

# Dignity Therapy for People with Motor Neuron Disease and Their Family Caregivers: A Feasibility Study

Samar M. Aoun, BSc(Hons), MPH, PhD,<sup>1</sup> Harvey M. Chochinov, MD, PhD, FRSC,<sup>2</sup>  
and Linda J. Kristjanson, BN, MN, PhD<sup>3</sup>

## Abstract

**Background:** There are calls to explore psychological interventions to reduce distress in patients with motor neuron disease (MND) and their family caregivers. Dignity therapy is a short-term psychotherapy intervention shown to alleviate distress for people with life-limiting illnesses.

**Objectives:** To assess the acceptability, feasibility, and effectiveness of dignity therapy to reduce distress in people with MND and their family caregivers.

**Methods:** The study used a repeated-measures design pre- and post-intervention. Acceptability and feasibility were assessed using participants' ratings of the helpfulness of the intervention across several domains and time and resources required. Effectiveness measures for patients included: dignity-related distress, hopefulness, and spiritual well-being; and those for family caregivers included burden, hopefulness, anxiety, and depression.

**Results:** Twenty-seven patients and 18 family caregivers completed the intervention. Dignity therapy was well accepted, including those patients who required assisted communication devices. The feasibility may be limited in small or not well-resourced services. There were no significant differences in all outcome measures for both groups. However, the high satisfaction and endorsement of dignity therapy by patients suggests it has influenced various important aspects of end-of-life experience. Family caregivers overwhelmingly agreed that the dignity therapy document is and will continue to be a source of comfort to them and they would recommend dignity therapy to others in the same situation.

**Conclusions:** This is the first dignity therapy study to focus on MND and on home-based caregiving. Results established the importance of narrative and generativity for patients with MND and may open the door for other neurodegenerative conditions.

## Introduction

ALTHOUGH MOTOR NEURON DISEASE (MND) is a relatively rare disease, with an annual incidence of approximately 2 per 100,000, approximately half of patients die within 2.5 years of symptom onset and 1.2 years of diagnosis<sup>1,2</sup> and the burden of disease for the individual affected and family is substantial.<sup>3-5</sup> Disease progression is often rapid, with high levels of disability changing over months rather than years and the consequent need for support, including assistance with feeding, communication, movement, transferring, toileting, and other personal daily living tasks.<sup>6,7</sup> From diagnosis, people with MND experience relentless loss.<sup>8</sup> Previous research has shown that patients with MND

experience significantly more negative emotions, particularly hopelessness and helplessness, than patients with cancer.<sup>9</sup>

A comprehensive review of the literature revealed that the experience of diagnosis, assisted ventilation, cognitive changes, and end-of-life decision-making create challenges within a short time for MND caregivers, underscoring the need for supportive interventions.<sup>3</sup> People with MND and their families often describe their care experiences as unrelenting and worse than cancer because of the progressive nature of the disease and the hopelessness of recovery.<sup>4,10,11</sup>

To date, there are few nonpharmacologic interventions specifically designed to lessen the suffering or existential distress that patients experience toward the end of life.

<sup>1</sup>School of Nursing and Midwifery, Curtin University, Perth, Western Australia, Australia.

<sup>2</sup>Department of Psychiatry, University of Manitoba CancerCare Manitoba, Winnipeg, Manitoba, Canada.

<sup>3</sup>Swinburne University, Melbourne, Victoria, Australia.

Accepted September 4, 2014.

Dignity therapy is one such therapy that has been shown to alleviate distress in individuals with end-stage cancer<sup>12</sup> and in older patients<sup>13</sup> and has demonstrated benefits for family members by moderating their bereavement experience.<sup>14</sup> The randomized controlled trial (RCT) findings<sup>12</sup> showed that dignity therapy outperformed standard care and client-centered care in a palliative care population in which 96% had malignant conditions.

Dignity therapy is a novel, brief approach based on an empirically validated model of dignity in terminally ill people.<sup>15–17</sup> Patients are invited to discuss issues that matter most or that they would most want remembered about their life. Sessions are transcribed and edited with a final version (generativity document) returned to the patient, for the patient to bequeath to a family member or a friend, thus becoming part of a personal legacy.

In the MND literature, there is a paucity of research on development and implementation of psychological interventions. This feasibility study answers such calls to explore studies of existing psychological therapies as a step to reduce distress in patients with MND and their family caregivers.<sup>18</sup>

### Objective

Our study aimed to test the acceptability, feasibility, and effectiveness of dignity therapy for people living with MND patients and their family caregivers while still engaged in caregiving. The hypotheses were that: (1) dignity therapy would be acceptable and feasible for both patients and caregivers; (2) dignity therapy would reduce dignity-related distress and increase quality of life, hopefulness, and spiritual well-being in patients; (3) dignity therapy would decrease caregiver burden, depression, and anxiety and improve caregiver hopefulness.

### Methodology

Ethical approval was obtained from Curtin University Human Research Ethics Committee. The study design is cross-sectional using a single intervention group and repeated measures pretesting and posttesting.

### Setting and participants

Participants were recruited from the support organization, the MND Association of Western Australia (MNDWA) where they were registered as members (not all people with MND in Western Australia are members). All patients were diagnosed by neurologists. MNDWA sent out invitations to

patients and their family caregivers living in the metropolitan and rural areas of Western Australia (2011–2013). We planned to recruit 50 patients, and where available, their family caregivers.

### Intervention (see Table 1 for questions protocol<sup>19</sup>)

The therapy was provided by a psychologist who completed a training workshop with the therapy originator, Harvey Max Chochinov. At the first visit, the therapist reviewed the patient and family caregiver information and consent forms (which were mailed to participants prior to the visit) and sociodemographic and baseline outcome measures were collected from the patient and family caregiver. A dignity therapy session followed within 2–3 days, was tape recorded, and a verbatim transcript was prepared within 1–2 days. Another appointment was made to edit the transcript during which the participant was invited to make corrections, clarifications, or additions as desired. In the last dignity therapy visit, the final bound transcript was provided to the participant with as many copies as the participant requested. Post-testing occurred with both the participant and family caregiver 1 week after the final dignity therapy document has been provided. Posttesting questionnaires were sent out and returned by post to reduce the response bias.

### Eligibility criteria

The patient was eligible if he or she had a diagnosis of MND, was at least 18 years of age, able to read and speak English, and able to provide informed consent and has achieved, prior to the interview, a score of less than 15 on the Short Blessed Orientation-Memory-Concentration test (BOMC), which is a cognitive screening test.<sup>20</sup> Family caregivers needed to be at least 18 years of age, able to read and speak English, provide informed consent, and be involved in the patient's daily care.

The feasibility of facilitated communication with people with MND (keyboard, handwriting, communication board, etc.) was also explored.

### Outcomes for acceptability and feasibility

Outcomes for acceptability were measured through ratings of participants' views on whether the intervention has helped them and their family with response options ranging from strongly disagree to strongly agree. Patient feedback was elicited on their experience and included improvement in quality of life, spiritual well-being, greater sense of having control on one's own

TABLE 1. DIGNITY THERAPY QUESTIONS PROTOCOL<sup>19</sup>

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- Tell me a little about your life history, particularly the parts that you either remember most or are most important? When did you feel most alive?
  - Are there things that you would want your family to know or remember about you?
  - What are the most important roles you have played in your life (family, work, community service, etc.)? Why were they important to you and what did you accomplish?
  - What are your most important accomplishments, and what makes you feel most proud?
  - Are there things that you feel need to be said to your loved ones or things that you would want to say again?
  - What are your hopes and dreams for your loved ones?
  - What have you learned about life that you want to pass along to others? What advice or guidance would you wish to pass along to your child(ren), husband, wife, parents, other(s)?
  - Are there important words or instructions you would like to offer your family?
  - In creating this permanent record, are there other things that you would like included?
-

life, feeling more respected and understood, and having a heightened sense of dignity. Caregiver feedback was elicited on their experience in terms of a benefit to the patients and themselves, reduced sense of caregiver stress, greater sense of hope, and better preparation for end of life.

Outcomes for feasibility were measured through the number of visits by therapist, number of days to complete the therapy, time taken by therapist to deliver the therapy including interviewing, editing, and travel.

### Outcomes for effectiveness

The patient outcome measures for effectiveness were:

- Dignity-related distress as measured by Patient Dignity Inventory (PDI)<sup>21</sup> was the primary outcome. This 25-item measure evolved directly from the empirical model of dignity in the terminally ill<sup>17</sup> and corresponds to each of the models' themes and subthemes, including physical distress, social support, dependency, existential distress, and peace of mind. This measure has been shown to have good face, internal, test-retest, and concurrent validity (coefficient  $\alpha=0.93$ ). PDI has 5 response categories ranging from 1=not a problem to 5=an overwhelming problem. Higher scores indicate higher distress.
- Quality of life as measured by the Amyotrophic Lateral Sclerosis Assessment Questionnaire-5 (ALSAQ-5).<sup>22</sup> This is a disease-specific health-related quality of life instrument for use in studies of patients with MND/ALS, and has been found to be a valid measure similar to the longer version. It has 5 response categories from 0=never to 4=always. Higher scores indicate lower quality of life.
- Spiritual well-being as measured by the Functional Assessment of Chronic Illness Therapy-The 12-item Spiritual Well-Being Scale (FACIT-sp 12).<sup>23</sup> This is a brief self-report measure designed to assess the nature and extent of a person's spiritual well-being with two subscales (Faith, Meaning/Peace). It has strong internal reliability (coefficient  $\alpha=0.87$  for the total scale, 0.88 for the faith factor, and 0.81 for the meaning factor) and 5 response categories from 0=not at all to 4=very much. Higher scores indicate higher spiritual well-being.
- Hopefulness as measured by the Herth Hope Index (HHI).<sup>24</sup> This brief 12-item self-report measure of hope consists of three dimensions: temporality and future, positive readiness and expectance, and interconnectedness. HHI is reliable (coefficient  $\alpha=0.97$ ) and it has 4 response categories between 1=strongly disagree and 4=strongly agree. Higher scores indicate higher hopefulness.

The family caregiver outcome measures for effectiveness were:

- Caregiver burden as measured by the Zarit Burden Interview (ZBI-12) was the primary outcome.<sup>25-27</sup> The brief 12-item version has demonstrated comparable results to the full version (ZBI-22) and is reliable (coefficient  $\alpha=0.87$ ). It has 5 answer categories between 0=never and 4=nearly always. Higher scores indicate higher caregiver burden.
- Anxiety and depression as measured by The Hospital Anxiety and Depression Scale (HADS), which is a 14-

item instrument, structured as a four-point Likert scale, widely used as a screening tool for anxiety and depression in terminally ill people.<sup>28</sup> It is deemed reliable and valid with Cronbach's alpha for the anxiety subscale ranging from 0.68 to 0.93 and the depression subscale ranging from 0.67 to 0.90.<sup>29</sup> Higher scores indicate higher anxiety and depression. Cutoff scores of 8 and higher identify possible cases, 11 or higher identify probable cases.

- Herth Hope Index (as above for patients).

### Analyses

SPSS, version 22 (IBM SPSS, Armonk, NY) was the software package used for statistical analysis. Analyses were done on all participants with available data at baseline and at the end of the study intervention. Data were summarized using standard descriptive measures. Preintervention versus post-intervention comparisons were carried out using the paired *t* test if the data were normal or the Wilcoxon signed-rank test if it was not. All comparisons were carried out on a two-tailed basis.

### Results

#### Response rate and attrition

MNDAWA posted invitations to a total of 147 members of the association on three occasions between June 2011 and May 2013, with 35 clients responding (23.8% response rate). However, only 27 patients completed the study (23% attrition rate): 3 withdrew before consenting, 2 withdrew after consenting, 2 died before study completion, and 1 did not pass the cognitive screen. Eighteen family caregivers agreed to participate (9 patients did not have family caregivers or their caregivers did not participate either for lack of time or the patient did not want their partner included).

#### Demographic measures

Two-thirds of patients were male ( $n=18$ ), 82% were married ( $n=22$ ), and 26% lived in a rural area ( $n=7$ ). The mean age was 64.3 years (standard deviation [SD] 10.7), ranging from 32 to 81 years. Twenty-six percent of participants had been diagnosed for less than 1 year ( $n=7$ ), 33% for 1 to 2 years ( $n=9$ ), 15% for 2 to 3 years ( $n=4$ ), and 26% more than 4 years ( $n=7$ ).

Two-thirds of patients had participating family caregivers who were all spouses residing with the care recipient ( $n=18$ ); 72% were women ( $n=13$ ), with a mean age of 59.9 years (SD 11.8) ranging from 38 to 80 years; 56% of family caregivers reported spending 12 hours or more per day caring for their partners ( $n=10$ ).

#### Acceptability

The highest mean scores of acceptability (over 4) were achieved for patients in terms of: helpfulness of the intervention to them (with 88.9% agreeing or strongly agreeing) and to their families (81.5%); in their satisfaction with dignity therapy (92.6%); and their recommendation of dignity therapy to other patients (77.8%; Table 2). The majority of patients found dignity therapy helped them feel closer to their loved ones (70.4%) and gave them a sense of looking after unfinished business (66.7%; Table 2). The lowest mean scores of acceptability (3 or less) were achieved for patients

TABLE 2. ACCEPTABILITY OF DIGNITY THERAPY BY PATIENTS WITH MOTOR NEURON DISEASE ( $N=27$ ) AND A COMPARISON WITH A PREVIOUS STUDY

	Current study mean <sup>a</sup>	SD	% Agree/strongly agree <sup>b</sup>	2011 study <sup>c</sup> mean
Dignity therapy was helpful to me	4.27	0.604	88.9	4.23
Dignity therapy was helpful as any other health care aspect	3.54	0.859	59.3	3.63
Dignity therapy improved my quality of life	3.42	0.758	48.1	3.54
Dignity therapy has given me a sense of looking after unfinished business	3.73	0.533	66.7	3.35
Dignity therapy improved my spiritual wellbeing	3.38	0.637	44.4	3.27
Dignity therapy lessened my sense of sadness or depression	3.08	0.977	37.0	3.11
Dignity therapy lessened my sense of feeling a burden to others	2.92	0.935	25.9	2.81
Dignity therapy helped me feel more worthwhile and valued	3.54	0.761	51.9	3.38
Dignity therapy helped me feel like I am still me	3.69	0.884	63.0	3.81
Dignity therapy has given me a greater sense of having control over my life	3.19	0.749	33.3	3.02
Dignity therapy helped me accept the way things are	3.50	0.949	59.3	3.39
Dignity therapy made me feel more respected and understood by others	3.35	0.977	48.1	3.16
Dignity therapy made me feel I am still able to fill an important role	3.69	0.970	63.0	3.62
Dignity therapy was satisfactory	4.31	0.549	92.6	4.26
Dignity therapy made me feel life is more meaningful	3.58	0.643	55.6	3.55
Dignity therapy heightened my sense of purpose	3.35	0.797	44.4	3.49
Dignity therapy heightened my sense of dignity	3.38	0.852	55.6	3.52
Dignity therapy lessened my sense of suffering	3.31	0.736	44.4	2.86
Dignity therapy made patient feel more hopeful	3.00	0.849	33.3	—
Dignity therapy increased my will to live	2.96	0.978	29.6	2.94
Dignity therapy helped me feel closer to their loved ones	3.72	0.936	70.4	—
Dignity therapy has or will be of help to my family	4.08	0.702	81.5	3.93
Dignity therapy could change the way my family sees/appreciates me	3.52	1.046	59.3	3.58
Dignity therapy could change the way my health care providers see/appreciate me	3.23	0.951	37.0	—
I would recommend dignity therapy to other patients or family members who are dealing with MND	4.26	0.619	77.8	—

<sup>a</sup>Mean of scores: 1 = strongly disagree; 2 = disagree; 3 = neither agree nor disagree; 4 = agree; 5 = strongly disagree.

<sup>b</sup>Percent of those who agreed or strongly agreed with the statements.

<sup>c</sup>Chochinov et al.<sup>12</sup>;  $n=108$ .

SD, standard deviation; MND, motor neuron disease.

TABLE 3. ACCEPTABILITY OF DIGNITY THERAPY BY MOTOR NEURON DISEASE FAMILY CAREGIVERS ( $N=18$ )

	Mean <sup>a</sup>	SD	% Agree/strongly agree <sup>b</sup>
Dignity therapy was helpful to my family member	4.22	0.647	88.9
Dignity therapy has given my family member a heightened sense of purpose or meaning	3.78	1.060	55.6
Dignity therapy helped increase my family member's sense of dignity	3.56	0.984	44.4
Dignity therapy has helped prepare my family member for the end of life, whenever that may occur	3.33	0.970	50.0
Dignity therapy was as important as any other aspect of their care	3.61	0.979	61.1
Dignity therapy helped reduce my family member's suffering	3.22	1.003	38.9
Dignity therapy helped increase my family member's hopefulness about the future.	3.17	0.857	33.3
Dignity therapy document helped me during this time of our life	3.33	1.085	50.0
Dignity therapy helped me prepare for the end of life of my family member, whenever that may occur	3.11	0.832	27.8
Dignity therapy was helpful to me in reducing my feelings of stress as a family caregiver	3.00	0.907	33.3
Dignity therapy helped me feel closer to my family member	2.94	0.938	33.3
Dignity therapy increased my hopefulness about the future	3.11	0.758	33.3
Dignity therapy document will continue to be a source of comfort for my family and me	3.83	0.618	72.2
I would recommend dignity therapy to other patients or family members who are dealing with MND	4.00	0.686	77.8

<sup>a</sup>Mean of scores: 1 = strongly disagree, 2 = disagree, 3 = neither agree nor disagree, 4 = agree, 5 = strongly disagree.

<sup>b</sup>Number and percent of those who agreed or strongly agreed with the statements.

in terms of dignity therapy being helpful in increasing the will to live (29.6%), in lessening sense of feeling a burden to others (25.9%) and in improving hopefulness (33.3%; Table 2).

The majority of family caregivers felt that dignity therapy was helpful to their family member (88.9%) and more than 70% reported that the generativity document will continue to be a source of comfort to them and their families and they would recommend it to others dealing with MND. One-third felt that dignity therapy improved their feelings of hopefulness, that of their family member, or reduced their stress (33.3%); 50% felt it helped prepare them for end of life (Table 3).

**Feasibility**

Dignity therapy took between 3 and 7 visits to complete with an average of 4 visits per patient. The median duration in days to complete the therapy was 36 days, ranging between 14 and 113 days. A median of 4 copies were requested ranging from 1 to 20 copies and 59% requested an e-copy as well as a hard copy. The median number of pages was 19, ranging from 7 to 57 pages.

The therapist’s time to conduct the interview and edit, review, and hand over the document was as follows. Interviewing time: an average of 2 hours per patient per visit or a total of 8 hours for the average 4 visits. Editing time: 2 occasions of editing for an average of 2 hours per occasion or a total of 4 hours editing per transcript. Therefore, on average, 12 hours were needed to deliver the therapy per patient. Travel time: the therapist’s time was increased because of the travel to participants’ homes, particularly those in rural areas where seven patients lived approximately 200 to 300 kilometers from the city of Perth (or 400- to 600-kilometer return trips necessitating 4 to 6 hours driving and overnight stays). In addition, we need to factor in the transcription time and cost of a transcription service employed for the 27 transcripts.

**Potential effectiveness**

There were no significant changes pre- and post-intervention for patients in terms of measures on dignity related distress (primary outcome), quality of life, spiritual wellbeing and hopefulness (secondary outcomes; Table 4).

There were no significant pre/posttest changes for family caregivers in terms of measures on caregiver burden (primary

TABLE 5. POTENTIAL EFFECTIVENESS FOR CAREGIVERS: PRE- AND POST-INTERVENTION MEANS FOR CAREGIVER BURDEN, HOPEFULNESS, ANXIETY, AND DEPRESSION

Caregivers n=18	Mean	SD	p values
Pre-ZBI-12	12.76	8.012	
Post-ZBI-12	16.29	11.224	0.055
Pre-Herth hope	38.35	4.595	
Post-Herth hope	36.71	4.524	0.109
Pre-HADS anxiety	7.53	3.659	
Post-HADS anxiety	6.88	4.328	0.250
Pre-HADS depression	4.35	3.334	
Post-HADS depression	4.41	3.906	0.904

SD, standard deviation; ZBI-12, Zarit Burden Interview; HADS, Hospital Anxiety and Depression Scale.

outcome), hopefulness, anxiety, and depression (secondary outcomes). Both anxiety and depression scores were below 8, which is the cutoff score for possible cases (Table 5).

**Discussion and Conclusions**

Dignity therapy was well accepted by patients with MND and their family caregivers with nearly 90% of each group stating that dignity therapy was helpful. As presented in Table 2, the patients’ responses from this study were surprisingly comparable to those of the intervention group with cancer (n=108) of the RCT by Chochinov et al.<sup>12</sup> It is noteworthy that nearly 60% of patients and just more than 60% of caregivers reported dignity therapy was as important as any other aspect of their health care. As significant, approximately two-thirds of patients indicated that dignity therapy helped them attend to unfinished business, made them feel like they were still themselves, and that they were capable of filling an important role. Caregivers overwhelmingly agreed that the dignity therapy document is and will continue to be a source of comfort to them and nearly 80% said they would recommend dignity therapy to others in the same situation. To our knowledge this is the first study undertaken for people with MND and in particular assessing its usefulness for family caregivers while still engaged in home based caregiving rather than postbereavement. A previous study has focused on family members of the frail elderly resident in long-term care facilities.<sup>30</sup>

There were no significant differences in dignity-related distress levels between pre- and post-intervention for the patients. This may be because of the small sample size and also to the low base rates of distress, which precluded being able to demonstrate significant post-intervention improvements in the primary outcome measure of distress. It may well be that patients with MND who did not accept the invitation to participate were too ill and had much higher distress levels or the possibility of self-selection by participants who were in less distress. These findings mirror the ones reported by Chochinov et al.,<sup>12</sup> despite them enrolling a much larger sample size (326 patients with cancer) with 42% of assessed patients not eligible because they were too ill to participate. However, it is worth noting that the dignity-related distress overall score was higher for MND patients pre- and post-intervention (49.82–49.14) compared to that of residents of care homes (39.00–40.22).<sup>30</sup> For the secondary

TABLE 4. POTENTIAL EFFECTIVENESS FOR PATIENTS: PRE- AND POST-INTERVENTION MEANS FOR QUALITY OF LIFE, SPIRITUAL WELL-BEING, HOPEFULNESS, AND DIGNITY-RELATED DISTRESS

Patients n=25	Mean	SD	p values
Pre-ALSAQ 5	9.44	3.895	
Post-ALSAQ 5	9.28	3.770	0.735
Pre-FACIT Total	30.76	10.084	
Post-FACIT Total	31.04	9.628	0.822
Pre-Herth Hope	38.60	5.132	
Post-Herth Hope	36.76	6.540	0.207
Pre-PDI	49.82	15.723	
Post-PDI	49.14	12.833	0.679

SD, standard deviation; ALSAQ 5, Amyotrophic Lateral Sclerosis Assessment Questionnaire-5; FACIT, Functional Assessment of Chronic Illness Therapy; PDI, Patient Dignity Inventory.

outcome measure, hopefulness scores of patients did not differ from those of residents of care homes.<sup>13</sup> Caregivers' scores on hopefulness did not differ from those of care recipients, their anxiety and depression levels were low and stayed the same pre- and post-intervention, but they tended to feel more burdened at post-intervention presumably because of the quick progression of the disease. However, the possibility of the intervention itself causing additional burden cannot be ruled out and warrants investigation in future larger studies with a control group.

In general, it took longer to deliver the therapy to this population group (on average 12 hours per patient) compared to residents in care homes (11 hours)<sup>13</sup> and palliative care patients in a hospice (6.3 hours).<sup>31</sup> However, such time comparisons between studies may not be accurate because editing durations may fluctuate as people get better at doing the interviews, i.e., the material is better organized and easier to edit. Furthermore, the average duration of therapy from the initial interview to handing over the approved document was longer in this study, 36 days compared to 32 days in Hall et al.<sup>13</sup> and less than 14 days in the setting of oncology palliative care.<sup>12</sup> Longer completion times were mainly due to speech impairment, the patient going into the hospital for surgery or respite care, family and work commitments, and requesting more time to work on the document. The majority of family caregivers assisted with the interview and editing process, which extended the duration of the dignity therapy visits.

For a number of participating patients, while the loss of speech impacted the duration of the interview, it did not seem to affect the acceptability of the intervention, which was successfully completed using assisted communication devices, particularly the lightwriter ( $n=3$ ), and a combination of e-mail ( $n=3$ ), pen and paper ( $n=3$ ), and spousal assistance ( $n=3$ ). Several caregivers commented that it would have been better if dignity therapy was undertaken earlier in the diagnosis while the care recipient was still able to communicate more themselves. Eleven participants had suffered from the bulbar onset of the disease in which speech is affected early in the disease.

Results raise concerns regarding the feasibility of the therapy. Staff training, interviewing, editing, and transcribing as well as travel costs may make this too impractical for smaller organizations with limited resources. However, the therapy might be feasible for larger services who may consider offering dignity therapy in their setting.

The number of participants enrolled in this study was relatively small, but similar to samples reported in other feasibility studies. The qualitative analysis of the interviews will provide richer details on the experience of patients and caregivers with dignity therapy, which could inform other studies in the neurodegenerative field. Recruitment and retention of participants is particularly challenging when there is a quick progression in disability as is the case of MND. Hence it is recommended that dignity therapy is undertaken earlier in the disease trajectory. Despite that there were no demonstrable significant changes across standard measures, which may be a power issue or a floor effect (meaning it is hard to show improvements on items that are low to begin with), the high satisfaction and endorsement of dignity therapy suggests it has influenced various important aspects of end-of-life experience. It is worth noting that there are likely other less tangible effects of dignity therapy that need

to be further understood in view of the very strong recommendation of caregivers regarding others undertaking this therapy (78%). Therefore, the next step would be to shift attention towards sorting out an explanation for the salutary effects of dignity therapy. Another limitation of the study is the lack of a control group. To ascertain the effectiveness of dignity therapy for patients and family caregivers, future studies should consider an RCT design with a control group receiving a friendly visit or standard care, similar to Julião et al.<sup>32</sup> in which dignity therapy was shown to alleviate depression and anxiety in patients.

Most individuals with MND live at home, where their psychosocial functioning is inextricably tied to the extent and quality of support they receive from family members. Therefore, it is important to design and evaluate effective interventions and find ways to deliver them to families. Results from this study point to the value of narrative and generativity for patients with MND. Future studies, based on these insights, could lead to more streamlined interventional strategies for people with MND and those with other neurodegenerative conditions, where the disease progression may be relatively slower but have similar profound physical, social, and psychological consequences for both the individual and family.

#### Acknowledgments

This research is supported by a linkage grant from the Australian Research Council and the Motor Neurone Disease Association of Western Australia (LP0991305). Many thanks to the MNDAWA for assisting in the recruitment and to the patients and their family caregivers who generously contributed to this research despite their difficult circumstances. The research team acknowledges Dr. L. Oldham for contributing to the design of the project, Ms. B. Bentley for conducting the interviews, Ms. K. Deas for assisting with the data collection, and Ms. D. Howling for assisting with the analysis.

#### Author Disclosure Statement

No competing financial interests exist.

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Address correspondence to:  
 Samar M. Aoun, BSc(Hons), MPH, PhD  
 School of Nursing and Midwifery  
 Curtin University  
 GPO Box 1987  
 Perth  
 Western Australia 6845  
 Australia

E-mail: s.aoun@curtin.edu.au