Communication, Information Gathering and Use Among ALS Stakeholders: Diagnosis and Care

by

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ABSTRACT

The pre and post diagnostic phases of amyotrophic lateral sclerosis (ALS) care are inherently uncertain for patients and caregivers. The emotional character of ALS related communications requires that providers have a high degree of skill and tact. Understanding patient and caregiver preferences is an important component of high-quality clinical communication. External information seeking is common pre and post diagnosis, indicating a degree of information deficit among this disease group. In this qualitative work I investigate the information and communication needs of ALS patients (n=12), caregivers (n=10), and providers (n=13) during diagnosis and over the disease course (n=35). Using qualitative content analysis methods I explore the ALS communication environment by: 1) analyzing the ALS diagnostic experience of my cohort, including their communication needs and information preferences. 2) identifying the information seeking practices of patients and caregivers over the course of their disease. The use and value of this information is also explored. Preferences related to information source and content are discussed. This thesis also provides recommendations for clinicians that are focused on the delivery of a diagnosis. It also emphasizes the importance of ALS patient and caregiver support groups as a medium of information exchange and facilitator of successful adaptation.

PREFACE

This is an original work by Mackenzie Moir. This research project received research ethics approval from the University of Alberta Research Ethics Board, Project Name "ALS Care Decision Making: Understanding Clinician-Patient/Caregiver Communications", No. 00064853, originally approved on 22nd of August 2016.

DEDICATION

I dedicate this work to the incredible individuals who make up the ALS community.

I can never repay you for so generously gifting me that which was of incalculable value to you, your time.

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First, I would like to thank the patients and caregivers who fought through tears, laughter, and fatigue to share their incredible stories with me.

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GLOSSARY OF TERMS

ALS	Amyotrophic lateral sclerosis
ALS DI	Amyotrophic lateral sclerosis Depression Inventory
BDI	Beck Depression Inventory
EFNS	European Federation of Neurological Societies
GP	General Practitioners
PBA	Pseudobulbar Affect
SPIKES	**S**etting up an interview, assessing the patient's **P**erception, obtaining the patient's **I**nvitation, giving **K**nowledge and information to the patient, addressing the patient's **E**motions with empathic responses; and **S**trategy and summary.

CHAPTER 1: INTRODUCTION AND OBJECTIVES

Amyotrophic lateral sclerosis (ALS) is a devastating neurodegenerative disease that results in the atrophy and paralysis of skeletal muscle; it is incurable and nearly always fatal. As such, emotional distress often characterizes the ALS diagnostic experience. This emotional distress is exacerbated by the onset of heterogenous and unexplained symptoms, which leads to frustrating diagnostic clinical encounters. After diagnosis, the unpredictable and progressive nature of the disease requires patients and caregivers to adapt to the illness over the course of their care.

Information gaps and the uncertainty pre- and post-diagnosis compound patient and caregiver frustration. Thus, clinical communications play a crucial role, as do the information needs and preferences of individual patients and caregivers. Identification of the evolving needs and preferences of patients and caregivers over the course of ALS requires responsiveness and adaptability from healthcare providers in the form and content of their communications.

My thesis research, therefore, aims to:

(1) analyze the experiences and preferences of patients, caregivers, and healthcare providers in receiving or providing an ALS diagnosis;
(2) analyze patient and caregiver motivations for seeking information outside of clinical encounters;
(3) identify the external information sources used and the specific topics of interest, and their respective value to patients and caregivers; and
(4) understand the information and communication preferences of patients and their caregivers about disease progression, therapy and prognosis, and the extent to which these are accounted for by healthcare providers.

My research was conducted with patients and caregivers recruited from the ALS Clinic in Edmonton, Alberta as part of a larger project on clinical communications to improve the lives of ALS patients and their caregivers. As an exploratory study, it had the further aim of determining the feasibility of interview-based qualitative research with ALS patients and their caregivers.

In my thesis, I use the term “patient” as defined by Canada’s Strategy for Patient-Oriented Research: “anyone who has personally lived the experience of a health issue” and the term

"caregiver" to refer to informal caregivers, including family and friends, who have lived experience of ALS (http://www.cihr-irsc.gc.ca/e/49232.html). Finally, I use the term "provider" to refer to all healthcare providers, including allied health professionals, primary care clinicians, and specialists.

In this chapter, I first provide a background literature review and rationale for my thesis research, followed by an outline of this paper-based thesis.

BACKGROUND AND RATIONALE

Amyotrophic Lateral Sclerosis

ALS is a progressive motor neuron disease that results in both upper and lower motor neuron degeneration and paralysis of voluntary skeletal muscles (Hobson, Harwood, McDermott, & Shaw, 2016). Primary symptoms of ALS typically include muscular atrophy, fasciculations, and spasticity (Moore, McDermott, & Shaw, 2008). ALS is incurable and nearly always fatal; it has a 50% mortality rate within three years of symptom onset (Mitchell & Borasio, 2007), usually from respiratory failure (Hobson et al., 2016; Talbot, 2004). Riluzole is currently the only approved pharmacological agent in Canada that has been demonstrated to slow the progress of ALS; it improves survival rates by an average of two months (Miller, Mitchell, Lyon, & Moore, 2003). Interventions such as non-invasive positive pressure ventilation (NIPPV) have also been shown to improve survival (Chiò et al., 2009).

Management of ALS relies on symptomatic control and relief (Hobson et al., 2016; Moore et al., 2008) of progressively worsening and complex clinical needs (Corcia & Meininger, 2008). However, ALS symptoms are phenotypically heterogeneous, with individual differences in symptom onset, clinical motor and extra motor features, prognosis and progression being the norm (Swinnen & Robberecht, 2014). Symptoms include spasticity, fasciculations, pain, pseudobulbar affect, anxiety and depression, respiratory insufficiency, problems of the upper and lower gastrointestinal tracts, atrophy and weight loss (Jackson, McVey, Rudnicki, Dimachkie, & Barohn, 2015), as well as heterogenous forms of cognitive impairments (Montuschi et al., 2015; Phukan et al., 2012).

Clinical management of this challenging symptomology requires a multidisciplinary and comprehensive approach (Radunović, Mitsumoto, & Leigh, 2007). Multidisciplinary care may

also provide the support necessary for optimal decision making among patients and caregivers (Hogden, Foley, Henderson, James, & Aoun, 2017). Early discussion of difficult subjects between patients and health care providers, such as end of life and respiratory interventions, allows for preparation and planning in anticipation of change (Andersen et al., 2012; Munroe et al., 2007) prior to the manifestation of debilitating symptoms. It is therefore crucial that ALS patients and caregivers be provided with pertinent information and are communicating effectively with their healthcare providers in anticipation of loss and physical decline.

Loss and ALS

The progressive physical and functional decline typical of ALS (Hobson et al., 2016; Talbot, 2004) is connected with feelings of unremitting loss among patients (Foley, Timonen, & Hardiman, 2014). This experience of loss is multidimensional and includes physical ability, personal independence, and the fulfillment of social roles (Sakellariou, Boniface, & Brown, 2013). In particular, the loss of autonomy and the ability to communicate is a primary concern among ALS patients and has a profound impact on decision making to initiate life-sustaining interventions like ventilatory support (Lemoignan & Ells, 2010).

Physical deterioration impacts the self-esteem and sense of control among ALS patients (King, Duke, & O'Connor 2009). Physical limitations that prevent participation in important activities, alongside occupational restrictions, have a profound impact on personal identity and the fulfillment of social roles (Brott, Hocking, & Paddy, 2007). Indeed, the loss associated with ALS impacts some of the most intimate domains of personhood, including sense of self, social identity, and expectations for the future (Brown & Addington-Hall, 2008; Locock, Ziebland, & Dumelow, 2009).

Among caregivers, day to day and home care requirements for ALS patients increase as physical capacity declines. These responsibilities commonly become all-consuming for caregivers, leading to increased isolation and a loss of independence (O'Brien, Whitehead, Jack, & Mitchell, 2012).

Uncertainty and ALS

ALS patients have described fear of the unknown, particularly as it relates to the future they feel they have lost due to their illness (Hugel, Grundy, Rigby, & Young, 2006). A fear of the unknown is a "lower order" fundamental form of fear from which commonplace "higher

order fears" (e.g., a fear of heights) are logically derived (Carleton, 2016a). Unknowns refer to "the perceived absence of information at any level of consciousness" (Carleton, 2016b; p.31). Fear of the unknown is not surprising, because newly diagnosed patients and caregivers often know very little about their illness (Abdulla et al., 2014; Chiò et al., 2008; Davis & Turner, 2010). A perceived absence of relevant information, the experience of uncertainty, and related distress have all been well documented among patients and caregivers within the ALS literature (Galvin, Gaffney, Corr, Mays, & Hardiman, 2017; Hughes, Sinha, Higginson, Down, & Leigh, 2005; Lemoignan & Ells, 2010; O'Brien, Whitehead, Jack, & Mitchell, 2011; J. Oh & Kim, 2017; Pavey, Allen-Collinson, & Pavey, 2013).

Uncertainty among ALS patients may manifest due to emerging symptoms, delayed diagnosis, and testing (Galvin et al., 2017; O'Brien et al., 2011; Pavey et al., 2013), in addition to concern for the future, variable progression, loss of independence, and fear of the unknown (Hugel et al., 2006). Uncertainties and fear have also been documented concerning practical matters such as the anticipation of future disability, proximal cause of death and choice around major interventions, such as assisted ventilation (Hughes et al., 2005; Lemoignan & Ells, 2010).

It is likely that the pervasive experience of uncertainty is at least partially due to individual differences in ALS progression, functional decline, and disease trajectory (Swinnen & Robberecht, 2014).

Hope and Control

While not synonymous, hope and control within the ALS context are conceptually related. No single definition currently exists for hope, however, the term broadly refers to having a positive orientation toward one's future (Sachs, Kolva, Pessin, Rosenfeld, & Breitbart, 2013; Sullivan, 2003). Oster and Pagnini (2012) found hope to be high among samples of ALS patients and correlated with an increased desire for continued life. Hope in ALS patients may be tied to finding meaning in one's life, rather than being dependent on continued survival alone (Centers, 2001). Finding meaning in terminal illness may depend on "a shift in individual priorities and goals", with the pursuit of these goals facilitating the creation of new forms of hope when survival is no longer a likely option (Sachs et al., 2013; p. 122). The shift in priorities may require that patients replace expectations of survival with the achievement of more realistic goals like comfort, dignity, intimacy, and salvation (Sullivan, 2003).

The opposite of hope, hopelessness, has been defined as a "way of thinking in which negative expectations about the future are pervasive" (Beck, Weissman, Lester, & Trexler, 1974; Ganzini, Silveira, & Johnston, 2002, p. 315). Hopelessness may engender feelings of entrapment during times of crisis, such as being diagnosed with a terminal illness; it may result in behavioral responses that involve little, ineffective or inappropriate action (see Plahuta et al., 2002 for discussion). Hopelessness in ALS patients is associated with a sense of lack of control over one's health and a lack of purpose in life (Plahuta et al., 2002).

With increasing disability, ALS patients perceive a shift of control to powerful others (for instance, physicians) (Goldstein, Atkins, & Leigh, 2003). However, the wide-ranging loss experienced by ALS patients prompts some to find different ways to re-exert control (Foley et al., 2014; King et al., 2009), while accounting for their level of disability (Lee et al., 2001). This regaining of a sense of control is an important coping strategy among ALS patients (King et al., 2009) and is a predictor of well-being (Real, Dickhaus, Ludolph, Hautzinger, & Kubler, 2014). As a result, information seeking may be a useful coping strategy among ALS patients (Larsson, Nordin, & Nygren, 2016; Tramonti, Bongioanni, Fanciullacci, & Rossi, 2012) that enables a reassertion of control and the provision of hope (Fanos, Gelinas, Foster, Postone, & Miller, 2008). Information seeking may enable patients to play a more active role in their care (Broom, 2005).

Information needs and clinical communications

ALS patients and caregivers desire disease-related information at diagnosis, and insufficient information may cause concern and distress (Bolmsjo & Hermeren, 2001; Chiò et al., 2008; Hughes et al., 2005; O'Brien et al., 2011). Beyond diagnosis, people facing terminal illness have high levels of information need about prognosis and symptom management at all stages of their disease, including end of life (Parker et al., 2007). Indeed, Silverstein et al., (1991) found that the overwhelming majority of ALS patients in their study wanted "as much detail as possible" about their illness. Specifically, ALS patients are interested in information about their symptoms and how their illness will progress (Ang et al., 2015; Bolmsjo & Hermeren, 2001; Silverstein et al., 1991) as well as about home supports and care over the disease course (Oh & Kim, 2017). Patients require information on respiratory and communication equipment, life-sustaining treatment, ventilator care (Silverstein et al., 1991), and about the psychological

impacts of ALS, particularly in the areas of cognitive dysfunction and emotional lability (Wicks & Frost, 2008). Further, patients and caregivers are interested in information on ALS research and therapies (Abdulla et al., 2014; Chiò et al., 2008), because these topics provide hope (Fanos et al., 2008).

Caregivers express information needs on the provision of day to day management and physical care for patients at home (Larsson, Fröjd, Nordin, & Nygren, 2015; O'Brien et al., 2012), as well as on options for assistive equipment, public resources, and programs, including how to access them (Bolmsjö & Hermérn, 2003; Cipolletta & Amicucci, 2015; O'Brien et al., 2012; Williams, Donnelly, Holmlund, & Battaglia, 2008).

Given this diversity of information needs, clinicians need to engage in empathetic communication practices that provide encouragement and are respectful of individual patient differences that continue throughout the course of patient care (Abdulla et al., 2014; Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008). The effective delivery of a diagnosis may set the tone of the therapeutic relationship (Chiò & Borasio, 2004), and further clinical encounters need to appropriately pace information about progression (Dawson & Kristjanson, 2003; Hughes et al., 2005). The emotional sensitivity of ALS communications makes the delivery of the right information at the right time a major consideration in the delivery of quality care (Goodyear-Smith, 2005).

ALS diagnostic guidelines, such as the European Federation of Neurological Societies (EFNS), make recommendations for breaking an ALS diagnosis. Most recommendations promote a clear, empathetic, and honest approach to the disclosure of a 'bad news' diagnosis that preserves hope without providing false assurance (Andersen et al., 2012; Chiò & Borasio, 2004). Indeed, patients and their caregivers prefer discussion of hopeful topics, such as research, clinical trials, disease modifying therapies and topics that reduce uncertainty, such as future symptom management and disease outcome (Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008; McCluskey, Casarett, & Siderowf, 2004).

The devastating and heterogeneous nature of the illness makes clinical communication very challenging for all parties involved (Aoun, Breen, Edis, et al., 2016; Aoun, O'Brien, Breen, & O'Connor, 2018), with clinical encounters unlikely to meet all patient and caregiver information needs. ALS patients therefore engage in an array of information seeking behaviors

and strategies to fit their preferences (O'Brien, 2004), both before and after diagnosis (Abdulla et al., 2014; Chiò et al., 2008). Information seeking may be a response to the well-established experience of low information and high uncertainty that is common among ALS patients during these phases of care (Hughes et al., 2005; Lemoignan & Ells, 2010; O'Brien et al., 2011; Pavey et al., 2013). However, problems with the timing and content of information may be a potential driver for information seeking due to its importance when anticipating future change and preparing for clinical decision making (Hughes et al., 2005).

End of life Information Needs and medical aid in dying

Like other patients diagnosed with a terminal illness (Parker et al., 2007), ALS patients face complex decision making pressures and choices about end of life care. However, the progressive nature of their illness results in discussions with the clinical care team about end of life treatment preferences often being delayed (Munroe et al., 2007). It is therefore of interest whether ALS patients and their caregivers seek out information on end-of-life options outside of their clinical encounters.

Adding to the complexity of end of life communications is the recent availability of Medical Assitance in Dying (MAiD) in Canada. In *Carter v. Canada*, (2015), the Supreme Court of Canada ruled that the Criminal Code (1985) prohibitions on assisted suicide (s. 14 and s. 241) violated of s.7 of the *Canadian Charter of Rights and Freedoms* (1982). The case was brought by Kaye Carter and Gloria Taylor, the latter of whom had ALS. The Government of Canada responded with Bill C-14 (2016), *An Act to amend the Criminal Code and to make related amendments to other Acts (medical assistance in dying)*, that amended the *Criminal Code of Canada* to permit MAiD under certain conditions. MAiD is defined as **"(a)** the administering by a medical practitioner or nurse practitioner of a substance to a person, at their request, that causes their death; or **(b)** the prescribing or providing by a medical practitioner or nurse practitioner of a substance to a person, at their request, so that they may self-administer the substance and in doing so cause their own death." (Criminal Code of Canada, s. 241.1).

Those eligible for MAiD must be competent adults, covered by a provincial medical insurance plan, who are suffering from "a grievous and irremediable medical condition". Individuals seeking MAiD must have made a voluntary request, in the absence of external pressure or coercion, and have been given informed consent that includes information related to

palliative care options (Criminal Code of Canada, s. 241.2(1)). “Grievous and irremediable condition” is considered an illness that is serious and incurable, is in an advanced state and of irreversible decline, causes enduring and intractable physical and psychological suffering that is individually intolerable, and in which death is reasonably foreseeable (*Criminal Code of Canada*, s. 241.2(2)). In response to the change in Canadian criminal law, provinces have independently developed their own infrastructure for referral and delivery of MAiD within each of their own health care systems. Of the 1, 961 Canadians that completed a medically assisted death during 2017, 10-13% suffered from neurodegenerative disease (Government of Canada, 2018).

Thesis outline

This thesis is paper-based, meaning that there will be slight repetition between chapters, mostly in the use of literature and the explanation of research methods.

Chapter 2 provides additional details about my methods. It explains my epistemological and ontological assumptions, both of which shaped the design of this research and my interpretation of the data. I further provide a personal statement that reflects on the biases and preconceptions that may had influenced my qualitative research.

Chapter 2 also specifically outlines my sampling and recruitment methods, my semi-structured interview guides, the accommodations necessary to modify my interview methods to address the specific needs of the research participants, and the approach I used when analyzing these data. I then discuss the ethical considerations that informed the design of my research, how I enabled participants to provide informed consent and how they were protected and supported during and after their participation. The chapter ends with a discussion of study limitations.

Chapter 3, “Information Seeking Behavior and Communication Preferences Among ALS Stakeholders During Diagnosis”, is the first empirical research paper. In Chapter 3, I present my qualitative study where I analyze the experiences and preferences of patients, caregivers, and providers in receiving or providing an ALS diagnosis. I further present my analysis of patient and caregiver motivations for seeking information outside of clinical encounters, the information preferences of patients and their caregivers about disease progression and prognosis, and the extent to which these are accounted for by providers.

Chapter 4, “Information Sources and Use Among ALS Patients and Caregivers”, is the second empirical research paper. In Chapter 4, I address the following aims: 1) to understand the motivations of individuals living with ALS and their caregivers for seeking ALS-related information outside of clinical encounters; 2) to identify the external information sources and topics of interest; and 3) to analyze the value of this information to individuals living with ALS and their caregivers.

Finally, in Chapter 5, I conclude my thesis and provide a set of recommendations for providers on communicating about external information with patients and caregivers. I also argue for the value of patient support groups as an important source of information exchange and support. Recommendations related to the promotion of attendance and conduct are provided. The chapter concludes with suggestions for future research.

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CHAPTER TWO: METHODS

INTRODUCTION

In this Chapter, I outline my research related activities and methods. I first outline my philosophical assumptions. In this section I describe my epistemological & ontological claims to provide the reader with an understanding of how they influenced the approach and conduct of my work. Second, I describe my target population, its location, and how I screened and recruited study participants. Third, I describe the methods I used to conduct interviews, analyze collected data, and maintain study rigor. Fourth, I offer a discussion on the ethical considerations of studying this population and provide a description of how the needs of patients and caregivers were accommodated and how they were protected as a vulnerable population. Finally, I discuss the limitations of my exploratory study.

PHILOSOPHICAL FOUNDATIONS

The strength of my research design is partially grounded in my choice of assumptions and research paradigm, both of which are congruent with my views on the "nature of reality" (Mills, Bonner, & Francis, 2006, p. 26). The ontological assumptions used for this project mostly (but not completely) fall under those within both the positivist (realism) and post-positivist (critical realism) tradition. I agree with the 'realism' ontological assumption that reality exists and that it is independent of the human mind and reject the critical realist ontological notion of a separately existing social reality (Annells, 1996; Denzin, 1994). My methods and work assume a single reality. Similar to the critical realist ontology, I accept that universal truths and laws exist, but only partially accept the notion that their discovery is, as a general rule, "near impossible" (Levers, 2013, p. 3). I also reject the post-positivist notion that there are useful facts outside what can be "physically observed" (Crossan, 2003, p. 53). This work assumes that all observation occurs within reality, including the capture of recollected data comprised of past experiences.

In general, I agree that inquiry, and qualitative inquiry in particular, should focus on the observation of the results and outcomes of causal forces, rather than the forces in and of themselves (Clark, MacIntyre, & Cruickshank, 2007; Levers, 2013). In this vein, it makes sense that the goal of research is, more often than not, the incremental movement towards knowledge

through the observation and discovery of "partial segments or approximations of" the truth (Clark, 1998; Levers, 2013). Thus, I posit that claims to truth should be approached through the use of reason *in combination* with observation. This position differs slightly from the critical realist position which favors the use of 'reductive reasoning' when generating casual explanations (Clark, MacIntyre, & Cruickshank, 2007; Levers, 2013; McEvoy & Richards, 2003). I also hold that every effort should be made to fully integrate newly discovered facts, or 'approximations of truth', into larger preexisting systems of knowledge. The combined use of observing relevant findings and using inductive reasoning to explain and integrate them into existing knowledge is what guided my approach to inquiry within this exploratory study.

The positivist epistemological influences in my work assume that all existent facts reside in a reality that can be apprehended due to its governance by natural law (Denzin, 1994). I also assume that inquiry occurs in the real world and that the any method of inquiry exists separately and independently from the inquirer (Annells, 1996; Levers, 2013; McEvoy & Richards, 2003). Both these assumptions, including those outlined further above, have impacted my research in a few important ways. First, it allows for individual conceptions of loss and value to be directly connected with the natural laws that govern an objective reality that is shared by all participants. These natural laws include everything from the neurodegenerative pathology of ALS to the forces of gravity that further confine and limit the mobility of ALS patients. Second, this assumption also allows for a clearer connection between the individual experience of uncertainty and the absence of needed ALS related information. This connection assumes that information is an existent material entity that can be sought out and consumed. Lastly, the separation between the method of inquiry and the inquirer assumes that any knowledge product is the result of my analysis of recollected experiences as told by individual participants. Thus, the knowledge manufactured by my work is the product of my analysis of participant experiences alone, not a "co-creation" between myself and participants.

PERSONAL STATEMENT

As of September 2018, I have spent a decade studying in health-related fields and later engaged in the provision of clinical care. I graduated in April 2012 with a Bachelor of Science and Nursing from York University and worked as a Registered Nurse from August 2012 to August 2015 at Kingston General Hospital. During that time, I was involved in palliative and

end of life care and supported numerous patients and families as they came to terms with a terminal diagnosis and transition into the final phase of life. I have always considered this work to be a privilege. I similarly consider my participation in this research and the completion of this thesis as an extension of this privilege.

My approach to this research was undoubtedly influenced by my nursing training and clinical experience. My professional experience delivering palliative and end-of-life care provided me with some obvious advantages alongside some more subtle biases. One advantage of this clinical experience was my understanding and practical comfort when discussing sensitive subjects, such as end-of-life, with my study participants. My clinical practice also provided me with the interviewing skills required to dig deeply into the histories, experiences, emotions, views, and opinions of participants. My training also provided me with the ability to appropriately pace conversations when they became too emotionally difficult, while still addressing sensitive issues. Skillful pressing on some sensitive topics ensured that the views and thoughts of my participants were more fully captured, allowing for a more robust and accurate understanding and representation of their lived experiences.

These advantages also came with some biases, the most obvious of which would be my overall clinical approach to my study. I was less interested in building theory than in conducting patient centered and clinically oriented research that gave rise to actionable recommendations. My practical intent likely influenced the direction of interviews, my probing on specific themes, and the development of my recommendations. As a result, my thesis does not build a comprehensive theory on ALS communications, but instead offers recommendations intended to improve the clinical interactions and day to day lives of ALS stakeholders.

Another source of bias comes from the fact that I have never lost a close family member, experienced poor health, or been disabled. Because of this, there were times where I struggled to appreciate the now obvious importance of some parts of my data. My initial non-understanding of the importance of experiential information about practical day to day management and assistive equipment is a good example of this. It can be hard to understand the value of information about pillows and handicap friendly venues when you can physically support yourself, are not in pain, and have the liberty of unhindered movement. It seems strange to consider going to the washroom as an overwhelming ordeal. But if you can understand how

difficult this task is for patients with ALS and their caregivers, the value of equipment information that makes this ordeal bearable, or no longer required, becomes instantly obvious. These examples illustrate a particularly difficult struggle for me, especially at the beginning of this project when I was only beginning to fully appreciate the depth of my data. To manage the bias created by this experiential gap, I made sure to document my observations, impressions, and thinking over the course of this project. My consistent practice of personal introspection was informed by these observations. The insights generated from this reflection were very helpful in managing my personal bias and added to the development of the ideas and concepts central to my analysis.

RESEARCH METHODS

Sampling and Recruitment

ALS management frequently occurs within a triad of care, consisting of the patient, a caregiver, and a provider. Because of this, my supervisory committee and I agreed that inclusion of these groups was required to elicit a full understanding of ALS communications. I based my recruitment of participants on similar procedures and criteria used in other studies and via purposive sampling (Albert et al., 2005; Chiò et al., 2008; Stutzki et al., 2014).

The guiding principle used in determining the theoretical sample size in qualitative research is *data saturation*, which occurs when data no longer generates novel theoretical insights or core properties of the categories you have created (Charmaz, 2006). Based on previous work, we estimated that fifteen to twenty participants in each category would be a reasonable recruitment limit for our sample. However, because my study was exploratory, I did not base my sample size on reaching data saturation.

Similar to other qualitative studies (Bolmsjo & Hermeren, 2001; Bolmsjö & Hermérn, 2003; Burchardi, Rauprich, Hecht, Beck, & Vollmann, 2005; Foley, Timonen, & Hardiman, 2014; Hogden, Greenfield, Nugus, & Kiernan, 2012), I anticipated interviewing 5-10 patients; 5-10 caregivers (not necessarily patient/caregiver dyads); 5 specialists (neurologists and palliative care specialists) who diagnose and/or care for patients with ALS; 5 general practitioners (GPs) who currently or had cared for patients with ALS; and 5 nurses who had cared for patients with

ALS. However, I did not recruit any general practitioners and only recruited one palliative care specialists working in the community. See Appendix 1 for recruitment criteria.

Using a purposive snowball sampling approach, I recruited 12 ALS patients and 10 caregivers from the ALS Clinic, located in the Kaye Edmonton Clinic, Alberta, Canada. I also contacted patients and caregivers who had previously consented to participate in research. Eligible patients had to be over the age of 18 and mentally competent. The clinic coordinator confirmed competence and intact cognitive status before recruitment. Caregivers eligible for participation had to be over the age of 18 and have current or previous experience providing informal care to someone affected by ALS. Health care providers eligible for participation were over the age of 18 and had professional clinical experience caring for ALS patients. Among this group, I recruited nurses (n=5), neurologists (n=7), and primary care providers (n=1). Recruited neurologists came from three Canadian provinces, Alberta, Quebec, and Saskatchewan.

I employed a two-step process when recruiting patients and caregivers into my study. The first step involved approaching patients and caregivers during their clinic visits, briefly introducing the study, providing participants with condensed literature explaining the study (see Appendix 2), and determining the preferred method and direction of further contact. The second stage of recruitment involved a second appointment, usually within the homes of participants. During this second encounter, I provided participants with a lengthy and through explanation of my project. I provided a more comprehensive package of study information (see Appendix 3) (which was sometimes emailed in advance depending on preference) and gave them ample time to ask questions about their participation and the study at large. This was often followed by the provision of written informed consent (see Appendix 4).

After the provision of written and informed consent, I administered a set of surveys to both patients and caregivers (see Appendix 5). Two of these surveys were designed to screen patients and caregivers for depression. I administered The ALS Depression Inventory (ALS-DI) to patients and the Beck Depression Inventory (BDI) to Caregivers (Beck & Beamesderfer, 1974; Kübler, Winter, Kaiser, Birbaumer, & Hautzinger, 2005). No caregiver scored greater than 20 (20-28 indicates moderate depression; 29-63 indicates severe depression) on the BDI, and therefore none were excluded from the study. However, if they had scored over 20, I would have notified the caregivers' family physician.

ALS patients who scored over 23 on the ALS-DI were not eligible to participate. A score of or >23 was taken as suggestive of depressive symptoms, the need for referral, and more thorough evaluation. When a patient scored over 23 on the ALS-DI, I notified Dr. Wendy Johnston, Director of the ALS Clinic, of the result. I did not continue to interview five patients who scored over 23 on the ALS-DI. If a participant had a score that suggested depressive symptoms, Dr. Johnston either managed their psychiatric care at the ALS clinic, contacted their family physician (as per the consent form), or offered a referral to Dr. Anne Maccocia, the clinical psychologist for the neurosciences program.

DATA COLLECTION

Instruments

Patients completed the ALS-DI, a previously validated 12-item survey designed to detect depressive symptoms in ALS patients by eliminating questions related to "somatic or motor related symptoms" while accounting for "progressive physical impairment" (Hammer, Häcker, Hautzinger, Meyer, & Kübler, 2008, p. 214; Kübler et al., 2005). Caregivers completed the BDI, a widely used 21-item survey that assesses for the presence and intensity of depressive symptoms in both clinical and general populations (Beck, Steer, & Carbin, 1988). Patients also completed: 1) the Hearth Hope Index, a 12-item Likert survey that is capable of assessing 'hope' in a clinical setting (Herth, 1992); 2) The Edmonton Symptom Assessment Scale, a 10-item survey designed to assess symptom distress among palliative populations (Bruera, Kuehn, Miller, Selmser, & Macmillan, 1991); and 3) The Patient Dignity Inventory, a 25-item survey designed to assess end-of-life dignity-related distress (Chochinov et al., 2008). These were all important tools for understanding the current psychological and physical state of participants (see Appendix 5).

Semi-Structured Interviews

Similar to previous work (Bolmsjo & Hermeren, 2001; Bolmsjö & Hermérn, 2003; Burchardi et al., 2005; Foley et al., 2014; Hogden, Greenfield, et al., 2012; Hogden, Kiernan, & Greenfield, 2012), I conducted semi-structured interviews (Charmaz, 2006; Denzin, 1994) with ALS patients, caregivers, and healthcare providers. I developed separate guides for each stakeholder group (see Appendix 6). All guides contained separate sections related to diagnosis, care, end of life, medical assistance in dying and information gathering. All guides used a semi-structured, open-ended approach. This use of a semi-structured guide allowed interviewees to

explore topics on their own while facilitating the natural emergence of unanticipated findings. In one instance involving profound verbal impairment, I conducted a joint interview with a caregiver and patient together. The caregiver provided interpretation on behalf of the patient, while supplementing the patient's comments with their own information regarding their care experience. This interview was counted as a single patient interview. I excluded the supplemental information from the caregiver in my analysis to the best of my ability. Participants set the pace and length of interviews. I also took care to make sure that participation was flexible enough to accommodate the needs and limitations of all participants.

Concerns about the feasibility of my data collection strategy were voiced by my supervisory committee and considered at the outset of this project. These concerns most often centered around the ability of patients and caregivers to emotionally tolerate subject matter around end of life and medical assistance in dying. A second concern involved the ability for patients to physically tolerate these interviews, given the breadth of topics I intended to explore. In total, my participants provided 74 hours 13 minutes and 37 seconds of digital audio recording. Interviews lasted between 31 minutes to 4 hours and eight minutes. Interviews with providers were usually completed in a single session and on average lasted for one and a half hours. In contrast to providers, interviews with patients and caregivers were frequently completed over multiple sessions as required. This flexibility was key to the feasibility of my data collection strategy.

In contrast to the initial concerns, patients and caregivers tolerated these interviews remarkably well. The length of patient interviews (12) ranged from one to 4.13 hours and lasted an average of 2.4 hours. Caregiver interviews (10) ranged from nearly 1.73 to 3.16 hours and lasted an average of 2.5 hours.

Emotional upset occurred often among participants and at times required short breaks from interviewing. Episodes of emotional upset occurred most frequently among patients. During episodes of emotional upset, I reminded participants that they could stop the interview and skip questions. One patient blamed their emotional upset on "the syndrome". Pseudobulbar affect (PBA), or "uncontrollable episodes of laughter or crying that are excessive for or incongruent with the underlying emotion and situation" is prevalent among ALS patients (Thakore & Pioro, 2017, p. 1). While I was not competent to identify which patient participants

may have exhibited symptoms of PBA, I made a conscious effort to account for its potential impact in setting the pace of the interview. Only one patient requested to skip over content related to end of life and medical assistance in dying. All other participants completed the interview and were often pleased with the discussion. One participant did not fully complete an interview due to physical decline.

In summary, patients and caregivers tolerated discussions of topics related to disease history, diagnosis and care experience, end of life, medical assistance in dying and information gathering. While at times difficult, some participants expressed satisfaction with the interview and offered a wealth of rich experiential data. Given my experience in this exploratory study, an interview-based expanded study, if conducted with caution and tact, would be both feasible and likely to generate valuable data.

DATA ANALYSIS

Once interviews were completed, I used a professional service to transcribe the audio files of my interviews. I verified completed transcripts against audio recordings to ensure their accuracy. I then imported verified transcripts into N-Vivo 11^{TM} qualitative software for analysis. Interviews were first 'chunked' into parts that corresponded with thematic sections of the semi-structured guide. These included descriptions of diagnosis, care, end of life, medical assistance in dying and information gathering. I then analyzed transcripts using an incident to incident (open coding) approach (Charmaz, 2006; Lawrence & Tar, 2013). I coded individual transcripts using the constant comparison method (Glaser & Strauss, 1967), which involved an iterative reanalysis of transcript data as new concepts emerged (Charmaz, 2006). I discussed these emerging themes on a regular basis with my supervisor, Dr. Wendy Johnston and her research associate, Ms Westerly Luth. These meetings allowed for a consensus to develop around the validity and coherence between certain codes (e.g., "openness" and "honesty") and concepts (e.g., "good communication") which eventually developed into broader themes (e.g., "good diagnosis experience").

Member Checking

To ensure the rigor of my data, I engaged in member checking. I offered interviewees the opportunity to comment on summaries or entire copies of their individual transcripts (Charmaz,

2006). Depending on the preference of the participant, member checking occurred both in person and via email. When done in person, I reviewed summaries or full transcripts together with the participant. Via email, I provided participants either a full copy or summary of their transcript and asked them to comment.

Comments most often involved minor corrections to the text or, occasionally, clarification of concepts and unclear statements. One caregiver participant commented that they hoped they did not come off as too "negative" and, after reading our conversation, described being more encouraged to now read an ALS manual that was provided to them. They stated:

> *There was one correction on my part. I declared the clinics didn't give us much. Sitting here, I see we have a binder labeled, "A Manual for People Living with A.L.S."... Needless to say, I've not previously opened it, but now have my curiosity going. Making a point of seeing what it entails.*

In another instance of member checking, I asked a patient participant to clarify a statement they made about their diagnostic experience, specifically what they in retrospect would have liked to have been told. They clarified by stating:

> *As I travel through this ALS journey I suppose there will always be new items of discussion that may arise. I don't think it is possible to capture everything at the onset of the disease because everyone's journey is unique.*

Follow up conversations like these greatly enriched my analysis. First, follow ups provided a welcome form of guidance and confirmation that I was on the right "conceptual track" as I shaped the direction of the project together with Dr. Johnston and Ms Luth. Second, these assured me that I was accurately representing the experiences, values, and opinions of my participants. Third, given the complexity and richness of the data, member checking also provided some necessary clarifications. Finally, engaging in the lengthy process of writing summaries improved how I thought of my project, my data and the stories of my participants.

Transferability

Distinct conceptions and efforts at achieving rigor, validity, reliability and quality are clear when comparing qualitative and quantitative approaches to research (Golafshani, 2003). Considering this distinction, generalizability may be an inappropriate criterion when attempting

to judge the validity and quality of qualitative research, the focus of which is to examine "a specific issue… in a certain population… of a focused locality… [and] in a particular context" (Leung, 2015, p. 326). Transferability, or the degree to which qualitative findings may be transferred to other contexts, is an important consideration when judging the trustworthiness of qualitative research (Korstjens & Moser, 2018). In this thesis, I have attempted to provide a detailed description of the experiences of my participants and of my research process, allowing readers and other researchers to judge their applicability in other contexts, including their own.

ETHICAL CONSIDERATIONS

RECRUITMENT

Patients and Caregivers and Health Care Professionals

My recruitment process explicitly removed the attending physician of the ALS Clinic, Dr. Johnston, from any related activities. Recruitment efforts mainly took place in the ALS Clinic, Kaye Edmonton Clinic, Edmonton, Alberta. First, I worked with the clinic coordinator to identify patients that fit my study criteria. The clinic coordinator and I used the weekly care roster during clinic days to identify who was eligible. The clinic coordinator approached potential participants to gauge if they had any interest in hearing about the research currently available at the clinic. If patients and caregivers expressed interest in participating, I approached them and provided them with an information sheet outlining what the research project was about, the expectations of participation, what their rights would be as a participant, how I would protect their confidentiality, what the outputs of the research would be, and how I would present my findings (see Appendix 2). Next, I asked patients and caregivers if they had a preferred method of contact, if I could initiate contact, and when would be an appropriate time to contact them to book a screening appointment would be. I informed potential participants that the provision of consent and participation in the study was entirely voluntary and would not affect their care in any way.

In addition to direct contact, I used recruitment posters with study contact information posted at the ALS Clinic at Kaye Edmonton Clinic, Edmonton, Alberta, Canada (see Appendix 7). Dr. Johnston contacted colleagues, specialists, and nurses who currently provided care to

ALS patients, or had done so in the past. She identified physicians and nurses who met the study criteria. I recruited these suggested participants through a mix of convenience and snowball sampling methods. Depending on the mode of introduction, I communicated with specialists, GPs and nurses by either email or phone.

Screening and Informed Consent

After reading the study information (see Appendix 3) (or going over it with the participants), I encouraged participants to ask questions about the study and articulate any concerns they had. I also informed them that they could withdraw from the study at any point and refuse to answer questions. I explained that they had 3 months after their interview to withdraw their responses from the study. None did. To accommodate for level of disability, participants could provide either written or oral consent. I informed participants that the consent process and interview could take place in as many sessions as required for completion. I also told participants in advance about the screening process that would begin immediately after providing consent. I informed participants that Dr. Johnston or I would notify their family physician if their results on the ALS-DI or the BDI were indicative of depressive symptoms. I did not similarly screen providers, and most provided consent prior to participating in the study. Two physicians provided consent and completed interviews over the phone at a later date (see Appendix 3 and 4).

Participant Vulnerability

Depression, suicidality, cognitive impairment and frontotemporal dementia are associated with ALS (Fang et al., 2008; McLeod & Clarke, 2007; Miller et al., 2009). As a vulnerable population, my study included several inclusion and exclusion criteria that were intended to make sure that participants were not unintentionally harmed or coerced into participation (see Appendix 1). Recruited participants had to meet our basic inclusion and exclusion criteria, provide the contact information of their primary care provider, and consent to allowing researchers to contact their healthcare provider if they displayed signs of depression or suicidality over the course of the study (see Appendix 4).

I interviewed patients and caregivers about their experiences with ALS. Covering this subject matter resulted in the discussion of topics that were emotionally challenging for some participants. Despite this, my clinical experience as a registered nurse allowed me to pace

difficult conversations appropriately. Using this training, I made sure not to push participants beyond their emotional limits and did not include sensitive topics of discussion when required or requested. I also warned participants in advance about question topics so they could emotionally prepare themselves before participating in the interview.

Confidentiality

Given the relative rarity of ALS, the target population within Alberta was small, potentially making participants identifiable. This made participant confidentiality a significant concern. I anonymized all data before use in presentations and this thesis. I gave each participant a pseudonym. I kept a master list linking participants to descriptors in a locked filing cabinet within a research space at the School of Public Health – located at 3-093 at Edmonton Clinic Health Academy. I stored transcripts and email interviews on a secure and password protected School of Public Health terminal.

LIMITATIONS

This research was limited to ALS and did not include the study of other devastating motor neuron conditions with slightly different forms of clinical presentation, affected groups, and lethality (primary lateral sclerosis, spinal muscular atrophy, Kennedy's disease). Given that this was also an exploratory project, its scope was limited to patients and caregivers receiving treatment within the province of Alberta, Canada.

My study also did not include other allied healthcare professionals, who are involved in the delivery of multidisciplinary care, such as speech-language pathologists, dieticians, and respiratory technicians.

Given the burden of ALS in daily life, participants who agreed to participate in my study were likely to view research endeavors in a favorable light. This frame of reference may be partially due to the previous academic training of participants, their respect for knowledge production, or their coping successes. The screening process we engaged in using the ALS-DI and the BDI likely further selected for participants who had a greater sense of control and a more positive outlook on life.

Given the primarily qualitative methods of data collection and analysis, the results of the study are primarily geared towards discovery – not generalizability. It is likely, however, that the

methods and findings in this work are transferrable to other similar contexts. Given that my data depended on the recollection of long, complicated, and emotionally charged experiences, recall bias among participants was also likely. Further, given that the experience of patients, caregivers, and providers may be significantly affected by the environment in which healthcare is delivered, the experience of similar stakeholders is likely to differ outside Alberta. This use of a single site is likely to limit the overall generalizability of our results.

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CHAPTER 3: INFORMATION SEEKING BEHAVIOR AND COMMUNICATION PREFERENCES AMONG ALS STAKEHOLDERS DURING DIAGNOSIS

INTRODUCTION

Amyotrophic Lateral Sclerosis (ALS) is a degenerative motor neuron disease that causes near total paralysis of skeletal muscles (Hobson, Harwood, McDermott, & Shaw, 2016). ALS is incurable and nearly always fatal, with a 50% mortality rate within three years of symptom onset (Mitchell & Borasio, 2007), highlighting the need for rapid and accurate diagnosis. The ALS diagnostic experience has been characterized as prolonged, emotionally distressing, and uncertain due to the emergence of unexplained symptoms, low disease awareness and frustrating interactions with healthcare providers (Galvin, Gaffney, Corr, Mays, & Hardiman, 2017; O'Brien, Whitehead, Jack, & Mitchell, 2011; Pavey, Allen-Collinson, & Pavey, 2013).

The symptoms and progression of ALS are variable and may cause profound disruption to the lives of patients and caregivers (Bury, 1982; Pavey et al., 2013). While a diagnosis improves clarity, the manner of its delivery may have an ongoing impact on the newly formed therapeutic relationship (Chiò & Borasio, 2004).

ALS diagnostic guidelines, such as the European Federation of Neurological Societies (EFNS), make recommendations for breaking an ALS diagnosis that include the provision of printed materials about the disease (Andersen et al., 2012). The guidelines promote a clear, empathetic, and honest approach to the disclosure of a 'bad news' diagnosis that preserves hope without providing false assurance (Chiò & Borasio, 2004). This desire to balance hope with honesty among ALS physicians has been found in the results of previous surveys (Aoun, Breen, Edis, et al., 2016). Indeed, patients and their caregivers prefer discussion of hopeful topics, such as research, clinical trials, disease modifying therapies and topics that reduce uncertainty, such as future symptom management and disease outcome (Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008; McCluskey, Casarett, & Siderowf, 2004).

Communications that address these values and preferences are associated with better patient and caregiver satisfaction (Abdulla et al., 2014; Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008; McCluskey et al., 2004) and should initially be based on the patient's understanding of their illness (Andersen et al., 2012; Baile et al., 2000). This starting point is likely to be influenced by external information sources, as ALS patients and caregivers seek out hopeful information and investigate sources of uncertainty, such as symptomology, clinical course, and outcomes (Abdulla et al., 2014; Chiò et al., 2008).

Accordingly, in this qualitative study I explore and analyze the experiences and preferences of patients, caregivers, and healthcare providers when receiving or providing an ALS diagnosis. I further explore and analyze patient and caregiver motivations for seeking information outside of clinical encounters, the information preferences of patients and their caregivers about disease progression and prognosis, and the extent to which these are accounted for by healthcare providers.

METHODS

Participants

I recruited ALS patients (n=12) and caregivers (n=10) (9 formed patient-caregiver dyads) being treated in Edmonton, Canada, using a purposive and snowball sampling method. Patients and caregivers eligible for participation were over the age of 18 and were without cognitive deficiencies, as determined by the clinic coordinator. Caregivers eligible for participation were currently or had previous experience providing informal care to someone affected by ALS. Provider participants comprised seven neurologists who specialized in ALS, one palliative care physician that worked in the community, and five nurses.

Clinic staff asked patients and caregivers if they were interested in research opportunities, and I approached those who were with verbal and written information about the study. For those who expressed interest in participating further, I scheduled a preliminary meeting to describe the study protocol and answer questions. I administered the ALS Depression Inventory (ALS DI) and the Beck Depression Scale (BDI) to consenting participants and excluded patients if they scored over 23 on the ALS DI and excluded caregivers if they had a score of 20 and above on the BDI. Five patients scored over 23 on the ALS DI, with no caregivers meeting the 20 and above

threshold of the BDI. I continued with the semi-structured interview with those participants who met the inclusion criteria and consented to participate in my study.

I also recruited healthcare providers (n=13) over the age of 18 using a purposive and snowball sampling method across three Canadian provinces, Alberta, Quebec and Saskatchewan. Of these, physicians had clinical experience with ALS patients, and were either ALS specialists or were primary care providers. Nurses had experience caring for ALS patients, usually on an outpatient basis.

Data Collection

I performed a limited chart review of patient health histories to collect demographic information and the time between reported symptom onset and the confirmation of the ALS diagnosis. I used a semi-structured interview guide that covered ALS diagnosis, care, advanced disease, medical assistance in dying, and information gathering behaviors (see Appendix 6). I chose this partially open-ended approach so that items of importance to participants could emerge during discussions (Charmaz, 2006; Denzin, 1994). I also created field notes immediately after completing each interview to capture initial impressions and emerging concepts, which were integrated into subsequent interviews and my analyses.

Qualitative Content Analysis

I verified verbatim transcripts of interviews against their corresponding digital recording to ensure accuracy. I imported transcripts into N-Vivo 11^{TM} qualitative software for analysis. I employed the use of open coding to identify themes (Charmaz, 2006; Lawrence & Tar, 2013), using a constant comparison method (Glaser & Strauss, 1967). This involved an iterative reanalysis of transcripts as new concepts emerged from the data (Charmaz, 2006). I discussed these emerging themes on a regular basis with two other members of my research team. These meetings allowed for consensus to develop around the validity and coherence of themes. I then used member checking to ensure the accuracy of the data and the validity of my analyses (Cho & Trent, 2006); I provided either interview summaries or full transcripts back to participants for comment. I then reviewed and cross checked my interpretation of responses with participants in person or via email. The few responses that were provided were then integrated into my final analysis.

Ethics

The study protocol was approved in August of 2016 by the Health Research Ethics Board-Health Panel at the University of Alberta, Canada (see Appendix 8).

RESULTS

The majority of patients (10) and caregivers (8) had heard of ALS prior to receiving a diagnosis. Level of pre-diagnostic knowledge varied considerably between participants. Patients and caregivers made frequent reference to knowing about "Lou Gehrig's Disease". Six patients and four caregivers described some form of information seeking before receiving an ALS diagnosis. Patients spent 15 months on average obtaining a diagnosis, with a range from 5 to 20 months.

External Information Seeking Prior to Diagnosis

Patients and caregivers frequently described the uncertainty associated with the emergence of unexplained ALS symptoms, such as the need to "brush teeth with two hands" (Caregiver 2), and frustration with the protracted diagnostic experience (Table I, 1 and 2). Reactions to the onset of symptoms varied but were typically described as emotionally distressing and as a cause for uncertainty, except when recognized by those with a family history of ALS (Table I, 3).

Uncertainty and a knowledge deficit led some patients and caregivers to engage in information seeking pre-diagnosis. Some participants were motivated to clarify the clinical terms used by their physicians (Table I, 4). Other participants didn't think to look up information because they either recognized symptoms as ALS related (Table I, 3) or symptoms were too non-specific to prompt further knowledge seeking (Table I, 5). Family history of ALS, pop culture sources, and awareness campaigns like the "Ice Bucket Challenge" (a viral ALS awareness and research fund raising campaign that hit its peak in the summer of 2014) contributed to participants' passive understanding of ALS before the emergence of symptoms (Table I, 6 and 7). However, prior experience with ALS within a friendship network was not always helpful due to the variation in onset of symptoms (Table I, 8).

Providers described patients as ranging from knowing little to being very knowledgeable at the time of their ALS diagnosis (Table II, 1). Provision of a diagnosis to those with little knowledge was more challenging, because they were not prepared for the bad news (Table II, 2).

Communication of an ALS Diagnosis

Patients described positive diagnostic encounters as characterized by clarity, thoroughness, patience, sensitivity, support, openness, and honesty with the opportunity to ask questions (Table I, 9). Patients also appreciated efforts made by providers to make them more physically comfortable (Table I, 10). However, not all diagnostic experiences were positive. Participants described negative or sub-optimal experiences as brief, sometimes resulting in emotional shock (Table I, 11). Some patients described the distress associated with a terse delivery of their diagnosis, or outright dismissal (Table I, 12 and 13), whereas one patient was angered when he realized that his diagnosis was used as a teaching moment for medical students (Table I, 14). Another was upset when the physician ignored their family history of ALS (Table I, 15). Experienced providers described a stepwise method for delivering an ALS diagnosis that accounted for the current understanding of ALS as expressed by the patient (Table II, 3), including his/her information preferences and additional needs (Table II, 4). Physicians described a standard set of information provided to patients (Table II, 5), sometimes aided by diagrams and metaphor (Table II, 6). Providers also recognized the occasional need to provide accurate information when they detected misconceptions about clinically complex and emotionally sensitive topics (Table II, 7), or the potential for misconception because of information seeking by patients between appointments (Table II, 8). Providers recognized that the diagnostic encounter enabled patients to "move on" (Provider 13).

Table I. Patient and Caregiver Perspectives

Symptom onset and time to diagnosis
1. "[After struggling to get up stairs]. I'm thinking, okay, I'm really out of shape. I got to get better as time goes on, but it never happened. I was very frustrated [And I was left with] you know, guilt. What did I do that caused me to be here? What could I have done differently?....Why am I stumbling about and why am I not getting better?" *Patient 5*
2. "I expressed frustration [to my family doctor]. I think it was after the fact. Why did it have to take so long? Why didn't they believe me the first time I went and see them or something? She just kind of dismissed it and said, well, now you know, and it really didn't make any difference … Six months of not knowing, in her mind, was not a problem." *Patient 8*
3. "So, because we have multiple family members with ALS, it was never something that we didn't discuss, that we always knew that there was a possibility…Right from the beginning, I recognized the symptoms of her having ALS before she did, so we just had to be patient with that." *Caregiver 6*
Motivation for seeking further information on ALS
4. "[Neurologist] said there was motor neuropathy… So I googled that and there are so many forms but when you look under motor -- at least the document I looked at, ALS was down the way. Motor neuropathy could be just injured neurons." *Patient 5*
5. "I really didn't know what to be looking for. I mean he had a limp. I was thinking spinal problems or, you know, something like that." *Caregiver 11*
6. "You know, I knew it was Lou Gehrig's disease and I watched the movie on Lou Gehrig. But even in the movie, there wasn't a whole lot pertaining to this disease. It was more him as a baseball player. Right? And of course, the ice challenge." *Patient 10*
7. "I learned from the ice bucket challenge that my symptoms were similar. So, I started doing a little bit of my own research on Dr. Google…everything paralleled precisely what the onset symptoms would be for ALS." *Patient 11*
8. "I was really in denial. [Before diagnosis] I thought it was something else... [Because] We had a guy on our [sports] team, he passed away with ALS. I knew him quite well. So…. I thought I knew all the symptoms … But I didn't know there was all these different types of ALS." *Caregiver 7*
Characteristics of a positive diagnostic experience
9. "[ALS Neurologist] went over to all the findings…then said, yes, this is consistent with ALS … I came away from there thinking [they] explained it very well. I didn't feel that [they were] being cruel or unreasonable or cold about it at all. [It] was matter of fact… it felt like a compassionate place to be in that room with [them]… [They] didn't rush. [They] took [their] time. [They] gave me lots of opportunity to ask questions." *Patient 8*
10. "[They] was very helpful and for me even to get up on, when I get cold, of course, you're just about half-naked when you're trying to do the testing right on the ***bare skin*** and stuff eh? [They] made sure [They] turned up. You know, kept it warmer, helped me to get up and down off the table and up the stairs that she had in there." *Patient 6*

Characteristics of a negative diagnostic experience

11. "It was fairly abrupt like it was shocking actually to get that information within a few minutes…As I left the clinic, I was in a little bit of shock. … [the delivery] took probably less than one minute." *Patient 11*
12. "[The neurologist] just said, "It's not good." First of all, he said, "Well, there's been a change. Whatever" And then I go, "So that's not so bad?" He goes, "Oh no, this is not good news. You have um. You know. We feel it's neurological, neuromuscular disorder or illness, disease, and, you know, ALS." And I said, "So that's when I have – What, I have two or three years to live?" He goes, "Oh, it might be a little longer." Then I asked to see [ALS Specialist] and shook his hand…That was it, three to four minutes." *Patient 7*
13. "I mean by this time, I was crawling up the stairs to get up here because I didn't trust my balance anymore…. I was getting fasciculation in my left leg… [So I got a referral from my GP]….Three months later I went back to the neurologist [for the second time] and I told her, I said, "I'm afraid whatever this is, is ALS." And she said, "Well, it can't be because your leg is getting stronger." So she sent me away." *Patient 8*
14. "What I didn't like about [my diagnosis] was that it was almost like it was he was telling the students…watch how he reacts when he gets told because it's so devastating…This is not a stage show. This is an open and honest communication where this is the worst kind of uh, you know uh, news you can give me… I didn't care for that experience at all… [It was] probably a ten-minute conversation…. And then after that meeting, they all left, then-then I broke down by myself." *Patient 4*
15. "When he got the [genetic] report… [he] said, "I still don't believe you have it."… [that made me] upset because just from the symptoms of my mom and my brother I knew that it was starting. So that part was upsetting." *Patient 6*

Information needs of patients and caregivers at time of diagnosis

16. "[After receiving the diagnosis we said] We're all in for anything. Any research related…whether it was drug related or the MRI, body donation, like anything, we're all in. So that to me is another healthy attitude." *Caregiver 2*
17. "[We] wanted to know a fix for it. We wanted to know if it was hereditary because we had the [two children]… That was the biggest thing… I know it was always something in the back of my head that he could have passed it on to the [children]." *Patient 9*
18. "[When I got to the ALS Clinic] they could not put a label to what I have, so it was labeled as ALS and that I would then go become part of the ALS Clinic… [So at that point] I'm thinking, okay, if I have it, at least if I'm with the clinic and the team and they provide support, then I will get help or whatever, and so I was okay with that….I was more upset when it was unknown what it was going to be…When I knew it was ALS, I think by that point, yeah, that's it. That's my life so it's okay…[and I felt better] because the unknown was gone" *Patient 5*
19. "[At the time of diagnosis] I would have liked to have heard of… [treatment and alternative treatment] prior experience of other people with the disease, just more personalized information as far as how the disease progresses. If there are things that would be beneficial, I would like to know in advance or before symptoms get too bad…[Crying] [It's important…] for hope…" *Patient 3*

Communication about progression and prognosis

20. "[ALS Physician told me] Just how everyone is different. He said that the version I have [slow progressing upper limb onset] is the best of the worst, and I get that [after seeing other ALS patients] … [and him telling me that] was beneficial - because you've got this bad disease but it could be worse." *Patient 4*
21. "[ALS Neurologist] said it could be up to five years… which gave my sister hope in that. [The fact that they] said three to five years… [After she told them] I think [My family] were really focused on it… I kind of felt bad. [Three months later] My sister goes, [she asked the ALS Clinic staff] "Five years? Look how much she's deteriorated in the last three months." … My sister was quite angry." *Patient 7*
22. "[However] if they could have told me that I had slow progression and that I'm going to be around for many years, I would have loved that… this three to five-year average for life expectancy is probably a misguidance and maybe it scares people [because it doesn't separate different onset and progression groups]." *Patient 5*
23. "Well, given the how long I have lived, I would probably have liked to have heard that that was a possibility. Now, implicit in what [ALS neurologist] said was that not everybody dies that quickly, but that's not what you tend to hear." *Patient 8*

Table II. Provider Perspectives

Symptom onset and diagnostic experience
1. "Oh, that can be really surprising. There's some that know everything about it and some have had relatives…Then there are others who, if you use the term ALS, they don't know what you're talking about." *Provider 8*
2. "I think the most challenging situations are when people coming not having any information at all. [Because] people almost come in being prepared to some extent for the worst. They've already almost moved on to their next step." *Provider 13*
Provider approaches to communicating an ALS diagnosis
3. "I [provide the diagnosis] over two visits and not sort of data dump everything at once… I summarize [their] clinical history for them, establish that rapport… At the first visit I wouldn't really communicate too too much. And there's really a bit of an interplay between sort of what I think their understanding [is]." *Provider 3*
4. "It depends on the individual. I find that the more I practice, the more I realize that it's an individual approach. Some patients want to know a lot at the beginning. I answer their questions. Some patients don't…I do a warning shot followed by what the disease is. I give my reasons for saying it. I give reasons for hope and [the practical] things that we will do" *Provider 9*
5. "Well, I try to explain what it is and it's disappearance of nerve cells…There's no way we know right now to replace them … it can involve your arms and legs causing paralysis which can progress over time, or it can involve the nerve cells controlling your swallowing and speech and breathing… that there's a variety of different types. Some people have survived for years with versions of this. There's only one treatment out called Rilutek or riluzole that may slow down the disorder a little bit…but despite all that, there's a team of specialized people here that try and help through the course of this and you just go day by day with it." *Provider 8*
6. "I sit down and I usually draw a diagram actually for them showing very schematically the motor neuron pathways…I find that when I schematically draw it out for them like that and explain the diagnostic process, it makes more sense for them…. With that diagram, it's a lot easier for me to explain why I'm doing these investigations and why I need to stick needles into a bunch of muscles during the examination." *Provider 12*
7. "Am I going to choke to death?" or, "Am I going to suffocate?" those are really rare conversations in the first visit, but [If] I get the sense that people are distressed by what they have seen with other people with ALS. I think at that point, I do have to step in and say that quality of life is important. People don't choke to death. People don't suffocate to death because we have interventions." *Provider 13*
8. "[After a diagnosis is given] I know people go in the Net and everything. I make sure to talk right away the first meeting about the things that scare them the most… [But only if] there's some sort of prompting, [otherwise] I won't go. [But] when I see that the patient is going there, I won't let him [go] one or two months making his own assumptions." *Provider 10*
9. "[Patients are frequently bringing in treatment information, but] …I [only] discuss [the proven therapies in ALS]…and that anything outside the confines of the clinical trial is not proven and should be approached with caution. I simply can't advise if they want to know about a particular supplement… [or a] compound that hasn't been studied." *Provider 9*

Communications about ALS progression and prognosis

10. "How much longer do I have?" That's one of the more common questions that comes up, if they have already thought this was ALS or have been told this was ALS. It's interesting some won't come out and say it directly…They'll ask those kind of things or, "What's going to happen next?" *Provider 13*
11. "I'll tell them some people have been with me for five years, some people have been with the clinic for over ten years and the first response is like "I'll take ten years." [It provides them with a] little bit of hope" *Provider 4*
12. "I'd say it's variable… I do try to give information, [about different rates of progression between groups] but I said everybody's different and you have to be really careful and I think because there's still people that come back and say, "You told me I won't live this long," and they actually carry on for quite a while." *Provider 8*
13. "I know there's an uncertainty there. This makes it a little hard too, yes. You- you're giving a range, and some patients will not understand that you cannot be a lot more precise than that." *Provider 11*

Information Needs at the time of Diagnosis

At the time of diagnosis, participants described a desire for information about treatment and research (Table I, 16) and about the heritability of ALS (Table I, 17). Information about future care and support (Table I, 18 and 19) and the heterogeneity of prognosis and progression reduced uncertainty or provided hope (Table I, 20). Discussions on these topics either occurred or were wished for, retrospectively (Table I, 19). However, providers struggled to discuss research and alternative treatments and cautioned patients and caregivers about reliance on external information (Table II, 9).

Communication about Progression and Prognosis

For some patients, the discussion of the variability in progression provided hope (Table I, 20). However, for others, the emotional impact of an overly optimistic prognosis caused distress and anger (Table I, 21). Nevertheless, some patients were concerned with being given a general prognosis of three to five years for all forms of ALS (Table I, 22) and others preferring to hear of the possibility of living longer (Table I, 23). Emphasizing the need for communications to be responsive to patient preferences, one patient wanted no further negative information about his/her prognosis, preferring to "keep focused on today on being positive about life even with living with ALS" (Patient 2).

Providers were generally responsive when patients subtly indicated that they wanted to discuss prognosis (Table II, 10). Some providers probed to determine if the patient was ready to discuss sensitive topics (Table II, 4 and 8). Providers focused on individual differences in hope and optimism as a cue for the framing of emotionally sensitive information (Table II, 11). However, explanation of this variability also posed challenges if it was misunderstood by patients and caused frustration due to a lack of precision (Table II, 12 and 13).

DISCUSSION

The participants in my study described their experiences in receiving or providing an ALS diagnosis. Patients and caregivers described both positive and negative experiences, while providers described their actions as largely conforming with best practice recommendations for communicating an ALS diagnosis (Andersen et al., 2012). My results further describe the external information seeking behaviour and information preferences of patients and their

caregivers on diagnosis, disease progression, and prognosis and the extent to which these are accounted for by healthcare providers.

External Information Seeking Prior to Diagnosis

Similar to other studies (Abdulla et al., 2014; Chiò et al., 2008), most patients and caregivers in my study reported knowing little about ALS before being diagnosed, despite most previously hearing of it. During their 'diagnostic journey', patients and caregivers quickly came face to face with the limitations of their own knowledge and the knowledge of their primary care providers. Poor disease familiarity among providers (Chiò, 1999; Mitchell et al., 2010; O'Brien et al., 2011) resulted in delayed or inappropriate referrals for specialty investigations and, in some cases, misdiagnosis (Cellura, Spataro, Taiello, & La Bella, 2012; Chiò, 1999; Donaghy, Dick, Hardiman, & Patterson, 2008; Household & Swash, 2000; O'Brien et al., 2011; Paganoni et al., 2014). Referral to a neurologist was therefore a crucial step in the diagnostic journey of participants in my and other studies (Nzwalo, de Abreu, Swash, Pinto, & de Carvalho, 2014).

For those who reached the limits of their knowledge, the ensuing uncertainty resulted in information seeking pre-diagnosis (Galvin et al., 2017; O'Brien et al., 2011; Pavey et al., 2013; Abdulla et al., 2014). Patients and caregivers described existing awareness of ALS from their social circles and pop culture, which shaped their initial understanding of ALS (Abdulla et al., 2014). While some information caused misconceptions and had a negative impact, ALS fundraising and awareness campaigns, such as the ALS Association's 2014 Ice Bucket Challenge (http://www.alsa.org/fight-als/ice-bucket-challenge.html), combined with some interactions with primary care providers, were useful as a starting point when seeking further information.

Communication of an ALS Diagnosis – The Patient and Caregiver Perspective

The patients and caregivers in my study described positive diagnostic experiences as patient-centered and long enough to build rapport, allowing for questions to be answered by providers. Respect for dignity and privacy was important, as reported previously (Andersen et al., 2012; Chiò & Borasio, 2004). The time taken to deliver the diagnosis, along with the building of rapport, has been reported to correlate with higher ratings of provider ability and patient satisfaction (Aoun, Breen, Howting, et al., 2016; McCluskey et al., 2004). Despite the emotional difficulty of receiving a diagnosis, participants described several positive provider characteristics like patience and empathy while also being matter of fact and compassionate.

Similar characteristics have been described by patients with ALS elsewhere (Aoun, O'Brien, Breen, & O'Connor, 2018).

Negative diagnostic encounters, on the other hand, were terse and dismissing of patient concerns; they resulted in distress or frustration. Such encounters are unfortunately common (O'Brien et al., 2011; Pavey et al., 2013) and are important factors related to the satisfaction of ALS patients and caregivers (Aoun, Breen, Howting, et al., 2016; McCluskey et al., 2004). Increasing recognition of this importance has resulted in an expansion of accepted practice recommendations to include the use of clear language, empathetic delivery, rapport building and openness to answering patient questions (Chiò & Borasio, 2004; Andersen et al., 2012). Irrespective of the quality of diagnostic encounters, participants described the emotional shock coupled with relief from the unknown that accompanied a diagnosis (Aoun et al., 2018; O'Brien et al., 2011; Pavey et al., 2013), followed by a refocus on problem solving and adaptation (Galvin et al., 2017). Both relief and fear at diagnosis among patients with rheumatoid arthritis were noted in seminal work done by Bury, (1982, p.173), who described a "confirmation of the worst" among those who knew or suspected their diagnosis beforehand. Despite the emotional trauma of a poorly communicated diagnosis (Pavey et al., 2013), ALS patients were relieved by explanations of unexplained and frightening symptoms.

Communication of an ALS Diagnosis – The Provider Perspective

Healthcare providers seek to reduce uncertainty through the provision of relevant information to patients and caregivers. This role may begin with the resolution of a "perceived absence of information" via the provision of the ALS label and related information during diagnosis (Carleton, 2016). Providers in my study described step-wise diagnostic approach for the delivery of highly sensitive information, calibrated against the patient's understanding of his or her illness. This approach accords with the SPIKES protocol, which provides a non-prescriptive approach to delivering bad news (Baile et al., 2000). The six steps of the SPIKES protocol are: (1) **S**etting up an interview; (2) assessing the patient's **P**erception; (3) obtaining the patient's **I**nvitation; (4) giving **K**nowledge and information to the patient; (5) addressing the patient's **E**motions with empathic responses; and (6) **S**trategy and summary. Originally developed for breaking bad news to patients with cancer, the SPIKES protocol has been

correlated with patient satisfaction and perceptions of provider ability when delivering an ALS diagnosis (Aoun, Breen, Howting, et al., 2016; Baile et al., 2000; McCluskey et al., 2004).

Misconception and low information about ALS have been found in small samples of the general public (Davis & Turner, 2010). Likewise, ALS patients and caregivers also have low to non-existent knowledge of ALS at diagnosis (Abdulla et al., 2014; Chiò et al., 2008). Providers in my study often used prompts and active listening when addressing low levels of understanding and misconceptions about ALS among patients. These approaches allowed for the frank discussion of emotionally sensitive topics early in the disease course and, during later encounters, decreased uncertainty between appointments. The potential for misunderstanding highlights the need to assess patient understanding during clinical encounters, particularly when delivering a terminal diagnosis (Andersen et al., 2012; Baile et al., 2000). However, differences in patient and caregiver preferences in my study means that the "minimal set" of information that should divulged during an ALS diagnosis remains unclear (Abdulla et al., 2014; Chiò et al., 2008, p. 54).

Communications about prognosis and disease progression

The effective delivery of a diagnosis may set the tone of the therapeutic relationship (Chiò & Borasio, 2004), therefore, satisfactory communications that are empathetic, provide encouragement, and are respectful of individual patient differences should continue past their valued use during diagnosis (Abdulla et al., 2014; Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008). Patients and caregivers in my study were interested in topics that reduced uncertainty, including heritability, disease progression and prognosis. Participants did not frequently express a desire for information about end of life during diagnosis. While the provision of ALS related knowledge at diagnosis has been linked to patient-caregiver perceptions of provider skill, information about end of life may be less important when compared to other topics (Aoun, Breen, Howting, et al., 2016; McCluskey et al., 2004). Among other factors, patient ambivalence, family dynamics, and a general unwillingness to discuss emotionally sensitive topics that deal with the end of life may partially explain these differences in information preference throughout the disease course (Almack, Cox, Moghaddam, Pollock, & Seymour, 2012; Sampson et al., 2011; Slort et al., 2011; Stewart, Goddard, Schiff, & Hall, 2011).

Providers described the difficulty of explaining uncertain and emotionally sensitive information about ALS prognosis and progression, and the consequences for patients and caregivers when this information was incorrect, including emotional distress. These sentiments were echoed by some patient participants, who wished they had received more accurate information on progression and prognosis at the time of diagnosis. Providers in my study recognized the challenge of individual differences in pathology, patient disease knowledge, information preferences, and individual interpretations when delivering bad news (Abdulla et al., 2014; Ptacek & Eberhardt, 1996; Swinnen & Robberecht, 2014). This clearly impacted the approach they took when delivering an ALS diagnosis. Prognostic ambiguity and uncertainty in disease trajectory similarly increase the challenge of appropriately timing discussions about care and end-of-life options for providers (Almack et al., 2012; Kiely, Stockler, & Tattersall, 2011). In accordance with related guidelines, most attempted an individualistic patient centered approach to appropriately time the provision of sensitive information in diagnostic and post-diagnostic clinical visits (Andersen et al., 2012; Baile et al., 2000).

LIMITATIONS

My study has a number of limitations. First, while I discussed my thematic analysis with other members of the study team, I did not use any interrater reliability measures between two coders, impacting the validity of the findings. Nevertheless, I enhanced the validity of my analysis through the use of fieldnotes, constant comparison methods, and member checking. Second, my study had a small sample size and recruitment of patients and caregivers was limited to one specialized ALS clinic in Edmonton, Alberta, which may limit the generalizability of my conclusions. However, given the thick descriptions provided by participants, alongside a detailed outline of my research methods, it is plausible that these findings might be transferable to other contexts. Third, while the providers were recruited across Canada, they represented those with experience caring for ALS patients and therefore did not reflect the experiences of primary care providers. Encounters with latter category of provider were more negatively viewed by patients and caregivers. Fourth, given the complexity and emotional sensitivity of the diagnostic experience, it is likely that the descriptions offered by participants were influenced by recall bias.

CONCLUSION

Patients and caregivers described an emotionally distressing and uncertain experience when trying to find answers to the emergence of unexplained symptoms that later proved to be due to ALS. This uncertainty often drove patients and caregivers to seek out external information pre- and post-diagnosis. Similar to other studies, patients and caregivers had specific information and communication preferences during their clinical encounters, which, when respected by providers, reduced uncertainty, alleviated distress, improved satisfaction, and provided hope. While there were a number of shared communication preferences, information preferences were more heterogenous. This heterogeneity suggests the need for providers to elicit information preferences on an individual basis and address them as needed. The providers in my study generally described utilizing currently available best practices in the delivery of a diagnosis and communications about prognosis and disease progression. Particularly with regard to the use of a gradual approach to disclosure, and when addressing emotional needs and uncertainty through the use of clear and empathetic language. These patient-centered practices enabled the sensitive delivery of information relevant to the clinical state of the patient, while respecting individual information and communication preferences.

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CHAPTER 4 – INFORMATION SOURCES AND USE AMONG ALS PATIENTS AND CAREGIVERS

INTRODUCTION

The management of amyotrophic lateral sclerosis (ALS) depends on the control of progressively worsening symptoms that are difficult to predict, resulting in complex information and clinical needs among patients and caregivers (Corcia & Meininger, 2008; Hobson, Harwood, McDermott, & Shaw, 2016). All of the information needs of persons living with ALS, including their caregivers, are unlikely to be met within these clinical encounters (Abdulla et al., 2014; Chiò et al., 2008). Information seeking outside of those encounters is therefore common and may be a useful coping strategy among persons living with ALS (Larsson, Nordin, & Nygren, 2016; Tramonti, Bongioanni, Fanciullacci, & Rossi, 2012) and important for the provision of hope (Fanos, Gelinas, Foster, Postone, & Miller, 2008). Indeed, ALS researchers have explored the approaches taken by those affected by ALS in attempting to exert and regain of control over their lives (Foley, Timonen, & Hardiman, 2014; King, Duke, & O'Connor, 2009), while accounting for level of disability (Lee et al., 2001). In this study, I therefore aimed to: 1) understand the motivations of individuals living with ALS and their caregivers for seeking ALS-related information outside of clinical encounters; 2) identify the external information sources and topics of interest; and 3) analyze the value of this information to individuals living with ALS and their caregivers.

Persons living with ALS and their caregivers require a diversity of information on care and supportive needs over the course of the illness (Oh & Kim, 2017). The unpredictable and progressive nature of ALS drives the need for information that helps individuals to anticipate future change and adapt to ongoing physical decline (Hughes, Sinha, Higginson, Down, & Leigh, 2005; Lemoignan & Ells, 2010). However, individual preferences in the management of ALS related uncertainty may act as modifiers to information seeking behaviors (Abdulla et al., 2014; Chiò et al., 2008; Goodyear-Smith, 2005; O'Brien, 2004).

Loss in human affairs is universal, and creates unpredictability, fear, and new threats to an individual's sense of control (Murray, 2001), including for those living with ALS (Brown &

Addington-Hall, 2008; Goldstein, Atkins, & Leigh, 2003; Sakellariou, Boniface, & Brown, 2013). A deteriorating sense of purpose in life and an externalized sense of control (Plahuta et al., 2002) may lead to feelings of hopelessness, the impact of which is well described in the ALS literature (Ganzini, Johnston, & Hoffman, 1999; Ganzini, Johnston, McFarland, Tolle, & Lee, 1998; Ganzini, Silveira, & Johnston, 2002).

End of life communication is uniquely complicated, prone to several challenges (Bowman, 2000; Galushko, Romotzky, & Voltz, 2012), and in need of improvement according to Canadian patients and family members receiving palliative care (Heyland et al., 2010). After Carter v. Canada 2015, Canada passed Bill C-14 (2016) which amended s. 14, s. 227, and s. 241 of the *Criminal Code of Canada* to enable Medical Assistance in Dying (MAiD) for Canadian patients suffering from a "a grievous and irremediable medical condition" *(Criminal Code of Canada* s. 241.2, 1985). However, the majority (82%) of Canadian physicians involved in diagnosing and caring for ALS patients report feeling unprepared for the rollout of programs related to MAiD (Abrahao et al., 2016). The added communication challenges of MAiD as an end of life option may drive patients to seek out related information from external sources. My exploratory study in one Canadian province is one of the first to examine how persons living with ALS and their caregivers access information outside of clinical encounters, including their opinions on the value of that information.

METHODS

Participants

I recruited individuals living with ALS (patients) and caregivers from the ALS Clinic, located in the Kaye Edmonton Clinic, Alberta, Canada, using both purposive and snowball sampling methods. Eligible patients were over the age of 18 and were without cognitive deficiencies, as determined by the clinic coordinator. Eligible caregivers had current or previous experience providing informal care to someone affected by ALS.

Clinic staff asked potential participants if they were interested in research opportunities, and I approached those who were with verbal and written information about the study. For those who expressed interest in participating further, I scheduled a preliminary meeting to describe the study protocol and answer questions. I administered the ALS Depression Inventory (ALS DI)

and the Beck Depression Inventory (BDI) to consenting participants. I excluded five patients who scored over 23 on the ALS DI and no caregivers, because all scored below 20 on the BDI. I then conducted semi-structured interviews with consenting participants who met the inclusion criteria.

Data Collection

I performed a limited chart review of patient health histories to collect demographic information and the time between reported symptom onset and the confirmation of the ALS diagnosis. I used a semi-structured interview guide that covered ALS diagnosis, care, advanced disease, medical assistance in dying, and information gathering behaviors (see Appendix 6). I chose this open-ended approach so that items of importance to participants could emerge during discussions (Charmaz, 2006; Denzin, 1994). I also created field notes immediately after completing each interview to capture initial impressions and emerging concepts, which I integrated into subsequent interviews and my analyses.

Qualitative Content Analysis

I verified verbatim transcripts of interviews against their corresponding digital recording to ensure accuracy. I imported transcripts into N-Vivo 11™ qualitative software to facilitate my analysis. I used open coding to identify themes (Charmaz, 2006; Lawrence & Tar, 2013), using a constant comparison method (Glaser & Strauss, 1967). This involved an iterative reanalysis of transcripts as new concepts emerged from the data (Charmaz, 2006). I discussed these emerging themes on a regular basis with two other members of the research team. These meetings allowed for consensus to develop around the validity and coherence of themes. I then used member checking to ensure the accuracy of the data and the validity of my analyses (Cho & Trent, 2006); I provided either interview summaries or full transcripts back to participants for comment. Full transcripts and written summaries were reviewed with participants, either in person or via email. General interpretation of interview was agreed upon between myself and participants. Most changes were very minor and easily integrated into my final analysis.

Ethics

The study protocol was approved in August of 2016 by the Health Research Ethics Board-Health Panel at the University of Alberta, Canada (see Appendix 8).

RESULTS

I interviewed 22 participants, of whom 12 were patients (7 women and 5 men) and 10 were caregivers (5 women and 5 men); there were 9 patient-caregiver dyads. Patients ranged from 47-77 years of age.

Information seeking outside of clinical encounters

Most patients (10) and caregivers (8) had heard of ALS prior to receiving a diagnosis, frequently referring to knowing about "Lou Gehrig's Disease". Six patients and four caregivers described looking up information before being provided with a diagnosis. Some participants described uncertainty about symptoms and disease progression as a motivation for seeking information (Table I, 1), while others were reluctant to read material prior to being emotionally "ready". (Table I, 2). Some addressed their knowledge deficit through extensive research due to an intellectual need (Table I, 3) or for pragmatic planning purposes in anticipation of ALS progression (Table I, 4). Other participants took a day-to-day management approach (Table I, 5).

Information sources and topics

Both patients and caregivers sought out information from the Internet, including patient blogs and patient fora, and followed stories on news media (Figure 1). The ALS Society provided information directly through an information binder and home visits by staff and indirectly by facilitating a support group for people living with ALS and their caregivers to share information and experiences (Figure 1).

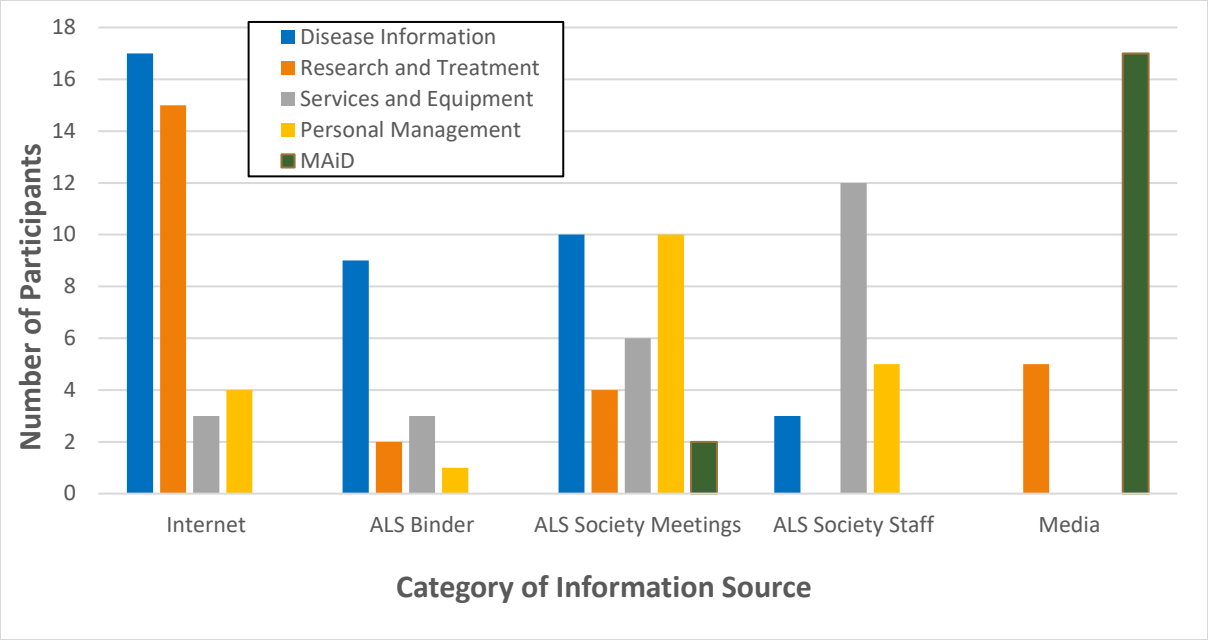

Figure 1. ALS-related topics by category of information source mentioned by participants.

On the internet, participants searched for specific information on management of symptoms, how to adapt as new symptoms emerged, and equipment (Table II, 1). Participants also sought out disease-related information to aid in decisions about the need for future home renovations (Table II, 2). Participants sought information on alternative therapies from peer-support websites, such as PatientsLikeMe (Table II, 3) and from clinic websites and the media on stem cell therapies (Table II, 4). In addition, most participants were aware of news on Medical Assistance in Dying (MAiD) via the media (Table II, 5).

ALS Society staff provided literature that contained information on ALS related topics of interest to participants (Table II, 6) and responded to inquiries about equipment (Table II, 7). Participants sometimes learned about equipment options at patient support meetings (Table II, 8). Participants also exchanged experiences and practical solutions at support meetings. One caregiver described a discussion of end of life advice, including the value of the "peace of mind" that came with being prepared (Caregiver 2). Three patients described discussions of "right to try" legislation that purportedly enables terminal patients to access experimental therapies under certain conditions (Patient 4, 6, and 7). Participants appreciated the practical information and solutions to day-to-day problems that came from others at these meetings (Table II, 9), including information about services and finances (Table II, 10).

Table I. Quotations about motivations for seeking or not seeking further information on ALS.

Motivations for seeking or not seeking further information on ALS
1. "[The changes I could expect in the future] weren't explained. In terms of ongoing assessment, there was no real discussion as far as how the disease normally or typically progresses. I'm just shooting from the hip. I'm assuming my hand and arm function will get worse, that my speech will get worse, but we haven't really discussed any of it." *Patient 3*
2. "The ALS Society gave me a book about ALS. I think I read a couple of pages of that. I just put it away. Now, I think I'm ready to read about it…Maybe I just didn't want to -- see, maybe I didn't want to know about things… but I'm going to read it now. I guess I'm ready. I guess I was in denial too." *Patient 7*
3. "…I had to. Once [Patient] was diagnosed, I started doing a whole bunch of research on the disease, the breakdown like a cellular level because I needed to understand it and that was just me, because I just needed to understand that." *Caregiver 2*
4. "[This is] a learning curve that's faster and you've got to adopt and change your lifestyle on a weekly basis. …The lack of information is probably the most terrifying of all, when you don't know [how to intervene]… if I have no knowledge what to do next, what good am I for [my spouse's] care? You've got to be on top…We need information. Our brain requires it to go forward…" *Caregiver 1*
5. "We haven't really asked too many questions. Pretty much know what's going to happen just from reading on the Internet, but we don't really delve into it that much… It's just more of a day-to-day thing. So like when something else happens, well, it will be that time for that to happen. I'm not really one to live my life on what's going to happen. I'd rather just deal with it today." *Patient 10*

Table II. Quotations about information sources and topics

Information source and content
1. "I have gone back to [a number of blogs] over the years. They tended to be around specific things, either mechanical or dietary. I have a lot of problems with incipient pressure sores… [So] I did a lot of reading on people who had pressure sores and how they were trying to prevent them or how they were coping… [I also] went on the Internet and researched slings, and I found pictures of people doing exactly what I thought should be possible [as my current lift didn't fit my needs at the time]… It's hard to find out how other people have solved those problems." *Patient 8*
2. "Definitely thought about different things and just normal everyday things like looking at the house and how it would be to make this a handicapped house and things like that…. Just from what I read [online] in the first kind of 3 months." *Patient 10*
3. "I'm currently looking at the Deanna Protocol. I have ordered the supplements and we'll see how that works… So that's one that's been talked about quite a bit online. There have been people on the PatientsLikeMe website that have gotten very good results from it." *Patient 3*
4. "You can't help but see this stuff on the media… and there is heaps of testimonials whether it's bogus or whether they've been true. I mean there is patients there telling you that they receive the stage two stem cell therapy and miraculously their progression has stopped or some might even tell you that it's reversed." *Patient 11*
5. "I follow some stuff on the news, like I followed where they're trying to, [pass legislation on Medical Assistance in Dying], I follow that a little bit on the news what's happening with Parliament and stuff but not a whole lot." *Patient 6*
6. "The lady from the society came out and provided quite a bit of literature in the form of a binder, which was very helpful, and that binder was inclusive of everything in terms of treatments, alternative treatments, pathophysiology, availability of equipment and all that stuff." *Patient 3*
7. "We can call the ALS Society and ask for something [equipment wise], but then it ultimately goes through [their] occupational therapist… If I have any questions about the equipment, I ask them." *Caregiver 7*
8. "I got a bidet. When I thought of a bidet, I thought of what the queen uses, something that's 100 years old. But I wouldn't have learned about that at the hospital. [I learned about it by talking about it with other patients and family members affected by ALS at support meetings, then] You go into the backroom [of the ALS Society] there and see all the stuff that they have and it's just waiting for you to, "Okay, I need this, I need this."" *Patient 4*
9. "You're learning different information [at the ALS society meetings]. The practical day-to-day things are what is most important and [immediate]… You learn from [providers] as well but…it's more clinical as opposed to the day-to-day things" *Patient 5*
10. "We go around the table and everyone gets to share and we share a lot. There's one lady… [and] her husband… [they] didn't know about the disability tax credit. So we knew about it, we filled them in…. Home care…we learn about that and I

wouldn't have learned about that had I not gone to those first meetings and had great conversations [with other patients]…" *Patient 4*

Table III. Quotations about participant impressions of the value of information.

The Value of ALS Society Support Groups: Disease Insight, Coping and Inner Strength
1. "And like, there's that exchange of information. So there's one lady there … she's in rough shape. She can't move… I guess it's important to see that to understand what your future's going to be. And uh, so I think from a learning perspective, I learned more there than I do at the ALS clinic because you see, you see the people, you hear their experiences." *Patient 4*
2. "Everybody, uh "this is what's happened to them in the last month" and how they're feeling. And it's good to see when ones that haven't progressed to anything worse than what they were the month before, they're still doing okay and everybody's happy to see each other. And if you haven't been there they'll say, "Oh, I missed you. Where were you? How come you didn't come last month?" So you feel like you belong." *Patient 6*
3. "[After hearing someone else was going on a vacation, I thought] "Wow! I have got upper limb onset and I would be so happy if five years down the road, I was going to [location]" that's one of the human elements there where you get an opportunity to talk face to face with ALS patients in various stages of the disease… I think adds a lot of insight into how you want to interpret your own situation." *Patient 11*
4. "I am more aware of that and we become stronger just saying, "Well, this is us. This is the way it is…. [And] So you look and you go, "Wow, these people are coping and." It helps you picture in. You see them going through it and they still enjoy things. … [And] How they are adopting…." *Patient 7*
5. "[It's] the practical things like -- one of the things I think that's the most useful comment made [during a meeting], "in this" [business] "you've got to learn to laugh a lot because there's a lot of negatives, A LOT of negatives. You just got to find a way to laugh." *Caregiver 1*
Hope for a therapy or a cure
6. "I think for anybody with something fatal, you want to have some hope that maybe one day there'll be a cure. So that's what keeps you I think hanging on is hope that maybe things will level out a little bit or maybe there will be a cure or maybe you can live in under a year or so. I think hope is a very important ingredient of maintaining your mental stability….[however the variability of the disease also] gives you some hope because your destination point isn't always going to be identical to someone else's. It gives just a little bit of hope that you can manage to some degree. You can manage your destination if you wish to." *Patient 11*
7. "I think I have looked at just about every stem cell therapy company and what they offer because you want to weed through a lot of the false hope like, what are they claiming is really happening like the testimonials and stuff like that only have hope. They don't have anything else. There is nothing. All these people live on hope and then they see [treatment and research information] they become very hopeful." *Caregiver 2*
8. "So, there's a thing about right to try. I'm at a point that stem cells, I don't know. To me, I can't see them really rejuvenating what I've lost. But absolutely, I would do stem cells in a minute. Like, why not? I would take any drug in a minute." *Patient 7*

Table IV. Quotations about media coverage of Medical Assistance in Dying

Participant descriptions and views of MAiD media coverage
1. "I'll watch [media coverage on MAiD] it if it comes on, but I'm not going to go out and look for it." *Patient 4*
2. "[Media coverage is] probably having an effect on me like whether I would want to have to travel to a different country to do that or stay in Canada… [So it has influenced me in] just knowing that I don't have to leave Canada if that's the path that I chose." *Patient 10*
3. "The quality of it can be all over the map, but some of what I have read over the years has been quite useful I thought. Some of the anecdotal stuff I find less satisfying because I always feel like there is so much more to the story that we don't know." *Patient 8*
4. "I think it's amazing to me that everybody seems to have an opinion on [MAiD], and some of the people who have opinions on it are so remote from the actual circumstance of being involved personally about having a loved one having a fatal disease. I'm very happy that there is not as much media coverage around [MAiD] now because I think the media coverage…I think was too -- it was extreme." *Patient 11*

Participants Views on the Value of the Information

Participants expressed the greatest value in ALS Society support meetings. While these meetings enabled participants to observe the heterogeneity in disease progression and symptoms and to compare themselves to others (Table III, 1), the meetings also built a sense of community (Table III, 2). Comparison of disease progression provided insight into what could be accomplished, even with more advanced symptoms, such as going on a vacation (Table III, 3). Some participants described the value of observing others living with ALS who were coping and enjoying life, despite suffering from profound loss and functional decline (Table III, 4), as well as maintaining a positive outlook and a sense of humour (Table III, 5).

However, participants also expressed their hope for a cure, the importance of that hope for emotional wellbeing, the hope derived from the variability of disease progression, and the ability to manage that "destination if you wish" (Table III, 6). While some participants expressed skepticism about the "false hope" provided by exaggerated claims around stem cell therapies (Table IV, 7), others would try those or new drug therapies "in a minute" (Table IV, 8).

Most participants had heard about MAiD through the media and it was mentioned on occasion at patient support groups (Figure 1). Most did not describe actively seeking out information about MAiD beyond media reports (Table IV, 1). For some participants, the information provided during media coverage of MAiD changed their perspective and understanding of the subject (Table IV, 2). One participant described the quality of media coverage as highly variable, particularly when focused on personal anecdotes (Table IV, 3). Another participant found that viewpoints expressed in the media coverage were extreme and expressed by individuals without lived experience of the issues (Table IV, 4).

DISCUSSION

Most of the participants in this Edmonton-based exploratory study had a low level of knowledge about ALS at the time of diagnosis. About half of participants in my study sought information from sources outside of their clinical encounters before receiving a diagnosis. Participants expressed their motivations for seeking or not seeking further information, their sources and topics of information, their opinions on the value of the information and its source, and their opinions on information on MAiD.

Motivations for information seeking outside of clinical encounters

Some participants in my study sought out information to address knowledge gaps and reduce uncertainty over the course of ALS, often driven by personal preference (O'Brien, 2004). As described in other studies, participants discussed the pressure to adapt to unpredictable change, progressive functional decline, uncertainties about the future, and a fear of the unknown (Hugel, Grundy, Rigby, & Young, 2006; Hughes et al., 2005; King et al., 2009; O'Brien, Whitehead, Jack, & Mitchell, 2012). Loss is thought to be conceptually related to uncertainty and a degraded sense of control (Murray, 2001). For those who are chronically ill, information seeking may be a means to exert control by deciding to "do something" rather than being told "what to do" (Broom, 2005). This re-exertion of control is closely related to personal autonomy (Foley et al., 2014; King et al., 2009). In addition, information seeking is an important coping strategy for people living with ALS (Hecht et al., 2002; Larsson et al., 2016; O'Brien, 2004; Tramonti et al., 2012; Young & McNicoll, 1998).

Information sources and topics

Participants described accessing information from sources previously identified in the ALS (Abdulla et al., 2014; Chiò et al., 2008) and cancer literature (Rutten, Arora, Bakos, Aziz, & Rowland, 2005). These sources included the internet, printed materials (ALS Society Binder), discussions with patients and caregivers at support meetings, and news media. Participants also sought out information on topics that have been previously described, such as ALS symptomology and progression (Abdulla et al., 2014; Ang et al., 2015; Bolmsjo & Hermeren, 2001; Chiò et al., 2008; Silverstein et al., 1991). Specific to progression, participants searched for information on practical concerns (Aoun et al., 2013; Oh & Kim, 2017), such as appropriate assistive equipment (Bolmsjö & Hermérn, 2003; Williams, Donnelly, Holmlund, & Battaglia, 2008), public services, available resources, and how to practically manage ALS patients when providing care at home (Cipolletta & Amicucci, 2015; Larsson, Fröjd, Nordin, & Nygren, 2015; O'Brien et al., 2012)

The Value of Information – Support Groups and Hope

Participants in this study found great value in face-to-face support groups. This finding is supported by Oh et al., (2014) who found that while encounters with other ALS patients were initially frightening for some, participants described these interactions as an important illness related event. As in other studies, hope and positive thinking were found to be important

methods of coping (Brown & Addington-Hall, 2008; Larsson et al., 2016; Tramonti et al., 2012). These encounters also allowed some participants to positively reframe their situation, while others learned coping mechanisms, such as humor, while developing constructive attitudes and achieving realistic adaptation and quality of life goals (King et al., 2009; Young & McNicoll, 1998).

Hope has been found to be high in people living with ALS, despite facing their own mortality, and this hope is correlated with an increased desire for continued life (Oster & Pagnini, 2012) and a stronger sense of control (Brown & Addington-Hall, 2008; Goldstein et al., 2003). Hope plays an important role in facilitating disease acceptance (Centers, 2001). Indeed, the gathering and consumption of hopeful external information may facilitate a "shift in individual priorities and goals", particularly following the recognition that continued life is no longer possible (Sachs, Kolva, Pessin, Rosenfeld, & Breitbart, 2013, p. 122). At this stage, priorities shift from expectations of survival to hope for comfort and dignity (Sullivan, 2003).

Similar to other studies, some participants derived hope from information about potential cures or alternative therapies (Fanos et al., 2008), which were expected to slow progression and provide symptomatic relief (Bedlack, Joyce, Carter, Paganoni, & Karam, 2015; Vardeny & Bromberg, 2005; Wasner, Klier, & Borasio, 2001). Participants expressed hope in stem cell therapies, despite significant and well documented physical and financial risks to ALS patients in exchange for little or no benefit and an unlikelihood of effectiveness in the near future (Chew, Khandji, Montes, Mitsumoto, & Gordon, 2007; Piepers & van den Berg, 2010). They were aware that information aggressively promulgated by stem cell clinics that provide unproven therapies was likely to be unreliable or inaccurate (Marcon, Murdoch, & Caulfield, 2017; Murdoch, Zarzeczny, & Caulfield, 2018).

Medical Assistance in Dying

Finally, given the prominence of media coverage of MAiD in Canada over the course of this study (2015-2018), it is notable that my participants did not describe actively seek out information on this topic as an end-of-life option. Since Bill C-14 came into effect in June 2017, Canada's provincial health systems, including Alberta Health Services, have independently developed infrastructure for referral and delivery of MAiD. During 2017, 1,961 eligible

Canadians accessed a medically assisted death, 10-13% of which suffered from neurodegenerative disease (Government of Canada, 2018).

Other than an exploration of opinion of neurologists specializing in ALS care (Abrahao et al., 2016) and the experiences of Canadian physicians providing MAiD (Khoshnood et al., 2018) no studies conducted in Canada have explored ALS patient perspectives of MAiD. My exploratory study of participants in Alberta indicates that those living with ALS and their caregivers do not actively seek out information on MAiD from external sources. Most described hearing about MAiD in the news media; few described "following" the topic. Others expressed general dissatisfaction with the coverage as polarizing and of poor quality. Nevertheless, one participant expressed satisfaction in knowing that the MAiD option was available, without having to leave Canada. Aware that their illness was terminal, some participants described not yet being prepared to make important advanced disease and end of life decisions. This may explain why participants did not actively seek out information on this topic as they did with more hopeful topics, such as research and treatment.

LIMITATIONS

This study has several limitations. First, I asked participants to describe past experiences, which potentially introduced recall bias into my data. Second, I used no interrater reliability measures to ensure the rigor of my study. Instead, I used field notes and the constant comparison methods and discussed my thematic coding with Dr. Wendy Johnston and her research associate, Ms Westerly Luth. Third, I did not collect detailed information as to how information sources and preferences changed throughout the disease course. This lack of data limited me from analyzing how these behaviors and preferences may have changed in the face of increasing disability. Fourth, the structure of our interview guide did not probe participants about information sources and content regarding end of life options in the same way it did for MAiD. This likely biased the responses we received from participants who described their information seeking behavior and preferences related to MAiD. Finally, patient participants all received treatment at a single clinic site in Edmonton, Alberta. It is therefore likely that some of the identified drivers of information seeking may not be entirely transferrable to other clinics and regions in Canada.

CONCLUSION

ALS patients and caregivers frequently experience an absence of key information, the need for which is often driven by progressive symptoms, functional decline, and complex forms of loss. This absence of information creates emerging uncertainties over the disease course. Participants described information seeking as facilitating disease adaptation and useful forms of coping. The use of external information allowed participants to address challenges, manage uncertainty, find new forms of meaning, and maintain hope. To our knowledge this work is the first examination of how ALS patients and caregivers consume different types of external information over the course of their illness. This improved understanding may allow health care providers to better locate information gaps among stakeholders that are crucial for adaptation, including how these gaps may be addressed.

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CHAPTER 5 - CONCLUSION

SUMMARY OF FINDINGS

In this thesis, I have attempted to demonstrate how preferences and the experience of uncertainty add to the complexity of the amyotrophic lateral sclerosis (ALS) communications environment. Because patients, caregivers, and providers make up the triad of ALS care, it is crucial to consider these perspectives when attempting to understand what makes this communication environment so challenging.

In Chapter 2, I addressed my methods aim: to assess the feasibility of an interview-based qualitative study with participants living with ALS or their caregivers recruited from the ALS Clinic in Edmonton, Alberta. I determined that patients and caregivers physically and emotionally tolerated discussions of topics related to disease history, diagnosis and care experience, end of life, medical assistance in dying and information gathering. Accommodations included: conducting interviews over multiple sessions, conducting a patient interview with a caregiver present to assist in communications, providing breaks during periods of emotional upset, and reminding participants that they could decline to answer emotionally distressing questions. Given my experience in this exploratory study, an interview-based expanded study, if conducted with caution and tact, would be both feasible and likely to generate valuable data. Indeed, some participants appreciated the opportunity to share their experiences. However, interview-based methods will be more difficult with patients in the later stages of ALS who are facing greater communications challenges.

In Chapter 3, I addressed two substantive aims of my thesis: (1) to analyze the experiences and preferences of patients, caregivers, and providers in receiving or providing an ALS diagnosis; and (2) to understand the information preferences of patients and their caregivers about disease progression, therapy and prognosis, and the extent to which these are accounted for by healthcare providers. Patients and caregivers frequently described experiencing uncertainty and emotional upset when attempting to find answers and explain symptoms before the confirmation of a diagnosis. Participants also described what made the eventual delivery of an ALS diagnosis both a positive and negative experience. They described positive experiences as

patient-centric, with empathetic delivery that was not rushed. In contrast, negative experiences were terse and dismissive of patient concerns. Similar to other studies, patients and caregivers identified specific information and communication preferences during their clinical encounters, which, when respected by providers, reduced uncertainty, alleviated distress, improved satisfaction and provided hope. Uncertainty drove participants to seek out additional information as a means of reducing uncertainty, preserving hope, and gaining information about the practical solutions required for daily adaptations throughout the disease course.

In line with best practices for the delivery of bad news, provider participants described a careful stepwise approach when delivering bad news and in providing information both pre- and post-diagnosis. However, seven of my provider interviewees were experts in the care of ALS patients, while most of the negative encounters described by patients and participants were with primary care physicians or physicians who did not specialize in ALS. Thus, while experienced providers accounted for the preferences identified by patients and caregivers, less experienced clinicians might not employ best practices in the delivery of bad news. All participants acknowledged the challenges in delivering accurate information on progression and prognosis at the time of delivery, given the heterogeneity of ALS.

In Chapter 4, I addressed another substantive aim of my thesis: to analyze patient and caregiver motivations for seeking information outside of clinical encounters, identify the external information sources used and the specific topics of interest, and their respective value to patients and caregivers. Most patient and caregiver participants sought information outside of clinical encounters at some point, while close to half pursued information prior to confirmation of the ALS diagnosis. Some information sources were described as highly valuable, especially patient support groups facilitated by the ALS Society. The need for further information was often driven by progressive symptoms, functional decline, and complex forms of loss. The use of external information allowed participants to address challenges, manage uncertainty, find new forms of meaning, and maintain hope. Indeed, one key finding was the extent to which patients and caregivers sought out hopeful information on experimental therapies and research, while only passively following news media coverage on medical assistance in dying (MAiD).

In the following sections, I outline recommendations that address the information and communication challenges I identified. These recommendations are informed by both the

existing ALS literature and my analyses. I then discuss study limitations and make recommendations for future research.

RECOMMENDATIONS

Recommendation 1 – Improve the delivery of bad news by healthcare providers

The effective delivery of a bad news diagnosis has been a long-standing topic of concern within the palliative and ALS literature (Johnston, Earll, Mitchell, Morrison, & Wright, 1996; Ptacek & Eberhardt, 1996). This has resulted in a recognized need to identify and examine best practices in the delivery and content of an effective, humane, patient-centered diagnosis.

Elements of best practice in the delivery of bad news have been consolidated into the SPIKES protocol for the delivery of bad news. As described in Chapter 3, the six steps that make up the acronym SPIKES are: (1) **S**etting up an interview; (2) assessing the patient's **P**erception; (3) obtaining the patient's **I**nvitation; (4) giving **K**nowledge and information to the patient; (5) addressing the patient's **E**motions with empathic responses; and (6) **S**trategy and summary. Thus a major component of the SPIKES protocol is the identification of a patient's understanding of his/her disease and what information s/he would like to have in the moment, as well as addressing emotional reactions to the delivery of a diagnosis (Baile et al., 2000).

Within the ALS literature, adherence to the SPIKES protocol during the delivery of a diagnosis has been associated with improved patient satisfaction and a higher rating of perceived provider skill (Aoun, Breen, Howting, et al., 2016; McCluskey, Casarett, & Siderowf, 2004). Other important qualities of effective communication of a diagnosis also included offering empathetic responses, suggesting realistic goals, exploring expectations, discussing what patients and family members hope for, and the creation and follow through on a plan of referral and support (Aoun, Breen, Howting, et al., 2016)

The breaking of bad news to ALS patients has been conceptually broken down into both process and content (Chiò & Borasio, 2004; Johnston, et al., 1996). Communication process concerns itself with style, language, and context of communication. Expert opinion and early research recommended the use of language that can "transmit warmth, empathy, and respect, preserving hope, but without false reassurance" while also being straightforward, honest, and

sensitive to patient receptivity towards information (Beisecker, Cobb, & Ziegler, 1988; Chiò & Borasio, 2004, p. 197). Chiò et al., (2008) found that patient satisfaction with the communication of a diagnosis was associated with an impression that their providers understood their feelings. Other work has emphasized the value placed on directness, clarity, the presence of another person for support, being able to ask questions of their provider, and the avoidance of unnecessary and pessimistic information (Johnston, et al., 1996). Clinical guidelines also emphasize the importance of the setting in which the diagnosis takes place, the use of hopeful and honest reassurance, and warm, caring, empathetic, and respectful language (Andersen et al., 2012). Finally, the provision of enough time during a diagnostic encounter to allow for emotional processing is also an important component of patient satisfaction (Aoun, Breen, Howting, et al., 2016; McCluskey et al., 2004).

Communication content refers to the information that is provided during a diagnostic or other clinical exchange (Chiò & Borasio, 2004). The range of information needs that exist during the delivery of a diagnosis and thereafter are vast (Abdulla et al., 2014; Chiò et al., 2008; Oh & Kim, 2017). Personal preferences play a major role in the seeking out of information among ALS patients (O'Brien, 2004). Instead of arguing for the development of a recommended minimal set of information to be given during a diagnosis, it may instead be more useful to advocate for continued practice of eliciting related preferences from patients and caregivers. Some of the earliest work in this area found that ALS patients expect their diagnosing providers to be sensitive to the amount of information they are prepared to hear (Beisecker et al., 1988). However, providers may face significant challenges when attempting to develop this sensitivity during a single encounter – especially when meeting with a newly diagnosed patient. This challenge, alongside individual differences in information tolerance, may require a diagnosis to be delivered in stepwise installments and at a pace ultimately determined by the patient (Andersen et al., 2012). Similar to previous work, providers participating in my study described a stepwise approach to the delivery of a diagnosis (Aoun, Breen, Edis, et al., 2016; Aoun, Breen, Howting, et al., 2016). This approach was key to ensuring comprehension of information and the preservation of hope.

Participants in my study described the information types that were of value when receiving a diagnosis, including those identified in other studies: ALS symptoms, outcome, and

information related to assistive equipment (Chiò et al., 2008; Silverstein et al., 1991). While the provision of information has been linked to patient satisfaction during the delivery of a diagnosis, some types of content seem to be preferred over others. For example, patients place value on information related to how their diagnosis was established, the certainty of their diagnosis, the current state of ALS knowledge, current trends in research, and existing clinical trials of therapies and other interventions (Aoun, Breen, Howting, et al., 2016; Chiò et al., 2008). In contrast, patients may view information about end of life options and gastronomy insertion as much less important than other topics, particularly during the delivery of a diagnosis (McCluskey et al., 2004).

Information content clearly plays an important role in the delivery of an ALS diagnosis. The effective delivery of a diagnosis may have future implications as the first step in providing palliative care, while also setting the tone of the therapeutic relationship (Borasio, Sloan, & Pongratz, 1998; Chiò & Borasio, 2004). Despite stating that they had been provided with all relevant information, patients and caregivers remain likely to seek out information from multiple external sources post diagnosis (Abdulla et al., 2014; Chiò et al., 2008). This behavior indicates that ongoing information needs develop and persist post- diagnosis, in spite of the best efforts of health care providers when breaking bad news.

Finally, experienced providers in my study accounted for the preferences identified by patients and caregivers and generally followed best practices in delivery of a diagnosis. Patients and caregivers described poor diagnostic encounters from less experienced healthcare providers. This points to the need for continuing medical education for healthcare providers to both understand best practices when delivering a terminal diagnosis and to continue to reinforce the need for patient-centric, effective, and empathetic communication of bad news.

Recommendation 2– Improve information availability and communication over the course of ALS through support groups

While the information and communication preferences of patients and caregivers affected by ALS are well described during the diagnostic period, less is known about how these change over course of the disease. There is significant information demand after diagnosis as patients and caregivers often have to adapt to unpredictable change, functional decline, uncertainties about the future, and a fear of the unknown (Hugel, Grundy, Rigby, & Young, 2006; Hughes,

Sinha, Higginson, Down, & Leigh, 2005; King, Duke, & O'Connor, 2009; O'Brien, Whitehead, Jack, & Mitchell, 2012).

The adaptation pressures faced by ALS patients often create information gaps over the course of their illness (Oh & Kim, 2017). Patients may be interested in diagnosis related information, prognosis, outcome, and how death from respiratory failure occurs (Aoun et al., 2013; Chiò et al., 2008; Lemoignan & Ells, 2010); hopeful information about treatment and research (Abdulla et al., 2014; Aoun et al., 2013; Chiò et al., 2008); and the clinical course and progression of ALS (Abdulla et al., 2014; Ang et al., 2015). Patients and caregivers place importance on education on practical and day to day care when adapting to functional changes at home (Larsson, Fröjd, Nordin, & Nygren, 2015; O'Brien et al., 2012), and these practical needs may increase over time (Weisser, Bristowe, & Jackson, 2015). Alternatively, need for information about unmet practical needs among caregivers may decrease as they gain experience in providing care (Weisser et al., 2015).

In response to the need to adapt to ALS progression, patients and caregivers frequently seek out additional information after the delivery of a diagnosis (Abdulla et al., 2014; Chiò et al., 2008), and I identified this trend in my research. The seeking out of external information allows patients to adapt to progressive pathological change. However, as also identified in my analysis, it is important to note that in addition to need, individual preferences and fear may influence the information seeking behaviors of patients (Hughes et al., 2005; O'Brien, 2004).

Providers must therefore attempt to provide patients with the right information at the right time, while accounting for the potential emotional sensitivity of the information (Goodyear-Smith, 2005) and its impact on decision making (Lemoignan & Ells, 2010). Patient and caregiver participants in my study described several adaptation pressures and a need for additional practical and experiential information. They found greatest value in the exchange of experiential knowledge and practical information during ALS Society support meetings. However, they distinguished between the type and purpose of the information provided by the ALS clinic versus what was provided and exchanged at patient support groups. While several participants described the value of the latter, one participant explained the limitations of the information exchanged in such fora due to the absence of ALS 'survivors' and differences in motivating psychology when compared to support groups for other illnesses.

People have often asked me why ALS patients don't get together and "Simple, because there are no survivors."... there are no people who have gone down this road and come out the other end and then sort of like the -- a lot of the cancer support groups, they rely heavily on the psychology of survival. It doesn't seem to work the same way with ALS because people don't think in terms of surviving it. - Patient 8

This limitation has implications for the accumulation and exchange of experiential and practical information for those who are and will be newly diagnosed.

In light of this discussion, I specifically recommend that:

a. Healthcare providers should encourage participation in support groups:

Some participants in my study expressed hesitation before attending support groups facilitated by the ALS Society. Patients and caregivers may, therefore, benefit from provider encouragement when deciding whether they should attend patient support meetings. This encouragement and potential increase in attendance may provide more patients and caregivers with the information they need to adapt to the changes and challenges they face in their daily lives. The practical utility of support groups, alongside other intangible benefits, such as a sense of community, should be emphasized by providers. Furthermore, providers should identify and discuss patient and caregiver concerns and fears about attendance. Patient and caregiver understanding of the purpose of these groups, including provider experiences of their benefit, should be discussed. The writing of 'support group prescriptions' may be a useful way to broach the subject with patients, invite discussion of concerns, and "formalize" their attendance as an important and recommended component of care.

b. The open, patient-centered format of support groups should be preserved:

My analysis suggests that patient support meetings served as a means of information exchange, facilitated successful coping, and provided a sense of community for attendees. According to participants, much of the generated value from these meetings depended on the open discussion of "everything" that was related to daily life with ALS. This open and unrestricted format facilitated the generation and exchange of useful information that was of interest to patients and caregivers. This "ground up", patient-centered approach to the broaching and discussion of topics is possibly the key to the practical utility and adaptive value of the information gathered from these support meetings. While some moderation from staff may be

appropriate, an open form of discussion should remain as the guiding principle of these support groups. In addition, support group moderators might keep a repository of the issues discussed matched to possible resources or solutions.

STUDY LIMITATIONS

My thesis research has several limitations. First, as a qualitative study, this work aimed to describe a complex clinical process, not predict its outcomes. The nature of the data, small sample size, and recruitment from one ALS clinic in Edmonton, Alberta limits the generalizability of my findings, but some findings are likely transferable to similar contexts. In addition, most of my provider participants were experienced in the clinical care of ALS patients, and did not represent the range of healthcare providers encountered by ALS patients and caregivers over the course of the disease.

Second, my work relies on participants' descriptions of past experiences. These experiences often dealt with emotionally difficult subject matter, periods of extreme stress, and significant disruption. At times, these descriptions also relied on the recall of events that occurred several years ago. The nature of these experiences and the significant time since their occurrence likely added a degree of recall bias into our data.

Third, I was the sole coder of my interview transcripts and therefore did not employ any interrater reliability measures. Nevertheless, to improve the rigor of my work, I: 1) wrote field notes immediately post interview; 2) used the constant comparison method when analyzing interview transcripts; 3) discussed the development of my themes and concepts with my supervisor, Dr. Wendy Johnston and her research associate, Ms Westerly Luth; and 4) engaged in a member checking exercises with all participants.

Fourth, while participants described the use of specific information sources and content, my study is limited by the non-longitudinal nature of my data. This prevented me from determining if information-seeking preferences, sources, and content changed over time. More specifically, my current data does not contain information related to how these behaviors change in the face of increasing disability.

Finally, I asked specific questions about MAiD but did not focus questioning on other end-of-life options. This collection oversight likely introduced bias into our data, particularly when

compared to what misleadingly looks to be a nonexistent interest in information related to end of life options in Figure 1 Chapter 4.

FUTURE RESEARCH

My research has demonstrated the viability of an interview-based study with ALS stakeholders. Further research should expand the study to other sites to explore regional differences in the evolving needs and preferences of patients and caregivers over the course of ALS, including the responsiveness and adaptability of healthcare providers in the form and content of their communications. Specifically, further research should focus on diagnostic information preferences in both content and style in the delivery of an ALS diagnosis and mechanisms to implement best practices in the delivery of bad news.

In addition, further research should focus on how the communication needs and preferences of patients change over the disease course, including further research on communication of end-of-life options. As MAiD becomes increasingly available across Canadian provinces, further research will be required on best practices in communication about its availability and suitability compared to palliative care.

Finally, on a practical note, given my finding of the value of patient support groups, research should be undertaken on alternative modes of providing support groups to more advanced patients and those in rural or remote areas, such as online fora.

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APPENDICES

APPENDIX 1. RECRUITMENT INCLUSION AND EXCLUSION CRITERIA

Patients: We are interested in interviewing patients about their experience communicating and making decisions related to their ALS care. Patient participants may be male or female, over the age of 18 and cognitively competent in a decision-making capacity. Patient participants must have a current diagnosis of ALS and be actively receiving specialized care by a neurologist or palliative care specialist. They may also be at any stage of their disease course, or at any point in their end of life decision making. Patient participants will primarily be recruited from the Neurology Clinic at Kaye Edmonton Clinic at the University of Alberta.

For recruitment, there will be two separate episodes of contact between patients and research staff. First, patients will be asked if they are interested in participating in research by the study coordinator, and if they would be willing to speak to someone about participating. If they indicate an interest in being contacted about research, patients will be asked if they would a) prefer to contact the researcher or b) consent to be contacted by the researcher. Patients will also be given an opportunity to speak directly to a research staff member at the clinic in order to be given recruitment materials (pamphlet) and the contact information of the staff member.

The second stage of patient recruitment will involve an in-depth explanation of the study, an opportunity to ask researchers questions about the study, and the signing of a research participation consent form, and a consent form for the release of medical records. During the second stage, patient participants will be informed of the risks associated with participating. Patients will be reminded that participation is totally voluntary, and that they are under no obligation to participate – and that their refusal will not affect their care in any way.

Immediately after consent, patient participants will be expected to complete the ALS Depression Inventory. In the event a patient participant has a clinically relevant score from the inventory (>23), they will be ineligible for participation in the study and their health care

provider will be informed using the contact information they've provided during the consent process by the PI, Dr. Wendy Johnston.

Caregivers of Patients with ALS: This group will comprise of individuals who are providing informal care to patients with ALS. These people will be recruited alongside patients during clinic visits, or through snowballing if the patient visits the clinic on their own. Caregivers may be male or female, and must be over the age of 18. Caregivers should be providing informal care in the following ways to qualify: physical care, assistive care, personal care, nutritional care, assistance with activities of daily living, assistance with appointments, etc. However, care may not be strictly limited to these activities, and will not be considered hard inclusion criteria.

Caregivers will be approached about recruitment in the same fashion as patient participants, and will involve a two stage process. First, caregivers who accompany patients to ALS clinic visits will be approached by the coordinator, and be asked if they would be interested in participating in any research currently being run out of the clinic. Caregivers will be asked if they a) prefer to contact the researcher or b) consent to be contacted by the researcher. They will also be given the option to speak directly to a member of the research staff in order to be given recruitment materials (pamphlet) and the contact information of the staff member.

The second stage of recruitment will involve an in-depth explanation of the study, an opportunity to ask researchers questions about the study, and the signing of a research participation consent form. During the second stage, caregiver participants will be informed of the risks associated with participating. They will also be reminded that participation is totally voluntary, that they are under no obligation to participate, and that their refusal will not affect the clinical care of the individual they provide informal care to in any way.

Immediately after they have consented, caregiver participants will be expected to complete the Beck Depression Inventory. In the event caregivers scores are clinically significant (= or > than 20), they will be ineligible for participation in the study. Their primary health care provider will be informed using the contact information they've provided during the consent process by the PI, Dr. Wendy Johnston.

Health Care Providers for People with ALS: Health care practitioners may be male or female and must be over the age of 18. Providers who have not worked with ALS patients, or have only

cursory experience in managing ALS, will not be included. We will also exclude providers who do not have direct experience in the management of ALS care, as they have not had to practice within a context that requires communicating and discussing care options with patients. Alternative/naturopathic workers will not be included.

Provider participants will be approached for recruitment through snowballing or directly by the research staff. Specialists and nurses working at the ALS clinic will be approached directly by a member of the research team. Additional specialists who diagnose and refer to the ALS Clinic will be contacted directly by letter, email, or by phone. General practitioners and nurses who care for ALS patients outside the ALS clinic will be contacted through snowballing methods. These individuals will be contacted by phone, email or letters if they agree to be contacted about participation in the study.

APPENDIX 2 - CONDENSED INFORMATION SHEET

Study information

ALS Care Decision Making: Understanding Clinician-Patient/Caregiver Communications

The Study: Researchers from the School of Public Health and the Faculty of Medicine and Dentistry, University of Alberta, are interested in understanding the information needs of ALS patients and their caregivers. Our goal is to improve communication between ALS patients/caregivers and their healthcare providers about an ALS diagnosis, ongoing care and end-of-life decisions.

Participants: We are recruiting ALS patients, their caregivers, and healthcare providers.

What will you be asked to do? We would like to speak with you about your perspectives on ALS Care decision-making, the information sources you rely on, and conversations with healthcare providers. We have approximately 20 questions that we would like to ask you. The interview would take approximately 1-1.5 hours in one sitting. However, we can be very flexible in how we conduct the interview. We can cover the questions in multiple sessions. You can also respond to the questions in your own time, using any method you choose, including email.

Confidentiality: Your identity will be kept confidential. We will not use your name or other identifying information in any publications or presentations based on our research.

Consent: Participation in this study is voluntary. If you decide to participate, your participation will in no way affect any clinical care.

If you are interested in participating in this study, please contact:

Mackenzie Moir
School of Public Health
University of Alberta

APPENDIX 3 - FULL INFORMATION SHEET

Patient Information Sheet

Information Sheet for the University of Alberta Research Project:
ALS Care Decision Making:
Understanding Clinician-Patient/Caregiver Communications

What is this study about?

This study aims to inform clinical communications between healthcare providers, ALS patients, and their caregivers. The communications may be about diagnosis, ongoing care, and end-of-life decision making. We are particularly interested in how communications have been impacted by media attention and legal reforms on medical aid in dying. We are also interested in your opinions on any external sources of information about ALS you have accessed. We value all perspectives on any of these issues.

Who is doing the study?

Dr. Tania Bubela, School of Public Health and Dr. Wendy Johnston, Faculty of Medicine and Dentistry, at the University of Alberta, are responsible for this study. Mackenzie Moir is the primary contact for the study. He is a registered nurse who is currently an MSc student and research assistant in the School of Public Health. Westerly Luth, School of Public Health, is the research associate coordinating the study.

What will we ask you to do during our first meeting to discuss your participation in the study?

During our first meeting we will explain the study and ask for your consent to participate.

During our first meeting, we will ask you to complete some standard, short questionnaires that help us understand your current symptoms. These questionnaires can be completed orally or on a touchscreen. They are called the *Heath Hope Index*, the *Edmonton Symptom Assessment Scale*, and the *Patient Dignity Inventory*. You may already have been asked to complete some or all of these by your healthcare team.

We will also ask you to complete a short questionnaire called the *ALS Depression Inventory*. The questionnaire can be completed orally or on a touchscreen. If the results of this questionnaire indicate that you might be depressed, we will tell you and your treating physician. Then you can receive the appropriate care. We will not continue with the study, and your responses to all questionnaires will be deleted from our records.

What will we ask you to do during the study?

There are three parts to this study:

1) We would like to interview you about your communications with healthcare providers. These communications may be about diagnosis, ongoing care, and end-of-life decision making. We would also like to ask you about any discussions you have had about medical aid in dying. Finally, we would like to ask you about your views on other sources of information that you have used to understand ALS and its care.
2) Once we have analyzed all the interviews, we will invite you to comment on a summary of our findings. We want to make sure that we have properly understood and represented your thoughts and opinions. If you decide to give us feedback, we will go over the summary of our findings with you and record your comments. We will collect your comments in person, by telephone, or electronically depending on your preference. It should take less than an hour to provide feedback, but we will take as long as you require to gather your feedback.
3) We would also like your permission to review and summarise your ALS-related medical records from the University of Alberta ALS Clinic and from your family doctor from a year before your diagnosis. Reviewing your medical records related only to your ALS diagnosis and care will help us to understand how you were diagnosed and the discussions you have had with your healthcare team about your ongoing care.

How will we conduct the interview?

We are very flexible in how we conduct the interview, and we can decide on the best options for you, including:

- In one sitting. The interview would take about 1-1.5 hours. There are 23 questions. With your permission, we will digitally record the interview.
- If you prefer, we can conduct the interview in as many short sessions as you need.
- We can also give you the questions. You can then answer the questions in your own way in your own time. For example, you can respond to questions by email or you can use a word processor and give us the file.

Where will we conduct the interview?

We can conduct the interview anywhere where you feel comfortable, for example, in a private room when you visit the University of Alberta ALS Clinic. We can also come to your home or any other location in the greater Edmonton area.

What are the benefits and risks of participating in this study?

Benefits: Your participation will help to inform best practices for clinical communications between healthcare providers, ALS patients, and their caregivers. The communications are about diagnosis, ongoing care, end-of-life decision making, and medical aid in dying. We are also interested in the understanding how external sources of information are used by patients and their caregivers.

Risks: Some of the interview questions about end-of-life care and medical aid in dying are likely to cause emotional distress. You may choose not to answer any questions that you find distressing. We are not aware of any long-term risks of participating in this study.

What are your rights as a research participant?

- Participation in this study is completely voluntary
- You are free to withdraw from this study ***without*** having to give a reason.
- You can withdraw any portion or all of your answers within **90 days** following the end of the interview process.
- If you choose to withdraw from the study, we will destroy all paper records related to your participation and delete all digital files.

Will I be paid to be in the research?

- To thank you for your time you will receive a grocery card ($25).
- If you travel to the University of Alberta for your interview, you will receive a gas card ($25) and $12 for parking following your interview.
- If you withdraw from the study, you will still get the grocery card and relevant travel repayment.

What type of personal information will we collect?

We will collect your name and contact information so that we can send you the summary of our findings. In addition, with your permission, we will ask you for your Alberta Health Care Number so that we can access your medical records. We will delete that number from our records as soon as we have received your medical records.

How will we protect your confidentiality?

Interviews:

- We will give all digital files (audio or other communications, such as email) an identifier (e.g., Participant 1).
- We will keep the list that links your contact information with your digital files in a locked filing cabinet in a secure office at the School of Public Health, University of Alberta.
- If you participate in a face-to-face interview, we will have the audio file transcribed by a professional transcriptionist who has signed a confidentiality agreement.
- Only study staff who have signed a confidentiality agreement will have access to the digital files.
- We will remove any potentially identifying information from any transcripts prior to storing them on a secure server at the School of Public Health, University of Alberta.
- We will not use your name in any of publications and presentations that result from the study. Instead, we will use the general identifier (e.g., Participant 1).
- We will destroy all paper records and delete digital files 5 years after the end of the study.

Review of Medical Records:

- Only staff with read-only authorization from Alberta Health Services will access your medical records at the University of Alberta ALS Clinic. These are stored on a digital system called *e-clinician*. The staff will create a summary of the reasons for your visits to the Clinic to understand what may have been discussed and when. The summary will be transferred via an encrypted, secure USB key to a secure server at the School of Public Health, University of Alberta. No identifying information will be collected, except for year of your birth and month/year of diagnosis. We will use the same identifier (e.g., Participant 1) for all digital files.
- With your permission, Dr. Johnston at the University of Alberta ALS Clinic will request your medical records from your primary healthcare provider (e.g., your family physician). We would like to request these records from one year prior to your diagnosis. On average, symptoms begin to show approximately one year before diagnosis. The paper copies of your medical records will be sent via secure courier to Dr. Johnston. We will store them in a locked cabinet in Dr. Johnston's secure research space in the Clinical Sciences Building at the University of Alberta. The study staff will summarize the reasons for any ALS-related visits. Once we have completed the summaries, we will destroy the paper medical records in compliance with Alberta Health Services policies. The summary will be transferred via an encrypted, secure USB key to a secure server at the School of Public Health, University of Alberta. No identifying information will be collected, except for year of your birth and month/year of diagnosis. We will use the same identifier (e.g., Participant 1) for all digital files.
- By signing this consent form you are agreeing and giving permission to the study team to collect, use and disclose anonymous information about you from your personal health records as described above.

Risks to confidentiality:

(1) Given the small size of the ALS community in Alberta, it is still possible that people might guess who you are because of what you have said.
(2) If you choose to respond to interview questions via email or send us a document with your answers, we cannot guarantee the security of email. We suggest that you only use a password-protected email account. The University of Alberta uses gmail, however, all email accounts are password protected.

How will we use the information?

We will analyse the answers to our interviews for the main themes that are common and different between participants using qualitative research software called NVIVO. Our analysis will be presented in an MSc Thesis, in academic publications, in popular/lay summaries of our research findings and in presentations. The analyses will contribute to the development of best practice guidelines on clinician-ALS patient/caregiver communications.

Ethics Review

The study protocol has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, **contact the Research Ethics Office at (780) 492-2615**.

Study Contacts

Mackenzie Moir is the primary contact for this study. You can contact Mackenzie or Dr. Tania Bubela, if you have any further questions:

Mackenzie Moir
School of Public Health
University of Alberta

Dr. Tania Bubela
School of Public Health
University of Alberta

Caregiver Information Sheet

Information Sheet for the University of Alberta Research Project:
ALS Care Decision Making:
Understanding Clinician-Patient/Caregiver Communications

What is this study about?

This study aims to inform clinical communications between healthcare providers, ALS patients, and their caregivers. The communications may be about diagnosis, ongoing care, and end-of-life decision making. We are particularly interested in how communications have been impacted by media attention and legal reforms on medical aid in dying. We are also interested in your opinions on any external sources of information about ALS you have accessed. We value all perspectives on any of these issues.

Who is doing the study?

Dr. Tania Bubela, School of Public Health and Dr. Wendy Johnston, Faculty of Medicine and Dentistry, at the University of Alberta, are responsible for this study. Mackenzie Moir is the primary contact for the study. He is a registered nurse who is currently an MSc student and research assistant in the School of Public Health. Westerly Luth, School of Public Health, is the research associate coordinating the study.

What will we ask you to do during our first meeting to discuss your participation in the study?

During our first meeting we will explain the study and ask for your consent to participate.

During our first meeting, we will ask you to complete a short questionnaire called the *Beck Depression inventory*. If the results of this questionnaire indicate that you might be depressed, we will tell you and Dr. Johnston, Clinical Director of the University of Alberta ALS Clinic. She can then refer you for the appropriate care. We will not continue with the study, and your responses to the questionnaire will be deleted from our records.

What will we ask you to do during the study?

There are two parts to this study:

1) We would like to interview you about your communications with healthcare providers. These communications may be about diagnosis, ongoing care, and end-of-life decision making. We would also like to ask you about any discussions you have had about medical aid in dying. Finally, we would like to ask you about your views on other sources of information that you have used to understand ALS and its care.
2) Once we have analyzed all the interviews, we will invite you to comment on a summary of our findings. We want to make sure that we have properly understood and represented your thoughts and opinions. If you decide to give us feedback, we will go over the summary of our findings with you and record your comments. We

will collect your comments in person, by telephone, or electronically depending on your preference. It should take less than an hour to provide feedback, but we will take as long as you require to gather your feedback

How will we conduct the interview?

We are very flexible in how we conduct the interview, and we can decide on the best options for you, including:

- In one sitting. The interview would take about 1-1.5 hours. There are 23 questions. With your permission, we will digitally record the interview.
- If you prefer, we can conduct the interview in multiple, shorter sessions.

Where will we conduct the interview?

We can conduct the interview anywhere where you feel comfortable, for example, in a private room when you visit the University of Alberta ALS Clinic. We can also come to your home or any other location in the greater Edmonton area.

What are the benefits and risks of participating in this study?

Benefits: Your participation will help to inform best practices for clinical communications between healthcare providers, ALS patients, and their caregivers. The communications are about diagnosis, ongoing care, end-of-life decision making, and medical aid in dying. We are also interested in understanding how external sources of information are used by patients and their caregivers.

Risks: Some of the interview questions about end-of-life care and medical aid in dying are likely to cause emotional distress. You may choose not to answer any questions that you find distressing. We are not aware of any long-term risks of participating in this study.

What are your rights as a research participant?

- Participation in this study is completely voluntary
- You are free to withdraw from this study ***without*** having to give a reason.
- You can withdraw any portion or all of your answers within **90 days** following the end of the interview process.
- If you choose to withdraw from the study, we will destroy all paper records related to your participation and delete all digital files.

Will I be paid to be in the research?

- To thank you for your time you will receive a grocery card ($25).
- If you travel to the University of Alberta for your interview, you will receive a gas card ($25) and $12 for parking following your interview.
- If you withdraw from the study, you will still get the grocery card and relevant travel repayment.

What type of personal information will we collect?

We will collect your name and contact information so that we can send you the summary of our findings

How will we protect your confidentiality?

- We will give all digital files related to your interview an identifier (e.g., Caregiver 1).
- We will keep the list that links your contact information with your digital files in a locked filing cabinet in a secure office at the School of Public Health, University of Alberta.
- Your interview will be transcribed by a professional transcriptionist who has signed a confidentiality agreement.
- Only study staff who have signed a confidentiality agreement will have access to the digital files.
- We will remove any potentially identifying information from any transcripts prior to storing them on a secure server at the School of Public Health, University of Alberta.
- We will not use your name in any of the publications and presentations that result from the study. Instead, we will use the general identifier (e.g., Caregiver 1).
- We will destroy all paper records and delete digital files 5 years after the end of the study.

Given the small size of the ALS community in Alberta, it is still possible that people might guess who you are because of what you have said.

How will we use the information?

We will analyse the answers to our interviews for the main themes that are common and different between participants using qualitative research software called NVIVO. Our analysis will be presented in an MSc Thesis, in academic publications, in popular/lay summaries of our research findings and in presentations. The analyses will contribute to the development of best practice guidelines on clinician-ALS patient/caregiver communications.

Ethics Review

The study protocol has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, **contact the Research Ethics Office at (780) 492-2615**.

Study Contacts

Mackenzie Moir is the primary contact for this study. You can contact Mackenzie or Dr. Tania Bubela, if you have any further questions:

Mackenzie Moir
School of Public Health
University of Alberta

Dr. Tania Bubela
School of Public Health
University of Alberta

Provider Information Sheets

Information Sheet for the University of Alberta Research Project:

ALS Care Decision Making: Understanding Clinician-Patient/Caregiver Communications

What is this study about?
This study aims to inform clinical communications between healthcare providers, ALS patients, and their caregivers about diagnosis, ongoing care, end-of-life decision making, and external sources of information accessed by patients and their caregivers. We are particularly interested in how communications have been impacted by media attention and legal reforms on medical aid in dying. We value all perspectives on this latter issue.

Who is doing the study?
Dr. Tania Bubela, School of Public Health and Dr. Wendy Johnston, Faculty of Medicine and Dentistry, at the University of Alberta, are responsible for this study. Mackenzie Moir is the primary contact for the study. He is a registered nurse who is currently an MSc student and research assistant in the School of Public Health. Westerly Luth, School of Public Health, is the research associate coordinating the study.

What will we ask you to do?
There are two parts to this research:

1) We will interview you about your clinical communications with ALS patients and their caregivers related to diagnosis, ongoing care, end-of-life decision making, and external sources of information accessed by patients and their caregivers. We will also ask you about how to communicate about medical aid in dying. We anticipate the interview will take approximately one hour. With your permission, we will digitally record the interview.
2) Once we have analyzed all the interviews, we will invite you to comment on a summary of our findings to ensure that we appropriately understood and represented your thoughts and opinions. If you decide to give us feedback, we will go over the summary of our findings with you and record your comments. We will collect your comments in person, by telephone, or electronically depending on your preference. It should take less than an hour to provide feedback, but we will take as long as you require to gather your feedback

What type of personal information will we collect?
We will collect your name and contact information so that we can send you the summary of our findings.

What are the benefits and risks of participating in this study?
Benefits: Your participation will help to inform best practices for clinical communications between healthcare providers, patients with ALS, and their caregivers about diagnosis, ongoing care, end-of-life decision making, and external sources of information accessed by patients and their caregivers.

Risks: Some of the interview questions about end-of-life care and medical aid in dying may cause emotional distress. You may choose not to answer these questions. We are not aware of any long-term risks of participating in this study.

What are your rights as a research participant?

- Participation in this study is completely voluntary
- You are free to withdraw from this study ***without*** having to give a reason.
- You can withdraw any portions or all of your comments in the interview with 90 days following your interview.
- If you choose to withdraw, your responses will be deleted from the server and not included in our analysis.

Will I be paid to be in the research?

- To thank you for your time you will receive a grocery card ($25).
- If you travel to the University of Alberta for your interview, you will receive a gas card ($25) and $12 for parking following your interview.
- If you withdraw from the study, you will still get the grocery card and relevant travel repayment.

How will we protect your confidentiality?

- We will assign the digital audio file of your interview an identifier (e.g., Healthcare Provider 1) prior to it being sent to a professional transcriptionist.
- Your contact information linked to the identifier will be kept separately in a secure office at the School of Public Health, University of Alberta.
- Only study staff who have signed a confidentiality agreement will have access to the recording and the interview transcript.
- We will remove any potentially identifying information from the transcript prior to storing it on a secure server at the School of Public Health, University of Alberta.
- We will not use your name in any of publications and presentations that result from the study. Instead, we will use a general description (such as "Healthcare Provider 1").
- We will destroy all paper records and delete digital files 5 years after the end of the study.

Given the small size of the ALS community in Alberta, it is still possible that people might guess who you are because of what you have said.

How will we use the information?

We will analyse the interview transcripts for the main themes that are common and different between different interviewees using qualitative research software called NVIVO. Our analysis will be presented in an MSc Thesis, in academic publications, in popular/lay summaries of our research findings and in presentations. The analyses will contribute to the development of best practice guidelines on clinician-ALS patient/caregiver communications.

Ethics Review

The study protocol has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, **contact the Research Ethics Office at (780) 492-2615**.

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Mackenzie Moir is the primary contact for this study. You can contact Mackenzie or Dr. Tania Bubela, if you have any further questions:

Mackenzie Moir
School of Public Health
University of Alberta

Dr. Tania Bubela
School of Public Health
University of Alberta

APPENDIX 4 – INFORMED CONSENT FORMS

Patient Consent Form
CONSENT FORM

To Participate in the University of Alberta Research Project:

ALS Care Decision Making: Understanding Clinician-Patient Communications

Investigator
Mackenzie Moir
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Supervisor
Tania Bubela
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Please read the following carefully:

I read and received a copy of the information sheet provided for the research project or have had the research project explained to me in a satisfactory manner.

I understand the possible benefits and risks of participating in this study.

I have had an opportunity to ask questions and discuss this study.

I understand that I can quit taking part in this study at any time without giving a reason up to 90 days following the end of the interview(s).

I understand that I can refuse to answer specific interview or survey questions, or end the interview or survey process at any time I wish without giving a reason.

I discussed confidentiality with the person who explained the research information letter.

I consent to the use an audio recorder for this research.

I consent to allowing the research team to access my personal health care information related to my ALS diagnosis, care, and management.

I understand that the data will be safely stored for five years.

I understand that the information I provide, including my personal health information, may be used in presentations, reports, and guidelines.

I understand that my primary care physician may be contacted by the primary investigator of this study (Dr. Wendy Johnston) if there is concern related to the status of my mental health.

I agree to participate in this study.

I agree with the above statements ❑ Yes

❑ No

_____ _____

Print Name Signature

Date: _____

Participant telephone number: _____

Participant email: _____

Participant mailing address: _____

_____

_____

_____

Family doctor name: _____

Family doctor telephone number: _____

The plan for this study has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, contact the Research Ethics Office at (780) 492-2615.

Caregiver Consent Form
CONSENT FORM

To Participate in the University of Alberta Research Project:

ALS Care Decision Making: Understanding Clinician-Patient Communications

Investigator
Mackenzie Moir
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Supervisor
Tania Bubela
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Please read the following carefully:

I read and received a copy of the information sheet provided for the research project or have had the research project explained to me in a satisfactory manner.

I understand the possible benefits and risks of participating in this study.

I have had an opportunity to ask questions and discuss this study.

I understand that I can quit taking part in this study at any time without giving a reason up to 90 days following the end of the interview(s).

I understand that I can refuse to answer specific interview or survey questions, or end the interview or survey process at any time I wish without giving a reason.

I discussed confidentiality with the person who explained the research information letter.

I consent to the use an audio recorder for this research.

I understand that the data will be safely stored for five years.

I understand that the information I provide may be used in presentations, reports, and guidelines.

I understand that my primary care physician may be contacted by the primary investigator of this study (Dr. Wendy Johnston) if there is concern related to the status of my mental health.

I agree to participate in this study.

I agree with the above statements ❑ Yes

❑ No

_____ _____

Print Name Signature

Date: _____

Participant telephone number: _____

Participant email: _____

Participant mailing address: _____

_____

_____

_____

The plan for this study has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, contact the Research Ethics Office at (780) 492-2615.

Provider Consent Form
CONSENT FORM

To Participate in the University of Alberta Research Project:

ALS Care Decision Making: Understanding Clinician-Patient Communications

Investigator
Mackenzie Moir
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Supervisor
Tania Bubela
Department of Public Health Sciences
University of Alberta
Edmonton, AB, T6G 1C9

Please read the following carefully:

I read and received a copy of the information sheet provided for the research project or have had the research project explained to me in a satisfactory manner.

I understand the possible benefits and risks of participating in this study.

I have had an opportunity to ask questions and discuss this study.

I understand that I can quit taking part in this study at any time without giving a reason up to 90 days following the end of the interview(s).

I understand that I can refuse to answer specific interview or survey questions, or end the interview or survey process at any time I wish without giving a reason.

I discussed confidentiality with the person who explained the research information letter.

I consent to the use an audio recorder for this research.

I understand that the data will be safely stored for five years.

I understand that the information I provide may be used in presentations, reports, and guidelines.

I agree to participate in this study.

I agree with the above statements ☐ Yes

☐ No

_____ _____

Print Name Signature

Date: _____

Participant telephone number: _____

Participant email: _____

Participant mailing address: _____

_____

_____

_____

The plan for this study has been reviewed for its adherence to ethical guidelines by a Research Ethics Board at the University of Alberta. For questions regarding participant rights and ethical conduct of research, contact the Research Ethics Office at (780) 492-2615.

APPENDIX 5 – INSTRUMENTS

ALS Depression Inventory (ADI-12)

ALS-Depression-Inventory (ADI-12)

This questionnaire consists of 12 statements with 4 possible answers: 'I fully agree', 'I agree', 'I don't agree' and 'I do not agree at all'. Please read every statement precisely and think about to what extent the statement is applicable to you in the last two weeks including today. Please mark (tick) the right answer for you. Please cross only one answer at a time and do not leave any statements open.

		I fully agree	I agree	I don't agree	I do not agree at all
1	I am happy and I smile often.	☐	☐	☐	☐
2	I can appreciate life despite my circumstances.	☐	☐	☐	☐
3	I can get away from it all and I am often relaxed.	☐	☐	☐	☐
4	I feel alive and vital.	☐	☐	☐	☐
5	More often than not I am sad.	☐	☐	☐	☐
6	I have lost all interest in family and friends.	☐	☐	☐	☐
7	Most often I feel empty.	☐	☐	☐	☐
8	There is nothing that I look forward to or that I can enjoy.	☐	☐	☐	☐
9	I often feel lost and abandoned and don't know how to carry on.	☐	☐	☐	☐
10	I look forward to every new day.	☐	☐	☐	☐
11	I often wish I were dead.	☐	☐	☐	☐
12	I feel like I have lost all of my energy.	☐	☐	☐	☐

Beck Depression Inventory

Beck's Depression Inventory

1.
0 I do not feel sad.
1 I feel sad
2 I am sad all the time and I can't snap out of it.
3 I am so sad and unhappy that I can't stand it.

2.
0 I feel discouraged about the future.
1 I feel I have nothing to look forward to.
2 I feel the future is hopeless and that things cannot improve.

3.
0 I do not feel like a failure.
1 I feel I have failed more than the average person.
2 As I look back on my life, all I can see is a lot of failures.
3 I feel I am a complete failure as a person.

4.
0 I get as much satisfaction out of things as I used to.
1 I don't enjoy things the way I used to.
2 I don't get real satisfaction out of anything anymore.
3 I am dissatisfied or bored with everything.

5.
0 I don't feel particularly guilty
1 I feel guilty a good part of the time. I
2 feel quite guilty most of the time.
3 I feel guilty all of the time.

6.
0 I don't feel I am being punished.
1 I feel I may be punished.
2 I expect to be punished.
3 I feel I am being punished.

7.
0 I don't feel disappointed in myself.
1 I am disappointed in myself.
2 I am disgusted with myself.
3 I hate myself.

8.
0 I don't feel I am any worse than anybody else.
1 I am critical of myself for my weaknesses or mistakes.
2 I blame myself all the time for my faults.
3 I blame myself for everything bad that happens.

9.
0 I don't have any thoughts of killing myself.
1 I have thoughts of killing myself, but I would not carry them out.
2 I would like to kill myself.
3 I would kill myself if I had the chance.

10.
0 I don't cry any more than usual.
1 I cry more now than I used to.
2 I cry all the time now.
3 I used to be able to cry, but now I can't cry even though I want to.

11.
0 I am no more irritated by things than I ever was.
1 I am slightly more irritated now than usual.
2 I am quite annoyed or irritated a good deal of the time.
3 I feel irritated all the time.

12.
0 I have not lost interest in other people.
1 I am less interested in other people than I used to be.
2 I have lost most of my interest in other people.
3 I have lost all of my interest in other people.

13.
0 I make decisions about as well as I ever could.
1 I put off making decisions more than I used to.
2 I have greater difficulty in making decisions more than I used to.
3 I can't make decisions at all anymore.

14.
0 I don’t feel that I look any worse than I used to.
1 I am worried that I am looking old or unattractive
2 I feel there are permanent changes in my appearance that make me look unattractive
3 I believe that I look ugly.

15.
0 I can work about as well as before.
1 It takes an extra effort to get started at doing something.
2 I have to push myself very hard to do anything.
3 I can't do any work at all.

16.
0 I can sleep as well as usual.
1 I don't sleep as well as I used to.
2 I wake up 1-2 hours earlier than usual and find it hard to get back to sleep.
3 I wake up several hours earlier than I used to and cannot get back to sleep.

17.
0 I don't get more tired than usual.
1 I get tired more easily than I used to.
2 I get tired from doing almost anything.
3 I am too tired to do anything.

18.
0 My appetite is no worse than usual.
1 My appetite is not as good as it used to be.
2 My appetite is much worse now.
3 I have no appetite at all anymore.

19.
0 I haven't lost much weight, if any, lately.
1 I have lost more than five pounds.
2 I have lost more than ten pounds.
3 I have lost more than fifteen pounds.

20.
0 I am no more worried about my health than usual.
1 I am worried about physical problems like aches, pains, upset stomach, or constipation.
2 I am very worried about physical problems and it's hard to think of much else.
3 I am so worried about my physical problems that I cannot think of anything else.

21.
0 I have not noticed any recent change in my interest in sex.
1 I am less interested in sex than I used to be.
2 I have almost no interest in sex.
3 I have lost interest in sex completely.

Edmonton Symptom Assessment Scale

Capital Health

CARITAS HEALTH GROUP

Edmonton Symptom Assessment System: Numerical Scale
Regional Palliative Care Program

Please circle the number that best describes:

No pain	0	1	2	3	4	5	6	7	8	9	10	Worst possible pain
Not tired	0	1	2	3	4	5	6	7	8	9	10	Worst possible tiredness
Not nauseated	0	1	2	3	4	5	6	7	8	9	10	Worst possible nausea
Not depressed	0	1	2	3	4	5	6	7	8	9	10	Worst possible depression
Not anxious	0	1	2	3	4	5	6	7	8	9	10	Worst possible anxiety
Not drowsy	0	1	2	3	4	5	6	7	8	9	10	Worst possible drowsiness
Best appetite	0	1	2	3	4	5	6	7	8	9	10	Worst possible appetite
Best feeling of wellbeing	0	1	2	3	4	5	6	7	8	9	10	Worst possible feeling of wellbeing
No shortness of breath	0	1	2	3	4	5	6	7	8	9	10	Worst possible shortness of breath
Other problem	0	1	2	3	4	5	6	7	8	9	10	

Patient's Name _____

Date _____ Time _____

Complete by *(check one)*
- ☐ Patient
- ☐ Caregiver
- ☐ Caregiver assisted

BODY DIAGRAM ON REVERSE SIDE

Herth Hope Index

Herth Hope Index

	Strongly disagree	Disagree	Agree	Strongly agree
1. I am optimistic towards life	☐	☐	☐	☐
2. I have short and long term plans	☐	☐	☐	☐
3. I feel very lonely	☐	☐	☐	☐
4. I can see the possibilities amidst the difficulties	☐	☐	☐	☐
5. My faith comforts me	☐	☐	☐	☐
6. I am afraid of the future	☐	☐	☐	☐
7. I can remember happy and pleasant times	☐	☐	☐	☐
8. I feel very strong	☐	☐	☐	☐
9. I feel capable of giving and receiving affection/love	☐	☐	☐	☐
10. I know where I want to go	☐	☐	☐	☐
11. I believe in the value of each day	☐	☐	☐	☐
12. I feel my life is useful and worthy	☐	☐	☐	☐

Patient Dignity Inventory

Patient Dignity Inventory

For each item, please indicate how much of a problem or concern these have been for you within the last few days

1. Not being able to carry out tasks associated with daily living (e.g., washing myself, getting dressed).
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

2. Not being able to attend to my bodily functions independently (e.g., needing assistance with toileting-related activities)
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

3. Experiencing physically distressing symptoms (such as pain, shortness of breath, nausea).
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

4. Feeling that how I look to others has changed significantly.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

5. Feeling depressed.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

6. Feeling anxious.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

7. Feeling uncertain about my illness and treatment.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

8. Worrying about my future.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

9. Not being able to think clearly.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

10. Not being able to continue with my usual routines.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

11. Feeling like I am no longer who I was.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

12. Not feeling worthwhile or valued.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

13. Not being able to carry out important roles (e.g., spouse, parent).
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

14. Feeling that life no longer has meaning or purpose.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

15. Feeling that I have not made a meaningful and lasting contribution during my lifetime.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

16. Feeling I have 'unfinished business' (e.g., things left unsaid, or incomplete)
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

17. Concern that my spiritual life is not meaningful.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

18. Feeling that I am a burden to others.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

19. Feeling that I don't have control over my life.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

20. Feeling that my illness and care needs have reduced my privacy.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

21. Not feeling supported by my community of friends and family.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

22. Not feeling supported by my health care providers.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

23. Feeling like I am no longer able to mentally 'fight' the challenges of my illness.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

24. Not being able to accept the way things are.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

25. Not being treated with respect or understanding by others.
1 ☐ Not a problem 2 ☐ A slight problem 3 ☐ A problem 4 ☐ A major problem 5 ☐ An overwhelming problem

APPENDIX 6 – SEMI-STRUCTURED INTERVIEW SCRIPT

Semi-Structured Interview Guide – ALS Patients

Preamble

Thank you for taking the time to participate in an interview about your experiences.

I would like to remind you that your participation in this research is entirely voluntary. You do not have to answer questions if you do not want to. You may stop the interview at any time, for any reason. You may also withdraw your answers up to 90 days after the interview process ends. If you do so, we will destroy any record of your participation.

Your information will be kept confidential. Any quotations from interviews that we use in publications and presentations will not include your name but an identifier such as Participant 1. However, we cannot guarantee that someone would not be able to identify you based on your views.

I am going to ask you questions on your experiences and discussions with your healthcare team about your diagnosis and on-going care. I am then going to ask you questions about any conversations you have had with your healthcare team on choices at the end-of-life. Finally, I am going to ask you some general questions about any information sources you may have used to research ALS.

Questions

General background

1. Please tell me a little about yourself.
   [PROMPT: before ALS diagnosis]

Diagnosis

2. Please describe how you were diagnosed with ALS?
   [PROMPT: from symptoms to confirmatory diagnosis]
3. Who explained your diagnosis to you and, if you can remember, what was discussed during that visit?
   [PROMPT: what was explained well? What was not explained well? Was there something you wanted/needed the doctor to say/do that they did not?]
4. Was there someone other than a healthcare provider who was with you during your diagnosis? If so, what role did they play?

Care

5. Please describe what care you have been receiving since your diagnosis and by whom.
   [PROMPT: on disease course that may include changes in breathing and mobility]
6. Please describe how the changes in your life due to ALS and the associated care options for symptom management were explained to you and by whom?

[PROMPT: what was explained well? What was not explained well? Was there something you wanted/needed the healthcare provider to say/do that they did not? How prepared were you for the conversations?]

7. How has ALS affected your ability to communicate with your healthcare providers?
8. Have you participated in any clinical studies, and if so, what have they involved?
9. Was there someone other than a healthcare provider who accompanies you during your healthcare visits? If so, what role did they play?

Advanced disease state

10. Have you thought about the end-of-life choices you'd like to make?
    [PROMPT: influences on views about end of life, such as, spirituality, religion, culture, worldview]
11. Have you talked to a healthcare provider about your end-of-life care?
    - IF YES:
      - What triggered the conversations about end of life with your health care provider?
        [PROMPT: Do you bring it up initially, or did the health care provider?]
      - How did you feel talking about end-of-life with your health care provider?
      - What happened during the conversation?
    - IF NO:
      - Do you want to?
      - Who should start that conversation?
      - What would you want to know?
12. Have you talked to a health care provider about advance directives?
    - IF YES:
      - What happened during the conversation?
      - Do you feel adequately informed about advance directives?
    - IF NO:
      - Do you want to?
      - What would you want to know?
13. Was there someone who helped you while you were thinking about end of life issues? If so, what role did they play?

Medical aid in dying (if not already raised in answers to questions on advanced disease state)

The next few questions are about medical aid in dying, also known as physician assisted dying. We value all perspectives on this issue.

14. What are your views on medical aid in dying?
    [PROMPT: influences on views about medical aid in dying, such as, spirituality, religion, culture, worldview]
15. Have you talked to your healthcare provider about medical aid in dying?
    - IF YES:
      - What triggered the conversation about medical aid in dying with your health care provider?
        [PROMPT: Do you bring it up initially, or did the health care provider?]

        - How did you feel talking about medical aid in dying with your health care provider?
        - What happened during the conversation?
    - IF NO:
        - Do you want to discuss medical aid in dying?
        - Who should start that conversation?
        - What would you want to know?
16. [If relevant] Was there someone who helped you while you were thinking about medical aid in dying? If so, what role did they play?
17. How do you feel about media coverage of medical aid in dying and has it influenced your views on the issue?

Information gathering

18. What did you know about ALS before your diagnosis?
19. What sources of information, other than your healthcare provider, have you used to gather information about ALS?
20. How credible do you find those sources?
21. Have you discussed these sources with your healthcare provider?
    IF YES:
    - How did your healthcare provider respond to these sources?
22. Have you taken any actions or are you planning on taking any actions based on the information sources you have read?
    [PROMPT: clinical trial participation, stem cell clinic visits, lifestyle/nutrition modifications]
23. Has someone helped you access other sources of information or have you discussed these sources with others? If yes, can you describe those conversations?

End of Interview

24. Is there anything else you would like to add, or are there any questions we should have asked that we did not?

Thank you very much for your participation in the interview. We will contact you with out summary of the interviews and provide you with the opportunity to provide feedback on our understanding of your answers.

Semi-Structured Interview Guide – Caregivers

Preamble

Thank you for taking the time to talk to me about your experiences.

I would like to remind you that your participation in this research is entirely voluntary. If you do not feel comfortable answering a question, you may decline to do so. You may stop the interview at any time for any reason. You may also withdraw your answers up to 90 days after this interview. If you do so, we will destroy any record of your participation.

Your information will be kept confidential. Any quotations from interviews that we use in publications and presentations will not include your name but an identifier such as Caregiver 1. However, we cannot guarantee that someone would not be able to identify you based on your views.

I am going to ask you questions about your experiences caring for people with ALS throughout their disease course – from onset of symptoms and diagnosis to care at the end-of-life. The goal of the study is to improve communications between ALS patients, their caregivers and health care providers.

Questions

General background

1. Please tell me a little about yourself.

Diagnosis

2. Were you involved during [your _]'s diagnosis?
   [PROMPT: from symptoms to confirmatory diagnosis]
3. If yes, who explained the diagnosis to you and, if you can remember, what was discussed during that visit?
   [PROMPT: what was explained well? What was not explained well? Was there something you wanted/needed the healthcare provider to say/do that s/he did not?

Care

4. Please describe the care that your [*relationship to ALS patient, e.g., spouse*] has been receiving since his/her diagnosis and by whom.
   [PROMPT: care related to breathing, feeding and mobility]
5. Please describe how the disease course for ALS and the associated care options for symptom management were explained to you and by whom?
   [PROMPT: what was explained well? What was not explained well? Was there something you wanted/needed the healthcare provider to say/do that they did not? How prepared were you for the conversations?]
6. Has your [*relationship to ALS patient, e.g., spouse*] participated in any clinical studies, and if so, what have they involved for you?
7. What support have you received from healthcare providers in caring for your [*relationship to ALS patient, e.g., spouse*]?
   [PROMPT: on adequacy of support]

8. Are there others, outside of healthcare providers, to whom you turn for support and what role do they play?

Advanced disease state

9. Have you discussed end-of-life choices with your [*relationship to ALS patient, e.g., spouse*]?
   [PROMPT: influences on views about end of life, such as, spirituality, religion, culture, worldview]
10. Have you talked to a healthcare provider about end-of-life care for your [*relationship to ALS patient, e.g., spouse*]?
    a. IF YES:
        - What triggered the conversations about end of life with that health care provider?
          [PROMPT: Who brought it up initially?]
        - How did you feel talking about end-of-life with that health care provider?
        - What happened during the conversation?
    b. IF NO:
        - Do you want to?
        - Who should start that conversation?
        - What would you want to know?
11. Have you talked to a health care provider about advance directives?
    a. IF YES:
        - What happened during the conversation?
        - Do you feel adequately informed about advance directives?
    b. IF NO:
        - Do you want to?
        - What would you want to know?
12. Has someone other than a healthcare provider helped you think about end of life issues? If so, what role did they play?

Medical aid in dying (if not already raised in answers to questions on advanced disease state)

The next few questions are about medical aid in dying also known as physician assisted dying. We value all perspective on this issue.

13. What are your views on medical aid in dying?
    [PROMPT: influences on views about medical aid in dying, such as, spirituality, religion, culture, worldview]
14. Have you talked to your healthcare provider about medical aid in dying?
    a. IF YES:
        - What triggered the conversation about medical aid in dying with a health care provider?
          [PROMPT: Who initiated the conversation?]
        - How did you feel talking about medical aid in dying with the health care provider?
        - What happened during the conversation?

b. IF NO:
    - Do you want to discuss medical aid in dying?
    - Who should start that conversation?
    - What would you want to know?

15. [If relevant] Has someone other than a healthcare provider helped you think about end of life issues? If so, what role did they play?
16. How do you feel about media coverage of medical aid in dying and has it influenced your views on the issue?

Information gathering

17. What did you know about ALS before your [*relationship to ALS patient, e.g., spouse*]' s diagnosis?
18. What sources of information, other than your healthcare provider, have you used to gather information about ALS?
19. How credible do you find those sources?
20. Have you discussed these sources with your [*relationship to ALS patient, e.g., spouse*]'s healthcare provider?
    IF YES:
    - How did your healthcare provider respond to the sources?
21. Have you taken any actions or are you planning on taking any actions based on the information sources you have read?
    [PROMPT: clinical trial participation, stem cell clinic visits, lifestyle/nutrition modifications]
22. Has someone helped you access other sources of information or have you discussed these sources with others? If yes, can you describe those conversations.

End of Interview

23. Is there anything else you would like to add, or are there any questions we should have asked that we did not?

Thank you very much for your participation in the interview. We will contact you with out summary of the interviews and provide you with the opportunity to provide feedback on our understanding of your answers.

Semi-Structured Interview Guide – General Practitioners and Specialists

Preamble

Thank you for taking the time to talk to me about your experiences.

I would like to remind you that your participation in this research is entirely voluntary. If you do not feel comfortable answering a question, you may decline to do so. You may stop the interview at any time for any reason. You may also withdraw your answers up to 90 days after this interview. If you do so, we will destroy any record of your participation.

Your information will be kept confidential. Any quotations from interviews that we use in publications and presentations will not include your name but an identifier such as HealthCare Provider 1. However, we cannot guarantee that someone would not be able to identify you based on your views.

I am going to ask you questions about your experiences caring for people with ALS throughout their disease course – from onset of symptoms and diagnosis to care at the end-of-life. The goal of the study is to improve communications between ALS patients, their caregivers and health care providers.

Questions

General background

1. Please tell me a little about your history and medical practice.
   [PROMPT: position as generalist/specialist; training; length of time in practice]
2. [For General Practitioners] – What number of ALS patients have you seen in your past and current practice/work as a nurse?

Diagnosis – For General Practitioners and Specialists

3. How do you diagnose ALS?
4. What is your approach in communicating an ALS diagnosis?
   [PROMPT on the content of the communication and the development of the communications approach]
   - What do patients know about ALS when you give them their diagnosis?
5. What are the challenges in giving an ALS diagnosis, and how do you overcome these?
6. How do you respond to patient questions?

Care – For General Practitioners and Specialists

7. What is your approach in communicating about symptom management with ALS patients?
   [PROMPT on when?; Immediately following diagnosis (why?); upon a return visit]
8. What are the challenges in communicating about managing symptoms with patients and how do you overcome these?
9. What are the most difficult care interventions to talk about with patients?
10. How do you adapt your approach in response to progressive disability during the disease course?

11. How do you talk to caregivers about the patient's symptom management and ongoing care?

Advanced disease state

12. When do your communications shift from symptom management alone to end-of-life care?
13. How do you talk about end-of-life with patients?
14. How do you talk about end-of-life with caregivers?
15. What are the biggest challenges in talking about end-of-life with patients? With caregivers?
    [PROMPT on how challenges are overcome]

Medical aid in dying

The next few questions are about medical aid in dying also known as physician assisted dying. We value all perspective on this issue.

16. What are your views on medical aid in dying/ physician assisted death?
    [PROMPT: Is medical aid in dying something that should be offered as part of medical practice? Would you offer medical aid in dying as part of your practice?]
17. Have ALS patients and/or their caregivers raised the issue of medical aid in dying in the course of your practice?
    a. If yes, can you describe the conversation that took place?
    b. If no, how would you respond to a patient wishing to discuss medical aid in dying in your practice?
18. Do you feel adequately prepared to discuss medical aid in dying with ALS patients and their caregivers, and if not, what training or information do you feel you need?

External sources of information

We would now like to shift the interview to discuss the information gathering behaviour of patients and their caregivers.

19. What do patients and their caregivers understand about ALS at the time of diagnosis?
    [PROMPT: have patients/caregivers researched ALS as a possible diagnosis prior to the confirmatory visit?]
20. Which external sources of information do patients and their caregivers wish to discuss with you?
    [PROMPT: Internet, media, family/friends]
21. What is the content of that external information, and how is it understood by patients/caregivers?
    [PROMPT: clinical trials, stem cell therapies, medical aid in dying, other]
22. How do you respond to the questions brought to you by patients and their caregivers based on external information?

End of Interview

23. Is there anything else you would like to add, or are there any questions we should have asked that we did not?

Thank you very much for your participation in the interview. We will contact you with out summary of the interviews and provide you with the opportunity to provide feedback on our understanding of your answers.

Semi-Structured Interview Guide – Nurses

Preamble

Thank you for taking the time to talk to me about your experiences.

I would like to remind you that your participation in this research is entirely voluntary. If you do not feel comfortable answering a question, you may decline to do so. You may stop the interview at any time for any reason. You may also withdraw your answers up to 90 days after this interview. If you do so, we will destroy any record of your participation.

Your information will be kept confidential. Any quotations from interviews that we use in publications and presentations will not include your name but an identifier such as HealthCare Provider 1. However, we cannot guarantee that someone would not be able to identify you based on your views.

I am going to ask you questions about your experiences caring for people with ALS throughout their disease course – from onset of symptoms and diagnosis to care at the end-of-life. The goal of the study is to improve communications between ALS patients, their caregivers and health care providers.

Questions

General background

1. Please tell me a little about your history and work in healthcare.
   [PROMPT: training; current position; length of time in healthcare]
2. What number of ALS patients have you seen in your past and current work as a nurse?

Diagnosis

3. What kinds of questions do ALS patients and their caregivers ask you about their diagnosis?
4. How do you respond to these questions?
   [Prompt on which are most challenging]

Care

5. What is your approach in communicating about symptom management with ALS patients?
6. What are the challenges in communicating about managing symptoms with patients and how do you overcome these?
7. What are the most difficult care interventions to talk about with patients?
8. How do you adapt your approach in response to progressive disability during the disease course?
9. How do you talk to caregivers about the patient's symptom management and ongoing care?

Advanced disease state

10. How do you talk about end-of-life with patients?
11. How do you talk about end-of-life with caregivers?

12. What are the biggest challenges in talking about end-of-life with patients? With caregivers?
    [PROMPT on how challenges are overcome]

Medical aid in dying

The next few questions are about medical aid in dying also known as physician assisted dying. We value all perspective on this issue.

13. What are your views on medical aid in dying/ physician assisted death?
    [PROMPT: Is medical aid in dying something that should be offered as part of medical practice? Would you participate in a medically assisted death as currently allowed?]
14. Have ALS patients and/or their caregivers raised the issue of medical aid in with you?
    a. If yes, can you describe the conversation that took place?
    b. If no, how would you respond to a patient wishing to discuss medical aid in dying?
15. Do you feel adequately prepared to discuss medical aid in dying with ALS patients and their caregivers, and if not, what training or information do you feel you need?

External sources of information

We would now like to shift the interview to discuss the information gathering behaviour of patients and their caregivers.

16. Which external sources of information have patients and their caregivers wished to discuss with you?
    [PROMPT: Internet, media, family/friends]
17. What is the content of that external information, and how is it understood by patients/caregivers?
    [PROMPT: clinical trials, stem cell therapies, medical aid in dying, other]
18. How do you respond to the questions brought to you by patients and their caregivers based on external information?

End of Interview

19. Is there anything else you would like to add, or are there any questions we should have asked that we did not?

Thank you very much for your participation in the interview. We will contact you with out summary of the interviews and provide you with the opportunity to provide feedback on our understanding of your answers.

APPENDIX 7 – RECRUITMENT POSTER

Do you or someone you know have amyotrophic lateral sclerosis (ALS)?

ALS Care Decision Making: Understanding Clinician-Patient/Caregiver Communications

The Study: Researchers from the School of Public Health and the Faculty of Medicine and Dentistry, University of Alberta, are interested in understanding the information needs of ALS patients and their caregivers. Our goal is to improve communication between ALS patients/caregivers and their healthcare providers about an ALS diagnosis, ongoing care and end-of-life decisions.

Participants: We are recruiting ALS patients, their caregivers, and healthcare providers.

What will you be asked to do? We would like to speak with you about your perspectives on ALS Care decision-making, the information sources you rely on, and conversations with healthcare providers. We have approximately 20 questions that we would like to ask you. The interview would take approximately 1-1.5 hours in one sitting. However, we can be very flexible in how we conduct the interview. We can cover the questions in multiple sessions. You can also respond to the questions in your own time, using any method you choose, including email.

Confidentiality: Your identity will be kept confidential. We will not use your name or other identifying information in any publications or presentations based on our research.

Consent: Participation in this study is voluntary. If you decide to participate, your participation will in no way affect any clinical care.

Please Contact: Mackenzie Moir
School of Public Health
University of Alberta

APPENDIX 8 – ETHICS APPROVAL

Approval Form

Date:	August 19, 2016
Study ID:	Pro00064853
Principal Investigator:	Tania Bubela
Study Title:	*ALS Care Decision Making: Understanding Clinician-Patient/Caregiver Communications*
Approval Expiry Date:	Friday, August 18, 2017

Approved Consent Form:	Approval Date	Approved Document
	8/19/2016	Information Sheet - ALS Caregivers.docx
	8/19/2016	Information Sheet - ALS Patients.docx
	8/19/2016	Consent Form for Caregivers.docx
	8/19/2016	Consent Form.docx
	8/19/2016	Medical Information Release Consent Form.pdf
	8/19/2016	Information Sheet - Healthcare Providers.docx
	8/19/2016	Consent Form for Health Care Providers.docx

Sponsor/Funding Agency:	The James and Jeanie Brown ALS Research Fund		
Sponsor/Funding Agency:	Alberta Innovates Health Solutions	AIHS	Canada

Thank you for submitting the above study to the Health Research Ethics Board - Health Panel . Your application, including the following, has been reviewed and approved on behalf of the committee;

- ALS Study - Recruitment Poster (7/26/2016)
- ALS Study - Recruitment Sheet (7/26/2016)
- Patient Dignity Inventory (7/22/2016)
- Herth Hope Index (7/22/2016)
- ALS Depression Inventory (7/22/2016)
- Beck Depression Inventory (7/22/2016)
- ESAS (7/22/2016)
- Interview Questions - ALS Caregivers (7/26/2016)
- Interview Questions - ALS Patients (7/26/2016)
- Interview Questions - Healthcare Providers (7/27/2016)
- Research Protocol (7/26/2016)
- Chart Review Form (8/16/2016)

The Health Research Ethics Board assessed all matters required by section 50(1)(a) of the Health Information Act. Subject consent for access to identifiable health information is required for the research described in the ethics application, and appropriate procedures for such consent have been approved by the HREB Health Panel. In order to comply with the Health Information Act, a copy of the approval form is being sent to the Office of the Information and Privacy Commissioner.

A renewal report must be submitted next year prior to the expiry of this approval if your study still requires ethics approval. If you do not renew on or before the renewal expiry date (Friday, August 18, 2017), you will have to re-submit an ethics application.

Approval by the Health Research Ethics Board does not encompass authorization to access the patients, staff or resources of Alberta Health Services or other local health care institutions for the purposes of the research. Enquiries regarding Alberta Health approval should be directed to (780) 407-6041. Enquiries regarding Covenant Health approvals should be directed to (780) 735-2274.

Sincerely,

Anthony S. Joyce, Ph.D.
Chair, Health Research Ethics Board - Health Panel

Note: This correspondence includes an electronic signature (validation and approval via an online system).