____

When did your child die (month/year)? _____

Where did your child die?

- Home
- Canuck Place
- Other hospice
- BC Children's Hospital
- Local hospital

Appendix E

Audit Trail: Initial Coding/Themes Evolution

Date: 2022-08-11

Initial codes (table imported from NVivo)

Name	Files	References
acceptance	2	8
advance directives	2	4
advocating for your child	4	13
anger	2	3
autopsy	1	2
body in fight mode	1	1
Canuck Place	4	13
caring HCP	4	12
closeness	2	4
connection with child	4	13
connection with other parents	3	3
Coping	4	32
counselling	3	6
death	2	6
death location	4	6
decline	4	14
denial	2	4
difference between settings	4	15
Doing your own google research	1	2
Empathy	3	24
expertise of HCP in LLC	4	8
Feeling safe	2	4
Fiinancial stressors	2	3
Focusing on the now	3	6
gratefulness for being given timeline	1	2
grief being the new normal	3	5

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Name	Files	References
grief of life imagined	3	10
guilt feelings	2	4
HCP downplaying severity of condition	3	8
Hope	4	12
Impact on siblings	2	3
importance of HCP communication	4	25
importance of the little things	4	5
important memories	4	10
initial diagnosis	4	6
Judgment from HCP	2	8
lawsuit	1	1
leaving a legacy	2	2
limitations of the child	2	3
Maintaining control	4	16
maintaining normalcy	4	9
Maintaining role as parent	4	12
meaning of words	2	2
meaningful life	3	12
moving city	1	1
Multiple hospital visits	3	7
multiple losses	2	11
new normal	3	7
not understanding diagnosis	2	2
ongoing grief	3	12
online support	1	1
Onset of grief	3	5
parent health problems	2	3
parent's interpretation - initial diagnosis	1	3
parent's work	1	1
Post death grief	4	13
preparedness for decline symptoms	3	8

AG IN PARENTS WHOSE CHILD HAS A NMLLC

Name	Files	References
PTSD-like	1	1
right vs wrong decision	2	5
sadness	4	10
sharing knowledge of diagnosis	2	3
shock	1	1
sleep deprivation	2	2
social isolation	3	5
space between hope and grief	2	6
stress	2	4
support	4	20
supportive connection - instrumental	2	2
timeline	1	2
transparency from HCP	4	24
Uncertainty	4	17
unexpected death	2	3

Date: 2022-08-29

Codes starting to become organized into themes

Name	Comments	Description	Files	References
Acceptance of diagnosis			2	8
advance directives			2	4
anger			2	3
autopsy			1	2
body in fight mode	??? unsure how to rename and where it fits yet	Parent described her body being in “fight mode”	1	1
Importance of Canuck Place	Could be merged with “difference between settings	Parents described the impact of receiving support from CPCH, but also the impact of the physical space.	4	13
Emotional and physical connection to the child	Closeness and connection to the child merged together			

AG IN PARENTS WHOSE CHILD HAS A NMLLC

Theme:				
Healthcare providers				
caring HCP			4	12
Empathy			3	24
expertise of HCP in LLC			4	8
difference between settings (hospital vs hospice)			4	15
HCP downplaying severity of condition			3	8
importance of HCP communication			4	25
Judgment from HCP			2	8
sharing information about diagnosis/prognosis/changes in status			2	3
transparency from HCP			4	24
connection with other parents			3	3
Coping			4	32
counselling			3	6
death			2	6
death location			4	6
decline			4	14
denial			2	4
Doing your own google research			1	2
Feeling safe			2	4
Financial stressors			2	3
Focusing on the now			3	6
Benefit of being given timeline	Merge with timeline		1	2
Theme: Grief				
grief being the new normal			3	5
grief of life imagined			3	10
space between hope and grief			2	6
ongoing grief			3	12
Onset of grief			3	5
Post death grief			4	13
guilt feelings			2	4
Hope			4	12
Theme: Meaning Making				
importance of the little things			4	5
important memories			4	10
meaningful life			3	12
leaving a legacy			2	2

AG IN PARENTS WHOSE CHILD HAS A NMLLC

initial diagnosis			4	6
lawsuit			1	1
limitations of the child			2	3
Theme: Preserving role as a parent/changing parental role		Parents want to stay parents, but also see their role change as they also become caregivers		
Maintaining control			4	16
maintaining normalcy			4	9
Parents as advocates			4	13
right vs wrong decision		Parents being confident they are making the right decision for the child	2	5
Maintaining role as parent			4	12
meaning of words			2	2
Multiple hospital visits			3	7
Theme: multiple and ongoing losses			2	11
social isolation		Social isolation experienced by parents	3	5
moving city			1	1
parent's work		Parents having to stop working or change jobs	1	1
Impact on siblings		Sibling experiencing social isolation, not going to daycare, etc.	2	3
new normal		With every loss comes a new normal	3	7
not understanding diagnosis			2	2
online support			1	1
parent health problems			2	3
parent's interpretation of initial diagnosis			1	3
preparedness for decline symptoms			3	8
PTSD-like			1	1
sadness			4	10
shock			1	1
sleep deprivation			2	2
stress			2	4
support			4	20
supportive connection - instrumental			2	2
Uncertainty			4	17
unexpected death			2	3

Date: 2022-09-01

Email between myself and thesis supervisors to discuss refinement of themes/subthemes



Joanie Maynard <maynardjoanie@gmail.com>

Codes organization

Joanie Maynard <maynardjoanie@gmail.com>

Thu, Sep 1, 2022 at 4:52 PM

To: "Jennifer M.L Stephens" <jsteph35@uwyo.edu>, Karen Cook <kcook@athabascau.ca>

Hi,

I keep trying to figure out what makes the most sense as far as organizing codes into themes... so far this is where I'm at (see attached picture):

Theme 1: Initial diagnosis and stepping into a world of uncertainties

Theme 2: Onset of grief

Theme 3: Grief (emotional response to grief, grief of a life imagined, omnipresence of grief, the space between hope and grief)

Theme 4: Multiple losses/Anticipatory losses

Theme 5: Preserving parental role (advocating, involvement in care and decision-making, maintaining connection with child)

Theme 6: Meaning making (doing the normal things, focusing on the now, leaving a legacy)

Theme 7: hope

Theme 8: Impact of healthcare providers and healthcare settings

Theme 9: Supportive connections (counselling, family, community, etc.)

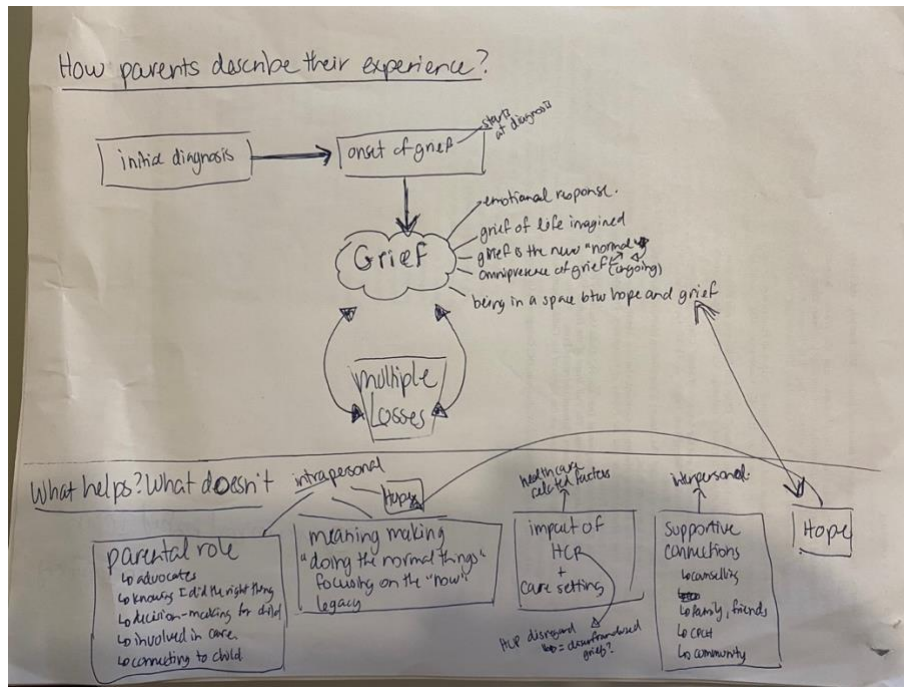
Thinking of grouping Themes 5-9 into interpersonal factors, healthcare related factors and intrapersonal factors. Not sure yet.

Themes 1-4 answer my first research question (how do parents describe their experience of AG, and the other themes answer my other research question (how do parents describe factors impacting their experience of AG?)

Thoughts? Comments?

Thanks,
Joanie

AG IN PARENTS WHOSE CHILD HAS A NMLLC



Date: 2022-09-18

Updated organization of themes and subthemes

Emotional Pandemonium

Theme 1 – Initial Strike

Subtheme: State of shock/What it means for the future?
 Subtheme: Onset of grief

Theme 2 – Walking Wounded

Subtheme: Ongoing losses and the omnipresence of grief
 - Grief of a life imagined
 - Grief being the new normal
 Subtheme: Uncertainty

Theme 3 – Coping

Subtheme: Parental role/maintaining control
 Subtheme: Hope and meaning making
 Subtheme: Impact of supportive connections
 - HCP and difference in health care settings

AG IN PARENTS WHOSE CHILD HAS A NMLLC

Theme 1 and 2 are part of this rollercoaster of intense emotions filled with uncertainties, which I called an “emotional pandemonium”. **Theme 1** is all about the “initial strike”, i.e. when parents sees that something is wrong with their child or the event that leads to a diagnosis. And then they ask themselves: what does this mean for the future? That’s also the onset of their grief journey. I named **Theme 2** “walking wounded”, because that’s what parents remind me of. Walking wounded are people who are injured but still walking. Parents are grieving but still have to keep on going.

Theme 3 is kind of a standalone theme. Theme 1 and 2 describes parents grief journey, which is a mix of grieving what has been lost while grieving what will be lost (anticipatory). Theme 3 describes factors that impact their grief, positively or negatively.

I have a decent amount of subthemes and subthemes of subthemes. Can I have subthemes of subthemes? Or maybe I can integrate the data into the “first level” subtheme instead of making it a subtheme of a subtheme.

Appendix F

Athabasca University Ethics Approval and Renewal Certificates



CERTIFICATION OF ETHICAL APPROVAL

The Athabasca University Research Ethics Board (REB) has reviewed and approved the research project noted below. The REB is constituted and operates in accordance with the current version of the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS2) and Athabasca University Policy and Procedures.

Ethics File No.: 24257

Principal Investigator:

Ms. Joanie Maynard, Graduate Student
Faculty of Health Disciplines\Master of Nursing

Supervisor:

Dr. Karen Cook (Co-Supervisor)
Dr. Jennifer Stephens (Co-Supervisor)

Project Title:

Anticipatory grief in parents who have a child with a non-malignant life-limiting condition

Effective Date: March 15, 2021

Expiry Date: March 14, 2022

Restrictions:

Any modification or amendment to the approved research must be submitted to the AUREB for approval.

Ethical approval is valid *for a period of one year*. An annual request for renewal must be submitted and approved by the above expiry date if a project is ongoing beyond one year.

A Project Completion (Final) Report must be submitted when the research is complete (*i.e. all participant contact and data collection is concluded, no follow-up with participants is anticipated and findings have been made available/provided to participants (if applicable)*) or the research is terminated.

Approved by:

Date: March 15, 2021

Barbara Wilson-Keates, Chair
Faculty of Health Disciplines, Departmental Ethics Review Committee

Athabasca University Research Ethics Board
University Research Services, Research Centre
1 University Drive, Athabasca AB Canada T9S 3A3
E-mail rebsec@athabascau.ca
Telephone: 780.213.2033

AG IN PARENTS WHOSE CHILD HAS A NMLLC



CERTIFICATION OF ETHICAL APPROVAL - RENEWAL

The Athabasca University Research Ethics Board (REB) has reviewed and approved the research project noted below. The REB is constituted and operates in accordance with the current version of the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS2) and Athabasca University Policy and Procedures.

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Principal Investigator:

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Faculty of Health Disciplines\Master of Nursing

Supervisor:

Dr. Karen Cook (Co-Supervisor)
Dr. Jennifer Stephens (Co-Supervisor)

Project Title:

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Effective Date: March 14, 2022

Expiry Date: March 14, 2023

Restrictions :

Any modification or amendment to the approved research must be submitted to the AUREB for approval.

Ethical approval is valid *for a period of one year*. An annual request for renewal must be submitted and approved by the above expiry date if a project is ongoing beyond one year.

A Project Completion (Final) Report must be submitted when the research is complete (*i.e. all participant contact and data collection is concluded, no follow-up with participants is anticipated and findings have been made available/provided to participants (if applicable)*) or the research is terminated.

Approved by:

Date: March 01, 2022

Carolyn Greene, Chair
Athabasca University Research Ethics Board

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Telephone: 780.213.2033



CERTIFICATION OF ETHICAL APPROVAL - RENEWAL

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Ethics File No.: 24257

Principal Investigator:

Ms. Joanie Maynard, Graduate Student
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Supervisor/Project Team:

Dr. Karen Cook (Co-Supervisor)
Dr. Jennifer Stephens (Co-Supervisor)

Project Title:

Anticipatory grief in parents who have a child with a non-malignant life-limiting condition

Effective Date: March 14, 2023

Expiry Date: March 14, 2024

Restrictions:

Any modification/amendment to the approved research must be submitted to the AUREB for approval prior to proceeding.

Any adverse event or incidental findings must be reported to the AUREB as soon as possible, for review.

Ethical approval is valid *for a period of one year*. An annual request for renewal must be submitted and approved by the above expiry date if a project is ongoing beyond one year.

An Ethics Final Report must be submitted when the research is complete (*i.e. all participant contact and data collection is concluded, no follow-up with participants is anticipated and findings have been made available/provided to participants (if applicable)*) or the research is terminated.

Approved by:

Date: March 10, 2023

Paul Jerry, Chair
Athabasca University Research Ethics Board

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