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Kelsey Drewry Date

Narrative Interview for the Identification of Supportive Care Needs for Patients with Amyotrophic Lateral Sclerosis: An ethical analysis and pilot study

By

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Master of Arts

Bioethics

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# Narrative Interview for the Identification of Supportive Care Needs for Patients with Amyotrophic Lateral Sclerosis: An ethical analysis and pilot study

By

Kelsey Drewry
B.S., Oregon State University, 2014

Advisor: Kathy Kinlaw, MDiv

An abstract of
A thesis submitted to the Faculty of the
James T. Laney School of Graduate Studies of Emory University
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2017

## Abstract

Narrative Interview for the Identification of Supportive Care Needs for Patients with Amyotrophic Lateral Sclerosis: An ethical analysis and pilot study

By Kelsey Drewry

Amyotrophic lateral sclerosis (ALS) is a neurodegenerative disease that causes patients to progressively lose control of voluntary muscles responsible for movements like chewing, walking, talking, and breathing. No cure exists and disease progression is highly variable. Due to the severity of symptoms and their impact upon patient and caregiver quality of life, the need for palliative and supportive care services for patients with ALS has been clearly identified. However, recent studies suggest a need to tailor these services to the needs of patients with neurological disorders, but little research has been published to this effect for ALS. This paper discusses the ethics of palliative and supportive care, their intersection with narrative ethics, and a proposed research method that integrates patient-centered principles from each of these disciplines to elicit care needs from patients' stories of their disease experience. The paper also describes a pilot study to test the methodology of illness narrative interview and thematic analysis to identify supportive and palliative care needs for patients with ALS and their caregivers. Ten patient and family groups with ALS functional rating scores between 27-44 participated in a two-tiered, semi-structured illness narrative interview. Grounded theory guided the thematic analysis of interview transcripts. Sixty-one themes emerged and were grouped into two distinct genres, six domains, and eighteen categories. All patients discussed the importance and helpfulness of the love and support of their family. Faith communities and spiritual practices, maintaining a positive outlook, and altruistic action were also amongst the most predominant themes in the "Expressed Needs" genre. Most prevalent in the "Observed and Experienced Changes" genre were themes of appreciating the value of life and time, thinking about death and dying, and concern for the wellbeing of family. The most commonly identified unmet care need was psychological support for depressive symptoms. The multifaceted needs expressed by participants cohere with the multidisciplinary approach of supportive and palliative care and were well elicited by the narrative interview approach. The results of this pilot study suggest that illness narrative interview and thematic analysis may be utilized effectively to elicit palliative and supportive care needs.

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## Chapter 1: Understanding Supportive and Palliative Care

The primary aims of this study are to: (1) assess the efficacy of illness narrative interview and thematic analysis as a means to elicit supportive care needs for patients with amyotrophic lateral sclerosis, and (2) to evaluate identified needs with respect to current tools for assessing supportive and palliative care needs among patients with neurologic conditions. Of note, due to common misconceptions about the nature of palliative care and to honor the explicit preferences of care team at the Emory ALS Center where this study was conducted, the term *supportive care* has been used instead of *palliative care* in all communications with patients (including all IRB approved documents) in order to avoid potential for distressing associations between *palliative* and *end-of-life* care. This conflation is understandable and common amongst patients and providers alike throughout the healthcare system. As described by Bruera and Hui, supportive, palliative, and hospice care are similar in kind, but vary in their implementation with respect to the progression of a patient's condition [1].

### *Supportive Care: A new guise for early-intervention palliation*

The term *supportive care* emerged in the early 1990's, defined explicitly in 1994 by Page as:

> the provision of the necessary services for those living with or affected by cancer to meet their informational, emotional, spiritual, social, or physical needs during their diagnostic, treatment, or follow-up phases encompassing issues of health promotion and prevention, survivorship, palliation, and bereavement…In other words, supportive care is anything one does for the patient that is not aimed directly at curing his disease but rather is focused at helping the patient and family get through the illness in the best possible condition. [2]

Other scholars have argued for a narrower definition of supportive care as a practice concerned with the management of adverse effects of treatment, again in the specific context of advanced cancer care [3]. For the purposes of this study the first, broader definition of supportive care originally outlined by Page has been adopted as a surrogate term for *palliative care* (which will be defined and discussed subsequently). This concept of supportive care has been adapted more widely to address suffering from a variety of serious diseases beyond cancer. Supportive care may benefit anyone "living with or affected by" life-threatening or life-limiting disease, especially a disease with a trajectory as variable as ALS.

Corroborating the concerns expressed by the Emory ALS Center physicians at the conception of this project, a recent study conducted in the context of advanced cancer care suggested that use of the term *supportive care* is less likely to cause distress in patients and families as compared to *palliative care*. The physicians surveyed reported that they were more likely to refer patients to *supportive care* [4]. A subsequent survey of oncologists revealed that physicians were significantly more likely to refer newly diagnosed patients to *supportive care*, because *palliative care* was perceived as being synonymous with hospice, decreasing hope, and being less appropriate for treatment of side effects [5]. Further, in a before-and-after name change comparison, Dalal and colleagues found that when *supportive care* was used, the number of referrals made to inpatient palliative care services increased, and referrals occurred earlier in the outpatient setting [6]. Substantiating these findings from the patient perspective, it has also been shown that the term *supportive care* is associated with better understanding, more favorable impressions, and higher perceived need by patients as compared to *palliative*

*care* [7]. Importantly, the authors of these studies understand palliative and supportive care to be the exact same set of services, but see *supportive care* as a new term that may be used to remove negative associations with *palliative care* and improve the quantity and timeliness of patient referrals to these beneficial services.

As stated previously, this study employs the term *supportive care* in all patient communications to avoid any distress it may cause patients and family members, as well as to mitigate the impact of any negative associations with *palliative care* that may serve as a barrier to the sharing of insightful perspectives and experiences. Throughout this manuscript, *palliative and supportive care* will be used together as a single unit. This is done, in part, to reflect their related nature, but also in recognition of the distinctions made by Bruera and Hui. In their view, *supportive care* and *palliative care* provide the same or similar services, and maintain equivalent aims of whole-person and family support, but are appropriately invoked at different stages in illness progression, largely to promote maximum and timely referrals (see Figure 1) [1, 3]. It seems that current trends in both literature and practice are to replace the term *palliative care* with *supportive care,* at least in the early-intervention setting to remove barriers to referral. However, *palliative care* is not completely replaced by the term *supportive care,* as it may continue to be utilized during the advanced- and end-stages of progressive disease. Because of the variability in disease progression and symptom severity characteristic of ALS and experienced by the participants of this study, the use of *supportive and palliative care* together throughout this manuscript reflects the diversity of the participating patient population. The choice to group these terms together attempts to encompass current definitions expressed in the literature without excluding or mislabeling services that

remain under the purview of palliative medicine by definition. Figure 1 provides a visual representation of the relationship between hospice, palliative, and supportive care to assist in conceptualizing the relationship of these three fields.

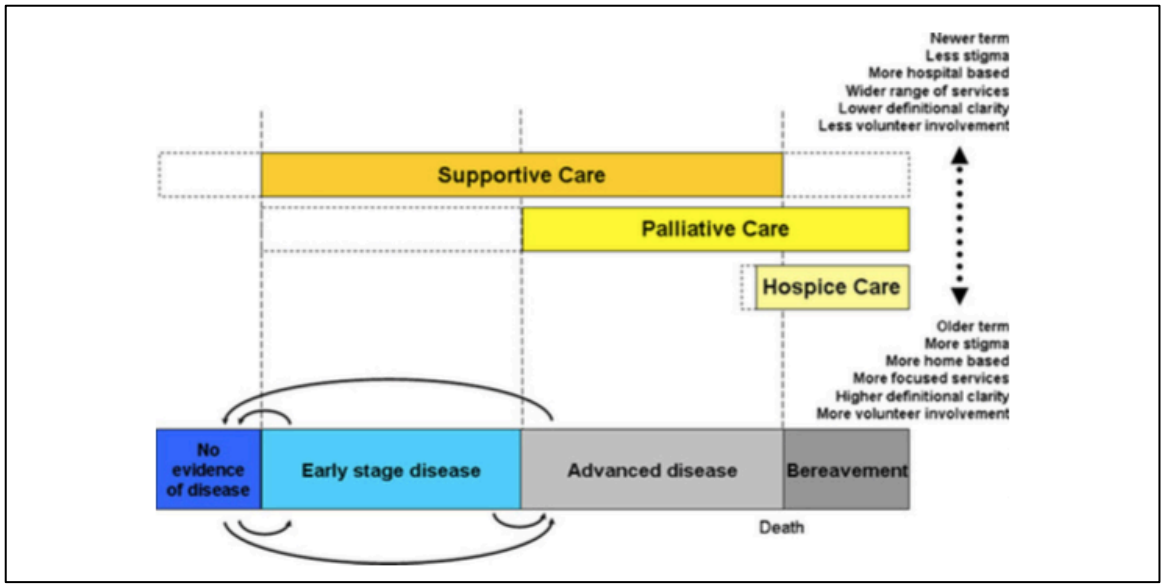


**Figure 1-** Temporal and progressional relationship between supportive, palliative, and hospice care. Image from Hui *et al* 2013. "Different stages of disease are depicted at the *bottom*, with *solid arrows* showing that patients can shift from one stage to another. The patient population for "supportive care," "palliative care," and "hospice care" is shown by the *horizontal bars* above…the *dashed boxes* illustrate the evolving nature of these definitions to expand on their scope of service…Other distinguishing features among the three terms are listed on the right hand side." [3]

As depicted in Figure 1, the framework of Bruera and Hui describes *hospice care* as a subset of *palliative care,* which is part of *supportive care*. In combining *palliative* and *supportive care* for the purposes of this investigation, it is essential that the two not be subsequently conflated with *hospice care*. In order to distinguish the important differences between *hospice care* and *palliative/supportive care*, further discussion of the two fields follows.

***Distinguishing and Defining Palliative Care***

The word *palliate* is derived from the Latin *pallium*, meaning to cloak or cover. In practice, palliative medicine seeks to ameliorate or mitigate suffering, rather than to simply mask it. Where as curative medicine is focused on the removal or treatment disease, it may be insufficient to alleviate symptoms of suffering associated with severe illness. Palliative care, then, is intended to supplement curative endeavors with a model of interdisciplinary care designed to mitigate patient and family experience of illness-related suffering. As defined by the World Health Organization (WHO),

> Palliative care is an approach that improves the quality of life of patients and their families facing the problem[s] associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual. [8]

As outlined in this definition, the central focus of palliative care is the prevention and relief of suffering within its many domains—physical, emotional/psychological, social/relational, and spiritual/existential. The inherent moral assumption behind this definition of palliative care is that suffering is *bad*—an assertion not limited to the palliative arena, but widely embraced by medicine as a whole. Though curative medicine embraces the idea of *necessary pain*, that is, some quantity of discomfort that can be justified by future improvements in condition, medicine generally understands pain and suffering, in their many manifestations, to be negative components of the human condition deserving of treatment and care [9, 10]. Other schools of thought exist, which embrace and understand suffering in an entirely different manner, but discussions of such ideologies are beyond the scope of this investigation. For the purposes of this discussion,

pain and suffering will be regarded as sufficiently negative to warrant concern and attempts at palliation (whenever such attempts are desired by the recipient of such care).

If we understand the objective of palliative and supportive care to be the mitigation of pain and suffering caused by severe illness, then we must further clarify our concepts. Pain, epistemologically speaking, is relatively easier to characterize than suffering. Pain is generally understood as the physical experience of discomfort due to illness or injury [11]. Science and medicine recognize the general mechanism of pain to be neurological in nature, an effect of biochemical signaling cascades within the body [12, 13]. Suffering, however, is more challenging to define. Though it is often associated with reference to pain, suffering may be best understood as a response to damage, injury, or loss of an aspect (or aspects) of an individual's personhood—including the lived past; the family's lived past; one's culture and/or society; roles, associations and relationships; one's body, unconscious mind, political being, secret life, and/or one's perceived future [14, 15]. The nature of such injuries and their diverse effects are not well understood, but prevention and/or amelioration of the associated experience of suffering related to serious illness are the aim of palliative care.

This field is distinguished by its holistic and inclusive approach to the patient experience, a characteristic that has been argued to date back to its roots in Asklepion medicine and retained due to the epistemological challenges of suffering [16]. Where other medical specialties tend to become increasingly narrow in purview, palliative care aims to improve patient well-being by addressing a multitude of factors that contribute to one's experience of life and illness. Accompanying the distinctive holistic nature of palliative care is the broad scope of its focus. While the traditional patient—the

individual suffering from severe chronic or life-threatening illness—is the primary concentration of the palliative care team, the broader focus of care extends to the patient's family and caregivers, recognizing that disease-based suffering is not limited to the patient experiencing illness [17].

In discussing palliative medicine, it is essential to clearly delineate the goals of care, applicable patient populations, and implications of palliative consultation. A 2014 study of barriers to palliative care referrals revealed that providers had limited knowledge regarding the nature of non-hospice palliative care, how it differs from hospice, what it offers patients, families, and providers, when it is indicated, and how to access it [18]. While some of these issues may be overcome by employing the term *supportive care* in place of *palliative care*, it remains essential to continue educational endeavors regarding the distinction between hospice and palliative/supportive care. To avoid perpetuating damaging conflations between *palliative*/*supportive care* and *hospice/end-of-life care*, we must further clarify two common misconceptions about these fields prior to further discussion.

First, non-hospice palliative care is not elected instead of or in exclusion to curative or "disease-directed" interventions. In ideal circumstances, palliative care is a supplement to conventional disease management procedures, minimally at first, then increasing as the disease progresses, the patient's condition declines, and/or curative measures are exhausted (see Figure 2). If the patient responds to curative measures, palliative support can be tapered off as symptoms lessen [19, 20]. This model of integrative palliative care allows for patients to not only experience the benefits of etiological symptom removal provided by curative medicine, but also to gain from

supportive care designed to mitigate the challenging physical, emotional, and relational aspects of the illness experience that may otherwise be left unaddressed.

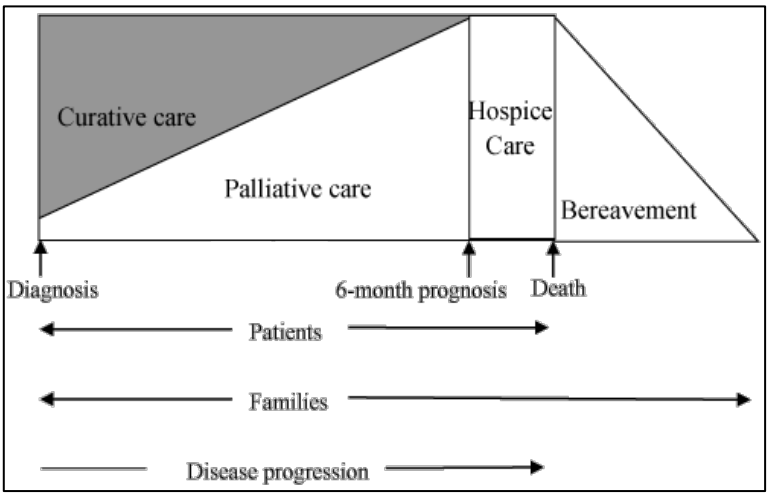


Figure 2- Model of palliative care integration throughout illness trajectory. Image from Guo *et al*. [19]

The second essential distinction is that palliative care is not equivalent to hospice or end-of-life care. Though the two are often erroneously conflated, there are important differences between these disciplines that must be clarified in order to minimize patient rejection of palliative care services and to maximize appropriate provider referral. There is a pervasive misconception that palliative medicine is reserved for end-of life care or is indicative of a terminal prognosis, appropriate only for individuals who are in the process of dying. This unfortunate misunderstanding likely derives from the close evolutionary history of hospice and palliative medicine, but must be corrected if the benefits of supportive/palliative care are to be more widely accessed.

As referenced in the context of American health care, *hospice* is most closely linked to the Medicare Hospice Benefit—a program offered to all Medicare beneficiaries

with the aim of providing access to high quality end-of-life care. In order to be eligible for the Medicare Hospice Benefit, an individual must have a terminal diagnosis and an expected prognosis of six months or less, confirmed by two physicians [21, 22]. Enrolling in hospice is accompanied by an informed decision made by, or on behalf of, the patient to forego further curative treatment and to direct focus toward improving the time left to live. In direct contrast, neither a terminal diagnosis nor poor prognosis is a prerequisite for the recommendation or receipt of palliative care. In fact, palliative care provided in conjunction with curative treatment has been demonstrated to not only improve quality of life and decrease depression, but also to increase median survival for patients with metastatic cancer [23].

In the context of non-hospice palliative care, the focus is on managing symptoms that decrease quality of life for patients suffering from life threating or life-limiting illness. This includes the assessment and management of physical symptoms like pain, fatigue and insomnia, dyspnea, and gastrointestinal symptoms, and non-physical causes of suffering such as psychosocial distress, functional, financial, spiritual, and family concerns [1, 24]. Most often, members of a palliative care team provide these services, though patients may be referred to a tertiary care provider for necessary specialist care.

It is unequivocally true that palliative care is a large component of hospice care and the Medicare Hospice Benefit—this is sensible, as the mitigation of suffering and improvement of quality of life is a primary concern for quality end-of-life care. However, while palliative care is certainly appropriate during the dying process, it is simply untrue that palliative care is *only* appropriate for patients who are dying.

The increasing demand for and emphasis on non-hospice palliative and supportive care stems from a fundamental historical change in the etiology of death. Modern medicine has enabled both the prolongation of life and of the dying process; death is no longer a sudden, unpredictable event. Instead, a long process of decline now commonly precedes actual dying. As the duration of this process has increased, medicine has been faced with a paradigm shift in order to maintain care and compassion for its most vulnerable patients.

> A patient used to receive a diagnosis of a life-threatening disease and a treatment plan was laid out with little attention paid to the consequences of the treatment or what will be done if the treatment fails to arrest the disease. And it was only in the last few days or weeks of life that a patient was offered comfort care measures. Today, as compassionate healthcare providers it is incumbent upon us to introduce comfort care early in the process. [25]

Modern integrative palliative and supportive care offers to provide comfort throughout the illness trajectory and allow for the needs of patients and families to be met throughout all stages of disease. A dramatic change is therefore necessary in the recognition of what these fields have to offer, and focus must be on the benefits of palliative and supportive care for those living with chronic and progressive diseases.

***The Evolution of Hospice, Palliative, and Supportive Care***

It is likely that much of the confusion and conflation that exists between *palliative* and *hospice care* is due to their close evolutionary history. The origin of palliative medicine itself is intertwined with the substantial changes in the typical human life course during the twentieth century. In 1900, the average life expectancy in the United States was forty-seven years. By 2000, it had increased to seventy-seven [26, 27]. The advent of germ theory and subsequent advances in biomedicine and public health largely account for the incredible magnitude of this change, and new technologies completely altered the way in which individuals receiving care in biomedically developed countries experienced illness and death.

As described by bioethicist Daniel Callahan, the prevailing imperative of biomedical research "…stems from the view that medicine has an almost sacred duty to combat all the known causes of death." [28] As medicine became increasingly successful at prolonging life, societal perceptions of death began to change. Describing modern social attitudes toward death, Phillipe Ariés characterizes the current phase of Invisible or Forbidden Death as predicated on the unconscious or theoretical belief that death is avoidable with sufficient investment in medical care and scientific research [29, 30]. Theoretically, such a framework may have been appropriate for modeling the treatment of, and reduction of mortality from, infectious disease. However, the paradigm of the scientific method, with its footing in early twentieth century germ theory, did not adapt to the changing patterns of death. This tension has led physicians and the public alike to perceive death not as a natural process inherent to life, but rather as a failure of modern medicine [1, 31]. As such, death has become stigmatized, forbidden, and largely removed

from public observation and discourse [29, 30]. With these concurrent social and biomedical developments, the American population began to live longer lives with increased incidence of chronic illness and age-associated medical spending, accompanied by increasing durations of disease-related suffering and bereavement inadequately addressed by the scientific model of medicine.

These factors led to recognition of the need to care for individuals proceeding through such challenging experiences; and thus, contemporary hospice and palliative care were inspired. While the first hospices date back hundreds of years to religious organizations caring for the ill and dying, the foundation of the modern hospice movement is credited to Cicely Saunders and colleagues, with the opening of St. Christopher's Hospice in the United Kingdom in 1967 [32]. There, major clinical studies were undertaken to gather information about pain control and the chemistry behind it, providing the first research into palliative methodology. Research from St. Christopher's demonstrated the first empirical evidence recounting the prevalence of pain and suffering at the end of life, and it was this recognition that spawned the palliative care movement in the hospice setting. As a large proportion of St. Christopher's patients were afflicted by malignant disease, oncological care has deeply influenced the conceptual model for palliative medicine [32].

In descriptions of her experiences treating St. Christopher's patients at the end of life, Saunders outlined the idea of *total pain*, which she defined as the physical symptoms, mental distress, social problems, and emotional difficulties that accompany terminal cancer [33, 34]. The work of St. Christopher's hospice and others that followed

was aimed at alleviating this *total pain*. Saunders' original definition and goal is still echoed in the current WHO definition of palliative care.

Hospices proliferated throughout the United Kingdom during the 1970s and 1980s, funded largely by local charities, especially those concerned with caring for individuals with cancer [31]. Within the growing network of hospices, general practitioners (GPs) were largely responsible for the provision of palliative services. While committed to the philosophy and intent of palliative care, there was a distinct need for training in the new medical skills of care for the dying. The Association for Palliative Medicine of Great Britain and Ireland was formed in response, and strove to meet these needs while continuing to support the GPs in their provision of palliation. The United Kingdom thus became the first country to recognize palliative medicine as a medical specialty [1, 35].

The term *palliative medicine* itself was coined by Balfour Mount in 1974, in hopes of circumventing the negative connotations and terminal implications of the word *hospice* [34]. Mount forged a new model of care that combined the hospice services of St. Christopher's with the philosophy of American thanatology through a rich, collaborative dialogue with contemporaries Cicely Saunders and Elizabeth Kubler-Ross [1, 36]. He upheld the holistic approach of hospice, believing, "We're body, mind, and spirit. These are interdependent, and problems in each domain profoundly influence well-being in every other domain," but he adopted a secularized, integrated model to leverage the existing administrative and funding structures of the healthcare system [1, 37]. To the man who defined the term, palliative care was a combination of internal medicine and rehabilitation medicine accompanied by clinical pharmacology. A year after defining

*palliative medicine*, Mount opened the first palliative care unit (PCU) at McGill University in Montreal, Canada, which was followed shortly thereafter by a similar program at St. Boniface General Hospital in Winnipeg. These programs became models for the WHO and hospital PCUs across the globe [37]. The integration of the original PCUs into teaching hospitals, coupled with Mount's own strong ties to oncology, shifted the focus of the new *palliative care* from terminally ill cancer patients to much earlier points in the disease, significantly distinguishing it from hospice and providing the grounds for future broadening to earlier illness and non-cancer diagnoses. Shortly thereafter, Saunders and other UK leaders in the hospice movement agreed that the scope and nature of palliative care were distinct and defined the field; *hospice* was best reserved to describe the form of *palliative care* provided at the end of life [1, 36].

In the United States, the specialty of palliative care arose out of hospice and cancer programs in the mid-to-late 1980s. During the next decade, the Institute of Medicine called for the development of palliative care expertise to address the deficiencies in the healthcare system's approach to end-of-life care [38]. Soon afterwards, a model role for the physician in this new field was described [39]. By 1996, the number of hospitals supporting specialized palliative care teams had grown to 275 [40]. In 2006, hospice and palliative medicine became a recognized specialty by the American Association of Medical Colleges, and there now exist nearly 100 hospice and palliative medicine fellowship programs in the United States [40]. Today, the number of hospital-based palliative care programs in the US has increased to about 1300 [41].

Though Saunders, Mount, and other disciplinary founders were intentional in differentiating palliative care and hospice, distinction between these practices often

remain unclear even in today's medical facilities. In contexts where palliative care is a consultation or integrated service, it may be less associated with hospice or end-of-life care. However, in hospital settings, an inpatient palliative care unit often functions dually as a hospice, serving as a relocation destination for ICU patients in their last hours or days of life. This particular model is an ideal target for quality improvement research, as the patients and families following such a trajectory have little time to benefit from palliative efforts.

Despite repeated demonstrations of the benefits offered by palliative care to patients suffering from a variety of disorders, an Australian research team recently noted that provision of palliative services and referrals to specialist palliative care were disproportionately low [18]. In order to improve the recognition of patients' needs and to increase referral rates, Grigis *et al* developed the *Palliative Care Needs Assessment Guidelines* in 2006 [42]. The focus of this team was on patients suffering from cancer and associated complications. Subsequently, as the broader value of palliative care was recognized, a palliative care needs assessment tool for progressive disease (PC-NAT: PD) was developed. Because the PC-NAT: PD was created for patients suffering from any progressive disease, there is an opportunity for research to determine whether we can continue to improve access to and application of palliative care by informing needs assessments with disease-, illness-, or injury-specific components of suffering. That is, do patients with progressive neurologic, cardiovascular, or oncologic conditions experience unique forms of suffering, and might they subsequently benefit by condition-specific tailoring for assessment of their particular palliative care needs?

***Palliative Care in Neurology***

Today, palliative care is a common component of treatment for cancer patients [43]. Studies have demonstrated that patients with metastatic cancer receiving palliative care in addition to curative treatment have improved quality of life, decreased rates of depression, and increased median survival [23]. Such improvements in patient outcomes have helped popularized palliation as an element of care for patients with chronic and progressive diseases other than cancer. The palliative lens shifted to neurology in the late 1990's when the American Academy of Neurology's Ethics and Humanities Subcommittee stated that, due to the nature of neurologic illness and the patient's inevitable decline, the integration of palliative care into neurological clinics was essential [44].

A clearly identified need exists for incorporating palliative care into treatment plans for patients suffering from the progressive neurodegenerative disorder amyotrophic lateral sclerosis (ALS) [45, 46]. The often rapid decline and death of patients suffering from this disease coupled with the high symptom burden are classic indicators for the necessity of palliative care intervention for both patients and caregivers. Non-motor symptoms including pain, depression, existential fears, social isolation, fatigue, and constipation, amongst others, are large components of the patient's experience of neurologic illness, but may be overlooked by the treating neurologist in his effort to manage the primary diagnosis and other ALS-associated symptoms that appear to more obviously affect quality of life [47, 48]. However, there is significant evidence that, over time, non-motor symptoms are among the most function-limiting experiences for patients and affect caregiver burden and quality of life (for both patient and caregiver) more than

motor symptoms [49-51]. Thus, amelioration of these symptoms is an important component in completely addressing both patient and caregiver suffering. Since the potential for patients with neurologic disorders to benefit from palliative care was identified, increasing research has been undertaken with the aims of fulfilling unmet needs.

Recently, the integration of palliative care and neurology has led to the development of the nascent field of neuropalliative care. Neuropalliative care provides a palliative approach tailored to the unique needs of patients, families, and caretakers affected by chronic neurological conditions [52]. The identified amenability of the neurologist's clinic to function jointly with palliative care has been recognized and is the subject of developing academic and clinical research [53, 54].

The model for palliative care delivery in the context of progressive neurological disease may vary from other clinic organization in other specialties within the hospital setting. While in many contexts the neurologist functions as a consultant, he or she may be considered a primary care provider by patients suffering from ALS and other neurodegenerative disorders [55]. Thus, the neurologic specialist is not only responsible for managing symptoms directly related to neurologic disease progression; he or she is also tasked with aiding in the maintenance of quality of life. Despite this essential obligation, a survey of US neurologists suggested that many lacked knowledge about basic palliative care principles, despite the fact that much of the treatment for ALS can also be appropriately described as palliative in nature [56-58].

Since no therapy has been approved to slow, halt, or reverse the progression of ALS, all of the clinical interventions employed are targeted at preserving quality of life

and reducing the impact of motor symptoms. The poor prognoses associated with the disease have created a clinical environment especially sensitive to symptom management, and the practice—intentional or coincidental—of neuropalliative care [59].

To further explore current practice, let us consider the model of care delivery implemented by the Emory University ALS Center. Here, patients are typically seen once every six months during one of the multidisciplinary ALS clinic days. Following check in, a patient and his family are shown to a private room where they are visited by members of an interdisciplinary team of physicians, nurses, and other care providers. Amongst the day's consultations, patients are seen not only by their neurologist, but also by respiratory, occupational, and physical therapists, a nutritionist, a social worker, a speech pathologist, and members of support organizations like the Muscular Dystrophy and ALS Associations. Each of these consulting individuals is committed to both proactive and reactive amelioration of any suffering experienced by the patient, with an overarching aim of maintaining a high quality of life for the longest possible duration. Not only does this model constitute a palliative approach, it provides patients with high quality preventive palliative care. Much of the discussion within the clinic is focused on prophylactic measures to address symptoms that have not yet occurred, but are on the horizon.

### *Best Practices & Moral Obligations*

Closer examination of the practical role of the neurologist in the management of neurodegenerative disorders such as ALS prompts questions about the duties of the physician. Is providing palliative care a moral obligation for neurologists? How does the

provision of palliative services interface with legal and ethical mandates regarding quality of care?

In the context of this discussion, it may be reasonable to take the standard of care as a minimum obligation, both legally and morally. However, where the law does not set a mandatory upper bound, ethical ideals may dictate the minimal obligatory practices of good medicine. Unquestionably, physicians are legally obligated to practice in compliance with relevant professional organizations' practice guidelines. A physician deviating from the accepted standard of care, even with good intentions, may be held legally liable for negligence or malpractice.

One of Medicine's guiding moral responsibilities is the principle of nonmaleficience, or a physician's obligation to not cause unavoidable or unnecessary harm, pain, or suffering to his patients [9]. Morally relevant potential harms include both acts of commission and acts of omission. It seems clear that any provider, upon encountering a patient with disease-related suffering and identifiable need for palliative care, is morally obligated to assist the patient in obtaining access to necessary services. If no such effort is made, the physician allows harms (suffering) to persist that could otherwise be mitigated, thus violating duties of technical competence (in identifying palliative care needs) and professionalism (in providing or referring patients to palliative care services) [60]. Not only must the physician have the technical skills in his or her field of expertise to provide care that at least meets the minimum standards of practice, but he or she also ought be sufficiently aware and attentive to identify potential indicators of unmet palliative needs [17]. Subsequently, the tenet of non-abandonment obliges the provider, upon identifying a palliative need or suffering in one of its forms, to aid in

provision of supportive care. The secondary obligation engendered here may involve provision of palliative services by the physician him- or herself, conversation with the patient and family about the offerings of supportive care, and for referral to a specialist palliative care team.

The context of the patient encounter will substantially affect the associated obligations. Some subspecialty surgeons, for example, have minimal duties to provide palliative care due to practical limitations like the duration of the clinical encounter and primary obligations engendered by the expertise of the specialist and particular therapeutic purpose of the patient's visit. However, even the specialist must be aware of the factors that contribute to palliative needs for patients and families, be diligent in assessing these needs during personal contact with the patient and family, and provide referrals as necessary in recognition of the humanity and suffering of his patients. These are positive moral obligations placed on the physician beyond his legal duties to meet standards of care. In order to fulfill the tenets of a medical practice intended to care for and promote the wellbeing of whole persons, the physician must be sufficiently informed and sensitive to patient's non-physical needs, and he must be cooperative with palliative specialists. Failures to meet palliative needs in this area may not be legally enforceable, but the recognition and appropriate treatment of pain and suffering is certainly a moral mandate of the medical profession.

The obligation of physicians in the care for patients with life-threatening or life-limiting disease is substantially different when compared to conditions for which curative treatment exists. As stated previously, the role of the neurologist working with progressive neurodegenerative diseases often becomes one of primary care, as the patient

sees this specialist most frequently and relies on him or her for management of disease-related symptoms [55]. This provider's intimate knowledge and proximity to the patient should increase his duty to be sensitive to *total pain* and multidimensional suffering. Since there is no curative treatment to be offered, the neurologist has positive legal and moral obligations to palliation of symptoms. Here, palliative measures are the only recourse provided by standards of practice and provide the only manner in which a physician can treat his patient. Clearly, the physician is not obligated to individually providing each of the many facets of palliative care, but he must be responsible for ensuring his patients' needs are being met, and either contacting, coordinating, or cooperating with the palliative care team as appropriate.

These requirements function for a generalized patient group, but might physician obligations differ for individual patients? Rita Charon's work has emphasized the role of narrative competence in medical practice (described further in Chapter 2), and these approaches are particularly relevant in the context of life-threatening and life-limiting illness. However, it is far from possible for physicians to spend hours conducting thorough narrative interviews with each patient and caregiver to completely understand the individual's circumstances. He or she may, through careful listening and narrative consideration, divine particular needs derived from individual characteristics. But, it remains unreasonable to assume that heavily scheduled physicians, nurses, and medical providers can, in practice, tease apart individual narratives to assess nuanced palliative needs or ensure all needs are being met. To this end, tools have been developed to both screen for the necessity of palliative services and to monitor needs during care, though the available tools leave little room for narrative nuance, nor are they tailored to specific

diseases or the likely needs of specific patient populations. Thus, the proposed methodology may assist physicians in fulfilling their moral obligations to alleviate suffering by determining more patient-centered and disease-specific needs for palliative care.

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## Chapter 2: Ethics and Narrative in Palliative and Supportive Care

Finding literature that discusses palliative care ethics outside the context of end-of-life care is challenging. This may result from the co-evolution of palliative and hospice care as discussed in Chapter 1. Though non-hospice palliative care is now receiving increasing recognition, perhaps promoted by its rebranding as supportive care, palliative care is still predominantly utilized by patients receiving treatment for cancer. Studies of early intervention palliative and supportive care also predominantly occur in the cancer context [1]. However, according to 2014 National Vital Statistics data (most recent available), eight of the fifteen most common causes of death are non-cancerous chronic or degenerative diseases [2]. Just like those receiving supportive and palliative care for cancer prior to approaching a terminal prognosis, the large number of patients suffering from chronic and progressive non-malignant diseases are equally deserving of the supportive measures provided by a palliative care team, despite the fact that their prognoses may be substantially longer than those of advanced-stage cancer patients. In fact, one might argue that patients with chronic or progressive illnesses that are life-threatening or life-limiting have an increased need for supportive and palliative care, since the duration of their suffering has the potential to be much longer. However, non-cancer diagnoses are associated with reduced access to palliative care [3].

Increasing the focus on supportive and palliative care for non-cancerous chronic and progressive disease demands a novel examination of the ethics of palliative care in the non-hospice setting. When a patient's timeline for care is substantially extended beyond the months, weeks, or days typical of hospice patients, the traditional ethical

discussions that revolve around end-of-life decisions—including debate regarding double effect, opiate dosing, futility, and withholding or withdrawing life-sustaining therapies—do not contribute adequately to discussion of the ethics and moral assumptions of palliative care for the patient who has been diagnosed with a life-threatening or life-limiting illness, but who may not be imminently dying. Thus, a broader framework for the ethics of non-hospice palliative and supportive care is increasingly as much of the duration of this care will occur outside of the end-of-life context.

The primary moral assumption to be addressed is the meaning of *care*. Whereas the traditional medical model tends to portray good care as providing an accurate diagnosis and excellent treatment, palliative and supportive care have operationalized a distinct understanding of how to attend to patients, known as patient-centered caring. This method of service provision is based on recognition and deep respect for patients as individuals outside of the context of their illnesses—as people who operate in a social world, who are listened to, engaged, informed, and respected by their physicians [4-6]. Patient-centered caring understands a corresponding obligation for physicians to provide care that coheres with the patient's unique values and fits best with their individual circumstances.

Certainly, this type of recognition is not exclusive to best care practices in supportive and palliative contexts. However, in the case of life-threatening and life-limiting illness, they become increasingly important due to the intensive nature of medical interventions and the effects that serious illness can have on a patient's identity, as well as the identity and welfare of the patient's family [7]. As described by Eric Cassel, thorough healing occurs when an individual's well-being is restored through

reinstated health—fostered by good care in the diagnosis and treatment model—and a return of the self as a whole, functioning person, which may be fostered by patient-centered caring [8]. The foci and implicit ethical obligations of the patient-centered caring model clearly cohere with the holistic aims of palliative and supportive care. As much of life occurs embedded in social contexts outside of the clinic, and as suffering and spiritual distress have no medically defined etiology, the fields tasked with improving quality of life and preventing and relieving suffering in its many forms must understand the patient as a person and seek his or her deep involvement in his or her own care.

Associated with patient-centered care in palliative and other medical settings is a moral framework encompassed by Nel Noddings' "ethics of care". This system describes the role of a caregiver as being comprised of: "(1) attentiveness, an orientation toward being aware of the other person's need; (2) responsibility, a commitment to take care of that need; (3) competence, the capacity to provide good care; and (4) responsiveness, recognition of the unique perspective and position of the care receiver." [9, 10] The ethics of care is based on the caregiver's endeavor for an empathetic understanding of the illness experience of the one cared-for. This undertaking is narrative in nature, as it demands an intricate understanding of the patient's perspective and unique situation. An ethics of care, supplemented by narrative compassion, provides the foundation for a strong, trusting patient-provider relationship grounded in understanding and respect, necessary for the challenges of prolonged degenerative illness.

Though the necessary patient narrative may not encapsulate the entirety of an individual's experiences, the stories patients tell and the ways in which they are told can provide valuable insight into the nuances of a patient's worldview and understanding of his or her condition. Distinguished from an oral history characteristic of clinical encounters, an illness narrative is broader in scope and tends to provide more information about the patient's values, modes of reasoning, and the meaning of illness for the individual. Recognizing the value of this sort of knowledge both in and out of the medical context, a broad and incorporative field of narrative scholarship arose that privileged complete, first-hand stories about a challenging decision, event, social construct, or experience.

Narrative scholarship as a whole, including narrative ethics, is a relatively new approach being applied to academic work throughout many disciplines. Due to the novelty of narrative research and the diverse range of substrates to which it can be applied, there is no general consensus about what "narrative ethics" is or what it means to be "doing narrative ethics". The following section will describe the various approaches to conducting narrative work, focus on its role in medical practice and research, and argue for the particular appropriateness of narrative ethics and methods in palliative and supportive care.

### *Definition and Functions of Narrative Ethics in Medicine*

The most general justification for narrative ethics comes from a theme depicted by Kenneth Burke—specifically that we, as humans, use stories as "equipment for living" [11]. Narratives—composed of sequences of events over time, the agents involved, and

the connections between them—are epistemological tools by which we learn to feel and think and interact with society [12]. Such "equipment" works to shape our understanding of the world and the situations we encounter throughout our life. Stories enable us to grapple with "lived situations, celebrate…triumphs and encompass…tragedies" [13]. Thus, a discussion of narrative ethics is essential, as these lived stories have also taught us the "correct" and "appropriate" ways to interact with others [14].

In describing narrative ethics, Adam Newton outlines a bi-directional relationship between the literary and the philosophical. By tying them together with the term *narrative ethics,* Newton ascribes narrative structures ethical status while making apparent that ethical discourse depends on narrative structure [15]. In short, narrative ethics can be understood as the application of narrative concepts and methodology derived from the fields of philosophy and literary criticism to questions of moral understanding and evaluation [16]. These tools can be employed in a variety of contexts, all of which embody distinct facets of "doing narrative ethics". The applications of the field may be as diverse as literature and ethical inquiry themselves, but the focus of this discussion will be on the role of narrative in clinical research and biomedical ethics, further specified to care for chronic neurodegenerative disease. To begin to understand the influence the field of narrative ethics may exert in this context, Hilde Lidemann Nelson outlines five "things to do with stories" that have moral implications [17].

First, scholars like Martha Nussbaum and Rita Charon have described the benefits of reading, listening to, or viewing stories, proposing that when one attends carefully to the nuances and complexities of narrative in any of its forms, one's moral sensibilities are sharpened [18, 19]. This point builds upon the mimeticist argument that even fictional

narrative is representative of the structures of everyday experience [15]. In the words of Barbra Hardy, “The novel does not invent its structures, but heightens, isolates, and proceeds to analyze the narrative forms, methods and motions of perception and communication; sometimes explicitly, always implicitly, the novel is concerned to analyze the narrative forms of everyday life.” [20]

While Nussbaum focuses more generally on the use of literature in developing moral emotion, Charon’s methods are specifically applied to medical practice. In 2001, Charon published her first article on *narrative medicine*, a clinical practice fortified by *narrative competence*, or “the capacity to recognize, absorb, metabolize, interpret, and be moved by stories of illness.” [21] These abilities are developed by rigorous training in close reading, attentive listening, reflective writing, and by bearing witness to suffering, and they enable a more compassionate, humane, and effective practice of medicine [19, 21, 22]. Of course, narrative medicine is not equivalent to narrative ethics, but it represents some applications for the work of stories in moral education, which may extend to changes in practice and behaviors for physicians influenced by the methodology.

A second avenue for using narratives relies on their ability to assist one in making sense of complex experiences. In this capacity, one tells a story to make moral sense of an event by choosing particulars from the many details about the occurrence and reflecting upon them with respect to relevant moral ideas. To this effect, Margret Urban Walker describes how an individual can turn to his or her own narrative history to inform a challenging decision with evidence about what best reflects who he or she is and who

he or she wants to be. This reflective practice of narrative storytelling helps one develop a "strong moral self-definition" by which a given course of action can either be ratified or repudiated [23].

Similarly, Laurel Richardson recognizes stories as both a "mode of reasoning *and* a mode of representation" [24]. In this view, an individual can use his own story to reason through a personal decision or use his narratives to represent himself to others. Reasoning and representation are not limited to first-person applications, though. An individual can construct a narrative in which he imagines himself as the moral agent or subject and can then proceed to picture the outcomes of different decisions. In this way, narrative exercises foster both empathy and moral imagination by allowing decision makers to "try on" outcomes prior to issuing a final choice on a matter [25]. Martha Montello refers to this function as the "moral laboratory", in which agents can temporarily inhabit different lives [26]. In the medical context, a provider with sufficient narrative data may be able to employ moral imagination to immerse herself in the life, values, and worldview of her patient, thus fostering a practice of compassionate and patient-centered care.

The first person illness narrative is a primary example of sense making through story telling. Arthur Frank has described three primary structures of the illness narrative that, when listened to by healthcare professionals**,** assist in understanding the patient's experience and re-contextualizing suffering as a bond between patient and provider rather than an isolating force. These structures include (1) the restitution story in which the patient tells of getting sick, suffering, being treated, and being restored to health; (2) the chaos story where disability can only increase, pain will never remit, physicians are either unable to understand what is wrong or unable to treat it successfully; and (3) the quest

story, in which the patient understands illness as a condition from which something can be learned, and this learning can be passed on to others [27]. Narrative forms primarily function to help patients make sense of their own experiences—though they may not be constructed consciously—but, with careful attention, they can assist providers in understanding what gives meaning and value to a patient and, thereby, inform care providers about conflicting forces that may be causing difficulty for the patient [28].

The third role of stories is one common in medical ethics. Here, a particular situation is analyzed with respect to formal topics and its relation to paradigm cases. In a clinical case, for example, medical indications, patient preferences, context of care, and quality of life may be used to evaluate a patient's story, enabling one to note similarities and differences between the scenario under consideration and a paradigm case that guides moral action in "cases of this sort". One can then reason by analogy about the best course of action for the case at hand [29]. This approach, known as casuistry, holds that moral reasoning does not demand universal principles applied rigidly. Rather, ethical decisions occur most effectively through familiarity with intimate details of a given case and historical president in similar cases [30].

Fourth, one can perform literary analysis on stories by applying the tools of textual criticism to explicit narratives, 'stories people tell about their lives', or social practices that are treated as literary texts [31, 32]. Thus, literary scholars may contribute to ethical discourse by "uncovering moral meanings of health and illness, identifying what counts as a virtuous practice of medicine, noting how ethical responsibilities are

assigned within this practice…or attending to a host of other matters that bear directly on the moral values operating within medicine." [17]

Finally, the fifth "thing to do with stories" is invoke them to make a moral point. Fables, parables, and anecdotes have long been used to teach children "right and wrong" or provide warnings about the dangers of certain actions. Moral philosophers frequently construct stories about hypothetical situations in order to comment on the plausibility of a moral point. Paradigm cases of goodness and badness are often used to justify a given conclusion in both professional and non-professional reasoning [31]. In this way, bioethical and legal cases are invoked as setting precedent, providing concrete examples of moral norms applied to particular stories. Similarly, individuals employ stories to justify thinking and actions. Patients recount past experiences or the stories of friends to explain why they made particular treatment decisions, requested a specific physician, or avoided medical care altogether. In all of their forms these invoked narratives constitute an evidence base for both teaching and explaining moral decisions.

While Nelson's five applications of narrative toward moral work help establish a picture of how narrative ethics functions, they do not make central theses of the field obvious. Distilled, and framed in the context of moral questions in medicine, narrative ethics posits that (1) each moral situation is unique and cannot be completely addressed by abstract, law-like universal principles; (2) the appropriateness of any medical decision is contingent upon individual details and the fit of a given course of action within a patient's life story. The life story or stories of the patient form the basis for narrative

reflective equilibrium; and (3) moral justification by narrative reflective equilibrium is not conducted to reach a definitive correct ethical solution to a question, but rather to explore tensions between individual and shared meanings, open up dialogue, and challenge established norms. [16]

While discourse in disciplines ranging from sociology to anthropology to nursing is increasingly interested in topics like literature and medicine and the medical humanities, our lived experiences continue to add to our ongoing narratives of self and society. We continue to reflect upon these stories and use them to ascribe meaning to new or confusing information, and importantly, our stories and the empathy engendered by listening to them promote the formation of authentic connections between individuals.

### *Interactions between Principlist and Narrative Ethics*

Not unlike palliative and supportive care, the rise of narrative in medicine is understood partly as a reaction to the increasing prevalence of chronic disease and the subsequent change in the caring nature of the physician-patient relationship [7, 22, 33]. Thus, where the universal and abstract maxims of scientific or moral theory may have once been considered the optimal impartial companions of medical and ethical judgment, more nuanced approaches became necessary to manage chronic illnesses mired with heterogeneous presentations and caring interactions which attempted to address patients' non-medical lives. Patient narratives of illness, sometimes referred to as "lay narratives", gained prevalence in academic literature as the means to inform the individual considerations that were now inextricable from good biomedical care.

The new influence of narrative particulars was expressed not only by the increasing emphasis of care in the physician-patient relationship, but also by a shift in moral thinking. In a 1993 retrospective article for the *Journal of the American Medical Association*, Edmund Pellegrino argued that medical ethics was undergoing a shift analogous to the 1970s' transition from the "Hippocratic ethic," with its emphasis on duty and virtue, to that of Beauchamp and Childress' principlism, which was understood to be better suited for a more heterogeneous society. According to Pellegrino, medical ethics was then beginning a new metamorphosis to "anti-principlism" [34]. Novel approaches to questions of ethics in medicine included the ethics of care, feminist ethics, communitarian ethics, and narrative ethics, amongst others [22, 35].

These "anti-principlist" approaches to recurrent moral questions are not without their critics. Narrowing our scope back to narrative applications in medical ethics, casuistry has been both the predominant challenger to principlist approaches to ethics as well as the primary target of criticism [36]. Effectively, the tension between casuistry and principlism has centered amongst concerns of sensitivity to the particulars of a case, objectivity, and the ability to produce a defensible moral resolution. Beauchamp and Childress, for example, express concern about the assertion of casuistry that paradigm cases can inform moral judgments with their facts alone. These authors and others continue to argue that in order for cases to be recognized as analogous, they must be connected by a documented and morally relevant norm or maxim [30]. In determining "which maxim should rule the case and to what extent", a case may be subject to bias, and valuable moral information may be lost to potentially analogous cases [37].

Casuists and narrativists alike have critiqued the principlist approach to bioethics, citing its failures to account for appropriately nuanced application of moral theory to particular cases. Critics of Beauchamp and Childress have posited that a principlist approach provides no satisfactory resolution for situations in which two principles are in conflict with one another without adopting a narrative technique [38]. Arthur Kleinman argues that principled attempts

> to answer wholesale each and every one of the serious dilemmas faced by patients, families, and clinicians (and in a standardized manner, yet!) contain a dangerous hubris which falsifies the existential experience of illness as much as that of healing… they must recognize that human problems cannot be reduced to simplistic formulas and stereotyped manipulations that treat patients and their families as if they were overly rational mannequins. [39]

This assessment is a common criticism of the inability of various moral philosophies to account for irrationality and particulars of circumstance in ethical deliberation. As a critique of principlist ethics, it raises concerns regarding neglect of relationships, emotion, and deeply held belief. For Kleinman, addressing the experiential and personal components of illness is the moral core of medicine, especially in the context of chronic illness [39].

However, nearly twenty-five years after Pellegrino touted the paradigm shift to anti-principlism, it seems that bioethics has not abandoned a reliance upon principlism as a major mode of moral discourse. Rather, a sort of integrated approach that incorporates both narrative particularity and appeals to universal principles has taken root. This framework, formally termed by Rawls as “reflective equilibrium” operates on the thesis that ethical justification “occurs through a reflective testing of moral beliefs, moral principles, judgments, and theoretical postulates with the goal of making them coherent.”

[30] A decision or set of moral beliefs is then justified if it maximizes coherence of the relevant beliefs [40].

Even Rita Charon, champion of narrative medicine, does not understand principlist and narrative approaches to be incompatible. Rather, in her view they are complementary approaches [41]. Howard Brody argues that principlism has "irreducibly narrative roots" [42], and that, "rather than having narrative and nonnarrative ethics, we have ethics in which the use of narrative is explicit and ethics in which its use is implicit." [25] And McCarthy asserts that, "a 'good' principlist pays attention to the uniqueness of each moral situation and so, has narrativist tendencies, and a 'good' narrativist has a view to multiple stories and shared meanings and, so, is inclined toward principlism." [16] These symbiotic accounts of narrative and principlist ethics in the clinical setting provide the most functional model for applied clinical ethics. When utilized coherently, they provide medical professionals an ethical anchor in the form of easily learned tenets of moral practice while maintaining the moral importance of particulars.

Upon review of the aims and techniques of narrative in medicine, this approach seems particularly salient for research in palliative and supportive care. The holistic focus of both practices promotes care that is sensitive and compassionate to the individual experience of illness, and that generates moral considerations from within that framework. Thus, in an attempt to better understand the moral and experiential components of patients living with neurodegenerative disorders like ALS, narrative analysis is an appropriate research methodology. In the words of Howard Brody,

"Exactly how would one go about addressing matters of moral importance in one's life *other* than by means of narrative?" [25]

### *A Narrative Framework of Study*

This study will apply the lens of narrative ethics to examine a cohort of first person narratives about the experience of ALS. By studying these autobiographical accounts, sometimes called "narratives of witness", we may be able to elucidate palliative and supportive care needs of patients with ALS and, in doing so, may also begin to gain a better understanding of the moral framework(s) utilized by patients suffering from neurodegenerative disease [43]. The experimentally identified needs may take the form of unique ethical considerations embedded in narrative themes or experiential components common amongst patients, or they may be more clinical in their presentation as biological or psychological care needs. Thus, in this study, the narrative plays at least two roles. It is both the substrate—as analyzed textual data—and a descriptive tool used to explore the clinical and moral significance of findings.

Importantly, while the work presented in this study does engage in the comparison of stories, it must be distinguished from casuistry in that, here, the patient is the author of his own story, and paradigm cases are not used for analogous reasoning [43]. Certainly, the reasoning that takes place in medical practice may be similar to casuistic approaches as physicians compare details of a particular patient's case to generalized scientific and moral knowledge [44]. However, distinguishing these stories from those utilized in pure casuistic analysis or even in reflective equilibrium is the issue of authorship. Unlike presented or paradigm cases, illness narratives are jointly

constructed by the patient, the medical team, and the patient's family. Thus, the narrative approach employed here upholds the primacy of the patient's story, but also looks to represent the voices of those who, while not experiencing the disease directly, will be significantly impacted by the outcome of particular moral or medical resolutions [45, 46].

Methodologically, the analysis of patients' stories will borrow from the sociological approach of Grounded Theory in the formation of descriptive codes that are inductive, comparative, and iterative [47]. However, instead of attempting to generate pure theory about social interaction or influence, themes will be analyzed for practical content. The analysis seeks to address: What palliative care needs arise as themes in the data? Are these thematically expressed needs met or unmet by current clinical practice? Are these needs identified by current palliative care needs assessment tools? Further, do common themes of moral thought arise in these illness narratives? If this is the case, how might these narrative moral frameworks function in the setting of clinical neurology?

The operating moral hypothesis is that patient experiences expressed in narrative form contain information that is both medically and morally salient for the provision of patient-centered supportive and palliative care. By engaging patients in narrative research, we will discern whether there are unrecognized needs, moral frameworks, and modes of reasoning and valuing that arise distinctly in the context of the neurodegenerative illness ALS.

***Influence of Narrative Research on Clinical Care and Health Policy***

While the aims and findings of narrative research may be academically interesting, do these methods have application in the clinic? Do narrative practices transcend the limitations of clinical research and practice by healthcare providers? It seems that there are three loci of narrative interventions in the healthcare institutions: (1) clinical research, (2) medical education and individual practice, and (3) institutional policy.

Narrative research in the clinical setting is becoming increasingly common, especially in studies aimed to improve the quality of palliative care by deepening our understanding of the illness experience. Some of these investigations are descriptive, seeking primarily to increase sensitivity to the experiential nuances of serious illness. For example, studies have been conducted to investigate the impact of terminal illness in young adults on their parents, to examine the concept of hope for patients and caregivers at the end of life, or explain the day-to-day difficulties experienced by hospice nurses. [48-50]

In addition to descriptive narrative research, designed to sensitize readers to the many facets of the experience of illness and caregiving, a distinct branch of applied methodology has evolved toward a more clinical task. These studies apply the tools of qualitative analysis to health narratives to describe patient needs, barriers to optimal care, the successes and failures of medical interventions, and the lasting effects of medical treatment, and some suggest resolutions to identified issues [51-53]. Additionally, the potential for narrative acts themselves to have therapeutic effect has recently been described—specifically, storytelling has been shown to enhance self-management for

patients living with chronic disease as well as reduce pain and improve wellbeing in patients with cancer [54, 55]. These results cohere with the healing effects of storytelling theorized by narrative scholars like Frank and Kleinman.

It is important to note that, while seemingly similar, these narrative investigations are categorically distinct from patient reported outcomes (PROs) research. PROs are not narrative in nature and constitute only responses to discrete questions concerned with symptom presentation not knowable by an observer. For example, headache frequency and severity cannot be determined without patient input, but the responses to such prompts certainly do not constitute a story in the sense it is invoked by narrative ethics and research. The aims of PRO research are quantitative rather than qualitative, designed to investigate prevalence and severity of relevant symptoms numerically. [56]

As previously discussed, physicians and other healthcare providers can enhance their practice utilizing the skills of narrative competence outlined by Rita Charon. Narrative medicine involves not only engagement of individual providers, but has evolved into a unique component of medical education, to promote moral sensitivity and patient-centered caring in medicine. As more medical schools embrace these methodologies, the influence of narrative practice increases. Still, it remains limited in scope by practitioners' individual commitments.

Narrative research may change the practice habits of individual providers who engage with the literature or receive training in narrative medicine. But to what extent might this field of study exert influence on healthcare's institutional practices and policies?

Howard Brody posits that the primary institutional impact of narrative ethics is political. Namely, by bringing the multiple perspectives and stories required by good narrative practices to the fore, narrative ethics stands to democratize medical ethics. The reweighted privileging of experience and voice attributes expertise to those who may traditionally have been overlooked or unheard in deliberative processes [42].

Further, the role of narrative research in public health was actually recognized long before its influence began to gain traction in clinical medicine. Examples of narrative interviewing are prevalent in public and global health studies, as well as throughout the literature of medical sociology and anthropology. For example, one study used narrative interviewing to investigate a community's perspectives on breast cancer interventions in order to better understand the low rates of treatment amongst community members, then designed a new health promotion strategy to reflect the particular concerns reflected thematically in the narrative data [57]. Numerous other studies have followed this model, investigating barriers to care in a chosen population through narrative interview followed by amending interventional practices to meet the narratively elicited preferences of the population [58-61].

Though not always scholarly in nature, illness narratives in any medium can influence policy. Some accounts are directly targeted at politicians and certain pieces of legislation, as in the case of patient testimony or invoked patient stories in the law-making process. The prevalence and calculated use of illness narratives about a particular disease, illness, disorder, or other threat to health may influence budgetary decisions and legislation, thereby affecting the healthcare institutions from the top down [62]. Books like Atul Gawande's New York Times Best Seller, *Being Mortal: Medicine and what*

*matters in the end* and Joe Fins' *Rights Come to Mind: Brain injury, ethics, and the struggle for consciousness* are written for the public, physicians, and policy makers alike, and use intertwined personal, patient, and societal narratives in an effort to reshape thinking and direct policy and practice towards components of healthcare thought to be underappreciated and underserved [63, 64].

### *The Nature of Evidence in Supportive and Palliative Care*

The close relationship of biomedical science and medical practice has long emphasized the physical and easily measurable components of illness in both medical research and therapeutic care. Technological developments accompanied by mathematical models have made things like enzymatic deficiencies, hormone levels, and cell counts biological parameters that can be measured, statistically analyzed, and compared. This quantifiable physical data provides the grounds for a modern "evidence-based medicine". Biomedical science is the realm of definite, knowable, physical data. In these figures we can be confident (within a given interval), we can isolate variables, we can analyze and control for a given set of parameters. This methodology has allowed for incredible developments in medical technology and practice, accounting for the improvement and saving of innumerable lives. Those components of practice that fall outside the scope of empiric knowledge have been dubbed "the art of medicine" [65]. However, the functionality of this dualist perspective, wherein the social and experiential is distinct from the medical, has come under increasing scrutiny as healthcare has begun to expand its lens from disease-centered to person- and patient-centered practices.

Palliative care, in particular, requires a reach beyond the traditional definition of "evidence" and expands into the investigation of concepts classically deemed immeasurable by empirical science. In this field, concerned with addressing intangible symptoms like suffering and strained relationships, the nature and epistemological challenges of the subject require a less physical form of evidence. For example, there is no assay to quantify suffering, or even to distinguish it from pain. So, how do we collect and analyze data to provide appropriate evidence for an empirical practice of palliative

care if the experiences of interest are not solely physical and not currently measurable by biomedical means? How do we innovate in a field that often relies on interpersonal skill as much as technical prowess?

The current study explores the role of narrative research in augmenting the practice of palliative medicine. The medical and health policy literature is rife with calls for the development of more patient-centered methodologies and the necessity of increased evidence for palliative and supportive care intervention. In designing surveys, assays, and evaluative scales, the patient's voice has been lost, and patient-centeredness has become heuristic-centeredness. Numbers cannot express grief. The nature of a changing spousal relationship from lover to caretaker cannot be quantified. And we, the outside observer, cannot know the experiential facets of illness that most greatly affect our patients if they manifest in ways the biomedical establishment does not traditionally recognize as significant.

Employing narrative research to develop a new evidence base will allow for the design of more patient-centered practices in which needs have been elicited from those experiencing illness and its effects. These metrics have the potential to be more sensitive to actual patient needs and may allow us to assess the need for more integrated and holistic practices. Emphasizing and integrating this kind of information does not threaten the practice of traditional biomedical research, nor the good therapeutic care based on its discoveries. Rather, it serves as a supplement that may further enhance the medical establishment's ability to completely care for its patients, even where restorative technologies may fail to cure completely.

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## Chapter 3: Rationale and Methodology

### *Rationale*

Currently, the European Federation of Neurological Science task force recommends that palliative care measures should be implemented as soon as an ALS diagnosis is made, citing that early referral to palliative services before a patient reaches the terminal phase is essential for building relationships with palliative care staff members that will be beneficial for optimal end-of-life care [1]. However, there remains much discrepancy on the actual timing of palliative care referral, and no standard for the initiation of palliative care intervention exists internationally or in the United States [2-4]. In the context of palliative and supportive care in neurology, the budding field of *neuropalliative care,* scholars are still working to evaluate specific needs and to determine appropriate timing for intervention. In a recent review, Boersma and colleagues identified several unmet areas of research in the developing field of neuropalliative care that must be addressed, including patient-centered studies to identify and characterize palliative care needs in neurology and evaluate patient preferences for addressing these needs [5]. Similarly, a *Neurology Today* opinion piece by Gina Shaw provided the objective, "We want to be able to identify people who are no longer slowly progressing, but are hitting a stage of higher disease and symptom burden that would demand a palliative approach. If we start intervening early, we hope to have a higher impact..."[6]

Much of the current literature on palliative needs in ALS focuses on evidence-based guidelines for patients who have already reached the advanced stages, with discussion aimed primarily at addressing end-of-life needs that could be covered by

hospice, rather than a model of early-intervention, integrated palliative and supportive care. Studies summarizing palliative care intervention in ALS understand palliative care to shift focus from disease modification to symptom management [2, 3]. However, as previously discussed, this understanding is both inappropriate in terms of current definitions of palliative care and the integrated palliative/therapeutic approach. A recent comprehensive evidence-based review by Karam and colleagues provides substantial detail about the physical components of palliative care for patients with ALS, distinct from a focus on the end-of-life stage. Left out of this review, though, are the non-physical components of the palliative care model. Reference to depression, the only psychological symptom discussed, is limited to patients' experiences of pain [7]. An article on supportive care needs in ALS discussed prevalence of depression and anxiety and suggested that, since no ALS-specific management guidelines exist, in these symptoms "multidisciplinary management including psychological support, palliative care and physical therapy is recommended, along with standard drug treatments used in other diseases." [8] Neither spiritual nor relational suffering associated with the disease were mentioned, even in the context of "supportive care". Narrative information about the patient experience of illness may provide ALS-specific evidence regarding any desire for treatment of psychological symptoms of depression and anxiety, and of other concerns such as suffering, relational challenges, and spiritual distress.

A host of tools exist are regularly employed in research endeavors to quantitatively and qualitatively assess the efficacy of palliative care for patients with ALS, their families, and their caregivers [2]. A recent technical brief on currently available assessment tools in palliative care found "a paucity of tools to assess…cultural,

ethical and legal domains, and patient-reported experience" [9]. The same review suggested that caregivers completing palliative care needs assessment questionnaires felt the information gathered was pertinent to physicians but of little impact to patients or families [9]. In an effort to address these gaps and enhance access to palliative care by improving needs identification, it seems that the tools of narrative ethics are very well suited to help illuminate patient needs and expectations from the emerging field of neuropalliative care.

Recent studies from academic teams throughout Europe have suggested the necessity of establishing more patient-centered practices for palliative care and its evaluation in patients with neurodegenerative disorders, but little research has been published to this effect, especially within the specific contexts of ALS or the US healthcare system [3, 10, 11]. A team of researchers in Italy led by Claudia Borreani identified unmet needs of patients with multiple sclerosis (MS) and their caregivers through interviews. A large portion of these unmet needs were beyond the scope of traditional medical intervention, but rather were psychosocial in nature. The team went on to construct a home-based palliative care program on the basis of these findings, and is now in the process of assessing the efficacy of the program in a randomized controlled clinical trial [12]. A literature review indicates that no narrative work like that of Borreani *et al* has yet been published in the United States or in the context of the distinct motor neuron disease ALS. The narrative work demonstrated by the Italian team and proposed here provides a meaningful example of first-person experience informing clinical evaluation of patient needs and represents an opportunity to inform patient-

centered practices with patient voices, leading to the development of narratively-informed care programs.

***Methods***

*Sample and Recruitment:* This study has been deemed exempt by the Emory University Institutional Review Board. Adult patients of varying age diagnosed with ALS were recruited from the Emory ALS Center in Atlanta, Georgia. Ten (10) adult patients and accompanying adult family members participated. Participants ranged in disease progression and symptom burden as measured by the ALS Functional Rating Score Revised (ALSFRS-R). The ALSFRS-R is a validated test of global function for patients with ALS used by healthcare providers to monitor disease progression and as a standard outcome measure for clinical research. The scores range from 0 to 40—higher scores are associated with more retained function [13].

Eligible patients were identified by the multidisciplinary care team on the basis of:

- Age >18
- ALS diagnosis
- ALSFRS-R score
- Appointment date scheduled within the study period
- English-speaking and capable of autonomous response (verbal, written, or technologically assisted)
- Initial team assessment of amenability to participation.

While this pilot study was not intended to represent the full diversity of the ALS patient population, efforts were made to include patient participants representing diversity in age, age of onset, gender and ethnicity. Potential patient participants were first seen by care providers during their appointment at the multidisciplinary ALS Clinic Day at the ALS Center. The attending neurologist asked the patient if they were interested in speaking to "a student" (Kelsey Drewry [KD]) about a research project. In order to minimize any potential coercive influences, healthcare providers did not discuss any particulars of study participation with patients. If the patient agreed to speak with the student, KD met with the patient and family members to discuss the purpose and methods of the study. After this conversation, patients were left with an IRB-approved patient information sheet. After the clinical appointment was complete, KD met with potential participants again to discuss their interest in completing an interview and answer any questions regarding the study purpose and methodology.

*Interview Procedure:* Interviews were conducted the same day in private clinic rooms in the ALS Center. Before beginning the interview, the study purpose and interview format were reviewed with the participants. Any remaining questions about the study were answered. Participants were informed of their right to decline to answer any questions or withdraw from participation at any time. A waiver of signed consent under 45 CFR 46.117(1)(c)(1) was granted. Thus, the participant's consent for participation was indicated verbally and recoded as a consent form signed and dated by KD. If additional family member were participating, each participant was asked to participate in the verbal consent process.

Interviews lasted between 15 and 60 minutes. The interview itself was two-tiered. The first question, "Can you tell me about your experiences?" was the unstructured, open-ended question component. Following this, a semi-structured interview based on a modified form of the McGill Illness Narrative Interview was conducted [14]. The interview questions can be found in Appendix Document 2. Interviews were audio recorded for verbatim transcription.

*Data Analysis:* Grounded theory guided the analysis of interview transcripts [15]. Analyses were conducted by KD with consultation from advisor, Kathy Kinlaw, using QSR International's NVivo 10 qualitative analysis software. Coding was done line-by-line based on meaning and content. The coding process was open and iterative, using markers identified during analysis. Second level coding was used to group similar themes into categories [16]. Third level coding was then undertaken to produce domains comprised of similar categories [17]. Data analysis led to both explicitly stated palliative and supportive care needs as well as care needs deduced from ALS-related changes and experiences (physical, emotional/psychological, social/relational, and spiritual/existential). The distinct modes in which needs arose led to the grouping of domains into two distinct genres, "Expressed Needs" (explicit statements about care needs) and "Experienced & Observed Changes" (deduced care needs from reported experiential information).

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## Chapter 4: Results

### *Participant Characteristics*

Ten patient and family groups participated in interviews at the Emory ALS Center between February and April 2017. The average ALSFRS-R (ALS Functional Rating Score-Revised) for patients was 34 (range 27-44). Table 1 summarizes participant characteristics. Interviews lasted an average of 31 minutes (range 15-54).

**Table 1-** Characteristics of ten ALS patient and family/caregiver groups who participated in semi-structured illness narrative interviews.

| Characteristic | Sub-Characteristic | N (%) |
|---|---|---|
| Patient Gender | Male | 9 (90%) |
| | Female | 1 (10%) |
| ALSFRS-R* | | 34.4, 4.4 (27-44) |
| Non-Patient Participants | None | 3 (30%) |
| | Spouse | 5 (50%) |
| | Adult Child | 3 (30%) |
| | Close Friend | 1 (10%) |

*Mean, SD (range)

### *Elicited Themes*

Following thematic analysis of the ten interview transcripts, sixty-one themes grouped into two genres, six domains, and eighteen categories emerged (see Table 2a & b). The two genres that have been theorized, "Expressed Needs" and "Observed & Experienced Changes" denote two broad and distinct types of themes that arose from the data. The distinguishing characteristic between Genres 1 and 2 was the nature of reporting by the participant. Genre 1, "Expressed Needs" encompasses themes related to the necessary physical, psychosocial, spiritual, and medical needs of the patients, as well as any care desired that the patient was not already receiving (Table 2a). Themes in

Genre 1 represent care needs that were either explicitly stated as being helpful, being necessary, being desired, or having been sought out by the patient or participating family member(s). In contrast, Genre 2 comprises themes linked to the physical and psychosocial changes experienced by the patient, family, and caregivers as a result of ALS diagnosis and disease progression (Table 2b). Palliative and supportive care needs arising from Genre 2 were deduced during analysis and do not reflect directly stated patient or family member care needs or desires.

*Genre 1*

**Table 2a-** Genre 1, "Expressed Needs": Domains, Categories, and Themes

| Domain | Category | Theme | Frequency (N) |
|---|---|---|---|
| **Physical/ Cognitive** | Symptom Management | Physical, Occupational, & Respiratory Therapy | 7 |
| | | Medical & Assistive Devices | 8 |
| | | Weight & Nutrition Management | 4 |
| | | Pain Management | 2 |
| | | Pseudobulbar Affect Management | 2 |
| | Personal Care | Assistance with Dressing | 4 |
| | | Assistance with Personal Hygiene/Bathing | 5 |
| | | Assistance with Eating | 2 |
| | | Assistance with Toileting | 1 |
| | Mobility | Assistance Moving Around the Home | 2 |
| | | Transportation Assistance | 3 |
| | | Wheelchair Accessibility | 2 |
| **Psychosocial** | Psychological Wellbeing | Control Over or Intention to Improve Symptoms | 5 |
| | | Being Realistic/Accepting Disease | 4 |
| | | Hope for Cure | 4 |
| | | Opportunities to Participate in Research | 5 |
| | | Altruistic Actions | 5 |
| | | Positive Outlook/Humor | 7 |
| | | Making the Most Out of It | 8 |
| | | Dealing with Fear & Uncertainty | 6 |
| | | Dealing with Sadness & Depression | 8 |
| | | Planning for End of Life | 2 |
| | Relationships | Love & Support from Family | 10 |
| | | Assistance & Support from Friends | 7 |
| | | Support from ALS Community | 5 |
| **Spiritual** | Benefit from Faith | Support from Faith Community | 8 |
| | | Trust in Faith Practice for Positive Outcome | 6 |
| | Spiritual Support | Addressing Issues of Theodicy and the Question, "Why me?" | 5 |
| | | Addressing Loss of Faith | 1 |
| **Management of Care & Information** | Information | Honesty from Provider | 2 |
| | | Value of Information About Disease & Care | 8 |
| | Interventions & Assistance | Preventive Interventions | 3 |
| | | Assistance with Using Medical Devices | 2 |
| | Access & Quality | Concerns About Cost of Care | 2 |
| | | Concerns About Wasting Time & Money On Therapies | 2 |
| | | Desire for More Individualized Care | 2 |
| **Additional Care Desired** | Home Care | Increased Access to Home Health or Supportive Care | 4 |
| | | Increased Information about Home Health Care & Access | 2 |
| | Experimental/ Unproven Therapy | Complimentary and Alternative Medicine | 5 |
| | | Stem Cell Therapy | 1 |
| | | Medical Marijuana | 1 |
| | | Dietary Supplements | 3 |

*Genre 1- Physical/Cognitive Domain.* This domain contained three categories concerned with the patient's medical and non-medical care needs to manage the physical and cognitive symptoms of ALS, and the corresponding needs for assistance resulting from diminished physical capacities. Medical and assistive devices were mentioned most frequently. Eight of ten patients brought up the need for or use of assistive devices like walkers, wheelchairs, the Trilogy ventilator, and drugs like Riluzole and Nudexta. Seven patients discussed physical, occupational, and respiratory therapy; most found the therapeutic recommendations easy to incorporate into daily life, though some assistance was needed in learning to use assistive devices.

Two patients reported symptoms associated with pseudobulbar affect (PBA)—uncontrolled crying or laughter that may be inappropriate or disproportionate in a given social context [1]. Interestingly, the prevalence in this study's participants (N=2, 20%) coheres with the reported prevalence estimates for PBA in ALS of 9.4%-34.5% [2]. One patient had just begun taking the drug (Nudexta) to manage symptoms; the other reported no intervention, only embarrassment about her increased emotional displays.

Personal care and hygiene arose as a theme in half of the interviews (N=5). None of the patients expressed shame or embarrassment regarding their need for assistance with bathing, toileting, dressing, or feeding. The need for assistance ranged from minor aid such as buttoning shirts or adjusting collars to comprehensive care.

> *You know, now I'm completely dependent on somebody else to prepare my meals, to help me to bathe, to help me to, to, to toileting. I mean, it's, it's just like, I mean I'm totally dependent on somebody else… to drive me somewhere, get me dressed, undressed, put me to bed, you know all those things.* (Patient)

Issues of decreased mobility and the need for accessible personal and public spaces seemed to cause patients the most concern and frustration of topics in this first physical/cognitive domain. Many (N=7) expressed dissatisfaction with the feeling of a "loss of independence" or the new challenges associated with visiting public spaces or attending social events due to physical restrictions and inaccessibility of spaces to wheelchairs or walkers.

> *The most difficult thing is trying to go to family affairs and things like that where, where homes are not accessible to me. I can't get up stairs, uh, and most homes have 2 or 3 stairs to get into, or what have you, or they're bi-levels or tri-levels or what have you. And that…bothers me, you know because I really can't get into those places.* (Patient)

*Genre 1- Psychosocial Domain.* This domain consisted of the inter- and intra-personal factors that helped patients and family members live with ALS. The psychological factors discussed as being helpful to living with ALS centered around accepting the disease and being realistic about the implications of diagnosis, remaining positive and hopeful, and the importance of feeling some control in managing symptoms.

The importance of retaining a positive outlook arose in 70% of interviews. Many patients mentioned the importance of positivity in association with acknowledgment that sadness did occur, but returned to the idea of utilizing humor and positivity to cope with the challenges of ALS's physical symptoms. Similarly, the theme of "making the most of it"—continuing to live life as normally as possible—was addressed by 80% of patients. They expressed desires to continue with hobbies, traveling, daily activities, and "having fun". Several noted that these intentions were tenable with slight adaptations to the activities.

Very interestingly, half of patients emphasized the importance of altruistic action through participation in research and community outreach.

> *You know, information is a, is a big key to understanding what's going on. Not just for me - for the doctors and anybody else. And I, I just put it like this, if nothing else comes out of me having this condition, if I can help anybody else, um… I'm not helpless as long as I can say or do something that helps somebody in the future or helps somebody now.* (Patient)

Another patient described the thinking of himself and his wife:

> *Um, I think as we walk down this path, if we can help someone else, um, if it's to listen or talk or be encouraging or just be there, I mean um, even if it's a total stranger… as I said earlier, um, ALS is a community, but also a family.* (Patient)

Only two patients brought up planning for the end of life, but in those instances it was associated with a positive psychological effect as patients felt it would help reduce the burden on their family. Several patients (N=4) expressed concern for the psychological wellbeing of their family members, with and without reference to advanced care planning. Six patients expressed an obligation to show strength and leadership, to keep morale high and keep one's family happy, and to minimize burdensomeness. Depression was mentioned by four (4) participants in association with patients feeling like their family was suffering as a result of the patient's condition.

> *You really do see the pain in your family. Because it's something you're living with, and so you're writing a prime directive and things like that. And I feel like it's very difficult for your family members. And I can see where depression could set in, because it, you're at a point where you see that you're hurting your family. and sometimes you just want that to stop... I do feel that uh, you know, I don't want to hurt my family. Um but I, I see the joy that they have with me, so as long as I can be with them then I think I'm making them happy. That's what I want to do.* (Patient)

All patients discussed the necessity of support, both physical and psychological, from their immediate family. The value of assistance and emotional support from friends was emphasized in 70% of interviews. The ALS community, a category including both support groups and patients' friends with ALS, was also described as very helpful in half of interviews (it was not mentioned in the other 5).

Spiritual practices & faith communities were emphasized as being very supportive and helpful both psychologically and socially to patients and families in eight interviews.

> *One of my things I've said is, "How do people go through all of this and not know the Lord? How in the world do they make it?" And somebody said, "I don't think they do." 'Cause, I don't understand how you can go through this—in a way it's suffering—but how do you get through it without your faith in God? And I think it has got to be impossible or near impossible to just try to trudge through all of that and not have a support system and the family of believers to be there to support you through encouragement, and comfort during those times.* (Wife of Patient)

A patient reported,

> *You know, if I didn't have faith this, this thing could get you, have you thinking some strange thoughts. You know, because you go through this 24 hours a day, 7 days a week... It's a rough deal.* (Patient)

Interestingly, 5 patients specifically mentioned asking "Why me?" in the spiritual context. Some struggled with their faith, but found it restored, while one patient reported having lost his faith entirely due to his experience with ALS.

*Genre 1- Management of Care and Information Domain.* This domain dealt with the aspects of managed medical care that patients described as necessary and helpful. 80% of patients described information as being valuable and essential, and expressed gratefulness for the ability to have a large amount of information about the disease and its management, as well as the ability to have honest discussions with care providers.

Several participants (patients and family members, N=3) expressed appreciation for the timing of interventions, specifically citing the value of preventive and proactive care in maintaining the quality of life.

Two (2) patients expressed some concerns about their care. These patients raised concerns about the cost of care, a fear of wasting time and money on ineffective therapies, and perceived insufficient individualization of care. One (1) patient expressed frustration over his inability to receive care he desired due to the fact that his medical presentation did not meet the requirements for insurance reimbursement, despite the fact that he very much felt he needed care.

> *The system's kind of flawed I think because… it kind of covers a, a base or… it's like a blanket care system, it's not like individual. And I understand that that's almost financially not feasible, but I guess people who have the money, they can kind of tailor whatever kind of support they need. But when you don't have the money, you can only rely on what's available through the system which kind of blankets everybody… And, like for me, I could use some assistance at home, but because I still drive a car and I'm not home bound, then they're not willing to listen to me on that.* (Patient)

*Genre 1- Additional Care Desired Domain.* This domain centered around participant statements of desire for supportive or therapeutic care in addition to the services they were receiving at the Emory ALS Center. Increased access to home health care or general assistance at home was a prominent theme in this topic. Many patients also had strong interest in complimentary/alternative medicine, especially chiropractic treatment and acupuncture. Patients also expressed desire to try experimental or unproven therapies like stem cell treatments, using medical marijuana to ease pain, and any "nutrient or supplement that might help".

*Genre 2*

*Genre 2, "Observed & Experienced Changes"* comprises themes linked to the physical and psychosocial changes experienced by the patient, family, and caregivers as a result of ALS diagnosis and disease progression (Table 2b).

**Table 2b-** Genera 2, "Observed & Experienced Changes": Deduced Domains, Categories, and Themes.

| Domain | Category | Theme | Frequency (N) |
|---|---|---|---|
| **Physical** | Weakness | Upper Body Weakness | 8 |
| | | General Weakness & Tiredness | 5 |
| | Mobility | Trouble Walking & Falling | 8 |
| | | Change in Living Situation | 4 |
| | Communication | Changing Speech | 3 |
| | Nutrition | Weight Loss | 3 |
| **Psychosocial** | Major Perspective Change | Value of Life & Time | 9 |
| | | Change in Identity | 5 |
| | | Thinking About Death & Dying | 6 |
| | Response to Loss of Independence/Physical Abilities | Feeling Burdensome | 4 |
| | | Frustration | 4 |
| | | Concern for Family's Wellbeing | 6 |
| | Change in Family Dynamics | Shift from Carer/Provider to One Who is Taken Care Of | 5 |
| | | Increasing Spousal and Family Closeness | 8 |
| | Change in Social Interactions | Increased Empathy/Understanding for Others | 3 |
| | | Others Feeling Sorry for Patient | 5 |
| | | Loss of Friendships | 4 |
| | | Loss of Employment | 5 |

*Genre 2- Physical Domain*. This domain includes reported physical changes that affected the lived or observed experiences of patients and their family members. Figure 3 summarizes these results.

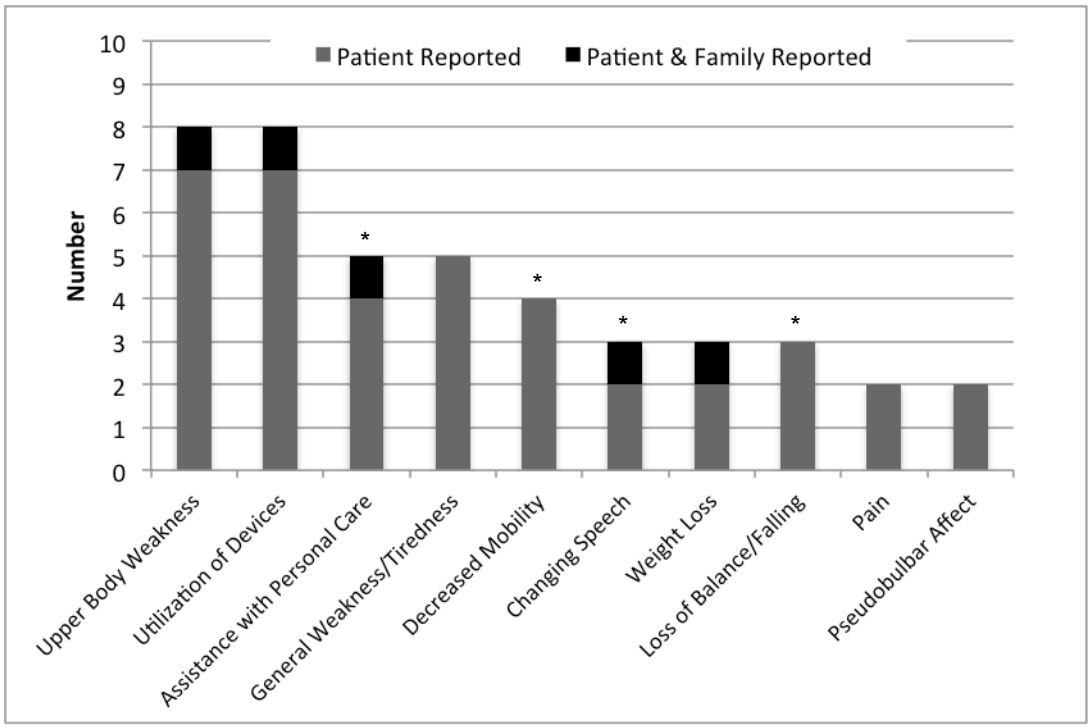


**Figure 3-** Prevalence of symptoms and physical changes reported by patients and family members affected by ALS during semi-structured illness narrative interview. * indicates symptoms monitored and reflected by the ALSFRS-R.

The most commonly noted physical changes experienced by patients were upper body weakness and reliance on medical and assistive devices. These themes arose in 80% of interviews, broached by both patient and family member participants.

> *I was doing things on my job like dropping tools and, and the weird thing about it is although I was losing strength in my hand, I didn't know I was losing strength. So when I th- when I, mentally I was holding something just like I always did, but physically I wasn't. I feel like I'm applying the same strength to hold it, but then I couldn't hold it… As far as the physical issues, there's all kinds of gadgets out there to help, and I think they're improving on them even as we speak.* (Patient)

Five patients addressed general weakness and tiredness. Three patients discussed weight loss, changing speech, and falls. Only two participants mentioned current pain and pseudobulbar affect symptoms (not related).

*Genre 2- Psychosocial Domain.* This domain comprised the psychological and social changes patients and their family members reported throughout the experience of ALS. The theme with the highest prevalence throughout all of the interviews and in both genres of data was the value of life and time. This theme was discussed in 9 interviews by both patient and family member participants ($N_{patients}$=9, $N_{family}$=3). It was reiterated several times in many of the interviews (N=4), arising in different contexts throughout.

> *That life is very precious and you need to enjoy everything, every moment. What can happen to the quantity and quality of life can be changed rapidly.* (Patient)

Another patient echoed this sentiment stating,

> *Life is precious. And that old cliché or saying, "Life is too short"? Guess what? It is. I mean, I, there are some days I can't imagine having more fun than I do that particular day. But, you know, God puts up with me and he allows me more time and fun and laughter. Good stuff.* (Patient)

Six patients discussed how ALS had affected their thinking about death and dying. A majority (N=5) did not express fear or discomfort with the idea of death, rather, they accepted it.

> *I just, just the fact that I've lived a good life and uh, I, if my life ended tomorrow, I wouldn't be disappointed. and hopefully when I do go, I'll go to a good place, wherever that place is, you know.* (Patient)

Three of the six patients broaching the topic of death and dying described finding comfort in the fact that they had "lived a good life." These patients tended to be older, and received their diagnosis later in life.

Five interviews reported a change in identity as a result of the disease. These changes were described by both patient and family member participants.

> *It's been, been a life-changing experience- mentally, physically, emotionally, everything.* (Wife of Patient)

> *I would say number one it's made me a different person, its opened my eyes to um, uh, really appreciate what you have.* (Patient)

Patients commonly reported negative psychological responses to their changing physical abilities. These included frustration with their inability to accomplish tasks or participate in activities they previously had. Frustration or some degree of psychological difficulty on the patient's part was also associated with the change in social role within the family caused by physical limitations. Patients felt that they were forced to abandon their role as a carer in the family and accept the role of the one-cared-for. While this seemed to cause distress, it was also associated with increasing spousal and family closeness in half of the interviews.

> *When you have a, a significant other that's used to you doing everything too, it's just as difficult for them to adjust as it is for me to adjust. Um, sometimes you would think I was just not doing things because I wanted her to do it for me. I'm like, "No, that's not the case." You know? She would say things like, "Well You just did that last week." I said, "Yeah, but I can't do it this week." And then two weeks later I was able to do it again- you know, those type of things. So she thought I was just acting like a baby sometimes, you know? I'm like, "No, I only ask for help because I need help." You know, so, um, once I started understanding how to adjust to it psychologically, it was better for me in my relationship.* (Patient)

Another patient stated,

> *I realized my wife became more friendly, more supportive. You know, more uh caring and loving and supporting. I thought it would have been opposite, but it did not.*

And a third patient reflected,

> *It's made me appreciate our relationship and for however long time we have together. Um, I know she's never come out and said, "Why don't you go to another part of the cabin or the house, you're bugging me." But I do sense, and sometimes I feel the need for more closeness.*

Social interactions outside of the family involved patients (N=3) reporting increased understanding of and empathy for others suffering from significant illness, as well as increased compassion for caregivers. Many (N=5) also reported negative social interactions, specifically their dislike for others "feeling sorry for me".

> *I mean, a lot of people feel sorry for you. I really don't like that, but I understand why they feel that way. And they, a lot of it's cause they care about you and they love you and, and uh there's some people who don't know you at all, and they, they still feel sorry for you. And uh that's, that's the part of it I don't like, you know? That doesn't help me for you to feel sorry for me. You know? Um, it's okay if you want to understand what I'm going through. But just to feel sorry for me? You know, that is not something I'm fond of.* (Patient)

Four patients reported friendships ending due to ALS. In some instances this was caused by their inability to participate in activities that had previously constituted the core of the relationship, some felt that friends distanced themselves because they didn't know how to respond or interact with the patient after diagnosis, and one patient reported that within his culture, significant disease and disability in men is viewed so negatively that his friends abandoned him.

### *Notable Language Use*

Though it did not arise as a deductive theme, the frequency with which patients and family member participants utilized plural and plural possessive pronouns (we, us, our) in reference to "their" diagnosis, medical care, participation in research, and other aspects of disease management was notable during analysis. Statements like "we were told we had ALS" (Patient, Interview 1), "we got a swallow study done" (Wife of Patient, Interview 2), and "our doctors" (Patient, Interview 9) occurred in 60% of interviews. The implications of this result for group autonomy will be explored in the discussion.

### *Intervention Areas*

Synthesizing these reported needs and experiential changes leads to a theoretical multidimensional approach to holistic management including medical care (to manage physical and cognitive symptoms, provide information and opportunities for patients to participate in research), psychosocial support (to address family needs, changing personal and familial roles, support patient and family psychological wellbeing, and to promote social programs like disease associations and support groups), domestic support (to provide access to home health and supportive care, assist with personal care and transport), administrative (to assist patients with receiving desired care and help with cost of care concerns), and public health and policy (to address issues of cost of care and eligibility for and access to services). These domains overlap to form a complex system of best care for patients, all encircled by public health and policy issues (not specifically addressed by this study). The theorized multidimensional approach resulting from the

findings of this study coheres with the literature's emphasis on best care for patients with ALS coming from a multidisciplinary clinic [3].

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## Chapter 5: Discussion

The aims of this study were to explore the palliative and supportive care needs of patients with ALS and their family members through illness narrative interview and thematic analysis. The semi-structured narrative interview methodology and qualitative analytics employed did give rise to thematically identified supportive and palliative care needs in this pilot study population. Further study participants would be needed to completely explore the effectiveness of this methodology. Recruitment of a broader range of participants would provide additional information about needs. For example, a revision of the interview format to include a written response modality could be considered to include the voices of patients who can no longer communicate verbally or have respiratory insufficiency that makes a long verbal interview exhausting. Despite these necessary improvements, narrative interview proved a valuable tool in eliciting supportive and palliative care needs.

### *Overview*

Participants in this study reported overall happiness with the care they were receiving. In fact, in 7 of the 10 interviews, patient and family member participants explicitly stated that they were very satisfied with the care and resources provided by the ALS center and associated organizations.

> *I'd say [the study facility] has played a big part. Um, it's hard to describe unless you've been there and to get this diagnosis… it's like, "oh my gosh, what do I do? Where do I go? What do I say? Who do I tell?" You feel completely lost. And coming here, I mean, I just can't imagine, and this is no lie, just because we're on tape and you're interviewing… but in my opinion, Emory is the tops. I would not go anywhere else unless Dr. A*

> *chased me out the door. Seriously, um, this diagnosis is a downer. But here, I feel a sense of hope.* (Patient, Interview 1)

> *I'm really happy with the care here at the clinic. And I probably shouldn't say care as much as I mean information. Because it has kept me ahead of the curve, uh, in terms of things that I need, everything that I needed, I have, I've had prior to getting to that point, that uh, that I had to have it… it's one of those things that, that was always brought to my attention, that you should be ahead of the game all the time. And quite frankly, the clinic, Dr. [B], Dr. [A] have kept me, you know, ahead of that.* (Patient, Interview 3)

The honesty and positive attitude of care providers coupled with the quantity and quality of information were cited as primary contributing factors to participants' satisfaction with care. When asked, participants could think of few unmet needs or additional care they desired, other than general hope for a cure. The unmet needs or desires that were expressed fell into the category of holistic supportive care and were not clinical in nature.

### *Methodological Limitations*

Because of the small sample size and single-center recruitment, these results may be taken as a positive indication of experiences of these specific patient participants with the care team, but no broad conclusions may be drawn about the quality of care provision at the ALS Center or by multidisciplinary ALS management in general. While a majority of participants were happy with their care, the effects of selection/participation bias must not be overlooked. On average, patient participants were not experiencing severe symptoms of ALS as reported by the ALSFRS-R scores, which averaged 34.4 on a scale of 0 to 40 (higher score associated with more retained function) [1]. This is not to diminish the challenging lived experiences of the patients interviewed, but rather to say

that those with greater progression may have additional explicit supportive and palliative care needs that are not represented by this study due to recruitment limitations.

The recruitment method, interview methodology, and setting may have discouraged patients with lower ALSFRS-R scores and higher symptom burden from participating. The verbal interview may have been exclusionary, too difficult, or too time intensive for patients with respiratory insufficiency or non-verbal communication modalities. It is possible that a written list of open-ended prompts and written responses may have provided a more inclusive means for these patients, though a questionnaire would have removed the possibility of dialogue, follow up questions, notation of and response to non-verbal cues, and limited the interaction between participants in group settings. A "take-home" written response would likely have garnered more participants of varied progression and ALSFRS-R score, as the timing and duration of interview (occurring after long clinical appointments) were also deterrents to patients who were approached but decided not to participate. Some were too tired after their appointment, while others expressed concern about the amount of time required for the interview and subsequent effects on their commute home.

The identification of potential patient participants by members of the clinic team did not seem to directly influence the pool of potential participants. However, it did seem that some patients understood the study to be concerned with quality assessment of specific practices at the clinic rather than academic in nature despite the patient information materials and discussions with interviewer KD. The assumed association between KD and the clinic may have affected the participant population by reducing the number of patient participants who were dissatisfied with their care, thereby excluding

potential valuable needs. For example, one potential participant told KD, “You don’t want to talk to me, I don’t have anything good to say [about my care]” (paraphrased). Thus, in future studies it would be essential to more clearly communicate that the researchers/interviewers are not associated with the clinic, hospital, or interested in quality assessment in any way. It seems very likely that patients who are incompletely satisfied with their care may have a greater number of unmet palliative care needs than those who are satisfied and subsequently eager to participate.

The limitations in recruitment and participation being noted, narrative interview was indeed useful in assessing these patients’ needs and learning about their experiences. The verbal narrative format allowed participants to structure responses freely and dictate the content and direction of the interview to a large extent. This flexibility enabled more detailed and person-centered responses than more structured modalities and grants an exceptional ability to understand the impacts of this disease in the life of the individual patient and his or her family. Additionally, many patients explicitly thanked interviewer KD for the opportunity to share their stories and experiences, and were appreciative of the person-centered nature of the study and its aims. These statements may be associated with the therapeutic nature of storytelling. Narrative expression has been argued to help bring meaning into one’s life, validate and reaffirm experiences, and connect one with oneself and others [2]. Storytelling in research may also foster a feeling of resilience in participants, as they are able to communicate and reflect upon their strength in dealing with challenging circumstances [3]. The opportunity to tell one’s narrative may also provide participants a chance to feel heard and individually valued in the clinical setting. One patient, unprompted, emphasized the importance of increasing the individualization

of care. He felt that because all ALS patients at the clinic receive similar services, his individual and unique needs were overlooked or unaddressed, either by clinic staff, practices, or health/insurance policy in general. This patient believed that narrative and person-centered practices both in research and medical practice would vastly improve the quality of care and illness experience for individual patients.

### *Expressed Needs*

*Additional Care.* Evidenced by the themes that arose during analysis, patients' medical needs for physical symptom management were well met, and participants were generally satisfied with their care. As stated, the most commonly referenced additional care desires were chiropractic treatment and acupuncture, home health care, and experimental therapies.

Chiropractics and acupuncture are categorized as *complimentary and alternative medicine* (CAM), or non-mainstream interventions used in addition to (complimentary) or in place of (alternative) standard medical treatment. Studies suggest that just over 50% of patients with ALS utilize CAM interventions, citing expectations of being cured (10%), improving their condition (30%), or slowing disease progression (50%) [4, 5]. Though chiropractic treatments and acupuncture are not currently discussed in therapeutic guidelines for the care of patients with ALS, they are undergoing evidence-based review to determine their appropriate roles in supportive care for patients with cancer and are offered as components of comprehensive treatment plans at some cancer centers [6, 7]. Acupuncture has been demonstrated to provide relief from cancer-related

pain and chemotherapy-related nausea and vomiting, but its role in symptom alleviation for ALS is not yet well understood [8]. Some studies have demonstrated putative benefits relating to reductions in pain and spasticity, while others have suggested improvements in motor function in both animal and human studies [9-11]. However, methodological errors and dubious experimental design afflict these publications, and independent evidentiary validation remains crucial before any evidence-based recommendations can be made regarding a therapeutic role for acupuncture in ALS [12].

Chiropractic care is utilized by cancer patients to help alleviate joint and muscle stiffness throughout the body [13]. Currently no evidence exists for the use of chiropractic treatment for patients with ALS, but there is evidence for its efficacy in treating chronic spinal pain leading some to recommend its use for symptom relief in multiple sclerosis [14]. It is possible that a similar role may exist for symptom management in ALS.

The relationship between CAM therapies and traditional western biomedical intervention has been the subject of both scrutiny and change in recent years. Academic medicine has seemingly abandoned the oppositional stance—in which providers were professionally obligated to oppose utilization of CAM—to a more integrated approach. However, as argued by Kaptchuk and Miller, an integrative approach may be inappropriate due to discordant philosophies and ethos in biomedical and CAM fields [15]. The authors continue to argue for a pluralistic approach, which "recognizes unbridgeable epistemological differences in the methods of developing medical knowledge and validating treatments, but acknowledges that both mainstream medicine and CAM can offer clinically valuable treatment options for patients in the light of

informed choices based on their preferences and values" [15]. Assuming this unified perspective on the relationship between the two disciplines allows providers to conduct more open conversations about the potential benefits of complimentary therapies with patients, coherent with the aims of a narrative framework of care. Additionally, conducting these conversations in a manner that does not seek to impose the ethos of the biomedical model may allow patients to more effectively convey their values and reasoning to the provider, thus providing the necessary narrative information for the physician to enter the "moral laboratory" and engage in more empathetic moral reasoning when making recommendations about complimentary therapies. The appropriate provision of information to patients regarding access of desired CAM interventions like chiropractic care and acupuncture may address one of the expressed patient needs identified in this study. While no evidence currently exists demonstrating therapeutic benefit from CAM treatment of ALS, utilizing these services may provide the patient a means to feel they are "doing something" to manage their condition—a theme that emerged as a helpful psychological tool for coping with the disease.

Further exploring the role of CAM treatments in the care of patients with ALS would require studies to evaluate any potential physical and/or psychological benefit. However, in the current absence of supporting data, social questions arise regarding the recommendation of complimentary interventions. Beyond concerns about direct (albeit inadvertent) physical harms to the patient that may result from these therapeutic attempts, issues of fairness and resource allocation must be considered. First, what is the appropriate way for insurance to address reimbursement for interventions with no current evidence base? While one can certainly argue for the importance of the potential for

psychological, if not physical, benefit from CAM interventions for patients with ALS, it is not obvious if this is a just allocation of limited insurance funds that may otherwise subsidize treatments with a more robust evidence base. If insurance coverage is not widely available for CAM interventions, is it appropriate for providers to recommend or discuss these treatments with patients who may or may not be able to afford it? There is potential for such discussions to increase finance-based stresses by presenting uncertain and perhaps untenable hope, an outcome that opposes the palliative goals of minimizing disease-based hardships. However, a provider declining to discuss these avenues with his patient may experience distress at what he or she may feel is incomplete honesty in communication. Here, a potential role of narrative knowledge may be seen. Though it is unreasonable to expect insurance to cover or not cover CAM therapies on an individual basis, a provider with good narrative knowledge of his patient's values, goals, and modes of reasoning may be able to make well-reasoned decisions about the extent to which discussion/recommendation of CAM modalities is ethical.

Home health care is an established component of care for individuals with some disability due to disease or dysfunction and is a Medicare-funded benefit [16]. While a few participants of this study reported experience with home health care, several desired more information or access to home health services for which they could not currently receive reimbursement. An administration-level intervention to work with patients and assist with understanding the nature of home care, services provided, and qualifying conditions for insurance coverage may be beneficial. Social work services may also be of value toward meeting these needs.

During interviews some patients reported frustration with needing assistance at home but being ineligible for any insurance benefit because they could still preform certain disqualifying functions, such as driving. A more complete understanding of specific home health care needs for patients with ALS is necessary, and may be a valuable avenue of future research aimed at improving the patient experience of neurodegenerative disease. Particularly, understanding the nature of "effective" home health care as well as its effects on patient and family quality of life and social/relational stress will contribute to more thorough care in this area. It may be of additional interest to investigate any potential relationship between receipt of home health care and the need for familial counseling, as one may speculate that much household tension may be related to an unmet need for home-based care. Ultimately, changing access to home health care for the ALS patient population would require evidence-based intervention at the policy level, and may necessitate specific provisions for ALS patients in recognition of the unique impacts and needs associated with the disease.

The expressed desire to utilize experimental and/or unproven therapies must be addressed with great care by a clinician, due to the potential for negative impact on the patient's health and disease management. Again, information and patient-provider communication plays an essential role. Evidenced by the themes of participant appreciation for information and honest discussion with healthcare providers, the ability to discuss patients' interest in experimental treatment should not be a barrier. However, it is important to acknowledge the unintended moral consequences of physicians' decisions regarding which studies to address with patients—disclosure or non-disclosure of

information about developing experimental therapies may enable or inhibit patient decision-making [17].

Similarly, provider and federal regulatory considerations of compassionate use of experimental therapies may also substantially alter the decisions available to a patient [18]. Ethical tensions here must be considered as there are strong implications for the wellbeing of individual patients receiving drugs through compassionate use policies, but making access too broad may obstruct the collection of meaningful evidence regarding an experimental therapy's efficacy.

*Altruistic Action.* The finding that half of patients interviewed expressed altruistic action as an essential part of coping with their illness is very interesting, especially within the narrative framework of this study. Some patients explicitly stated interest in participating in ALS-related research in order to help others with the disease at a future date, while other participants altruistic aims were more open-ended and general. This suggests an important mode of reasoning and valuing present within the participant population. Whether the desire to act altruistically is directly associated with the experience of severe illness is a question not addressed by this study, though a fascinating avenue of future bioethical research.

As discussed in Chapter 2, one of Arthur Frank's theorized illness narrative structures is the *quest story* in which the patient frames his illness as an experience from which something may be learned, and that learning may be passed on to others [19]. It seems that many ALS patients and caregivers may frame their experiences in this way, and even find doing so helpful. Accompanied by Richardson's postulation that stories

may serve as both a "mode of reasoning and a mode of representation," patients may tell their illness narrative as a quest story to help bring meaning to the challenges they face, which may otherwise be very difficult to justify.

### *Observed Needs*

*Psychological.* Though patients never explicitly stated a desire or need for professional help to manage emotional/psychological or social/relational challenges, the greatest observed unmet needs appeared to exist in the psychosocial domain. Several patients mentioned struggling with depressive symptoms or displayed depressive symptoms during the course of the interview. One patient, when responding to the question, "Has this [disease] changed the way you think or feel about yourself?" began to sob and stated, "I feel worthless." Others mentioned the potential to be depressed, or "I could see how that could get you down," though they did not specifically say they were experiencing depressive symptoms themselves. None of the participants mentioned mental health services during interviews. Coupled with the finding that patients with motor neuron disease—the neurodegenerative disease category of which ALS is a subtype—experience more hopelessness, demoralization, and suicidal ideation than patients suffering from metastatic cancer, these reports and observations merit significant consideration [20].

Deepening our understanding of the nature and effects of depression, anxiety, and psychological needs on the patient and caregiver experience of ALS is an important aim for future research. Of particular interest are the effects that result from improving the availability and utilization of mental health interventions. Specifically, if issues like depression are managed more successfully, what are the effects on the patient and family

experiences? Is there any change in decision-making trends, expressed moral frameworks or values, or shared narratives for patients whose mental health needs are addressed? Are there associated improvements in prognosis, social health, or overall reduction in the amount of suffering experienced by patients and their family members? If managing emotional/psychological needs demonstrates functionality in improving patient and caregiver experience and outcomes, investigation into potential prophylactic measures to circumvent or proactively address factors that may lead to psychological distress would be of great interest.

In addition to the queries above, research into the impacts of social norms or constructs (i.e. stigma, shame, etc.) on patients' interest or reporting of mental health care needs in the context of neurologic illness would be fruitful. This trajectory of inquiry may also help to inform considerations about how care teams may best address the potential need for psychological support for patients who do not explicitly ask for such care. Treating these sensitive issues with consideration and compassion is essential to minimize their deleterious effects on patient and family wellbeing.

In addition to noting the increased incidence of psychological distress, the study conducted by Clarke and colleagues also suggested that feelings of being "useless" or a "burden" are more common in patients with neurologic disorders due to the specific physical and cognitive disabilities associated with neurologic diseases [20]. Self-perceptions of uselessness and burdensomeness were stated verbatim during patient responses in this study, corroborating the findings of Clake *et al* while simultaneously demonstrating the utility of narrative methodology to elicit important psychological

symptoms. These results further evidence the necessity of improved recognition and management of mental health needs for patients with ALS.

*Spiritual.* Of the eight (8) interviews in which faith was discussed, spiritual practices were reported to be essential or incredibly helpful in coping with ALS in seven. Participants reported both psychological support from their spiritual beliefs as well as social support from their faith community. These results cohere with the findings of researchers such as sociologist Ellen Idler and psychiatrist Harold G. Koenig, whose findings indicate that religious beliefs and institutions help individuals face serious illness and death [21-26]. Correspondingly, the work O'Brien *et al* suggests that spiritual beliefs help sustain patients with ALS/MND and aid both patients and family members in making sense of their experiences [27].

A majority of the patients reporting spiritual beliefs had struggled with the question, "Why me?" That is, participants could not understand what the karmic or spiritual rationale was for the extreme hardship they were granted alongside the diagnosis of ALS. These reports suggest potential benefit from consultation with a chaplain, as this interaction may help patients and family members come to a better understanding of the intersection of their faith and lived experiences.

Only one patient reported losing his faith completely as a result of ALS. He was incapable of rectifying his diagnosis and other unassociated negative life experiences with his spiritual beliefs, and thus gave them up altogether. Earlier availability of spiritual support services may have been particularly helpful in this case and others like it.

Further narrative research may be useful in exploring the degree to which patients and family members feel spiritual or religious issues affect their quality of life. While Idler and Koenig have both shown that faith is helpful in dealing with serious illness, what effect does spiritual distress exert on other domains of physical and psychosocial health? It seems possible that difficulties with one's faith, spiritual, or religious beliefs may be related to other psychological symptom presentation. Further, it would be interesting to explore the ways in which patients' religious beliefs and practices (or lack thereof) affect their medical decision-making and means of moral reasoning throughout illness.

*Social.* Social challenges reported by participants occurred in both the familial and non-familial settings. Within the family, patients expressed frustration or sadness associated with changing familial roles and the necessity of relying on family member caregivers to perform basic tasks and activities. The manner in which these experiences were described suggested that the shift from carer to care-receiver might be particularly distressing for patients. Further narrative investigation into the experience of changing familial and social roles and its association with identity construction (and damage) would be of great interest. Group or family counseling may serve as a beneficial supportive care intervention for patients and families experiencing challenges related to changing social roles.

The extent to which issues of intra-family stress due to role reversal occurs is unknown and may be small, as all patients (N=10) interviewed in this study expressed receiving meaningful and necessary support from their immediate family. While it is

possible that the ubiquity of expressed appreciation and necessity of family member help may have resulted from family member presence during some interviews, this theme arose even in interviews where no additional participants were present (N=3). Patients reported discussions of compassion for the caregiver occurring in support group settings provided by the ALS Association (ALSA), and emphasized the importance of this practice. Referral to programs like ALSA or similar support groups may assist in promoting healthy intra-family interactions and reducing psychosocial burden for patients and caregivers.

Non-familial social interactions were reported as beneficial and helpful in seven (7) interviews. While this held true for close friends, five (5) patients cited frustration with responses to their illness from acquaintances, distant family, and the general public. These participants discussed experiencing extreme dislike for instances in which others expressed feeling sorry for the patient, stared, questioned the status or reality of patients' disability, or abandoned their friendship entirely.

One is left to wonder why these distressing responses are so prominent. It may be that individuals responding in this manner are either uncomfortable with (in)obvious manifestations of severe illness, or they may simply be uncertain about how to respond to a friend, acquaintance, or stranger who is experiencing significant illness. Addressing these issues is incredibly challenging and must largely take place on the public health level of intervention. Campaigns to promote awareness and sensitivity to disability (visible or not) and increasing discussion of how to interact with individuals who are or become disabled in an empathetic manner may improve patients' social experiences.

Undoubtedly, these issues will persist to some degree and, unfortunately, in extreme cases like those experienced by one participant, may be exceedingly difficult to address due to cultural barriers. A recent review of palliative care assessment tools found no tools that adequately addressed supportive care needs arising out of particular cultural practices or stigmas (for example, an association of illness with weakness), an area in need of continued research [28].

Palliative care guidelines for neurology suggest that neurologists must have fundamental proficiency with the palliative care skills of communicating bad news, assessing and managing non-motor symptoms, assessing caregivers, and end of life care planning. Referral to specialist palliative care is recommended for more complex issues, including spiritual and psychological care [29]. However, palliative care needs assessment heuristics for spiritual/existential care are currently very limited and provide a much-needed avenue for future research [28]. It seems that in order to improve care and more fully support patients through the progression of ALS, increased access to the psychosocial services provided by a palliative care team, ethics consultation, social work, and/or psychological care is necessary, even for patients and caregivers with relatively low symptom burden (as reported by the ALSFRS-R).

***Autonomy and Decision-Making***

None of the patient participants taking part in the study had diagnosis of comorbid dementia or similar cognitive disorder that could have affected their decision-making capacity. Following accepted ethical and legal guidelines, these patients are entitled to

make autonomous and independent medical decisions [30]. However, the frequency of plural and plural possessive pronoun use by patients to discuss medical and illness experiences as occurring to the patient and spouse together suggests that individual autonomy may be overly narrow in the context of ALS. Organic utilization of phrases like “we have ALS” and “our doctors” by patients and their spouses suggest that they do not view decision making or even the disease experience as occurring to themselves exclusively.

Palliative care and ethics consultation services extend care and consideration to family members and caregivers in a way that appears coherent with these findings. It may be the case that in the context of ALS the provisions of informed consent ought to be adapted to fit with innately expressed patient/spouse joint autonomy in order to respect both the patient-depicted joint experience of disease and medical care as well as the large impact that caregiving has on the spouse. Of course, this would demand a much larger evidence base and substantial multidisciplinary deliberation before any recommendation could be made to this effect.

Jointly conducted informed consent or adapted communication practices to respect joint autonomy may be very challenging to institute and employ. A robust cadre of studies, based in both narrative and other methodologies, would be necessary to suggest who ought to be involved in joint decision-making endeavors and the role of each participating party (i.e. is decision-making democratic or are the voices of certain decision-makers privileged?). Surely, issues of spouse disagreement or instances of parties struggling to accept the implications of a diagnosis or course of care could significantly impede care delivery. It seems unlikely that a single care provider could

independently advise and manage medical decision making for a patient if multi-party medical autonomy were instituted—this poses a potential role for the consistent involvement of an ethics consultant or committee to assist in facilitating group decisions. Further investigation including rigorous ethical analysis, patient and health provider interview, and outcomes-based pilot studies are required before any recommendation can be made on the topic of adapted multi-party autonomy.

***Methodology***

The methodology of narrative interviewing to elicit palliative and supportive care needs for patients suffering from ALS demonstrated utility. The first interview question, "Can you tell me about your experience?" coupled with follow-up and clarifying questions by the interviewer was successful at eliciting patients' illness narratives, though the stories tended to be slightly disjointed with respect to chronology. Participation of family members contributed both positively and negatively to the chronology of the illness narrative, as they would sometimes interrupt with details from another point in the story or help a patient to remember dates and sequences of events more accurately. Family member participation may have precluded some details of familial interaction from being shared by the patient due to concerns for the feelings of other participants present. However, it offered a unique dually constructed narrative that accounted for both patient and caregiver experiences simultaneously. The completeness and integrity of the narratives may be improved by conducting interviews with patient and family members together and separately in order to provide space for each to speak privately with the interviewer.

The semi-structured component of the interview functioned well to fill out participants' narratives by focusing on aspects of the illness experience that were sometimes overlooked. It is of note that many patients addressed each of the topics covered by the semi-structured section independently during the first open-ended question.

Together, the open-ended and semi-structured interview components successfully provided information about the meaning of illness in one's life, modes of reasoning about illness, and values of both patient and caregiver participants. Assessing patient needs in this way avoids the imposition of an extrinsic value system that may incorrectly prioritize the aspects of illness experience and their need to be addressed. Allowing these themes to arise organically through a guided conversation allows participants to prioritize care needs and values that might otherwise be overlooked by researchers who are unfamiliar with the firsthand experience of neurodegenerative illness and caretaking. Narrative interview methodology like that utilized in this study also shifts the balance of power between the researcher and participant, as the participant is able to freely dictate the content of their interview (through both inclusion and exclusion). In more traditional survey studies, the researcher narrowly defines what is important to the study and it may be difficult for participants to convey additional information they feel is important.

While directly reported unmet needs were low, likely due in part to the relatively early stages of disease in participating patients and the broad spectrum of care offered by the multidisciplinary clinic at the participating ALS center, unmet needs were identified nevertheless. Multi-level interventions based on the findings may be necessary to address the care needs deduced from thematic analysis of the interviews. Many of the

interventions fell under the purview of supportive and palliative care, while others would be best addressed by non-neurologic medical specialties, ethics services, community resources, administration, or public health. The finding of unmet palliative and supportive care needs in this study's small, relatively early stage ALS patient cohort suggests the importance of early-intervention palliative and supportive care for the ALS population. Expanded study of similar methodology and outcomes research is necessary to provide an evidence base for these putative conclusions.

Studies assessing palliative and supportive care needs and outcomes may be particularly amenable to illness narrative research. The goal of these fields is to ameliorate suffering that occurs in conjugation with severe illness, but the meaning, nature, and alleviation of suffering is incredibly personal and varies on the basis of a host of non-medical factors. Narrative methodologies, such as the one tested here, provide a sensitive and person-centered technique to evaluate needs and outcomes in a way that is meaningful to participants. Unlike other methodologies, narrative interview does not reduce the participant's experience to numerically scaled or short-answer responses. Instead, it allows patients and caregivers to structure the interview in the way they feel is most meaningful and accurate. Doing so may provide unique insights to research teams and novel perspectives for continuing research while more effectively addressing the complex dimensions of severe illness necessary for effective palliative and supportive care.

Though narrative based research as conducted here faces significant practical limitations due to the necessary time and personnel resources required for study, it may provide unique benefits both methodologically and ethically. Unlike non-narrative

methods of needs assessment research, patient and caregiver participants may themselves derive benefit from participation due to feelings of being heard and valued as an individual, and having an opportunity to share their narrative in the clinical context. These potential benefits make illness narrative based research distinctive due to the value and viability of results and simultaneous participant benefit.

### *Conclusion*

This study conducted ten semi-structured illness narrative interviews of patients with ALS and their family members then utilized thematic analysis of interviews to identify unmet palliative and supportive care needs. Though the interview format, timing, and setting require adaptation to improve recruitment and inclusivity of more severely affected patients, the method did elicit important themes from participating individuals. Explicitly stated and deductively determined supportive and palliative care needs were revealed through interviewing and analysis, demonstrating distinctive care needs rooted in the challenging and diverse presentation of ALS and its symptoms. Additionally, special considerations may be necessary to acknowledge patient-spouse joint autonomy constructed in the context of the ALS experience. However, further study is necessary to completely examine this phenomenon. The deduction of supportive and palliative care needs in the participating population emphasizes the importance of early intervention palliative and supportive care for patients with ALS.

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# Appendix

## *Illness Narrative Interview Questions*

*Part I- Unstructured*

1. Can you tell me about your experiences?

*Part II- Semi-Structured*

The following questions are adapted from the McGill Illness Narrative Interview (MINI): *Generic Version for Disease, Illness or Symptom [1]*.

2. When did you experience symptoms or difficulties for the first time? [*Let the narrative go on as long as possible, with only simple prompting by asking*, 'What happened then? And then?']

3. Other than physicians, did you see a helper or a healer of any kind? If so, tell us about your visit and what happened afterwards.

4. Have you had any tests (diagnostic, therapeutic, other) for you ALS? Have you participated in any studies?

5. Has the [ALS center] clinic team (or other healer[s]) give you any recommendations to follow?

8. How are you dealing with each of these recommendations? Are they easy for you to follow?

9. Are there any tests, interventions, or care you expected to receive [for ALS] that you did not receive?

13. Have you sought any help or care outside of this clinic?

14. Is there any help or care that you would like to receive that you do not currently receive?

15. How has this [ALS] changed the way you live?

16. Has it [ALS] changed the way you feel or think about yourself?

17. Has it [ALS] changed the way you look at life in general?

18. Has it [ALS] changed the way that others look at you?

19. How have your relationships to your family members changed?

20. What has helped you through this period in your life?

21. How have your family or friends helped you through this difficult period of your life?

22. How has your spiritual life, faith or religious practice helped you go through this difficult period of your life?

23. Is there anything else you would like to add?

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