

**“You do realize that I'm like totally winging this?”:
Exploring the diagnosis, disclosure and coping experiences
of persons living with ALS**

by

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Author's Declaration

I hereby declare that I am the sole author of this thesis. This is a true copy of the thesis, including any required final revisions, as accepted by my examiners.

I understand that my thesis may be made electronically available to the public.

Abstract

Background: Amyotrophic Lateral Sclerosis (ALS), an incurable motor neuron disease, primarily affects those between the ages of 60-79, and sees an approximate post-diagnosis life-expectancy of only 2-5 years. The condition has an unpredictable but ultimately terminal trajectory that poses a number of challenges for patients, caregivers and healthcare providers. One of these major challenges is the need to make sure that patients' quality-of-life is as high as possible throughout the disease course. Many factors have been shown to influence quality-of-life, including patients and caregivers' ability to cope and adapt to the changes associated with the disease. There is some evidence to suggest that the manner in which healthcare providers present the information and empathize with their patients' thereafter, in addition to patients' initial reactions to hearing their diagnosis, may hold some predictive value or have an impact on subsequent coping-related outcomes. But still, our knowledge regarding the relational, communicational and psychodynamic forces that occur within the process of diagnostic disclosure is relatively limited. Therefore, further investigation of patients and caregivers' experiences in the conversations surrounding their ALS diagnosis is necessary to enhance guidelines and practices towards improved coping and quality-of-life support for people living with ALS.

Objectives: The purpose of this study was to explore the experiences of persons living with ALS in diagnosis and disclosure, with specific attention paid to their experiences in hearing their diagnosis, and their preferences for care and coping support. The perspectives of those living with the disease and those of their caregivers were considered.

Methods: Data were collected from a sample of 18 people consisting of persons with ALS/ PLS ($n = 9$), family caregivers ($n = 7$), a professional caregiver ($n = 1$), and one past caregiver ($n = 1$).

Data were collected during individual ($n = 5$), dyad ($n = 8$) and group ($n = 9$) interviews that were conducted using interview guides comprised of a series of open-ended questions related to the study's research questions. The interviews were audio-recorded and transcribed verbatim. A thematic analysis of the acquired data were conducted using methods outlined by Braun & Clarke (2006) in order to establish major themes. Coding was done using NVivo 11 software.

Results: The data revealed six major themes that specifically relate to the ALS diagnosis process, eight themes relating to the diagnostic disclosure process, and eight themes pertaining to the coping process. Three major themes were established with regards to participants' desired experiences for support in their coping: hope stimulating conversations and activities, ongoing information provision aimed at curbing uncertainty, and independence, autonomy, lifestyle and normalcy supported throughout the disease course.

Significance: This project serves as an initial step in bridging the relevant gaps in our knowledge and understanding toward improved patient-centred care practices in diagnosis, disclosure, care and coping support for persons with ALS. There is also potential for the project's findings to guide practice and policy developments to benefit the care of persons with other illnesses characterized by short and unpredictable trajectories.

Keywords: *Amyotrophic Lateral Sclerosis (ALS), Diagnostic Disclosure, Coping, Quality-of-life, Qualitative methods, Thematic Analysis*

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Dedication

To my mom, who has been my primary resource for strength and encouragement;
to my brother, who watches over me;
to my sister, who keeps me grounded;
to my husband and children, who offer me unconditional love;
and to my dad, who inspired this project.

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List of Abbreviations

ALS – Amyotrophic Lateral Sclerosis

PLS – Primary Lateral Sclerosis

MND – Motor Neuron Disease

MNDCS – Motor Neuron Disease Coping Scale

ACP – Advance Care Planning

MRI – Magnetic Resonance Imaging

EMG – Electromyography

HCP – Healthcare Provider

PSW – Personal Support Worker

PWALS – Person with ALS

PWPLS – Person with PLS

DTA – Declined to Answer

CCAC – Community Care Access Centre

LHIN – Local Health Integration Network

Chapter 1: Introduction & Overview

ALS (Amyotrophic Lateral Sclerosis) is a devastating neurodegenerative disease (Amyotrophic Lateral Sclerosis Society of Canada Canada [ALS Canada], 2016). Though we once thought of it as a relatively rare condition, it has been found to affect an increasing number of people (Strong & Rosenfeld, 2003, Arthur et al., 2016). This is particularly troubling given that ALS illness trajectories are always terminal, despite varying in length, and are typically characterized by many challenges related to functional and, sometimes, cognitive decline (ALS Canada, 2016). Because of the condition's incurable nature, health-related quality-of-life for persons with ALS has become an important focus in the literature (Pagnini, 2018; ALS Canada, 2016; Clarke, Hickey, O'Boyle & Hardiman, 2001; Miller et al., 2009).

The concept of quality-of-life has proven to be complex, and its condition, affected by many factors, both directly and indirectly (Pagnini & Castelnuovo, 2016; Matuz, Birbaumer, Hautzinger & Kübler, 2015; Pagnini, 2018). In particular, patients' and caregivers' ability to cope and adapt to the changes associated with disease have been associated with self-perceived quality-of-life (Matuz, Birbaumer, Hautzinger & Kübler, 2010; Montel, Albertini & Spitz, 2012a; Matuz et al., 2015; Jakobsson Larsson, Nordin & Nygren, 2016). With that, there is some evidence to suggest that the way patients react to hearing an unfavourable diagnosis like ALS, and the manner in which healthcare providers present the information and empathize with their patients' in relaying such diagnoses, may hold some predictive value or have an impact on subsequent coping-related outcomes (Sparks, Villagran, Parker-Raley & Cunningham, 2007; McCluskey, Casarett & Siderowf, 2004; Shaw, Brown & Dunn, 2013; O'Brien, Whitehead, Jack & Mitchell, 2011). Conceringly, our understanding of the relational, communicational and psychodynamic intricacies that exist within the process of diagnostic disclosure is relatively

limited (Pavey, Allen-Collinson & Pavey, 2013). Therefore, a better understanding of patients and caregivers' experiences in the interactions surrounding an ALS diagnosis is an asset in attempts at enhancing guidelines and practices towards improved coping and quality-of-life support for people living life-limiting illnesses.

Chapter 2: Background & Literature Review

2.1 ALS & Its Impact

ALS is a progressive and incurable multi-system disorder characterized by muscle wasting, severe disability, and unpredictable illness trajectories. Its symptoms are caused by rapid brain and spinal cord nerve cell degeneration, and typically lead to death within three years of initial symptom onset (ALS Canada, 2016; Oliver, Borasio & Johnston, 2014; Murray & Butow, 2016). Sadly, the nature of the disease necessitates a great deal of effort in coping and caregiving, such that many sufferers, caregivers, and care providers experience stress in the uncertainty that results from the disease's burden (Hecht et al., 2003; Pagnini et al., 2010). Patients and their families also indicate a deep-rooted sense of worry and a loss of control over their health and bodies after diagnosis (ALS Canada, 2016; Oliver et al., 2014).

Current research data tell us that, in Ontario, about five in every 100,000 people live with ALS (Wolfson, Kilborn, Oskoui & Genge, 2009). Canadian national survey statistics indicate that our health system sees about two new cases in every 100,000 people each year (ALS Canada, 2016; Chio et al., 2013; Strong & Rosenfeld, 2003). It is predicted that the global incidence of ALS will increase by an average of nearly 70% by the year 2040 (Arthur et al., 2016). The average annual cost of care for each individual patient, which is currently estimated to be between \$30,000 - \$70,000 in Canada and the US, is also expected to rise significantly (Gladman, Dharamshi & Zinman, 2014; Miller et al., 2014).

2.2 ALS & the Aging Population

The anticipated increase in diagnosed cases of ALS has been attributed, largely, to our aging population. As we are becoming increasingly aware, the proportion of people over the age of 60 years is rapidly expanding. The number of older adults, worldwide, is expected to double from 1 billion to 2 billion between 2020 and 2050 (Clegg et al., 2013). Thus, by 2050 older adults will account for more than 20% of our global population (Bloom, Canning & Fink, 2010). This shift is accompanied by significant challenges, predominantly due to the association between advanced age and a higher risk of disease and frailty (Clegg et al., 2013; Arthur et al., 2016).

Over time, our bodies slowly lose their ability to function optimally. This decline occurs at both the physiological and physical level, and precipitates gradual increases in susceptibility to various conditions, and vulnerability to medical complications and death. As such, ALS is most commonly diagnosed in later adulthood, with older age considered a major prognostic factor (Arthur et al., 2016; Logroscino, et al., 2015; Robberecht & Philips, 2013).

Research shows that ALS can be classified as an “age-dependent neurodegenerative disease like Parkinson’s and Alzheimer’s disease” (Logroscino, et al., 2015, p. 142). Its rate of incidence peaks around the eighth decade of life, while the majority of those living with the condition are men and women between the ages of 60-79 years (Arthur et al., 2016; Wolfson et al., 2009; Logroscino, et al., 2015). Hence, as our older adult population expands, so will the number of people living with ALS, warranting increased research efforts aimed at ameliorating relevant healthcare services (Arthur et al., 2016; Andersen et al., 2012).

2.3 Current Challenges & Opportunities

2.3.1 ALS in an Evolving System

Compared to other chronic conditions, ALS trajectories are considered relatively short, and unpredictable. Where some related disorders are associated with slow, predictable disease progression, and decade, or multi-decade, long life-expectancies, people with ALS typically survive less than 5 years, with fluctuating rates of decline throughout (Eisen & Calne, 1992; Tom et al., 2015; Rodriguez et al., 2015). This represents a significant challenge for our healthcare system which faces increasing demand for a range of long-term chronic care services. These services are usually tailored to those conditions that are more wide-spread, better understood, and more easily managed. Meanwhile, our system remains best suited to support patients in urgent medical crises, through predictable, short-term recoveries (Goldsmith, 1990; Marchildon, 2013; Ministry of Health and Long-Term Care (MOHLTC), 2007; Zwar et al., 2017; Arthur et al., 2016).

Given the current state of our healthcare system, patient-provider interactions are often centred around patients' physical symptomatology (Wagner, Austin & Von Korff, 1996; Goldsmith, 1990; MOHLTC, 2007). These interactions remain characterized by a diagnose-cure dynamic, just as was the case amid the post-world war II period, where a curative culture was maintained as a means of combatting the common communicable diseases of the era (MacIntosh, Rajakulendran, Khayat & Wise, 2014). However, despite dedicated efforts aimed at forwarding a cultural shift, the ideals of a patient-centred paradigm have yet to be completely embedded within the various layers of our system. Met by healthcare providers' general lack of familiarity with the unique nature of ALS, patients' needs are often left unaddressed, and their expectations unmet (Oliver et al., 2014; Boyd, Cooper & O'Brien, 2016; Jakobsson Larsson, 2016).

2.3.2 The Unique Needs of Persons with ALS

ALS' relentless attack on the body's physical functioning, as well as its incurable nature, make the disease an unfavourable diagnosis. People are often reported to respond to hearing a diagnosis of ALS with initial shock and distress, anger or worry, and, in many cases, disbelief and denial (Hogden, Greenfield, Nugus & Kiernan, 2012a; Hugel, Grundy, Rigby & Young, 2006; Goldstein & Leigh, 1999). In effect, these reactions are said to be quite common across various life-limiting conditions. In fact, many people faced with bad news about their health are believed to experience some level of denial or wishful thinking throughout their disease course (Zimmerman, 2004; Baile et al., 2000). Some studies suggest that these reactions can negatively affect coping, and potentially hinder participation in subsequent care planning, and should therefore be mitigated (Hogden et al., 2012a; Brown & Addington-Hall, 2008; Andersen et al., 2012). In response to such evidence, general guidelines for diagnostic disclosure have been developed and implemented to aid healthcare providers in effectively delivering the bad news to their patients, while identifying and dealing with unfavoured coping mechanisms such as denial (Baile et al., 2000; Andersen et al., 2012; Miller et al., 2009).

Though well received, many of the current guidelines that exist were not developed and tested specifically for use in ALS care. When it comes to communication and relational care, current practices in ALS care are, and have, typically been guided by parameters outlined for cancer care focused initiatives (Bolmsjo & Hermeren, 2001; Baile et al., 2000; Miller et al., 2009; Astrow et al., 2008). Though such initiatives have generated a valuable basis in knowledge from which care recommendations for ALS can continue to be developed, research on how cancer care-informed recommendations might be tailored to ALS care is still lacking (Baile et al., 2000; Miller et al., 2009).

There are indeed some similarities between conditions like ALS and advanced cancer, as in their association to aging, their unpredictable trajectories, and the related feelings of distress and anxiety. But, peoples' experiences in ALS are, in many cases, much different from those in advanced cancer (Hogden et al., 2012a; ALS Canada, 2016; Bolmsjo & Hermgren, 2001; Walton et al., 2016). As one of the participants in Hogden et al.'s (2012a) study eloquently articulated: "Cancer is a walk in the park by comparison [to ALS]...where you don't have some sort of fighting chance, some glimmer of hope." (p. 833). This suggests that there may be important psychodynamic and psychosocial factors that mark key differences between cancer and ALS care needs, and that pose a unique challenge for sufferers.

The vast majority of people with ALS eventually succumb to respiratory failure. This is often following extensive diagnostic investigation, potential pharmacotherapy, and rigorous efforts in care and support; including counsel on the use of breathing supports, feeding tubes, mobility aids and communication devices. The myriad of complex medical decisions that befall those affected by ALS soon after diagnosis, matched by uncertainty, and unpredictability in disease progression, make contending with ALS a very unique and personal experience (de Sousa et al., 2017; Hogden et al., 2012a; ALS Canada, 2016; Andersen et al., 2012; Miller et al., 2009).

Overall, we know little about the efficacy and transferability of current tools and practices for persons with ALS (Baile et al., 2000; Schellenberg, Schofield, Fang & Johnston, 2014). Moreover, the relationship between methods of diagnostic disclosure, resultant reactions and relational dynamics should be further investigated as marked areas of potential differences in need. These matters may need to be acknowledged in the adaptation of various guidelines and

recommendations, and in healthcare provider training (Baile et al., 2000; Aoun et al., 2018; Miller et al., 2009; de Sousa et al., 2017; Schellenberg et al., 2014).

2.3.3 The Diagnosis Process

The term ‘motor neuron disease’ (MND) denotes a series of conditions that are characterized by nerve cell degeneration, or more specifically, the degeneration of the motor neuron cells. ALS is the most common type of MND (Oliver et al., 2014; Strong & Rosenfeld, 2003; ALS Canada, 2016). Other classifications of MNDs include progressive muscular atrophy (PMA) and primary lateral sclerosis (PLS), which are diagnoses made based on specific presentations of symptoms (Oliver et al., 2014; Strong & Rosenfeld, 2003).

Diagnosing ALS is typically a very involved process, as there is no one test that can determine, with certainty, whether or not the presented symptoms are related to the disease (Oliver et al., 2014; Boyd et al., 2016; Eisen, 1999). The *El Escorial* criteria for the diagnosis of Amyotrophic Lateral Sclerosis, originally developed in 1990 by the World Federation of Neurology are applied worldwide by clinicians and scientists in efforts to identify and classify symptoms of the disease as they are related to either upper or lower motor neuron degeneration (Brooks, 1994). These criteria define subsets of clinical features that guide suspected, possible, probable and definite diagnoses of the condition, as well as recommended methods of ruling out mimicking conditions, including electromyography (EMG) and magnetic resonance imaging (MRI) testing (Brooks, 1994; Oliver et al., 2016; Strong & Rosenfeld, 2003).

Primary healthcare clinicians are often Canadians’ first point of contact within our healthcare system (Canadian Institute for Health Information [CIHI], 2016). For ALS patients, and those presenting possible symptoms of the condition, primary care services offer referral and

access to multidisciplinary care clinic teams that ideally work in a coordinated way to facilitate the diagnosis process, help guide efforts in symptom management, and support the prevention of medical complications and premature death (CIHI, 2016; Andersen et al., 2015; Mitsumoto & Rabkin, 2007).

A patient's journey is likely to begin with complaints of initial symptoms such as muscle cramps, weakness and fatigue; shortness of breath, numbness and/or tingling in the extremities. These initial symptoms are usually first discussed with a primary care physician who considers the case, then refers the patient to a specialized neurologist for testing, and disease confirmation, if appropriate. Once ALS has been confirmed through the reasonable exclusion of other conditions, the patient's specific needs are assessed and a care team is assembled (Mitsumoto & Rabkin, 2007; ALS Canada, 2016; Andersen et al., 2012).

There exist a number of challenges and barriers to patients' optimal care (Miller et al., 2009; Andersen et al, 2012). First, a major systemic issue for persons with ALS is the low rates of specialized clinic attendance (Andersen et al., 2012; Miller et al., 2009; Rooney et al., 2015; Hogden et al., 2017; Stephens et al., 2015).

In Canada, ALS clinics represent a gold standard of care for patients with the condition (Andersen et al., 2012; Miller et al., 2009). In such clinics, patients receive specialized care by way of facilitated access to interdisciplinary healthcare services. Led by neurologists, specializing in MND, patients are to be individually assessed, monitored, consulted and informed. Those who make use of the services that specialized ALS clinics have to offer have been found to experience fewer medical complications, reduced hospital admissions, improved quality-of-life and prolonged survival, as compared to those who visit general clinics (Andersen et al., 2012; Miller et al., 2009; Rooney et al., 2015). Patients are often highly encouraged to

complement the care they receive from their family doctor, homecare provider, community supports and others, with visits to an ALS clinic (Rooney et al., 2015; ALS Canada, 2016).

However, many people, namely older adults, tend not to frequent ALS clinics until severe symptom onset, if at all (Andersen et al., 2012; Rooney et al., 2015; Stephens et al., 2015; Hogden et al., 2017). This presents a number of missed opportunities to maximize care outcomes, and perhaps suggests ongoing system and organizational-level insufficiencies related to patient education and accessibility (ALS Canada, 2016; Andersen et al., 2012; Miller et al., 2009; Rooney et al., 2015; Stephens et al., 2015).

Second, because a diagnosis is often a prerequisite to gaining access to specialized and funded services, many people go without until an accurate diagnosis is reached. This can take upwards of a year, and misdiagnosis is relatively common in the initial stages of the process (Boyd et al., 2016; ALS Canada, 2016; Oliver et al., 2014). The often convoluted process of arriving at a confirmed diagnosis of ALS can often affect patients' relationships with their care provider's, and thus their overall satisfaction in their care, making good communication an essential part of the process (ALS Canada, 2016; Hogden et al., 2017; Mishler, 1984).

2.3.4 Communication & the Care Process

Patients' knowledge about the services available to them, and how or when to best use them, as well as their overall benefit from having access to specialized ALS clinics, are closely tied to decision-making support through coordinated care and communication. This is such that rich care experiences and optimal outcomes are facilitated by the active incorporation of various stakeholder perspectives into the decision-making process, and the support and guidance offered

by healthcare providers (Rooney et al., 2014; Miller et al., 2009; Hogden et al., 2017; Mishler, 1984).

Thus, the reality that many patients are not receiving the valuable care services that ALS clinics have to offer may suggest that there are gaps or shortcomings in care providers' communication and relational practices. Because patient-provider relationships are seen as "linch-pins in the health care process" (Boyer & Lutfey, 2010, p. S81), such that individual patient experiences in trust, interaction, and quality in care are directly related to their health outcomes, and service utilization, it is likely that the way in which providers currently interact and communicate with persons with ALS might be impacting their engagement with specialized services. If patients are not being appropriately inspired to seek out services, or informed of these services' unique benefits and the different avenues they can take to access them, it is no surprise that attendance might suffer. Thus, a specific emphasis on the need for enhanced communication between healthcare providers and their patients is warranted.

Further, the idea of 'hope' as central to patients' experience in terminal illness suggests that patients' cognitions and complementary core beliefs play a pivotal role in shaping their experiences in coping with their illness, and in the care process (Snyder, Wroblewski, Parenteau & Berg, 2004). But, as of yet, we know little about how to instill hope in patients recently diagnosed with ALS through effective communication practices. We also need to know more about how a sense of hope might uniquely impact their experiences in care (Andersen et al., 2012; Miller et al., 2009). It is for this reason that an exploration of patients' perspectives on what influences their cognitive realities in health and care is important. Too, a more profound understanding of ALS patients' thought processes, emotions and behaviours may open the door

to the discovery of alternative influential cognitive processes and provide insight to inform tailored practical suggestions for fostering hope and alternatives.

According to Mishler (1984), patient voices and their understandings of their problems and circumstances are an important part of effective and appropriate medical practice. In typifying various patterns of communication in medical settings, he emphasizes the importance of acknowledging patients' personal and social contexts in order to best understand the realities of their conditions to be able to offer the most appropriate care and treatment. Mishler (1984) indicates that most patient-provider communications tend to be one-sided, in that it is usually the provider that does all of the talking. Correspondingly, he advocates for open dialogue and patients being given the opportunity to ask questions of their providers.

Our understanding of patient-provider communication have since evolved to further emphasize considerations of not only communication processes between patients and their physicians, but also communication as it relates to patients and their families, gendered preferences, cultural competence and varying communicational limitations, may they be physical, developmental or otherwise (Mude, Simon, Scherz & Parham, 2012; Cooper-Patrick et al., 1999; Cooper & Roter, 2003; Cooper et al., 2003).

The complexity of the ALS journey just in terms of service navigation requires a great deal of communication to support effective coordination and continuity. From the beginning stages of the care journey that typically starts during the diagnosis process and continues through to the later stages of disease care until death, it is certainly important that healthcare providers are able to maintain open lines of communication for patients and their families to be able to ask questions and understand their disease course. Adapting to patients changing communicational needs in relation to their physical decline is also likely to be necessary (Oliver et al., 2014). In

addition, patients' communicational needs have been shown to differ depending on where they live (e.g. at home, in hospital, etc.), and in accordance with their culture and spiritual beliefs, as well as their gender, such that gender accordance among care providers and patients can be advantageous (Oliver et al., 2014; Mude et al., 2012; Cooper-Patrick et al., 1999). Research advocates for improved provider training programs and increased knowledge regarding individual patient needs and preferences (Oliver et al., 2014; Mude et al., 2012).

2.3.5 Diagnostic Disclosure as a Critical Period for Intervention

The tools and guidelines currently available for diagnostic disclosure do not offer specific instructions for how to go about disclosing the diagnosis of ALS towards successful coping (Miller et al., 2009; Buckman, 2005; Andersen et al., 2012). A large number of physicians still report experiencing a great deal of stress and uneasiness when breaking the news of terminal illness to patients. Meanwhile, many patients say that they were left feeling under-informed and discontented after hearing the news (McCluskey et al., 2004; Connolly, Galvin & Hardiman, 2015; Shaw et al., 2013). As there are indications of remaining gaps, it is evident that there is still a need for practice-level improvements to be made to the disclosure process (McCluskey et al., 2004; Schellenberg et al., 2014).

As a sensitive and stressful step in the care process, diagnostic disclosure is a critical period for intervention which is potentially predictive of future coping-related outcomes (Sparks et al., 2007; Shaw et al., 2013; O'Brien et al., 2011). Thus, there is likely to be great benefit to working towards improving current guidelines and recommendations by expanding our knowledge base about peoples' experiences in the process. A deeper understanding of varying ALS care experiences in diagnostic disclosure may lay the foundations for improving the process

could be improved to better facilitate subsequent coping and quality-of-life (Shaw et al., 2013; O'Brien et al., 2011; McCluskey et al., 2004; Schellenberg et al., 2014).

2.3.6 Past & Present Approaches to Diagnostic Disclosure

The enduring short-term, recovery oriented paradigm has influenced the system-, organizational-, and individual-level activities in care provision, as it has shaped the development of many of the tools, programs, and services currently in use. This is especially true with regard to the tools and strategies used to guide the communication of a terminal diagnoses (Wagner et al., 1996; Goldsmith, 1990; Nolte & McKee, 2008).

Prior to the 1950s, concealing information to protect the provider's reputation and the patients' sensibilities was the norm in medicine (Jutel, 2017; Sisk, Frankel, Kodish & Isaacson, 2016b; Sisk, Bluebond-Langner, Wiener, Mack & Wolfe, 2016a; Cathell, 1890; Sutro, 1915). During the 19th and early 20th century, doctors were seen to have a great deal of authority as 'truth' holders and disseminators. Doctors were given discretion to share such 'truth' as they saw appropriate (Jutel, 2017).

During this time, most sources of ailment were communicable and curable, and there was less of a focus on the care of those with chronic and terminal illnesses. This lack of focus persisted until improved hygiene and vaccinations lowered risks of infection, and cancer and other chronic illnesses became more prevalent (Nolte & McKee, 2008). Amid the beginning stages of the shift from an acute to a chronic disease care-oriented paradigm, our limited knowledge of non-communicable diseases led decision-makers to advocate for stress mitigation and hope preservation by means of withholding diagnostic and prognostic information (Sisk et al., 2016a; Sisk et al., 2016b; Nolte & McKee, 2008; Jutel, 2017).

It is now widely accepted that patients have the right to be informed, but even so, we are still playing ‘catch up’ in our development of best-practice methods that fit the current paradigm (Nolte & McKee, 2008; Sisk et al., 2016a; Sisk et al., 2016b).

Two modern tools commonly used for diagnostic disclosure include the six-step SPIKES protocol, described by Baile et al. (2000), and the parameters outlined by Andersen et al. (2012) of the EFNS task force. The SPIKES protocol lays out a series of steps to be followed during conversations in which bad news is delivered. These steps are aimed at guiding healthcare providers in gathering information, transmitting relevant information, providing support, and encouraging patients’ engagement in care planning (Baile et al., 2000). The EFNS task force lists a number of recommendations to be considered prior to and during conversations in which a diagnosis of ALS is to be relayed. Among some of the recommendations listed are: a comfortable quiet setting in which to communicate the bad news, an understanding of who the patient is, and what their communicational preferences are, prior to disclosure, and the use of simple and carefully worded statements (Andersen et al., 2012).

Both of these tools have been correlated with improvements in patients’ satisfaction surrounding the manner in which the news of their illness was broken to them (Connolly et al., 2015; Shaw et al., 2013; McCluskey et al., 2004). However, even with significant advancement since the 1980s, a large number of physicians still report experiencing a great deal of stress and uneasiness when breaking the news of terminal illness to patients, while many patients indicate that they are left feeling under-informed and discontented (Sisk, et al., 2016a; Sisk et al., 2016b; McCluskey et al., 2004; Schellenberg et al., 2014).

Notwithstanding our systems efforts to promote well-being and quality-of-life through adaptive chronic care provision, current parameters for diagnostic disclosure tend to be vague

and insufficient to adequately support healthcare providers in effectively facilitating conversations that focus on outcomes, rather than just treatment options, with their patients. This risks negative experiences in both care provision and receipt (Marchildon, 2013; MOHLTC, 2007; Andersen et al., 2012; McCluskey, et al., 2004; Sisk et al., 2016a).

2.3.7 Current Models of Coping for Persons with Chronic Illness

Coping is a complex process whereby individuals attempt to manage the demands of a stressful situation (Lee et al., 2001). This process typically involves the use of various tools and resources to mitigate and/or work through the stress. For persons with ALS, the ability to cope in their disease is a key factor in survival and maintaining quality-of-life. But, despite its significance, few studies have broached the topic (Lee et al., 2001; Matuz et al., 2010; Jakobsson Larsson et al., 2016). Our current understanding of the various coping strategies that individuals with ALS use in their disease tend to revolve around the ideas of ‘processes’ and ‘resources’ (Lazarus, 1993; Hobfoll, 1989). Denial and avoidance are commonly discussed in the literature, as are patients’ desires to maintain control, variable levels of individualized efforts in resilience, and the importance of social support (Matuz et al., 2010; Jakobsson Larsson et al., 2016; Rabkin, Wagner & Del Bene, 2000). In addition, a number of studies have shown that older adults tend to show greater resilience than their younger counterparts (Folkman, Lazarus, Pimley & Novacek, 1987; Felton & Revenson, 1987; Jakobsson Larsson, Nordin, Askmark & Nygren, 2014; Matuz et al., 2010). This, perhaps, suggests that there are learned components of coping capacity, and offers potential opportunities for practice improvement by way of learning and understanding what it is that makes older adults more resilient, or better able to cope with their diagnoses (Matuz et al., 2010; Jakobsson Larsson et al., 2014).

One of the most commonly referenced models for coping with chronic illness is Lazarus and Folkman's (1984) transactional stress-coping model, which has been adapted for a broad variety of chronic diseases including ALS and other MNDs (Jakobsson Larsson et al., 2016; Pagnini & Castelnuovo, 2016; Manne, 2002). This model highlights a series of complex interactions, or transactions, between various contextual factors, perceptions, and expectations that ultimately shape the individual's psychosocial adjustment to their illness. The model identifies two separate, but sometimes overlapping, styles of coping: problem-focused and emotion-focused (Lazarus and Folkman, 1984; Manne, 2002; Swash, 2010; Matuz et al., 2010; Matuz et al., 2015; van Groenestijn et al., 2011).

Subsequent research has determined that those who adopt problem-focused styles of coping tend to fare better than those who adopt more emotion-focused styles (Graven et al., 2014; Lazarus, 1993; Hobfoll, 1989). Problem-focused styles of coping have often been associated with greater resilience, as compared to emotion-focused styles of coping, which have been associated with avoidance and denial (Graven et al., 2014; Matuz et al., 2015). As avoidance and denial can pose a significant challenge for healthcare providers in their attempts to engage patients in care and support-seeking towards better coping, efforts may be made to intervene or divert related behaviour (Graven, 2014; Matuz et al., 2015; Hogden et al., 2012a).

Other theories of coping revolve around ideas about cognitive processing (Janoff-Bulman, 1992) and social comparison (Gibbons & Gerrard, 1991). Common themes among the aforementioned models of coping are summarized in table 1.

Table 1: Common Themes among Models of Coping

Common Theme	Transactional Model (Lazarus & Folkman, 1984)	Cognitive Processing Model (Janoff-Bulman, 1992)	Social Comparison Model (Gibbons & Gerrard, 1991)
Loss	Illness is viewed as a stressor induced by the threat of harm/ loss of functioning.	Illness is perceived as a loss of ‘previous’-self.	Illness is perceived as a loss of optimal functioning
Illness Experience	Stress responses and coping are associated with cognitions and perceptions of controllability of their condition.	Individuals work to integrate their illness experience into their pre-existing beliefs of themselves and the world round them.	Illness is viewed as a threat to sense-of-self.
Uncertainty	Illness is considered threatening and disruptive, requiring ongoing re-evaluations of one’s illness experience.	The unpredictable nature of illness motivates individuals to reassess their core beliefs and perceptions.	Health problems increase uncertainty, and thus pursuit of certainty.
Appraisal	Coping is the product of cognitive and behavioural efforts driven by ongoing evaluations of individual and contextual factors, against accessible resources for adjustment/ internal regulation (either emotion-focused or problem-focused).	By focusing on the positive aspects of their illness experience, and searching for meaning, individuals attempt to shape new identity/ core beliefs.	Ill individuals tend to place themselves on a spectrum of severity and focus on those who are worse off (downward social comparison) to reduce perceived threat

These models highlight the complex and dynamic nature of coping with a life threatening illness. In particular, Janoff-Bulman’s (1992) cognitive processing model of coping addresses the struggle that lies in adapting to a new way of thinking of one’s own functioning, and finding

purpose as one learns to navigate the world in a new way; while Gibbons & Gerrard's (1991) social comparison model of coping emphasizes individuals' tendency to want to compare, contrast, and seek information about their peers in an effort to try to place themselves in their condition, along a spectrum of severity.

These models of coping help us visualize and differentiate patients' various coping styles, and reveal the common goals for mitigating stress responses and managing cognitive dissonance. That said, what we still lack is a comprehensive conceptualization of the models' applicability in various contexts, through diagnosis and disclosure to successful coping and high quality-of-life, from which personalized coping support plans may be fashioned (Manne, 2002; Graven et al., 2014).

2.3.8 Understanding Patients' Use of Coping Strategies

Although we do have some insight into the long-term progression of peoples' processes in coping, what we currently know about coping in the early stages of ALS is extremely limited. It is important to note and understand the different coping strategies that patients use throughout the disease process, particularly in the initial stages. With a deeper understanding of these different approaches to coping, the system may begin to improve patient-centred practices towards greater quality-of-life and, potentially, increased longevity (Jakobsson Larsson et al., 2016; Tramonti, Bongioanni, Fanciullacci & Rossi, 2012).

A quantitative study by Jakobsson Larsson et al. (2016) seeking to gauge the difference between initial and later-term coping strategies revealed that patients with ALS use strategies "such as support, positive action, independence, [and] positive thinking...to a greater extent compared to strategies such as avoidance/venting, information seeking" (p. 238); and that these

strategies often do not change over time. Data were collected with the Motor Neuron Disease Coping Scale (MNDCS) among other measures of affect and functionality. The MNDCS is A 22- item questionnaire that is specific to MND, and is used to determine the extent to which common coping strategies are individually relevant (Jakobsson Larsson et al., 2016; Lee et al., 2001). Data collected using this, and similar scales, present a solid basis for understanding the generalized relevance of certain coping strategies, but are unable to offer concrete explanations of why patients adopt certain coping strategies and what those strategies mean to them, relative to their well-being (Jakobsson Larsson et al., 2016; Lee et al., 2001; Montel et al., 2012a; Montel et al., 2012b).

Hogden et al. (2012a) offer a rich qualitative description of several factors that often influence patients' participation in decision-making related to specialized multidisciplinary care processes. They identified six themes within three major categories representing structural, interactional and personal influencing factors. Within those three categories, the themes related to supportiveness in the decision-making environment, and patients' relations to their diagnosis; patients' responses to deterioration, their level of engagement with their care team, their outlook on life, their perceptions of control, as well as their attitudes towards planning for the future. Hogden et al. (2012a) emphasize denial and resilience as two common approaches to coping, and note denial as a significant hindrance to engagement in learning and decision-making for persons with ALS. But they also remark that even under optimal conditions, some patients still struggled to engage in the decision-making process. This suggests gaps in our understanding of patients' use of coping strategies, specifically in relation to their perceived function, utility, and motivating factors.

Other qualitative works have also lent themselves to informing the development of patient-centered engagement tools, understanding individual patient narratives, and interpreting patients' change and adaptation patterns (Hogden et al., 2012b; Hogden, Greenfield, Caga & Cai, 2016; Brown & Addington-Hall, 2008; King, Duke & O'Connor, 2009). Such studies have paved the way for current qualitative research geared towards making practical improvements within our system. Further exploration of patient coping-related experiences that might inform proactive approaches to patient engagement and coping support could ultimately offer a different conceptualization of what coping looks like on an individual basis, and foster resiliency in such a way that quality-of-life is regarded above all else (Hogden et al., 2012a; Hogden et al., 2012b).

2.3.9 The Importance of Successful Coping

The harsh nature and volatility of ALS poses a number of challenges for various stakeholders including the need to maximize time and efficiency in determining patients' care needs, and mobilize the appropriate resources. This is especially true when it comes to caring for older adults with the condition, as they face additional layers of complexity associated with comorbidity, increased susceptibility to frailty, and unique psychosocial issues (Mitsumoto, 2009; Logroscino, et al., 2015; Miller et al., 2014, Andersen et al., 2016; Hardiman, Van Den Berg & Kiernan, 2011; Paganoni et al., 2014).

Too, the rate at which older persons' with ALS lose their ability to communicate their wants, needs, and wishes varies drastically from person to person. Some older people experience a steep decline in health resulting in a reduced physical, and sometimes cognitive, capacity within a few months of being diagnosed, while others experience long periods of high functioning (Arthur et al., 2016; Miller et al., 2014; Boyd et al., 2016). This variability makes it

extremely important for patients to become engaged in decision-making and care-planning processes as early as is therapeutically feasible, in favour of patient-autonomy, satisfaction in care, and high quality-of-life (Oliver et al., 2014; Hecht et al., 2003; Pagnini et al., 2010; Hogden et al., 2012a).

Successful coping significantly affects patients' level of engagement in decision-making (Hogden et al., 2012a). Given this reality, and the importance of patients' participation in their care, it is necessary to gain further insight into the relevant piece of the coping puzzle, including diagnostic disclosure and patients' reactions to it, to be able to ultimately support engagement and the end goal of improved quality-of-life for all persons living with ALS (Hogden et al., 2012a; Manne, 2002; Graven et al., 2014).

2.3.10 Decision-making & Advance Care Planning for ALS

Patients' desire to have a greater sense of control over their healthcare at the end of life is becoming increasingly well-known (Brinkman-Stoppelenburg, Rietjens & van der Heide, 2014). Unfortunately, for persons with ALS, their ability to effectively communicate their wants and wishes can be lost very soon after they learn of their diagnosis. This leaves their family members and healthcare providers at risk of making hasty medical decisions on their behalf, without documentation of what the patient might have wanted, or not wanted, for their care under certain circumstances (Brinkman-Stoppelenburg et al., 2014; Hossler, Levi, Simmons & Green, 2011).

Advance care planning (ACP) can be considered an important and empowering tool that can aid patients in ensuring that they have complete control over their care, both present and future. Requiring deep reflection and ongoing discussion, it has been shown to help persons with terminal illnesses such as ALS not only recognize, but appreciate and voice their priorities in,

and at the end-of-life, while easing moral distress, and improving cohesion among members of the patients' circle of care (Hossler et al., 2011; Ke et al., 2017). In addition, engaging patients in ACP has been shown to reduce stress and anxiety, improve patient-experiences, as well as care outcomes (Brinkman-Stoppelenburg et al., 2015; Murray et al., 2016). Current recommendations urge providers to start ACP discussions early to maximize the process' usefulness (Andersen et al., 2012). But, often, patients with ALS do not engage in such end-of-life discussions until late into their disease trajectory, when necessity strikes (Levi et al., 2017). In fact, a significant proportion (>50%) of people are neither engaging in, nor benefitting from the process of ACP, at all (Canadian Hospice Palliative Care Association (CHPCA), 2008; Levi et al., 2017). Fewer are going on to complete their advance directives (30%) to have the outcomes of the ACP discussion documented for the record (Connolly et al., 2015; Hossler et al., 2011; Borasio et al., 2001; Murray & Butow, 2016; Levi et al., 2017; CHPCA, 2010).

Concerns about the timing of ACP discussions have also been brought up. For some patients, introducing the topic too early may be overwhelming and, in some cases, damaging; while initiating the conversation late in the disease course may rob patients of the opportunity to make important decisions for their end-of-life care (Connolly et al., 2015; Levi et al., 2017). Thus far, we do not know enough about the subject to be able to offer specific guidelines for when and how to present patients with opportunity to participate in ACP in support of optimal care outcomes (Connolly et al., 2015).

There has been speculation as to the apparent lack of participation in ACP. The majority of the reasons cited have to do with relational, communicational, and informational barriers, and patients' perception of their disease state, which are all factors in coping success (Hogden et al., 2012; Connolly et al., 2015; Seeber, Hijdra, Vermeulen & Willems, 2012; Lazarus & Folkman,

1984). Our current understanding of patient engagement in ACP emphasizes the patient-provider relationship for which the groundwork is often laid when patients learn, or receive confirmation of, their diagnosis (Hogden et al., 2012a). It is at this point that the disease and its expected course are typically explained, patients' initial needs and goals are established, and the 'tone' of care is set. As has been discussed above, poor delivery of bad news has the power to harm clinical relationships and cause patients to disengage from the care process (McCluskey et al., 2004; Baker & Graham, 2004). This suggests that the point at which the news of ALS is broken to a patient and the succeeding moments are significant ones that should to be well attended to (Miller et al., 2009; Andersen et al., 2012).

2.4 Summary & Implications

We know that ALS is a terrible and unpredictable disease that people attempt to cope with in various ways (Lee et al., 2001; Matuz et al., 2010; Jakobsson Larsson et al., 2016). But, our current understanding of the diagnosis, disclosure, and coping experiences of persons with ALS is limited, while current tools and practices aimed at supporting patients through these processes remain inadequate (Lee et al., 2001; Matuz et al., 2010; McCluskey et al., 2004). Thus, we require a clear picture of these experiences in order to establish a path towards improving the effectiveness of current tools and practices in empathetic disclosure and coping support. Such advancements in our understanding are likely to support not only improvements in patients' overall satisfaction with their care in diagnosis, disclosure, and coping support, but also important developments in other aspects of care, such as communication, planning, and decision-making, towards better quality-of-life for persons living with ALS (McCluskey et al., 2004; Shaw et al., 2013; O'Brien et al., 2011).

As improving quality-of-life has become key areas of focus in the literature, the current project, which aimed to gain insight into key pieces of the quality-of-life puzzle, is a timely one (Pagnini, 2018; Miller et al., 2009; Murray & Butow, 2016; Miller et al., 2014). Additionally, as the number of people diagnosed with ALS continues to increase, the need for a clear understanding of the care needs of people living with ALS will become greater. As such, it continues to be important to push for significant advancement, so that the anticipated care improvements may be available to fulfill the increasing demand and to mitigate suffering.

Chapter 3: Study Rationale & Objectives

3.1 Objectives & Study Rationale

The current study sought to explore the interconnectedness and interdependence of the diagnosis, disclosure, and coping experiences of persons living with ALS through their eyes and through the eyes of their caregivers. Underlying relational, communicational, informational, and psychosocial forces that occur within the population of persons living with ALS, amid their hearing, conceptualizing and taking charge of their disease was a focus; as was participants' knowledge about perceived shortcomings of current approaches in disclosure and coping support, and their preferences for related care support.

Specifically, the current study aimed to answer the following research questions:

- 1) What are the experiences of adults living with ALS surrounding the receipt of their diagnosis?
- 2) What are the experiences of adults living with ALS in coping and coping support?
- 3) What experiences do adults with ALS desire when it comes to their coping support?

Previous studies have sought to identify facilitators and barriers to engagement, understand the emotional needs of persons with ALS in diagnostic disclosure, and to conceptualize the psychosocial realities of sufferers (Hogden et al., 2012a, Hogden et al., 2012b, Pavey et al., 2013; McCluskey, 2004; King et al., 2009; Foley, O'Mahony & Hardiman, 2007; Miller et al., 2009; Andersen et al., 2011). However, few so far have looked into the nature of these phenomena in adults with ALS. Even fewer have aimed to understand the coping

experiences of members of this population, in relation to their experiences in receiving confirmation of their diagnosis, and in coping during this process and thereafter (Montel et al., 2012a; Hogden et al., 2012a; Hogden et al., 2012b; Miller et al., 2009; Andersen et al., 2012). With a growing older adult ALS patient population and no known cure, it is thus necessary to do the work to understand and fill the knowledge gaps (Arthur et al., 2016).

Chapter 4: Methods & Research Design

4.1 Qualitative Methods & Research Design

As discussed above, we currently know very little about adults with ALS' experiences in diagnosis and disclosure, nor do we have a clear understanding of their experiences in discussing, or participating in, coping, let alone how these processes are interconnected or interdependent (ALS Canada, 2016; Murray & Butow, 2016; Oliver et al., 2014; Hogden et al., 2012a; Hogden et al., 2012b). This limited understanding hinders researchers and clinicians' ability to improve the process for the population, as a clear picture of the major practical gaps is first required.

Qualitative approaches to health service research have proven to be particularly useful in the "early stages of inquiry" (Sofaer, 1999, p. 1102). Such methods have the power to uncover patterns and themes that can be used to build meaningful descriptions of the complex and dynamic realities of study participants. In their nature, they are best suited for research inquiries that aim to gain a deep understanding of the particular phenomena, rather than quantify their facets (Sofaer, 1999). It is for this reason that a qualitative research design for data collection and analysis was chosen to address the aforementioned research questions.

Applying a qualitative methods to answer the study's research questions (outlined below) allowed for the capturing of participants' shared experiences in diagnosis, disclosure and in coping, which will serve as a basis for future exploration and potential quantitative investigation. Additionally, the obtained qualitative data are suitable for interpretation, from which patterns and themes could be deduced, toward broadening our knowledge for the sake of continued practice, program and policy development (Crabtree & Miller, 1999; Creswell, 2007; Sofaer, 1999).

4.1.1 Recruitment

A total of 18 participants (See section 4.2.4 for Sample Summaries) were recruited through the ALS Society of Canada. Initially, the ALS Society's Regional Manager for Ontario Central-West/ Waterloo Wellington served as a gatekeeper, and, thereby, mediated contact with patients and caregivers in Ontario.

No participants were excluded based on their gender, socio-economic status, race, ethnicity or religious affiliation. Patients diagnosed with the various ALS types and MND sub-types, as well as caregivers, were included in the sample in order to facilitate data triangulation. Eligibility of caregivers to participate in the study was based on self-report of their involvement in the care of someone diagnosed with ALS, whether they were a partner, family member, or friend. No specific level of caregiver involvement was required.

Copies of the recruitment flyer (See Appendix A) were provided. Upon receiving participants' contact information, the researcher contacted potential interviewees via either telephone or email to explain the objectives of the study and the study procedures. The researcher then set up a date and time to meet with interested participants on an individual basis to provide them with more information about the study, including what they would be asked to do if they decided to participate. An information letter was provided at this time (See Appendix B). Persons who agreed to participate in the study were given the opportunity to either proceed with the hour long interview at that time, to sign up for another interview time, or complete a participant contact form (See Appendix C) so that the researcher could communicate with the potential participant by email or phone, in the event that they needed more time to think about their participation. Participants who completed a contact form were contacted, thereafter, using a recruitment script (See Appendix D). In order to be eligible for participation, participants were

required to sign a consent form (See Appendix E) before participation. Participants received a feedback letter (See appendix F) following their participation.

4.1.2 Interview Guide Development

Semi-structured interviews consisting of a series of open-ended questions are commonly used for data collection in qualitative research (Creswell, 2007; Aronson, 1995; Kuehl & Newfield, 1991). This type of approach in interviewing gives participants the opportunity to speak freely to their lived experiences (Creswell, 2007; Webb & Kevern, 2001). For this reason, the current study used semi-structured interviews to capture participants' thoughts, perceptions, and experiences in diagnosis, disclosure, and coping, and to answer the indicated research questions.

Spradley (1979) suggests the importance of asking open-ended questions regarding the 'descriptions', 'culture' and 'contrasts' within the experiences of interest. It was on this basis, and in consulting the relevant literature, that the study's interview guides were developed. In addition, strategies for collecting rich data were employed based on the strategies for qualitative research outlined by Ritchie & Lewis (2003) and common practices in interviewing detailed by Marshall & Rossman (1995) and Seidman (1991) that aided the researcher in helping participants feel more comfortable in the interview scenario, thereby enabling them to share their rich experiences with greater ease. These strategies included the incorporation of questions that facilitated rapport building and conversation closure at the beginning and end of an interview, respectively, as well as procedures in active listening.

In October 2017, a group of nine focus group participants consisting of patients ($n = 3$) and caregivers ($n = 6$), including one professional caregiver, one past-caregiver (patient passed

away), were recruited through one of the ALS Society of Canada's regional support groups to gather participants' perspectives in a group context, and also to identify if refinements might be necessary to the interview guides, one with questions tailored for use with patients, and one tailored for use with caregiver participants (See Appendix G & H).

The focus group interview was then discussed with the researcher's supervisor and some modifications were made to the interview guide for clarity, and to help the interview flow better.

Finally, the study's interview guides were circulated among committee members with expertise in qualitative research to verify and validate the established questions and question probes, to ensure that the questions posed would be able to efficiently garner relevant and rich data for analysis. The committee's feedback was used to develop the final versions of the study's interview guides (See Appendix I & J).

4.1.3 Data Collection

Data were collected using a participant demographics form (See Appendix K) and the tailored interview guides, as described above (See Appendix G-J). The demographic form allowed for the collection of contextual information regarding the participants, while the interview guides facilitated the exploration of each participant's realities, and what they believed about their experiences of the processes of diagnosis and disclosure, and in coping. The interview questions were posed either in person or over the phone, after receiving participants' consent. Individual and dyad interviews were conducted based on the indicated preferences of the participants. Interviews were audio-recorded and transcribed verbatim (Creswell, 2007; Spradley, 1979; Aronson, 1995).

Throughout the research process, the researcher's thoughts and ideas were recorded as field notes. This was done to stimulate ongoing information processing, and to encourage reflexivity, thereby mitigating 'contamination' of participant responses (Webb & Kevern, 2001). The collection of data from multiple data sources, including through general and side-bar conversation, served to enhance the richness of the data collected in the interview process (Creswell, 2007; Wimpenny & Grass, 2000).

4.1.4 Sample & Setting

The current study involved the participation of 18 participants, conforming to Creswell's (2007) recommendation for a minimum sample size of 10 participants, to achieve a solid understanding of participants' shared experiences, or saturation and credibility in a qualitative study. Creswell (2007) also suggests some variability based on the nature and scope of the study, and the richness and thickness of the collected data. Saturation was not reached in the current study after the 10 individual and dyad interviews were conducted. In pursuit of saturation, the focus group data were also included in the analysis, acknowledging potential limitations (See section on limitations).

Given the challenges of sifting through data from multiple perspectives (i.e. patients and caregivers) in attempting to understand an individual perspective, a higher sample size was believed to be necessary to reach saturation. However, saturation was still not reached after 18 participants had been interviewed, meaning that new ideas were still being brought up during the last interview (Creswell, 2007; Fusch & Ness, 2015).

Only adult persons with ALS and caregivers for adult persons living with ALS participated in the study. All participants garnered their experiences in the context of the

Canadian healthcare system (namely in southwestern Ontario). The majority of participants with ALS or similar were community dwelling ($n = 7$), while two lived in hospital ($n = 2$). Some had regular, in-home, ongoing additional personal PSW support to varying degrees ($n = 3$), while others were solely cared for by their caregivers and/or family members and friends ($n = 4$).

Though the median age for symptom onset is about 62 years, it can range from 40-70 years (Hardiman et al., 2011; Wolfson et al., 2009; Arthur et al., 2016). It is for this reason that the study sought to collect data from and about adults (both male and female) between the indicated age range and beyond, rather than restrict eligibility to at least 62 years, or the standard age of 65. Extending the age to include those between their 30th and 80th years, allowed for the opportunity to hear a broader range of experiences.

The thoughts and perceptions of caregivers regarding the experiences of patient participants were also captured. The reason for seeking both perspectives (patients and caregivers) was to further broaden the sample. Patients' and caregivers' experiences were expected to complement one another, and offer a platform for data triangulation (Creswell, 2007; Bryant & Charmaz, 2007). The incorporation of various media (i.e. over-the-phone or in-person) and formats (group, dyad, individual) of data collection in the current study was also intended to facilitate data triangulation (Creswell, 2007; Marshall, Cardon, Poddar & Fontenot, 2013).

Thus, the study sample consisted of persons with ALS ($n = 9$), family caregivers ($n = 7$), a professional caregiver ($n = 1$), and one past caregiver ($n = 1$). The majority of the participants with ALS or similar were male ($n = 5$), while the majority of the caregiver participants (including past and professional) identified as female ($n = 7$). Participants reported diagnoses of various types and MND sub-types: probable/ atypical ALS ($n = 2$), confirmed ALS ($n = 6$), confirmed PLS ($n = 1$), confirmed ALS – slow progressing ($n = 3$), with some overlap between

categories, where participants were given more than one related diagnosis due to initial misdiagnoses and/or multiple opinions from different healthcare providers. These diagnoses were reported to have occurred between the years of 2010 to 2017, many of which were made in 2016 (n = 4). All participants described a path to diagnosis that began with either a visit to a family doctor or walk-in clinic physician upon symptom onset, followed by a referral to an ALS clinic for specialized diagnostics and care (clinics primarily in London, ON, Hamilton, ON, and Toronto, ON). ALS diagnoses were made by specialized neurologists. A minority of participants were directly referred to a neurologist by another type of specialist (n = 2).

Some participants described significant speech impairments (n = 3), one participant relied on eye controlled assistive technology to communicate. Some participants disclosed significant upper limb (n = 8) and lower limb (n = 9) physical impairment, requiring the use of mobility aids (e.g. walkers, wheelchairs, canes, leg braces, etc.) and/or home renovations/ accommodations/installations (e.g. ramps, lifts, widened doorways, bathroom modifications, relocation, etc.). All participants with ALS indicated significant general and progressing physical impairment, some of which whose disease had progressed to the point of ‘bed-bound’ status (n = 2).

Data were collected from the study’s participants in a focus group interview (n = 9), in individual interviews (n = 5), and in dyad interviews (n = (2 x 4) 8). Some of the participants from the focus group opted to participate in individual/ dyad interviews as well (n = 4). One of the participants who participated in both the focus group interview and a dyad/individual interview moved to a different city within southern Ontario prior to completing the dyad/individual interview. All participants lived in southern Ontario, excepting one who lived in British Columbia. Some participants described rural experiences (n = 3). The focus group

interview took place on October 12th, 2017. The Individual and dyad interviews were conducted between April 5th, 2018 and June 20th, 2018.

The average age of participants with ALS was 55 years, ranging between approximately 30 and 70 years (rounded for anonymity). The average age of all caregiver participants was 43.75 years, ranging between approximately 25 and 60 years (rounded for anonymity). The average age of caregivers, as stated, represents all types of caregiver participants in the sample excepting one family caregiver who declined to complete the demographics form. See Tables 2 and 3 for detailed sample summaries; summaries edited for confidentiality.

Table 2: Participant Sample Summary (Focus Group Interview)

n =	Gender	Role/ Relationship	~Age	Locale	Interview Format	Approximate Date/ Nature of Diagnosis
1	F	Caregiver – Professional	40	Large Urban City (Greater Toronto Area), ON	Group	
2	F	Past Caregiver – Daughter	45	Small Town (Waterloo Region), ON	Group	
3	F	PWPLS	65	Small Town (Waterloo Region), ON	Group	2017 – Confirmed/ PLS
4	F	Caregiver – Daughter	35	Small Town (Waterloo Region), ON	Group	
5	F	Caregiver – Sister	DTA	DTA	Group	
6	F	PWALS	45	Medium City (London Metro Area), ON	Group	September 2015 – Confirmed/ Slow progressing
7	M	Caregiver – Boyfriend	45	Large Urban City (Greater Toronto Area), ON	Group	
8	M	PWALS	55	Large Urban City (Kitchener Metro Area), ON	Group	Spring 2016 – Confirmed/ Progressed ALS
9	F	Caregiver – Cousin	50	Large Urban City (Kitchener Metro Area), ON	Group	

Table 3: Participant Sample Summary (Individual and Dyad Interviews)

n =	Gender	Role/ Relationship	~Age	Locale	Interview Format	Approximate Date/ Nature of Diagnosis
1	M	PWALS	65	Large Urban City (Kitchener Metro Area), ON	Individual	October 2016 – Probable/ Atypical
2	M	PWALS	30	Large Urban City (Greater Vancouver Area), BC	Individual	May 2016 – Confirmed/ Limb onset
3	F	PWALS	55	Large Urban City (Kitchener Metro Area), ON	Dyad	2015/2016 – Confirmed
4	M	Caregiver – Son	25	Large Urban City (Kitchener Metro Area), ON	Dyad	
5	M	PWALS	60	Large Urban City (Greater Toronto Area), ON	Individual	2015 – Confirmed/ Atypical; Lower MND Variant
6	M	PWALS	50	Large Urban City (Kitchener Metro Area), ON	Dyad	Spring 2010 – Confirmed/ Slow progressing
7	F	Caregiver – Wife	50	Large Urban City (Kitchener Metro Area), ON	Dyad	
8	F	Caregiver – Wife	60	Large Urban City (Kitchener Metro Area), ON	Individual	
9	F	PWALS	70	Large Urban City (Kitchener Metro Area), ON	Individual	2010 – Confirmed/ Slow progressing
10	F	PWALS	45	Medium City (London Metro Area), ON	Dyad	September 2015 – Confirmed/ Slow progressing
11	M	Caregiver – Boyfriend	45	Large Urban City (Greater Toronto Area), ON	Dyad	
12	M	PWALS	55	Large Urban City (Kitchener Metro Area), ON	Dyad	Spring 2016 – Confirmed/ Progressed ALS
13	F	Caregiver – Cousin	55	Large Urban City (Kitchener Metro Area), ON	Dyad	

4.1.5 Thematic Analysis

The current research project employed methods of thematic analysis as outlined by Braun & Clarke (2006) in processing the qualitative data. Kitzinger & Barbour (1999) describe this process as: “Drawing together and comparing discussion of similar themes and examining how these related to the variation between individuals and between groups” (p.16). Specific measures taken based on Braun & Clarke’s (2006) refined guidelines for data analysis are outlined in sections below.

According to Braun & Clarke (2014), “the field of health and wellbeing scholarship has a strong tradition of qualitative research—and rightly so” (p. 1). Specifically, they highlight the value of thematic analysis as an approach to qualitative inquiry, presenting it as ‘theoretically flexible’, existing within both the essentialist and the constructionist paradigms (Braun & Clarke, 2014; Braun & Clarke, 2006). This flexibility served as an advantage in the context of the current research project, as it facilitated the analysis of the similarities and differences among the patient and caregiver perspectives, and enabled the process of deriving meaning from their differing and shared experiences. It also supported the emergence of unexpected patterns and themes that might serve as important and beneficial insights in the limited body of ALS care-related literature (Braun & Clarke, 2006).

The thematic analysis conducted for the purposes of this project aimed to understand the experiences of persons with ALS through the eyes of patients and their caregivers, and took place under epistemological assumptions aligned with the constructionist paradigm. Under this system of belief, themes were generated through an analysis of participant-described events and incidents, with the understanding that people build their own realities as they navigate through and create meaning from the world around them. The constructionist paradigm recognizes and

embraces the complexity of human life, which provided a suitable basis for understanding people in their attempt to cope with a terminal illness; ‘what they do’ and ‘how they do it’ (Braun & Clarke, 2006; Charmaz, 2008. The data analysis process occurred inductively, meaning that the researcher did not attempt to fit the data into an existing framework or hypothesis, but rather let the data ‘tell the story’ (Braun & Clarke, 2006).

The data from recordings were transcribed verbatim and analyzed for significant statements and meanings, phrasings and mechanics using NVivo 11 software (Creswell, 2007). The Braun & Clarke (2006) guided thematic analysis of the acquired data took place across the following phases:

- 1) **Reading of the transcripts & identifying significant patterns:** the researcher became familiar with the data by reading through each of the verbatim transcripts beginning to end. Electronic copies of the transcripts were accessed from a secured computer folder and read on screen. The transcripts were read through twice. During the second read-through, attention was paid to relevant and significant patterns that emerged.
- 2) **Documenting relevant & significant patterns:** the relevant and significant patterns identified in the first phase of the analysis were documented by way of a reduction of the data into codes and categories. The electronic copies of the transcripts were uploaded into the NVivo 11 software, which was used to facilitate the generation of these codes and categories. The lists of categorized codes were then compiled into one comprehensive document.

- 3) **Theming & describing codes & categories facilitated by generated meaning:** the comprehensive document of codes and categories compiled in phase two was printed out and pulled apart so that the codes and categories could be manipulated and combined, then labelled, with the benefit of having a visual representation. The codes and categories were laid out on a flat surface to facilitate this process. The manipulation of these codes and categories yielded overarching themes that accurately represented the data. Relevant and exhaustive descriptions of the labeled themes were then drafted, including explanations of inconsistencies and ‘fit’.
- 4) **Revision and validation of themes and descriptions:** the themes and description formulated in phase three were reviewed, and their accuracy in reflecting the essence of the data and the indicated epistemological paradigm was assessed across several read-throughs. The original electronic copies of the interview transcripts were then revisited to resolve the described inconsistencies and issues of ‘ill-fit’. Descriptions were adapted accordingly.
- 5) **Defining the established themes:** the established themes and exhaustive descriptions (including extracted relevant quotations] were used to formulate definitions that clearly represented each aspect of the experiences being explored. These definitions were written up in a succinct document to be presented to study participants for the purposes of member-checking.
- 6) **Isolation of significant themes and member check:** in this final phase, the most significant themes were isolated, then the study’s participants were consulted as part of

the member-checking process. All participants were presented with the isolated themes, and asked to confirm or refute the findings.

The established themes and validated findings were used to offer a rich description of participants' lived experiences through diagnosis, disclosure, coping, and preference for coping support.

4.2 Enhancing Qualitative Rigour

'Rigour' pertains to the validity, reliability, and objectivity of research as key features of trustworthy academic work.

For qualitative research, reflexivity, as it relates to responsibility and honesty, is also vital (Baxter & Eyles, 1997; Lincoln & Guba, 1985). The criteria for qualitative rigour, according to Baxter & Eyles (1997), are as seen in Figure 1. Those criteria are: *credibility*, *transferability*, *dependability* and *confirmability*.

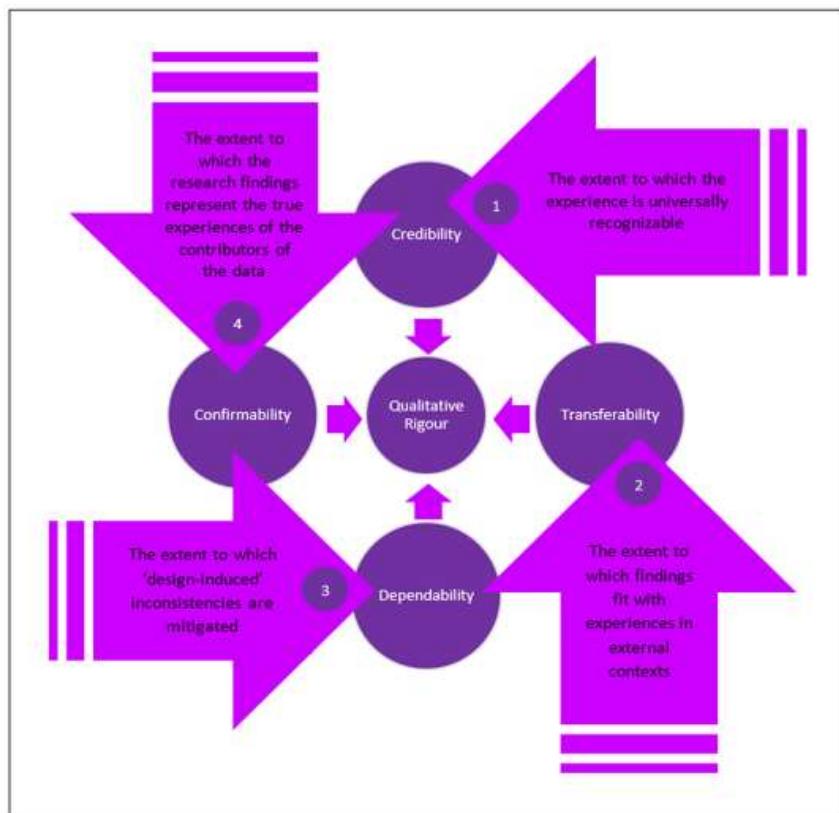


Figure 1: Criteria for Qualitative Rigour (adapted from Baxter & Eyles, 1997)

Baxter & Eyles (1997) state that *credibility*, the central tenet of rigorous qualitative research, is the extent to which an experience is universally recognizable by way of key themes

that emerge in its reduction or simplification. *Transferability* on the other hand, “refers to the degree to which findings fit within contexts outside the study” (Baxter & Eyles, 1997, p. 515).

Dependability has to do with the degree to which consistency in the study design is matched or maintained to ensure that no ‘design-induced’ inconsistencies arise. Dependability is closely linked to credibility. *Confirmability* relates to objectivity, in that it is the extent to which the research findings represent the true experiences of the contributors of the data and not the perspectives or biases of the researcher (Baxter & Eyles, 1997).

Baxter & Eyles (1997) provide useful suggestions to aid researchers in achieving the aforementioned criteria. Based on these suggestions, the current study employed the following techniques to enhance the qualitative rigour of the study (See Table 4).

Table 4: Enhancing Qualitative Rigour

Criterion	Credibility	Transferability	Dependability	Confirmability
Technique(s)	Member-checking to verify and validate collected data and research findings	Interviewing participants across various care settings	Audio-recording data	Reflecting on perceptions, preconceptions, beliefs, and values to account for and separate personal biases from the data
		Thick descriptions	Verbatim transcriptions	Process notes/ note-keeping
	Triangulation with the literature, across various mediums of data collection and different participants		Debriefing/ investigator triangulation	Member-checking

4.3 Ethical Considerations

There is always potential for ethical issues to arise during the research process. Therefore, it was necessary to have considered, and to continue to consider, possible issues to safeguard against undue harm, and to ensure, as much as feasibly possible, that the values and rights of participants were/are respected and protected (Creswell, 2007; Creswell & Poth, 2017).

In the case of the current study, the participants' physical, psychosocial, and cognitive capacities were of particular relevance. This study recruited participants of, but not limited to, a vulnerable population, who experience various forms of frailty. For this reason, it was imperative that the researcher consider their potential physical, psychological, and/or cognitive limitations that, at times, required close attention, such as when attempting to gain informed consent (ALS Canada, 2016; Murray & Butow, 2016). The following is a summary of the ethical issues and implications relevant to the current study:

There were no anticipated risks associated with this study, as was recognized by the University of Waterloo's Office of Research Ethics (See Appendix L). Participation was completely voluntary and participants were able to withdraw from the study at any time. In addition, participants were given the option to refuse to answer questions at any point during the study. Though, all participants who began the interview process answered every question without indicated concern. One participant withdrew from the study after having initially agreed to participate before signing the consent form. The participant asserted self-declared ineligibility as their reason for opting out of participation. The participant was thanked for their interest in the study and reminded that there would be no consequences or 'hard feelings' for rescinding their agreement to participate. Another participant agreed to participate in the focus group interview, but refused to complete the demographics form. This participant was thanked for their

participation and also reminded that there would be no consequences or ‘hard feelings’ for opting not to complete the form.

All participants were required to sign the consent form prior to participation in order to be eligible to participate in the interview process. The consent form was verified and approved by the Office of Research Ethics, and was carefully reviewed with each participant prior to participation. Time was allotted for questions pertaining to the study as needed.

In consideration of participants’ capacity to provide informed consent, the consent forms were amended to allow an opportunity for proxy consent. Proxy consent was required for patient participants in advanced stages of their disease. In these cases, the patient participants were asked to assent (as they were able) to have their caregiver or family member offer written consent for their participation in the study on their behalf. Proxy consent was obtained from three participants. Every effort was made to gain consent from the individual themselves, beginning with an initial conversation aimed at gauging the participants sense of the study’s aims, and their appreciation for the risks and benefits of their participation in the study. When it was observed or indicated that the individual could not physically provide their own written consent, proxy consent was considered. No participants indicated or demonstrated a level of cognitive capacity that might have made it difficult for them to provide consent.

Participants’ confidentiality was respected throughout the research process. In this regard, no personal or identifying information was attached to the participants' responses. Each participant was assigned an identification/ reference number, which was used to organize the data. Only the participants’ identification/ reference number can be seen on the transcripts. Completed demographics forms were and will continued to be kept separate from interview transcripts. All data and participant information was, and will continue to be, stored in a secured

and locked cabinet located in an office in the Lyle Hallman Building at the University of Waterloo. Electronic files containing study data was, and will continue to be, password-protected. These files will be destroyed after five years from the date of study completion. Audio-tapes, transcripts, and data files generated in analysis will remain anonymous. Participants have been, and will be, identified only by their identification/ reference number or role (e.g. patient or caregiver) in the current report and in prospective manuscripts for publication, respectively. Participants have not been, and will not be, named while explaining the study results, or associated with any cited quotations

Confidentiality, as it pertains to participants' identity, was, and will continue to be, respected to the fullest extent possible by law. This means that, during the study, it was acknowledged that in the event that a participant disclosed that they were at risk of harming themselves or someone else, or that a child or elder was being harmed or neglected, confidentiality would have had to be breached, and the appropriate authorities would have had to be notified. No such events transpired during the course of the study. Moving forward, there are no conditions under which the confidentiality regarding identity cannot be guaranteed, other than those specified by local legislation, as per the above, if previously unheeded information should arise.

Only the research team had, and will continue to have access to the data collected, and all contributing members had to, and will have to sign an agreement to maintain confidentiality, otherwise known as a declaration of non-disclosure (See Appendix M). If, for any reason, a re-analysis of the data should take place a declaration of non-disclosure form will have to be signed by new members of the research team.

4.3.1 Statement of Positionality

A challenging aspect of qualitative research is that it requires researchers to get very close to study participants and collected data without significant influence from their individual opinions and experiences. But, some level of bias may come into play. If not carefully attended to, the researcher's preconceived notions might serve as a barrier to research integrity. To mitigate bias and subjectivity, the researcher minimized the figurative, and in some cases, physical distance between herself the study's participants. She also made a conscious effort to focus on immersing herself into the experiences being described by those being researched, rather than their lives as they exists at the personal level. This was complemented by diligent practices in note-writing and reflexivity throughout the study (Creswell, 2007, Charmaz, 2006).

In certain cases it is necessary for the researcher to offer a complete disclosure of their understanding and experiences with the research topic and/ or target population. This involves a statement of positionality that details their relevant social, personal, political, and professional views. This facilitates reflexivity in the researcher, and informs potential readers of the lens through which data were collected, analyzed and interpreted (Creswell, 2007, Charmaz, 2006). Therefore, given the researchers own lived experience and motivations for conducting the current study, and in order to uphold the values of honesty and integrity to the utmost standard as it is relevant to the conducted research endeavors, the following statement of positionality was offered prior to the study's commencement:

"My interest in diagnosis, disclosure and coping research for persons living with ALS are a direct reflection of my personal experiences supporting a member of my family through their disease in care. Over the past two years, I have played a role in supporting this individual both in the home and in institution-based care. This experience has, in part, shaped my

understanding of the ALS experience. My aim with the current study was to further explore the experiences of persons with ALS in care, specifically, with regard to diagnosis, disclosure and coping. I acknowledge that I, with my worldview, shaped by my experiences with the subject matter prior to beginning the study, acted as primary vessel for data collection and analysis. I also acknowledge that this means that despite practicing critical self-awareness while maintaining full-disclosure of my assumptions and biases throughout the research process, the results of the analysis conducted may reveal remnants of my preconceptions.

Chapter 5: Findings

5.1 Major Findings

Table 5 below provides a summary of the main themes that categorize each of the processes of interest: diagnosis, diagnostic disclosure, and coping. As specified, these themes were established through a thematic analysis conducted using the acquired interview data. See Appendix N & O for depictions of the codes, nodes and thematic maps generated in this analysis.

The data revealed six major themes that specifically related to the ALS diagnosis process, eight themes relating to the diagnostic disclosure process, and eight themes pertaining to the coping process. Three major themes were established with regards to PWALS/ PLS' desired experiences for support in their coping. All 25 themes are intended to represent the experiences of persons with ALS/ PLS, based on the thoughts expressed and experiences described by both persons with ALS/ PLS and caregivers.

Table 5: Summary of Major Category Themes

Experience Category & Corresponding Themes		
Diagnosis	Diagnostic Disclosure	Coping
Initiated by changes in functional capacity or unusual sensations	Verbal in-person report of conclusion	Rationalizing and reasoning
Theory generating/ Self-diagnosis of treatable condition	Confirmation of suspected diagnosis	Desire to maintain independence as long as possible
Trial and error	Concrete diagnosis as pivotal	Ongoing transition period
Process of elimination	Care team convening	Making peace
Symptom management	Access granting	Keeping mind occupied
Referrals to specialist and/or clinic	Willpower initiating	Maintenance of normalcy
	Information seeking	Social network involvement
	Limited information provision	Care team relationship development
Support Preferences		
Hope stimulating conversations and activities		
Ongoing information provision aimed at curbing uncertainty		
Independence, autonomy, lifestyle and normalcy supported throughout disease course		

5.1.1 Major Themes Related to the Diagnosis Process

Participants disclosed that their path through diagnosis was initiated by changes in functional capacity or unusual sensations. For example, one participant with ALS said:

“Before [I was diagnosed] it was about my blood sugar...The family doctor...was sending me for measurements of my blood sugar...one day...I was trying to do push-ups...and I couldn’t do it. I couldn’t do it anymore...so, that was when I went to [my family doctor]...” — [PWALSI-1]

Participants described a series of events triggered by their initial symptoms that involved a visit to either their family doctor or a doctor at a walk-in clinic. It was indicated that this visit then instigated initial tests ordered by the doctor, and processes of theory generating and self-diagnosis. These processes are demonstrated in the following statement made by a participant with ALS:

“...I started to feel like I was being electrocuted in my feet and legs... it just went from my toes to my knees. And then as time continued to pass...[it was] moving up my body... I pushed the doctor... to give me a possibility of what it could be because I was getting a little bit frustrated. And...my wife and I had been reading tens of dozens of articles on the Internet; and gone back and forth to each other trying to cross-reference my symptoms...we came up with all sorts of things...” — [PWALSI-5]

Both patients and doctors seemed to have been speculating as to what might be causing the symptoms, as illustrated by the statement above, as well as the following remarks made by a PWALS and a caregiver, respectively:

“I was a...typical, healthy guy and noticed some atrophy in my left hand. Specifically in the muscles that control the thumb...I was a cliché quote, unquote, muscly kind of dude...I assumed that I pinched a nerve in my arm while exercising or something like that...” — [PWALSI-2]

“...initially, the degree of weakness was so mild...that's why I thought that it would be something else...in terms of...weight loss...they were just thinking his nutritional status was not very good...[that] he wasn't getting enough protein...Then they [thought]...maybe other conditions also cause weight loss symptoms...” — [CAR1-8]

The conversations that were had surrounding the first symptoms presented to participants' primary care physicians (or first points of contact) involved a lot of back and forth in testing, and referral to various specialists, otherwise characterized as a lengthy process in trial and error. As one participant with ALS indicated: "...*it was a bit of a revolving doors...of characters who sort of confirmed my diagnosis...people in and out...*" [PWALSI-2]. In some cases, misdiagnosis and ineffective surgery or treatment aimed at symptom management were also experienced. One participant with ALS said: "[*It was] all just sort of symptom management while we were trying to figure out what the problem was and once targeted we could treat it...hopefully*" [PWALSI-5].

. While others stated:

"...my hands started getting weak. I could barely turn the keys to unlock my car. So, I went to the doctor and they said, somethings not right... And then...[the doctor] said: "I think you have carpal tunnel". So they did the surgery on both hands and that didn't really help...Then that doctor said it must be something different..." — [PWALSI-9]

"I was a PSW and I fell in the bathtub...it was drop foot...and I knew something wasn't right...When I fell in that bathtub, I was fine. I could run and jump until I fell in that bathtub...I kept working...and then I went to [the doctor]...and I was getting acupuncture. For many years I kept going and trying different thing all the time..." — [PWALSD-3]

The initial back and forth was ultimately followed by a determination, through a process of elimination, that the presented symptoms were most likely neurological in nature. This is said to have prompted final referrals made either to first, a local neurology specialist, or directly to a major city clinic neurologist for similar assessments and observations of symptom progression. One participant with ALS who asserts: "...*basically they rule everything out, and then as you continue to get worse, they say: "Hey you got ALS"*" [PWALSI-2], attests to this. Two other participants explain similar experiences:

"...it's not an obvious thing like you're going to break a leg, where you go into any ER and they put a cast on you, and you're out the door type of thing. When it is a diagnosis of exclusion and you don't know what it is...you got to figure out what it is or what it isn't" — [PWALSI-5]

“...they try to rule out everything else. Then they say you probably have ALS. But, [they] refer [me] to a team in [a major city]...and they basically did the same tests, and said the same thing: that I probably have ALS. Then they said about nine months later they would follow-up...and then they said: “okay, you have ALS”. — [PWALSD-6]

This led some directly to acceptance, while others sought a second opinion first:

“...I went to [the clinic]...I [told] them, I don’t know what I have. I don’t know if I have ALS or what I have. So, I asked for a second opinion and they said I had a neurological disease. So I said, I don’t know what I have, but I know somethings not right here” — [PWALSD-3]

Overall, the process of diagnosis was described by participants as one characterized by a great deal of back and forth, frustration, and hopeful theorizing. Participants indicated putting a great deal of effort into ascertaining a label that best suited the symptoms that they were experiencing.

5.1.2 Major Themes Related to the Diagnostic Disclosure Process

According to the participants of this study, all conversations surrounding diagnosis and prognosis were conducted in-person, regardless of the healthcare provider that was relaying the information. These conversations were indicated to have been ongoing for nine months to about a year. In two cases, a confirmed diagnosis was reached after two to three years, and in one case, a confirmed diagnosis was reached after just a matter of weeks:

“...there was maybe a year...maybe nine months of activity before [I received a confirmed diagnosis]...but that’s the day essentially, that they exhausted all of treatments and were monitoring my progression. Then they essentially concluded and voiced to me that I had ALS” — [PWALSI-2]

“I think it wasn’t until [three years later] that we were really told: “Okay, at this point between three different hospitals, we have run every conceivable test...and in the absence of finding anything else that we could pin this on we have to conclude that you have lower motor neuron onset ALS” — [PWALSI-5]

The process of receiving a confirmed diagnosis was described as a procedure in confirming the suspected, such as for PWALSI-5 who continued on to say: “*...and between the first neurologist and the second I had done a lot of research, so I had already decided that's what I had. So, it wasn't a surprise*”. A similar experience was described by this PWALS who discussed the following:

“...I've always had terrible luck. So right off the bat, and given me reading the Wikipedia page and noticing the physical symptoms I sort of concluded independently...and with my pessimism...that I had ALS. But yeah, I was noticing continued muscle atrophy, continued weakness, gradual, but obvious, over those nine months. And when I finally got the word what it was, it was one of these things that I had been expecting to hear I think” — [PWALSI-2]

After receiving a confirmed diagnosis, participants described a subsequent process of convening a multidisciplinary care team. For example, one PWALS says: “*...once I got a confirmed diagnosis from the specialized ALS clinic, it's been a whole care team of a half dozen individuals that I see quite regularly*” [PWALSI-2]. Participants perceived their concrete diagnosis as pivotal, such that it sparked a will to push forward and reassess personal priorities. Like for this participant who declared: “*I can't go anywhere...I have too much to live for!*” [PWALSD-3].

Similar was true for this PWALS who indicated similar:

“So essentially what happened...from that day forward it's like: okay, well, you have three to five years or whatever math they give you....and pardon my language, but it's like: fuck that! I'm not going in the office anymore. You know what I mean? Like what's in it for me?” — [PWALSI-2]

In trying to digest the diagnostic and prognostic information that was presented to them, participants said that they took it upon themselves to educate themselves about their condition:

“So I told [my wife] that night when I got home from work. And...when we went to bed, we started looking it up on the internet. And that's how we realized that's all we had for those six weeks [until our next appointment]” — [PWALSD-6]

Participants criticised the system for long waits between appointment times, and complained that one of their most significant struggles in the diagnostic disclosure process was the limited amount of information provided about the condition they were just diagnosed with. One PWALS stated: “*...I knew what I had when I went to the first neurologist...when she told me I probably have ALS. But, I didn't even know what that was, and she wouldn't tell me*” [PWALSD-6].

While their caregiver added: “*...when you first read [the pamphlets they give you] ...it's sickening. You just read what it is...you don't know. Just to be thrown [the information], with no lead up to it is awful*” [CARD-7].

Participants discussed that having a concrete diagnosis was helpful, in that it granted them access to support from social organizations like local ALS Society of Canada chapters and Community Care Access Centres (CCAC)¹, and eligibility to participate in treatment trials and clinical studies:

“I am going to be trying that new medicine...hopefully in the next few weeks...[through the ALS clinic]...So I was waiting for the phone call today...[from the ALS clinic neurologist]...” — [PWALSD-3]

“...[we filled] out a million forms...So I'm now on the wait list for an interview for...direct funding. And I now have...through [the] LHIN; doing personal support hours 1 hour daily. Which basically is: come in make me a coffee, shower me...Cool. I'm hooked up with [the local hospice], but I haven't heard back from them yet...” — [PWALSD-10]

Generally, experiences in the process of diagnostic disclosure were described as relieving rather than shocking. Participants described having been grateful to be through the diagnosis process, such that they were then able to begin to focus on establishing next steps, and understanding care and treatment options with a consistent care team.

¹ CCACs were local Ontario home and community care service organization, now part of Ontario's Local Health Integration Networks (LHINs).

5.1.3 Major Themes Related to the Coping Process

Study participants began their coping journey after noticing initial symptoms of the disease. They began this process by rationalizing and reasoning, trying to logically draw together a sequence of events that might have led to them developing their condition. For example, one PWALS said: “*...I feel that if I hadn't fallen in that bathtub, I would have been fine today*” [PWALSD-3]. While another said: “*If...they could have stopped the wiggling in my shoulders, I wouldn't have lost the use of my arms. Neither would I have [had]the wiggling's in my thighs...*” [PWALSI-1]. This reasoning and rationalizing, eventually evolved into more existential personal reflection, such that participants were saying: “*I just still don't understand why it's me. It doesn't seem fair. I don't know, I don't understand it...it's not like we have it in our family*” [PWALSD-3]. Another participant said:

“You know I never smoked, moderate drinker, exercised regularly. We had a heart smart diet....we tried to do everything we possibly could to mitigate...and then something like this happens...” — [PWALSI-5]

Generally speaking, the coping process was characterized as an ongoing transition period in which they were constantly anticipating further physical decline, or mourning the loss of previous physical ability. As one participant eloquently articulated”

“...we sort of said: we're just going to have to continue to make it up as we go along. But...I am mourning the loss of things that I used to do, and completely take for granted. Like brushing my teeth, shaving, showering. I mean...that's when it really hits home, when you get to the point where...your most basic caring needs, that you've essentially been doing since childhood, and you've been doing it without any conscious thought whatsoever...[are] just automatic at that point. Then all of a sudden to be struggling with it. That is surely difficult to contemplate. It's very distressing. But, you've really got no choice but to suck it up. And now I've finally got to the point where I have to save my dignity. You know it's slowly being stripped away, and you have to somehow create a new normal where you are willing to accept the help of others” — [PWALSI-5]

And as articulated by the same person, this involved making peace with the circumstances, and proactively working to adapt to change; often, on a daily basis:

“...there’s no sense being frustrated...and I should know. It’s more...like mourning. So I think that in order to adapt to this...to keep my wits about me... [I] make the best of it. I don’t really have time to dwell. I have to make my peace with these increasing limitations...as quickly as possible. But as I said now it’s almost on a daily basis. I’m saying: well I’m not going to be doing that again” — [PWALSI-5]

One participant anticipated having to move in with their family as a means of coping with the ongoing transition:

“So for the time being now....I live on my own. I haven’t had to move in with family yet. That’s going to happen in the near future as I’m now getting to the stage where it’s becoming difficult and unsafe to sort of be on my own” — [PWALSI-2]

Other participants planned moves into more accommodating homes, and fretted for those that had to do home renovations:

“...we were in a two storey home, and we were starting to think: okay, this is not going to work. So then we’re starting to search for a bungalow. Then...by the grace of God this [house] was on the market. It had everything. Wide doorways, a bathroom that was accessible. But, I can’t even imagine what it would be like for the families who would have to go through the renovation process. And for someone who’s faster progressing and you try to renovate to make things comfortable for them...renovations take time. I couldn’t imagine the horror of having to deal with that” — [CARD-7]

Some participants accepted deliveries of mobility aids that they didn’t currently need:

“...*[pointing to the corner]* that chair is a lift chair. It helps you sit. It reclines and also comes forward so you can get out easier...*[But]* I don’t need yet” [PWALSI-9].

One PWALS opted to participate in counselling to facilitate the anticipated transition:

“One thing that’s going great is the society is also providing me with free counseling. They referred me to a therapist...I’ve been seeing her...once a month, for the last two years kind of thing, and bringing people with me as needed to sort of work through any friction...So that’s something that I assume will support me and can assist and continue to support me through this next transition of me having to move in with my dad” — [PWALSI-2]

Keeping one's mind occupied was also a common means of coping. Whether in hospital or living at home in the community, participants saw great value in immersing themselves in brain stimulating activities; one caregiver explained:

"When he has his TV, he likes to watch sports and news and shows. He'll listen to music on the TV...Sometimes, he'll type out things on his [eye-controlled assistive technology] machine, and save them. You know, instructions for staff, or things to tell me, things to tell the hospice fellows...It's slow in the moment, so he likes to have things prepared" — [CARD-13]

Relatedly, a PWALS had the following to say:

"So obviously I'm spending a lot more time in bed or propped up in a chair. But I still got my brain candy. I'm still working on estate planning and trying to transfer my business clients to other people..." — [PWALSI-5]

A desire to maintain independence and normalcy, as it related to their desired lifestyle, for as long as possible was often a motivating factor for participants' actions. For example, one caregiver said:

"Things have changed...we used to like camping a lot...but now, when our friends are camping, we'll go visit them for the day. We'll barbecue with them, then come back home. We won't stay over, but at least we try..." — [CARD-7]

This statement demonstrates an effort to maintain continue participating in the activities that they've previously enjoyed, as best possible.

One PWALS suggested a strong preference for independence by relay this story:

"I resisted having to use the walker or the wheelchair for three months. But, then I fell on my face too many times...literally, I fell on my face...My arms weren't strong enough to break my fall. So, when I fell forwards I landed on my face. We had to call the ambulance...to come help pick me up" — [PWALSD-6]

Another participant explained that he believed that his previous independence was something that gave him great pride; a character trait, in a sense, that he wanted to be remembered for. He indicated that it was very important to him that his friends, family and colleagues remember him as a strong, capable person. He said:

“...everybody knows me as being fit and trim, and running around like a proverbial chicken....[so] I just sort of went off the grid...I prefer that people remember me as being someone with vitality, and the picture of good health which I was. So...we're trying to sort of keep a lid on it at this point”. — [PWALSI-5]

A PWALS that participated in the focus group stated:

“I felt pitied by everybody else. People would see my arm dragging and open doors for me or ask me what happened or tell me everything will be okay—and it's like...why are they being so nice to me?” — [GRP-6]

Further, another PWALS declared: “*I just try to keep going forward every day and live my life normal*” [PWALSD-3]. This participant continued on to say:

“I used to take care of people. I loved my job. I'd rather take care of people, than have people take care of me. I used to cut the grass, I used to do the vegetable garden. I worked 78 hours every two weeks...I would come home, cook supper, [and] do dishes. Now my life's really changed. Now, I got to sit here and watch everybody else do it. It's not easy” — [PWALSD-3]

But, this participant shared that, in face of her longing for her previous independence, she maintains an active social life that brings her a great deal of happiness and excitement: “*But we're lucky because we went to Kid Rock, and we had my nephew's support there too. I've got a lot of family members*” [PWALSD-3].

Another participant with ALS, living in hospital, described similar sentiments:

“...[I like to feel connected] to the catholic community across Canada...[most days]...I [sit] here all by myself...and then...after my snack...they bundle me up and put me in the bed...that's it...[when friends and family come]... I get to talk to them. I get to talk to other people” — [PWALSI-1]

For many participants, their relationships with their healthcare providers were also a focus in their coping. One participant with ALS summarized:

“I try to form a very human and sort of like honest personal connection to my clinicians, both on the clinical side, and sort of humanistic support side. And that's what I find most valuable in terms of interacting with these people. You know, some of the bullshit aside, in dealing with, and as individuals...and people...as we go with this journey together right” — [PWALSI-2]

In addition, patients alike describe their connection to specialized community organizations as incredibly valuable in terms of their coping: For example, one caregiver said: “*...[my family member] really likes the ALS support group. So, he's really hoping to be able to get out to those groups*” [CARD-13]. A PWALS living at home stated:

“...the ALS society... [goes] above and beyond in terms of providing personal support and a 'just like a family'-type of atmosphere that I know I can reach out to...knowing and having that genuine, honest, reliable security and consistency has been valuable to me I think...[they are a tool] in my toolbox in case I need them. So that's been very helpful and comforting for me” — [PWALSI-2]

In general, the process of coping was described by participants as one that began with rationalizing and reasoning, and evolved to include mechanisms of making peace with the given circumstances, and later, a focus on engaging in activities that nurtured patients' sense of independence, autonomy and normalcy, with emphasis on maintaining their previous and/or desired lifestyle as much as possible.

5.1.4 Major Themes Related to Support Preferences

The interview data revealed three major themes related to participants' preferences for support in care and coping. Most commonly, participants expressed a desire to participate in more hope stimulating conversations and activities with their healthcare providers. Both patients

and caregivers indicated that they believed that the conversation and activities that they experienced upon receipt of the diagnosis activities tended to be hope depleting and discouraging. For example, one caregiver described the following during the focus group interview:

“I remember when my sister was diagnosed...the doctor told her, her diagnosis...I saw that she got this huge look on her face, and the doctor just said ‘there, there’. I thought that was awful... my sister said...it makes you feel like there’s no hope...for you...that’s not right...”— [GRP-5]

Likewise, a PWALS stated: “*...[the doctors] just said it was ALS...beyond that... all that they’ve said is that it’s a progressive disease and that there’s nothing they can do about it....that’s the painful fact*” [PWALSI-1].

Participants suggested gentler presentations of prognostic information, paired with conversations that revolved more around treatment or potential treatment options:

“...on the day...it would have been tremendously helpful during that very bleak and terminal [conversation]...to hear that: don’t worry, there’s a stem cell treatment just around the corner, and you’re going to be fine”— [PWALSI-2]

The professional caregiver who participated in the group describe a scenario where a tailored approach to information presentation might be beneficial:

“I had a client that was convinced it was lime disease. So eventually we had to just say okay it’s lime disease...I think if calling it lime disease makes her happy then do it. [But] that might not work for someone else...”— [GRP-1]

Other participants discussed healthcare providers giving people a sense that there is something that they can proactively do to maintain their wellbeing and curb helplessness:

“...on that day...they gave me a list of vitamins and supplements to take...I take them every day of my life after that. It’s vitamin C, D and E, Coq10, ginseng...They describe it as being as beneficial as a mother’s milk is to a baby. So I said why not? So I spend about 100 bucks a month

on that...[which is] okay [because] I swear that's [why I've lived so long]....other people...they don't last but a couple of years. Or less" — [PWALS-9]

"...I don't think we ever lost even today our optimism that there could be something... so that why I'm prepared to participate in trials...it just seems to be that if we're given the option of doing something or nothing...I'm going to do something" — [PWALS-5]

One PWALS, semi-jokingly, proposed the presentation of a sort of token to relay condolences and boost optimism, to be provided by healthcare providers during the diagnostic disclosure process when he said: "*Maybe balloons or something...like sorry you have ALS...*" [PWALSI-2].

Another common concern for participants, as considered in the preceding sections in this chapter, was the limited information they received after receiving their confirmed diagnosis. Given this concern, participants of this study recommended that healthcare providers provide detailed information about what to expect in their condition on an ongoing basis. Uncertainty in their ongoing transition through their declining capacity for independence was very common for participants, and it was suggested many times over that verbal discussion and written information aimed at curbing this uncertainty would be incredibly beneficial. As one caregiver articulated:

"I think every patient would like to know what's happening [to them]. Especially when you are seeing such a decline in your abilities...you would want to know...and I think that was one of [our] very great frustrations. Because I think my husband felt that if he were given the proper diagnosis, and that was accurate; then that there would be something, somewhere, a treatment that hprocess could look into...I know with ALS there is no particular treatment that like: hear you go, now you know, and take it, and now off you go and everything will be good. [But] I still think he would like to have been told...I think patients knowing what they have, and even to be able to prepare for [it]...knowing more information about what is happening to me?...how long am I going to be like this?...How long am I going to be fine this way?... Or what I can do?...to learn something...something to help with coping...[Because] the uncertainty is always a very difficult thing..." — [CARI-8]

One PWALS had this to say:

"...the next time I went to a team meeting...I mentioned it [to the ALS clinic neurologist] that...I managed to contact the ALS society and I started going to the support group meetings...and she

looked at me and went: oh, I meant to mention that there's a meeting in [the same city as the ALS clinic]...Thanks...well...I'm in [a different city] so I figured it out on my own...I kind of get the feeling that [the ALS clinic neurologist] expects that I know what I'm doing... And I'm like: You do realize that I'm like totally winging this?...So...more information would be good in relation to ALS....Before it's actually required..." — [PWALS-10]

Reinforcing the idea that PWALS want more information about their condition to help prepare them for future decline, and to guide them to supportive resources. This participant's statement also highlights the concern of patients being required to self-advocate, and seek out information on their own.

Independence, autonomy, lifestyle and normalcy were also discussed in the previous sections of this chapter. Suggestions related to how persons with ALS and closely related conditions could be better supported to maintain these aspects of their experience, were put forward by participants. These suggestions are a reflection of the lived experiences of patients and caregivers, as they were described in the interview process.

Some participants described accessibility in the community as being one of their main challenges in maintaining their active lifestyle. As this caregiver relates:

"I think a lot of it is the time. The extra time it takes for everything and having to think through every scenario. No day is ever simple. When you got to go out to load up the van to go somewhere...[and] find a washroom, that's successful. If you're going to McDonald's or something there's no double family washroom in there. So you have to kind of plan things around situations like that...A couple years ago we went to [a mall]...We thought it would be a fun day to go shopping...[then] take the subway downtown to go to the Blue Jay game. So...we shop....Of course we are rushing around, we're having a lot of fun, and then we're trying...to go down get on the subway. There is no accessible way to get from [the] mall down to the subway. There's no elevator. There's no ramps. There's nothing! Like, we were trying to have just a nice day out. It was enough for me to rip my hair out" — [CARD-7]

Other participants suggested that increased awareness and understanding among healthcare providers and related professionals about the importance of maintaining one's independence and autonomy is critical:

I went to a clinic and...I asked about a feeding tube...because I'm getting to the point where I can't use my arms. I'm like: What am I supposed to do? ...Do you guys use feeding tubes at [this] point?...[They said] no, that's when somebody feeds you....or you're just going to have to ask for more help. I'm like do you know how many times a day I'm asking people for help....and you're honestly going to sit there with your completely ambulatory body and pat me on the knee and say: you're just going to have to ask for more help? ...Fuck you. I'm serious. Lucky that I don't have a left arm because I was seriously thinking of throat punching her, and I like her. She's one of my favorite people at the clinic" — [PWALSD-10]

The participants that lived in hospital described restrictive hospital policies that deny them access to entertainment and activities that they enjoy, while hospital staff don't seem to understand their needs. One of them said: "*...they won't allow me to move with the walker. They hold on to me, saying that, I'm going to fall down...They are over doing it. [My care team members] are all on the extreme end of things...*" [PWALSI-1]. While the other stated: "*It appears that [hospital staff] are not too informed about how to treat this disease...often I feel like I am in a straight-jacket...it seems like there is nothing anybody can do*" [PWALSD-12].

Two caregivers talked about logistical issues being barrier to friends and family being able to visit their love ones in hospital:

"...a bed became available at [the current hospital] first, so that's why he's here....it's not great for people who want to come see him. I mean he's from [another city], so most of the people come to see him are from there. And I work there so I come see him when I can...it's unfortunate that there aren't more beds available in his home community..." — [CARD-13]

"I think there is a lot of deficiency...Number one is because even though the...facility gives access to television programs, sometimes they still just cut off...[certain] channels without asking. And then the patients are left without any [of that] support. The other is that some...friends might...come visit him, but they can't afford to pay \$15 or \$20 for parking every time. But they would come...but cumulatively that's a lot to bear...so maybe there could be some kind of provision for these kind of supporters to get breaks on these types of things" — [PWALS-8]

By and large, patients and caregivers described preferences for care and support relating understanding their condition and its expected trajectory, wanting more information options for treatment and care that would help them preserve their independence, autonomy, desired lifestyle and normalcy for as long as possible.

5.2 Member Check Results

Member checks were carried out as a means of enhancing the rigour of the qualitative study. These member checks were done via email. Participants were offered copies of their interview transcripts, the thematic maps, and a table summarizing the emergent themes. Each participant was contacted individually, and their feedback was used to guide the final framing of the research findings. Feedback regarding the interview data, the analytical process, and derived themes was generally positive. Some participants sought clarification regarding the terms used, and the graphics created during the analytical process. Most participants just offered confirmation of their accordance with the themes presented. Clarification was provided upon request.

The following are excerpts from email communications that took place between the researcher and participants during the member-checking process:

Table 6: Member Check Results

Excerpts
<i>“That's awesome...very impressive format and true to our discussion. Yes reflects my experience well” [PWALSI-9]</i>
<i>“Thank you for sending the results and the presentation of the analysis - I am finding it very informative representation of the results. Some questions: ‘confirmation of suspected’ term is not clear, [and] do the sizes of the circles represent relative/ ie. bigger=greater emphasis? ” [CARI-8]</i>
<i>“Your summary looks great. It makes complete sense to me” [PWALSI-2]</i>

These excerpts serve as examples that reflect the general consensus among participants.

Not all feedback received is reflected here.

Chapter 6: Discussion & Limitations

6.1 Discussion

Many of the themes that emerged from the data align with those found in the literature. For example, the findings consistent with the work of Hogden et al. (2012a), who determined that the nature of the terminal prognosis associated with ALS/ PLS can make it difficult to manage dynamics in healthcare provision and receipt, as well as patients' engagement in care. The current study found this also to be the case, specifically relating to the challenge of managing the uncertain and ongoing transition into increased dependence before death. This challenge seemed to be connected to psychological coping mechanisms such as rationalizing and reasoning, and hopeful theory generation. This highlights a need for an established protocol for hope stimulating, and uncertainty curbing care practices.

Interestingly, the majority of participants said that they were not surprised by their diagnosis at the point of formal disclosure. Participants indicated that after having gone through a lengthy process in trial and error, and having done their own theorizing and research throughout, receiving confirmation of their diagnosis came as more of a relief than a shock. Participants described the internet as a significant tool in their quests to identify the cause of their symptoms. Patients and caregivers talked about doing their own research using the internet a tool, then bringing the results of their searches and discussions to their healthcare providers for further consideration and conversation. Having the resources to be able to conduct their own independent research seemed to offer participants a sense of control and authority during the diagnosis process, which was often long and tedious. Consequently, the process of diagnostic disclosure occurred more as an emergent conversation that concluded with a confirmation of the suspected diagnosis, rather than as a one-time report of surprising test results.

Once they received confirmation of the suspected diagnosis, participants were, in a sense, happy to be able to put a name to their condition, and begin exploring their treatment options. In the case of this sample, it was most common for participants to experience more of an intense reaction to their initial symptoms, as they worried for their ability to continue to perform optimally in life and at work. Participants began to reason and rationalize increasingly as they grew more aware of the limited treatment options that existed for their condition; this is until they slowly began to make peace with their circumstances, potentially facilitated by extended rather than abrupt processes in diagnostic disclosure, or confirmation as it could otherwise be known.

One participant had never received a concrete diagnosis, and indicated that he felt removed from the diagnosis process, such that he believed he was put through diagnostic testing without being made fully aware of what tests were being completed, and why. This participant opted to avoid conversations of future planning and prognosis. Rather, he elected to talk about information provision without describing a will to make peace with his circumstances, or take proactive steps to adapt to an ongoing transition into progressive dependence. This suggests that perhaps patients' full awareness procedures, protocols and justifications in the progress being made towards a diagnosis is a critical component of care that could foster success in coping.

In general, participants indicated that the use of coping strategies like rationalizing and reasoning were consistently present from the point of initial symptom onset to present. Processes in peace-making were commonly described by both participants living with a form of ALS that was slow progressing, and by persons who said they had experienced a steep decline in functional ability soon after their diagnosis. The participants that reported an approximate date of diagnosis that was more than two years prior described a greater awareness of the coping

mechanisms that did, and did not work for them, as well as a better understanding of the resources that were most appropriate given their circumstances. This is excepting one participant who demonstrated a significant amount of self- and circumstantial- awareness despite having been diagnosed about two years prior. This participant was the youngest PWALS in the sample, and resided in a Canadian province other than Ontario (unlike all others in the sample), suggesting that age and locale may be influential factors in coping and/or support seeking processes. Above all, participants' desire to maintain independence, autonomy, lifestyle and normalcy, as well as hope to fair better than the average, and certainty in their expectations what's to come, were indicated to be the main sources of motivation in their coping.

Further, Jakobsson Larsson et al. (2016) discuss coping mechanisms such as support seeking and positive action, avoidance and information seeking, with the latter two being less common among the ALS population. The current study determined that continuous information seeking was actually quite common among persons with ALS. Participants described this as a means of informing their own understanding of their condition in order to curb uncertainty, given the limited and sometimes untimely information they received from healthcare providers.

By and large, participants described information provision as generally lacking in their healthcare experience. Though concerns of overwhelming patients with too much information, too soon, have been discussed by research groups such as McCluskey et al. (2004) and Connolly et al. (2015), the current findings suggest that persons with ALS and the related want more information about what to expect in their condition, and about available resources, than they are currently receiving. As indicated by both patients and caregivers, such information would facilitate coping by providing people with a platform from which to create their own plans for proactive action towards better management of their current and anticipated needs.

Moreover, participants indicated that they did not feel that their needs were understood by healthcare providers, particularly in hospital. Participants living in the community expressed a general satisfaction with the care they received, but also placed more emphasis on the importance of being understood by their friends and family, rather than their healthcare providers. The issue of communication among HCPs and their patients, between HCPs of various disciplines, as part of an effective care team, and between patients, families and/or caregivers and HCPs and/or allied professionals within community settings was also raised frequently in interviews. This is such that participants discussed frustration in having numerous HCPs involved in their care, some of which they see once and then never again. Females and males alike articulated a desire for consistency and constancy in care provision.

The ALS Society of Canada and CCAC/ LHIN were commonly referenced as major sources of community support, and important facilitators of independence, autonomy, lifestyle and normalcy. A sense of community, support in advocacy, and understanding were among the most cited benefits. The participants that lived in hospital were not as connected to these organizations, but desired many of the supports that they offered, especially a broad understanding about ALS related needs. This suggests a potential opportunity to further explore the re-creation of the essential and relevant aspects of these organizations within hospital settings.

Reassuringly, current recommendations for diagnostic disclosure, as laid out by Baile et al. (2000) and Andersen et al. (2012), advise verbal in-person reports of diagnosis and prognostic information. According to participants, this is indeed the way such conversations are typically carried out. In addition, the general process of care as described in the literature was corroborated by the findings of this study. Participants of the current study explained that their

journeys through the care system began with the presentation of mild symptoms to a primary care physician, followed by a process of elimination in diagnosis, and a referral to a neurology specialist and/ or an ALS clinic team in a major city that provides them access to specific supports. This path is consistent with the descriptions of multiple sources both in and outside of Canada.

Some of the study's participants indicated that they had garnered the majority of their relevant experiences in rural settings. Boyd et al. (2016) raise the issue of rural experiences as it relates to ALS care and time to diagnosis. They report no difference in time to diagnosis between rural and urban dwelling participants. Similarly, this study revealed no differences in accounts of rural and urban participants with regards to their diagnosis, disclosure and coping experiences. Both groups discussed challenges getting to ALS clinic appointments, with general community accessibility and coping support availability. Only one participant, who resided in Toronto near an ALS clinic, expressed ease of access. It appeared that it was not the size of the city or town that made a difference, but the location and limited number of clinics.

Related to later life or advance care planning, only two participants addressed the topic directly. These participants described the process of receiving their confirmed diagnosis as an instigator to their will to engage in future planning. While others also described receiving a confirmed diagnosis as pivotal and will initiating, they rather framed the experience around life priority reassessment.

Several factors should be considered in the examination of the current study's findings. First, the average age of participants was lower than expected for an age-related condition (Logroscino, et al., 2015). Anecdotal evidence gathered through general conversation with the study's primary gatekeeper, and through observation, indicated that the older adult participants

potentially eligible for the study were unable to participate due to logistical, mobility and/ or capacity related issues. The majority of potential older adult participants were relatively progressed in their disease, and therefore, either were uninterested in discussing their challenges, and the devastating nature of the condition, or were unable to communicate in a manner that could be accommodated by the technology available for the purposes of this study.

Second, individual, dyad and group interview data were used in the study's analysis. The main reason for this was feasibility. Though many participants were comfortable participating in individual interviews, others indicated that they preferred to participate as a dyad. The persons with ALS who participated in the study as part of a dyad expressed that they desired their caregivers to be present to facilitate communication, and to be there for caregiving and general support. The voice of the persons with ALS tended to be dominant in all interview forms, as was intended. In one case, the caregiver spoke to his own experience rather than to the experience of the PWALS. This information was disregarded in the analysis. Generally speaking, the thoughts and perspectives shared by caregivers about the patient experience were complementary rather than contaminating.

The benefit of the individual interviews was such that participants' unique and individual experiences could be isolated without concern that another party might be influencing their responses. Individual interviews conducted with patients can be thus seen as more advantageous in this regard, given that it is their voice and experiences that were the main focus of the study. Caregivers who participated in individual interviews were also able to share their take on their counterparts' experiences, although there was no way of validating, in the moment, the described experience from the patient perspective. Advantageously, many of the individual interviews conducted with caregivers were complemented by individual interviews with their specific

patient counterparts on different dates. This provided an opportunity to cross-reference and validate the described experiences.

With regards to the dyad interviews, there was initial concern regarding their execution related to the potential for caregivers' voices to be dominant and contaminating the patients' perspectives during the interviews. For this reason, the researcher took great care in ensuring that participants understood the study aims, potentially facilitating the process described above for the dyad interviews in which patient views were paramount. In addition to this favourable dynamic, the dyad interviews also provided a depiction of the real-world dynamics that occur between patients and their caregivers.

A focus group interview was conducted in the preliminary stages of this study; part of the objectives for this process was to identify possible refinements needed to in the patient and caregiver interview guides. The focus group interview data were also included in the analysis. Acknowledging that the interview guides used in the focus group interview would be different from the final versions of the guides used in the individual and dyad interviews, this analysis was carried out to find the emergence of similar themes.

Overall, the various methods of data collection used as part of this study may serve some benefit, as the emergence of similar themes amid different settings suggests the credibility of those themes. In addition, having had patients and their caregivers present together for interviews, granted opportunities for both parties to provide details of the events being described, towards formulating complete accounts of their experiences. The dyad and focus group formats also gave the researcher an opportunity to observe the dynamics that occur between PWALS/PWPLS and caregivers, as well as those that occurred between PWALS/ /PWPLS and

other PWALS//PWPLS, and fellow caregivers, which further facilitated the painting of a clear picture of participants' diagnosis, disclosure and coping experiences.

In light of these considerations, what the data reveals is that what the participants want is a supportive structure around them that enables them to uphold a lifestyle that fits with their 'normal', and allows them to feel autonomous and independent. They do not want to have to 'wing it' in the face of uncertainty with no hope of living a life that isn't dictated by their condition or by a healthcare system that does not understand what it is like to live with ALS. What exactly that supportive structure looks like is to be determined. But, based on the evidence presented by this study, practice may begin to improve with the understanding that walking patients through every step of the diagnosis journey, including what tests are being completed, and why, in addition to keeping them informed of process next steps, and support options, even before a diagnosis is reached, is imperative. These endeavours are likely to be beneficial for coping and the maintenance of quality-of-life.

6.2 Limitations

The main limitation of this study relates to the nature of the method used for analysis. Thematic analysis is one of the most widely used methods in social science and health research, but, because it does not impose any rigid expectations or guidelines in the analysis process, some have criticized the validity of the approach (Braun & Clarke, 2006; Braun & Clarke, 2014). That said, the flexibility of the approach was important for current research project, as it facilitated the process of formulating meaning from the experiences described by both patients and caregivers, and enabled the emergence of unexpected patterns, themes, and insights from the data (Braun & Clarke, 2006). A phenomenological approach, for example, could have been considered, as it

boasts the capacity to help the researcher “understand the lived experience of individuals and their intentions within their ‘lifeworld’” (Crabtree & Miller, 1999, p. 28). But, the approach would have been too rigid to be able to capture the voices of both patients and caregivers regarding the experiences of persons with ALS through the processes of interest.

Another potential limitation of this study has to do with the manner in which data were collected. As described above, individual, dyad and focus group interviews were used to collect qualitative data for analysis. The individual and dyad interviews were conducted using the finalized versions of the interview guides, while the focus group interview was conducted using the preliminary versions of the interview guides. The use of different interview guides can be considered a potential limitation of this study. This is because participant responses might have been different depending on which interview guide was used to guide conversation. For example, the original interview guide might have compelled the researcher to pose the questions in a way that the final version did not, prompting different responses. That said, the difference between the preliminary and finalized interview guides were fairly minor. In addition, the fact that similar themes arose from all transcripts regardless of the interview format is reassuring.

Other limitations of this study include the unintentional omission of older PWALS’ voices. Due to the recruitment issues described above, it was the case that few persons with ALS over the age of 65 years participated in the study. This can be considered a limitation of the current study, as the experiences of those with ALS, potentially with the highest needs were not captured. It might also serve as a justification for careful consideration of recruitment methods in future research.

Further, by the end of the analysis phase of this study new ideas were still emerging from the data, meaning that saturation was not reached. This is certainly a study limitation, but does

not invalidate the results presented. Rather, it too serves as a justification for further consideration and future research aimed at recruiting additional participants that are able to speak to their experiences in diagnosis, disclosure and coping, as did those in the current study. This would complement the findings of the thematic analysis conducted.

Finally, the researcher acknowledges that the interpretation of the data collected may have been influenced by her lived experience, and thus her personal bias, despite the precautions taken.

Chapter 7: Conclusion

Despite some limitations, this study presents results that are an important contribution to the literature. Serving as an initial step in bridging the gaps in our knowledge and understanding regarding the experiences of persons with ALS/ PLS in diagnosis, disclosure, and coping, the current study reveals specific opportunities for further exploration toward improved patient-centred care and support practices. This study validated experiences identified in the literature, and established a reframing of those experiences within a previously unaddressed population.

The study uncovered three categories of support preferences identified in the analysis of the qualitative data: hope stimulating conversations and activities, ongoing information provision aimed at curbing uncertainty, and independence, autonomy, lifestyle and normalcy supported throughout the disease course. These outputs are of value, as they aid in narrowing the focus of ongoing efforts in practice improvement.

This thesis project was conducted within the context of our Canadian healthcare system. Given that this environment offers a unique opportunity to explore patients' experiences within a complex and evolving system, the study results could potentially lend themselves to improved service provision in settings equally as complex or similar (Marchildon, 2013). With that, the adult ALS population presented a unique opportunity to understand such complex health circumstances, while comprehending their perceptions and experiences has revealed opportunities for process and practice improvement (Logroscino, et al., 2015).

Overall, the study offers a reframing of current conceptualizations of what it means to empower and support patients through life-limiting illnesses. In addition, its results provide insight into the unique struggles and support needs of persons with ALS. This greater understanding can aid in efforts to better support patient-provider relationships and

communication, improved efficiency and effectiveness of healthcare practices, and the increased availability and accessibility of important programs and services. These efforts could in turn lead to greater understanding and empathy, as well as greater quality-of-life, for persons with ALS.

Chapter 8: Knowledge Translation & Future Directions

8.1 Knowledge Translation

A final report of this study's findings will be drafted for publication hereafter. In addition, a summative presentation of the study outcomes will be presented to patient and caregiver groups in the local area. A copy of the presentation will also be forwarded to the ALS Society of Canada's main branch for further dissemination.

8.2 Future Directions

This project serves as an initial step in bridging the relevant gaps in our knowledge and understanding toward improved patient-centred care practices in diagnosis, disclosure and coping support. However, further research will be necessary to corroborate the study results and address the healthcare provider perspective. It will be important to obtain additional quantitative evidence to complement the qualitative research findings and ensure saturation. Moreover, it will be necessary to incorporate the voice of healthcare providers that play a role in supporting people living with ALS and other closely related conditions, so that barriers and resource limitations that they might face in trying to fulfill the needs and desires of their patients can be pinpointed. These current and future findings, together, could support the development of evidence-based recommendations for community and hospital-based care practices related to the availability and accessibility of specific programs, services and accommodations.

Additional research surrounding the impacts of other factors and dynamics on the quality-of-life of persons living with ALS is warranted. Religious-, cultural-, age-, and gender-specific needs will be an important area of continued exploration. The differences and similarities in PWALS' experiences in various communities, hospitals, and amid different healthcare funding

structures will also be important matters to explore. An accumulation of further research findings is likely to guide practice and policy developments to benefit the care of persons with chronic or terminal illnesses with short and unpredictable trajectories.

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Appendix A

Recruitment Flyer

ARE YOU CANADIAN?

DO YOU LIVE WITH AMYOTROPHIC LATERAL SCLEROSIS (ALS)? OR DO YOU CARE FOR SOMEONE WHO LIVES WITH ALS?

PARTICIPANTS NEEDED FOR RESEARCH IN RECEIVING AND COPING WITH A DIAGNOSIS OF ALS

We are looking for interested people to take part in a study aimed at *understanding how persons with ALS receive and cope with their diagnosis of ALS.*

As a participant in this study, you would be asked to: *share your thoughts and opinions on the diagnosis, disclosure and coping processes in either a one-on-one or two-person interview at a location that suits on your comfort level.*

Your participation would involve 1 session,
which will take approximately 45-60 minutes.

For more information, or to participate in this study,
please contact:
Kathleen Pauloff
Student Researcher
School of Public Health and Health Systems
at
519-888-4567 ext. 35879 OR
Email: kat.pauloff@uwaterloo.ca

This study has been reviewed by, and received ethics clearance
through a University of Waterloo Research Ethics Committee.

Appendix B

Information Letter

Date: _____

Study Name

Exploring the diagnosis, disclosure & coping experiences of persons living with ALS

Researchers

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Introduction

You are being invited to participate in a research study called “Exploring the diagnostic disclosure & coping experiences of persons living with ALS” conducted by two researchers: Dr. Paul Stolee and Kathleen Pauloff. This study is being conducted as part of Kathleen’s Master’s thesis project.

Your participation in this study is entirely voluntary, so it is up to you to decide whether or not to take part in this study.

Before you decide, it is important for you to understand what the research study involves. This letter will provide you with information about the study. It will explain the purpose of the research, your role in the research and potential benefits, risks and discomforts.

Please take the time to read the following information carefully.

Who is conducting the study?

This study is being conducted by two researchers: Dr. Paul Stolee and Kathleen Pauloff, both of whom are from the School of Public Health and Health Systems at the University of Waterloo.

What is the purpose of the study?

The purpose of this study is to learn about adult persons experiences in receiving and coping with their diagnosis of ALS. Specifically, we would like to hear from patients and caregivers about what their thoughts and experiences are during those processes.

What will happen?

You are being invited to participate in an hour-long (approximate) discussion about your experiences or your understanding of someone's experiences in receiving a diagnosis of ALS. We also want to better understand the coping needs of persons living with ALS. The interviewer will ask you about the conversations surrounding these events, and what was good or bad about the experiences. Specifically, you will be asked about how you or the person you care for reacted to the news, and what strategies were used to cope or make sense of the diagnosis. Based on your response to these questions, the researcher may ask you to provide further details about your experiences, then ask you to reflect on what optimal care would look like to you, in both or either of these processes. The conversation will take place as an individual interview or in a group setting. You may also request the attendance the person who provides care for you, if that is what you would prefer. The interview will be scheduled at your convenience. With your permission, the interview will be audio-recorded.

By providing your insights on your experiences, you will help us gain a better understanding of this research topic. Listening to your experiences will be central to the process of gaining an understanding of adult patients and caregivers' experiences in receiving the diagnosis of ALS, and in coping.

Where will the study take place?

The study will take place wherever you feel most comfortable. Examples of places where interviews may take place include at your local ALS Society of Canada Chapter or at your home.

Will the study help you or others?

We do not know if being in the study will help you, but we hope to learn about what adult persons with ALS and their caregivers experience in receiving and coping with a diagnosis of ALS. We hope that we learn in this study will allow us to make recommendations to help other people in the future.

Will the study harm you?

These are one hour conversations so we don't expect this to bother you. However, if the conversations are upsetting to you, we will stop the conversation and can make sure you have someone to talk with to get help.

Is your participation voluntary?

Your participation in the study is completely voluntary and you may choose to withdraw from participating at any time, up to the point of publication. You may withdraw your consent to participate over the phone, in person or via email. Your decision whether or not to participate has no effect on the care you receive or your relationship with the ALS Society of Canada, or any other affiliated organization. You can decline to participate in the study without penalty. If you agree to participate, you will be able to talk about whatever you are comfortable with. If there is a question you do not want to answer, you may say, "I don't want to answer that question."

Can you change your mind or decide not to answer a question?

You can change your mind and stop being part of the study at any time, up to the point of publication. Your decision to stop, or to refuse to answer particular questions, has no effect on your healthcare or any support provided through the ALS Society of Canada, now or in the

future. If you decide to leave the study, all of the data collected from you will be immediately destroyed.

What will happen to your information?

All information you give during the conversation will be held in confidence. Your information will be kept in a locked filing cabinet at the University of Waterloo, School of Public Health and Health Systems, and will be accessed only by members of the research team. Your name will not appear on any of the data.

Only the project team will have access to entire interviews. With your permission, anonymous quotations may be used in the following way(s):

- in teaching and demonstration materials
- in scholarly papers, articles and other publications, and
- in presentations at academic, healthcare conferences

Confidentiality, as it pertains to your identity, will be respected to the fullest extent possible by law. This means that, in the event that you disclose that you are at risk of harming yourself or someone else, or that a child or elder is being harmed or neglected confidentiality will have to be breached, and the appropriate authorities will be notified. Electronic files containing study data will be password-protected, and will be destroyed after a minimum of 5 years. Audiotapes, transcriptions, questionnaires and data files will remain anonymous such that no names will be associated with the data. Each participant will be assigned an identification number, which will be used to organize the data. There are no conditions under which the confidentiality regarding your identity cannot be guaranteed, other than those specified by local legislation, as per the above.

Who can I contact if I have any questions?

If you have questions about the research or about your role in the study, please feel free to contact Dr. Paul Stolee by phone at (519) 888 4567 x 35879 or by e-mail (stolee@uwaterloo.ca) or Kathleen Pauloff by phone at (519) 888 4567 x 35879 or by email (kat.pauloff@uwaterloo.ca). This research has received clearance through a University of Waterloo Research Ethics Committee. If you have any comments or concerns with this study, please feel free to contact please contact the Chief Ethics Officer, Office of Research Ethics, at 519-888-4567, ext. 36005 or ore-ceo@uwaterloo.ca

What will happen after the study is over?

The researchers will ask if you would like to be contacted in the future to go over the findings and give your opinions on the results. If you do not want to be contacted in the future, you may indicate this preference without penalty and without any consequences to your healthcare or your relationship to the ALS Society of Canada.

Conclusion

We are excited about this study and are looking forward to listening to your experiences and insights on receiving and coping with the diagnosis of ALS. We sincerely hope that you will consider participating.

Appendix C

Participant Contact Form

Please circle one (below):

Patient

Caregiver

Other

Specify: _____

Name: _____

Please provide either your full name, first name, initials or a pseudonym

Telephone #: *Please complete any or all of the following.*

Home#_____

Work # _____

Cell # _____

Email Address:

I wish to be contact about my potential participation in the study entitled “Exploring the diagnosis, disclosure & coping experiences of persons living with ALS”.

YES NO

Appendix D

Recruitment Script

Hello, my name is Kathleen Pauloff, a Masters student working under the supervision of Dr. Paul Stolee in the Geriatric Health Systems research group at the University of Waterloo School of Public Health and Health Systems. I understand that you have expressed interest in participating in a research study that we are conducting.

To review how you would contribute to our study: we are hoping to talk to you about your Experiences or your understanding of someone's experiences in receiving a diagnosis of ALS. We also want to better understand the coping needs of persons living with ALS. Your participation in this study will involve an hour-long (approximate) interview, in which, the interviewer will ask you to talk about your experiences in receiving a diagnosis of ALS or similar, or as someone who has cared for a person who has gone through the ALS diagnosis process or similar. She will ask you about the conversations surrounding the event, and what was good or bad about the experience. You will then be asked about your or your loved one's reaction(s) to the news, and what strategies were used to cope or make sense of the diagnosis. Based on your responses to these questions, the researcher may ask you to provide further details about your experiences, then ask you to reflect on what optimal care would look like to you, in both or either of these processes. The conversation will take place as an individual or dyad interview, or in a group setting. You may also request the attendance of the person who provides care for you or the person you provide care for, if that is what you would prefer. The interview will be scheduled at your convenience. With your permission, the interview will be audio-recorded.

I would like to assure you that this study has been reviewed by, and received ethics clearance through a University of Waterloo Research Ethics Committee. However, the final decision to participate is yours.

Are you still interested in participating?

If you are interested in hearing more about the study and participating, please contact Kathleen Pauloff by phone at (519) 888 4567 x 35879 or by email (kat.pauloff@uwaterloo.ca).

If you would be comfortable setting up a time for an interview, please email Kathleen with your preferred time and date from the list below.

List potential dates/times

I am happy to answer any other questions you may have via email, over the phone or in person.

Participants to be reminded of the following:

- You may decline to participate in any part of the interview and may terminate the interview at any time.
- The interview will be audio-recorded to facilitate collection of information, and later transcribed for analysis.
- All information regarding your identity will remain confidential, will be stored in a secure location, and will be destroyed after a minimum of 5 years.

If no longer interested in participating: Thank you for your time.

Appendix E

Consent Form

By signing this consent form, you are not waiving your legal rights or releasing the investigator(s) or involved institution(s) from their legal and professional responsibilities.

I have read the information presented in the information letter about a study being conducted by Kathleen Pauloff of the Department of Public Health and Health Systems at the University of Waterloo. I have had the opportunity to ask any questions related to this study, to receive satisfactory answers to my questions, and any additional details I wanted.

I am aware that I have the option of allowing my interview to be audio recorded to ensure an accurate recording of my responses.

I am also aware that excerpts from the interview may be included in the thesis and/or publications to come from this research, with the understanding that the quotations will be anonymous, and used in the following way(s):

- in teaching and demonstration materials
- in scholarly papers, articles and other publications, and
- in presentations at academic, healthcare conferences

I was informed that I may withdraw my consent at any time without penalty by advising the researcher.

This study has been reviewed and received ethics clearance through a University of Waterloo Research Ethics Committee (ORE#22512). If you have questions for the Committee contact the Chief Ethics Officer, Office of Research Ethics, at 1-519-888-4567 ext. 36005 or ore-ceo@uwaterloo.ca.

For all other questions contact **519-888-4567 ext. 35879** or by email at kat.pauloff@uwaterloo.ca. You can also contact my supervisor, Professor **Paul Stolee** at **519-888-4567 ext. 35879** or email stolee@uwaterloo.ca.

With full knowledge of all foregoing, I agree, of my own free will, to participate in this study.

YES NO

I agree to have my interview audio recorded.

YES NO

I agree to the use of anonymous quotations in any thesis or publication that comes of this research.

YES NO

Participant (or Proxy) Name: _____ (Please print)

Participant (or Proxy) Signature: _____

Witness Name: _____ (Please print)

Witness Signature: _____

Date: _____

When this study is completed, we will write a summary of the results. Would you be interested in receiving a copy of the report?

YES, please e-mail me a summary of the results. My e-mail address is:

YES, please mail me a summary of the results. My mailing address is:

NO, I do not wish to receive a summary of results

Appendix F

Feedback Letter

Date: _____

Dear Participant,

I would like to thank you for your participation in this study entitled “Exploring the diagnosis, disclosure & coping experiences of persons living with ALS”. As a reminder, the purpose of this study is to learn about adult persons experiences in receiving and coping with their diagnosis of ALS.

Participation in this study is voluntary. It involved an interview of approximately 45-60 mins, at which time data were collected. The data collected will contribute to a better understanding of how adult persons with ALS receive and cope with their diagnosis, and how they engage in care. Our hope is that the information you have provided will help improve practices in healthcare.

This study has been reviewed and received ethics clearance through a University of Waterloo Research Ethics Committee (ORE#22512). If you have questions for the Committee contact the Chief Ethics Officer, Office of Research Ethics, at 1-519-888-4567 ext. 36005 or ore-ceo@uwaterloo.ca.

For all other questions contact me, Kathleen Pauloff (see contact information below). Please remember that any data pertaining to your identity will be kept confidential. Once all the data are collected and analyzed for this project, I plan on sharing this information with the research community through seminars, conferences, presentations, and journal articles. If you are interested in receiving more information regarding the results of this study, or would like a summary of the results, please provide your email address, and when the study is completed, anticipated by August 2018, I will send you the information. In the meantime, if you have any questions about the study, please do not hesitate to contact me by email or telephone as noted below.

Sincerely,



Kathleen Pauloff

Student Investigator

Phone: 519-888-4567 ext. 35879

Email: kat.pauloff@uwaterloo.ca

You can also contact my supervisor, Professor Paul Stolee at 519-888-4567 ext. 35879 or email stolee@uwaterloo.ca.

Appendix G

Focus Group Interview Guide (Patient)

Section 1: Building Rapport

1. Tell me a little bit about yourself.

Prompt: Where are you from? What is your family life like? Could you walk me through a day in your life? Or the last few months/ years of your life?

Section 2: Diagnostic Disclosure Experiences

2. Please describe your experiences (i.e. how you felt, what you thought, etc.) in your conversation(s) with the healthcare provider(s) that broke the news to you.

Prompt: What was positive about your experiences, if anything? What was challenging about the process, if anything? How did the conversation surrounding your diagnosis with your healthcare provider(s) play out? What was your level of comfort in the situation? How did you feel in the conversation? What went through your mind during the conversation? Did you feel that you were given too little information? Too much information? Enough information.

3. How did you react when the healthcare provider(s) broke the news to you?

Prompt: How did you act? What was your emotional response? What was your physical response? What did you say? What types of questions did you ask, if any?

Section 3: Coping Experiences

4. What does coping mean to you?

Prompt: What is good coping? What is bad coping? How do you normally cope with tough situations?

5. How did you cope during and the conversation you had with the healthcare provider(s) that gave you the news of your diagnosis.

Prompt: What did you do to make sense of the information presented by the healthcare professional(s)? How did you relate to the diagnosis? Did you accept it? Did you believe it might have been wrong?

Section 4: Engagement Experiences

6. What are three words you might use to describe your level of involvement in your care, in the decision-making, and/or in planning? Please explain.
7. What are your thoughts about the conversation(s) you had/ are having with your healthcare provider(s) about making decision for your care?

Prompt: What questions were/ are you asked? What topics were/ are brought up? What decisions were/ are you asked make? How 'in control' do you feel?

8. In a perfect world, what would your journey from diagnosis to where you are now look like?

Prompt: What works? What doesn't? What are important things to consider? What do you wish would have been done differently? What do you think might have made things easier, more comfortable, or clearer? What might have helped you to become more involved, or feel more in control?

9. What benefits do you see in improving the way healthcare providers discuss diagnoses with their patients?
10. What benefits do you see in being more involved, or in control of your care, if any?

Section 5: Closure

11. What are any other thoughts you have regarding conversations about the diagnosis, and/ or coping and your level of involvement in your care?
12. Please tell me one of your greatest achievements in life today, or tell me about one thing in your life that never fails to put a smile on your face.

Prompt: Have you ever won an award or been recognized for doing something cool? Is there a person that makes you happy? Do you have a favourite activity?

Appendix H

Focus Group Interview Guide (Caregiver)

Section 1: Building Rapport

1. Tell me a little bit about yourself.

Prompt: Where are you from? What is your family life like? Could you walk me through a day in your life (either personal or professional)?

Section 2: Diagnostic Disclosure Experiences

2. Please describe your experiences (i.e. how you felt, what you thought, etc.) in the conversation(s) had with the healthcare provider(s) that broke the news to your loved-one or client.

Prompt: What was positive about your experiences, if anything? What was challenging about the process, if anything? How did the conversation surrounding your diagnosis with the healthcare provider(s) play out? What was your level of comfort in the situation? How did you feel in the conversation? What went through your mind during the conversation? Did you feel that too little information was given? Too much information? Enough information?

3. How did your loved-one or client react when the healthcare provider(s) broke the news?

Prompt: How did they act? How did you perceive their emotional response?? What did they say? What types of questions did they ask, if any?

4. How did you react when the healthcare provider(s) broke the news to your loved-one or client?

Prompt: How did you act? What was your emotional response? What was your physical response? What did you say? What types of questions did you ask, if any?

Section 3: Coping Experiences

5. What does coping mean to you?

Prompt: What is good coping? What is bad coping? How do you normally cope with tough situations? How do you believe your loved-one or client normally copes with tough situations?

6. How do you believe your loved-one or client coped during and after the conversation had with the healthcare provider (s) that broke the news of their diagnosis to them?

Prompt: What did they do? What did they say?

7. How did you cope with hearing the diagnosis?

Prompt: What did you do? What did you say? What thoughts went through your mind? Did you have concerns or worries?

Section 4: Engagement Experiences

8. What are three words you might use to describe your loved-one or client's level of involvement in their care, in the decision-making, and/or in planning? Please explain.
9. What are your thoughts about the conversation(s) had with healthcare provider(s) about making decision for your loved-one or client's care?

Prompt: What questions were asked? What topics were brought up? What decisions had to be made? How 'in control' do you believe your loved-one or client felt?

10. In a perfect world, what would your loved-one or client's journey from diagnosis to where they are now look like?

Prompt: What works? What doesn't work? What do you wish could have been done differently? What do you think might have made things easier, more comfortable, or clearer? What might have helped your loved-one or client feel more involved, or more in control?

11. What benefits do you see in improving the way healthcare providers discuss diagnoses with their patients?
12. What benefits do you see in patients being more involved, or in control of their care, if any?

Section 5: Closure

13. What are any other thoughts you have regarding the diagnosis process, conversations about the diagnosis, and/ or your loved-one or client's level of involvement in your care?
14. Please tell me one thing you admire about your loved-one or client.

Prompt: Have they ever won an award or been recognized for doing something cool? What is their favourite activity? What are they good at?

Appendix I

Dyad/ Individual Interview Guide (Patient)

Each participant will receive a copy of the study's information letter and consent form. Before being asked to sign the consent form, the researcher will read through the information letter with the participant and answer any questions they might have. After all initial questions related to the nature and purpose of the study have been answered, and the participants indicates that they would like to continue on to participate in the study, they will be asked to sign the front side of the study consent form. Should they sign, the following questions and probes will posed:

Approximate interview duration: 45- 60 minutes

Section 1: General/Background Information & Building Rapport

13. When were you diagnosed with ALS? What were the circumstances surrounding your diagnosis?

Prompt: How long ago were you diagnosed with ALS? How long before you received your diagnosis did you start experiencing symptoms? What was the process between visiting your doctor about your initial symptoms of ALS and being diagnosed like for you?

Section 2: Diagnostic Disclosure Experiences

The following questions are intended to capture participants' experiences in diagnostic disclosure. Section context and aim (i.e. "Let's now talk about you being given your diagnosis. I would like to know about that conversation and how you reacted to hearing your diagnosis.") to be introduced to participant prior to posing the following questions:

14. Please describe your experiences (i.e. how you felt, what you thought, etc.) in your conversation(s) with the healthcare provider(s) that broke the news to you.

Prompt: How did you react when the healthcare provider(s) broke the news to you? What was positive about your experiences, if anything? What was challenging about the process, if anything? How did the conversation surrounding your diagnosis with your healthcare provider(s) play out?

15. Now that you have received your diagnosis, thinking back, how do you think you would have liked to receive the news about your diagnosis?

Prompt: Is there anything the healthcare provider could have done differently? What would have been helpful?

Section 3: Coping Experiences

The following questions are intended to capture participants' experiences in coping. Section context and aim (i.e. "Let's now talk about coping. I would like to know about how you have coped since hearing your diagnosis.") to be introduced to participant prior to posing the following questions:

16. How did you cope after learning about your condition? Tell me about this process and how it has impacted your life.
17. Is there anyone you lean on for support in your condition? If yes, who? If no, explain what kind of support is or would be helpful to you.
18. What are your experiences in coping after hearing your diagnosis?

Prompt: What do you do to cope? What do you find helpful in coping? What challenges your ability to cope?

19. Have you noticed any changes in your coping over time? If yes, what changes have you noticed? How would you describe your past and current coping styles? If no, what aspects of your coping have remained dominant and constant?
20. What are your thoughts about/ experiences regarding the conversation(s) you had/ are having with your family, friends or healthcare provider(s) about coping, if any?

Prompt: What questions were/ are you asked? What topics were/ are brought up? Do you feel understood and/or supported? Why or why not? Is there anyone in particular you've felt most supported by? Who? How have you been supported in terms of your coping?

Section 4: Preferences & Suggestions for Improvement

The following questions are intended to capture participants' preferences and suggestions. Section context and aim (i.e. "Let's now talk about coping the 'shoulda, woulda, coulda'. I would like to know about what you think could have been different and/or better in your journey through hearing your diagnosis and coping.") to be introduced to participant prior to posing the following questions:

21. In a perfect world, what would your journey from diagnosis to where you are now look like?

Prompt: What works? What doesn't? What are important things to consider? What do you wish would have been done differently? What do you think might have made things easier, more comfortable, or clearer? How could you have been better supported in coping?

22. As we discussed, my research focuses on helping healthcare providers in discussing diagnoses with their patients, and in supporting them in coping. On that basis, what do you think, from your experience, might be helpful or important for me to know?

Section 5: Closure

23. What are any other thoughts you have regarding the diagnosis process, conversations about the diagnosis, and/ or your ability to cope/ support in coping?

Participants will then be thanked for participating the study using the follow script template:

"Thank you so much for participating in this study; your insights are very valuable and will play a role in improving the way healthcare providers inform people of their diagnoses and support them in coping. I will be speaking with others to learn about their experiences as well. After that, I will go through a formal process of reading through what everyone has said and drawing out the major themes. During this process, I will contact you, if I may, and ask you to complete what is called a 'member check'. This just means that I will ask you to verify that what I've captured, is what you intended to say, and is true to your experiences. After all the information is verified, I will put together a final report, which I am happy to pass along to you, if you'd like."

Participants will then be asked to complete the reverse side of the consent form, giving permission for the researcher to contact them thereafter, and to indicate whether or not they would like to receive a copy of the final report.

Appendix J

Dyad/ Individual Interview Guide (Caregiver)

Each participant will receive a copy of the study's information letter and consent form. Before being asked to sign the consent form, the researcher will read through the information letter with the participant and answer any questions they might have. After all initial questions related to the nature and purpose of the study have been answered, and the participants indicates that they would like to continue on to participate in the study, they will be asked to sign the front side of the study consent form. Should they sign, the following questions and probes will posed:

Approximate interview duration: 45- 60 minutes

Section 1: General/Background Information & Building Rapport

1. When was your loved one diagnosed with ALS? What were the circumstances surrounding their diagnosis?

Prompt: How long ago were they diagnosed with ALS? How long before they received their diagnosis did they start experiencing symptoms? What was the process between visiting their doctor about their initial symptoms of ALS and being diagnosed like for them?

Section 2: Diagnostic Disclosure Experiences

The following questions are intended to capture patients' experiences in diagnostic disclosure. Section context and aim (i.e. "Let's now talk about your loved one being given their diagnosis. I would like to know about that conversation and how your loved one reacted to hearing their diagnosis.") to be introduced to participant prior to posing the following questions:

2. Please describe your loved one's experiences (as you understand them) in their/your conversation(s) with the healthcare provider(s) that broke the news to them.

Prompt: How did you react when the healthcare provider(s) broke the news to you? What was positive about your experiences, if anything? What was challenging about the process, if anything? How did the conversation surrounding your diagnosis with your healthcare provider(s) play out?

3. Thinking back, how do you think your loved one would have liked to receive their news about their diagnosis?

Prompt: What, if anything, could the healthcare provider have done differently? What would have been helpful?

Section 3: Coping Experiences

The following questions are intended to capture participants' experiences in coping. Section context and aim (i.e. "Let's now talk about coping. I would like to know about how your loved one has coped since hearing your diagnosis.") to be introduced to participant prior to posing the following questions:

4. How did your loved one cope after learning about their condition? Tell me about this process and how it has impacted their/your life/lives.
5. Is there anyone your loved one leans/ leaned on for support in their condition? If yes, who? If no, explain what kind of support might be/ have been helpful to them.
6. What are your loved one's experiences in coping after hearing their diagnosis?

Prompt: What does/ did your loved one do to cope? What do/ did they find helpful in coping? What challenges/ challenged their ability to cope?

7. Have/ did you notice(d) any changes in your loved one's coping over time? If yes, what changes have/ did you notice(d)? How would you describe their past and/ or current coping styles? If no, what aspects of their coping have/ did remain(ed) dominant and constant?
8. What are your thoughts about/ experiences regarding the conversation(s) that were had/ are being had with your family, friends or healthcare provider(s) about your loved one's coping, if any?

Prompt: What questions were/ are asked? What topics were/ are brought up? Do you think your loved one feels/ felt understood and/or supported? Why or why not? Is there anyone in particular you've felt has been most supportive? Who? How has your loved one been supported in terms of their coping?

Section 4: Preferences & Suggestions for Improvement

The following questions are intended to capture participants' preferences and suggestions. Section context and aim (i.e. "Let's now talk about coping the 'shoulda, woulda, coulda'. I would like to know about what you think could have been different and/or better in your loved one's journey through hearing their diagnosis and coping.") to be introduced to participant prior to posing the following questions:

9. In a perfect world, what would your loved one's journey from diagnosis to where they are now/ beginning to end look like?

Prompt: What works? What doesn't? What are important things to consider? What do you wish would have been done differently? What do you think might have made things easier, more comfortable, or clearer? How could your loved one have been better supported in coping?

10. As we discussed, my research focuses on helping healthcare providers in discussing diagnoses with their patients, and in supporting them in coping. On that basis, what do you think, from your experience, might be helpful or important for me to know?

Section 5: Closure

11. What are any other thoughts you have regarding the diagnosis process, conversations about the diagnosis, and/ or a patient's ability to cope/ support in coping?

Participants will then be thanked for participating in the study using the follow script template:

"Thank you so much for participating in this study; your insights are very valuable and will play a role in improving the way healthcare providers inform people of their diagnoses and support them in coping. I will be speaking with others to learn about their experiences as well. After that, I will go through a formal process of reading through what everyone has said and drawing out the major themes. During this process, I will contact you, if I may, and ask you to complete what is called a 'member check'. This just means that I will ask you to verify that what I've captured, is what you intended to say, and is true to your experiences. After all the information is verified, I will put together a final report, which I am happy to pass along to you, if you'd like."

Participants will then be asked to complete the reverse side of the consent form, giving permission for the researcher to contact them thereafter, and to indicate whether or not they would like to receive a copy of the final report.

Appendix K

Patient Demographics Form

Reference Number: _____
For office use only

Please circle one (below):

Patient Caregiver Other Specify:_____

Age:_____

Please circle one (below):

Male Female Otherwise Identified

Current City:_____

Appendix L

Ethics Approval

Dear Researcher:

The recommended revisions/additional information requested in the ethics review of your application for the study:

Title: Tailored engagement: Understanding the needs of older adult persons with ALS in receiving and coping with a diagnosis, and in subsequent participation in care ORE #: 22512
Faculty Supervisor: Paul Stolee (stolee@uwaterloo.ca) Student Investigator: Kathleen Pauloff (kat.pauloff@uwaterloo.ca)

have been reviewed and are considered acceptable. A University of Waterloo Research Ethics Committee is pleased to inform you this study has been given ethics clearance.

A signed copy of the notification of ethics clearance will be sent to the Principal Investigator (or Faculty Supervisor in the case of student research). Ethics approval to start this research is effective as of the date of this email. The above named study is to be conducted in accordance with the submitted application (Form 101/101A) and the most recent approved versions of all supporting materials.

University of Waterloo Research Ethics Committees operate in compliance with the institution's guidelines for research with human participants, the Tri-Council Policy Statement for the Ethical Conduct for Research Involving Humans (TCPS, 2nd edition), Internalization Conference on Harmonization: Good Clinical Practice (ICH-GCP), the Ontario Personal Health Information Protection Act (PHIPA), and the applicable laws and regulations of the province of Ontario. Both Committees are registered with the U.S. Department of Health and Human Services under the Federal Wide Assurance, FWA00021410, and IRB registration number IRB00002419 (Human Research Ethics Committee) and IRB00007409 (Clinical Research Ethics Committee).

Renewal: Multi-year research must be renewed at least once every 12 months unless a more frequent review has otherwise been specified by the Research Ethics Committee on the signed notification of ethics clearance. Studies will only be renewed if the renewal report is received and approved before the expiry date (Form 105 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/renewals>). Failure to submit renewal reports by the expiry date will result in the investigators being notified ethics clearance has been suspended and Research Finance being notified the ethics clearance is no longer valid.

Modification: Amendments to this study are to be submitted through a modification request (Form 104 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/modifications>) and may only be implemented once the proposed changes have received ethics clearance.

Adverse event: Events that adversely affect a study participant must be reported as soon as possible, but no later than 24 hours following the event, by contacting the Chief Ethics Officer. Submission of an adverse event form (Form 106 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/report-problems>) is to follow the next business day.

Deviation: Unanticipated deviations from the approved study protocol or approved documentation or procedures are to be reported within 7 days of the occurrence using a protocol deviation form (Form 107 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/report-problems>).

Incidental finding: Anticipated or unanticipated incidental findings are to be reported as soon as possible by contacting the Chief Ethics Officer. Submission of the incidental findings form (Form 108 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/report-problems>) is to follow within 3 days of learning of the finding. Participants may not be contacted regarding incidental findings until after approval has been received from a Research Ethics Committee to contact participants to disclose these findings.

Study closure: Report the end of this study using a study closure report (Form 105 - <https://uwaterloo.ca/research/office-research-ethics/research-human-participants/renewals>).

You are responsible for obtaining any additional institutional approvals that might be required to complete this study.

Best wishes for success with this study.

Karen Pieters, MPH
Manager
Office of Research Ethics
EC5, 3rd floor
[519.888.4567 ext. 30495](tel:519.888.4567)
kpieters@uwaterloo.ca

Appendix M

Declaration of Non-Disclosure

I acknowledge that, in my capacity as a member (or staff, employee) of _____, I will have access to certain confidential information. This information includes, but is not limited to the following: files, data books, diagrams, records, studies, protocols, reports, draft publications, interviews, surveys, samples, schedules, appraisals, computer programs, and statistical information. Confidential information may be oral, written, or electronic.

I understand that all _____ members must sign a Declaration of Non-Disclosure when they commence their association with the _____. This includes undergraduate and graduate students conducting research within the _____ and temporary members or visiting faculty from other institutions. Under this declaration, members consent to keep all matters to which they are privy related to all projects being conducted at the _____ confidential.

I agree that during my association with the _____ and for a period of five years after termination of employment or association with the _____, I shall not disclose to any other person, firm or corporation, any confidential information relating to any projects, other than for the specific purposes required by my duties within the _____, without previous consent in writing from the Director of the _____ or his/her designate.

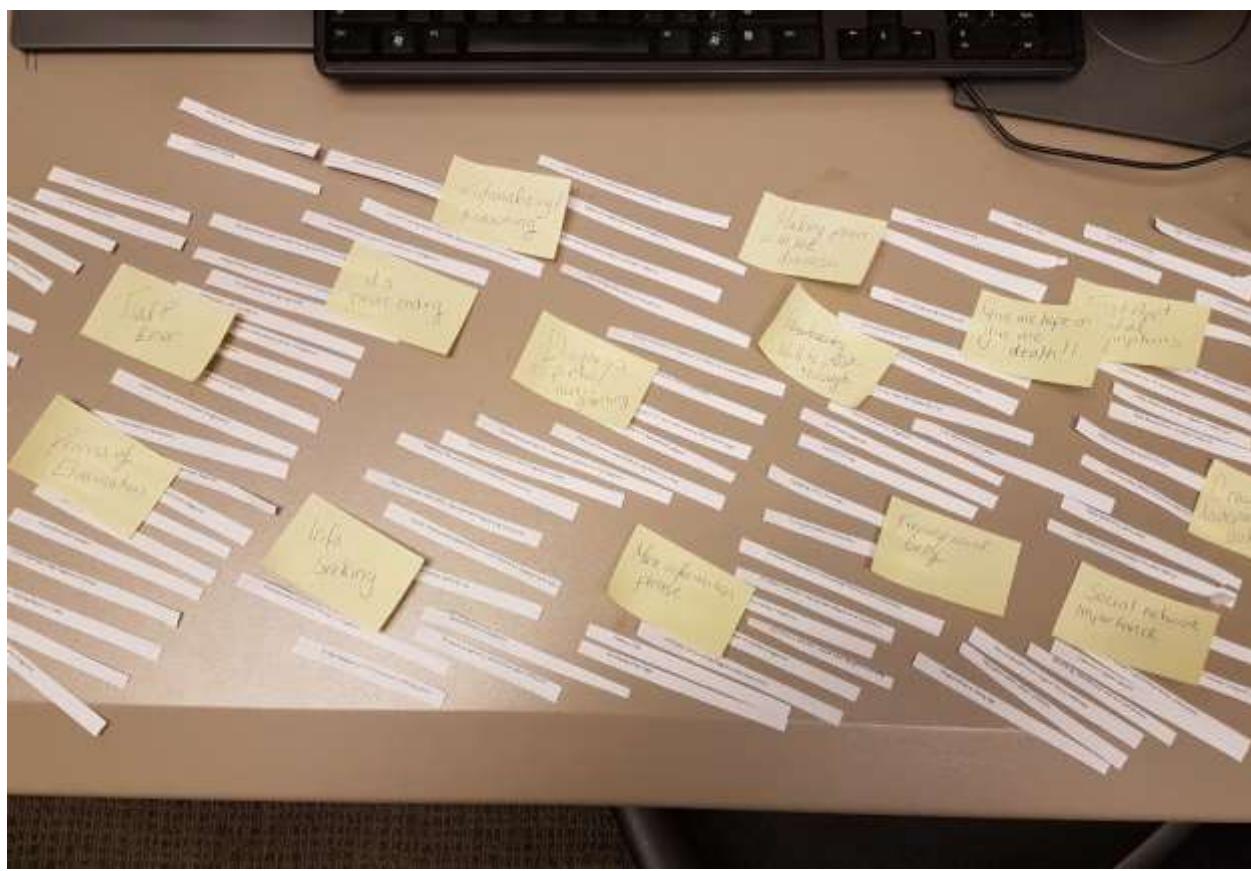
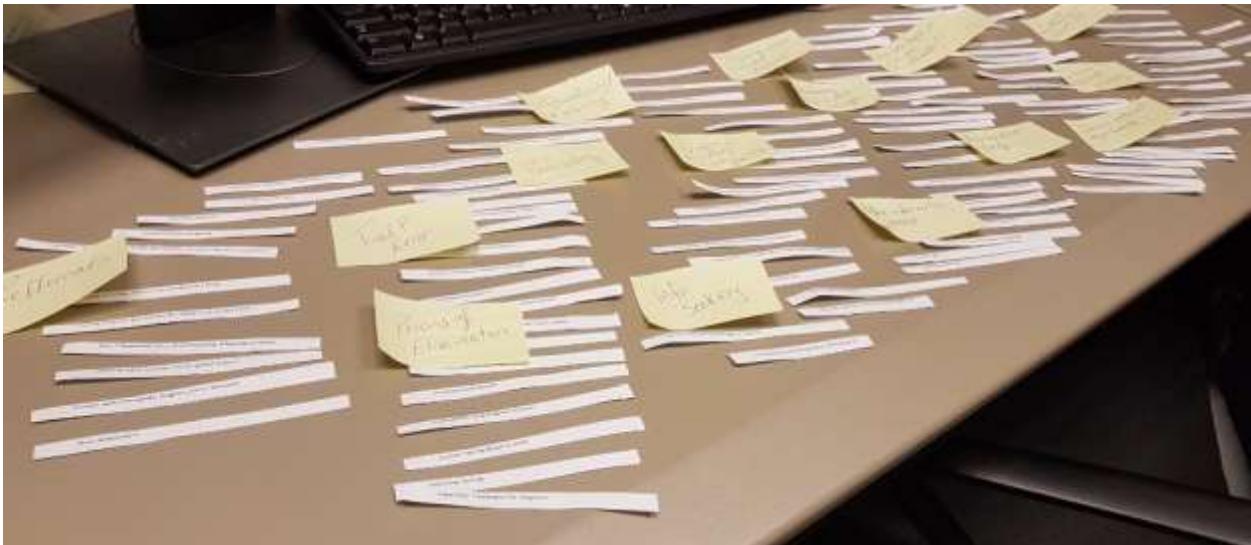
I also understand that I am required to notify the Director of the _____ or his/her designate immediately of any breach of my obligations or conflict of interest under this agreement which comes to my attention.

By signing and returning a copy of this document to the Director of _____ or his/her designate, I confirm my understanding and acceptance of the above clause and will comply with these clauses. I also agree that my obligation to comply with the above will survive my termination of association with the _____ for a period of five years.

Signed: _____
Name (printed): _____
Witness: _____
Date: _____

Appendix N

Codes & Nodes



Appendix O

Thematic Maps

