

Original Article

Cite this article: Meira MdV, Silva Rsd, Chochinov HM, Medeiros MOSFd, Ferreira MMdM, de Góes Salvetti M (2024). Effects of Dignity Therapy on individuals with amyotrophic lateral sclerosis: Case studies. *Palliative and Supportive Care* **22**, 517–525. <https://doi.org/10.1017/S1478951523001888>

Received: 30 September 2023

Revised: 05 November 2023







Accepted: 28 November 2023

Keywords:

Palliative care; psychological stress; brief psychotherapy; amyotrophic lateral sclerosis

Corresponding author:

Rudval Souza da Silva;
Email: rudsouza@uneb.br

Mariana do Valle Meira, M.Sc.¹ , Rudval Souza da Silva, Ph.D.^{1,2} ,
Harvey Max Chochinov, B.Sc.³ , Maria Olivia Sobral Fraga de Medeiros, M.Sc.⁴ ,
Marilaine Matos de Menezes Ferreira, Ph.D.⁴  and
Marina de Góes Salvetti, Ph.D.⁵ 

¹Postgraduate Programme in Nursing and Health, Nursing School at the Federal University of Bahia, Salvador, Bahia, Brazil; ²School of Nursing, Bahia State University, Senhor do Bonfim, Bahia, Brazil; ³Department of Psychiatry, University of Manitoba, Winnipeg, MB, Canada; ⁴School of Nursing, Bahiana School of Medicine and Public Health, Salvador, Bahia, Brazil and ⁵School of Nursing, São Paulo University, São Paulo, Brazil

Abstract

Objectives. To analyze the effects of Dignity Therapy (DT) on the physical, existential, and psychosocial symptoms of individuals with amyotrophic lateral sclerosis (ALS).

Methods. This is a mixed-methods case study research that used the concurrent triangulation strategy to analyze the effects of DT on 3 individuals with ALS. Data collection included 3 instances of administering validated scales to assess multiple physical symptoms, anxiety, depression, spiritual well-being, and the Patient Dignity Inventory (PDI), followed by the implementation of DT and a semi-structured interview.

Results. The scale results indicate that DT led to an improvement in the assessment of physical, social, emotional, spiritual, and existential symptoms according to the score results. It is worth noting that the patient with a recent diagnosis showed higher scores for anxiety and depression after DT. Regarding the PDI, the scores indicate improvements in the sense of dignity in all 3 cases, which aligns with the positive verbal reports after the implementation of DT.

Significance of results. This study allowed us to analyze the effects of DT on the physical, existential, and psychosocial symptoms of individuals with ALS, suggesting the potential benefits of this approach for this group of patients. Participants reported positive effects regarding pain and fatigue, could reflect on their life trajectories, and regained their value and meaning.

Introduction

Suffering is inherent to the human experience, but it is often more prevalent among individuals with chronic degenerative diseases who undergo pain, distress, and processes of finitude. Nursing care can assist in alleviating suffering, particularly in the final stages of life. Cicely Saunders advocated that suffering becomes unbearable only when no one cares (Saunders 1991).

When it comes to individuals with motor neuron diseases, existential suffering is profound, and studies indicate that this suffering can be greater than that experienced by cancer patients (Aoun et al. 2015). The suffering observed in individuals with degenerative diseases like amyotrophic lateral sclerosis (ALS) can be attributed to the negative emotional burden linked to hopelessness and a sense of helplessness, frequently encountered during the rapid and continuous progression of the disease, accompanied by an ongoing loss of mobility.

ALS, a degenerative motor neuron disease, advances progressively, widely, and irreversibly without a known cause. Its primary characteristic is muscular atrophy, which leads to complete functional dependence and significant physical, emotional, and social changes. While ALS is quite rare, the patients who develop it have a life expectancy of 3–5 years, often associated with significant existential suffering (Amato and Russell 2016; Quinn and Elman 2020).

The perception of being mentally aware but trapped within one's body brings about a succession of losses, both for the individual facing illness and their family (Leite Neto et al. 2021). The rapid degradation of functionality affects self-esteem, hope, and the perception of dignity, intensifying existential suffering (Amato and Russell 2016; Aoun et al. 2015).

In this context, preserving dignity through measures that prioritize autonomy, reinforce self-care, and value human beings represents more than a resource for the quality of life for individuals in palliative care; it can be seen as an essential human necessity (Chochinov 2012).

The Dignity Model, proposed by Dr Harvey Max Chochinov initially for terminally ill cancer patients, considers 3 major dimensions that influence the perception of dignity: concerns related to the illness, a repertoire of dignity-conserving responses, and social dignity (Chochinov 2002, 2012). This model aims to promote reflections on critical issues for individuals facing

life-threatening or limiting conditions due to an incurable and progressive illness, encompassing feelings experienced in the process of finitude (Chochinov 2002).

Based on this Dignity Model, Chochinov introduced Dignity Therapy (DT), a brief and individualized form of psychotherapy designed to facilitate moments of reflection and alleviate existential suffering. It has been applied to individuals in palliative care, particularly in the field of oncology (Chochinov 2002; Chochinov *et al.* 2011).

In addition to alleviating suffering, the DT allows the people facing death to create a legacy document. This document results from a recorded interview guided by a therapist. It is a moment when a person is invited to recount aspects of their life to be remembered by family and friends. Discovering purpose and meaning in your life and expressing final words or advice to loved ones is possible. After the recording, the therapist presents the patient with the opportunity to edit the interview transcript and prepare the legacy document together. Thus, the therapy is completed when this document is produced, and it can be shared with loved ones, whether family and/or friends (Chochinov 2012).

Given the limited number of studies on the application of DT in individuals with neurological diseases, especially ALS, there is a recognized need to test its effects in this patient group. Studies have been developed with family caregivers of people with ALS, and the results of this intervention are that the end-of-life experience has improved and helps family members during bereavement (Bentley *et al.* 2014). Other studies with these patients have recommended that DT be undertaken earlier in the disease trajectory, which points to the value of narrative and generativity for patients with motor neuron disease (Aoun *et al.* 2015).

Thus, this study aimed to analyze the impact of DT on the physical, existential, and psychosocial symptoms of individuals with ALS.

Method

A mixed-methods case study was conducted using the concurrent triangulation strategy (Creswell and Creswell 2023), applied to 3 patients with ALS. This type of study is widely used in social sciences, particularly psychology (Schuelke and Rubenstein 2020). Concurrent triangulation involves the simultaneous collection of qualitative and quantitative data directed toward the same concept and encompasses the simultaneous execution of both approaches (Creswell and Creswell 2023). In this study, the idea of interest was the physical, existential, and psychosocial suffering of individuals with ALS.

The potential participants for this study were recruited from the neuromuscular outpatient clinic of a private university in Northeast Brazil, recognized for its expertise in providing care to individuals with ALS. The inclusion criteria included patients with a confirmed diagnosis of ALS and 18 years and older. Patients without a confirmed diagnosis or those with limitations in oral communication were excluded.

The initial selection involved 10 individuals diagnosed with ALS, aged 18 years and older, with 7 males and 3 females. Out of these, 7 were excluded: 1 person did not have a confirmed ALS diagnosis, 1 passed away before data collection began, 1 had speech deficits (unintelligible language), and 4 declined to participate in the study. As a result, 3 patients agreed to participate and were included in the study.

The data were collected through interviews and the administration of research instruments in person at the patients' residences

or through remote means facilitated by technology. The entire process of administering the scales took approximately 30 min. Data collection occurred between October 2021 and April 2022. The Research Ethics Committee approved the study under protocol number 4.952.826. To maintain anonymity and confidentiality, pseudonyms (Geranium, Lily, and Sunflower) were used when describing the cases, symbolizing the personalities of each participant chosen by themselves. The study design is depicted in Fig. 1.

Qualitative and quantitative data were collected simultaneously and independently. An instrument with sociodemographic data, including age, gender, marital status, religion, education, employment status, and time since disease diagnosis, was used for participant identification. Quantitative data collection was conducted through video calls, with the administration of the 4 scales (Patient Dignity Inventory [PDI], Hospital Anxiety and Depression Scale [HADS], Functional Assessment of Chronic Illness Therapy – Spiritual Well-Being [FACIT-Sp], and Edmonton Symptom Assessment Scale [ESAS]), 2 days after they consented to participate in the research, using platforms like Google Meet® or WhatsApp®. The researcher would read and explain the structure and possible answers to the participants according to each scale. So, he answered the inquiries during the application of each scale.

The following scales were used at 3 distinct time points: PDI (Chochinov *et al.* 2008), which is a product of the Dignity Model and aims to assess the sense of dignity across multiple dimensions using 25 indicators. Scores can range from 0 to 5, with higher values suggesting issues related to the importance of dignity in that dimension. The HADS (Zigmond and Snaith 1983) is composed of 14 items divided into anxiety and depression scales, with its score distributed across possible cases (above 8 points), probable cases (above 11 points), and severe disorders (above 15 points). The FACIT-Sp (Peterman *et al.* 2002) comprises 12 items, subdivided into Meaning/Peace (items 1–8) and faith (items 9–12), and its responses are expressed on a 5-point Likert scale, with 0 corresponding to “none” and 4 corresponding to “very much” spiritual well-being sensation. Finally, the ESAS (Paiva *et al.* 2015) assesses symptoms such as fatigue, pain, nausea, anxiety, depression, appetite, well-being, sleep, and dyspnea, with scores related to the intensity of symptoms ranging from 0 to 10 points, where higher values correspond to worsening occurrences (Fig. 1).

At “baseline” or baseline, the scales (quantitative data) were administered, as well as at “time point 1,” 7 days after the DT session, and at “time point 2,” 28 days after the therapy. Between the baseline and “time point 1,” the intervention sessions (DT) took place. The number of sessions varied from 1 to 2 due to patient fatigue, each lasting between 60 and 180 min. A digital recorder captured the narrative, and a legacy document was produced based on the recording. This document was shared with each patient after being read, approved, and edited after all data collection processes. Two of the authors participated in a DT Training Workshop with Dr Chochinov.

After the intervention and before the seventh day post-intervention, a session with the participant was arranged to conduct the interview (qualitative data). A semi-structured script was used to comprehend the effects of DT concerning physical, psychosocial, and existential suffering. During the interview, the researcher asked 3 questions: (a) Tell me what it was like for you to undergo DT. (b) After completing DT, how have you been feeling? (c) Regarding your physical, emotional, and spiritual symptoms, can you understand whether, after speaking during DT and generating the legacy document, they have improved or not?

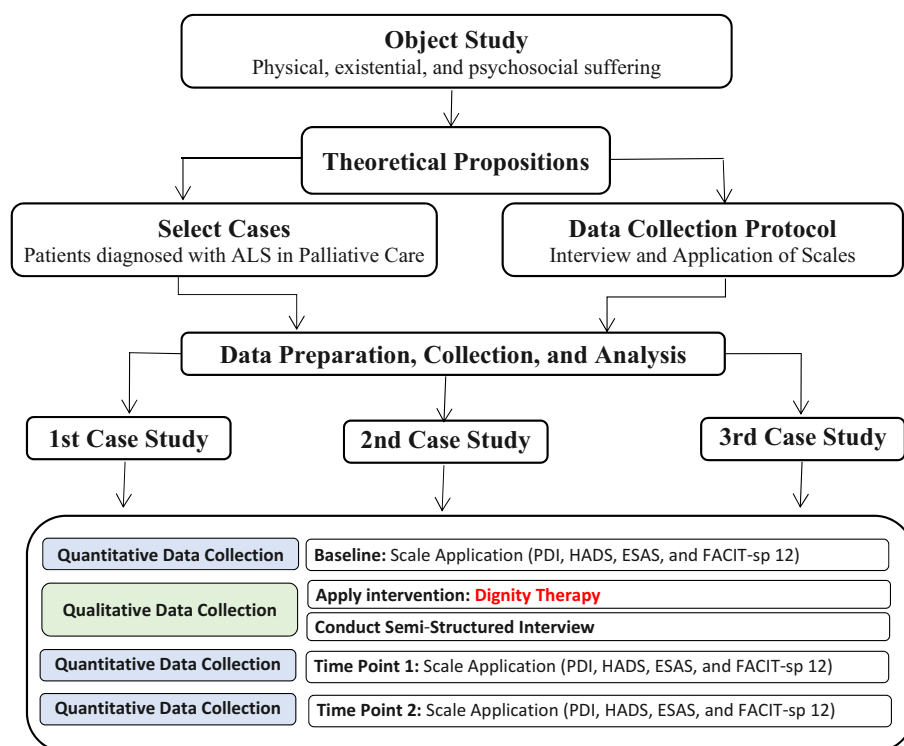


Figure 1. Representative diagram of the study design.

The interviews were transcribed and facilitated by an online simultaneous transcription program. Subsequently, they were edited and presented in a recorded virtual session with each participant via the “Google Meet” platform. During this session, feedback was provided, and some ideas were altered and reorganized per the participant’s requests. Ultimately, this process led to the creation of the legacy document handed over to the participant.

All meetings were prescheduled and took place in a private, calm, and comfortable setting (usually at the participants’ homes), both in person and/or remotely, based on the participant’s preference. Virtual communication platforms and applications such as “Google Meet,” “Zoom,” and “WhatsApp” were used. These measures were adopted to uphold patient confidentiality, dignity, and comfort, as all participants had impaired physical mobility (bedridden or dependent on orthotics).

The qualitative data obtained through semi-structured interviews were analyzed by comparing them with the quantitative data (Busetto et al. 2020) obtained from the administration of the scales and their corresponding scores. These scores were analyzed using descriptive statistics.

Results

Qualitative analysis

The case studies are presented based on the 3 dimensions of the Dignity Model: illness-related concerns, dignity-conserving responses, and social dignity (Chochinov 2002).

Case 1 – Mr. Geranium

Concerns related to the illness

Male, 53 years old, born in a Northeastern Brazilian capital, widower, Catholic, residing with his only teenage son. He is an electronics technician, and at the time of data collection, he was

retired due to the disease and functional limitations. He maintained good cognitive acuity and reported that the first symptoms of ALS appeared in 2017, with a confirmed diagnosis in 2018. His mobility was significantly impaired, rendering him bedridden and with unintelligible speech. Few indicators of physical suffering were observed during scales and DT administration.

When questioned about stressful physical symptoms like pain, shortness of breath, fatigue, or nausea, he mentioned only pain and fatigue, which slightly improved after DT. Regarding his appearance to others, he stated that it was a “minor issue” in his life. Concerning psychological suffering, he indicated a history of seeking treatment for depression, attributing his sadness not to the disease itself but to the mourning process following the loss of his wife. He reported an improvement in sadness after undergoing DT and demonstrated a clear understanding of the disease and its treatment.

Dignity-conserving perspectives

He demonstrates a pursuit of spiritual comfort, as his faith in God is evident during DT and described in the legacy document, showcasing his involvement and devotion to the Catholic religion. Despite his physical limitations and dependence on family, friends, and caregivers for daily activities, he does not see himself as a burden to others. He maintains a positive attitude toward his life, attempting to maintain normalcy and highlighting his victories and joyous moments. He characterizes his challenges as insignificant compared to everything he has experienced until that point in his life. He mentions not being afraid of death but does feel an existential emptiness after losing his wife and worries about his son’s future.

Social dignity inventory

He exhibited emotional and hopeful tendencies during DT, particularly when addressing aspects of his childhood and adolescence marked by numerous sacrifices and efforts. Nonetheless,

he acknowledges succeeding in his profession, enjoying autonomy and control over his life, displaying solid relationships with his family, and benefiting from social and familial support. He emphasized the joy and satisfaction of building and providing for his family. Simultaneously, he recounted the suffering caused by the loss of his mother and wife, along with the diagnosis of ALS. He expressed concerns about the disease's implications for his life, especially concerning his son. While he acknowledged the loss of many functions due to ALS, he did not demonstrate sadness about it. He focused on cherishing moments with his son and witnessing his development into a "good person." This notion became his reason for living. While creating the legacy document, he displayed concern for preserving details, ensuring that nothing was lost, and safeguarding his story for future generations. Spiritual comfort prominently featured in his life, providing him with calmness and hope.

Case 2 – Mr. Lily

Concerns related to the illness

Male, 52 years old, born in the Northeastern Brazilian capital. He spent a significant portion of his life in the Southeastern region of Brazil. He is a Jehovah's Witness, married, and residing with his wife, 2 adult daughters, and 1 grandson. He worked as a systems analysis technician and is retired due to the disease, which has imposed functional limitations. He possesses good cognitive understanding and identified the first symptoms of ALS in 2017, although the diagnosis was only confirmed in 2021. He experiences severely compromised physical mobility and has difficulty walking but maintains intelligible speech and some movements in his upper limbs.

During the administration of scales and the intervention, indicators of physical suffering were evident in his discourse. When asked about stressful physical symptoms, he mentioned experiencing pain and fatigue, which had slightly improved after DT. He expressed that his appearance to others is not a concern in his life and did not express significant psychological suffering. He demonstrated a clear and conscious understanding of his disease and its treatment.

Dignity-conserving perspectives

He appears to be observant, committed, and helpful during the application of DT, recounting his life journey marked by struggles, love, and faith. For him, family always comes first, emphasizing unity, respect, and affection for all. Despite his physical limitations and dependence on family members for daily activities, he does not perceive himself as a burden to others. He reflects positively on his life, striving to maintain normalcy and emphasizing victories and joyous moments. He demonstrates concern for building a promising future for his wife and daughters, recounting his efforts to achieve this goal. He mentions not being afraid of death and cherishes the small moments he has experienced.

Social dignity inventory

He seems hopeful and spiritual, emphasizing faith as an essential part of his life. He acknowledges success in his profession and in establishing and maintaining his family, and he displays a positive relationship with his loved ones, indicating social solid and familial support. DT enabled him to reflect on living with limitations and overcoming obstacles day by day, mainly because family and faith are his reasons for living. He acknowledges the loss of many functions due to ALS, yet he does not express sadness regarding this matter. The disease is not prominent in his life, and he is referred

to as a "supporting character." He is concerned about preserving his story as a legacy document for future generations of his family.

Case 3 – Mrs. Sunflower

Concerns related to the illness

Female, 49 years old, evangelical, born in a Northeastern Brazilian capital, married but in the process of divorce. She is a mother of 2 children, one adult and another teenager, who lives with her. Her older son assists her by taking her to appointments and other places, while the younger one provides company and helps with household tasks. She is a professional seamstress, retired due to a disease that has caused functional limitations. Her cognitive acuity is preserved, and she mentioned identifying the first symptoms of ALS in 2012, though the diagnosis was only confirmed in 2022. Her physical mobility is considerably compromised, with difficulty walking, but she maintains motor control in her upper limbs and has intelligible, slightly nasal speech.

During the administration of scales and the intervention, she exhibited signs of physical suffering. When asked about stressful physical symptoms, she mentioned experiencing pain, fatigue, and nausea, noting slight improvement after DT. Concerning her appearance to others, she responded that it was not a significant problem in her life. She appeared to be a person who values her appearance, emphasizing the desire to be well-taken care of even though she will become entirely dependent on others for daily activities.

Psychological suffering was identified when she mentioned loss of autonomy and identity. This suffering was slightly reduced after undergoing DT. She appears to have a conscious understanding of her disease and treatment but is still adapting to the new reality.

Dignity-conserving perspectives

She appears to be highly emotional, affectionate, and nostalgic. During DT, while narrating her life journey, she portrays herself as a joyful person surrounded by companionship and love, highlighting her fulfillment as a mother. She conveys an image of a woman who is both vain and strong yet gentle, showing concern for and taking care of everyone around her. She feels like a burden to others and appears saddened when discussing the small desires she wishes to fulfill but is limited by her dependence on family members, such as going to the beach. Despite this, she tries to maintain normalcy, emphasizing her family as the reason for living. She shows concern about her children's future, particularly her younger son, and has doubts about the best person to assume guardianship in case she dies. She mentions not fearing death, describing in detail her wishes for when that moment comes. Despite regretting certain aspects of her journey as a mother and wife, she still values the moments she has experienced.

Social dignity inventory

She demonstrates perseverance, hopefulness, courage, and self-assuredness, defining with precision all her desires and expectations for the future. Despite her life challenges, she finds reasons to continue living with her family and faith. The memories shared during therapy made her laugh and become emotional, imprinting her essence in the legacy document. She mentions the loss of many functions and expresses some sadness about it, particularly regarding her autonomy. A slight improvement was observed after DT. She is concerned about preserving her story as a legacy document so that her family knows how she feels and how she would like to be treated and remembered in the future.

Table 1. Description of scale application, DT, and interviews

| Cases | 1 – Mr. Geranium | 2 – Mr. Lily | 3 – Mrs. Sunflower |
|-------------------|---|--|---|
| Baseline | The administration of the 4 scales took an average of 30 min, and the participants did not show any difficulties in understanding the questions addressed in the scales. | | |
| Application of DT | Conducted in a single in-person session lasting approximately 180 min. | Conducted virtually (via <i>Google Meet</i> ®), chosen by the participant, in 2 sessions due to exhaustion from speaking, with each session lasting approximately 120 min. | Conducted in person, in 2 sessions, due to the participant's exhaustion and emotions, with each session lasting approximately 120 min. The chosen location was her home, considering the participant's difficulty in mobility and prioritizing her privacy. |
| Interview | When asked about how it was to undergo the therapy and how they felt, they responded: | | |
| | <i>The therapy was good because I regained points I no longer remember or remembered very little [...] I realized that I achieved many things. When we bring these things out, it lifts our spirits psychologically. I felt very well! [...] After I did the Dignity Therapy, I am more balanced spiritually and emotionally [...].</i> | <i>Dignity Therapy was valuable because it made me recall unique moments that stay in our secondary memory [...] It's a therapy that's truly worth doing [...] it brings a certain dignity, a life improvement, and we manage to have a bit more joy! [...] Regarding my physical symptoms, it does provide some relief, but I can't say it was one hundred percent because I also use medications to help me [...].</i> | <i>Dignity Therapy for me was both scary and gratifying [...] I feel thankful and relieved to be able to say things that I wouldn't find the strength to say to my family [...] I think I got very emotional!</i> |
| Time point 1 | It also took place virtually, 7 days after the application of DT. The entire process lasted about 30 min, and it was a calm and seemingly enjoyable moment for the participant. | | |
| Time point 2 | The third administration of the scales, in all 3 cases, took place in the same manner as described in time point 1. | | |

DT = Dignity Therapy.

Quantitative analysis

The quantitative data are described for the 3 assessment time points.

Table 2 presents the results of the scores from the 4 scales (PDI, HADS, FACIT-Sp, and ESAS) in a comparative manner. It is based on the participants' responses (Table 1) regarding their perception of the empirical indicators of each respective scale, indicating whether there was an improvement in their well-being according to the timing of scale application before and after the completion of DT.

Discussion

This study described the application of DT to 3 patients diagnosed with ALS, facing physical, existential, and psychosocial issues. When invited to participate in the research, they appreciated the opportunity to speak with the researcher about their concerns. The results suggest that the reflections facilitated by DT can alleviate the suffering of these patients by emphasizing the value of the moments they have experienced throughout their lives.

A systematic review (Xiao et al. 2019) of DT indicates that this intervention is a promising approach to enhancing psychosocial well-being among patients in palliative care facing oncological diseases. Similarly, another study conducted with palliative care patients in Canada, Australia, and the United States used a scale to assess the impact of dignity after implementing the intervention, yielding positive results (Scarton et al. 2018).

DT is a relatively recent therapeutic intervention, and its studies have predominantly been conducted in Western countries. However, research conducted in a healthcare service in Southern Taiwan indicated that DT could enhance the sense of dignity and reduce feelings of sadness and even depression in patients with terminal oncological diagnoses. This study suggests the need for further research in other countries, considering cultural variations

and other diseases such as ALS, stroke, or renal insufficiency (Li et al. 2020).

The results of the current study conducted in Brazil suggest an improvement in signs of sadness, anxiety, and depression for these 3 patients. However, it is worth noting the case of the recently diagnosed ALS patient, who showed an increase in scores on the anxiety and depression scales. That said, this same patient demonstrated reduced dignity-related distress (PDI) and gratification with DT. DT showed positive effects on the physical, existential, and psychosocial symptoms of these patients with ALS by facilitating the retrieval of life experiences, achievement of goals, memories of happy moments, and accomplishments that were previously undervalued but proved to be of vital importance to Geranium, Lily, and Sunflower.

A multicenter study (Chochinov et al. 2011) conducted with palliative care patients in North American and Australian countries demonstrated that after the implementation of DT, participants considered it a helpful therapy that prioritizes quality of life and dignity. In addition, the intervention changed patients' and their families' perspectives regarding their finitude and understanding of what palliative care entails.

DT enabled participants to articulate and share memories, often painful and never mentioned before, awakening a sense of gratitude for being heard and understood. These feelings of freedom and relief experienced by patients after DT were also observed in another study (Julião et al. 2014) that aimed to determine the influence of DT on depression and anxiety in palliative care patients in Portugal.

As mentioned earlier, the results were presented based on the 3 categories that support the Dignity Preservation Care Model (Chochinov 2002), and they were analyzed considering their divergences and convergences.

This Care Model (Chochinov 2002) explicitly states that relevant issues inherent to the process of illness are directly connected to a person's dignity, so these issues can exacerbate levels of physical,

Table 2. The score results according to the scales [PDI, HADS, FACIT-Sp, and ESAS] applied in the 3 study time points

| Scales | Geranium | | | Lily | | | Sunflower | | |
|-------------------------------------|----------|----|----|------|----|----|-----------|----|----|
| | T0 | T1 | T2 | T0 | T1 | T2 | T0 | T1 | T2 |
| PDI (Min 25/Max 125) | 67 | 42 | 35 | 54 | 46 | 33 | 80 | 72 | 60 |
| HADS – Anxiety (Min 0/Max 21) | 7 | 6 | 6 | 7 | 3 | 0 | 13 | 11 | 14 |
| HADS – Depression (Min 0/Max 21) | 8 | 5 | 4 | 4 | 1 | 2 | 8 | 11 | 14 |
| FACIT-Sp – 12 (Min 0/Max 48) | 30 | 29 | 31 | 34 | 42 | 40 | 20 | 21 | 20 |
| ESAS (Min 0/Max 10) | | | | | | | | | |
| Pain | 7 | 6 | 5 | 7 | 2 | 1 | 8 | 6 | 5 |
| Fatigue (Weakness) | 5 | 7 | 4 | 7 | 4 | 7 | 6 | 7 | 5 |
| Nausea (Queasiness) | 0 | 0 | 0 | 0 | 0 | 0 | 0 | 6 | 7 |
| Sadness | 3 | 3 | 3 | 3 | 0 | 0 | 7 | 4 | 5 |
| Anxiety | 2 | 0 | 0 | 3 | 0 | 0 | 6 | 3 | 5 |
| Drowsiness | 3 | 5 | 2 | 0 | 0 | 2 | 5 | 6 | 7 |
| Appetite | 1 | 2 | 0 | 8 | 8 | 9 | 2 | 7 | 6 |
| Sense of well-being | 3 | 2 | 2 | 7 | 0 | 4 | 6 | 5 | 6 |
| Dyspnea | 0 | 0 | 0 | 7 | 0 | 0 | 2 | 4 | 3 |
| Sleep | 2 | 2 | 0 | 8 | 0 | 5 | 4 | 8 | 7 |

DT = Dignity Therapy; ESAS = Edmonton Symptom Assessment Scale; FACIT-Sp = Functional Assessment of Chronic Illness Therapy – Spiritual Well-Being; HADS = Hospital Anxiety and Depression Scale; PDI = Patient Dignity Inventory. T0, denotes the baseline (2 days before the DT); T1 denotes the first week of follow-up (7 days after the intervention); and T2 denotes the fourth week of follow-up (28 days after the intervention).

existential, and psychosocial suffering. Concerns related to the disease, encompassing both physical and psychological responses, are among the themes that directly affect a person's dignity in palliative care.

In all 3 case studies, regarding the level of independence, participants exhibited some degree of physical limitation, which implies the inability to perform their work functions and provide for their financial needs. They all receive social benefits through retirement and depend on their family members and/or caregivers to carry out activities of daily living, such as bathing, eating, and moving around.

ALS is a disease that affects each patient's physical, existential, and psychosocial dimensions. According to the International Classification of Functioning, Disability, and Health released by the World Health Organization in 2001, ALS is indeed a clinical condition that impacts a person's life from the perspectives of the body, the individual, and society (Sandstedt et al. 2018) in a way that requires the healthcare team, regardless of the severity of the disease, to conduct a thorough assessment regarding the level of impairment, activity limitations, and restrictions to apply person-centered interventions at the appropriate time (Bentley and O'Connor 2017).

In this context, DT elicited a positive response from these patients, particularly by helping them recover their life stories and

creating a legacy document that can be left to their loved ones before or after their death.

The age range of the participants varied between 49 and 53 years, with 2 of them being men and one woman. A study (Mehta et al. 2023) on the prevalence of ALS in the US population points to a lower prevalence of the disease among individuals aged 18–39 years and a higher prevalence among those aged 70–79 years, and more in men than in women. It is worth noting that diagnosing ALS is challenging in the early stages of the disease due to the complexity of distinguishing it from other conditions, such as spondylotic myelopathy, spinal muscular atrophy, multiple sclerosis, and other diseases (Norris et al. 2020).

Regarding technology-mediated interviews and even the application of DT via online platforms, the results have been promising, and studies already indicate that conducting online DT is feasible and well-received by patients. It reduces therapist time and clinical costs while also reaching individuals who might otherwise not be able to undergo DT (Bentley et al. 2020). The same study even suggests conducting DT sessions through email, which is particularly favorable for ALS patients who struggle with speech but can respond to questions via email.

During the post-intervention interviews, it was evident in all 3 patients that there were feelings of anguish related to the loss of functional capacity. This loss significantly impacted their level of independence, leading to a sense of unproductivity due to physical impairment. Among the participants, only Mr. Lily demonstrated the ability to reframe his current condition by applying his professional knowledge to engage in leisure activities to occupy his time.

On the other hand, all 3 participants maintained cognitive functions, which is often observed in patients diagnosed with ALS. In many cases, cognitive clarity remains until the end of life for individuals with ALS. These characteristics highlight palliative care teams' crucial role in providing care for ALS patients and their families (Fahrner-Scott et al. 2022).

Regarding the physical and psychosocial symptoms caused by the disease, there was a heightening of emotions during DT, such as sadness and longing. These emotions suggest psychological reactions indicating understanding the disease and perceiving finitude (Julião et al. 2014).

In the 3 case studies, the participants reported the perception of "improved dignity," also reflected in the comparative PDI (Chochinov et al. 2008), applied before and after the intervention. These scores gradually decreased, shifting from "a huge problem" to "a small problem." This change was even more noticeable 1-month post-DT.

Regarding anxiety and depression based on HADS (Zigmond and Snaith 1983) scores, both Geranium and Lily reported improvements after DT. Sunflower experienced an improvement in anxiety 7 days after the DT, which worsened after 28 days. In her interview, she mentioned feeling weakened, having headaches, and nausea, likely due to insomnia that had been affecting her for a few days. She reported that she had been dealing with depression for a long time and was quite distressed at the time of data collection due to a series of personal events, including her recent diagnosis of ALS and the process of separating from her partner, leading to a wave of concerns about the future of her children.

Results from a study (Chochinov et al. 2016) conducted in Canada on dignity-related distress among patients with non-oncological diseases, including those with ALS, demonstrate distinct and significant distress patterns within each group. In the ALS patient group, more significant dignity-related distress was

observed compared to the others. Some concerning issues were the inability to fulfill important social roles, failure to carry out daily tasks, feeling like a burden to others, losing control over their lives, and a sense of devaluation. It was also noted that they were more prone to experiencing depression and displaying excessive concern about the future.

Regarding the physical symptoms of ALS, the participants reported that pain interferes with their lives. During therapy, they indicated that they were most burdened by their disease when they felt hurt or that physical impairment hindered daily activities. DT seems to have some effect on pain and fatigue relief, as observed in the scores of the ESAS (Paiva et al. 2015). Mr. Lily also reported that DT provides “relief” from pain and fatigue.

DT mobilizes people’s emotions, transforming simple storytelling into a journey that brings reflections and new meanings. It is like watching a movie with real characters who realize they have been and are essential to others. This process enables them to reevaluate their lives, extracting a more positive perspective (Schuelke and Rubenstein 2020; Söderman et al. 2020). Initially, participants in all 3 case studies seemed unable to grasp the DT concept fully. However, during the intervention (DT), they became receptive and allowed themselves to engage with their emotions without concern for the attending therapist.

All participants claimed improvement related to the spiritual and emotional dimensions, mentioning a sense of “balance,” a restoration of dignity, resulting in “a bit more joy,” upon recalling positive moments experienced. When asked about the significance of DT, Geranium responded that it was a moment that made him realize the dimension of finitude, allowing him to put his essence on paper, referring to the resulting document as a “farewell letter.”

The legacy document generated through DT is a resource that clarifies the patient’s wishes and perspectives. This document will hold great value for those left behind. It can be operationalized in various ways, including a farewell letter, a video message, or a postcard with a brief greeting (Welsch and Gottschling 2021). The fact that the patient wrote the message imbues it with meaning, care, and love.

A research study (Beck et al. 2019) comprised of a qualitative analysis regarding the benefits and barriers related to DT showed that for some patients, dignity represented how much their life held meaning for others, transcending death. From this perspective, the study’s results suggest that DT promoted self-expression and connection with loved ones to these patients, awakening a sense of purpose and facilitating the expression of continuity of self. Completing their legacy document enabled moments of reflection, autonomy, and opportunities for interaction with the family, with the latter often being the focus of DT.

In this sense, it can be inferred that family bonds and connections with life can be immortalized by creating a legacy document. Family ties hold great significance to Geranium, Lily, and Sunflower. Hence, there was great care not to leave any significant memory or message out of the legacy document.

Due to their prominent roles as “heads of the family,” Geranium and Lily demonstrated control over their lives despite the limitations imposed by the disease. They could express their desires and wishes, which their family members largely fulfilled. Sunflower was going through a divorce process and seemed emotionally more fragile, partly due to the recent diagnosis and concerns for her younger child’s care in the face of her impending death. In her legacy document, Sunflower mentioned how she would like to be cared for by her family, yet she admitted not having the courage to express these wishes directly. Instead, she preferred

her desires to be known after the family members read the legacy document.

Legacy documents transcend death itself, as they allow for the awakening of future generations to the meaning of life and a sense of acceptance (Vuksanovic et al. 2017). Furthermore, understanding the disease makes patients more resilient, enabling them to reflect on their lives during the process of DT, marked by challenges and triumphs. Geranium mentions that upon diagnosis, he experienced significant suffering and even went through a process of denial, leading to a depressive disorder. However, with the support of his partner and family, he managed to comprehend and overcome that difficult time. Lily expressed a sense of being at peace with his finitude, yet there was still an improvement in his sense of dignity.

Geranium and Lily have had more time to adjust to their diagnoses than Sunflower, who was recently diagnosed and is still adapting to the idea of finitude. This is accentuated by her being the central figure in the family and having to face the changes on her own in the absence of a partner. When women become ill, particularly those with dependent children, family life becomes disrupted, leading to anticipatory grief (Kramar et al. 2023). This is consistent with the gradual emotional detachment observed in Sunflower’s case.

Regarding hope for the future, Geranium mentions not seeing significant prospects for himself, considering himself fortunate, having already achieved everything he desired. The other participants described their plans for the near future, thus allowing themselves to live in the moment.

A significant emphasis on religiosity was observed during DT among the participants. All 3 mentioned faith as a helpful strategy in preserving dignity, emphasizing that it helps them live in the moment despite the limitations imposed by the disease. Along with FACIT-Sp-12 scores, this corroborates high levels of spiritual well-being among the participants, even before the therapy, with only a slight variation post-DT. Spiritual well-being is reflected in the perception that small actions can reveal moments of true significance, consistent with the notion that ALS is a “supporting character” in their lives.

The third category of the Care Model is the “inventory of social dignity,” which encompasses external factors or challenges. They include privacy boundaries, social support, tone of care, feeling like a burden to others, and aftermath concerns, i.e., the responsibility that their death will place on those left behind (Chochinov 2002).

All participants are dependent on a caregiver, family, or otherwise. During DT, they sometimes expressed embarrassment about being a burden, feeling like a weight on their families. Lily mentions this less often, perhaps due to his more reserved nature or because he is less physically limited than Geranium and Sunflower.

While the duration of illness for Lily and Geranium is similar, the progression of the disease has been faster for Geranium. Sunflower reveals that she feels like a burden to her children. This is evident when she describes her desire to go to the beach or carry out some routine household activities, for which she depends on her older son. Since ALS is responsible for the degeneration of motor functions, it hampers the patient’s involvement in daily activities and social functioning (Quinn and Elman 2020).

All 3 participants emphasize the importance of social support. They have close-knit families that understand their health situations and try their best to assist them. However, their positive relationships with their families do not eliminate their concerns for their children and relatives. Two of the participants (Sunflower and Geranium) have underage children and worry about not being

alive to see them reach adulthood. Geranium is a widower and is still dealing with grief, while Sunflower is recently separated and feels uncertain about her child's father taking care of him.

All participants mentioned that they could extend the DT sessions by writing numerous legacies, as they were delighted and experienced a sense of well-being while engaging in DT, highlighting its importance. Each of their discussions showed a noticeable reawakening of dignity, which had been somewhat forgotten amidst the challenges of daily life with ALS.

Implications for practice

DT is a care tool grounded in a multidimensional perspective, which aligns with nursing principles. Any palliative care team member can be trained to administer DT if they are intuitive, good communicators, empathic, and compassionate. The application of DT can be conducted in person or through virtual technology, allowing for a broader reach. Further studies are needed, utilizing more robust methodologies to test DT on larger samples of patients with neurodegenerative disorders facing the inevitability of death.

Limitations

This study has strengths and limitations that need to be acknowledged. The small sample size and study design do not allow for the generalization of findings. However, the observed results provide a basis for more robust studies to test the benefits of DT for patients with ALS and other chronic degenerative diseases.

Conclusion

This study analyzed the effects of DT on the physical, existential, and psychosocial symptoms of individuals with ALS, suggesting the potential benefits of this approach for this patient group. Participants reported positive effects on pain and fatigue, reflecting on their life journey, reclaiming value and meaning, and recognizing challenges and achievements. Participants were able to address issues related to finitude, as well as express concerns for the future of underage children.

Funding. The Conselho Nacional de Desenvolvimento Científico e Tecnológico (CNPq), Research Productivity Scholarship.

Competing interests. The authors declare that there is no conflict of interest.

References

- Amato AA and Russell JA (2016) Amyotrophic lateral sclerosis. In Amato AA and Russell JA, 2nd edn. *Neuromuscular Disorders*. Burlington: Mc Graw Hill, 174–198.
- Aoun SM, Chochinov HM and Kristjanson LJ (2015) Dignity therapy for people with motor neuron disease and their family caregivers: A feasibility study. *Journal of Palliative Medicine* **18**(1), 31–37. doi:10.1089/jpm.2014.0213
- Beck A, Cottingham AH, Stutz PV, *et al.* (2019) Abbreviated dignity therapy for adults with advanced-stage cancer and their family caregivers: Qualitative analysis of a pilot study. *Palliative and Supportive Care* **17**(3), 262–268. doi:10.1017/S1478951518000482
- Bentley B and O'Connor M (2017) Counselling people with amyotrophic lateral sclerosis. *Age* **30**, 50–59. doi:10.5176/2315-4330_WNC17.131
- Bentley B, O'Connor M, Breen LJ, *et al.* (2014) Feasibility, acceptability and potential effectiveness of dignity therapy for family carers of people with motor neuron disease. *BMC Palliative Care* **13**(1), 12. doi:10.1186/1472-684X-13-12
- Bentley B, O'Connor M, Williams A, *et al.* (2020) Dignity therapy online: Piloting an online psychosocial intervention for people with terminal illness. *Digit Health* **20**(6), 1–8. doi:10.1177/2055207620958527
- Busetto L, Wick W and Gumbinger C (2020) How to use and assess qualitative research methods. *Neurological Research and Practice* **2**(14), 1–10. doi:10.1186/s42466-020-00059-z
- Chochinov HM (2002) Dignity-conserving care: A new model for palliative care: Helping the patient feel valued. *JAMA* **287**(17), 2253–2260. doi:10.1001/jama.287.17.2253
- Chochinov HM (2012) *Dignity Therapy: final Words for Final Days*. New York: Oxford.
- Chochinov HM, Hassard T, McClement S, *et al.* (2008) The Patient Dignity Inventory: A novel way of measuring dignity-related distress in palliative care. *Journal of Pain and Symptom Management* **36**(6), 559–571. doi:10.1016/j.jpainsymman.2007.12.018
- Chochinov HM, Johnston W, McClement SE, *et al.* (2016) Dignity and distress towards the end of life across four non-cancer populations. *PLoS One* **11**(1), e0147607. doi:10.1371/journal.pone.0147607
- Chochinov HM, Kristjanson LJ, Breitbart W, *et al.* (2011) Effect of dignity therapy on distress and end-of-life experience in terminally ill patients: A randomized controlled trial. *The Lancet Oncology* **12**(8), 753–762. doi:10.1016/S1470-2045(11)70153-X
- Creswell JW and Creswell JD (2023) *Research Design – International Student Edition: Qualitative, Quantitative, and Mixed Methods Approaches*, 6th edn. Thousand Oaks, CA: SAGE Publications, Inc.
- Fahrner-Scott K, Zapata C, O'Riordan DL, *et al.* (2022) Embedded palliative care for amyotrophic lateral sclerosis: A pilot program and lessons learned. *Neurology Clinical Practice* **12**(1), 68–75. doi:10.1212/CPJ.0000000000001124
- Julião M, Oliveira F, Nunes B, *et al.* (2014) Efficacy of dignity therapy on depression and anxiety in Portuguese terminally ill patients: A phase II randomized controlled trial. *Journal of Palliative Medicine* **17**(6), 688–695. doi:10.1089/jpm.2013.0567
- Kramar MJE, Caro MPG, Ternero AM, *et al.* (2023) Migrant women caregivers' experiences in end-of-life formal care. *Revista da Escola de Enfermagem da USP* **57**(spe), e20230031. doi:10.1590/1980-220X-REEUSP-2023-0031en
- Leite Neto L, Novais T, França Júnior M, *et al.* (2021) Repercussions of language difficulties in people with amyotrophic lateral sclerosis and the impact on their lives and on their caregivers. *Revista CEFAC* **23**(4), e14120. doi:10.1590/1982-0216/202123414120
- Li YC, Feng YH, Chiang HY, *et al.* (2020) The effectiveness of dignity therapy as applied to end-of-life patients with cancer in Taiwan: A quasi-experimental study. *Asian Nursing Research* **14**(4), 189–195. doi:10.1016/j.anr.2020.04.003
- Mehta P, Raymond J, Punjani R, *et al.* (2023) Prevalence of amyotrophic lateral sclerosis in the United States using established and novel methodologies, 2017. *Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration* **24**(1–2), 108–116. doi:10.1080/21678421.2022.2059380
- Norris SP, Likanje MFN and Andrews JA (2020) Amyotrophic lateral sclerosis: Update on clinical management. *Current Opinion in Neurology* **33**(5), 641–648. doi:10.1097/WCO.0000000000000864
- Paiva CE, Manfredini LL, Paiva BSR, *et al.* (2015) The Brazilian version of the Edmonton Symptom Assessment System (ESAS) is a feasible, valid, and reliable instrument for the measurement of symptoms in advanced cancer patients. *PLoS One* **10**(7), e0132073. doi:10.1371/journal.pone.0132073
- Peterman AH, Fitchett G, Brady MJ, *et al.* (2002) Measuring spiritual well-being in people with cancer: The Functional Assessment of Chronic Illness Therapy – Spiritual Well-being Scale (FACIT-Sp). *Annals of Behavioral Medicine* **24**(1), 49–58. doi:10.1207/S15324796ABM2401_06
- Quinn C and Elman L (2020) Amyotrophic lateral sclerosis and other motor neuron diseases. *Continuum (Minneapolis)* **26**(5), 1323–1347. doi:10.1212/CON.0000000000000911
- Sandstedt P, Littorin S, Johansson S, *et al.* (2018) Disability, and contextual factors in patients with amyotrophic lateral sclerosis – A three-year observational study. *Journal of Neuromuscular Diseases* **5**, 439–449. doi:10.3233/JND-180322

- Saunders C** (1991) *Hospice and Palliative Care: an Interdisciplinary Approach*. London: Edward Arnold, 116.
- Scarton L, Oh S, Sylvera A, et al.** (2018) Dignity impact as a primary outcome measure for dignity therapy. *American Journal of Hospice and Palliative Medicine* **35**(11), 1417–1420. doi:10.1177/1049909118777987
- Schuelke T and Rubenstein J** (2020) Dignity therapy in pediatrics: A case series. *Palliative Medicine Reports* **1**(1), 156–160. doi:10.1089/pmr.2020.0015
- Söderman A, Östlund U, Harstäde CW, et al.** (2020) Dignity conserving care for persons with palliative care needs – Identifying outcomes studied in research: An integrative review. *Palliative and Supportive Care* **18**, 722–740. doi:10.1017/S1478951520000139
- Vuksanovic D, Green HJ, Dyck M, et al.** (2017) Dignity therapy and life review for palliative care patients: A randomized controlled trial. *Journal of Pain and Symptom Management* **53**(2), 162–170.e1. doi:10.1016/j.jpainsymman.2016.09.005
- Welsch K and Gottschling S** (2021) Wishes and needs at the end of life: Communication strategies, counseling, and administrative aspects. *Deutsches Ärzteblatt International* **118**, 303–312. doi:10.3238/arztebl.m2021.0141
- Xiao J, Chow KM, Liu Y, et al.** (2019) Effects of dignity therapy on dignity, psychological well-being, and quality of life among palliative care cancer patients: A systematic review and meta-analysis. *Psycho-Oncology* **28**(9), 1791–1802. doi:10.1002/pon.5162
- Zigmond AS and Snaith RP** (1983) The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavica* **67**(6), 361–370. doi:10.1111/j.1600-0447.1983.tb09716.x