2011

A National Study of Amyotrophic Lateral Sclerosis Multidisciplinary Clinic Utilization

Jared L. Young
Philadelphia College of Osteopathic Medicine, jaredyo@pcom.edu

Philadelphia College of Osteopathic Medicine

Department of Psychology

A NATIONAL STUDY OF AMYOTROPHIC LATERAL SCLEROSIS MULTIDISCIPLINARY CLINIC UTILIZATION

By Jared L. Young

Submitted in Partial Fulfillment of the Requirements of the Degree of

Doctor of Psychology

June 2011

PHILADELPHIA COLLEGE OF OSTEOPATHIC MEDICINE
DEPARTMENT OF PSYCHOLOGY

Dissertation Approval

This is to certify that the thesis presented to us by Jared L. Young, MSW, MS on the 21st day of June, 2011, in partial fulfillment of the requirements for the degree of Doctor of Psychology, has been examined and is acceptable in both scholarship and literary quality.

Committee Members' Signatures:

Stephanie H Felgoise PhD, Chairperson

Donald P Masey, PsyD

Zachary Simmons, MD

Robert A DiTomasso, PhD, Chair, Department of Psychology

Acknowledgments

This dissertation was made possible with the guidance of committee members, family, colleagues, and friends. The patience and expertise of dissertation chair, Dr Stephanie Felgoise, was invaluable. Support of Dr Zachary Simmons and Dr Donald Masey provided understanding and mentoring. Special thanks to Kristen Labin and Sara Martin, who generously assisted with data analysis. To the admissions interviewers and faculty at Philadelphia College of Osteopathic Medicine, thank you for being the first step in this journey.

I would like to extend sincerest thanks to my friends and family for enduring constant educational conversations and offering endless encouragement. Most importantly, I would like to express my deepest gratitude to my exceptional wife, Allison Lynn Young, my most passionate supporter and unwavering confidant. To my wonderful daughter, Lauren, thank you for your patience and endless smiles. To my father, who passed away during the course of my educational pursuits; thank you for being an amazing model of how to be a better student, father, husband, and Christian.

I would like to dedicate this manuscript to the memory of my father, Dr Jerry R. Young. His guidance throughout my life has provided a foundation to always trust in God.

Abstract

There is no cure for the fatal progression of Amyotrophic Lateral Sclerosis (ALS). Optimization of quality of life is the primary ALS treatment goal (Simmons, 2005). Consolidating multiple appointments into one visit is advantageous due to the severity of muscular deterioration and mobility problems associated with the disease. Multidisciplinary Clinics (MDC) provide improved care coordination, accessibility to health care professionals skilled in treating ALS, and improvements in symptom control for patients with ALS.

The purpose of this study is to discuss the differences in quality of life, physical health status, and coping skills for individuals with ALS attending multidisciplinary clinics versus those receiving traditional, practitioner driven care. The investigation is part of a larger program of research designed to identify and address the psychosocial needs of individuals with ALS. The literature is void regarding factors connected to the reasons why individuals choose to attend an MDC, compared with receiving traditional practitioner driven care. Grounded theory was utilized to analyze and compare data by coding categories related to patient choice to utilize a multidisciplinary clinic or to utilize traditional care. This approach is specifically designed to provide supporting evidence that multidisciplinary clinics optimize quality of life for patients with ALS. This study was conducted through an online survey; 403 people initiated the survey, and 329 met inclusion criteria. Individuals who attend ALS clinics perceive Quality Of Life (QOL) as being higher in physical function and bulbar function, although those patients have lower levels of physical function, as measured by the ALSFRS-R. Thus, individuals attending a MDC differ in this survey from those who do not attend.

Table of Contents

Chapter 1

Statement of the Problem and Purpose of the Study

Amyotrophic lateral sclerosis (ALS), characterized by paralysis and death within three to five years of diagnosis, is a neurodegenerative disease for which there is no cure (Simmons, 2005). Symptomatic treatments are important in controlling major consequences of ALS; these include pain, sleep disorders, spasticity, emotional liability, and depression (Simmons, 2005). Multidisciplinary clinics (MDC) cater exclusively to patients with ALS to provide coordinated treatment and formulate interventions to improve quality of life (Zoccolella et al., 2007).

Multidisciplinary clinics offer a coordinated approach to care, utilizing specialty teams of neurology, nursing, physical therapy, occupational therapy, speech therapy, pulmonology, nutrition, psychology, social work, respiratory therapy, and representatives from ALS associations. Clinic staff works with outside health care professionals, home health care aides, and hospice caregivers to provide complete care. Consolidating multiple appointments into one visit is advantageous for ALS patients due to mobility problems associated with the disease and also for maximal interdisciplinary collaboration.

There is no cure for the fatal progression of ALS. Primary treatment goals for patients with ALS must include optimization of quality of life. Multidisciplinary clinics provide improved care coordination, accessibility to health care professionals skilled in treating ALS, and improvement in symptom control. These clinics also benefit caregivers of ALS patients by providing support and encouragement.

Individuals with ALS may choose to attend a MDC for personal reasons, health care beliefs, or other factors. ALS patients may perceive the fact that the MDC provides not only access to physicians specially trained in ALS, but also access to research and new treatments. Traynor (2003) found a significant increase in survival rates for ALS patients who attended a MDC. The prospective population-based study found that patients enrolled in a multidisciplinary ALS clinic cohort had a mean increase survival of 7.5 months longer than patients who used the general neurology clinic. The results suggest the multidisciplinary approach to care is associated with improved survival rates.

In a sample study designed to evaluate patient quality of life, Van den Berg (2005) found multidisciplinary clinics improve social functioning and mental health. The study sampled 208 patients with ALS; 133 patients attended a multidisciplinary clinic and 75 patients received general ALS care. The percentage of individuals who received adequate aids and appliances was higher in patients receiving care from a multidisciplinary clinic, compared with patients receiving general care. The results indicate patient quality of life is improved in multidisciplinary clinics as a result of increased access to services and referrals.

Current research does not document the reasons why individuals choose multidisciplinary clinics over other traditional practitioner driven care. Expert opinion in the field and research suggests that attending a MDC improves care, survival rates, and QOL: The reality is that many individuals choose not to receive care in these settings, and the research is void in describing the factors that contribute to ALS patients' utilization of MDC. Information from ALS patients is valuable in determining the factors that influence individual patient choice to utilize MDC. In a broader context, the information

validates the establishment of amyotrophic lateral sclerosis multidisciplinary clinics, and generalizes the clinic approach to other areas of the medical field. The focus of the current qualitative study is to understand the reasons why patients utilize the multidisciplinary clinic approach, the perceived benefits from this system of care, and to identify barriers for those who do not attend. In addition to qualitative descriptions, a series of responses from qualitative questions are analyzed to identify psychological, social, and support needs of ALS patients. Analyses may include recommendations for medical practitioners regarding provision of care or resources to improve quality of life, to decrease depression and anxiety, and to improve the current MDC approach.

The investigation is part of a larger program of ALS research designed to identify and address the psychosocial needs of individuals with ALS, their caregivers, and families. In 2008 the Penn State Milton S. Hershey Medical Center conducted a population-based study of the quality of life of individuals diagnosed with ALS. The study was designed to compare the QOL, differences in states of health of individuals who attended MDC with those who did not. The study was on-line, but a paper copy questionnaire was also available. Participants provided demographic information, completed the ALS Functional Rating Scale Revised, and the ALS Specific Quality of Life Revised measure. Four hundred and twenty-five individuals responded to the survey, 384 in USA, and 41 worldwide; 68% attended ALS clinic and 32% did not. A primary outcome was that there were no significant differences in the QOL reports by individuals with ALS who attend MDC, compared with those who do not attend a clinic. Future directions from that study were to evaluate the impact that a MDC has on patient QOL, and the reasons why patients attend an MDC versus stand alone care for their treatment.

A qualitative research approach was suggested in order to inquire about the reasons why individuals attend or do not attend a MDC through an open ended question.

This study is designed to answer questions raised from the original research completed at Penn State Milton S. Hershey Medical Center to understand the reasons why people choose different forms of care. The previous study at Penn State Milton S. Hershey Medical Center identified the fact that patients who attended a MDC were more likely to use supportive care of non invasive ventilator support, power wheel chairs, be administered oral Riluzole, and participate in experimental trials. The non-clinic patients receiving palliative care in the sample were more likely to receive no treatment and have a tracheotomy performed. The two groups were significantly different in medical interventions.

This study intends to understand the differences between groups. Collecting qualitative data will allow the researcher to perform comparisons between and within groups to identify each incident for similarities and differences for themes of acceptance of the disease, and other salient factors that individuals use to determine medical need, and maintain and improve QOL. The source of QOL may not be the medical interventions received at the MDC; it may rather involve some personality trait associated with acceptance and problem solving of the individual. This qualitative approach allows the researcher to differentiate one theme from another, and identify specific properties of a theme. The use of comparisons provides a meaning of events that might otherwise seem obscure, and discover variations as well as general patterns to generate new knowledge (Corbin & Strauss, 2008).

Chapter 2

Literature Review

Etiology of Amyotrophic Lateral Sclerosis

Amyotrophic lateral sclerosis is a neurodegenerative disease affecting motor neurons, characterized by paralysis and death within three to five years from time of diagnosis (Simmons, 2005; National Institute for Neurological Disorders and Stroke, (NINDS). There is no effective cure for ALS, the most common motor neuron disease, which is progressive and invariably fatal (Rocha, 2005). ALS, which targets nerve cells, is responsible for voluntary muscles (Rocha, 2005), and is hallmarked by gradual decline in function of all muscles under voluntary control and loss of ability to move arms, legs and body (NINDS). The onset of ALS is characterized by subtle symptoms frequently overlooked and includes twitching, cramping, stiffness of muscles, muscle weakness affecting an arm or a leg slurred speech and difficulty chewing or swallowing. The sequence of emerging symptoms and rate of disease progression varies; eventually patients can not ambulate and report loss in the use of extremities. Individuals develop difficulty swallowing and chewing, thus impairing their ability to eat normally, causing weight loss. ALS patients may die as a result of respiratory failure when the muscles in the diaphragm and chest wall fail, causing an inability to breathe without ventilator support (NINDS).

Prevalence rates. Annual incidence of ALS is 1 to 2 per 100,000 population and the prevalence is 6 per 100,000 (Rocha, 2005). A significant amount of ALS cases are sporadic; and five to ten percent are familial. ALS occurs in adults, with the highest frequency of onset between 40 and 60 years of age (NINDS). Male to female ratio is

1.5:1, and reaches unity at age 70 (Rocha, 2005). Statistics show that 20,000 Americans have ALS, and an estimated 5,000 people in the United States are diagnosed with the disease each year (Corcia & Meininger, 2008; Mitchel, & Borasio, 2007). ALS is a prevalent neuromuscular diseases worldwide, and people of all races and ethnic backgrounds are affected (Corcia & Meininger, 2008; NINDS). ALS occurs apparently at random with no associated risk factors in 90-95 percent cases (Corcia & Meininger, 2008; NINDS). Patients do not have a family history of the disease, and their family members are not considered to be at increased risk for developing ALS.

Treatment. Riluzole is the only medication that the Food and Drug Administration (FDA) has approved for the treatment of ALS (Simmons, 2005; Radunociv, Mitsumoto & Leigh, 2007). Riluzole, which is believed to reduce damage to motor neurons by decreasing the release of glutamate, (Simmons, 2005) does not reverse the damage already done to motor neurons. Clinical trials with ALS patients show that Rilozole prolongs survival by several months, and prolongs time prior to patient need for ventilation support (Radunociv et. al. 2007).

End of life issues. ALS is a progressive and terminal disease, and patients will experience inadequate nutrition, respiratory insufficiency, increasing discomfort, and psychological distress (Averill, Kasarskis & Segerstrom, 2007). Providing ALS patients comfort and a peaceful death is a primary goal during the terminal phase. End of life care is provided in various settings, dependent upon patient preferences, skills, abilities, and caregiver choice (Shoesmith & Strong, 2006). Individuals may choose to reside in hospital settings, hospice, or die at home. Shoesmith and Strong (2006) state that 50 percent of individuals die at home, with hospice support, as a result of respiratory failure.

Palliative care interventions can alleviate air hunger by providing oxygen; can provide anxiety treatment with benzodiazepines, and provide nausea treatment with antiemetics (Shoesmith & Strong, 2006).

Albert and colleagues, (2005) examined the wish to die in a prospective study of ALS patients. Participants with advanced ALS had a likelihood of death within six months. Patients and care givers were interviewed monthly with a psychosocial assessment and depression screening tool. Eighty participants were enrolled in the study; 53 died during the course of the investigation; ten expressed a wish to die, and three hastened their deaths (Averill, Kasarskis, & Segerstrom 2007). Individuals expressing a wish to die met diagnostic criteria for depression; however, differences were less when questions regarding suicide were excluded from the depression screening tool. Patients indicating a wish to die verbalized less optimism, less religiosity, and greater hopelessness. The author suggests that the wish to die is not linked solely to depression, and is a broader syndrome of "end of life despair" described by McDonald et al. (1994), which include suffering, loss of interest, absence of pleasure, and pessimism. Individuals hastening death reported increased control ratings and ratings of suffering decreased in the final weeks of life. Albert et al. (2005) concludes that gaining control over dying brought mental health benefits and an element of control over ALS.

Albert et al. (2005) found that patients expressing the wish to die were less likely to use nasal ventilation, and patients hastening death did not use nasal ventilation. Greater than 50% of the individuals who did not express a wish to die utilized the device. Ventilation devices do not substantially extend survival in ALS patients, yet use of ventilators may indicate a patient's interest in living.

Averill et al. (2007) discusses a continuum in which resilience and ability to find meaning in life are at one end, and psychological distress, hopelessness and pessimism are at the other end of the continuum, based on specific patient personalities, coping mechanisms, and protective factors; these include: social support, socioeconomic status, and history of depression or other mental health disorders. At the time of diagnosis premorbid conditions are magnified, suggesting that health care practitioners need to evaluate patient mental health status prior to an ALS diagnoses to determine, accurately, mental health needs, and provide appropriate interventions impacting QOL.

Depression. ALS does not typically impair mind or intelligence; however, several studies suggest that ALS patients may have alterations in cognitive functions of decision making and memory (Simmons, 2005). Awareness of progressive loss of function leads to anxiety and depression, and lower quality of life (NINDS; Kurt, Nijboer, Matuz, & Kubler, 2007). Kurt and colleagues (2007) found the prevalence rates for depression in ALS patients ranging from 0% to 44%. Reported prevalence rates of depression vary, leading researchers to question the traditional depression screening tools used to diagnose depression in ALS patients (Kurt et al. 2007; Simmons, 2005). Diagnosing of depression is complicated because somatic complaints of loss of appetite, weight loss, psychomotor retardation, insomnia, and hypersomnia are symptoms resulting from ALS, but also mimic signs of depression. Averill et al. (2007) conclude that clinical depression may not be as prevalent for vulnerable populations as initially suspected by health care practitioners, although patients with depression and anxiety have lower quality of life, psychological distress, and lower satisfaction with life. (Vignola et al. 2008).

Multidisciplinary Clinics

Progression of ALS cannot be cured or stopped. The primary treatment goal for patients with ALS is the optimization of quality of life. Multidisciplinary clinics provide an interdisciplinary approach to care in order to promote the highest quality of life. A multidisciplinary clinic has the availability of different professionals and perspectives, providing improved care coordination and easing accessibility for patients, thought to lead to an improvement in symptom control and improved quality of life. Multidisciplinary clinics are hypothesized to benefit caregivers and are encouraged for ALS patients in order to experience improved quality of life.

Cancer multidisciplinary clinics. Current and emerging research supports the efficacy and use of MDC across a wide range of medical specialties. Ducharme and Colleagues (2005) found usual care, compared with a MDC specializing in congestive heart failure, reduced the number of hospital readmissions and hospital days, and improved patients' quality of life. Ducharme et al. (2005) randomly assigned 230 patients to standard care and an MDC specializing in heart failure. The MDC group of patients received access to cardiologists and allied health professionals, and at the end of a six month period, fewer patients in the intervention group required readmission (Ducharme et al., 2005). Both groups were given the Minnesota Living with Heart Failure questionnaire. The interventions group had substantial improvement in emotional and physical quality of life scores, compared with the control group (Ducharme et al. 2005).

Gabel and colleagues (1997) found that MDC for breast cancer increased patient satisfaction by including family members and friends in the decision making process, and decreased the amount of time from diagnoses to treatment. Anonymous questionnaires

were used to evaluate patient satisfaction of 177 patients in the intervention group who received care in a MDC setting, compared with 162 patients in a control group who were evaluated and treated at a hospital (Gabel, Hilton, & Nathanson, 1997). A retrospective chart review was completed on both groups to determine the amount of time between diagnoses and treatment. The study found increased patient satisfaction for individuals enrolled in a MDC, compared with the control group who were receiving traditional care, supporting the conclusion that MDC improve patient QOL.

ALS Multidisciplinary Clinics. A single study conducted in Southern Italy found that a multidisciplinary approach to care did not improve ALS survival rates (Zoccolella et al., 2005). Of 126 patients registered over a two year period, 84 were seen in a multidisciplinary clinic and 42 in general neurology clinics. There were no significant differences in onset, age, sex distribution, or interventions between the two groups, although more people attending multidisciplinary clinic received Riluzole. The median survival times from symptom onset were 26 months versus 32.3 or diagnosis 17.6 months versus 18 (Zoccolella et al., 2005). The results were not significantly different, and analyzing those with bulbar onset independently made no difference (Zoccolella et al., 2005).

Traynor and colleagues (2003) studied 345 Irish residents diagnosed with suspected to definite ALS from January 1996 to December 2000. Traynor found 24 percent of the patients used the multidisciplinary ALS clinic, and the remaining patients accessed general neurology clinics. The prospective population-based study showed that patients enrolled in a multidisciplinary ALS clinic cohort had a mean increase survival of 7.5 months longer than for patients using general neurology clinics (Traynor et al. 2003).

The multidisciplinary cohort received Riluzole and had more improved prognoses than patients attending a general neurology clinic. Results suggest the multidisciplinary approach to care is associated with improved survival rate. Patients with bulbar onset disease benefited form the aggressive management received in clinic type settings.

Van den Berg and colleagues (2005) found that multidisciplinary approaches to patient care improve quality of life, result, prominently in social functioning and mental health. The study sampled 208 patients with ALS; 133 patients attended a multidisciplinary clinic and 75 received general ALS care. No differences were found on the physical functioning scale; however, the percentage of patients who received adequate aids and appliance, compared with those patients not involved in an ALS clinic, was higher in patients in a multidisciplinary clinic. Results indicate patient quality of life is improved in multidisciplinary clinics as a result of increased access to services and referrals, and higher standard of care.

Local clinical scientist. Stricker (2006) defines the Local Clinical Scientist (LCS) model as a view of each clinical interaction as a research project; clinicians observe the effects, learn from observation and data, and apply learning to the next patient or research project. The treatment of individuals with ALS is complex, and requires the management of medical problems, mental health and social issues and multifaceted disabilities. As a result, the multidisciplinary approach to care has become more preferable. The goals of MDCs are to provide optimal care and basic research about ALS (Simmons, 2005).

Expert opinion considers the clinic approach as improving quality of life. Themes concerning reasons why individuals use this approach over traditional care provided by a

community physician are unknown. A local clinical scientist model allows the clinician working with an ALS patient at a multidisciplinary clinic to understand the salient factors that underlie the reasons why many use the clinic for care, and how this approach improves quality of life. LCS models provide the framework to inquire when the unique problem has no clear answers provided by current research. LCS models seek affirmation or contradictory evidence to the research question, and responds to the needs of the community (Stricker, 2006). Clinicians review each response from the patient as a research project and makes clinical changes when supporting evidence is obtained (Stricker, 2006).

Research on the reasons why ALS patients utilize a multidisciplinary clinic is limited, and leads the practitioner to question how this approach is better than treatment in other systems. Understanding benefits of a multidisciplinary clinic provides valuable information about the reasons why people use multidisciplinary care, and may be generalized to other disease models currently using this model, or considering the use of this delivery system. Practitioners in local settings are charged with combining observations and existing research to develop effective approaches (Stricker & Trierweiler, 2006). Flexibility provides community members with the ability to generate research questions and collaborate with researchers. Collection and analysis of data are carried out jointly, and the ALS community drives how knowledge will be used and new practices implemented.

Current research has not provided a clear understanding about the reasons why ALS patients utilize MDCs, leading local clinical scientists to attempt to understand the variance in data and explore this question. The qualitative method helps to bridge the gap

in quantitative research, and provides the medical community evidence-based practices. The information is valuable in determining how individuals select medical services and in a broader context validates the establishment of ALS clinics, and generalizes the approach to other areas of the medical field. A local clinical scientist model guides new provision of care and utilization of resources to improve quality of life.

Chapter 3

Hypothesis and Research Question

This research project was guided by the question of the reasons why ALS patients attend or do not attend a MDC, reasons for continued attendance, and what factors would influence attending a MDC. It was hypothesized that the reasons why an individual seeks care at a multidisciplinary clinic is to improve or influence patient quality of life through additional services and resources, durable medical equipment, access to mental health services, and access to clinical trials at a much higher rate. The study examined through open ended questions, the characteristics of individuals who do, or do not access care at multidisciplinary clinics, and explored what additional services, referrals, and treatments are offered. Individuals accessing MDC may have different coping styles that influence how they choose to receive care. Information from individuals affected by ALS would supply valuable data to practitioners, guide future interventions, and supply a new facet of quality of life to the clinical community. Based on these variables, the following were the proposed study hypotheses.

Hypotheses (H)

H1: Participants who attend an ALS MDC will have better problem solving skills than individuals who do not attend, based on Social Problem Solving Inventory-R.

H2: Participants who attend an ALS MDC will have overall better QOL than individuals who do not attend

H3: Participants who attend an ALS clinic will have higher levels of physical function than individuals who do not attend.

H4: Participants who attend a MDC will receive more treatments and services for maintaining QOL, compared with non-attendees.

Chapter 4

Methodology

Overview

This study utilized a mixed method of qualitative and quantitative approaches to study the differences between individuals diagnosed with ALS who access MDC and those who do not. Precise focus was on perceived QOL, functional rating, and social problem solving coping skills. Previous research has shown no difference between attendees and non-attendees with regard to QOL and physical functioning (Stephens, Walsh, & Simmons, 2008) Social problem solving skills have been shown to affect negative emotion of ALS patients and predict QOL in ALS caregivers (Murphy, Felgoise, Walsh, & Simmons, 2009). This study replicated the previous study, with the addition of evaluating social problem solving to determine if rational and systematic approach to problem-solving and decision-making may affect the personal choice of health care delivery or differentiate between those with higher and lower quality of life. Qualitative information was obtained to understand and explore the reasons why people choose to attend or not attend a MDC.

Qualitative information was obtained from participants who completed a series of open ended questions designed to solicit information regarding MDC utilization: 1) Why do you go to a MDC?, 2) What are the advantages and disadvantages of receiving care from a MDC?, 3) If you do not go to a MDC, please tell us the reasons why you do not, and 4) What are the circumstances in which a person not attending a clinic would consider receiving care at this type of center? The participants were asked objective questions concerning those who influenced their decisions to attend a clinic, how they

became aware of these services, what was their traveling distance to the clinic, and what additional services, referrals, and treatments were offered by attending an ALS clinic. Participants provided standard demographic information, including: 1) age, 2) gender, 3) ALS symptom onset, 4) site of ALS symptom onset, 5) date of ALS diagnoses, and 6) state, zip code, and country.

The study was conducted in the form of an on-line internet survey. The web based survey hosted on the internet utilized the survey software, "Survey Monkey". Control over who accessed the survey and the number of people completing the survey was limited. Penn State Milton S. Hershey Medical Center hosted the survey, and after completion of the survey, participants' web browser was returned to the homepage of the Penn State MSHMC ALS Clinic (http://www.alsphiladelphia .org/pennstatehershey/).

Design and Design Justification

Not all patients with ALS receive medical treatments from a MDC, opting to be treated by neurologists and health care practitioners outside of the MDC system. This study was designed to understand and explore various aspects of human experience in determining health care decisions. A mixed method approach of quantitative and qualitative research provides openness to new and unexpected discoveries emerging from the data, openness to the unexpected, coupled with a willingness to redirect innovative research as new insights emerge. The approach was intended to provide narrative accounts and interpretations, and sought to better understand how individuals make health care decisions, thus providing health care practitioners with valuable information in determining treatment options, and conceivably enhancing QOL. Quantitative

components of the study sought to identify potential differences between attendees and non-attendees' QOL, physical function, and social problem-solving coping skills.

Design and Design Justification

This was a mixed method study with qualitative and quantitative components. Study participants were surveyed, using a series of open ended questions designed to solicit information about the reasons why ALS patients attend a MDC, frequency of attendance, reasons for continued treatment at a multidisciplinary clinic, and under what circumstances a person not attending a clinic would consider receiving care at this type of center. Participants were asked objective questions concerning those who influenced their decisions to attend a clinic, those who provided knowledge of these services, how far they traveled to the clinic, and what additional services, referrals, and treatments were offered by attending an ALS clinic.

Group assignment. Question number one, How often have you attended a multidisciplinary clinic (MDC) provides six possible responses: 1) Never, and I have no plans to attend a MDC; 2) Never, but I have a scheduled appointment or plan to attend in the upcoming future; 3) One time for a single diagnostic visit, but I have no plans to attend again; 4) One time, and I have a scheduled appointment or plan to attend in the upcoming future; 5) More than 1 time, and I plan to continue my treatment at a MDC; 6) More than 1 time, and I do not plan to continue my treatment at a MDC. Participants were allowed to check only one box. Checking box numbers 1, 3, and 6 placed the respondent in the non-attendee group, and box number 2, 4, and 5 placed the participant in the attendee group.

Quantitative design. The quantitative component of the study asked participants to complete the ALS Specific Quality of Life Questionnaire (ALSSQOL-R), the ALS Functional Rating Scale Revised (ALSFRS-R), and a coping measure titled the Social Problem Solving Inventory Revised Short version. (SPSI-R: S). Participants were asked to provide demographic information of age, gender, onset, diagnosis information, country, state, and zip code. The study was conducted in the form of an anonymous, on-line, web based internet survey.

Sources of distress for people diagnosed with ALS include anticipation of suffering, treatment regimens, difficulty coping with life changes, and adjusting to inherent uncertainty and uncontrollability of the illness. Patients with ALS suffer from moderate to severe psychological distress and face substantial difficulties in coping with the illness. Differences in problem solving contribute to how patients perceive and control ALS, to feelings of hopelessness, and the ability to resolve day in and day out problems; all of these are impacted by individual ability to solve stressful problems.

Social problem solving is defined by D'Zurilla & Maydeu-Olivares (1995) as a self-directed, cognitive behavioral process in which a person identifies adaptive ways to address stressful everyday life situations. Ineffective problem-solving has been shown to be a significant mediator of stressful life experiences, and contributes to how people view the world and individual circumstances (D'Zurilla & Maydeu-Olivares, 1995) Individuals with ALS who are unable to problem solve effectively may be vulnerable to feelings of hopelessness, decreased ability to cope, and decreasing QOL.

Qualitative design. A qualitative research approach was chosen in order to understand and examine reasons, beliefs, and perceptions of ALS patients who utilize an

ALS multidisciplinary clinic, and to measure quality of life through quantitative analysis. The goal of the qualitative component was to describe and interpret the unique human experiences of patients who access a multidisciplinary clinic, understand individual interpretations of health care delivery, identify perceived benefits, and determine the type of treatment sought. Participants were asked open ended question to gather data about attitudes, perspectives, behaviors, and personality. This approach is useful when the researcher needs to know how individuals understand themselves, and the world.

The quantitative approach was chosen to evaluate study participants of the clinic and non clinic population in demographics, quality of life, problem-solving abilities, and health status, and evaluate descriptive differences between the two populations. Administration of standardized measures rather than qualitative questions enabled the researchers to measure differences in the population who attend or do not attend multidisciplinary ALS clinics.

In order to determine reasons why ALS patients utilize multidisciplinary clinics, this study incorporates individuals who access multidisciplinary clinics, and those who receive care from practitioners outside of the system. The investigator's hypothesized theme for the reasons why ALS patients utilize the multidisciplinary approach was identified through a qualitative analyses based on grounded theory by Corbin & Strauss, (2008), and standards for coding qualitative data in the field.

The qualitative research approach is designed to gain knowledge and understanding of under explored aspects of human experience, focusing on individual narrative accounts, interpretation, and contextual meanings (Corbin & Strauss, 2008). Grounded theory is defined as a methodology of analysis linked with data collection that

uses a systematically applied set of methods to generate a theory (Corbin & Strauss, 2008). This approach recognizes the importance of developing an understanding of an experience, phenomenon or process that is contextual grounded by individual knowledge and life experiences. Developing increased theoretical knowledge of patient experience is valuable in constructing theory and understanding about what is occurring within a situation by exploring phenomena, gathering data related to research questions, allowing theory to emerge from data; this the process gradually builds a holistic picture of what is being explored. Narratives from the participants are analyzed until participant themes are understood.

Participants

Participants were recruited through advertisements distributed by the ALS Association, Patientslikeme.com, and ALS Centers of Excellence. The ALS Association is a national organization with affiliate networks comprising both chapters and free standing support groups throughout the United States. Local chapters are multifaceted organizations responsible for carrying out the mission of the greater ALS National Association, and provide a variety of patient care initiatives. ALS support groups are collections of individuals that meet regularly for the purpose of providing mutual support and information to ALS patients, care givers, and families. ALS Association Chapters support patient services, community services, public awareness, advocacy, and research. A Research flyer and letter announcing the research project was sent to the National ALS Association. National and state chapters distributed information to registered ALS patients, and posted the study announcement in newsletters. Local chapters and center

directors notified ALS patients who utilize support groups and were registered with the chapters.

Patientslikeme.com is a web site for individuals to share information that can improve the lives of people diagnosed with ALS. Patientslikeme.com collects patient data, and provides data sharing partnerships with physicians, research organizations, and non profits. Patientslikeme.com advertised the study via a national web site.

The ALS Association certifies and supports regional institutions recognized as having experience and knowledge with ALS. Penn State Milton S. Hershey Medical Center is a Center of Excellence, and hosted this web based survey on the internet, using Survey Monkey software. Written notification of the study provided participants with detailed information regarding the research objectives, directions in how to access the survey through the URL of the research study internet site, and a point of contact for the study coordinator about questions or concerns regarding the study.

Individuals who identified as having been diagnosed by a health care professional with ALS or MND, including upper motor neuron and lower motor neuron disease were eligible to participate. The study drew ALS patients from across the United States, including individuals attending ALS MDC, and those receiving care from outside the MDC system. Ten participants self-identified residence outside of the United States and were included in the study. Participants were male and female, ages18 or older and were able to read and write English at a 6^{th} grade level.

Inclusion and Exclusion Criteria

Inclusion criteria. Individuals participating in the study were 18 years of age or older to avoid other motor neuron diseases of genetic origin that mimic ALS. Participants

had the capacity to understand the self-report questionnaires, and were able to read and write English on a 6th grade level. Participants were males or females who had been diagnosed with amyotrophic lateral sclerosis by a healthcare professional, and were at any stage in the disease process.

Exclusion criteria. Participants under the age of 18, or those not diagnosed with ALS were excluded. Screening procedures determined inclusion and exclusion criteria

The survey was hosted on the internet in public domain. Control over the numbers of people who accessed the survey was limited, including verification of an ALS diagnoses, and those who were completing the survey. The demographics portion of the survey asked participants to identify ALS symptom onset, date of diagnosis, and site of ALS symptom onset to verify diagnosis in this open enrollment study.

A survey with incomplete measures on the ALSSQOL-R, containing missing data of more than 3 missing responses were discarded from the study analysis because a total QOL score could not be calculated.

Surveys without dates of symptom duration for ALSFRS-R to allow calculation of length of illness were not included in the analysis.

Analysis of Risk / Benefit Ratio. This study carried minimal risk to the participants. There were no reports of physical discomfort, and there were no known physical risks associated with this study. Participants may have experienced some emotional discomfort in filling out the survey because of reflection on quality of life and physical functioning. Participants were instructed at the beginning of the survey that participation was voluntary; they were able to take breaks as needed, or stop their participation in answering the questionnaire at any time. Risks were described in a brief

introductory description at the beginning of the survey. The survey took approximately sixty minutes to complete.

No direct benefit was experienced by participants in this study. Results of the research may improve future treatment approaches, of development of treatment protocols, and improve the quality of life of individuals with ALS. In particular, results of this study may lead to a better understanding of the reasons why ALS patients access multidisciplinary clinics, of perceived advantages of this approach to care, and how multidisciplinary clinics improve patient quality of life. The survey may provide insight into the reasons why individuals with ALS do not attend a multidisciplinary clinic and the factors that affect their quality of life.

Procedures for maintaining confidentiality

This research involved anonymous responses to a web based survey. The research was voluntary and the responses are anonymous. All participants had to be 18 years or older to participate in the survey. No coding system was used to identify individuals, and no identifying IP addresses were collected. The health information collected was protected by law, as explained in the participating institutions privacy notices. The research records were electronically stored and the data were kept in a password protected computer file. In the event of any publication or presentation resulting from the research, no personally identifiable information will be shared.

Recruitment

The web based survey was hosted on the internet, using the survey software,"Survey Monkey". Control over those who accessed the survey and the number of people completing the survey was limited. The link to the survey was hosted on the Penn State

Hershey Medical Center ALS clinic website, National ALS Association website, and Patientslikeme.com. When the participant completed the survey the web browser was directed to the homepage of the Penn State MSHMC ALS Clinic (http://www.alsphiladelphia .org/pennstatehershey/).

Plan for Informed Consent Procedures

Research with human subjects requires prior consent. In order to assure that potential study participants fully understood what their participation entailed, informed consent was obtained from each individual involved in the study. As standard in informed consent procedures, participants were fully informed about the procedures, and benefits and risks involved in the proposed research. The main risk in the study was psychological because participants were asked personal questions about physical functioning and quality of life, questions routinely asked by health care professionals. Participants may have found the study pertaining to their experiences, thoughts, and feelings interesting and valuable. Health information was collected, and protected by law, as explained in the Milton S. Hershey Medical Center Privacy Notice. A copy of the privacy notice was available upon request.

Participants were required to demonstrate informed consent prior to participating in web-based survey research. Participants reviewed the explanation about the purpose of the study, procedures to be followed, and explanation of the risks and benefits of participating in the study. Individuals were asked to click on a button providing consent to participate. Thus, by performing the specific action of clicking on the “continue” button the participant clearly indicated that they had fully read information about the study. Participants were given access to Milton S. Hershey Medical Center Privacy

Notice, and they provided informed consent prior to viewing or completing any questions in the study. Taking part in this research was voluntary; participants were informed of their right to stop the survey at any time, and did not have to answer any questions for any other reason.

Measures

Instruments

ALS Specific Quality of Life Questionnaire-Revised

ALS-specific Quality of Life Questionnaire-Revised (ALSSQOL-R) is a 46 item questionnaire using a 0-10 point Likert scale, with 0 being the least desirable situation, and 10 the most desirable. The instrument contains 6 factors: (1) Negative Emotion, (2) Interaction with People and the Environment, (3) Intimacy, (4) Religion, (5) physical symptoms, and (6) bulbar function. Completion time averages 15 minutes. The ALSSQOL-R demonstrates concurrent, convergent, and discriminate validity for the overall instrument, and convergent validity for the subscales (Simmons, 2006).

ALS Functional Rating Scale-Revised

The ALS Functional Rating Scale-Revised (ALSFRS-R) is a ALS disease specific measure of global functioning (Montes et al., 2006) The rating scale is easily administered by the patient; it predicts survival time, and has been shown to be reproducible when administered by an evaluator. The 12 item questionnaire provides a score from worst function O to normal function 4 for each item. Total scores range from 0 to 48. Montes (2006) found the scale to be reliable when administered by an evaluator or patient. The ALSFRS-R questionnaire has shown good reliability and sensitivity to change over time. The ALSFRS-R predicts survival time, and correlates measures of

function and strength. Test results are reproducible when administered by the patient or evaluator and has been utilized in previous clinical trials (Montes et al., 2006). Kaufman and colleges found the ALSFRS-R to have exceptional intra rater and inter rater reliability when used as a primary outcome measure in a multi-center ALS trial. Scores are valid when compared with patient self ratings, and the instrument is sensitive to change. The self-administered version was used for this study.

Social Problem-Solving Inventory – Revised Short

The Social Problem-Solving Inventory – Revised Short version (SPSI-R: S) is a 25 item, self-report measure of social problem solving skills, and evaluates individual ability to resolve problems in everyday life (D'Zurilla, Nezu, & Mayeu-Olivares, in press). Completion time averages ten minutes. The measure asks subjects to report typical responses to current problems in general on a 5 point Likert scale, ranging from "not at all true of me" to "extremely true of me". The measure is based on a five dimensional model of problem solving. The inventory has a total of five scales; two scales are constructive, and three are dysfunctional dimensions of problem solving. Constructive dimensions are Positive Problem Orientation (PPO) and Rational Problem Solving (RPS). The PPO scale relates to an individual's constructive and problem solving cognitive set, and the RPS scale evaluates the rational and systematic application of effective problem solving strategies and performance. Dysfunctional dimensions are Negative Problem Orientation (NPO), Impulsivity/Careless Style (ICS), and Avoidance Style (AS). The NPO scale measures a dysfunctional or inhibitive cognitive emotional set. The ICS scale evaluates a deficient problem solving pattern characterized by active attempts to apply problem solving strategies and techniques that are impulsive and incomplete. The AS

scale assesses defective problem solving patterns characterized by procrastination, passivity or inaction. Each dimension converts a raw score to a standard score with a mean of 100 and a standard deviation of 15. Higher scores on the two adaptive scales, PPO and RPS, indicate potentially greater constructive or effective problem solving skill, and psychological well being. Higher scores on the three dysfunctional scores NPO, ICS, AS indicate potentially defective problem solving.

Procedure

Advertisement and recruitment of participants for the on-line, web-based survey was conducted through collaboration with the ALS Association, their affiliates, Patientslikeme.com, and Penn State Milton S. Hershey Medical Center. The ALS Association is a national organization with affiliate networks composed both of chapters and free standing support groups throughout the United States. A chapter is a multifaceted organization responsible for carrying out the mission of the ALS National Association goals and objectives and provides a variety of patient care initiatives. An ALS support group is a collection of people that meet for the purpose of providing mutual support and information to people with ALS, their care givers, and families.

Penn State Milton S. Hershey Medical Center provided a secure on-line server for the research, collected the on-line survey results, and stored data in a secure area at HMC. Through the various contacts of HMC with ALS society, ALS patients, and Center Directors, flyers and letters announcing the research were disseminated. HMC advertised the research through their website, and newsletter.

A Research flyer and letter announcing the research project was sent to the National ALS Association. These letters and information flyers were distributed to local

chapters. The Center Directors disseminated the flyers and letters directly to individuals registered with the chapters, to patients diagnosed with ALS, and to leaders of support groups throughout the United States. The Chapters and executive directors notified patients registered with chapters and asked support group leaders to notify individuals in group support.

Written notification of the study was provided in the form of a letter and study flyer. Information provided detailed the research objectives, directions in how to access the survey through the URL of the research study internet site, and a point of contact for the study coordinator concerning questions or concerns regarding the study. The announcement of the study was posted by website and newsletter through the Greater Philadelphia Chapter of the ALS Association, the Western Pennsylvania, and West Virginia Chapter of the ALS Association, and the Penn State Milton S. Hershey ALS Clinic.

The announcement provided participants with specific instructions on how to access the website, information on goals and objectives of study, as well as risks and benefits. Participants reviewed consent procedures, and clicked the button marked "continue" to indicate informed consent. Participants accessed the site providing informed consent, and began to answer the questionnaire. After completion of the survey, participants were instructed to click the "finish" button to indicate completion of the survey and allow the transmission of results to a secure server at Hershey Medical Center. The web browser redirected participants to the home page of Penn State MSHMC ALS clinic after completion of the study.

Chapter 5

Results

A total of 403 ALS patients started the on line survey from April 2009 to January 2010. Individuals self- identified as attendees of a MDC (N=240), or non attendees (N=89). Participants who did not answer the first question (N=74) were excluded as missing data. Question one, "How often have you attended a multidisciplinary clinic?" differentiates attendees and non attendees; thus, these respondents were unable to be classified either as attendees or non attendees, and were not included in the remainder of the analyses (see Table 1).

Descriptive statistics were used to examine the demographic characteristics of individuals who attend multidisciplinary ALS clinics, and those who do not attend such clinics, in order to look for descriptive differences between the two. Due to missing data on some measures, reported N values may vary between analyses.

Of the 233 attendees who reported gender, 62.7% (N=146) were male and 37.3% (N=87) female. Of the 84 non attendees who reported gender, 60.7% (N=51) were male and 39.3% (N=33) female. Differences between groups by gender were not significant (Phi Coefficient = -.018, p = .752). Male to female ratio of ALS patients is estimated at 1.3 to 1.6:1, thus the present sample was representative of the general population in terms of gender (Valdmanis & Rouleau, 2008). Mean age for both groups was approximately 58 years (nonattenders, N=83, X = 58.6, SD=10.01; attenders, N=235, X=57.6, SD=10.05), with no significant differences between groups based on age (t = .763 (144.36), p. = .447). ANOVA results showed a significant difference in mean symptom

duration between groups (attendees, N=233, X=59.8, SD=62.1; non attendees N=83, X=80.9, SD=84.4; F = 5.769, p. = 0.017).

Table 1

Categories	Frequency	Percent
Attendees		
Never, but I have a scheduled appointment or plan to attend in the upcoming future	18	4.5
One time, and I have a scheduled appointment or plan to attend in the upcoming future	20	5.0
More than 1 time, and I plan to continue my treatment at a MDC	202	50.1
Non Attendees		
Never, and I have no plans to attend a MDC	41	10.2
One time for a single diagnostic visit, but I have no plans to attend again	15	3.7
More than one time, and I do not plan to continue my treatment at a MDC	33	8.2
Did not answer question "missing"	74	18.4

Presented in Table 2, participants were similar in regard to site of ALS symptom onset: 1) limb, 2) bulbar, and 3) breathing (Phi Coefficient = .085, p = .319). No differences were found in regard to sporadic and familial ALS (Phi Coefficient = .024, p = .664). Most cases of ALS are sporadic; and five to ten percent are familial (Valdmanis & Rouleau, 2008). Participants who responded yes to, "Do you have a family member with ALS?" were representative of the general population in regard to sporadic or familial ALS.

Table 2

Site of ALS Symptom Onset - Attenders

Categories	Frequency	Valid Percent
Limb	174	74.4
Bulbar	59	25.2
Breathing	1	.4

Site of ALS Symptom Onset – Non Attenders

Categories	Frequency	Valid Percent
Limb	69	82.1
Bulbar	15	17.9
Breathing	0	0.0

Do you have other family members with ALS? - Attendees

Categories	Frequency	Valid Percent
Yes	16	6.9
No	216	93.1

Do you have other family members with ALS? – Non Attendees

Categories	Frequency	Valid Percent
Yes	7	8.3
No	77	91.7

Individuals who attend a MDC reported higher utilization of the following medications and assistance: 1) antidepressants, 2) medication to reduce cramping/spasticity, 3) sleeping medications, and 4) augmentative communication devices.

Non attendees reported higher utilization in the following areas: 1) medications to reduce saliva production, 2) pain medication, 3) treatments for constipation, 4) Percutaneous Endoscopic Gastronomy (PEG) or feeding tube, 5) tracheostomy or mechanical ventilation, and 6) power wheel chair. Non attendees also reported higher incident of receiving no treatment.

Attendees and non attendees report similar usage of treatment for 1) urinary urgency, 2) NIPPV or BiPAP, 3) complimentary or alternative medicine, and 4) “other” treatments (see Table 3).

Table 3

What treatments do you use for your ALS?
Attendees and Non Attendees

Categories	Attendees N, Percent (%)	Non attendees N, Percent (%)	P-value
Riluzole (Rilutek)	N=138, (57.5)	N=36, (40.4)	P=0.006*
Antidepressants	N=104, (43.3)	N=31, (34.8)	P=0.164
Medications to reduce saliva production	N=43, (17.9)	N=19, (21.3)	P=0.480
Medications to reduce cramping/spasticity	N=79, (32.9)	N=25, (28.1)	P=0.403
Pain medications	N=44, (18.3)	N=31, (34.8)	P=0.002*
Sleeping medications	N=75, (31.3)	N=25, (28.1)	P=0.580
Treatments for constipation	N=60, (25.0)	N=26, (29.2)	P=0.440
Treatments for urinary urgency	N=27, (11.3)	N=10, (11.2)	P=0.997
Percutaneous Endoscopic Gastronomy (PEG) or feeding tube	N=43, (17.9)	N=20, (22.5)	P=0.351
NIPPV or BiPAP	N=69, (28.8)	N=26, (29.2)	P=0.934
Tracheostomy or Mechanical ventilation	N=9, (3.8)	N=8, (9.0)	P=0.057
Augmentative communication device	N=61, (25.4)	N=15, (16.9)	P=0.102
Power Wheel Chair	N=106, (44.2)	N=47, (52.8)	P=0.163
Experimental medications	N=35, (14.6)	N=10, (11.2)	P=0.432

No treatment	N=10, (4.2)	N=9, (10.1)	P=0.040
Complementary and Alternative medicine (includes supplements, massage, holistic medicine, chiropractic care, acupuncture, etc)	N=70, (29.2)	N=26, (29.2)	P=0.993
Other (please specify)	N=192, (80.0)	N=71, (79.8)	P=0.483

Note. * = $p < .05$.

Individuals who attended a MDC reported higher utilization of health care professionals: 1) neurologist, 2), nurse, 3) respiratory therapist, 4) physical therapist, 5) occupational therapist, 6) mental health professional, 7) social worker, 8) dietician, and 9) speech therapist.

Individuals who do not attend a MDC report higher levels of health care utilization: 1) pastoral care chaplain, 2) complementary and alternative medicine, 3) hospice services and 4) experimental medications.

Clinic and non clinic attendees report similar utilization of: 1) pulmonologist, and 2) in home care (see Table 4).

Table 4

Response for Attendees and Non Attendees

Do you use or have your been referred to any of these health care providers as part of your care for ALS? Check all that apply

Categories	Attendees N, Percent (%)	Non attendees N, Percent (%)	P-value
Neurologist	N=196, (81.7)	N=64, (71.9)	P=0.053
Nurse	N=105, (43.8)	N=19, (21.3)	P=0.001*
Respiratory Therapist	N=118, (49.2)	N=25, (28.1)	P=0.001*
Pulmonologist	N=96, (40.0)	N=36, (40.4)	P=0.941
Physical Therapist	N=177, (73.8)	N=42, (47.2)	P=0.001
Occupational Therapist	N=144, (60.0)	N=37, (41.6)	P=0.003*
Mental Health Professional	N=53, (22.1)	N=15, (16.9)	P=0.298
Social Worker	N=109, (45.4)	N=25, (28.1)	P=0.004*
Pastoral Care or Chaplain	N=34, (14.2)	N=17, (19.1)	P=0.272
Dietician	N=107, (44.6)	N=14, (15.7)	P=0.00*
Speech Therapist	N=126, (52.5)	N=34, (38.2)	P=0.021
Complimentary and alternative medicine (Chiropractors, Naturopathic Doctors, Acupuncturists, etc.)	N=36, (15.0)	N=17, (19.1)	P=0.369
In Home Care	N=68, (28.3)	N=25, (28.1)	P=0.965
Hospice Service	N=15, (6.3)	N=15, (16.9)	P=0.003*
Experimental Medications	N=35, (14.6)	N=10, (11.2)	P=0.432

Note. * = $p < .05$.

In order to compare group means, Multivariate Analysis Of Variance (MANOVA) was conducted on the primary outcome measures of total ALSSQOL-R scores, total ALSFRS-R scores, and total SPSI-R scores between ALS patients who attend multidisciplinary clinics and those who do not attend clinics. A priori power analysis estimates a required sample size of 210 participants for one-way MANOVA with an effect size of .25 and a significance level of $\alpha = .05$.

Analyses were conducted with varying numbers of participants who meet inclusion criteria. ANOVA results showed a significant difference in overall quality of life in individuals who attend a MDC compared with non attendees, based on the total ALSQOL-R average score. ANOVA results also showed significant differences in participants who attend versus participants who do not attend a MDC on the Intimacy, Physical Function, and Bulbar Function subscales of the ALSQOL-R. ANOVA results revealed no significant difference between groups with regard to Negative Emotion, Interaction with people and the Environment, and Religiosity subscales (see table 5).

Table 5

ALSSQOL-R
Attenders and Non Attenders

Category	Attenders Mean, (SD), range	Non attenders Mean, (SD), range	P-value
ALSSQOL-R Average Total	N=229, X=6.4 SD=1.6, 0.8-9.6	N=81, X=5.7 SD=1.7, 0.2-9.2	P=0.004*
Negative Emotion	N=220, X=6.3 SD=2.0, 1.1-10.0	N=73, X=5.8 SD=1.9, 1.2-9.4	P=0.062
Interaction with people and the Environment	N=221, X=7.8 SD=1.7, 2.3-10.0	N=72, X=7.4 SD=1.5, 4.2-10.0	P=0.108
Intimacy	N=210, X=5.9 SD=2.0, 1.1-10.0	N=67, X=5.3 SD=2.1, 0.4-9.9	P=0.021*
Religiosity	N=223, X=5.4 SD=3.7, 0.0-10.0	N=75, X=6.3 SD=3.6, 0.0-10.0	P=0.069
Physical Function	N=224, X=6.2 SD=2.0, 0.0-9.8	N=78, X=5.3 SD=2.4. .3-10.0	P=0.004*
Bulbar Function	N=224, X=6.6 SD=2.6, 0.4-10.0	N=77, X=5.7 SD=2.7, 0.0-10.0	P=0.010*

*Note. * = p <.05.*

A total of 280 participants met inclusion criteria, and were included in the ALSFRS-R analysis. ANOVA results showed a significant difference between the two groups. Participants not attending a MDC reported overall better levels of physical function, as measured by the ALSFRS-R (see table 6).

Table 6

ALSFRS-R Total
Attendees and Non Attendees

Attend Status	N	Mean	Standard Deviation	F	P-value
Attendees	206	18.7	9.8	15.272	P=0.000
Non attendees	74	24.0	10.3		

Social Problem Solving Inventory – Revised: Short Form

Overall, a smaller number of participants (N = 274) completed the SPSI-R who also responded to the attend status. Attendee's and non attendee's mean scores on the SPSI-R and subscales were within average range compared with the SPSI-R normative sample, and did not differ significantly from one another (F = 1.599, p=.147).

Table 7
Social Problem Solving Inventory-R: S
Attendees and Non Attendees

Category	Attendees (N), Mean, (SD)	Non attendees (N), Mean, (SD)	P-value
PPO Scaled score	N=214, X=103.43, SD=16.08	N=69, X=102.62, SD=16.67	P=0.699
Negative Problem Orientation Scaled	N=210, X=95.31, SD=14.17	N=71, X=96.85, SD=15.65	P=0.362
Rational PS Scaled Score	N=213, X=104.44, SD=14.65	N=71, X=99.67, SD=15.71	P=0.032
ICS Scaled Score	N=214, X=97.21, SD=13.71	N=71, X=100.69, SD=14.47	P=0.069
AS Scaled	N=212, X=96.13, SD=12.90	N=70, X=97.45, SD=14.58	P=0.446
SPSIR Scaled	N=206, X=105.72, SD=12.94	N=68, X=102.08, SD=14.95	P=0.054

Qualitative Analysis

Data Coding

Participant response data were individually and collectively coded for analysis by the principal investigator and two doctoral level students in clinical psychology from Philadelphia College of Osteopathic Medicine. Free text responses were reviewed and categorized based on similarities. Individual responses identifying multiple themes were listed in multiple categories. Group processes, thoughts, and impressions of the coders were recorded by the investigator and were reported in manuscript form. Group consensus guided the process of drawing conclusions of emergent themes, providing greater validity to participant response data. A manuscript of the categories was

submitted to the larger ALS research team for comment and feedback. Cross comparisons of themes emerged in coding and were utilized to identify commonalities or differences. Subsequent responses of attendees and non attendees were differentiated and separated for qualitative analysis of respective viewpoints of treatment experiences. The purpose of the analysis was to evaluate similarities and differences of patients who attend or who do not attend a MDC.

Qualitative Results

Reasons for attending a multidisciplinary clinic

"If you do go to a multidisciplinary clinic, please tell us why you go. Please give us as much information and detail as you can."

228 participant responses were coded; 6 responses were excluded as "N/A" or reported as "non attending", (N=222). Convenience / travel / time was the predominant response (N=73, 32.9%), validating the fact that access to care standards influence quality of life. One participant commented on aspects of a MDC, and effect on his or her quality of life.

> "I have one appointment once every three months and all my health issues are addressed. If you will, it is one stop shopping rather than a multitude of appointments that wears greatly on me and my family. Also with full focus and expertise in the field they are able to help my issues from pain to frustration with lost abilities. The clinic also deals with other health issues; at my last appointment I had broken my arm and two vertebrae from a fall. Even though I had seen a doctor for the fractures they recommended and scheduled appointments for other procedures on my back. I decided to follow through with the recommended procedures which indeed helped with the pain I was dealing with. The ability to have all aspects of my changing conditions reviewed at one time and modified medical care immediately recommended regardless of the medical specialty required greatly simplifies my life. Bottom line they all care, share their individual observations at a team meeting and immediately recommend how to improve the quality of my life."

Specialist (N=70, 31.5%) was the second highest response given as a reason to attend a MDC. Integrative care (N=49, 22.1%) was the third highest.

> "The neurologist is a specialist in ALS. Appropriate PT, OT, RT, social services, etc., are provided by people with experience and competence in ALS. References for equipment are provided, including loan and rental equipment. The PT brought in the AFO vender and the company that fitted me for a power chair. There is outstanding knowledge of what Medicare and supplemental insurance will cover. There is prompt care and genuine concern and personal knowledge of me as a patient."

> "I go because I am given the attention and care from each specialist. I am never rushed it's a one on one time with them. They also make recommendations and suggestions and make further appointments for various things as I need them, like physical therapy, sleep study, lung evaluations, etc. If I need any supplies they are brought to me at my home from their loan closet. I have gotten a cane, a walker, a ream for the front door, a text board to help with my speech difficulties, a shower stool. All of this at no cost to me, if I need anything or any advice I have a registered nurse who comes to the house if need be. I feel that I matter as a person. I have been to two multi-disciplinary clinics and will go every three months. I think it's important too, that each person I see meets with the others to go over my case afterwards."

Participants found gaining information and knowledge (N=40, 18%), and evaluate progression (N=26, 11.7%) valuable facets of a MDC.

> "MDC was set up by the Mayo Clinic, at the Mayo Clinic. At each Clinic we see the specialists that can help us deal with each of our problems. For instance, at the latest clinic I saw the neurologist who did another evaluation of my progression. We saw a speech therapist, who evaluated my ability to speak. We met with a Physical therapist who measured me for a wheelchair. And we met with a Social Worker who set us up for attending Support Groups and connecting with people who could assist us in getting the equipment I will need shortly."

> "They provide most recent information available. Very helpful in giving suggestions to help me with daily functioning and to be able to maintain as much independence as this disease will allow. Helps me to keep track of all new research and trials that might be available for me to participate if I am eligible."

The final categories include 1) support and trust, 2) research and clinical trials, 3) cost and insurance coverage, 4) equipment, 5) diagnosis confirmation, and 6) expertise (see Table 9).

Table 9

If you do go to a Multidisciplinary Clinic, please tell us why you go. Please give us as much information and detail as you can.

Categories	Number of Responses	Percent
Convenience / Travel / Time	73	32.9
Specialist	70	31.5
Integrative care	49	22.1
Gaining information and knowledge	40	18.0
Evaluate progression	26	11.7
Support and trust	26	11.7
Research and clinical trials	22	9.9
Referrals	14	6.3
Cost and insurance coverage	7	3.2
Equipment	5	2.3
Diagnosis confirmation	3	1.4
Expertise	2	0.9

Advantages and disadvantages of attending a multidisciplinary clinic

"*Please tell us about any advantages or disadvantages from receiving care from a multidisciplinary clinic. Give as much detail and information as you can.*"

A total of 215 participants responded to this open ended question; 25 responses were excluded from the coding as incomplete. A total of 190 completed responses identified 244 advantages, and 101 disadvantages of receiving care at a MDC. Coders first stratified the two groups of responses as advantages and disadvantages. Responses in each group were coded and placed in a category.

Participant responses for MDC advantages were coded and placed in seven categories. The primary category convenience, integrated care / travel time (N=94) illustrates the importance of multidisciplinary care received at a MDC.

> "The team approach gives me a sense of quality care that I feel I could not get from a single doctor alone. ALS affects so many of the body's muscles and systems that having the team meet with me enhances my quality of life. The specialists know things that can help me to manage my care and make things easier for me."

> 'The facility is downtown. I live out in the suburbs so commuting to the office is perhaps not quite as convenient as it was being seen by a local neurologist, but I don't really feel like there's any comparison in terms of the quality of the care and its sophistication that I received. Again the advantages are that it provides one stop shopping. You are able to see the whole range of people providing care to you in one outing. I also think it's an advantage that they are all working at one centers so they can coordinate their activities in the care that they provide to me and the attention that they give to my wife."

Remaining categories of: 1) resources and information, 2 clinical expertise, 3) general support, 4) monitor progression, 5) treatment research and clinical trial, and 6) social support were recognized as advantages associated with MDC attendance (see Table 10).

> "They help you understand what's going on and what you can expect. Help get the correct (wheel chairs, walkers) things in general to make it easier to go on. Their advice both on the phone and over the internet take care of most any question. As this is really completely

> unknown to me and my wife they have been more help than they could ever believe. Even the smallest question is always taken care of."
>
> "The advantages are that all the medical professionals are at one location. They know more about the disease than any other doctor that I had been to."
>
> "My wife and I cannot imagine this disease without the experts in all divisions: Speech & swallowing; PT, OT, emotional therapy, sleep experts etc. plus at BID we have noon lunch with fellow patients in a social setting."

Participant responses were coded into nine categories related to disadvantages of attending a MDC. Two primary categories emerged, viz., travel distance (N=29), and a long exhausting day (N=24),

> "It's exhausting for the patient! Especially if you have to travel about 400 miles in a day."
>
> "I did not like the wait time and that it took almost a whole day to be seen by several practitioners. I found it fatigues me to be in this environment almost a whole day."
>
> "Went to MDC from time of diagnosis till January of this year when had surgery for feeding tube and vent. Getting to clinic is too difficult now, and the only time we go out is for medical necessity or major family event. If going out were easier, we would still go to the clinic."

Participants identified rigid and generic (N=15) wait time (N=14), schedule (N=7), limits of services and treatment (N=5), do not like doctor (N=4), unnecessary services (N=2), and environment (N=1) as disadvantages to attending a MDC (see Table 10)

> "There is a rote protocol for all patients without regard to individual variations which are many."

Table 10

Please tell us about any advantages or disadvantages from receiving care from a Multidisciplinary Clinic. Give us as much detail and information as you can.

Categories	Number of Responses
Advantages	
Convenience (integrated care / travel time)	94
Resources and information	44
Clinical expertise	43
General support/support from other patients	32
Monitor progression	15
Treatment research and clinical trials	11
Social support	5
Disadvantages	
Travel distance	29
Long exhausting day	24
Rigid and generic	15
Time / wait time	14
Schedule	7
Limits of services and treatment	5
Do not like doctor at MDC	4
Unnecessary services	2
Environment	1

Rationale for not attending a multidisciplinary clinic

"If you do not go to a multidisciplinary clinic please tell us why. Please give us as much information as you can."

100 individuals responded; 25 responded with "N/A" or incomplete response and were excluded from the coding. Travel (N=19, 25.3%) was recognized as the primary category for not attending a MDC

> "I don't go to a Multidisciplinary Clinic because it's in San Francisco and that's just too difficult for me to maneuver physically. It's tiring to drive in the commute even with my special van and motorized wheelchair. Very little parking for handicapped vans with ramps. Regular cars with placards park in them too and that almost always leaves me searching for a space. I just can't handle it."

A number of participant responses were coded in uninformed, not referred (N=13, 17.3%), and were unaware of the existence of a MDC.

> "I have not been spoken to about any potential benefits of going and I would be interested to see what a clinic visit could do for me."

Remaining participant responses were coded in the following categories: 1) felt like a lab rat, 2) depressing, no cure, 3) local doctor nearby, 4) didn't like doctor, 5) tiring / difficult, 6) none in area, 7) length of clinic time / poor clinic organization, 8) attend VA, 9) no need to attend, 10) cost and insurance, and 11) sub theme, utilizing alternative medications (see Table 11).

Table 11

If you do not go to a Multidisciplinary Clinic please tell us why. Please give us as much information as you can.

Categories	Number of Responses	Percent
Travel	19	25.3
Uninformed, not referred	13	17.3
Felt like a lab rat, depressing, no cure	9	12.0
Local doctor nearby	8	10.7
Didn't like doctor	8	10.7
Tiring / difficult	7	9.3
None in area	6	8.0
Length of clinic time / poor clinic organization	6	8.0
Attend VA	5	6.7
No need to attend	5	6.7
Cost and insurance	4	5.3
Sub theme, utilizing alternative medications	2	2.7

Circumstances influencing attendance at a multidisciplinary clinic

"If you do not attend a multidisciplinary clinic, under what circumstances might you attend an ALS clinic? Please give us as much information as you can."

A total of 87 participants responded; 21 responded with "N/A" or an incomplete response and were excluded from the coding (N=66). Nine categories were identified

from participant responses as circumstances influencing attendance at an MDC. However (N=6) participants indicated they would not consider attending a MDC.

The primary category was travel, closer to home, access (N=36, 54.5%). One participant response suggested willingness to attend a MDC if it were closer to home.

> If there was a clinic in my area I would attend regularly. But I live in a rural area and find the drive too tiring. My twice a year trip to my neurologist and trips to my respirologist, when the appointments can't be made for the same day, are all I can manage."
>
> "Money is the biggest factor, plus is hard to travel with all the equipment, such as pwr wheelchair, voice amp and BIpap unit. Plus I think the closest clinic is 400 miles away."

Other responses suggest participants were unaware or lacked information concerning the benefits of a MDC and were coded in category, understood function (N=9, 13.6).

> "Well, I would want to learn about it first. I am certainly open to the idea."
>
> "Would like to have information and an invitation."
>
> "When the time comes that I need help breathing or using my limbs. I have limited use of my left arm, fingers and left leg but still get around, slowly, without the use of a wheelchair. ALS of Cleveland and MDA of Toledo are both ready when I am."

Participants (N=7) indicated the potential of attending a clinic if physical health declined, considering that MDC has specialized treatments and providers. "When I start having difficulty breathing, talking, swallowing, etc., I will probably discuss the feasibility of attending the clinic with my neurologist," and "I might attend a Multidisciplinary clinic for my breathing and speech problems."

The remaining participant responses were coded in the following categories: 1) cure, 2) cost, 3) invitation, referral from a health care provider, 4) socialization, 5) trust doctor, and 6) less wait time (see Table 12).

Table 12

If you do not attend a multidisciplinary clinic, under what circumstances might you attend an ALS clinic? Please give us as much information as you can.

Categories	Number of Responses	Percent
Travel, closer to home, access	36	54.5
Understood function of MDC	9	13.6
Health declined and need specialized treatment	7	10.6
No chance I'll go, no hope	6	9.1
Cure	5	7.6
Cost	4	6.1
Invitation, referral from a health care provider	3	4.5
Socialization	3	4.5
Trust doctor	1	1.5
Less wait time	1	1.5

Travel and distance considerations

"How does travel distance to a multidisciplinary clinic or other health care professional influence your treatment decisions? Please give as much information as you can."

239 individuals responded; 10 responded with "N/A" or an incomplete response and were excluded from the coding (N=229). The majority of participant responses indicated that travel distance is not a factor in treatment decisions (N=144, 62.9%), In fact, one participant drives 225 miles to attend a MDC.

> "No I was told they were the best place so the distance of 225 miles each way didn't bother me a bit."

> "Going to a multiple disciplinary clinic is hard on someone in a wheelchair. There is no way that I could get the care that I get in Columbia South Carolina. The MDA sponsored clinic in Charlotte, North Carolina an unquestionably get better care to ALS patients."

> "I don't mind at all traveling this distance to the clinic because all personnel are very caring and empathetic. They all treat patients like their old friends with welcoming gesture. They take all patients as though their cases are most important one. Besides they have all the experts there in one place. I don't have to go different places for different appointments, especially for ALS patient who usually have problem in moving."

Participants (N=85) indicated that travel distance does influence treatment decisions. Responses were coded into six categories: 1) travel barriers: coordination, expense, and physically uncomfortable, 2) stage of disease progression make travel more difficult, 3) fatigue, 4) proximity to physician affects health care decisions, 5) frequency, and 6) time away from work (see Table 13).

> "It has everything to do with it. Traveling to San Francisco almost any time of the day is difficult and time consuming. Also it's extremely hard to park a modified can in a disabled parking spot because cars are in

them. The drive to and from is tiring, and I hate to ask someone to give up almost a whole day to go with me."

Table 13

How does travel distance to a Multidisciplinary Clinic or other health care professional influence your treatment decisions? Please give as much information as you can.

Categories	Number of Responses	Percent
Travel distance is not a factor in treatment decisions	144	62.9
Travel barriers: coordination, expense, and physically uncomfortable	48	20.9
Stage of disease progression makes travel more difficult	13	5.7
Fatigue	10	4.3
Proximity to physician affects health care decisions	10	4.3
Frequency	3	1.3
Time away from work	1	0.4

Chapter 6: Discussion

Summary of Findings

The national study of Amyotrophic Lateral Sclerosis multidisciplinary utilization was designed to investigate the characteristics, differences, and commonalities of patients with Amyotrophic Lateral Sclerosis attending multidisciplinary clinics with individuals not attending MDCs. Describing the reasons why individuals seek and attend treatment in an ALS MDC assists practitioners providing care to individuals with ALS. The primary rationale for conducting this study was to examine the differences in quality of life, states of health, and coping skills of patients with ALS attending MDCs compared with non attendees, and to describe specific service and treatment differences. Qualitative components of the study offer unique data to explore the reasons why ALS patients choose to attend or not attend MDCs, the advantages and disadvantages to attending, circumstances influencing attendance, and how travel distance impacts medical decision making. The study produced expected and significant findings.

The majority of the participants of this study were from the United States, ten individuals self-identified living outside of the United States, and were included in the sample. The sample was composed of individuals affiliated with the ALS Association, ALS affiliates, and Patientslikeme.com, self- identified as having ALS, and over the age of 18.

The two groups studied were similar by gender and age. Descriptive statistics did not find any differences between groups regarding ALS symptom onset, and sporadic and familial ALS. A significant difference was found between mean symptom duration

between groups. Individuals attending a clinic report longer symptom duration than individuals not attending a clinic.

Hypotheses (H)

Three of the four sets of hypotheses in this study were supported with significant findings. The four hypotheses were: 1) participants attending a MDC will have better coping skills, as measured by Social Problem Solving Inventory-R; 2) participants attending a MDC will have higher quality of life as measured by ALSSQOL-R; 3) participants attending a MDC will have higher levels of physical functioning, and 4) comparison of attendees and non attendees' overall utilization of treatment and professional services will demonstrate that a MDC provides individuals greater access to care.

Hypothesis (H1) theorized that participants attending an ALS MDC would have better problem solving skills than individuals who do not attend. Based on a previous study, social problem solving skills were shown to affect ALS patients with negative emotion, and predict QOL in ALS caregivers (Murphy, Felgoise, Walsh, & Simmons, 2009). Therefore, this study compared social problem solving skills between attendees and non attendees of a MDCs. It was theorized that participants attending MDCs may do so to obtain additional services and treatments influencing QOL.

H1 was not supported by this study. Problem solving skills did not differ between attendees and non attendees. Mean scores on the SPSI-R and subscales were within average range compared with the normative sample, and did not differ significantly between groups. Thus, both groups were similar and within a normative sample for problem solving and coping. Therefore, it does not appear that problem solving and

coping skills contribute to ALS patient quality of life or influence medical decision making.

Hypothosis (H2) stated that participants attending ALS MDCs would have overall better QOL than non attendees. Previous research compared QOL of ALS patients attending and non attending, and found no significant differences in the quality of life reports measured by ALSSQOL-R by individuals with ALS attending MDC ALS clinics, compared with non attendees (Stephens, Walsh, & Simmons, 2008).

Findings indicate that H2 was supported. This study found that individuals attending a MDC have better overall QOL than non attendees. This finding is significant when compared with previous research in which a difference was not found. This supports the fact that clinical expertise, and other resources provided by a MDC may serve to modify patient perceptions to the point where physical limitations are seen as having less negative impacts. Thus, a MDC may provide certain factors impacting QOL in this specific time and population. Establishing a causal relationship is difficult in a cross sectional study with an aim to describe the differences between QOL between groups without regard for what preceded or precipitated the health status found when the participant completed the survey. Thus, a cross sectional study is not able to establish a cause and effect relationship between attendees and non attendees, only the existence of different health related states.

Hypothesis (H3) indicates that participants attending ALS clinics will have a higher level of physical function than non attendees. Traditionally, MDCs provide patients greater access to services and treatment practitioners. In a previous study by

Penn State Milton S. Hershey Medical Center, disease duration and physical function did not differ between the two groups.

Findings indicate that H3 was supported. Findings reveal that individuals attending MDCs perceive QOL as being higher with respect to physical functioning and bulbar function than non attendees. However these individuals reported poorer overall levels of physical function as measured by the ALSFRS-R, with significantly shorter disease duration. It is possible that participants who attend a MDC are evaluated for pulmonary function and physical function, and are more aware of disease progression, compared with those who are not routinely evaluated by numerous specialist and health care providers. Thus, participants who attend a MDC may rate physical function lower as a result of increased contact with medical providers. Treatments received at MDCs may serve to modify perceptions of health, and serve to modify or alter health beliefs.

Hypothesis (H4) stated that participant attending MDCs would receive more treatments and services for maintaining QOL compared with non attendees. Findings indicate that H4 was partially supported. Differences are apparent between the two groups. Individuals attending MDCs report higher utilization in 1) antidepressants, 2) medication to reduce cramping/spasticity, 3) sleeping medications, and 4) augmentative communication devices.

Non attendees report higher utilization of: 1) medications to reduce saliva production, 2) pain medication, 3) treatments for constipation, 4) Percutaneous Endoscopic Gastronomy (PEG) or feeding tube, 5) tracheostomy or mechanical ventilation, and 6) power wheel chair. Significant amount of services are utilized by individuals outside the MDC. These services seem to reflect later stage disease

progression: PEG or feeding tube, mechanical ventilations, and power wheel chair. Individuals may not be able to access a MDC due to health and other supports, and choose not to attend. Qualitative responses support consideration that disease progression is a factor in treatment decisions. One participant described attending a MDC for many years; regrettably, his health declined, forcing him to change treatment from a MDC to an individual practitioner.

Attendees and non attendees report similar usage of treatment for 1) treatments for urinary urgency, 2) NIPPV or BiPAP, and "other".

Individuals who do not attend a MDC report higher incidence of receiving no care or treatment for ALS. This could be a contributing factor to higher utilization of hospice care and alternative medicine. Patients and families may elect not to participate in MDC for varied reasons, or choose not to receive any care.

To be expected, individuals attending MDCs report higher utilization of health care professionals: 1) neurologist, 2), nurse, 3) respiratory therapist, 5) physical therapist, 6) occupational therapist, 7) mental health professional, 8) social worker, 10) dietician, and 11) speech therapist. MDCs are developed to provide access to these services, and are accomplishing the goal. Additional supports and treatments may have a value and influence in quality of life.

Non attendees of MDCs report higher levels of health care utilization: 1) pastoral care chaplain, 2) complementary and alternative medicine, and 3) hospice services. The use of pulmonologist and in-home care was similar for both groups.

Non attendees of MDCs report greater utilization of community or pastoral care, and are searching outside of traditional medicine to receive care and support. One

particular finding is the rate of utilization for hospice services for non attendees. Individuals receiving care from hospice may not be afforded the same opportunity to utilize MDCs and may perceive quality of life differently, and thus influence the results. Individuals who attend an MDC report poorer levels of physical function; conversely, individuals not attending a clinic report higher use of hospice services, a treatment utilized with increasing frequency with disease progression. Individuals attending a clinic are more clearly aware of their disease progression and are also more knowledgeable about ALS. Rating of physical function may be lower based on this factor.

Individuals who choose to receive care from a solo practitioner may perceive that treatments are more effective and efficient in a private setting versus receiving care from a MDC setting where multiple providers are trying to see the same patient in a defined span of time. Qualitative responses reveal the MDC experience is long and exhausting. Respondents did not want to meet with the entire team of specialist in an attempt to shorten the day. Non attendees of an MDC may benefit from some of the resources and supports of the clinic; therefore, developing a flexible system of care to promote communication between MDC and solo practitioners becomes increasingly important in providing optimum care for the patient who chooses not to attend a MDC.

Qualitative findings

If you go to a multidisciplinary clinic, please tell us why you go

The specific aim of the study was to examine the characteristics of patients affected by ALS who attend MDCs. This was accomplished through a qualitative question, asking study participants who attend a MDC to provide information detailing reasons for attending. Individuals indicated that they attended because of convenience,

travel, and time. This result was expected because previous research found travel and convenience a significant factor in attending a clinic. One participant wrote: “everything done at one time-doctor, PT, OT, ST. Takes a ½ day but worth it + they are all ALS specialist”. Many participants expressed the idea that combining several appointments into one day reduced stress and provided increased access to care. Many responses referenced the phrase “one stop shopping” when describing the treatment received at a MDC.

A theme emerged regarding the specific care received from a MDC. Respondents described the importance of specialist trained in ALS and integrative care, including specialized services to deal with loss and frustration. Participants described initial interaction with health care professionals who were unfamiliar with ALS, and were encouraged to be seen by a team well versed in ALS treatment. In the case of ALS, participants responded that many health care professionals outside of a MDC do not feel comfortable with patients having ALS or these professionals do not have adequate training in caring for patients with ALS. Participants expressed concern with a lack of ALS specialist, especially in rural areas, resulting in long travel times to be seen by a health care practitioner. Yet they are willing to travel for hours to be seen by a specialist trained in ALS. Participants wrote of the ease and satisfaction of one appointment, addressing multiple health issues, coordinating care, and reducing stress on family and friends.

Participants were interested in gaining information and knowledge related to disease management and evaluation of disease progression. Participants want to have information on what to expect in the future, and suggestions to help with daily

functioning. Suggestions provided from MDC were perceived as helping with daily functioning, resulting in improved independence. Participants were curious and wanted to be involved in new clinical trials and research, and perceived the fact that MDC provided greater access to these opportunities.

Participant responses coded in the area of equipment was low. Conversely, as found in table 3 and 4, individuals who attend a MDC report higher utilization of equipment and services. One possible factor for a low incidence is that respondents used the phrase "one stop shopping" and "convenience", which may broadly be defined as services that include equipment. Additionally, respondents reference the fact that they meet with practitioners such as a physical therapist, occupational therapist and speech therapist who would be involved in arranging for equipment.

Advantages or disadvantages from receiving care from a multidisciplinary clinic

The second aim of the study was to determine the specific, perceived advantages or disadvantages from receiving care from an MDC, and explaining psychological, social, and medical concerns held by ALS patients.

The primary advantages identified for attending an MDC were convenience, integrated care, and travel time. Respondents consider MDCs to provide greater access to multiple providers with greater expertise in treating ALS. The phrase "one stop shopping" continues to be a theme in how participants describe experiences at an MDC, Respondents also spoke of the advantages of receiving care from multiple providers in one day, reducing multiple appointments with various treatment providers. Participants expressed a comfort in knowing that ALS specialists were available to provide assistance to the patient and family. Patients were willing to travel longer distances to attend a MDC

due to the perceived “quality of care and its sophistication”, and coordination of care between providers.

The prevalence rate of ALS is relatively low, compared with global neurological diseases, and disease states. The annual incidence of ALS is 2/100,000 population and the prevalence is 6/100,000 (Shoesmith & Strong, 2006). Generally cases are sporadic and it is theorized patients newly diagnosed with ALS are unfamiliar with the disease, do not have any friends or family diagnosed with the disease, and are not connected to any services. Respondents depend on MDCs to supply resources and information regarding disease management as well as information concerning the disease itself. Participants described the fact that a MDC provides a central location for information to assist the patient in understanding the disease of ALS and also what to expect. Information regarding wheel chairs and walkers, and general information “makes it easier to go on.”

Participants identified clinical expertise as an advantage of attending a MDC; attending provides an opportunity to be seen by a number of health care professionals who are specialists in the treatment of ALS. Participants describe early encounters with health care professionals who are not trained in ALS, and were not able to provide a correct diagnosis or supportive care. Participants recognize the fact that MDC are better equipped to provide specific ALS interventions or remedies to alleviate discomfort.

Participants of MDC interact with other patients, providing mutual support and a sense of not being alone. One participant described lunch with fellow patients in a social setting, and interaction with others outside of his immediate support group. The support of others with a similar illness, and support from staff at the MDC assists individuals and alters patients’ perspectives.

The primary disadvantage to attending a MDC was travel distance and length of day. Participants describe traveling “400 miles in a day” to attend an MDC, and feeling exhausted before the first specialist appointment. Waiting to see a specialist, which involved extending the length of the day, was seen as a disadvantage; multiple providers are trying to see the same patient in a defined period, and this involves wait times between specialists. Participants indicated clinics follow a rote protocol; they felt rushed through multiple appointments, and were not sure if the total number of specialist seen in one day was necessary. Development of a good MDC involves cooperation, communication between patient and provider, and assistance for the patient in understanding the benefit or reasons for treatment.

A third aim of the study asked participants who do not attend a MDC to identify reasons why they choose not to attend. The primary reason that participants choose not to attend a MDC was travel. Participants indicated that a MDC was located too far from home; there was the fatigue of travel, lack of care giver support to transport patient to a MDC, exhausting preparation time to travel, and progression of the disease, making travel too difficult. The complexity of the disorder, the need for involvement of multiple disciplines and recognition of the challenges of providing health care to individuals affected by ALS has led to the development of MDC as a means to improve coordination and integration of care, and has been shown to improve quality of life. MDCs have long been advocated by the National ALS Association as the optimal way to provide care. Yet only a few clinics are available within the United States, a primary barrier for care. Convincing facilities and organizations to expand funds and support to maintain ongoing programs or to develop new MDCs is particularly challenging. New and innovative ways

to reduce travel barriers are needed. Dorsey and colleagues (2010) found that telemedicine increases access to specialty care and improves quality of life for patients with Parkinson's disease who reside in remote locations greater than 130 miles from an academic movement disorders clinic. Features of telemedicine include specialized referral services, remote patient monitoring, patient consultation, and medical and health education. Utilization of new technology such as telemedicine may be an opportunity to expand services to and coordination with individuals who are not able to access a MDC.

Participants explained that were uninformed and also they were not referred; these constitute two reasons why that they had not attended a MDC. One participant wrote: "I have never had the opportunity, nor even heard of that option." This illustrates the continued need for education and information to individuals diagnosed with ALS, and also for coordination of services.

The fourth aim of the study was to examine the characteristics of individuals who do not attend a MDC, and what circumstances would influence their decision making to attend. Participants indicated that the primary reason for electing to be seen outside of a MDC was distance. If the clinic was available locally, participants would go. Financial costs of attending a clinic were described as factors in determining treatment decisions. Participants indicated a lack of understanding or a lack of awareness about the function of a MDC; they would consider a MDC if they had additional information, or if their health declined. Overall, participants recognize the benefits of a MDC and would consider this treatment option.

The fifth aim of the study was to determine how travel distance to a MDC influences treatment decisions. Participants (N=144, 62.9%) indicated that distance did

not influence decision making, and would make the trip regardless of time and travel, perceiving that the MDC provides greater care and expertise. Travel to a MDC was a common concern to participants of the study. Respondents planned, however, to continue attending a MDC regardless of the travel for the perceived benefits the MDC provided: 1) specialist, 2) integrative care, 3) information and knowledge.

Participants who indicated that travel influenced medical decisions described four primary barriers: 1) coordination, 2) expense, 3) physical discomfort, and 4) stage of the disease process and fatigue. Individuals not attending a MDC would consider care from a MDC if it were closer; if participants better understood the function of a clinic, and if health declined. Only (N=6), a small percentage, would not consider attending a MDC. Overall, participants explained that receiving care at a MDC had improved quality of life.

Significance of the findings

In this study, patients with ALS attending a multidisciplinary clinic differ from those who do not attend. First, participants who attend a MDC report a shorter duration of the disease compared with those who do not attend. Second, participants attending a MDC identify poorer overall levels of physical function. It is theorized that increased monitoring from the MDC could influence patients' perceptions regarding physical functioning. Third, participants attending a MDC report higher overall quality of life, and higher perceived QOL in relation to physical and bulbar function. These finding are significantly different from a previous study conducted by Penn State Milton S. Hershey Medical Center; disease duration and physical function did not differ between groups, and there were no significant differences in the QOL reports by individuals with ALS who attend a MDC compared with those individuals with ALS who do not attend ALS

clinics. Supports, specialists trained in ALS care, and integrative care may serve as factors changing patient perspectives, and altering quality of life. Participants attending MDCs report receiving additional treatments and higher utilization of professional services than non-attendees.

The literature is void regarding qualitative factors connecting the reasons why individuals choose to attend a MDC, compared with receiving traditional practitioner-driven care. This study categorized reasons why people attend a clinic, the advantages and disadvantages of a MDC, the reasons why people do not attend, the circumstances influencing a decision to attend, and how travel impacts medical decision making. To be expected, travel and access were primary influences; however, respondents are willing to travel long distances to be seen at a clinic, illustrating the fact that participants' perceived benefits of MDC supersede travel time. Individuals searching for information, specialized treatment, and support perceive the fact that the MDC is a "one stop shop", where identified medical needs are met, thus increasing quality of life. Those individuals attending a MDC indicated that the MDC approach consolidates many appointments into one day, reducing overall travel, time, and expense. Participants who reported travel distance was a disadvantage stated that this was the case, specifically as disease progression led to decreased mobility and need for medical technology. Primary circumstances influencing ALS attendance were travel and distance from home. Interestingly, (N=9) participants were not aware of the function of a MDC, illustrating a need to continue advertising the benefits, function and availability of ALS clinics.

Relevance of the study to the theory and practice of psychology

Individuals are increasingly aware and accepting of the benefits of psychological interventions and services for emotional, physical, and psychological stress. Kurt and colleagues (2007) examined depression and anxiety in individuals with ALS and found the primary focus of palliative therapy in ALS is in coping with physical symptoms, but the psychological aspects of the disease are neglected. Patients and caregivers could benefit from greater access to psychological services delivered in a MDC. Two participant responses made recommendations for MDC related to psychological needs; one response identified opportunities to expand services by including access to Primary Care Physicians (PCP). MDCs do provide psychological services and holistic care. Patients and care givers may not be aware of these and of other services and focus on primarily receiving care from medical professionals. Responses regarding psychological and PCP services were found in the qualitative section, soliciting participants to describe advantages and disadvantages of attending a MDC. Participants wrote:

> "Think psychiatric is another need which is not really addressed there"
>
> "The ALS MDC is very focused, synchronized and provides superb support. There are no disadvantages. This is how it should be. The only way they could make it better is to include a nutritionist and psychologist as well."
>
> "A central place for all medical needs is good. Unfortunately, they don't provide primary care."

Implications of research findings related to diversity

Practitioners are increasingly faced with providing care in a multicultural society. Language barriers and health literacy complicate health care delivery. MDC need to be increasingly aware of communication styles when delivering information or assessing

disease progression with individuals from different backgrounds. Safe and quality health care to individuals requires MDCs and practitioners to understand how patients' socioeconomic backgrounds affect health beliefs and behaviors. MDCs need to consider ethnic, cultural, and economic backgrounds of patients. This study found that non attendees utilized pastoral care or chaplains at higher percentages (Attenders N=34, 14.2; Non Attenders N=17, 19.1) than attendees. Individual religious beliefs may be one factor influencing how care is received and perceived. Understanding cultural competency and health care beliefs, including interaction with unfamiliar medical systems of care may increase access to medical care and affect quality of life.

Implications of research findings as related to advocacy

Current treatments cannot stop or reverse ALS, and the current focus of care is on maintaining or improving QOL. Patient Centered Medical Home (PCMH) is an approach to providing comprehensive care and maintaining quality of life. The American Academy of Pediatrics developed the medical home model for delivering primary care that is accessible, continuous, comprehensive, family-centered, compassionate, and culturally effective to coordinate care for children with multiple and special health care needs (Stange, Miller, Nutting, Crabtreee, Stwart, & Jaen, 2010; Peek, 2010). PCMH works in collaboration with patients and families to facilitate partnerships between physicians, specialists, and community resources to optimize treatment and coordination of care. PCMH provides services to equip patients with an understanding of specialty care, provides educational services, out of home care family support, and other links to private and public services important for the overall health of a person. PCMH de-fragments health care, and provides accessibility at the times of patients' greatest need. Advocating

for this model brings providers and patients together to plan care, and create strategies to reduce barriers.

Limitations

Limitations of this study include lack of relevant demographic information including: marital status, social supports, other care giver supports, and socio economic status. Certain factors may contribute to overall QOL, and provide a catalyst for a person to choose attendance at a MDC. Supports relevant to specific demographic information could assist the individual with transportation, encouragement, and be a significant factor in contributing to quality of life to the extent of altering patients' perspectives and expectations regarding physical limitations. Conversely, individuals devoid of resources may find it difficult to attend a MDC.

External validity is affected by selection bias. Individuals routinely evaluated at MDCs have social supports, and may be less vulnerable to depression and psychological distress. Patients with severe depression, with a lack of social supports, and with difficulty coping with ALS are less likely to volunteer for research. Study volunteers may not be typical of average ALS patients. The study participants will need access to a computer, skills in utilizing a computer, and depending on the progression of the disease adaptive devices or social supports to complete the survey. Possession of a home computer may be associated with greater financial stability, and higher socioeconomic class.

Participants connected to the ALS community were to be informed of the study, and notification of the study may not have reached ALS patients residing in rural areas

not connected to ALS supports, or individuals not affiliated with the ALS community. Individuals with low QOL may not be interested in responding to the survey.

Differentiation of attendees and non attendees was by self-selection in question one. All survey participants had full access to the qualitative questions and were not directed to respond to certain question based on attending status. It is possible that participants could have responded to all questions regardless of attendance status. A number of participants responded with N/A, potentially indicating that a participant read the question, realized it was not applicable based on attendance status, and did not continue. However, a participant could answer the question by mistake, influencing the results. Reviewing the first qualitative question one participant wrote "I have never attended." The risk was reduced by having multiple coders review the responses and agreeing on which response to include in the analysis. Future survey design should include limited access to survey questions for each group, decreasing the opportunity to respond accidently to a question.

Suggestions for Future Work

This study was an attempt to gather information about the reasons why individuals attend or do not attend a MDC, circumstances in which a person would consider attending, and how travel determines treatment decisions. The study examined differences in QOL, physical functioning, and coping skills to further understand how individuals make decisions regarding their ALS and how a ALS MDC improves quality of life.

Qualitative analysis revealed convenience, travel, time, specialists, and integrative care were primary reasons for individuals' attendance at a MDC; conversely, individuals

did not attend a clinic due to travel distance, long exhausting days, and physical discomfort.

Future considerations would be an evaluation of barriers found in accessing a MDC, travel distance, and physical environment. One participant responded to the advantages and disadvantages of a MDC by describing the difficulty traveling and the long exhausting day of appointments at the clinic. Especially noted were the wait times between specialist appointments, time with "nothing to do", and "lack of food and drink." Future directions could be an evaluation of the physical environment and ways to improve perceptions of attendees and non attendees of a MDC. A second participant response noted a negative experience, influencing his decision to attend.

> "The advantage is getting input from many disciplines in 'one stop'. Two disadvantages are that the day is exhausting and I don't necessarily need to see all of the "specialists" in the clinic (but I have to anyway). Additionally, the clinic is clinic is 55 miles from my home and I have to arrive by 8:15 A.M. meaning my day starts way too early. I'm tired before I get there. I hate the waiting in between specialists with nothing to do, nothing to eat or drink. The whole ordeal sometimes takes as long as 7 hours."

Future research considerations could include qualitative analysis of the impact of psychological interventions found at a MDC and ALS patients and care givers' perceptions of treatment. It could also include an evaluation of those aspects that were found to be valuable and also those that were not useful. This could provide practitioners with a better understanding of how ALS patients and care givers would prefer to receive psychological interventions from the beginning diagnosis to later stage of the disease progression when speech is affected or lost, and mobility to attend psychological services outside of the home is difficult.

Summary and Conclusions

Current treatments cannot stop or reverse ALS. The current focus of care is on maintaining or improving QOL. Individuals attending an ALS clinic receive additional resources, medical treatments, and support. Salient features of how a MDC impacts the overall QOL was examined in this study; further exploratory research is beginning to reveal the reasons why individuals choose to attend an ALS clinic, the perceived benefits of this delivery of care, and any perceived barriers. The qualitative approach provides the researcher the opportunity to better understand the nature or meaning of a particular human experience from a national sample of individuals.

One of the significant advantages of a national web based survey method is that the collection of data is representative of a larger population. The findings and results from this study can be generalized to reveal wider social patterns and trends, and implications and generalizations can be made from the data that represent unique perspectives and health care beliefs of individuals across the United States. In a broader context, results of the reasons why individuals attend a MDC are relevant to other disciplines in the medical field that utilize the MDC approach to care; these include oncology and heart failure. Understanding the reasons why individuals attend a MDC or choose not to attend will provide exploratory information to the National ALS Association and medical practitioners in developing service provisions exclusively to provide care for ALS patients; understanding these reasons also offers a unique perspective of potential barriers for individuals who receive care from traditional practitioner care. Previous research, as well as this study identified travel and distance to a clinic as significant barriers to accessing care. Specific interventions might target travel

and accessibility. Future directions may focus on opportunities to decrease barriers faced by individuals who do not attend a MDC.

This study is part of a larger program of research designed to identify and address the psychosocial needs of individuals with ALS and their families. The study sought to discover the reasons why individuals with ALS access MDC versus receiving traditional practitioner driven care; it also sought to measure objectively the coping skills, quality of life, and physical function between groups. The overarching goal was to describe the nature and meaning of these human experiences, and to develop new knowledge from the perspective of individuals with ALS.

This study delves beyond the obvious to gain subjective understanding into identifying categories or themes directly related to Quality Of Life perceptions of ALS patients. Attending a Multi-Disciplinary Clinic would improve the QOL for individuals with severe disabilities and with prospects of a debilitating disease. A particular finding is that patients attending a MDC ALS clinic perceive QOL as being higher with respect to physical function and bulbar function; however, these patients have poorer overall levels of physical function as measured by the ALSFRS-R. It cannot be assumed that QOL decreases as the number of symptoms or degree of disability increases, inferring that perceived satisfaction with life depends on subjective conditions versus objective measures. A participant responding to reasons why he or she attends a MDC wrote:

> "At this point I feel it will give me the best chance to deal with the challenges I expect to have in the future. It will allow me to monitor how the disease is progressing using one source rather than putting everything together on my own."

Multidisciplinary Clinics provide enhanced care coordination, increased accessibility to health care professionals skilled in treating Amyotrophic Lateral

Sclerosis, and improvements in symptom control for patients with ALS. MDCs offer a comprehensive environment for treatment of ALS and are appropriate with onset of symptoms to obtain accurate diagnosis in an efficient manner.

The Patient-Centered Medical Home supports the MDC model by organizing care around patients, by working in teams, and by coordinating and tracking care over time. The main concern for this model is the reduction in need for primary care physicians; yet, this may alleviate the shortage of PCPs. The structure of the Patient-Centered Medical Home facilitates partnerships between patients, physicians, and family members.

Lack of funding is the predominant reason for the small numbers of MDC locations. Access to MDCs is an issue because of mobility, loss of functionality, distance to MDC sites, and level of needed caregiver support: Use of telemedicine should be explored to improve patient confidence in the ability to manage care. Supports and resources provided for ALS patients in MDCs serve to alter the overall patient perspective of quality of life.

References

Albert, S. M., Rabkin, J. G., Del Bene, M. L., Tider, T., O'Sullivan, I., Rowland, L. P., & Mitsumoto, H. (2005). Wish to die in end-stage ALS. *Neurology*, 65, 68-74.

ALS Association. ALS association certified centers. Retrieved February 1, 2009, from http://www.alsa.org

Averill, A. J., Kasarskis, E. J., & Segerstrom, S. C. (2007). Psychological health in patients with amyotrophic lateral sclerosis. *Amyotrophic Lateral Sclerosis*, 8, 243-254.

Corbin J., & Strauss A. (2008). Basics of qualitative research: techniques and procedures for developing grounded theory (3rd ed.). Sage Publications.

Corcia, P., & Meininger, V. (2008). Management of amyotrophic lateral sclerosis. *Drugs*, 68(8), 1037-1048.

Dorsey, E. R., Deuel, L. M., Voss, T. S., Finnigan, K., George, B. P., Eason, S., Miller, D., Reminick, J. I., Appler, A., Polanowicz, J., Viti, L., Smith, S., Joseph, A., Biglan, K. M. (2010). Increasing access to specialty care: a pilot, randomized controlled trial of telemedicine for Parkinson's disease. *Movement Disorders*, 25(11), 1652-1659.

Ducharme, A., Doyon, O., White, M., Rouleau, J. L., Brophy, M. (2005). Impact of care at a multidisciplinary congestive heart failure clinic: a randomized trial. *Canadian Medical Association Journal*, 173(1), 40-45.

D'Zurilla, T. J., Nezu, A. M., & Maydeu-Olivares, A. (1995). Manual for the Social Problem-Solving Inventory-Revised. Unpublished manuscript, State University at New York at Stony Brook

Elliott, T. R., Shechuk, R. M., & Richards, J. S. (1999). Caregiver social problem-solving abilities and family member adjustment to recent-onset physical disabilities. *Rehabilitation Psychology*, 44(1), 104-123.

Gabel, M., Hilton, N. E., Nathanson, S. D. (1997). Multidisciplinary breast cancer clinics. *Cancer*, 79(12), 2380-2384.

Kaufmann, P., Levy, G., Montes, J., Buchsbaum, R., Barsdorf, A. I., Battista, V., Arbing, R., Gordon, P. H., Mitsumoto, H., Levin, B., & Thompson, J. L. P. (2007). Excellent inter-rater, intra-rater, and telephone-administered reliability of the ALSFRS-R in a multicenter clinical trial. *Amyotrophic Lateral Sclerosis, 8*, 42-46.

Kurt, A., Nijboer, F., Matuz, T., & Kubler, A. (2007). Depression and anxiety in individuals with amyotrophic lateral sclerosis. *CNS Drugs, 21*(4), 279-291.

McDonald, E., Wiedenfeld, S., Hillel, A., Carpenter, C., & Walter, R. (1994). Survival in amyotrophic lateral sclerosis: the role of psychological factors. *Archives of Neurology*, 51, 17-23.

Mitchell, J. D., & Borasio, G. D. (2007). Amyotrophic lateral sclerosis. *Lancet, 369*, 2031-2041.

Montes, J., Levey G., Albert, S., Kaufmann, P., Buschsbaum, R., Gordon, P. H., & Misumoto, H. (2006). Development and evaluation of a self administered version of the ALSFRS-R. *Neurology, 67*, 1294-1296

National Institute for Neurological Disorders and Stroke and National Institute of Health. *Amyotrophic Lateral Sclerosis Fact Sheet*. Retrieved December 10, 2008, from http://www.ninds.nih.gov/disorders/amyotrophiclateralsclerosis

Peek, C. J. (2010). Building a medical home around the patient: what it means for behavior. *Families, Systems & Health,* 28(4), 322-333.

Radunovic, A., Mitsumot, H., & Leigh, N. (2007). Clinical care of patients with amyotrophic lateral sclerosis. *The Lancet Neurology*, 6, 913-925

Rocha J. A., Reis, C., Simoes, F., Fonseca, J., & Mendes R., J. (2005). Diagnostic investigation and multidisciplinary management in motor neuron disease. *Journal of Neurology, 252*, 1435-1447.

Rowland, L. P., & Shneider, N. A. (2001). Amyotrophic lateral sclerosis. *The New England Journal of Medicine*, *344*(22), 1688-1700.

Shoesmith, C. L., & Strong, M. J. (2006). Amyotrophic lateral sclerosis; update for family physicians. *Canadian Family Physician, 52*(12), 1563-1569.

Simmons, Z. (2005). Management strategies for patients with amyotrophic lateral sclerosis from diagnosis through death. *The Neurologist 11*(5), 257-270.

Sorenson, E. J., Mandrekar J., Crum, B., & Stevens, J. C. (2007). Effect of referral bias on assessing survival in ALS. *Neurology, 68*, 600-602.

Stephens, H. E., Walsh, S.M., Simmons, Z. (2008). A national survey of quality of life for ALS patients in the United States. *Amyotrophic Lateral Sclerosis* 2008; 9(suppl 1):12.

Stange, K. C., Miller, W. L., Nutting, P. A., Crabtreee, B. F., Stwart, E. E., & Jaen, C. R. (2010). Context for understanding the national demonstration project and the patient centered medical home. *Annals of Family Medicine*, 8(1), 2-8.

Stricker, G. (2002). What is a scientist-practitioner anyway? *Journal of Clinical Psychology*, 58(10), 1277-1283.

Sticker, G. (2006). The local clinical scientist, evidence-based practice, and personality assessment. *Journal of Personality Assessment*, 86(1), 4-9.

Stricker G., & Trierweiler, S., J. (2006). The local clinical scientist: A Bridge between science and practice. *Training and Education in Professional Psychology*, S(1), 37-46.

Traynor, B. J., Alexander, M. Corr, B. Frost, E., & Hardiman, O. (2003). Effect of a multidisciplinary amyotrophic lateral sclerosis (ALS) clinic on ALS survival: A population based study, 1996-2000. *Journal of Neurology Neurosurgery and Psychiatry*, *74*, 1258-1261.

Valdmanis, P., & Rouleau, G., A. (2008). Genetics of familial amyotrophic lateral sclerosis. *Neurology*, 70, 144-152.

Van den Berg, J. P., Kalmijn, S., Lindeman, E., Veldink, J. H., De Visser, M., Van der Graaff, M, M., Wokke, J. H. J., Van den Berg, L. H. (2005). Multidisciplinary ALS care improves quality of life in patients with ALS. *Neurology*, 65, 1264-1267.

Vignola, A., Guzzo, A., Calvo, A., Moglia, C., Pessia, A., Cavallo, E., Cammarosano, S., Giacone, S.,Ghiglione, P., & Chio, A. (2008). Anxiety undermines quality of life in ALS patients and caregivers. *European Journal of Neurology*, 15, 1231-1236.

Zoccolella, S., Beghi, E., Palagano, G., Fraddosio, A., Guerra, V., Lepore, V., et al. (2007). ALS multidisciplinary clinic and survival. *Journal of Neurology*, 254, 1107-1112.

Appendix A: Questionnaire

Introduction:

A National Study of ALS Multidisciplinary Clinic Use

1. A National Study of ALS Multidisciplinary Clinic Use

Researchers under the direction of Dr. Zachary Simmons at the Penn State University College of Medicine are interested in learning about the quality of life, problem solving, and use of clinical services of persons with ALS (PALS) across the United States. This study will take approximately 60 minutes of your time to complete a set of questions abut your physical health state, your quality of life, and information about where you live and what type of care you receive for your ALS. The information that you provide in this survey is confidential and will be shared only with the research team. This survey is labeled with a code number and not with your name or other identifying information. If you should have any questions or concerns about this study, please contact the study coordinator, Beth Stephens, at (717) 531-003, 283395 or hstephens1@psu.edu.

Risks and Discomforts:

There is no physical discomfort, and there are no physical risks associated with this study. You may experience emotional discomfort and become upset due to thinking about how all these factors impact on your life. You may experience fatigue (tiredness), so please work at your own pace.

Only one questionnaire response will be accepted per computer.

If you have participated in a Quality of Life study in the past (i.e., in a clinic setting), you are still eligible to participate in this study.

2. Demographics

Age

___What is your age in years?

Gender

___Male

___Female

Site of ALS Symptom Onset

When did you first notice symptoms of ALS? If you do not know the exact date, please use “01” for month or day

MM/DD/YYYY

Site of ALS Symptom Onset

___Limb

___Bulbar

___Breathing

ALS Diagnosis

When were you diagnosed with ALS? If you do not know the exact date, please use “01” for month or day

MM/DD/YYYY

Do you have other family members with ALS?

___Yes

___No

Tell us where you live

State:

Zip / postal code:

Country:

3. Multidisciplinary Clinic Use

Some people with ALS attend a multidisciplinary clinic. We define a multidisciplinary clinic as a team of health care providers including: neurology, physical therapist, occupational therapist, nutritionist, speech therapist, mental heath clinician, social worker, nurse, and others. Other ALS patients do not received medical treatments from a multidisciplinary clinic; they go to neurologists, other doctors and health care practitioners.

How often have you attended a multidisciplinary clinic (MDC)?

___ Never, and I have no plans to attend a MDC

___ Never, but I have a scheduled appointment or plan to attend in the upcoming future

___ One time for a single diagnostic visit, but I have no plan to attend again

___ One time, and I have a schedule appointment or plan to attend in the upcoming future

___ More than 1 time, and plan to continue my treatment at a MDC

___ More than 1 time, and I do not plan to continue my treatment at a MDC

If you do go to a multidisciplinary clinic, please tell us why you go. Please give us as much information and detail as you can.

_____

_____

_____

_____

Please tell us about any advantages or disadvantages from receiving care from a Multidisciplinary Clinic. Please give as much detail and information as you can.

_____

_____

_____

_____

If you do not go to a Multidisciplinary Clinic please tell us why. Please give us as much information as you can.

_____

_____

_____

_____

If you do not attend a multidisciplinary clinic, under what circumstances might you attend an ALS clinic? Please give us as much information as you can.

_____

_____

_____

_____

If you attend a multidisciplinary clinic, who influenced your decision to attend a multidisciplinary clinic?

___ Spouse or other family member

___ Physician

___ Another ALS Patient

___ALS Association, Muscular Dystrophy Association, or other disease-based group

___No one. I made the decision on my own

___ Other

Other (please specify) _____

Who first told you about a multidisciplinary clinic?

___ Neurologist

___ Primary Care Physician

___ Internist

___ Nurse

___ALS Association, Muscular Dystrophy Association, or other disease-based group

___ Other: please specify _____

Distance of ALS Clinic

4. Treatments and Services for ALS

How does travel distance to a multidisciplinary clinic or other health care professional influence your treatment decisions? Please give as much information as you can.

_____

_____

_____

_____

What treatments do you use for your ALS?

___ Riluzole (Rilutek)

___ Antidepressants

___ Medications to reduce saliva production

___ Medications to reduce cramping/spasticity

___ Pain medications

___ Sleeping medications

___ Treatments for constipation

___ Treatments for urinary urgency

___ Percutaneous endoscopic gastronomy (PEG) or feeding tube

___ NIPPV or BiPAP

___ Tracheostomy or Mechanical Ventilation

___ Augmentive Communication device

___ Power Wheel chair

___ Experimental medications

___ No treatment

___ Complementary and Alternative Medicine (includes supplements, massage, holistic medicine, chiropractice care, acupuncture, etc)

___ Other (please specify)_____

Do you use or have you been referred to any of these health care providers as part of your care for ALS? Check all that apply

___ Neurologist

___ Nurse

___ Respiratory Therapist

___ Pulmonologist

___ Physical Therapist

___ Occupational Therapist

___ Mental Health Professional

___ Social Worker

___ Pastoral Care or Chaplain

___ Dietician

___Speech Therapist

___ Complimentary and alternative medicine professional (chiropractors, naturopathic doctors, acupuncturists, etc).

___ In Home Care

___ Hospice Services

Demographic Questions

- Today's Date
- Age (in years)
- Date of onset of ALS symptoms (month/year)
- Date of Diagnosis (month/year)
- Gender (male/female/prefer not to answer)
- Site of ALS symptoms (limb, bulbar, breathing)
- Do you have other family members with ALS (yes/no)
- Distance from nearest MDC to home in miles
- Zip code

Appendix: B Study Announcement

Persons with Amyotrophic Lateral Sclerosis (ALS)

Wanted for a Research Study

Researchers at Penn State College of Medicine are seeking participants for a research study on why individuals access a multidisciplinary clinic versus receiving traditional practitioner care, Quality of Life (QOL), and social problem solving skills of persons with ALS. The purpose of the study is to better understand the QOL, and problem solving skills of patients with ALS who receive different forms of care for the disease. Participants will complete a questionnaire (online or with a paper and pencil version) that asks questions about physical health status, quality of life, social problem solving skills, and use of medical services for care of ALS. Survey responses are confidential.

All persons 18 years of age or older, who have been diagnosed with the disease Amyotrophic Lateral Sclerosis are eligible to participate in this study.

To access the online questionnaire, go to our ALS Clinic website:
http://www.alsphiladelphia.org/pennstatehershey

For more information or to request a paper copy of the questionnaire, call the study coordinator, Beth Stephens, at 717-531-0003, extension 283395, or by email at hstephens1@psu.edu

Study Director: Zachary Simmons, MD, Department of Neurology, Penn State College of Medicine.

'This research study has been approved by the Institutional Review Board, under federal regulations, at Penn State College of Medicine, Penn State Hershey Medical Center.'
Appendix: C Hershey Medical Center IRB Abstract

Request for Waiver of Consent.doc (Orig. 10/09/05; Rev. 10/17/2005)

INSTITUTIONAL REVIEW BOARD
PENN STATE COLLEGE OF MEDICINE
Penn State Milton S. Hershey Medical Center

REQUEST FOR WAIVER OR ALTERATION OF CONSENT

IRB Protocol No.:

Protocol Title: A National study of Amyotrophic Lateral Sclerosis Multidisciplinary Clinic Utilization.

Principal Investigator: Zachary Simmons

According to the federal regulations, an IRB may waive the requirements to obtain informed consent or approve a consent procedure which does not include, or which alters, some or all of the elements of informed consent provided the IRB finds and documents that: (1) The research involves no more than minimal risk to the subjects; (2) The waiver or alteration will not adversely affect the rights and welfare of the subjects; (3) The research could not practicably be carried out without the waiver or alteration; and (4) Whenever appropriate, the subjects will be provided with additional pertinent information after participation.

To request a waiver or alteration of the requirement to seek informed consent of subjects, complete the following:

1. Indicate which of the following applies (Select one):

☐ a. Informed consent will not be obtained.

☐ b. A consent procedure will be used which does not include, or which alters, some or all of the elements of informed consent.
Explain which elements are not included or which elements are altered and why this is necessary:

2. Describe how the research meets all of the following four conditions:

a. The proposed research involves no more than minimal risk to the subjects because:
it is an online questionnaire that asks a national sample of patients with ALS about their medical service use, quality

b. The waiver or alteration will not adversely affect the rights and welfare of the subjects because:
participants can can chose not to complete the online survey or can stop participation at any time

c. The proposed research could not practicably be carried out without the waiver or alteration because:
it is an online survey available on our ALS Clinic website. informed consent is not practical for this study.

d. Whenever appropriate, the subjects will be provided with additional pertinent information after participation (Indicate why information will not be provided **or** explain why, how, and when information will be provided):
a summary of the results of the study will be posted on our ALS Clinic website

IRB. No. ____
Date
Office Use Only: _____

Institutional Review Board

Penn State College of Medicine

Penn STATE MILTON S. Hershey Medical Center

Protocol Summary Abstract

Title: A national study of Amyotrophic Lateral Sclerosis Multidisciplinary Clinic Utilization
Principal Investigator: Zachary Simmons, MD Dept./Div: Neurology

1. Rationale: The study is designed to discover why individuals with ALS access multidisciplinary clinics versus receiving traditional practitioner driven care, and to discuss the differences in quality of life, functional status, and coping skills for these two distinct ALS populations. The research project is guided by the following question: For what reasons did you decide to seek treatment and services at an ALS multidisciplinary clinic? The investigation is part of a larger program of research designed to identify and address the psychosocial needs of persons with ALS and their families. Hypothesizing that the findings will show a higher quality of life, coping skills, and significant differences in states of health of individuals with ALS attending MDC compared to individuals receiving traditional care.

2. Key Objectives: To examine the characteristics of persons affected by ALS who access a MDC clinic versus receiving traditional practitioner driven care. Collect Quality of Life, Functional Rating, and coping skills data on a national sample of patients with ALS.

3. Study Population: National sample of patients diagnosed with ALS and who are registered with the ALS Association.

4. Major Inclusion & Exclusion Criteria: Diagnosis of ALS

5. Method of Identification of Subjects/Samples/Medical Records: Participants will be notified of the study by the ALS Association through the distribution of flyers, letters, and postings on newsletters and websites. Dr. Simmons will notify his own patient population with a letter. No medical records will be used for this study. The investigators will not have access to the registry of patients with the ALS.

6. Consent Process and Documentation: The investigators request a waiver of informed consent. Completion of the anonymous survey will imply consent for participation in the research.

7. Allocation to Groups: Data will be analyzed by groups: 1) ALS patients receiving multidisciplinary care; 2) ALS patients receiving other forms of care.

8. Summary of Procedures: Participants will complete an anonymous survey on the internet that asks questions about why they do or do not attend an ALS Multidisciplinary Clinic, QOL, functional status, coping skills, and type of care and treatments used for ALS. Alternatively, participants can request a paper copy of the questionnaire and postage paid and addressed envelope to return the survey. No coding system will be used to identify survey responses.

9. Major Risks & Discomforts: Possible emotional discomfort from thinking about one's quality of life with ALS. A statement at the beginning of the survey will address this potential discomfort.

10. Potential Benefits: No benefits to the participant. The study will provide us with a greater understanding of why ALS patients attend or do not attend a Multidisciplinary Clinic, QOL, functional status, and

coping skills, from a national population of ALS patients, and will aid in the understanding of the association of QOL and coping skills with use of multidisciplinary care.

11. Privacy and Confidentiality: Response to the survey is anonymous. There is no code that links survey responses to individual participants. Data will be stored in a password protected and secure server in Neurology.

12. Qualifications and Research Experience of Principle Investigator: Zachary Simmons, MD has directed the ALS clinic at Penn State Hershey Medical Center since its inception in 1995, and has a long-standing research interest in understanding QOL in patients with ALS. Dr. Simmons has presented his research findings at national conferences and academic meetings and has authored numerous papers on QOL in ALS.

13. Study Site Location(s) The internet survey will be hosted by Survey Monkey ®. The survey will be posted on the website of the ALS Association and the website of the Penn State Hershey ALS Clinic.

14. References: Traynor, B. J., Alexander, M. Corr, B. Frost, E., & Hardiman, O. (2003). Effect of a multidisciplinary amyotrophic lateral sclerosis (ALS) clinic on ALS survival: A population based study, 1996-2000. Journal of Neurology Neurosurgery and Psychiatry, 74, 1258-1261.