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THE IMPACT OF DYSPHAGIA AND GASTROSTOMY ON QUALITY OF LIFE IN CAREGIVERS OF PATIENTS WITH AMYOTROPHIC LATERAL SCLEROSIS

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THE IMPACT OF DYSPHAGIA AND GASTROSTOMY ON QUALITY OF LIFE IN CAREGIVERS OF PATIENTS WITH AMYOTROPHIC LATERAL SCLEROSIS

THESIS

A thesis submitted in partial fulfillment of the requirements for the degree of Master of Science in Communication Sciences and Disorders in the College of Health Sciences at the University of Kentucky

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ABSTRACT OF THESIS

THE IMPACT OF DYSPHAGIA AND GASTROSTOMY ON QUALITY OF LIFE IN CAREGIVERS OF PATIENTS WITH AMYOTROPHIC LATERAL SCLEROSIS

There is little research studying the relationship between caregiver quality of life and gastrostomy, a palliative intervention recommended to manage dysphagia and malnutrition in patients diagnosed with Amyotrophic Lateral Sclerosis (ALS). To facilitate a more comprehensive assessment of treatment effectiveness and to better guide patients and their families, this study investigated the relationship between gastrostomy, caregiver strain, and patient disease-related factors. Patients with bulbar-onset ALS and their caregivers were recruited regardless of their decision to accept or decline future gastrostomy. Caregivers completed the Modified Caregiver Strain Index (MCSI) to assess levels of caregiver strain as an index of quality of life. Surveys were completed at 3-month intervals prior to gastrostomy and at a single time point following gastrostomy. Of 13 patient-caregiver dyads recruited, 1 dyad completed both phases of the study as of yet. This caregiver reported increased caregiver strain following gastrostomy. Medical interventions aimed at managing dysphagia, such as gastrostomy, may not have a predictable impact on caregiver strain, as indexed by the MCSI, or changes in caregiver strain may reflect characterological differences among patient-caregiver dyads. Other psychosocial factors within a given patient-caregiver dyad may be stronger predictors of caregiver strain, burden, and quality of life in caregivers.

KEYWORDS: Amyotrophic lateral sclerosis, dysphagia, gastrostomy, quality of life, caregiver strain, caregivers

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4/26/2019
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DEDICATION

To Zeke—I see the notes, you hear the melody, and together we enjoy the song.

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Chapter One: Introduction

Commensality, the act of eating with other people, is considered by many social scientists to hold a unique sociological, psychological, and anthropological value for human beings (Fischler, 2011; Sobal & Nelson, 2003). People often choose to share their daily meals in common meal despite being able to meet nutritional needs alone. Our selections in cuisine, methods of preparation, and commensal partners often extend beyond considerations of energy and nutritional nourishment and are inextricably linked to our social, cultural, and psychological quality of life (Barr & Schumacher, 2003). Given the deleterious effects on commensality by a disruption in an individual's ability to safely swallow, one would expect a resultant negative impact on quality of life. Amyotrophic lateral sclerosis (ALS) unequivocally does just this not only to the diagnosed individual but also to their partners, family members, and friends.

ALS is a progressive, terminal neurodegenerative disease characterized by the gradual denervation of motor neurons in the brain, brainstem, and spinal cord, ultimately resulting in paralysis of the voluntary muscles responsible for movement, speech, swallowing, and breathing (Wijesekera & Leigh, 2009). It is estimated that 5,600 individuals are diagnosed with ALS each year, with most individuals developing the disease between the ages of 40 and 70 years old (Robert Packard Center for ALS Research at Johns Hopkins, 2019). ALS is often described in three distinct clinical phenotypes depending on the origin of symptom onset within particular groups of muscles: spinal, bulbar, or respiratory (McDermott & Shaw, 2008). Bulbar-onset ALS, which follows a rapidly progressive and predictable course, is typified by symptoms emerging within the bulbar region of the brainstem, which houses the cranial nerves

responsible for speech, swallowing, and salivation (Hillel & Miller, 1989). The gradual loss of motor control in the bulbar musculature is considered, by some, to epitomize the most distressing and alienating symptoms of ALS: the loss of the ability to speak and swallow. Although bulbar symptoms are only part of the initial disease presentation in approximately 15-30% of individuals diagnosed with ALS, up to 85% of patients will eventually develop bulbar symptoms as motor neuron deterioration spreads to other muscle groups (Onesti et al., 2017). Onset of bulbar symptoms is a negative prognostic indicator, with some studies reporting a median survival range of 2 to 3 years from the earliest presentation of bulbar signs (Howard & Orrell, 2002; Mazzini et al., 1995; Stambler, Charatan, & Cedarbaum, 1998). Given the markedly reduced survival range and potentially traumatic symptoms experienced by patients diagnosed with bulbar onset ALS, it is imperative for medical recommendations and treatments to consider comfort and quality of life of the patient and their family.

Dysphagia, defined as difficulty in moving food or liquid from the mouth to the stomach, is an inevitable clinical symptom faced by individuals with bulbar onset ALS (Ruoppolo et al., 2013). Oral feeding ability becomes more inefficient and tiresome due to the progressive weakness and spasticity of the muscles involved in mastication and oral preparation (Rosenfeld & Strong, 2015). Concomitant impairments in limb mobility contribute to the individual's reduced ability to self-feed and maintain daily caloric needs. In addition to a reduction in the efficiency of swallowing, the safety of swallowing is simultaneously jeopardized. Deterioration in the respiratory, laryngeal, and pharyngeal musculature leads to reduced cough capacity and impaired airway protection, which

places the individual at risk for choking episodes, tracheal aspiration, and aspiration pneumonia (Park, Kang, Lee, Choi, & Kim, 2010).

While it is well established that dysphagia negatively impacts the swallowing-related quality of life of individuals diagnosed with ALS, it is unknown how dysphagia adversely affects quality of life in their caregivers as this assumption has been insufficiently studied (da Costa Franceschini & Mourao, 2015; Paris et al., 2013; Tabor, Gaziano, Watts, Robison, & Plowman, 2016). The responsibility of providing care to an individual diagnosed with ALS usually falls to the individual's partner or closest family member, and feeding was reported as the second most time-consuming task for caregivers of individuals with ALS (Chio et al., 2006). Additionally, the time devoted to caregiving, estimated to range from 5-15 hours per day, only increases with the progression of disease severity (Krivickas, Shockley, & Mitsumoto, 1997). This inevitability of increased caregiver strain for the caregiver highlights the importance of considering interventions that aim to improve, or at least maintain, the quality of life for both the patient and informal caregiver (Lo Coco et al., 2005).

While there is currently no known cure for ALS, various palliative interventions are employed to alleviate symptoms, prolong survival, promote independence, and maintain quality of life (Kiernan et al., 2011; McDermott & Shaw, 2008; Miller et al., 2009). To manage early dysphagic symptoms, compensatory strategies, including postural adjustments and dietary modifications, are recommended to increase the safety and efficiency of swallowing and maintain adequate nutrition and hydration (Wijesekera & Leigh, 2009). As frequently reported in the literature, management of dysphagia often falls upon a spouse or close family member of the patient due to the progressive physical

limitations and fatigue characteristic of the disease as well as the prohibitive costs of employing a professional caregiver (Stavroulakis et al., 2014, 2016; M. Trail, Nelson, Van, Appel, & Lai, 2004). Informal caregivers, who likely have very little training in dysphagia management, find themselves responsible for managing and troubleshooting the difficulties associated with eating and swallowing. Caregivers may experience increased caregiver strain due to the demands of food preparation for frequent small meals, compounding the substantial amount of time and effort already devoted to other routine care of the patient. Anecdotal and clinical observations suggest that family of individuals with dysphagia may also experience fear and anxiety due to the risk of their loved one choking or aspirating during a meal and the need for the caregiver to be physically present during all meals to assist with feeding or intervene if choking (Kasarskis, 2017; Stavroulakis et al., 2014). Mealtimes that were once a pleasurable, social experience devolve into a prolonged, taxing endeavor for both the individual diagnosed with ALS and their caregiver.

As dysphagic symptoms worsen in the presence of deteriorating bulbar musculature, early symptomatic management of dysphagia becomes less effective (J. Johnson et al., 2012; Radunović, Mitsumoto, & Leigh, 2007). The association between weight loss, malnutrition, and shortened survival in individuals with ALS has been previously demonstrated (Desport et al., 1999; Limousin et al., 2010). To counter the negative effects of malnutrition and weight loss, the American Academy of Neurology (AAN) recommends gastrostomy as an alternative or supplemental route to oral feeding (Miller et al., 2009; Miller et al., 1999). Current practice guidelines recommend gastrostomy tube insertion in patients with worsening dysphagia in the presence of

inability to maintain caloric requirements, weight loss greater than 10% of premorbid body weight, or forced vital capacity of 50% less than predicted value (Kasarskis et al., 2014).

The purported benefits of gastrostomy include weight stabilization, improved nutritional and fluid intake, and a safer route for medication; however, the evidence base on prolonging length of survival is equivocal and lacking in randomized controlled trials (Burkhardt, Neuwirth, Sommacal, Andersen, & Weber, 2017; Desport et al., 1999; Katzberg & Benatar, 2011; Mazzini et al., 1995; McDonnell, Schoenfeld, Paganoni, & Atassi, 2017; Mitsumoto et al., 2003; ProGas Study Group, 2015; Radunović et al., 2007; Spataro, Ficano, Piccoli, & La Bella, 2011; Stavroulakis, Walsh, Shaw, McDermott, & ProGas Study Group, 2013). Additionally, the impact of this specific medical intervention on quality of life in individuals with ALS and their caregivers lacks rigorous examination even though quality of life may be considered of critical importance in the discussion for or against gastrostomy. In the most recent Cochrane Review of enteral tube feeding in individuals diagnosed with ALS, only 20% of those individuals who met the criteria for enteral support actually accepted gastrostomy tube insertion (Katzberg & Benatar, 2011). This suggests that both physicians and patients are ambivalent about the value of gastrostomy. Perhaps the decision to proceed with gastrostomy must extend beyond objective medical criteria and consider psychological and psychosocial factors of the patient and their family (Brotherton & Abbott, 2009; Stavroulakis et al., 2013). In the face of terminal illness as overwhelming as ALS, quality of life must be considered a critical focus of our clinical care of this vulnerable population (Hardiman, Hickey, & O'Donerty, 2004).

Statement of the Problem

The decision to accept or decline gastrostomy must be guided by the preferences of the individual and their family, goals of treatment, and outcome data (DeLegge et al., 2005). ALS is a devastating disease for patients and their family members, and healthcare professionals need to provide clear, objective information regarding the anticipated benefits and burdens of gastrostomy (Squires, 2006). As of yet, the impact of gastrostomy on quality of life in individuals diagnosed with ALS and their caregivers has been insufficiently researched. Lacking an evidence-based understanding of the impact of gastrostomy on quality of life effectively inhibits informed, autonomous decision making among patients and their families. Due to the lack of effective pharmacotherapy, treatments that maximize quality of life in the individual and their family are critically important (Galvin et al., 2015; McDermott & Shaw, 2008). Physiologic improvement, e.g., increased weight gain, nutrition, hydration, and length of survival, in the absence of psychologic improvement, e.g., quality of life, may be perceived as little benefit for some individuals (Kurien et al., 2017). And worse, it is possible that for some individuals, gastrostomy effectively prolongs a debilitating state of reduced quality of life (Simmons, 2015). It is vital to obtain a comprehensive assessment of the treatment effectiveness of gastrostomy, including how this specific medical intervention impacts the quality of life in all individuals involved in the healthcare decision-making process (both patients and their caregivers).

Social support has been rated as one of the biggest determinants of quality of life in patients diagnosed with ALS, and multiple studies have documented the interdependence of patient and caregiver quality of life (Boerner & Mock, 2012;

Simmons, Bremer, Robbins, Walsh, & Fischer, 2000). Medical treatments that alleviate caregiver strain may improve the quality of care and social support provided to the patient. Gastrostomy is often recommended with the anticipation of a subsequent alleviation in the caregiver's physical burden of meal preparation and emotional burdens such as anxiety and fear surrounding the patient's swallowing and nutritional status. Quality of life in both patients and caregivers needs to be investigated as an outcome measure of treatment to remain consistent with the holistic person- and family-centered approach fundamental to palliative care (Hughes, Sinha, Higginson, Down, & Leigh, 2005; S. Johnson et al., 2017). Research into the effect of gastrostomy on caregiver quality of life aims to accomplish a more comprehensive understanding of the medical intervention's effectiveness and efficiency and, simultaneously, carer-proof the decision-making process (Al-Janabi, Nicholls, & Oyebode, 2016). Medical interventions that increase caregiver strain may decrease the effectiveness of the care they can provide and ultimately result in a cascade of negative ramifications. Thus, it is imperative to obtain a comprehensive assessment of the impact of gastrostomy on all individuals involved in the decision-making process.

Purpose of the Study

The AAN has called for further investigation via prospective trials to guide healthcare professionals in the treatment of individuals with ALS (ProGas Study Group, 2015). There is a need for a rigorous investigation into how gastrostomy impacts quality of life in caregivers of patients with ALS. This study ultimately aims to contribute to the understanding of the treatment efficacy of gastrostomy to better guide patients and their families in the decision to accept or decline gastrostomy and optimize the timing of

clinical recommendations for gastrostomy tube insertion. This study evaluates the impact of gastrostomy on quality of life in caregivers of patients with ALS and dysphagia. In the absence of randomized controlled trials, this study employed a prospective, longitudinal investigation into the quality of life of caregivers of patients diagnosed with ALS and neurogenic dysphagia through the decision to accept or decline gastrostomy.

Chapter Summary

Chapter 1 served to introduce dysphagia in individuals diagnosed with ALS, the impact of dysphagic symptoms on the individual and their family, and the current clinical recommendations and its implications on quality of life in both individuals diagnosed with ALS and their caregivers. Chapter 2 will comprise a review on the current literature investigating quality of life in caregivers of individuals with ALS and neurogenic dysphagia.

Chapter Two: Literature Review

The purpose of this literature review is to assess the extant literature relevant to determining the relationship between gastrostomy and quality of life in caregivers of patients with ALS and neurogenic dysphagia.

Caregiver Quality of Life

Caregiving is considered a "fertile ground for persistent stress" (Pearlin, Mullan, Semple, & Skaff, 1990, p. 583). The importance of considering the physical and emotional wellbeing of the caregiver of an individual diagnosed with a terminal illness can hardly be overstated. Caregivers who become overly burdened or stressed are at increased risk of compromises in their physical and/or emotional health, which impacts the care they provide to the care recipient (Cousins, Davies, Turnbull, & Playfer, 2002) . Although subsequent sections of this literature review accomplish an in-depth exploration of quality of life in caregivers of patients with ALS and/or dysphagia, some brief points on the broader topic of caregiving are necessary.

Scientific pursuits in caregiving research lack consensus in conceptualization and operationalization (Chou, 2000). Caregiver burden, caregiver strain, caregiver distress, and caregiver stress are often used interchangeably within the terminology of caregiver outcomes, and there is little agreement on the individual definitions of these terms (Braithwaite, 1992; Thornton & Travis, 2003). Consequently, there is great variability in conclusions regarding the main determinants of caregiver burden, caregiver strain, and caregiver distress and a simultaneous hindrance in the devising of suitable assessments to anticipate or identify vulnerable caregivers. Some studies reported patient-specific

impairments, disease progression, and duration spent in the role of caregiving were most influential in levels of caregiver burden and strain (Crespo, Lopez, & Zarit, 2005; Zarit, 2004). Other authors have disagreed, arguing that caregiver outcomes are more related to the variables that the caregiver brings to the caregiving situation, including personality traits and mental states (Burke et al., 2018; Cousins et al., 2002; Cumming & De Miranda, 2012; S. Johnson et al., 2017; Simmons et al., 2000; Zanetti et al., 1998). Poulshock and Diemling argued that caregiver burden could be conceived of as a psychological concept wherein "burden" is constituted by a caregiver's subjective interpretation of how their wellbeing is impacted by the difficulties pursuant to the patient's impairments and demands of the caregiving role (as cited in Cousins et al., 2002, p. 388). Viewing caregiver burden as a subjective phenomenon lends support to the findings of vastly different caregiver outcomes in similar caregiving situations.

Regardless of the lack of consensus in the caregiving literature, the caregiver's experience of increased caregiver burden/strain, reduced quality of life, and the increased propensity for this sequalae to surface during long-term care situations should not be minimized. Some authors conceived of the relationship between caregiver characteristics, caregiver strain, and caregiver quality of life within a "chains-of-risk" model (Litzelman et al., 2015). Caregiver strain is a function of a cluster of risk factors encompassing caregiver characteristics, demographics, and environmental resources. The accumulation of these risk factors through increased caregiver strain negatively impacts the health and quality of life of the caregiver. Previous studies highlighted the need to conduct early assessments of self-perceived levels of strain, burden, and quality of life in caregivers within long-term caregiving situations, and intervene before caregiver outcomes reach

clinically significant levels (Al-Janabi et al., 2016; Thornton & Travis, 2003). Further analysis into the conceptualization and operationalization of burden, strain, distress, and stress in caregivers of patients with chronic illnesses is beyond the scope of this study. For the purposes of this study, caregiver strain was considered an index of quality of life in caregivers. What follows is an exploration of the literature evaluating caregiver strain, burden, emotional and psychosocial characteristics, and quality of life in caregivers of patients with ALS and/or dysphagia.

Quality of Life in Caregivers of Patients with ALS

The concept of quality of life is multifaceted, broad-ranging, and difficult to define as either a quantitative or qualitative outcome measure (Hardiman et al., 2004). Understandably then, researchers have employed a variety of instruments in studying it. Pursuant to methodological disparities, it is difficult to discern and track commonalities in research results. However, there is broad consensus among researchers that quality of life among caregivers of patients with ALS may be significantly compromised by the challenges associated with the care of a patient suffering from a neurological disease (S. Johnson et al., 2017).

Much of the literature involving caregivers of patients with ALS has focused on how the disease and its inevitable progression may impact caregiver quality of life, psychosocial wellbeing, and levels of caregiver burden. The majority of studies have shown that the impact on caregiver quality of life has been negative. Investigators have found that the disease may negatively impact the caregivers' physical or psychological well-being (Krivickas et al., 1997). Significant levels of caregiver burden have been reported, and this burden is likely to increase as the disease worsens (Chio, Gauthier,

Calvo, Ghiglione, & Mutani, 2005; Gelinas, O'Connor, & Miller, 1998). According to Burke et al., approximately 50% of caregivers of patients with ALS report clinically significant levels of burden, highlighting the importance of considering caregiver burden and investigating interventions that may alleviate burden and improve quality of life in caregivers (Burke et al., 2017). Self-perceived quality of life among caregivers is adversely affected, and their satisfaction with life may significantly decrease (Bromberg & Forshew, 2002; Jenkinson, Fitzpatrick, Swash, & Peto, 2000; Lo Coco et al., 2005; Roach, Averill, Segerstrom, & Kasarskis, 2009).

However, other studies have reported that caregivers of patients with ALS rate their quality of life as fairly good (Marilyn Trail, Nelson, Van, Appel, & Lai, 2003). According to one study, while caregiver distress, anxiety, and depression were observed to increase over the course of the disease, these ratings did not meet pathological levels (Gauthier et al., 2007). Other studies have sought to understand if there is concordance between patient and caregiver in terms of quality of life and psychological factors; however, no broad consensus has emerged from these studies (Chio et al., 2005; Goldstein, Atkins, Landau, Brown, & Leigh, 2006; Jenkinson et al., 2000; Lo Coco et al., 2005; Olsson Ozanne, Strang, & Persson, 2010; Rabkin, Wagner, & Del Bene, 2000). Other literature is largely dedicated to investigating the contributors, predictors, and protective factors of caregiver quality of life, burden, strain, and distress rather than the impact of medical interventions such as gastrostomy (Burke et al., 2017; Burke et al., 2018; Chio et al., 2004; Gauthier et al., 2007). Due to the varied findings of quality of life in caregivers of patients with ALS, there is a need for a systematic study evaluating the burden of disease progression on caregivers.

While the aforementioned literature sets out to explore quality of life and burden of caregivers of patients with ALS in a broader context, there are fewer studies exploring how dysphagia specifically impacts caregiver quality of life. Additionally, there is very little research studying the nature of the relationship between caregiver quality of life and palliative interventions that manage dysphagia. In the subsequent sections, previous research into quality of life is explored from the unique perspective of caregivers who provide informal care to individuals with dysphagia, beginning first with literature on dysphagia as a result of non-ALS diagnoses and then leading into dysphagia due to ALS.

Quality of Life in Caregivers of Patients with Dysphagia

In studies conducted with caregivers of patients with dysphagia following stroke and traumatic brain injury, caregivers described how the cooking and serving of food became a vitally important task (Johansson & Johansson, 2009). They would spend a significant amount of time preparing a meal that was simultaneously safe, nourishing, and appetizing. For individuals who provide care to patients with dysphagia following non-surgical management for head and neck cancer, dysphagia can produce activity limitations and participation restrictions (Nund et al., 2014). Through thematic analysis of semi-structured interviews with 12 caregivers of patients with dysphagia without gastrostomy, researchers identified clinically significant levels of distress and decreased quality of life. Caregivers reported a sense of ill-preparedness when it came to the fulfilling their role in the management of dysphagia and its negative impact on meal preparation, social activities, and daily life as a family.

In patients with dysphagia who underwent chemoradiotherapy for treatment of head and neck cancer, 19 caregivers were asked to complete the Caregiver Quality of

Life Index-Cancer tool (CQOL-C) (Patterson, Rapley, Carding, Wilson, & McColl, 2013). Caregiver-dyads were only included in the study if the caregivers participated in meal provision and if patients reported experiencing dysphagia, as indexed by a score of less than 80 points on the M.D. Anderson Dysphagia Inventory (MDADI). No significant differences were observed in caregiver quality of life between pre-treatment and 3-months post-treatment; however, a significant improvement was observed in the 'burden' domain of the CQOL-C between three- and 12-months post-treatment. Authors reported a gradual decline in the number of patients who were reliant on gastrostomy during this period: 17 patients had gastrostomy at three-months post-treatment and only 11 patients had gastrostomy at 12-months post-treatment. Caregivers of patients who were gastrostomy-dependent at three-months post-treatment and 12-months post-treatment reported significantly lower quality of life scores on the CQOL-C. Authors concluded that with the increased requirements in the physical care of the patient's dysphagia comes with reduced quality of life reported in caregivers.

Dysphagia was associated with increased caregiver burden in all four studies selected for systematic review, although a divergence was observed in the tools used to measure caregiver burden (Namasivayam-MacDonald & Shune, 2018). Increases in general burden levels for caregivers was associated with self-reported difficulties in swallowing, gradual decline in feeding behaviors, and the operation of feeding tubes. Self-reported levels of emotional and psychological burden were related to the caregiver's concerns over the patient's nutritional intake, counterbalancing choking risks with desirable foods, and the conflicting process of accepting gastrostomy as eventuality. The authors of this systematic review highlighted the importance of understanding how

dysphagia may contribute to caregiver burden and the need for successful, evidence-based interventions that alleviate caregiver burden and, therefore, improve the quality of life of both the patient and the caregiver.

Building on the momentum of this systematic review, the same authors conducted a cross-sectional study of community-dwelling elderly adults and their spousal caregivers to further elucidate the type (financial, emotional, and/or physical burden) and degree of caregiver burden experienced by spouses of individuals with dysphagia (Shune & Namasivayam-MacDonald, 2019). Of the 422 care recipients surveyed, 17% of care recipients reported swallowing difficulties, and a significant association was found between the care recipient's self-reported dysphagia status and their respective caregiver's increased levels of emotional burden. For caregivers of spouses with swallowing difficulties, 70% of caregivers endorsed moderate to severe levels of emotional burden, 50% reported feeling depressed or hopeless within the past month, and 60% indicated their personal time was decreased due to the time devoted to caregiving. Despite the inherent limitations of self-reporting and cross-sectional analyses, clear methodological weaknesses were noted. Dysphagia status was determined via a single question posed to the care recipient: "any problems while chewing or swallowing while eating in the past month?" (Shune & Namasivayam-MacDonald, 2019, p. 5). Also, the presence or absence of caregiver burden was evaluated via a simple 5-point Likert scale. However, the results of this study cannot be ignored—dyadic health needs to be prioritized since the health and well-being of the caregiver and the care recipient are interdependent. Authors intimated the need for further longitudinal investigation into this association: what caregiver variables, care recipient variables, and/or dyadic variables

contribute to, or moderate, the relationship between the presence of dysphagia in care recipients and increased levels of emotional burden in caregivers?

Other authors evaluated the anxiety levels of caregivers of patients with neurological disorders, both with and without neurogenic dysphagia (Serel Arslan, Demir, & Karaduman, 2017). Based on the presence of dysphagia, participants were assigned to either the study group (dysphagic) or the control group (non-dysphagic). Of particular relevance to the present study, 24 of the 103 adult neurological patients assigned to the dysphagic study group were diagnosed with ALS. Results showed that caregivers of neurological patients with dysphagia reported higher momentary and long-lasting anxiety levels compared to caregivers of neurological patients without dysphagia. However, surprisingly, no relationship was observed between anxiety levels of caregivers and the severity of patients' dysphagia, types of feeding, or the degree of dependency of patient to caregiver. The authors concluded that the additional burden dysphagia placed on caregivers may be explained by four factors including chronic disruption in daily life of both caregiver and patient: diminution of valued social aspects of mealtimes, education of the caregiver and designation of the caregiver as the primary bearer of responsibility for the management of dysphagic symptoms in the patient, and potential feelings of insecurity or incapability in regard to administration of safe oral intake and symptom-related problem solving. This is highly relevant to our current research as it is the first study that includes a population of ALS caregivers (albeit only a small subset of the sample) when investigating the impacts of dysphagia on the caregiver. This study emphasizes the vital role caregiver involvement plays in dysphagia management, and

suggests that large, prospective studies are needed to evaluate the impact of dysphagia on ALS caregivers and potential interventions that alleviate caregiver burden.

Quality of Life in Caregivers of Patients with ALS and Dysphagia

Although the impact of dysphagia on quality of life of ALS caregivers has not undergone rigorous examination, some insight has surfaced through qualitative, retrospective studies that investigated the perspectives of patients, caregivers, and healthcare professionals regarding the decision to proceed with or forgo gastrostomy.

In a retrospective study exploring the perspectives of 10 patients and eight caregivers regarding the decision to proceed with gastrostomy insertion, participants described how the challenges of food preparation had a negative impact on both the caregiver's and the patient's quality of life (Stavroulakis et al., 2014). Altering food consistencies and thickening liquids was described as time consuming and, according to one patient-caregiver dyad, restricted their ability to travel since "the hotel couldn't handle it… they couldn't prepare the food in the way that was necessary." Other factors that were influential in the decision to proceed with gastrostomy insertion included significant weight loss, prolonged and effortful meals, and reduced swallow safety with frequent occurrences of choking, aspiration, and chest infection. While this small sample provided unique insight into the decision-making process of gastrostomy, it also highlighted the need for further investigation of the potential benefits of gastrostomy to assist patients and caregivers in making informed, timely decisions on whether they wished to proceed or forgo gastrostomy tube insertion.

In another study qualitative study, researchers interviewed healthcare professionals of patients with ALS to identify the diverse factors involved in the decision-making process of gastrostomy and discovered that the perceived effect on the patient's quality of life and the impact of the procedure on the family was pivotal in the decision to accept or decline gastrostomy (Martin et al., 2016). Many healthcare professionals reported the patient agreed to the gastrostomy procedure due to the positive impact they hoped it might have on their family while other healthcare professionals considered the patients' fear of the negative impact of gastrostomy on their family to be a decisive factor in the refusal of the intervention. For example, one healthcare professional stated, "[the caregiver] does everything for her… He's worn out, he's tired… I think it probably did [affect her decision]… I don't think she could put another thing on [the caregiver]" (Martin et al., 2016, p. 1372). The selection of healthcare professionals who were interviewed in this study was determined by the patients themselves; thus, the generalizations of these findings are limited. However, the authors provided valuable insights to support people with ALS and their caregivers. Ultimately, the authors' work implied a continued need for more evidence-based research on the impact of gastrostomy on the quality of life of patients and caregivers to facilitate informed decision-making regarding gastrostomy as a medical intervention.

Gastrostomy and Quality of Life in Caregivers of Patients with ALS

There is a paucity of research exploring the impact of gastrostomy on quality of life in patients with ALS and the findings, thus far, are equivocal (Katzberg & Benatar, 2011; Körner et al., 2013; Mazzini et al., 1995; McDonnell et al., 2017; Mitsumoto et al., 2003; ProGas Study Group, 2015; Zamietra et al., 2012). Similarly, there is a dearth of

research regarding the effects of gastrostomy on the caregiver's quality of life. Two retrospective studies and one prospective study are explored below.

Mitsumoto et al. conducted a stratified case-control study by retrospectively analyzing data derived from the ALS Patient Care Database (Mitsumoto et al., 2003). No significant differences were observed in caregiver burden between caregivers of patients with gastrostomy tubes and caregivers of patients without gastrostomy tubes; however, the instrument used to evaluate caregiver burden remained unclear. Authors concluded that these findings could be explained by either of the following observations: patients who proceeded with gastrostomy were farther along in disease progression and required greater amounts of care, or the instrument used to assess caregiver burden may not have been sensitive to the changes brought about by gastrostomy tube insertion. To properly investigate the question of whether gastrostomy tube insertion and use reduces the burden in caregivers, a prospective study utilizing sensitive, specific instruments to evaluate caregiver burden is needed.

In a retrospective, qualitative exploration, patients and caregivers described both challenges and benefits of gastrostomy (Stavroulakis et al., 2016). In the absence of alternatives, patients and caregivers described the potential benefits as outweighing the negatives; although, significant psychosocial concerns were noted. The benefits of gastrostomy included anxiety relief in both patients and caregivers, weight stabilization in patients, and prolonged survival. Researchers noted that concerns among some patient-caregiver dyads included the inability of patient and caregiver to share a meal, and its potential contribution to feelings of social isolation and depression, as well as caregiver discomfort and/or guilt when eating in the patient's presence, and finally the time and

burden associated with food administration and maintenance of gastrostomy equipment. These findings, although providing useful insight into the psychosocial concerns associated with gastrostomy, are limited due to small sample size as well as their retrospective character—findings were based on interviews with patients after they elected to proceed with gastrostomy. Another limitation of the study may explain, at least in part, the reason for inability to share a meal together; all patients interviewed in this study opted for late gastrostomy insertion and none maintained oral intake following the procedure. Earlier gastrostomy tube insertion may have yielded different results. The psychosocial impact of gastrostomy may be different in patients who elect for early gastrostomy insertion in anticipation of bulbar symptoms, and/or patients who opt to employ a combination of gastrostomy and oral intake for as long as tolerated.

Previous authors described how combining oral intake with enteral feeding after gastrostomy insertion may be beneficial for patients to gradually adapt to their new lifestyle and simultaneously improve their nutritional status while prolonging survival (Spataro et al., 2011; Squires, 2006). Patients may continue oral intake for pleasure until it becomes too distressing or they are unable to initiate a swallow. In the University of Kentucky ALS Multidisciplinary Clinic, patients are encouraged to continue eating and drinking as tolerated and for pleasure in addition to using the feeding tube. Future research should include a large sample of patients-caregiver dyads enrolled *prior* to the gastrostomy decision and followed longitudinally to further elucidate the impact of gastrostomy on patient and caregiver quality of life.

In a large, multi-center, prospective, longitudinal study, investigators recruited 345 patients with ALS and their caregivers to evaluate the changes in patient self-

perceived quality of life and caregiver strain following gastrostomy (ProGas Study Group, 2015). At baseline, before the gastrostomy procedure, and at 3-months following gastrostomy, patients completed the McGill Quality of Life questionnaire, and informal caregivers completed the Modified Caregiver Strain Index (Thornton & Travis, 2003). While no significant changes were discovered in patients' quality of life following gastrostomy, caregivers reported significantly greater levels of caregiver strain. Authors concluded that the observed increase in caregiver strain may be explained by the progressive nature of ALS. The motor disability of patients continues to increase due to gradual denervation of motor neurons, increasing their physical dependence on the caregiver. Additionally, authors did not offer statistical comparisons between caregiver strain and disease-related patient factors pre- and post-gastrostomy. It should also be noted that patient-caregiver dyads were enrolled as study participants only after agreeing to proceed with gastrostomy insertion. These aspects of the study confound the conclusion that can be made as to the factors related to increased caregiver strain following gastrostomy insertion.

The hypothesis that gastrostomy improves quality of life in caregivers by reducing caregiver burden has yet to be rigorously tested in a large, prospective study. Furthermore, studies with documented levels of increased caregiver burden following gastrostomy in patients with ALS have yet to be replicated. The current study would take up this task with respect to gastrostomy intervention and its impact on caregiver strain as an index of quality of life in caregivers of patients with ALS. Global or health-related quality of life instruments may not be sensitive to the specific changes in caregiver quality of life as impacted by gastrostomy, relative to caregiver burden. To understand

the specific factors that impact a caregiver's quality of life, investigators must employ instruments that are sensitive to these factors (Simmons, 2015).

Research Hypotheses

1. Following gastrostomy, caregiver strain, as indexed by scores on the Modified Caregiver Strain Index (MCSI), will decrease when compared to levels of caregiver strain prior to gastrostomy.
2. Caregiver strain will be positively correlated with the severity of the patient's functional impairments due to disease progression, as indexed by scores on the Amyotrophic Lateral Sclerosis Functional Rating Scale Revised (ALSFRS-R).
3. Prior to gastrostomy, caregiver strain will be positively correlated with the severity of the patient's self-reported symptoms of dysphagia, as indexed by scores on the Eating Assessment Tool (EAT-10). Increased severity of the patient's dysphagia will be associated with increased levels of caregiver strain.
4. Prior to gastrostomy, caregiver strain will be negatively correlated with patient-reported swallowing-related quality of life, as indexed by scores on the Swallowing Quality of Life Questionnaire (SWAL-QOL). Decreased swallowing-related quality of life in patients will be associated with increased levels of caregiver strain.

Chapter Summary

Chapter 2 reviewed the current literature on quality of life in caregivers, including individuals diagnosed with ALS and/or dysphagia, and the small amount of available

research on the impact of gastrostomy on caregiver quality of life. Chapter 3 outlines the methodology of the study.

Chapter Three: Methodology

The study was conducted within the University of Kentucky ALS Multidisciplinary Clinic, located at the Kentucky Neuroscience Institute. The Institutional Review Board (IRB) at the University of Kentucky (UK) approved participant recruitment and survey administration procedures.

Design

The study's observational, prospective, longitudinal design examined how gastrostomy impacts self-reported levels of caregiver strain in caregivers of patients diagnosed with ALS and neurogenic dysphagia. There are understandable ethical limitations of withholding a medical intervention; thus, patients and caregivers who met the inclusion criteria were recruited regardless of their decision to accept or decline gastrostomy in the future. Patients and their caregivers were enrolled in the study and followed throughout the disease progression, potentially in the presence of worsening dysphagic symptoms and the impending decision of whether to accept or decline gastrostomy.

Patients were blinded to the survey responses of caregivers. Due to impaired limb mobility, some patients were unable to complete their surveys independently outside of clinic and had to rely on caregivers for assistance to complete the surveys. In this case, a caregiver was not blinded to the survey responses of the patient. Research personnel anticipated if a patient required assistance to complete surveys and instructed caregivers to not influence the patient's survey responses.

Participants

A total of 26 participants, or 13 patient-caregiver dyads, were recruited to participate in the study from June 2018 to March 2019. To date, one patient-caregiver dyad completed surveys during both phases of the study— prior to gastrostomy and following gastrostomy. The results of this patient-caregiver dyad were presented as a single case study, with further results from other dyads pending.

Patient-caregiver dyads were recruited from a convenience sample of patients who were diagnosed with ALS and receiving healthcare services through the UK ALS Multidisciplinary Clinic. A patient and their caregiver were enrolled in the study together as a dyad; hence both the patient and caregiver were required to meet specific inclusion criteria. Patients were eligible to participate in the study based on the following five inclusion criteria: (1) diagnosis of ALS, (2) presence of bulbar symptoms, (3) consumption of an oral diet with no alternative means of nutrition, (4) no evidence of cognitive impairment, and (5) reliance on an informal caregiver for care and assistance (see Appendix A). For the first inclusion criterion, patient must have a diagnosis of ALS as defined by the revised El-Escorial diagnostic criteria (Brooks, Miller, Swash, & Munsat, 2000). Regarding the second criterion, the presence and severity of bulbar symptoms were evaluated by the neurologist and documented using the revised questionnaire-based ALS Functional Rating Scale (ALSFRS-R, Cedarbaum et al., 1999). Within the ALSFRS-R, the domain of bulbar functions encompasses three component scales: speech, salivation, and swallowing. Bulbar symptoms were considered present if a patient received a score of less than 4 on any of the three bulbar component scales. The third criterion for inclusion in this study specified the requirement for the patient to be

currently consuming an oral diet and not engaging in non-oral means of nutrition, i.e., the patient cannot already have a gastrostomy tube in place. The fourth inclusion criterion was based on the discretion of research personnel, and patient-caregiver dyads were excluded if the patient and/or caregiver exhibited overt cognitive impairment or appeared too unwell to participate in the study. The fifth and final inclusion criterion for the patient to participate in the study coincided with the single inclusion criterion for the caregiver—a caregiver was eligible to participate in the study if they were an adult individual (at least 18 years of age) who provided unpaid care and assistance to the patient with ALS. If the patient reported relying on multiple caregivers, the primary caregiver, as indicated by the most time devoted to care and assistance, was recruited to participate. Formal caregivers, defined as adult individuals who performed caregiving duties for monetary reimbursement, were excluded from the study.

Patient-caregiver dyads who met the above inclusion criteria were invited to participate in the study during their regularly scheduled clinic visit at the UK ALS Multidisciplinary Clinic. Informed consent was obtained from patients and caregivers who agreed to participate by signing consent forms approved by the UK IRB (see Appendix B). Participants were briefed on the purpose of the study, estimated time commitment to complete surveys, expected timeline of the study, deidentification and storage of survey responses, and their rights as participants in the study. Randomized study participant numbers were assigned to patient-caregiver dyads to deidentify the survey response data, and to link the survey response data within each dyad. Electronic data, including participant email addresses, clinical and demographic data, and survey responses, were stored within a database in REDCap, a secure online web application.

Physical forms associated with the study, including signed informed consent forms and paper survey responses, were filed in a locked office within the College of Health Sciences at the University of Kentucky.

Instrumentation

Since the purpose of this study was to evaluate the impact of gastrostomy on quality of life in caregivers of patients with ALS and neurogenic dysphagia, the focus was on the caregiver responses. Caregivers completed the Modified Caregiver Strain Index (MCSI, see Appendix C) to assess self-reported levels of caregiver strain as an index of quality of life (Thornton & Travis, 2003). The MCSI is a 13-item tool used to assess the subjective and objective determinants of caregiver strain across physical, financial, emotional, and social/personal domains. Responses for each survey item are quantified by the frequency of occurrence: 0 points for 'no', 1 point for 'yes, sometimes', and 2 points for 'yes, regularly'. Higher levels of caregiver strain are indicated by higher scores on the MCSI, with the total score ranging from 0 to 26. The MCSI represents a stable and reliable measure of caregiver strain over time, and increased caregiver strain has been associated with reduced quality of life in caregivers of patients with ALS, stroke, and other chronic disabilities (Burke et al., 2017; Litzelman et al., 2015). Additionally, the selection of the MCSI allowed cross-study comparisons with similar investigations into the impact of gastrostomy on caregiver quality of life (ProGas Study Group, 2015).

The Eating Assessment Tool (EAT-10, see Appendix D) is a 10-item symptom-specific screening tool used to assess the presence and severity of dysphagia (Belafsky et al., 2008). Dysphagic symptoms are rated using a 5-point scale ranging from 0 (no

problem) to 4 (severe problem), and the total score is calculated as a sum of all 10 items. The EAT-10 has been documented as a valid and reliable instrument to predict aspiration risk in patients diagnosed with ALS (Plowman et al., 2016). Patients who receive a total score of greater than 3 on the EAT-10 are at risk for unsafe airway protection and are twice as likely to experience penetration or aspiration while swallowing. Levels of caregiver strain are potentially impacted by the caregiver's perceptions of the patient's dysphagic symptoms and severity, as indexed by the EAT-10. Additionally, based on clinical experience, patients and caregivers have been observed to disagree on the degree of swallowing impairment. Both patient and caregiver were instructed to complete the EAT-10 to determine whether a concordance exists between patient's ratings of their swallowing impairment and the caregiver's ratings of the swallowing impairment, to assess the relationship between caregiver strain and caregiver's perceptions of the patient's swallowing impairment, and to evaluate how these perceptions of swallowing impairment may correlate to caregiver strain following gastrostomy.

The Swallowing-Related Quality of Life instrument (SWAL-QOL, see Appendix E) was administered to evaluate the patient's perception of how their dysphagic symptoms impact their quality of life and whether swallowing-related quality of life changes following gastrostomy (McHorney, Martin-Harris, Robbins, & Rosenbek, 2006; McHorney et al., 2002). The SWAL-QOL contains 44 survey items covering 11 domains of quality of life surrounding the meal experience that are potentially impacted by dysphagia. Swallowing-related quality of life domains include burden of dysphagia, eating desire, eating duration, food selection, communication, fear related to swallowing, mental health, social concerns related to swallowing, fatigue, sleep, and dysphagia

symptom frequency. Patients rate survey items using a 5-point Likert scale, with 1 indicating high frequency (very much true, almost always, strongly agree, all of the time, always true) and 5 indicating low frequency (not at all true, never, strongly disagree, none of the time, never true). A total SWAL-QOL score is derived ranging from 0 to 100, with higher scores representing higher ratings of quality of life related to swallowing. The SWAL-QOL questionnaire ends with options to indicate the currently consumed diet texture and liquid consistency and evaluate the patient's perception of their overall health: poor, fair, good, very good, or excellent. In patients diagnosed with ALS, lower SWAL-QOL scores were significantly correlated with increased dysphagia severity and the instrument differentiates patients who are safely swallowing versus patient who are more likely to experience unsafe swallowing (penetration versus aspiration) (Paris et al., 2013; Tabor et al., 2016).

Survey Procedure

Participants completed surveys during two phases of the study: pre-gastrostomy and post-gastrostomy. To reduce the time burden associated with completing surveys, participants were offered the option to complete surveys via mail or online. Within the REDCap database, an electronic link was generated and sent to the participant's email address. When clicked, the electronic link opened a webpage that presented surveys sequentially, with responses required for all survey items before the participant was allowed to proceed to the next survey. As a matter of convenience, participants were able to complete their set of online surveys across multiple sessions, if necessary. The electronic link maintained the participant's progress through the surveys. If the participant did not have reliable internet and/or email access or the participant preferred

to complete the set of surveys on paper, physical copies of surveys were provided. Each set of surveys contained a cover page (to maintain privacy of survey responses and to clearly distinguish whether the set of surveys was for the patient or the caregiver), the study participant identification number, and a stamped envelope addressed to the UK ALS Multidisciplinary Clinic.

Pre-Gastrostomy Phase. Participants within each patient-caregiver dyad completed surveys at or around each regularly scheduled visit, beginning with the clinic visit during which informed consent was obtained. At the UK ALS Multidisciplinary Clinic, patients were routinely scheduled for clinic visits once every 3 months. Accordingly, each patient-caregiver dyad completed surveys at intervals of approximately 3 months. Participants completed surveys during or following the clinic visit. If surveys were not completed within 1-2 weeks of the clinic appointment, participants were contacted and reminded to complete the surveys. Research personnel estimated surveys required between 20-30 minutes to complete.

The number of clinic visits a patient attends prior to electing to proceed with gastrostomy varies; however, due to the rapid disease progression and negative prognostic value of malnutrition and weight loss observed in patients exhibiting bulbar symptoms, patients typically attend three clinic visits prior to proceeding with gastrostomy tube insertion. Patient-caregiver dyads completed surveys at 3-month intervals until the patient elected to proceed with gastrostomy, or, in the case of declining gastrostomy, until the patient was no longer consuming an oral diet.

Post-Gastrostomy Phase. At the UK ALS Multidisciplinary Clinic, gastrostomy was offered based on the practical guidelines disseminated by the AAN (Miller et al., 2009;

Miller et al., 1999). Gastrostomy was recommended to patients in the presence of symptomatic dysphagia as indicated by self-reported symptoms during clinical interview, weight loss greater than 10% from stated usual adult body weight, inability to meet nutritional needs, or forced vital capacity (FVC) measured at 50% of predicted value. It is recommended that patients who wish to proceed with gastrostomy do so before their FVC dips below 50% of predicted value to reduce the risk of surgical complications. Eight weeks following the procedure, patient-caregiver dyads completed a final set of surveys. The choice to reassess caregiver strain at 8 weeks post-gastrostomy was a pragmatic decision, so as to allow for any potential influence of gastrostomy on levels of caregiver strain.

Outcome Measures

The primary outcome measure of the study was self-reported levels of caregiver strain, as indexed by the MCSI. Caregivers completed the MCSI on at least one timepoint prior to gastrostomy and at a single timepoint following gastrostomy.

Secondary outcome measures included caregiver perceptions of swallowing impairment and patient perceptions of swallowing impairment, as indexed by the EAT-10. Swallowing-related quality of life was evaluated by the patient using the SWAL-QOL. Perceptions of swallowing impairment and swallowing-related quality of life were collected on at least one timepoint prior to gastrostomy and at a single timepoint following gastrostomy. Other secondary outcome measures included patient clinical data collected at each clinic visit to assess disease progression and functional changes in impairment. Clinical data included weight (kilograms), body mass index, and forced vital capacity. The ALSFRS-R was used to track functional changes in changes in bulbar-specific symptoms and overall disease progression (Cedarbaum et al., 1999). Twelve

questions encompassing common tasks of daily living were rated by the neurologist on a 5-point scale, with a score of 0 indicating no function (inability) and a score of 4 indicating full function (normal ability). The selected tasks involved bulbar function (speech, salivation, swallowing), limb function (handwriting, cutting food, dressing and hygiene, turning in bed, walking, climbing stairs), and respiratory function (dyspnea, orthopnea, respiratory insufficiency). Scores for each of the 12 tasks are summed to produce a total score, ranging from 0 to 48. A lower total score on the ALSFRS-R suggests a greater degree of functional impairment. Total scores on the ALSFRS-R demonstrate strong correlations with objective measures of muscular and pulmonary functioning, and the progression of scores on the ALSFRS-R are significantly related to prognosis (Ruoppolo et al., 2013).

Chapter Summary

Chapter 3 served to outline the methodology employed to assess the relationship between caregiver strain and dysphagia severity both before and after gastrostomy. Chapter 4 presents the results of the study.

Chapter Four: Results

Participants

A total of 26 participants, or 13 patient-caregiver dyads, were recruited to participate in the study from a convenience sample of patients who were diagnosed with ALS and receiving healthcare services through the UK ALS Multidisciplinary Clinic. To date, eight dyads were withdrawn from the study, as indicated in Figure 4.1. Five dyads completed, or are currently completing, surveys during the pre-gastrostomy phase of the study. Of these five dyads, one dyad completed surveys during both the pre-gastrostomy and post-gastrostomy phases of the study, and the results of this patient-caregiver dyad are presented as a single case study below.

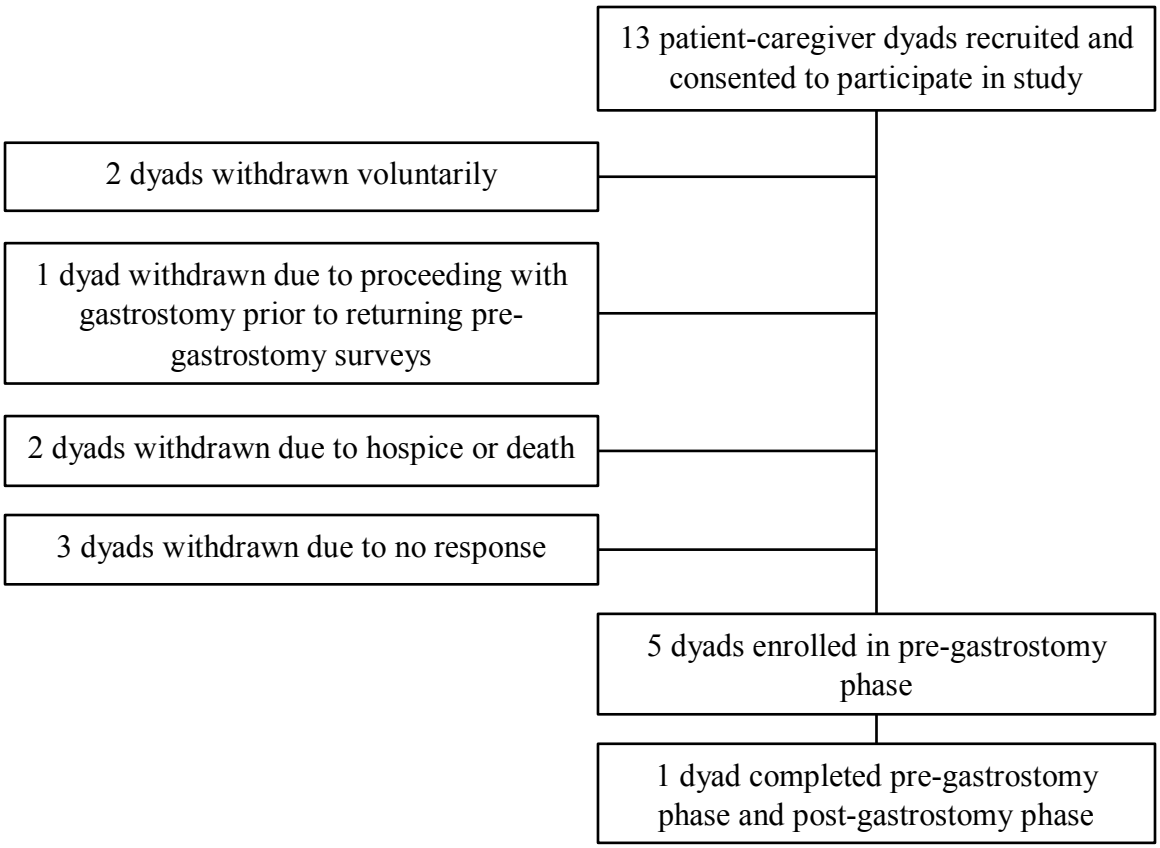


Figure 4.1: Profile of Participant Recruitment and Enrollment

Dyad Characteristics

The patient is a white male, age 75 years, and the informal caregiver is his female spouse. The patient is a retired healthcare professional and previously worked in mental health services. He reportedly was diagnosed in October 2018 after experiencing symptoms for 1 year, and his first clinic visit followed shortly thereafter. Participants consented to participate during their first clinic visit and completed pre-gastrostomy surveys via mail immediately following the clinic visit. The patient completed all pre-gastrostomy surveys: the EAT-10 and SWAL-QOL. However, the caregiver completed only the MCSI both pre- and post-gastrostomy. She did not complete the EAT-10 during the pre-gastrostomy phase. Research personnel attempted to follow-up multiple times; however, she was unable to complete and return the survey before the patient proceeded with gastrostomy. Thus, comparisons between patient and caregiver perceptions of the patient's swallowing impairments were not possible. The patient underwent gastrostomy tube insertion in December 2018. The patient and caregiver completed all surveys during the post-gastrostomy phase in January 2019.

Outcome Measures

Primary outcome measure. The primary purpose of this study was to assess the self-reported levels of caregiver strain at two time points: (1) before gastrostomy, when the patient was experiencing dysphagia and consuming an oral diet, and (2) after the patient proceeded with gastrostomy tube insertion. The caregiver completed the MCSI during both phases of the study, and the findings are reported in Table 4.1. The MCSI contains 13 items, with a total score ranging from 0 (no caregiver strain) to 26 (highest caregiver strain). The caregiver self-reported a caregiver strain score of 2 during the pre-

gastrostomy phase, and caregiver strain increased to 9 during the post-gastrostomy phase. Though the MCSI contains a total of 13 items, the caregiver's responses to 7 items were listed in Table 4.1 to elucidate any changes in her self-reported levels of caregiver strain between the pre-gastrostomy phase and the post-gastrostomy phase. The caregiver supplemented some of her survey responses on the post-gastrostomy survey with qualitative information. For the survey item 'There have been emotional adjustments', the caregiver wrote, "No arguments. My emotions are like a roller coaster." After the caregiver answered "yes, sometimes" to the survey item 'It is upsetting to find the person I care for has changed so much from his/her former self', she wrote, "Very different physically, he continues to be very awesome."

Table 4.1 Primary Outcome Measure: Caregiver Strain

	Pre-Gastrostomy	Post-Gastrostomy
MCSI, Total	2	9
My sleep is disturbed.	No	Yes, Sometimes
There have been family adjustments.	No	Yes, Sometimes
There have been changes in personal plans.	No	Yes, Sometimes
There have been other demands on my time.	No	Yes, Sometimes
There have been emotional adjustments.	No	Yes, On a regular basis "No arguments. My emotions are like a roller coaster."
It is upsetting to find the person I care for has changed so much from his/her former self.	No	Yes, Sometimes "Very different physically, he continues to be very awesome."
I feel completely overwhelmed.	Yes, On a regular basis	Yes, On a regular basis

Secondary outcome measures. The study assessed multiple secondary outcome measures to further elucidate the relationship between caregiver strain, dysphagia, patient swallowing-related quality of life, gastrostomy, and disease progression. Secondary outcome measures were assessed prior to gastrostomy and following gastrostomy and included patient's perception of his dysphagia severity (EAT-10), patient's ratings of his swallowing-related quality of life (SWAL-QOL), functional impairments due to disease progression (ALSFRS-R, Total), bulbar function due to disease progression (ALSFRS-R, Bulbar), weight (kilograms), body mass index, and forced vital capacity. Table 4.2 displays the patient's perception of his swallowing impairment (EAT-10). The patient's EAT-10 score was rated 34 (out of a total of 40) when he was consuming an oral diet and not relying on alternative means of nutrition (pre-gastrostomy phase). Following gastrostomy tube insertion, his EAT-10 scores increased to 36, suggesting he perceived his swallowing impairment to have increased following gastrostomy.

Table 4.2 Secondary Outcome Measure: Patient Dysphagia Severity

	Pre-Gastrostomy	Post-Gastrostomy
EAT-10, Total	34	36

The patient completed the SWAL-QOL before and after gastrostomy and Table 4.3 displays the results. The patient's total score on the SWAL-QOL decreased from 53.5 to 48.5 following gastrostomy, consistent with a reduction in the patient's overall swallowing-related quality of life. Decreased ratings of quality of life were observed in the domains of the burden of dysphagia, eating desire, eating duration, food selection, communication, fear, and dysphagia symptoms. The largest decline in swallowing-related quality of life scores from pre-gastrostomy to post-gastrostomy was within the domain of eating desire, with the patient indicating he cares less about eating, he is rarely hungry, and he no longer enjoys eating. Changes in the patient's survey responses within the domain of fear included increased fear of choking when eating and drinking but improvements in his perception of knowing when he might choke.

Following gastrostomy, the patient reported improvements in the four quality of life domains: mental health related to swallowing, social concerns related to swallowing, fatigue, and sleep. The largest improvement in scores from pre-gastrostomy to post-gastrostomy was within the domain of fatigue, with the patient reporting less weakness, tiredness, and exhaustion. Of note were the changes within the domains of mental health and social functioning. Regarding mental health related to swallowing, the patient reported slight reduction in the frequency of depression, annoyance, discouragement, and impatience regarding his swallowing problem and a slight increase in his frustration with his swallowing problem. In the domain of social concerns regarding his swallowing problem, the patient reported slight reduction in work/leisure changes, and a larger reduction in role changes with family and friends.

Table 4.3 Secondary Outcome Measure: Patient Swallowing-Related Quality of Life

	Pre-Gastrostomy	**Post-Gastrostomy**
SWAL-QOL, Total	53.5	48.5
SWAL-QOL, Burden	30	20
SWAL-QOL, Eating Desire	60	26.7
SWAL-QOL, Eating Duration	30	20
SWAL-QOL, Food Selection	50	30
SWAL-QOL, Communication	40	20
SWAL-QOL, Fear	70	60
SWAL-QOL, Mental Health	60	72
SWAL-QOL, Social Concerns	48	60
SWAL-QOL, Fatigue	66.7	86.7
SWAL-QOL, Sleep	80	90
SWAL-QOL, Symptoms	55.7	42.9
SWAL-QOL, Diet Texture	Soft, easy to chew foods like casseroles, canned fruits, soft cooked vegetables, ground meat, or cream soups.	All nourishment through a tube.
SWAL-QOL, Liquid Consistency	Liquids such as water, milk, tea, fruit juice, and coffee.	No liquids by mouth or limited to ice chips.
SWAL-QOL, Overall Health	Fair	Fair

Table 4.4 presents the patient clinical data during both phases of the study: pre-gastrostomy and post-gastrostomy. The patient's total scores on the ALSFRS-R increased from 32 to 35 following gastrostomy. Bulbar function on the ALSFRS-R was rated as 5 during the pre-gastrostomy phase and decreased to 4 during the post-gastrostomy phase. The decline in bulbar function was documented as a reduction in the patient's verbal speech ability. The patient's weight and body mass index increased following gastrostomy, from 82.1 kilograms to 83.9 kilograms and 26.7 to 27.4, respectively. Forced vital capacity decreased following gastrostomy from 70% to 60%.

Table 4.4 Secondary Outcome Measures: Patient Clinical Data

	Pre-Gastrostomy	Post-Gastrostomy
ALSFRS-R, Total	32	35
ALSFRS-R, Bulbar	5	4
Weight (kilograms)	82.1kg	83.9kg
Body Mass Index	26.7	27.4
Forced Vital Capacity (%)	70%	60%

Summary of outcome measures. One primary outcome measure and eight secondary outcome measures were assessed at two time points: pre-gastrostomy and post-gastrostomy. The caregiver reported increased levels of caregiver strain following gastrostomy. The patient reported increased severity of swallowing impairment, decreased swallowing-related quality of life, improved overall functional impairment, reduced bulbar function, increased weight and body mass index, and decreased forced vital capacity following gastrostomy.

Chapter Summary

Chapter 4 presented a case study of a single patient-caregiver dyad prior to and following gastrostomy. The fifth and final chapter provides a discussion of results, limitations of the study, and implications for future research.

Chapter Five: Discussion

Review of Purpose

Informed, autonomous consent is a necessary part of the decision to proceed with gastrostomy, and it is the onus of healthcare professionals to provide a clear explanation of the benefits and burdens of gastrostomy (DeLegge et al., 2005). There is little research studying the nature of the relationship between caregiver quality of life and palliative interventions that manage dysphagia in patients diagnosed with ALS (Katzberg & Benatar, 2011). A medical intervention that increases caregiver strain may decrease the effectiveness of the care they provide to the patient and ultimately result in a cascade of negative ramifications for both the patient and caregiver. To facilitate a comprehensive assessment of treatment effectiveness, and better guide patients and their families in their decision to proceed or decline gastrostomy, this study aimed to investigate the relationship between gastrostomy and caregiver strain as an index of quality of life in caregivers of patients with ALS and dysphagia. Secondary outcome measures were explored to evaluate the associations between caregiver strain and patient-specific factors such as swallowing-related quality of life, severity of dysphagia, and disease progression.

Review of Methodology

Thirteen patient-caregiver dyads were recruited to participate in the prospective, observational, longitudinal study. Patients and their informal caregivers were recruited to participate in the study prior to their decision regarding whether to accept or decline gastrostomy. To date, one patient-caregiver dyad completed surveys during both the pre- and post-gastrostomy phase. There were two phases of the study: pre-gastrostomy phase

and post-gastrostomy phase. Both patients and caregivers completed surveys at one or more time points, roughly every 3 months, during the pre-gastrostomy phase. Once the patient proceeded with gastrostomy, both the patient and the caregiver completed a final set of surveys.

The primary focus of the study was on the caregiver responses. To assess caregiver strain, caregivers completed the MCSI. Severity of the patient's dysphagia was assessed using the EAT-10, and both caregivers and patients rated their perceptions of the patient's swallowing impairment. The SWAL-QOL was employed to assess the patient's swallowing-related quality of life. The patient's overall disease-specific impairment and bulbar dysfunction was rated by the neurologist at each clinic visit using the ALSFRS-R. Clinical data, including weight, body mass index, and forced vital capacity, was collected to assess disease progression.

Discussion of Results

The study proposed four hypotheses. The first hypothesis, that caregiver strain would decrease following gastrostomy, was not supported. The second hypothesis proposed a positive relationship between caregiver strain and degree of the patient's functional impairment. This hypothesis was partially refuted: caregiver strain increased as the patient's overall function improved or stabilized. However, a decline was observed in the patient's bulbar function, specifically in the domain of speech. The third and fourth hypotheses compared levels of caregiver strain, severity of the patient's dysphagia, and the patient's swallowing-related quality of life. These final hypotheses could not be evaluated since outcome measures for this single patient-caregiver dyad were assessed at

a single time-point during the pre-gastrotomy phase. Thus, no comparisons could be made either within participants or between participants.

Caregiver strain. The single case study design prevented generalizing results to the wider population of caregivers of patients with ALS. Despite this limitation, some comments can be made regarding the increase in caregiver strain observed within this patient-caregiver dyad following gastrostomy. The hypothesis of this study was not supported in that the caregiver reported a higher level of self-perceived caregiver strain following gastrostomy, whereby the total MCSI score increased from 2 (pre-gastrostomy) to 9 (post-gastrostomy). The MCSI does not categorize scores to differentiate between clinically significant levels of caregiver strain; accordingly, it was difficult to conclude that meaningful change has occurred between levels of caregiver strain for this individual caregiver relative to gastrostomy. The ProGas Study Group previously investigated the relationship between gastrostomy and caregiver strain as indexed by the MCSI and discovered that caregiver strain significantly increased following gastrostomy placement (mean [SD] total MCSI score 9.9 [6.4] at baseline versus 11.8 [6.5] at 3 months; p=0.01) (ProGas Study Group, 2015). The caregiver participating in the current study reported comparatively less caregiver strain before and after gastrostomy. However, statistical analyses were not performed.

Additionally, for this particular patient-caregiver dyad, the pre-gastrostomy survey was completed following their first clinic visit when the patient's diagnosis was relatively recent. This may explain the caregiver's low total MCSI score during the pre-gastrostomy phase. The patient proceeded with gastrostomy tube insertion shortly thereafter due to the severity of dysphagia, difficulty maintaining adequate nutrition and

hydration, and declining forced vital capacity. While there were no dramatic changes in the patient's disease-specific impairments as detected by the ALSFRS-R in the time period between the pre-gastrostomy surveys and the post-gastrostomy surveys, the patient's speech was noted to have declined. In the presence of multiple confounding variables, it remains unclear what factor was most influential in the increase of caregiver strain post-gastrostomy: responsibilities involved in the use and maintenance the gastrostomy tube, the slight worsening of the patient's verbal speech, the chronic presence of dysphagia, the psychological distress of coping with a terminal illness, or a myriad of factors.

Gastrostomy is recommended as a symptomatic management of malnutrition and weight loss; however, the placement of a gastrostomy tube does not ameliorate bulbar symptoms. A gastrostomy tube will not return the pleasurable and social aspects of sharing a meal with others, as evidenced by the reduction in the patient's swallowing-related quality of life post-gastrostomy. Although patients are encouraged to continue oral intake as tolerated, the patient within the present case study reportedly ceased oral intake of food and liquid due to dysphagia severity. Previous literature called attention to caregiver reports of feelings of guilt when eating in front of the patient and engaging in an activity they once shared, while patients reported reluctance to stay in the same room when others were eating (Stavroulakis et al., 2016). The patient in this study continued to experience worsening verbal communication due to dysarthria, and previous studies have demonstrated the relative importance of loss of speech and mobility restrictions over swallowing impairments (Chio et al., 2005; Hecht et al., 2002). Multiple longitudinal studies documented stabilization in patient quality of life and a significant increase in

caregiver strain and burden over time (Creemers et al., 2014; Gauthier et al., 2007; ProGas Study Group, 2015). If gastrostomy acts to alleviate or increase the degree of caregiver strain in caregivers responsible for dysphagia management, the change may not be readily discernible, and may not alleviate the potentially devastating social, cultural, and psychological impacts of progressively worsening symptoms on quality of life in patients with ALS and their caregivers.

The literature on caregiver strain, burden, and quality of life is often restricted in scientific rigor owing to cross-study inconsistencies in operational definitions, instruments, and findings (Cousins et al., 2002). A conceptual analysis of caregiver burden identified the characteristics of the caregiver, the demands of caregiving, and the involvement in caregiving as predisposing factors to caregiver burden, i.e., factors that may increase the risk of experiencing caregiver burden (Chou, 2000). Due to the heterogeneity in the presentation and progression of ALS, some authors suggest that self-reported levels of caregiver burden reflect relative constancy throughout disease progression and that any reported fluctuations in caregiver burden are consistent with characterological differences among patient-caregiver dyads (Burke et al., 2018). Without a pre-morbid assessment of personality types, interpersonal dynamics, coping styles, and the potential effects of providing care in the presence of a terminal diagnosis, such characterological differences remain confounding variables that limit what conclusions can be drawn from the association between caregiver strain and gastrostomy in the framework of a terminal illness. Regardless, continued investigation into the impact of gastrostomy on caregiver strain are needed such that these limitations do not result from a

dearth of investigation but rather the limits of what clinical research in this field can definitively establish.

Further, it is unclear whether the current study places undue emphasis on dysphagia as a disability instead of focusing on what is most important for the particular patient and caregiver in the course of the disease progression. Previous studies suggest caregiver strain does not correlate with the patient's disease-specific variables but, rather, is correlated with intrinsic factors of the caregiver, the care recipient, and their dyadic relationship (Burke et al., 2018; Simmons et al., 2000). On the MCSI survey item regarding emotional adjustments, the caregiver provided an addendum, "no arguments, my emotions are like a roller coaster." Previous literature documented significant associations between increased caregiver burden, lower quality of life, and higher levels of distress, anxiety, and depression (Burke et al., 2017). In this study, the authors speculated that the observed reduction in a caregiver's quality of life and increased levels of self-reported caregiver burden were primarily influenced by the subjective experience of the caregiver, including the caregiver's resiliency, coping-styles, pre-morbid personality characteristics, and the extent of and reliance on support networks. In a study on caregivers of neurological patients with dysphagia, authors found correlations between the anxiety level of caregivers and the presence of dysphagia, but caregiver anxiety did not correlate with any patient-related factors, including the severity of dysphagia, the duration of dysphagia, the condition of a patient's dependency on the caregiver for eating and drinking, and the reliance on gastrostomy (Serel Arslan et al., 2017). Medical interventions aimed at managing dysphagia, such as gastrostomy, may not have a predictable impact on caregiver strain. Other psychosocial factors within a given patient-

caregiver dyad may be stronger predictors of caregiver strain, burden, and quality of life in caregivers of patients diagnosed with ALS.

Some studies found that caregiver quality of life may be modified or mediated by the degree of perceived social support more than physical limitations of the disease (Chou, 2000; Simmons et al., 2000). In the current study, no insight was obtained regarding the type or intensity of caregiving duties, the amount of time devoted to caregiving per day, the total duration spent in the role of an informal caregiver, or the ancillary employment of formal caregiving assistance. Based on clinical anecdotes, patients with increased dependency on a caregiver for eating and drinking are more demanding than patients who are able to self-feed or prepare their own meals. Although Serel Arslan et al. (2017) did not find a significant association between caregiver anxiety and condition of patient dependency on caregiver for eating/drinking, the relationship warrants further investigation. All of these are relevant factors in the assessment of caregiver strain, and the MCSI may not have been sensitive to these variables. Currently, there is no ALS-specific scale of caregiver strain. To evaluate outcome data for a specific medical intervention such as gastrostomy, there is a need for more sophisticated measure of caregiver strain in this vulnerable population.

Multidisciplinary Care. Multidisciplinary care for patients diagnosed with ALS is associated with positive treatment benefits, including increased duration of survival, reduced hospitalizations and mortality, and improved quality of life (Chio et al., 2006; Rosenfeld & Strong, 2015). As such, the sample of patients recruited to participate in the current study within the UK Multidisciplinary ALS Clinic may not be representative of other patients diagnosed with ALS and receiving their care elsewhere. Any conclusions

regarding the impact of gastrostomy on caregiver strain may not be generalizable to patients who are not enrolled in multidisciplinary care clinics.

Withdrawal rates. Of the 13 patient-caregiver dyads recruited to participate in the study, one patient-caregiver dyad completed surveys during both phases of the study: pre-gastrostomy and post-gastrostomy (see Figure 4.1). Four patient-caregiver dyads are currently enrolled in the pre-gastrostomy phase of the study, having completed surveys on at least one occasion. Eight patient-caregiver dyads were withdrawn from the study due to no response, voluntary withdrawal, entering hospice, or death. Data collection for this study is ongoing at the UK Multidisciplinary ALS Clinic, and three sites located within the United States plan to join this investigation. The addition of external sites aims to increase participant recruitment to a total of 60 patient-caregiver dyads to improve statistical power and sample representativeness.

Limitations

Due to ethical considerations, randomized and controlled clinical trials may not be feasible in pursuing this line of prospective investigation. Although careful steps were taken to improve the scientific rigor of this observational study, there were numerous limitations characteristic of survey research. Self-selection bias, the process of a patient agreeing to participate in a study, introduces bias that may affect the survey responses (Heffernan et al., 2004). Clinical anecdotes suggested that patients may have underreported the severity of their swallowing problems, while caregivers may have underreported the burden of caregiving. In addition, there were challenges in scheduling the surveys. Research personnel attempted to obtain completed surveys as close to 3-month intervals as possible but rescheduling of clinic appointments introduced variation

in the projected 3-month interval between visits to the UK ALS Multidisciplinary Clinic. Furthermore, occasional delays in survey completion occurred due to dyads misplacing paper copies of surveys, undelivered email surveys, and multiple reminders to complete survey responses.

Missing data were observed due to participants omitting responses within surveys, research personnel's inability to obtain all clinical data at every clinic visit, and concerns regarding the accuracy of forced vital capacity ratings in patients with severe bulbar symptoms. Conclusions regarding the relationship between levels of caregiver strain and the caregiver's perception of the patient's swallowing impairment were not possible as the caregiver did not complete all surveys during the pre-gastrostomy phase. Regarding the eight patient-caregiver dyads that were withdrawn from the study, this loss to follow-up may have reduced the representativeness of the sample and lead to biased estimates (Thompson & Levy, 2004). The research personnel offered electronic and mail-based surveys to increase the response rate. However, the response rate within the sample of participants was low and this potentially hindered the overall longitudinal analysis (Messina & Beghi, 2012).

Ample clinical discretion was exercised during the recruitment period. Though this was based on ethical considerations, this may have introduced a gate-keeping bias into the study (Aoun et al., 2012). Patients and their caregivers were excluded from participating in the study if they exhibited overt signs of cognitive impairment or if the symptomatology placed the patient in the end stages of ALS. Such exclusions of the more vulnerable patient-caregiver dyads may have represented an inadvertent selection bias. Although there was no formal screening or assessment of cognitive impairment,

participants were excluded from participating in the study if cognitive impairment was observed, whether diagnosed or clinically overt. Previous literature investigating quality of life in patients diagnosed with ALS and their caregivers is varied on the procedures for cognitive screening, with exclusion criteria ranging between clinical signs or symptoms, diagnoses of cognitive dysfunction, and brief cognitive screenings including the Montreal Cognitive Assessment (Creemers et al., 2014; Gauthier et al., 2007; Kurien et al., 2017).

During the process of obtaining informed consent from participants, research personnel reviewed the identical consent forms with each patient-caregiver dyad; however, there was no standardization of this conversation. Research personnel attempted to introduce and explain the purpose of the study in a consistent manner, but it was not possible to have the same conversation with each dyad. Despite instructions during the recruitment process, and reminders during the consenting process that their participation in the study and/or responses on the surveys would not impact the level of care received at the clinic, participant responses may have been impacted. Research personnel also encouraged participants to complete the surveys independently and to avoid influencing any survey responses. Outside of clinic, however, patients and caregivers potentially completed surveys in the presence of their dyadic partner.

Implications for Future Research

While the focus of this study was on the caregiver responses, this study represents a section of a larger, more comprehensive investigation into the relationship between gastrostomy and quality of life of both patients and their caregivers. Longitudinal data is currently being collected on swallowing-related quality of life in patients prior to and following gastrostomy tube insertion and in patients who decline gastrostomy. Literature

differs as to whether patients with ALS who decline gastrostomy may serve as a comparator group to those patients who elect to receive gastrostomy, or whether causal inference models may be more suitable to avoid confounding by indication (McDonnell et al., 2017). As of January 2019, an IRB modification was completed to add a cross-sectional component to the study to further elucidate the impact of disease-specific variables (bulbar symptoms versus limb symptoms). Patients without bulbar symptoms at their initial diagnosis were recruited to participate in the study to evaluate the impact of limb-onset symptoms on patient and caregiver quality of life and to investigate whether differences in self-reported caregiver strain exist between caregivers of patients with dysphagia and caregivers of patients without dysphagia. To further elucidate the impact of the dysphagia, gastrostomy, and disease-specific variables on emotional wellbeing of patients and caregivers, the Beck Anxiety Inventory and the Beck Depression Inventory were added to the battery of surveys (Bardhoshi, Duncan, & Erford, 2016; Beck, Steer, Ball, & Ranieri, 1996).

Previous literature has accomplished qualitative explorations into the experiences of patients and caregivers regarding gastrostomy and caregiver strain (Stavroulakis et al., 2016). During quantitative data collection of the current study, patients and their caregivers often volunteered unsolicited qualitative data along with their survey responses. These comments, next to their response on survey items, provided additional insight. In the future, this study will introduce a qualitative component to the current research design to encourage dialogue on the topic, obtain valuable opinions from patients and caregivers, and attempt to unpack the subjective phenomena behind changing levels of caregiver strain as it relates to gastrostomy.

Conclusions

It is imperative to evaluate the strain placed on caregivers of patients diagnosed with ALS throughout disease progression. ALS is a progressive, terminal illness and the inevitable worsening of a patient's impairments is likely to increase the physical and emotional demands placed on the caregiver. Caregivers who develop clinically significant levels of caregiver strain risk may become compromised in their physical and/or emotional health, which can, in turn, impact the care they provide to the patient. Medical recommendations and care provided within multidisciplinary clinics should consider interventions that address the well-being of caregivers in addition to patients (Boerner & Mock, 2012; Chio et al., 2005; Creemers et al., 2016).

The decision to accept or decline gastrostomy must be guided by the preferences of the individual and their family, goals of treatment, and outcome data (DeLegge et al., 2005). Healthcare professionals should provide clear, objective information of the anticipated benefits and burdens of gastrostomy (Squires, 2006). The ongoing investigation into the relationship between gastrostomy and quality of life in caregivers of patients diagnosed with ALS and dysphagia ultimately aims to contribute to the understanding of the treatment efficacy of gastrostomy to better guide patients and their families in their decision.

Chapter Summary

Chapter five discussed the results of this study, limitations, and implications for future clinical research.

Appendix A: Study Manual

Patients with ALS and Bulbar Symptoms

1. Identify qualified patient and caregiver dyads:
   - Patient must meet all of the following criteria to qualify for the study:
     - Patient has a confirmed diagnosis of ALS.
     - Patient exhibits bulbar symptoms, as indexed by a score of < 4 on at least one of the bulbar subscales of the ALS Functional Rating Scale (ALS-FRS-R). These subscales are listed at the end of the manual. The ALS-FRS-R is also located at http://www.outcomes-umassmed.org/als/alsscale.aspx
     - Patient does not have a PEG tube already in place.
     - Patient has an adult family member or friend who is considered the "primary caregiver" and they provide unpaid care and assistance.
   - New or current patients may be identified for qualification prior to or during the clinic appointment.
     - The research nurse or other study personnel may call the patient and/or caregiver at least 1 week prior to the clinic appointment to notify them of this study.
     - Interested patient/caregiver dyads may be asked to arrive 30 minutes prior to clinic appointment to meet with study personnel to further discuss the study and answer any questions.
     - Patient/caregiver dyads will be invited to participate in this study and asked to give consent to participate.
     - Patient/caregiver dyads will be consented in the clinic rooms during their appointments or in a separate consult room within the clinic.

2. Consent patient/caregiver dyads.
   - Approved research personnel will consent patients and caregivers by reviewing the entire IRB consent form and having the participants sign and date the last page.
     - Participants will be provided copies of the signed IRB consent forms.
   - Research personnel should highlight the following:
     - Please complete the surveys independently. If the patient requires physical assistance with completing the survey, the caregiver may assist them. Caregivers should be encouraged to allow patients to answer questions without caregiver input.
     - It should take approximately 20-30 minutes to complete the set of surveys.
     - Patient and caregiver will complete surveys around the time of each follow-up clinic appointment (approximately every 3 months).

- Within the "Caregiver" set of surveys, the first survey (EAT-10) will ask a series of questions regarding swallowing. The caregiver needs to fill out the survey from the *patient's* perspective. (Include a written reminder on the paper copy of the caregiver's EAT-10.)

– After consent forms have been completed, research personnel should politely request and document the caregiver's age (years) and relationship to the patient. This information is stored in the caregiver demographics section in REDCap (see Step 3).

3. Instruct & help participants complete the surveys.
   – Prior to survey administration:
      - Determine **how** the participants prefer to complete the surveys.
         - If the participants have reliable internet and email access, they may complete the surveys online.
            - Using The AWARE Study project in REDCap, an electronic link is generated and sent to the participant's email address. When clicked, this link takes the participant to a webpage where they are able to complete the set of surveys.
            - It is preferred that the patient and caregiver have separate email accounts; however, it is possible to send both the "Patient" set of surveys and the "Caregiver" set of surveys to the same email address. Care must be taken to ensure that the caregiver completes the "Caregiver" set of surveys, and the patient completes the "Patient" set of surveys. Each set of surveys must be clearly labeled when sending the electronic link in REDCap.
            - It is possible for a participant to complete the online surveys in multiple sittings. If the participant exits the webpage without completing the set of surveys, they may click the electronic link within the email and a webpage will open that allows them to resume their survey progress.
         - If the participants do not have reliable internet and email access or the participants prefer to complete the surveys on paper, hard copies of surveys may be provided.
            - Include a cover page on each set of surveys to maintain privacy of survey responses. On the cover page, clearly label the set as either "Patient Surveys" or "Caregiver Surveys".
            - Record the Participant ID on each set of surveys for identification purposes.

        - If participants are completing the surveys outside of clinic, provide an appropriately sized envelope, stamps, and clinic mailing address.
    - Determine **when** the participants prefer to complete the surveys.
        - Participants may complete the surveys during the clinic appointment (via personal electronic device, paper copies, or clinic iPad).
        - Participants may complete the surveys outside of the clinic appointment (via personal electronic device or paper copies).
- <u>During survey administration</u>:
    - Research personnel will send a link via REDCap *or* provide paper copies of the surveys.
        - Patients will complete the "Patient" set of surveys:
            - Eating Assessment Tool (EAT-10)
            - Swallowing Quality of Life Questionnaire (SWAL-QOL)
            - Beck Anxiety Inventory (BAI)
            - Beck Depression Inventory (BDI)
        - Caregivers will complete the "Caregiver" set of surveys:
            - Eating Assessment Tool (EAT-10)
            - Modified Caregiver Strain Index MCSI)
            - Beck Anxiety Index (BAI)
            - Beck Depression Inventory (BDI)
    - Participants may complete the surveys during the clinic appointment or outside of clinic.
        - If participants are completing the surveys outside of clinic, research personnel will encourage the participants to complete the surveys independently and within 1 week of their clinic appointment.
            - If the patient requires physical assistance with completing the survey, the caregiver may assist them. Caregiver should be encouraged to allow the patient to answer questions without caregiver input.
            - Research personnel will call with a reminder to complete and submit the surveys after 1-2 weeks.
        - If participants are completing the surveys during clinic, research personnel should encourage the participants to complete the surveys independently.
            - Patients who require physical assistance to complete the surveys will receive assistance from research personnel as needed.
            - Caregivers may be offered a separate room in which to complete their set of surveys. Research personnel will

inform the caregiver of when a multidisciplinary team member is ready to see both of them for health services. The caregiver may then pause their survey progress to rejoin their family member for the appointment. The caregiver may return to complete the surveys at a later time.
    - Research personnel will be available to answer any questions or concerns.

- Following survey administration:
    - Research personnel will collect paper copies of the completed surveys from the participant and transcribe the survey responses into REDCap.
        - Paper copies of completed surveys will be stored in CTW 124K.
    - Research personnel will input demographic data for each patient and caregiver into the 'Demographics' portion of the study in REDCap.
        - Patient Demographics:
            - Gender
            - Age
            - Weight (kg)
            - Body Mass Index (BMI)
            - Total ALS Functional Rating Scale (ALS-FRS-R)
            - Bulbar Subsection on ALS-FRS-R
            - Forced Vital Capacity (FVC)
            - Current assistive equipment, e.g., Cough Assist, Non-Invasive Ventilation, CPAP/BiPAP, AAC device(s), none.
        - Caregiver Demographics
            - Gender
            - Age
            - Caregiver relationship, e.g., spouse/partner, mother, father, sibling, child, friend, other.

4. During subsequent clinic visits, patient/caregiver dyads will be asked to arrive 30 minutes early in order to complete the surveys during the clinic appointment *or* the patient/caregiver dyads will be asked to complete the surveys outside of the clinic. The same process outlined above should be followed.

5. This survey process will be repeated at each clinic visit leading up to PEG placement (if patient elects to receive a PEG) and then once at 8 weeks post-PEG placement.

6. Any questions should be directed to Debra Suiter, PhD, CCC-SLP, (859) 218-5323 or debra.suiter@uky.edu.

ALS Functional Rating Scale – Bulbar Subscales

Item 1: Speech

4: Normal speech process

3: Detectable speech disturbance

2: Intelligible with repeating

1: Speech combined with non-vocal communication

0: Loss of useful speech

Item 2: Salivation

4: Normal

3: Slight but definite excess of saliva in mouth, may have nighttime drooling

2: Moderately excessive saliva; may have minimal drooling (during the day)

1: Marked excess of saliva with some drooling

0: Marked drooling; requires constant tissue or handkerchief

Item 3: Swallowing

4: Normal eating habits

3: Early eating problems – occasional choking

2: Dietary consistency changes

1: Needs supplement tube feeding

0: NPO (exclusively parenteral or enteral feeding)

The ALS-FRS-R is also located at http://www.outcomes-umassmed.org/als/alsscale.aspx

Appendix B: Consent Forms

Consent to Participate in a Research Study
The AWARE Study: Impact of Gastrostomy on Swallowing-Related Quality of Life in Patients with Amyotrophic Lateral Sclerosis and their Caregivers
Caregiver Consent

WHY ARE YOU BEING INVITED TO TAKE PART IN THIS RESEARCH?

You are being invited to take part in a research study about how feeding tube placement affects quality of life (QOL) for patients and their caregivers. You are being invited to take part in this research study because you are a caregiver of an individual with ALS who is receiving medical care through our multidisciplinary clinic. If you volunteer to take part in this study, you will be one of about 25 caregivers to do so at the University of Kentucky.

WHO IS DOING THE STUDY?

The person in charge of this study is Debra Suiter, PhD of University of Kentucky, Department of Rehabilitation Sciences. There may be other people on the research team assisting at different times during the study.

WHAT IS THE PURPOSE OF THIS STUDY?

This study will examine the impact of feeding tube (PEG) placement on quality of life (QOL) of patients with amyotrophic lateral sclerosis (ALS) and their caregivers. Many patients with ALS experience difficulty swallowing, and healthcare providers often recommend a PEG tube to meet their nutrition and hydration needs. Receiving nutrition and hydration via a PEG is thought to improve nutritional balance, stabilize weight, and possibly extend the patient's life.

This study will examine patient and caregiver perceptions about QOL prior to and following PEG placement. We hope to learn some of the concerns of patients with ALS and their caregivers to enhance communication in the future regarding PEG tube use and nutritional intervention.

ARE THERE REASONS WHY YOU SHOULD NOT TAKE PART IN THIS STUDY?

There are no known reasons that you should not take part in this study.

WHERE IS THE STUDY GOING TO TAKE PLACE AND HOW LONG WILL IT LAST?

The research procedures will be conducted at the Kentucky Neuroscience Institute. You will need to come to regularly scheduled clinic visit with the multidisciplinary ALS clinic. We will ask you to fill out questionnaires during today's clinic visit. You will be mailed study questionnaires 2 weeks prior to the next clinic visit. The total amount of time necessary to complete the questionnaires is approximately 30 minutes. We will collect those at each clinic visit prior to PEG placement and at one visit approximately 8 weeks following PEG placement. The number of clinic visits prior to a patient deciding to choose PEG placement varies from patient to patient, but on average, patients are seen for 3 clinic visits prior to PEG placement. Thus, your total time for participation in this study is anticipated to be approximately 2 hours.

WHAT WILL YOU BE ASKED TO DO?

You will be asked to answer questions about how you perceive the swallowing ability of the person for whom you are caring. We will also ask you questions regarding your feelings related to caring for an individual with ALS.

If you consent to participate in this study, we will give you these questionnaires at today's clinic visit. After this visit, we will mail the questionnaires to you approximately 2 weeks prior to each of your clinic visits and ask you to bring them with you when you return to clinic.

WHAT ARE THE POSSIBLE RISKS AND DISCOMFORTS?

There is a risk that your confidentiality may be breached. We will make every effort will be made to ensure the confidentiality of the responses you provide to our questionnaires. This includes assigning you a participant number that can be linked to your name on a participant list that will be kept in a locked filing cabinet in the Principal Investigator's office (Room 124D Charles T. Wethington Building). We will also store all questionnaires in a locked filing cabinet in the Principal Investigator's office. Data collected from the questionnaires will be entered into a computer file in a secured data base that only study personnel (the Principal Investigator and others involved in this research) will be able to access. No identifying information will be entered into this database.

WILL YOU BENEFIT FROM TAKING PART IN THIS STUDY?

You will not get any personal benefit from taking part in this study.

DO YOU HAVE TO TAKE PART IN THE STUDY?

If you decide to take part in the study, it should be because you really want to volunteer. You will not lose any benefits or rights you would normally have if you choose not to volunteer. You can stop at any time during the study and still keep the benefits and rights you had before volunteering.

IF YOU DON'T WANT TO TAKE PART IN THE STUDY, ARE THERE OTHER CHOICES?

If you do not want to be in the study, there are no other choices except not to take part in the study.

WHAT WILL IT COST YOU TO PARTICIPATE?

There will be no cost to you should you choose to participate in this study.

WHO WILL SEE THE INFORMATION THAT YOU GIVE?

We will make every effort to keep confidential all research records that identify you to the extent allowed by law. The answers you provide to the questionnaires will be stored in a secured database that is password protected and accessible only to study personnel including a research assistant and the Principal Investigator.

Your information will be combined with information from other people taking part in the study. When we write about the study to share it with other researchers, we will write about the combined information we have gathered. You will not be personally identified in these written materials. We may publish the results of this study; however, we will keep you name and other identifying information private.

We will make every effort to prevent anyone who is not on the research team from knowing that you gave us information, or what that information is. All paper copies of data will be stored in a locked filing cabinet in the Principal Investigator's Office (Charles T. Wethington Building, Room 124D). Only the Principal Investigator will have information that will link your responses to your name. For all other data, you will be identified by a randomly assigned participant number. Data collected from all participants will be entered into a database called RedCap for analysis. Again, no identifying information that could link you to participation in this study will be included in this database.

You should know, however, that there are some circumstances in which we may have to show your information to other people. For example, the law may require us to show your information to a court or to tell authorities if you pose a danger to yourself or someone else. Officials from the University of Kentucky may look at or copy pertinent portions of records that may identify you.

CAN YOUR TAKING PART IN THE STUDY END EARLY?

If you decide to take part in the study you still have the right to decide at any time that you no longer want to continue. You will not be treated differently if you decide to stop taking part in the study.

If you choose to withdraw from the study early, the data collected until that point will remain in the study database and may not be removed.

ARE YOU PARTICIPATING OR CAN YOU PARTICIPATE IN ANOTHER RESEARCH STUDY AT THE SAME TIME AS PARTICIPATING IN THIS ONE?

You may take part in this study if you are currently involved in another research study. It is important to let the investigator/your doctor know if you are in another research study. You should also discuss with the investigator before you agree to participate in another research study while you are enrolled in this study.

WILL YOU RECEIVE ANY REWARDS FOR TAKING PART IN THIS STUDY?

You will not receive any rewards or payment for taking part in the study.

WHAT IF YOU HAVE QUESTIONS, SUGGESTIONS, CONCERNS, OR COMPLAINTS?

Before you decide whether to accept this invitation to take part in the study, please ask any questions that might come to mind now. Later, if you have questions, suggestions, concerns, or complaints about the study, you can contact the investigator, Debra Suiter at 859-218-5323. If you have any questions about your rights as a volunteer in this research, contact the staff in the Office of Research Integrity between the business hours of 8am and 5pm EST, Mon-Fri at the University of Kentucky at 859-257-9428 or toll free at 1-866-400-9428. We will give you a signed copy of this consent form to take with you.

WHAT IF NEW INFORMATION IS LEARNED DURING THE STUDY THAT MIGHT AFFECT YOUR DECISION TO PARTICIPATE?

If the researcher learns of new information in regards to this study, and it might change your willingness to stay in this study, the information will be provided to you. You may be asked to sign a new informed consent form if the information is provided to you after you have joined the study.

WHAT ELSE DO YOU NEED TO KNOW?

There is a possibility that the data collected from you may be shared with other investigators in the future. If that is the case the data will not contain information that can identify you unless you give your consent/authorization or the UK Institutional Review Board (IRB) approves the research. The IRB is a committee that reviews ethical issues, according to federal, state and local regulations on research with human subjects, to make sure the study complies with these before approval of a research study is issued.

_____ _____
Signature of person agreeing to take part in the study Date

_____
Printed name of person agreeing to take part in the study

_____ _____
Name of [authorized] person obtaining informed consent Date

_____
Signature of Principal Investigator or Sub/Co-Investigator

Combined Consent and Authorization to Participate in a Research Study

The AWARE Study: Impact of Gastrostomy on Swallowing-Related Quality of Life in Patients with Amyotrophic Lateral Sclerosis and their Caregivers

Patient Consent

WHY ARE YOU BEING INVITED TO TAKE PART IN THIS RESEARCH?

You are being invited to take part in a research study about how feeding tube placement affects quality of life (QOL) for patients with ALS and their caregivers. You are being invited to take part in this research study because you have been diagnosed with ALS and are being cared for at our multidisciplinary clinic. If you volunteer to take part in this study, you will be one of about 25 people with ALS at the University of Kentucky.

WHO IS DOING THE STUDY?

The person in charge of this study is Debra Suiter, PhD of University of Kentucky, Department of Rehabilitation Sciences. There may be other people on the research team assisting at different times during the study.

WHAT IS THE PURPOSE OF THIS STUDY?

This study will examine the impact of feeding tube (PEG) placement on quality of life (QOL) of patients with amyotrophic lateral sclerosis (ALS) and their caregivers. Many patients with ALS experience difficulty swallowing, and healthcare providers often recommend a PEG tube to meet their nutrition and hydration needs. Receiving nutrition and hydration via a PEG tube is thought to improve nutritional balance, stabilize weight, and possibly extend the patient's life.

This study will examine patient and caregiver perceptions about QOL prior to and following PEG placement. We hope to learn some of the concerns of patients with ALS and their families to enhance communication in the future regarding PEG tube use and nutritional intervention.

ARE THERE REASONS WHY YOU SHOULD NOT TAKE PART IN THIS STUDY?

There are no known reasons that you should not take part in this study.

WHERE IS THE STUDY GOING TO TAKE PLACE AND HOW LONG WILL IT LAST?

The research procedures will be conducted at the Kentucky Neuroscience Institute. You will need to come to regularly scheduled clinic visit with the multidisciplinary ALS clinic. We will ask you to fill out questionnaires during today's clinic visit. You will be mailed study questionnaires 2 weeks prior to the next clinic visit. The total amount of time necessary to complete the questionnaires is approximately 30 minutes. We will collect those at each clinic visit prior to PEG placement and at one visit approximately 8 weeks following PEG placement. The number of clinic visits prior to a patient deciding to choose PEG placement varies from patient to patient, but on average, patients are seen for 3 clinic visits prior to PEG placement. Thus, your total time for participation in this study is anticipated to be approximately 2 hours.

WHAT WILL YOU BE ASKED TO DO?

You will be asked to answer questions about how your swallowing ability affects your daily activities and how you feel about your swallowing ability. The person assisting in your care will be asked questions regarding how they feel about your swallowing ability. The questionnaires used for this study are designed to find out how swallowing difficulty affects quality of life.

We will give you these questionnaires at today's clinic visit if you decide to participate in this study. After this visit, we will mail the questionnaires to your home approximately 2 weeks prior to each of your clinic visits and ask you to bring them with you when you come to clinic. We will mail you another set of questionnaires approximately 8 weeks following tube placement.

WHAT ARE THE POSSIBLE RISKS AND DISCOMFORTS?

There is a risk that your confidentiality may be breached. We will make every effort to ensure the confidentiality of the responses you provide to our questionnaires. This includes assigning you a participant number that can be linked to your name on a participant list that will be kept in a locked filing cabinet in the Principal Investigator's office (Room 124D Charles T. Wethington Building). We will also store all questionnaires in a locked filing cabinet in the Principal Investigator's office. Data collected from the questionnaires will be entered into a computer file in a secured data base that only study personnel (the Principal Investigator and others involved in this research) will be able to access. No identifying information will be entered into this database.

WILL YOU BENEFIT FROM TAKING PART IN THIS STUDY?

You will not get any personal benefit from taking part in this study. Your willingness to take part, however, may, in the future, help doctors better understand and/or treat others who have ALS.

DO YOU HAVE TO TAKE PART IN THE STUDY?

If you decide to take part in the study, it should be because you really want to volunteer. You will not lose any benefits or rights you would normally have if you choose not to volunteer. You can stop at any time during the study and still keep the benefits and rights you had before volunteering. If you decide not to take part in this study, your decision will have no effect on the quality of medical care you receive.

IF YOU DON'T WANT TO TAKE PART IN THE STUDY, ARE THERE OTHER CHOICES?

If you do not want to be in the study, there are no other choices except not to take part in the study.

WHAT WILL IT COST YOU TO PARTICIPATE?

You and/or your insurance company, Medicare or Medicaid will be responsible for the costs of all care and treatment you receive during this study that you would normally receive for your condition. These are costs that are considered medically reasonable and necessary and will be part of the care you receive if you do not take part in this study. There will be no additional costs to you for your participation in this study

WHO WILL SEE THE INFORMATION THAT YOU GIVE?
We will make every effort to keep confidential all research records that identify you to the extent allowed by law. The answers you provide to the questionnaires will be stored in a secured database that is password protected and accessible only to study personnel including a research assistant and the Principal Investigator.

Your information will be combined with information from other people taking part in the study. When we write about the study to share it with other researchers, we will write about the combined information we have gathered. You will not be personally identified in these written materials. We may publish the results of this study; however, we will keep your name and other identifying information private.

We will make every effort to prevent anyone who is not on the research team from knowing that you gave us information, or what that information is. All paper copies of data will be stored in a locked filing cabinet in the Principal Investigator's office (CTW 124D). Only the Principal Investigator will have information that will link your identity to your information. For all other data, you will be identified by a randomly assigned participant number. Data collected from all participants will be entered into a database called RedCap for analysis. Again, no identifying information that could link you to participation in this study will be included in this database.

You should know, however, that there are some circumstances in which we may have to show your information to other people. Officials of the University of Kentucky may look at or copy pertinent portions of records that identify you.

CAN YOUR TAKING PART IN THE STUDY END EARLY?

If you decide to take part in the study, you still have the right to decide at any time that you no longer want to continue. You will not be treated differently if you decide to stop taking part in the study.

If you choose to withdraw from the study early, the data collected until that point will remain in the study database and may not be removed.

ARE YOU PARTICIPATING OR CAN YOU PARTICIPATE IN ANOTHER RESEARCH STUDY AT THE SAME TIME AS PARTICIPATING IN THIS ONE?

You may take part in this study if you are currently involved in another research study. It is important to let the investigator/your doctor know if you are in another research study. You should also discuss with the investigator before you agree to participate in another research study while you are enrolled in this study.

WHAT HAPPENS IF YOU GET HURT OR SICK DURING THE STUDY?

If you believe you are hurt or if you get sick because of something that is due to the study, you should call Debra Suiter at 859-218-5323 immediately.

It is important for you to understand that the University of Kentucky does not have funds set aside to pay for the cost of any care or treatment that might be necessary because you get hurt or sick while taking part in this study. Also, the University of Kentucky will not pay for any wages you may lose if you are harmed by this study.

The medical costs related to your care and treatment because of research related harm will be your responsibility. You do not give up your legal rights by signing this form.

WILL YOU RECEIVE ANY REWARDS FOR TAKING PART IN THIS STUDY?

You will not get any rewards or payment for taking part in this study.

WHAT IF YOU HAVE QUESTIONS, SUGGESTIONS, CONCERNS, OR COMPLAINTS?

Before you decide whether to accept this invitation to take part in the study, please ask any questions that might come to mind now. Later, if you have questions, suggestions, concerns, or complaints about the study, you can contact the investigator, Debra Suiter at 859-218-5323. If you have any questions about your rights as a volunteer in this research, contact the staff in the Office of Research Integrity at the University of Kentucky between the business hours of 8am and 5pm EST, Mon-Fri at 859-257-9428 or toll free at 1-866-400-9428. We will give you a signed copy of this consent form to take with you.

WHAT IF NEW INFORMATION IS LEARNED DURING THE STUDY THAT MIGHT AFFECT YOUR DECISION TO PARTICIPATE?

If the researcher learns of new information in regards to this study, and it might change your willingness to stay in this study, the information will be provided to you. You may be asked to sign a new informed consent form if the information is provided to you after you have joined the study.

WHAT ELSE DO YOU NEED TO KNOW?

There is a possibility that the data collected from you may be shared with other investigators in the future. If that is the case the data will not contain information that can identify you unless you give your consent/authorization or the UK Institutional Review Board (IRB) approves the research. The IRB is a committee that reviews ethical

issues, according to federal, state and local regulations on research with human subjects, to make sure the study complies with these before approval of a research study is issued.

AUTHORIZATION TO USE OR DISCLOSE YOUR IDENTIFIABLE HEALTH INFORMATION

The privacy law, HIPAA (Health Insurance Portability and Accountability Act), requires researchers to protect your health information. The following sections of the form describe how researchers may use your health information.

Your health information that may be accessed, used and/or released includes:

- Gender, age, ALS-Functional Rating Scale data (which include information on speech, swallowing, and salivation), weight, height, body mass index, measures of lung function (forced vital capacity)

The Researchers may use and share your health information with:

- The University of Kentucky's Institutional Review Board/Office of Research Integrity.
- Law enforcement agencies when required by law.
- University of Kentucky representatives.
- UK Hospital

The researchers agree to only share your health information with the people listed in this document.

Should your health information be released to anyone that is not regulated by the privacy law, your health information may be shared with others without your permission; however, the use of your health information would still be regulated by applicable federal and state laws.

You may not be allowed to participate in the research study if you do not sign this form. If you decide not to sign the form, it will not affect your:

- **Current or future healthcare at the University of Kentucky**
- **Current or future payments to the University of Kentucky**
- **Ability to enroll in any health plans (if applicable)**
- **Eligibility for benefits (if applicable)**

After signing the form, you can change your mind and NOT let the researcher(s) collect or release your health information (revoke the Authorization). If you revoke the authorization:

- You will send a written letter to: Debra Suiter, 900 S. Limestone Avenue, CTW 124D, Lexington, KY 40536 to inform her of your decision.
- Researchers may use and release your health information **already** collected for this research study.
- Your protected health information may still be used and released should you have a bad reaction (adverse event).

The use and sharing of your information has no time limit.

If you have not already received a copy of the Privacy Notice, you may request one. If you have any questions about your privacy rights, you should contact the University of Kentucky's Privacy Officer between the business hours of 8am and 5pm EST, Mon-Fri at: (859) 323-1184.

You are the subject or are authorized to act on behalf of the subject. You have read this information, and you will receive a copy of this form after it is signed.

_____
Signature of Research Subject or Witness

_____
Printed name of research subject and Witness (if applicable)

_____ _____
Name of [authorized] person obtaining informed consent/HIPAA authorization Date

_____
Signature of Principal Investigator or Sub/Co-Investigator

Appendix C: Modified Caregiver Strain Index (MCSI)

Modified Caregiver Strain Index

Directions: Here is a list of things that other caregivers have found to be difficult. Please put a checkmark in the columns that apply to you. We have included some examples that are common caregiver experiences to help you think about each item. Your situation may be slightly different, but the item could still apply.

	Yes, On a Regular Basis=2	Yes, Sometimes =1	No=0
My sleep is disturbed (For example: the person I care for is in and out of bed or wanders around at night)	_____	_____	_____
Caregiving is inconvenient (For example: helping takes so much time or it's a long drive over to help)	_____	_____	_____
Caregiving is a physical strain (For example: lifting in or out of a chair; effort or concentration is required)	_____	_____	_____
Caregiving is confining (For example: helping restricts free time or I cannot go visiting)	_____	_____	_____
There have been family adjustments (For example: helping has disrupted my routine; there is no privacy)	_____	_____	_____
There have been changes in personal plans (For example: I had to turn down a job; I could not go on vacation)	_____	_____	_____
There have been other demands on my time (For example: other family members need me)	_____	_____	_____
There have been emotional adjustments (For example: severe arguments about caregiving)	_____	_____	_____
Some behavior is upsetting (For example: incontinence; the person cared for has trouble remembering things; or the person I care for accuses people of taking things)	_____	_____	_____
It is upsetting to find the person I care for has changed so much from his/her former self (For example: he/she is a different person than he/she used to be)	_____	_____	_____
There have been work adjustments (For example: I have to take time off for caregiving duties)	_____	_____	_____
Caregiving is a financial strain	_____	_____	_____
I feel completely overwhelmed (For example: I worry about the person I care for; I have concerns about how I will manage)	_____	_____	_____

[Sum responses for "Yes, on a regular basis" (2 pts each) and "yes, sometimes" (1 pt each)]

Total Score =

Appendix D: Eating Assessment Tool (EAT-10)

EATING ASSESSMENT TOOL (EAT-10)

Name: _____ _____ EMR# _____

Birthdate/Age: _____ Todays Date: _____

The purpose of the EAT-10 questions is to help measure swallowing problems. Answer each question by circling the number that matches how bad you feel the problem is for you.

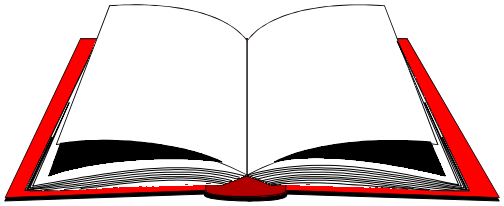
To what degree to you experience the following problems? **Circle an answer between 0 and 4**	0 = No problem				4 = Severe problem
1. My swallowing problem has caused me to lose weight.	0	1	2	3	4
2. My swallowing problem interferes with my ability to go out for meals.	0	1	2	3	4
3. Swallowing liquids takes extra effort.	0	1	2	3	4
4. Swallowing solids takes extra effort.	0	1	2	3	4
5. Swallowing pills takes extra effort.	0	1	2	3	4
6. Swallowing is painful.	0	1	2	3	4
7. The pleasure of eating is affected by my swallowing.	0	1	2	3	4
8. When I swallow food sticks in my throat.	0	1	2	3	4
9. I cough when I eat.	0	1	2	3	4
10. Swallowing is stressful	0	1	2	3	4
Add up the sum of the numbers you circled for a TOTAL EAT-10 Score:					

If your score is greater than 3 you may have swallowing problems. We suggest that you share your EAT-10 results with your doctor.

Reference: Belafky PC, Mouadeb DA, Rees CJ, Pryor JC, Postma GN, Alen J and Leonard RJ. Validity and reliability of the Eating Assessment Tool (EAT-10). *Ann Otol Rhinol Laryngol* 2008; 117(12):919-924.

Appendix E: Swallowing-Related Quality of Life Instrument (SWAL-QOL)

The SWAL-QOL SURVEY



Understanding Quality of Life in Swallowing Disorders

IMPORTANT NOTE: We understand that you may have a number of physical problems. Sometimes it is hard to separate these from swallowing difficulties, but we hope that you can do your best to concentrate ***only*** on your ***swallowing problem.*** Thank you for your efforts in completing this questionnaire.

1. Below are some general statements that people with ***swallowing problems*** might mention. In the last month, **how true** have the following statements been for you.

(circle one number on each line)

	Very much true	**Quite a bit true**	**Somewhat true**	**A little true**	**Not at all true**
Dealing with my swallowing problem is very difficult.	1	2	3	4	5
My swallowing problem is a major distraction in my life.	1	2	3	4	5

2. Below are aspects of day-to-day eating that people with ***swallowing problems*** sometimes talk about. In the last month, **how true** have the following statements been for you?

(circle one number on each line)

	Very much true	**Quite a bit true**	**Somewhat true**	**A little true**	**Not at all true**
Most days, I don't care if I eat or not.	1	2	3	4	5
It takes me longer to eat than other people.	1	2	3	4	5
I'm rarely hungry anymore.	1	2	3	4	5
It takes me forever to eat a meal.	1	2	3	4	5
I don't enjoy eating anymore.	1	2	3	4	5

3. Below are some physical problems that people with ***swallowing problems*** sometimes experience. In the last month, **how often** you have experienced each problem as a result of your swallowing problem?

(circle one number on each line)

	Almost always	**Often**	**Sometimes**	**Hardly ever**	**Never**
Coughing	1	2	3	4	5
Choking when you eat food	1	2	3	4	5
Choking when you take liquids	1	2	3	4	5
Having thick saliva or phlegm	1	2	3	4	5
Gagging	1	2	3	4	5
Drooling	1	2	3	4	5
Problems chewing	1	2	3	4	5
Having excess saliva or phlegm	1	2	3	4	5
Having to clear your throat	1	2	3	4	5
Food sticking in your throat	1	2	3	4	5
Food sticking in your mouth	1	2	3	4	5
Food or liquid dribbling out of your mouth	1	2	3	4	5
Food or liquid coming out your nose	1	2	3	4	5
Coughing food or liquid out of your mouth when it gets stuck	1	2	3	4	5

4. Next, please answer a few questions about how your ***swallowing problem*** has affected your diet and eating in the last month.

(circle one number on each line)

	Strongly agree	**Agree**	**Uncertain**	**Disagree**	**Strongly disagree**
Figuring out what I can and can't eat is a problem for me.	1	2	3	4	5
It is difficult to find foods that I both like and can eat.	1	2	3	4	5

5. In the last month, **how often** have the following statements about communication applied to you because of your ***swallowing problem***?

(circle one number on each line)

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
People have a hard time understanding me.	1	2	3	4	5
It's been difficult for me to speak clearly.	1	2	3	4	5

6. Below are some concerns that people with ***swallowing problems*** sometimes mention. In the last month, **how often** have you experienced each feeling?

(circle one number on each line)

	Almost always	Often	Sometimes	Hardly ever	Never
I fear I may start choking when I eat food.	1	2	3	4	5
I worry about getting pneumonia.	1	2	3	4	5
I am afraid of choking when I drink liquids.	1	2	3	4	5
I never know when I am going to choke.	1	2	3	4	5

7. In the last month, how often have the following statements **been true** for you because of your ***swallowing problem***?

(circle one number on each line)

	Always true	Often true	Sometimes true	Hardly ever true	Never true
My swallowing problem depresses me.	1	2	3	4	5
Having to be so careful when I eat or drink annoys me.	1	2	3	4	5
I've been discouraged by my swallowing problem.	1	2	3	4	5
My swallowing problem frustrates me.	1	2	3	4	5
I get impatient dealing with my swallowing problem.	1	2	3	4	5

8. Think about your social life in the last month. How strongly would you agree or disagree with the following statements?

(circle one number on each line)

	Strongly agree	**Agree**	**Uncertain**	**Disagree**	**Strongly disagree**
I do not go out to eat because of my swallowing problem.	1	2	3	4	5
My swallowing problem makes it hard to have a social life.	1	2	3	4	5
My usual work or leisure activities have changed because of my swallowing problem.	1	2	3	4	5
Social gatherings (like holidays or get-togethers) are not enjoyable because of my swallowing problem.	1	2	3	4	5
My role with family and friends has changed because of my swallowing problem.	1	2	3	4	5

9. In the last month, **how often** have you experienced each of the following physical symptoms?

(circle one number on each line)

	All of the time	**Most of the time**	**Some of the time**	**A little of the time**	**None of the time**
Feel weak?	1	2	3	4	5
Have trouble falling asleep?	1	2	3	4	5
Feel tired?	1	2	3	4	5
Have trouble staying asleep?	1	2	3	4	5
Feel exhausted?	1	2	3	4	5

10. Do you now take any food or liquid through a feeding tube?

(circle one)

No 1

Yes..... 2

11. Please circle the letter of the one description below that best describes the consistency or texture of the food you have been eating most often in the last week.

Circle one:

A. Circle this one if you are eating a full normal diet, which would include a wide variety of foods, including hard to chew items like steak, carrots, bread, salad, and popcorn.

B. Circle this one if you are eating soft, easy to chew foods like casseroles, canned fruits, soft cooked vegetables, ground meat, or cream soups.

C. Circle this one if you are eating food that is put through a blender or food processor or anything that is like pudding or pureed foods.

D. Circle this one if you take most of your nutrition by tube, but sometimes eat ice cream, pudding, apple sauce, or other pleasure foods.

E. Circle this one if you take all of your nourishment through a tube.

12. **Please circle the letter** of the one description below that best describes the consistency of liquids you have been drinking most often in the last week.

Circle one:

A. Circle this if you drink liquids such as water, milk, tea, fruit juice, and coffee.

B. Circle this if the majority of liquids you drink are thick, like tomato juice or apricot nectar. Such thick liquids drip off your spoon in a slow steady stream when you turn it upside down.

C. Circle this if your liquids are moderately thick, like a thick milkshake or smoothie. Such moderately thick liquids are difficult to suck through a straw, like a very thick milkshake, or drip off your spoon slowly drop by drop when you turn it upside down, such as honey.

D. Circle this if your liquids are very thick, like pudding. Such very thick liquids will stick to a spoon when you turn it upside down, such as pudding.

E. Circle this if you did not take any liquids by mouth or if you have been limited to ice chips.

13. In general, would you say your health is:

(circle one)

Poor	1
Fair	2
Good	3
Very Good	4
Excellent	5

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