

# 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>

- 
- 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-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

MNDWA 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

	<i>Current study mean<sup>a</sup></i>	<i>SD</i>	<i>% Agree/strongly agree<sup>b</sup></i>	<i>2011 study<sup>c</sup> mean</i>
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)

	<i>Mean<sup>a</sup></i>	<i>SD</i>	<i>% Agree/strongly agree<sup>b</sup></i>
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	0.055
Post-ZBI-12	16.29	11.224	
Pre-Herth hope	38.35	4.595	0.109
Post-Herth hope	36.71	4.524	
Pre-HADS anxiety	7.53	3.659	0.250
Post-HADS anxiety	6.88	4.328	
Pre-HADS depression	4.35	3.334	0.904
Post-HADS depression	4.41	3.906	

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	0.735
Post-ALSAQ 5	9.28	3.770	
Pre-FACIT Total	30.76	10.084	0.822
Post-FACIT Total	31.04	9.628	
Pre-Herth Hope	38.60	5.132	0.207
Post-Herth Hope	36.76	6.540	
Pre-PDI	49.82	15.723	0.679
Post-PDI	49.14	12.833	

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 MNDWA 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.

## References

1. Chancellor AM, Hendry A, Caird FJ, et al: Motor neuron disease: A disease of old age. *Scott Med J* 1993;38:178–182.
2. Bromberg MB: Quality of life in amyotrophic lateral sclerosis. *Phys Med Rehabil Clin N Am* 2008;19:591–605.
3. Aoun S, Bentley B, Funk L, et al: A 10-year literature review of family caregiving for motor neuron disease: Moving from caregiver burden studies to palliative care interventions. *Palliat Med* 2013;27:436–445.
4. Aoun S, Connors S, Priddis L, et al: Motor Neuron Disease family caregivers' experiences of caring, palliative care, and bereavement: An exploratory qualitative study. *Palliat Med* 2012;26:842–850.
5. Metropolitan Health and Aged Care Services Division, Victorian Government Department of Human Services: Motor Neurone Disease and Palliative Care: Interim Report on the MND Pathway Project. April 2008.
6. Oliver D, Aoun SM: What palliative care can do for motor neuron disease patients and their families? *Eur J Palliat Care* 2013;20:286–289.

7. Van Teijlingen ER, Friend E, Kamal AD: Service use and needs of people with motor neuron disease and their caregivers in Scotland. *Health Soc Care Community* 2001;9: 397–403.
8. Brown JB: User, caregiver and professional experiences of care in motor neuron disease. *Prim Health Care Res Dev* 2003;4:207–217.
9. Clarke DM, McLeod JE, Smith GC, et al: A comparison of psychosocial and physical functioning in patients with motor neuron disease and metastatic cancer. *J Palliat Care* 2005;21:173–179.
10. Locock L, Brown JB: All in the same boat? Patient and caregiver attitudes to peer support and social comparison in motor neuron disease (MND). *Soc Sci Med* 2010;71: 1498–1505.
11. O'Brien MR, Whitehead B, Jack BA, Mitchell JD: The need for support services for family caregivers of people with motor neuron disease (MND): Views of current and former family caregivers a qualitative study. *Disabil Rehabil* 2012;34:247–256.
12. Chochinov HM, Kristjanson L, Breitbart W, et al: Effect of DT on distress and end-of-life experience in terminally ill patients: A randomised controlled trial. *Lancet Oncol* 2011; 12:753–762.
13. Hall S, Goddard C, Opio D, et al: Feasibility, acceptability and potential effectiveness of DT for older people in care homes: A phase II randomized controlled trial of a brief palliative care psychotherapy. *Palliative Medicine* 2012;26: 703–712.
14. McClement S, Chochinov HM, Hack T, et al: DT: Family member perspectives. *J Palliat Med* 2007;10:1076–1082.
15. Chochinov H, Hack T, Hassard T, et al: DT: A novel psychotherapeutic intervention for patients near the end of life. *J Clin Oncol* 2005;23:5520–5525.
16. Chochinov H: Dignity conserving care: A new model for palliative care. *JAMA* 2002;287:2253–2260.
17. Chochinov H, Hack T, Hassard T, et al: Dignity in the terminally ill: A cross-sectional cohort study. *Lancet* 2002;360: 2026–2030.
18. Pagnini F, Simmons Z, Corbo M, Molinari E: Amyotrophic lateral sclerosis: Time for research on psychological intervention? *Amyotroph Lateral Scler* 2012;13:416–417.
19. Chochinov HM: *Dignity Therapy: Final Words for Final Days*. New York: Oxford University Press, 2012.
20. Katzman R, Brown T, Fuld P, et al: Validation of a short orientation-memory-concentration test of cognitive impairment. *Am J Psychiatry* 1983;140:734–739.
21. Chochinov HM, Hassard T, McClement S, et al: The Patient Dignity Inventory: A novel way of measuring dignity-related distress in palliative care. *J Pain Symptom Manage* 2008; 36:559–571.
22. Jenkinson C, Fitzpatrick R, Swash M, Jones G: Comparison of the 40-item Amyotrophic Lateral Sclerosis Assessment Questionnaire (ALSAQ-40) with a short-form five-item version (ALSAQ-5) in a longitudinal survey. *Clin Rehab* 2007;21:266–272.
23. Moadel A, Morgan C, Fatone A, et al: Seeking meaning and hope: Self-reported spiritual and existential needs among an ethnically diverse cancer patient population. *Psychooncology* 1999;8:1428–1431.
24. Buckley J, Herth K: Fostering hope in the terminally ill. *Nurs Stand* 2004;19:33–41.
25. Zarit SH: *The Hidden Victims of Alzheimer's Disease: Families Under Stress*. New York: New York University Press, 1985.
26. Bedard M, Molloy DW, Squire L, et al: The Zarit Burden Interview: A new short version and screening version. *Gerontologist* 2001;41:652–657.
27. Higginson IJ, Gao W, Jackson D, et al: Short-form Zarit Caregiver Burden Interviews were valid in advanced conditions. *J Clin Epidemiol* 2010;63:535–542.
28. Zigmond AS, Snaith RP: The Hospital Anxiety and Depression Scale. *Acta Psychiatr Scand* 1983;67:361–370.
29. Bjelland I, Dahl AA, Haug TT, Neckelmann D: The validity of the Hospital Anxiety and Depression Scale: An updated review. *J Psychosom Res* 2002;52:69–77.
30. Chochinov HM, Cann B, Cullihall, K, et al: DT: A feasibility study of elders in long-term care. *Palliat Support Care* 2012;10:3–15.
31. Montross L, Winters K, Irwin S: DT implementation in a community based hospice setting. *J Palliat Med* 2011;14: 729–734.
32. Julião M, Oliveira F, Nunes B, et al: Efficacy of dignity therapy on depression and anxiety in Portuguese terminally ill patients: A phase II randomized controlled trial. *J Palliat Med* 2014;17:688–695.

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

# Identifying and addressing the support needs of family caregivers of people with motor neurone disease using the Carer Support Needs Assessment Tool

SAMAR M. AOUN, PH.D.,<sup>1</sup> KATHLEEN DEAS, R.N.,<sup>1</sup> LINDA J. KRISTJANSON, PH.D.,<sup>2</sup> AND DAVID W. KISSANE, M.D.<sup>3</sup>

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

<sup>2</sup>Swinburne University, Melbourne, Victoria, Australia

<sup>3</sup>Faculty of Medicine, Nursing and Health Sciences, Monash University, Melbourne, Victoria, Australia

(RECEIVED August 14, 2015; ACCEPTED April 15, 2016)

## ABSTRACT

**Objective:** Family caregivers of people with motor neurone disease (MND) experience adverse health outcomes as a result of their caregiving experience. This may be alleviated if their support needs are identified and addressed in a systematic and timely manner. The objective of the present study was to assess the feasibility and relevance of the Carer Support Needs Assessment Tool (CSNAT) in home-based care during the period of caregiving from the perspectives of the family caregivers of people with MND and their service providers.

**Method:** The study was conducted during 2014 in Western Australia. Some 30 family caregivers and 4 care advisors participated in trialing the CSNAT intervention, which involved two visits from care advisors (6–8 weeks apart) to identify and address support needs. The feedback from family caregivers was obtained via telephone interviews and that of care advisors via a self-administered questionnaire.

**Results:** A total of 24 caregivers completed the study (80% completion rate) and identified the highest support priorities as “knowing what to expect in the future,” “knowing who to contact if concerned,” and “equipment to help care.” The majority found that this assessment process adequately addressed their needs and gave them a sense of validation, reassurance, and empowerment. Care advisors advocated the CSNAT approach as an improvement over standard practice, allowing them to more clearly assess needs, to offer a more structured follow-up, and to focus on the caregiver and family.

**Significance of Results:** The CSNAT approach for identifying and addressing family caregivers’ support needs was found to be relevant and feasible by MND family caregivers and care advisors. The tool provided a formal structure to facilitate discussions with family caregivers and thus enable needs to be addressed. Such discussions can also inform an evidence base for the ongoing development of services, ensuring that new and improved services are designed to meet the explicit needs of the family caregivers of people with a motor neurone disease.

**KEYWORDS:** Motor neurone disease, Amyotrophic lateral sclerosis, Carer support needs assessment tool (CSNAT), Support needs, Family caregivers, Service providers

## BACKGROUND

The significant psychological, social, and physical impact on family caregivers when providing home-based family caregiving for the terminally ill is well

Address correspondence and reprint requests to Samar M. Aoun, Curtin University, GPO Box U1987, Perth, Western Australia 6845, Australia. E-Mail: [s.aoun@curtin.edu.au](mailto:s.aoun@curtin.edu.au).



documented (Schulz & Beach, 1999; Aoun et al., 2005; Grande et al., 2009a; Stajduhar et al., 2010). Family caregivers' psychological outcomes can be improved if good support is received during caregiving (Ferrario et al., 2004; Grande et al., 2004; Kissane et al., 2006; Grande et al., 2009b). Identifying and addressing concerns early on leads to better health outcomes for carers (Grande et al., 2004; Grande et al., 2009a). However, adequate assessment of family caregivers' support needs by service providers is often informal due to the limited time available when their focus is primarily on the care recipient (Ewing et al., 2013).

Family caregivers of people with a motor neurone disease (MND) often describe their caring experiences as unrelenting due to the progressive nature of the disease and the relative hopelessness with respect to recovery (Locock & Brown, 2010; Aoun et al., 2012; O'Brien et al., 2012). Amyotrophic lateral sclerosis (ALS) is a progressive neurodegenerative MND with an incidence of 1–2 per 100,000 per year, a peak age at onset in the sixth decade of life, and a median survival of about 3.5 years from onset of symptoms (van Teijlingen et al., 2001; Leigh et al., 2003; Bromberg, 2008). People with an MND can progress rapidly to high levels of disability over a period of months rather than years, which intensifies their needs for support, including assistance with feeding, communication, movement, toileting, and other tasks of daily living (Oliver & Aoun, 2013).

Studies have reported that family caregivers suffer from anxiety, depression, strain, burden, fatigue, impaired quality of life, and reduced social contacts (Hecht et al., 2003; Chio et al., 2005; Goldstein et al., 2006; Aoun et al., 2013). While management of the physical symptoms in MND is paramount, attending to such family caregivers' psychosocial needs is crucial in order to prevent deterioration in their health outcomes (Goldstein et al., 2006; Oyebode et al., 2013). Most individuals with an MND live at home, where their psychosocial functioning is intimately connected to the extent and quality of support they receive from family members. Interventions to reduce caregiver burden and distress related to MND have been reported with varying success (Goldstein et al., 2006; Murphy et al., 2009; Aoun et al., 2014). It is thus important to design and evaluate effective interventions and find ways to deliver them to families living and caring for someone with a motor neurone disease (Pagnini et al., 2012; Aoun et al., 2013; Oliver & Aoun, 2013).

However, there is a lack of suitable tools for assessment of family caregivers' support needs during end-of-life home care (Hudson et al., 2010; Ewing & Grande, 2013), particularly for the period between diagnosis and end-of-life care (Goldstein et al., 2006; Oyebode et al., 2013).

The Carer Support Needs Assessment Tool (CSNAT) is a validated evidence-based tool used to identify family carer support needs in a systematic way, rather than employing the standard ad-hoc manner. As such, the tool also serves as a supportive carer intervention and, while carer-led, is facilitated by the health professional (Ewing et al., 2013; Ewing & Grande, 2013). The CSNAT adopts a screening format that is structured around 14 broad support domains. This format allows it to be brief but also comprehensive, enabling caregivers to identify the domains in which they require further support, which can then be discussed with health professionals. Each item represents a core family carer support domain in end-of-life home care, and these domains fall into two distinct groupings: those that enable the family caregiver to care and those that enable more direct support for themselves. There are 4 response options for each of the 14 CSNAT items that allow family caregivers to indicate the extent of their support requirements for each domain: "no more," "a little more," "quite a bit more," or "very much more" (Table 1). The health professional meets with the family caregiver to discuss priority needs and to formulate an action plan (as described in detail in the section on "The Intervention").

The CSNAT has been trialed using a stepped wedge cluster design within Silver Chain (a large community-based service provider in Western Australia) with 322 family caregivers of terminally ill

**Table 1.** Carer Support Needs Assessment Tool (CSNAT) domains (Ewing et al., 2013)

---

---

**Support that enables the family caregiver to care for the patient**

Do you need more support with:

- Understanding your relative's illness
- Knowing what to expect in the future when caring for your relative
- Managing your relative's symptoms, including giving medicines
- Providing personal care for your relative (e.g., dressing, washing, toileting)
- Knowing who to contact if you are concerned about your relative (for a range of needs, including at night)
- Equipment to help care for your relative
- Talking with your relative about his or her illness

**Support for the family caregiver in their caring role (more direct personal support)**

Do you need more support with:

- Having time for yourself in the day
  - Your financial, legal, or work issues
  - Dealing with your feelings and worries
  - Looking after your own health (physical problems)
  - Your beliefs or spiritual concerns
  - Practical help in the home
  - Getting a break from caring overnight
- 
-

people (mainly with cancer) and 44 nurses. The intervention group experienced a significant reduction in caregiver strain relative to controls ( $p = 0.018$ ,  $d = 0.35$ ) (Aoun et al., 2015b), and the feedback from family caregivers (Aoun et al., 2015a) and nurses (Aoun et al., 2015c) on using the CSNAT was positive. Although the CSNAT appeared to offer a practical approach to assessing and addressing family caregiver needs in cancer, it was deemed important to assess the extent to which the tool could be appropriate for use in other settings and among different disease groups. The present study was thus designed to implement and test the suitability of the CSNAT with family caregivers of people living with MND in the community across the entire spectrum of the caring experience, not only at the end of life.

## OBJECTIVE

Our aim was to assess the feasibility and relevance of the CSNAT in home-based care during the caregiving period from the perspectives of the family caregivers of people with MND and their service providers.

## METHODS

The study was conducted in Perth (in Western Australia (WA)) from April to July of 2014. It was approved by Curtin University Human Research Ethics Committee (SONM11-1014). All participants provided written informed consent to participate, and the ethics committee approved this consent procedure.

### Study Design

The study was designed to be descriptive and longitudinal. Family caregivers' support priorities were collected through the set of items on the CSNAT. Their feedback was obtained via semi-structured telephone interviews, while care advisors' feedback was gleaned from a self-administered questionnaire with open-ended questions. Feedback from both groups was obtained upon completion of the intervention (as described below). Family caregivers were considered to have concluded the study if they completed two CSNAT contacts with the care advisor (6 to 8 weeks apart).

### Participants

The study was conducted with primary family caregivers of clients of the Motor Neurone Disease Association of Western Australia (MNDWA) and their care advisors. This entity has in its database about 130 clients at various stages of disease progression, and the vast majority of people with MND in WA

are registered with the association. All adult caregivers (aged 18 years or older) who were caring at home and were able to read and write in English were eligible for the study, unless care advisors had concerns about a caregiver's ability to cope with research because of exceptionally high levels of distress. A primary family caregiver is defined as a person who, without payment, provides physical (and emotional) care to a person who is expected to die during the course of the period of caring. This care may be provided on a daily or intermittent basis.

The four care advisors working for the association were invited to participate. The standard practice of care advisors is to regularly visit clients at home, and their role involves complex case coordination, provision of disability aids and equipment, and delivery of information and facilitated support programs in order to enable people with MND to live as independently as possible for as long as possible. The care advisors attended a training session with the research team and had weekly contacts with the research officer, in line with previous work (Aoun et al., 2015b).

Participation in the feasibility study was voluntary for both groups, with no undue influence placed upon them. Family caregivers were assured that their decision would not in any way affect the supportive care they were then receiving or any care that they might receive in the future from any agency. Care advisors were also assured that their decision would not in any way affect their employment with the association.

### The Intervention

The intervention consisted of the following steps:

- the CSNAT was introduced to the family caregiver by the care advisor;
- the family caregiver was given time to consider which domains they required more support with;
- an assessment conversation took place where the care advisor and family caregiver discussed the domains where more support was needed to clarify the specific needs of the family carer, including what their top priorities were;
- a shared action plan was formulated where the family caregiver was involved in identifying the type of input they would find helpful (rather than delivery of the "standardized" supportive input that the service usually delivers);
- a shared review was planned within 6 to 8 weeks.

## Data Collection

The four care advisors working for the MND AWA, who regularly visit clients at home, introduced the study to the family caregivers who met the inclusion criteria, and they obtained written consent to trial the CSNAT and provide feedback to the researcher at the end of the trial. The care advisors collected the CSNAT data from the caregivers during their usual visits. For the purposes of this feasibility study, there was a baseline visit and then a follow-up visit within 6 to 8 weeks.

The researchers liaised regularly with care advisors during the data collection period to ensure that the research process was followed. They also collected feedback information from family caregivers after they had completed the study. Patient deaths were monitored with care advisors throughout the data collection period to ensure that recently bereaved family carers were not contacted by phone to complete the follow-up interview, which would have been insensitive.

Family caregivers were interviewed by an experienced research nurse, who telephoned at a prearranged time convenient for all (on average, within two weeks of completion of the intervention) to seek their feedback on the appropriateness, relevance, and benefit of the assessment process. Participants were given the opportunity to describe any other benefits or problems and ways of improving their experience of the CSNAT intervention. The questions asked at this phase were (as described in Aoun et al., 2015a):

- How easy or difficult was it for you to complete the CSNAT assessment of your support needs?
- Did you feel that completing the assessment process was helpful in getting the support you needed?
- Did this experience of identifying your needs affect what you did yourself?
- Did you feel that your needs as a carer were acknowledged/listened to in a way that was distinct from the needs of the patient?
- Do you think that the CSNAT assessment process could be improved in any way?

Care advisors preferred to give feedback by completing a self-administered questionnaire with open-ended questions in order to: (1) report on their experience with facilitating this process; (2) evaluate the benefits of or barriers to implementing the CSNAT with MND family caregivers; (3) suggest an optimal stage and time for administration and review; and (4) offer suggestions to assist with future

planning. Care advisors chose written feedback, as this method gave them time in their busy schedules to consider their answers and return the completed survey when convenient. An anonymous self-administered survey was sent to each care advisor and collected later by the researcher from the MND AWA office in a sealed envelope.

To get an indication of the disability of the care recipients and thus the burden this might pose for family caregivers, care advisors also completed a standard tool on the functional status of the person with MND using the Revised Amyotrophic Lateral Sclerosis Functional Rating Scale (ALSFRS-R), which has 12 items that assess activities of daily living (ADL) functions and changes in fine motor, gross motor, bulbar, and respiratory function (Cedarbaum et al., 1999). Higher scores are indicative of less impairment.

## Analysis

Descriptive statistics, using SPSS software (v. 22), were employed to describe the demographic characteristics of family caregivers and their support needs as identified by the CSNAT. Because of the small numbers involved, those reporting a need were grouped together (from “a little more” to “very much more”). Data from the interviews with caregivers were subjected to thematic analysis (Guest et al., 2012). Initial coding was carried out independently by the first two authors and was supported by NVivo 10 software. The interviews were not audiotaped, but meticulous notetaking allowed for verbatim transcripts. Transcribed interview notes were read and reread to identify keywords and key phrases, which were then grouped into categories labeled with codes. To enhance the credibility of our findings, the interviewer was involved in the analysis process so that consideration of the nonverbal context was assured. The major themes emerged after comparisons within and among individual interviews. These themes were initially identified independently, with differences resolved by discussion and returning to the data when necessary. Exemplars are provided herein to explain themes and describe how interpretations were reached.

The care advisors' written feedback data were subjected to content analysis by the first two authors following the same rigor, with responses grouped according to each survey item, ensuring that the context or explanation could be considered, and establishing overarching categories through comparison of content. Exemplars demonstrate how interpretations were reached (Hsieh & Shannon, 2005). Identified themes highlighted the relevance and feasibility issues for both groups.



## RESULTS

Some 30 family caregivers were recruited by 4 care advisors from the MNDWA, and 24 completed the study during the 4-month period (80% completion rate). Given the progressive nature of the neurological disease in this patient group, four patients died before the family caregiver completed the intervention. In addition, one carer declined due to her husband not wanting her to be involved any longer, and another went on an extended holiday and was not contactable. The final sample size was based on the number of clients visited regularly by the care advisors during the four-month study period and whose family caregivers met the selection criteria and agreed to participate.

Family caregivers completed two CSNAT forms (at a median interval of seven weeks), followed each time by a discussion about their support needs with the care advisors. For the first CSNAT contact, visits were face to face (79%) or by telephone (21%); for the second CSNAT contact, 46% of visits were face to face and 54% by telephone, in keeping with care advisors' usual practice. Feedback interviews by the researcher were undertaken on average 15.9 days after study completion, and interviews lasted an average of 12.4 minutes.

The majority of family caregivers found that the CSNAT form was easy to complete (83.3%), taking a median of 10 minutes (range = 3–20). All caregivers found the questions to be clear and appropriate, and they agreed that the instrument adequately addressed their support needs.

### Participants' Characteristics

The majority of family caregivers were female (75%), married (87.5%), and spouses/partners (79.2%), and 54% were retired. Their mean age was 63.8 years ( $SD = 12.9$ ) (Table 2). People with MND were predominately male (70.8%), with a mean age of 62.8 years ( $SD = 10.8$ ) and a median time since diagnosis of 20.5 months (range = 4–89). The ALS functional rating scale of fine motor, gross motor, bulbar, and respiratory function measured a median score of 27 (range = 9–46), indicating moderate functional impairment (Table 2).

Care advisors were female and had been working in the healthcare field in nursing or physiotherapy for 20 to 35 years and had worked as MND care advisors for 0.5 to 5 years.

### Family Caregivers' Support Needs and Provided Solutions

The top five support needs reported by family caregivers consisted of (Figure 1): (1) knowing what to expect

in the future (83%); (2) knowing who to contact if concerned (71%); (3) having equipment to assist in care (66%), (4) dealing with feelings and worries (58%); and (5) having time for themselves during the daytime (58%). When asked if there was anything else not addressed in CSNAT items (an item at the end of the form labeled "other"), one caregiver mentioned support "to communicate with other family members to help them cope with husband's illness and progressive decline." Another caregiver reported support "to communicate with wife, who lost her speech because of disease, and [caregiver] feels isolated from wife."

Care advisors documented their proposed solutions/action plans on the second half of the CSNAT form. The solutions put in place by the care advisors, in discussion with the caregiver, for "knowing what to expect in the future" consisted of discussions around end-of-life issues, advance health directives, future care, and the role of palliative care. For the second priority on "knowing who to contact if concerned," discussions centered around ambulance cover, referral to palliative care services, and a contact number at night and on weekends. For the third priority on "equipment to help care for your relative," information was provided on the association's equipment pool and the possibility of financial aid to rent equipment if required; a bedside commode was provided to aid with deteriorating mobility; and liaison with a disability service was made available in order to provide the next level of bathroom modifications. The solutions put in place for "dealing with your feelings and worries" consisted of information on various avenues for counseling and encouragement to attend the association's carer luncheon for social support. For the fifth priority on "having time for yourself in the day," care advisors liaised with service providers to increase the number of hours available for respite, discussed strategies for creating time for the caregiver, and encouraged caregivers to allow more people to help with relatives' care, giving caregivers more time for themselves.

### Family Caregivers' Experiences of the Assessment Process

Four themes emerged from the feedback interviews with family caregivers: (1) the overwhelming caregiver journey with MND; (2) CSNAT practicality and usefulness; (3) validation of the caregiver role and empowerment; and (4) reassurance of support.

#### *Theme 1: The Overwhelming Caregiver Journey with MND*

Feedback on the assessment process triggered caregivers to describe their overwhelming journey through



**Table 2.** Profile of family caregivers and people with MND (N = 24)

Family caregivers		<i>n</i>	%
Gender	Male	6	25.0
	Female	18	75.0
Age (years)	Mean ( $\pm$ SD)	63.8	$\pm$ 12.9
	Median (range)	66.5	(20, 80)
Marital status	Never married	1	4.2
	Divorced/separated	2	8.3
	Married	21	87.5
Cultural background	Australian	15	62.5
	British	7	29.2
	Other	2	8.3
Education	Primary	1	4.2
	Secondary	11	45.8
	Diploma/certificate/trade qualification	7	29.2
	Tertiary	5	20.8
Employment	Paid employment	7	29.2
	Retired/volunteer	13	54.2
	Household duties	3	12.5
	Other	1	4.2
Relationship to person with MND	Spouse or partner	19	79.2
	Parent	3	12.5
	Adult child	1	4.2
	Sibling	1	4.2
People with MND		<i>n</i>	
Gender	Male	17	70.8
	Female	7	29.2
Age (years)	Mean ( $\pm$ SD)	62.8	$\pm$ 10.8
	Median (range)	65.5	(38, 79)
Time since diagnosis (months)	Median (range)	20.5	(4, 89)
ALS functional rating scale	Median (range)	27	(9, 46)

the course of the disease. They often related their experience of personal stress:

I do have to go to see “a shrink”—It’s very stressful at times,” (FC18)

and shared how difficult they found coping with the losses brought about by MND:

They should bring in euthanasia— You wouldn’t put a dog through what MND does— I find it very difficult. It really rips you apart. (FC27)

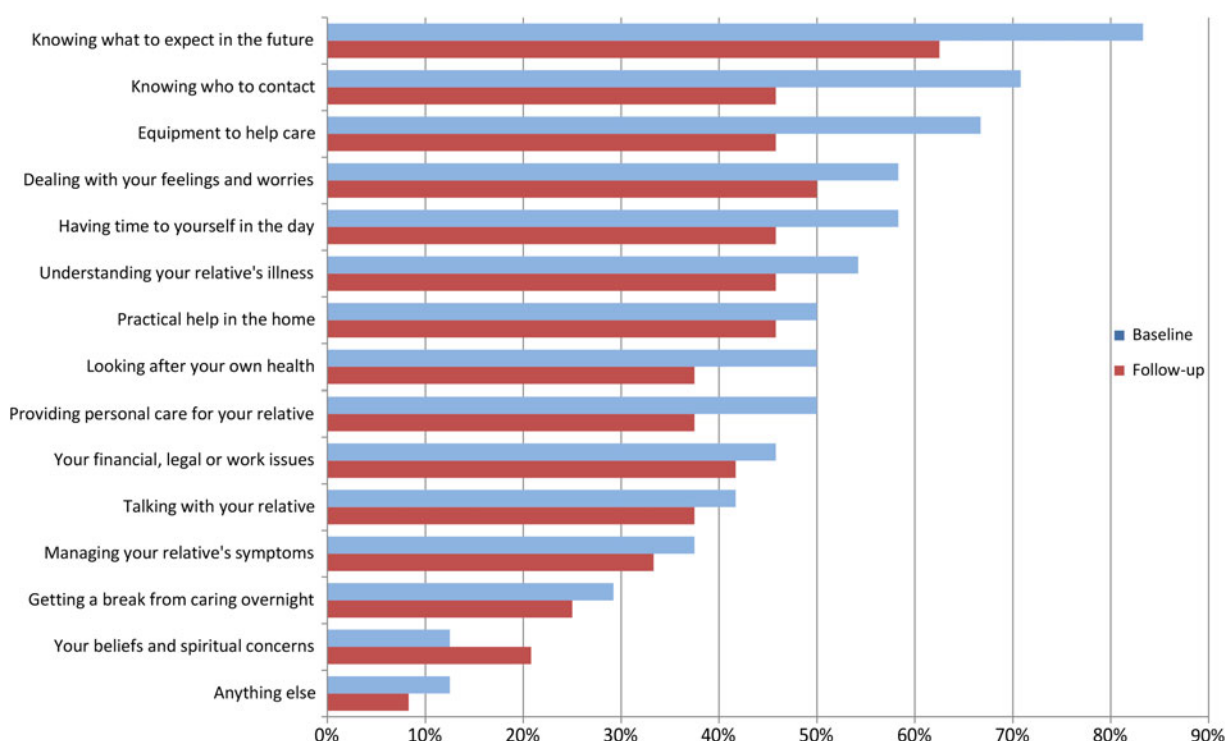
Expectations and acceptance of the personal demands of the caregiving journey were acknowledged by caregivers, as articulated by one participant:

Once you become a carer— “you have to throw part of yourself away.” I expected that. (FC14).

However, with the focus being primarily on the person with MND, the unmet needs of the caregiver were often missed:

I don’t think much of me— I have . . . been through breast cancer myself, and don’t need a lot. Yes, I did find it’s all “him, him, him.” I have come across that at times. I get a bit sick of it sometimes and think, “I’m here too!” (FC11)

I lost my partner 12 months ago, and I was his carer before, and now I’m caring for my son. It’s my whole



**Fig. 1.** Percentage of caregivers expressing need for more support with each Carer Support Needs Assessment Tool domain at baseline and follow-up ( $N = 24$ ).

life . . . I don't think of myself . . . I've got no issues really, but I mainly worry about how I'm going to cope. It's a terrible disease. (FC28)

Participants described being devastated by the hopelessness of the MND trajectory, comparing it to a cancer diagnosis, where there is often treatment and more support available and some hope of remission or recovery:

[The CSNAT] was very good. Maybe more detail about the fact that MND is terminal, unlike cancer, where some recover. (FC27)

Family caregivers commented on community's, friends', and health professionals' limited knowledge about the support available for people like them during the caregiving journey:

We're meeting people who have family and friends with MND, and they don't seem to know much about the MNDWA. Maybe the doctors don't tell them? When we go to the GP they say, "You know more about MND than I do." (FC11)

[I'm] looking at the support I need to give to other family members to help them cope with my husband's illness and progressive decline. (FC12)

## Theme 2: The CSNAT Practicality and Usefulness

Ease of completion of the CSNAT was considered important by family caregivers of persons living with MND, as they often have myriad forms to contend with. Caregivers described using the CSNAT as "Quite a good form— one of the better ones" (FC26), with one considering it essential to complete it by themselves: "I completed it on my own. You don't want someone else to influence what you need" (FC29).

Family caregivers appreciated the opportunity to rate their needs as listed in the CSNAT: "The scale was good to rate how much you needed, then the three [priorities] more thoroughly— was good to add more detail" (FC23).

The assessment process of working together with the care advisor was valued by caregivers:

The form was really well done . . . It puts those little stars up there to consider. It works with the two parts—the carer's answers and the care advisor's discussion— It can only work with the two together — It's very beneficial. (FC18)

The stage of MND trajectory and how this can affect the wide range of needs of the family caregiver were considered important when implementing the CSNAT, highlighting how these needs can change

rapidly. Some caregivers expressed this usefulness when their own needs changed as the disease progressed:

It was very easy. I was given the first form when it was early stages, and I didn't think I needed much. By the time of the second form, as it [MND] had progressed, my needs changed and the questions were more about what I needed then. It was helpful to talk to the care advisor about what she could do to help, going through the form together. (FC30)

As the disease progresses, you are more aware of symptoms. At first, you don't need much, and it would be "No" to nearly all [questions], but later it would be "Yes" to nearly all questions. (FC04)

The CSNAT was considered by caregivers as "a stimulus for conversation," prompting them to "think things through, and things to be put in place" (FC09):

It covered everything. Another box to say "not needed yet" would be helpful. I've been through the emotional stage, and now I'm in the practical stage and thinking about what needs to be done. (FC30)

### *Theme 3: Validation of the Caregiver Role and Empowerment*

It was evident that the assessment process could validate the caregiving role, as articulated by one participant:

The form made me think the role of carer was important— The fact they were being asked shows it is considered important ... The form shows some evidence someone is caring to ask. (FC18)

The CSNAT process allowed caregivers to reflect on what they needed or could do themselves:

It jogged me into thinking about what I might need—equipment, financial issues. The form had things I never even dreamed about needing. It made me realize what I can do at night if I need to call someone— I have a plan now, and I know I don't have to wait until the next morning. (FC30)

It focused your mind on issues and a method to address [them]— It's not something you can sweep under the carpet— An outcome resulted from going through the form. (FC12)

The CSNAT seemed to have helped when there were conflicting needs between family caregivers and pa-

tients, such as when caregivers felt restricted in accessing support for their own needs, as articulated by this participant:

[CSNAT] helped me to have counseling, and [the service] was helpful. My husband isn't wanting to be involved much. It can be a daunting process. (FC03)

The process of completing the CSNAT provided an opportunity for carers to consider their own needs when the focus was mostly directed toward patients:

Some of the questions I hadn't thought about. Yes, I think it was beneficial for me— This time it was, "Oh, this is about me!" (FC11)

The following participant wanted to go a step further and have the focus of the needs assessment to be specifically on the caregiver, in a way reaffirming the two domains that the CSNAT covered:

The distinction between the needs of the carer and the person cared for can sometimes be blurred. You as a carer tend to focus on "How can I improve my caregiving?" rather than looking at "What do I need as an individual?" Perhaps that can be accentuated—that this is looking specifically at you and your needs as a carer—distinct from the person cared for. (FC12)

### *Theme 4: Reassurance of Support*

A sense of relief was apparent when caregivers received the expertise and support provided by the MNDWA:

Care advisors see these people [with MND]— They know about the disease, whereas friends don't have an understanding of MND. So even just talking with the care advisors is a help. (FC25)

I found it very helpful— Yes, [the care advisor] was able to answer some of the questions straight away and explained what to do to get different things done. (FC30)

Completing the CSNAT assessment process involved discussion with the care advisor, which often prompted awareness by the caregiver of the need for support in patient symptom management. This was improved upon by family caregivers being encouraged to attend educational sessions at the MNDWA:

Definitely, especially from [the] MNDWA and the course they were doing [for caregivers]. It

increased my knowledge about the help available— Some things were a bit confronting, as we weren't at that stage. (FC03)

Participants were offered equipment or solutions to meet a particular need, as explained by one participant:

Yes, now I can help him out in many ways, but I can't lift, so that is the only thing I worry about. [The care advisor] is organizing a hoist for me, and that will help. (FC20)

I wasn't aware of all the equipment that was available— It's very good. I went along to a carer's lunch and was amazed at all the support available— We're so well looked after. The questions get you to think about things. You have a starting point and then can talk it through, and it gives you points you may not have considered. You have a rapport with the care advisor. (FC18)

Due to the potentially rapid deterioration associated with MND, end-of-life (EoL) issues are perhaps being considered earlier on in the disease trajectory than with other life-limiting diseases. The CSNAT can provide an opportunity to discuss this important issue where once it may have been overlooked or postponed:

One of the hardest [things] to discuss is EoL issues. [The CSNAT] focused my mind on the need to discuss this, and I ended up talking to people— I spoke to a counselor about EoL as a direct result of going through the survey. (FC12)

### *Care Advisors' Feedback*

All care advisors found the CSNAT format simple to complete and the questions easy to understand. They reported that the CSNAT helped identify issues that "perhaps would not have come up in a normal home visit or phone call," acknowledging that it had "been a springboard in several instances to allow a carer to explore their needs" (CA3), giving caregivers an opportunity to "verbalize their fears in a non-threatening way" (CA2), and "It made me realize that it paid to ask the question, even though I thought I sometimes knew what the answer might be" (CA1).

The CSNAT was considered by care advisors to assist in "providing a holistic approach to carers' needs" and was seen as highlighting the support provided for caregivers: "It does open doors for that ... It does let the carer be the focus of the support" (CA4). One care advisor explained, "It can uncover areas which may not have been recognized or adequately dealt with" (CA1), while another suggested that the CSNAT "acknowledges the important role carers play and the pressure put on them emotionally and physically" (CA2).

An important aspect of the CSNAT process was considered by care advisors to be "accountability and a documented record to assist the care provider" (CA3). They all advocated the CSNAT approach as an improvement over standard practice, as formalizing the process, and as providing a structured follow-up and a focus on the caregiver and family:

It is more comprehensive, provides a structured follow-up process, and there are aspects that are measurable. (CA3)

It formalizes the process and provides a means of documenting carers' needs. (CA4)

Consideration of caregiver and patient status and a sensitive approach were important when care advisors were introducing the CSNAT:

Finding the right time. There were instances when there had been an outpouring of issues. (CA1)

Sometimes when I planned to do it on a visit, it wasn't always appropriate (e.g., there were other pressing needs/issues). (CA4)

Using the CSNAT for regular reviews of caregiver needs was described by care advisors as offering "an opportunity to allow focus to be on carer rather than client— [and] allows them to have a safe place to recognize their needs, too" (CA4). An awareness of the changing and sometimes unpredictable needs of family caregivers was outlined as follows:

I think it is interesting to see how carer needs change over time and that their needs don't always follow the same trajectory as the person with MND. ... Sometimes what I perceive as a very stressful time for the carer they seem to sail through, whereas something minor [for me] at another time can unleash a great emotional tide for the carer. (CA1)

Another care advisor explained that it was useful to complete the CSNAT regularly,

because even if things don't change or deteriorate, it's again acknowledging their [family carers] needs. (CA2)



However, there is a conscious struggle to keep the focus on family caregivers along with the constantly pressing needs of care recipients:

If the time is right, the discussion points can have an immediacy that works very well. At times, though, even with the best intentions from all parties, it is often the person with MND whose needs are addressed first. It's good to be constantly reminding ourselves that the bigger picture of carer and family support have an equally important role. (CA1)

## DISCUSSION

Participants' involvement in this study provided them with an opportunity to share their difficult experiences, to gain increased insight into emotional concerns and enhanced awareness of supports, and to acknowledge their role as caregiver. In addition, participants described the benefits related to their increased timely access to support and better links to resources.

The CSNAT approach for identifying and addressing family caregivers' support needs was found relevant and acceptable by MND family caregivers and care advisors. For caregivers, a carer-led assessment process gave them a sense of validation, reassurance, and empowerment, as reflected in their narratives. Compared to standard practice, care advisors found the approach more comprehensive and formalized, similar to results found in previous studies using the CSNAT approach (Ewing & Grande, 2013; Aoun et al., 2015b; 2015a). It provided a structured follow-up process, a means of documenting caregiver needs, and a way to acknowledge their important role. The middle stage of the disease trajectory was suggested as when the CSNAT would best be administered for regular reviews because the time of receiving the diagnosis is highly emotional and the needs are not as easily identifiable in the early stages, when symptoms are not as advanced. By contrast, more changes occur toward the middle stages, and thus more care is required. The middle stages of the MND trajectory is the period when neurological symptoms are significantly developed and the person with MND requires more assistance from a family caregiver.

By structuring and reviewing caregiver needs two months apart, evidence was obtained of a steady reduction in their perception of needs, providing good evidence for the benefit of systematically repeating this review of needs using the CSNAT approach. The single domain that revealed a rise in need over time was caregivers' beliefs and spiritual concerns, which became more important over time. This could

reflect the benefit of caregiver reflection and recognition that a domain such as this can be valued.

Knowing what to expect as the illness progresses remained prominent for more than 60% at second follow-up, pointing to the continued need to educate and build the understanding that caregivers have about the future. A gradual educational process about care needs and about what to expect from the disease is clinically appropriate.

Communication issues are particularly important for people with MND and their family caregivers (Oliver & Aoun, 2013; Aoun et al., 2014) compared to most other life-limiting diseases. This is difficult for all types of MND, as deterioration occurs, but especially when symptoms include speech impairment, which suggests that health professionals need to integrate support with respect to all facets of communication for MND caregivers into their routine practice. Strain relating to loss of intimacy can be experienced by MND caregivers (Goldstein et al., 2006), as their partners' cognitive or physical ability to communicate diminishes, as evidenced in our and other studies (Oliver & Aoun, 2013; Oyebode et al., 2013), or if behavioral changes develop (Lillo et al., 2012). At another level, the needs of the broader family will likely depend upon the functioning of each group, their openness of communication, teamwork or cohesion, and their willingness to tolerate differences of opinion and remain mutually supportive (Kissane et al., 2006; Schuler et al., 2014). Communication issues were indeed raised in our study, but a larger national trial would be needed to warrant including a communication-related item to the CSNAT.

This is the first application of the CSNAT in an MND setting, a setting different from the one where it was developed in the United Kingdom (Ewing et al., 2013), and different from when it was further trialed in Australia (Aoun et al., 2015b) in home-based palliative care settings. In addition, the tool has been tested in our study earlier on in the caregiving journey and not just toward the end of life, a suggestion that was voiced in previous family caregiving research in the cancer field (Aoun et al., 2015a), and in MND (Aoun et al., 2014), where interventions were deemed beneficial earlier in the caregiving trajectory. Compared to caregivers who used the CSNAT in the cancer field (Aoun et al., 2015b), MND caregivers shared three of the top five priorities for support related mainly to direct carer support: "knowing what to expect in the future," "dealing with your feelings and worries," and "having time for yourself in the day." However, "knowing who to contact if you are concerned" and "needing equipment to help care" were more prominent priorities for MND caregivers in our study, reflecting the earlier timing in the caregiving journey, the rapidly progressive

nature of the disease, and the need to focus on practical help for patients.

## LIMITATIONS

Ours was a feasibility study with a small sample size, undertaken in one geographical location. Therefore, our findings cannot be generalized. Its limitations include the profiles of people who were not included in the study during the short period of data collection. The needs of caregivers who are still working; how single, separated, or divorced caregivers fare; and what special needs arise for parents, siblings, and children of patients with MND have not been explained in this cohort (Del Gaudio et al., 2012). Furthermore, care recipients were in the moderate stages of functional impairment, and there are significant challenges nearing the end of life. Such unaddressed needs specific to such circumstances could be explored in a larger national trial that would also ascertain the effectiveness of this assessment approach in improving caregivers' psychological outcomes in the MND setting.

While it may be considered a limitation in other contexts, eliciting written responses via a questionnaire from care advisors has worked well and has captured a breadth of opinion, in addition to being the care advisors' preferred form for providing feedback about their experience.

## CONCLUSIONS

Our results indicate that it is feasible to deliver this supportive intervention in the MND setting, particularly using the telephone to conduct follow-up assessments. Incorporating the CSNAT into the routine practice of MND care advisors would require minimal change in the structure of normal practice and minimal cost for the organization. Travel costs and interview times would not increase, as care advisors already visit patients and their family caregivers regularly, and follow-up assessment could be done by telephone, as per usual practice. Further inquiry into implementation in MND associations nationally and internationally is considered valuable, capitalizing on the systems already in place in these associations to provide support for family caregivers.

The CSNAT has provided a formal structure to facilitate discussions with family caregivers to enable needs to be addressed. Such discussions could also inform an evidence base for the ongoing development of services, thus helping to ensure that new or improved services are designed to meet the explicit needs of family caregivers of people with motor neurone disease.

## CONFLICTS OF INTEREST

The authors state that they have no conflicts of interest to declare.

## AUTHORS' CONTRIBUTIONS

SMA and KD conceived and designed the experiments. SMA and KD performed the experiments. SMA and KD analyzed the data. SMA, KD, LJK, and DWK interpreted the findings. SMA, KD, LJK, and DWK wrote the manuscript. All the authors read and approved the final manuscript.

## ACKNOWLEDGMENTS

The authors gratefully acknowledge the contribution of (1) the Motor Neurone Disease Association of Western Australia in helping to facilitate the project; (2) the care advisors in recruitment, data collection, and offering valuable advice; and (3) family caregivers in enriching the project with their feedback, given at a time of great difficulty. Many thanks are also offered to Ms. Denise Howting for assisting in the descriptive analysis.

## REFERENCES

- Aoun, S.M., Kristjanson, L.J., Currow, D.C., et al. (2005). Caregiving for the terminally ill: At what cost? *Palliative Medicine*, 19(7), 551–555.
- Aoun, S.M., Connors, S.L., Priddis, L., et al. (2012). Motor neurone disease family carers' experiences of caring, palliative care and bereavement: An exploratory qualitative study. *Palliative Medicine*, 26(6), 842–850.
- Aoun, S.M., Bentley, B., Funk, L., et al. (2013). A 10-year literature review of family caregiving for motor neurone disease: Moving from caregiver burden studies to palliative care interventions. *Palliative Medicine*, 27(5), 437–446.
- Aoun, S.M., Chochinov, H.M. & Kristjanson, L.J. (2014). Dignity therapy for people with motor neuron disease and their family caregivers: A feasibility study. *Journal of Palliative Medicine*, 18(1), 31–37.
- Aoun, S., Deas, K., Toye, C., et al. (2015a). Supporting family caregivers to identify their own needs in end-of-life care: Qualitative findings from a stepped wedge cluster trial. *Palliative Medicine*, 29(6), 508–517.
- Aoun, S.M., Grande, G., Howting, D., et al. (2015b). The impact of the Carer Support Needs Assessment Tool (CSNAT) in community palliative care using a stepped wedge cluster trial. *PLoS One*, 10(4), e0123012. doi: 10.1371/journal.pone.0123012. eCollection 2015.
- Aoun, S., Toye, C., Deas, K., et al. (2015c). Enabling a family caregiver-led assessment of support needs in home-based palliative care: Potential translation into practice. *Palliative Medicine*, 29(10), 929–938.
- Bromberg, M.B. (2008). Quality of life in amyotrophic lateral sclerosis. *Physical Medicine and Rehabilitation Clinics of North America*, 19(3), 591–605.
- Cedarbaum, J.M., Stambler, N., Malta, E., et al. (1999). The ALSFRS-R: A revised ALS functional rating scale that incorporates assessments of respiratory function. *Journal of the Neurological Sciences*, 169(1–2), 13–21.

- Chio, A., Gauthier, A., Calvo, A., et al. (2005). Caregiver burden and patients' perception of being a burden in ALS. *Neurology*, 64(10), 1780–1782.
- Del Gaudio, F., Zaider, T.I., Brier, M., et al. (2012). Challenges in providing family-centered support to families in palliative care. *Palliative Medicine*, 26(8), 1025–1033.
- Ewing, G. & Grande, G. (2013). Development of a Carer Support Needs Assessment Tool (CSNAT) for end-of-life care practice at home: A qualitative study. *Palliative Medicine*, 27(3), 244–256.
- Ewing, G., Brundle, C., Payne, S., et al. (2013). The Carer Support Needs Assessment Tool (CSNAT) for use in palliative and end-of-life care at home: A validation study. *Journal of Pain and Symptom Management*, 46(3), 395–405.
- Ferrario, S.R., Cardillo, V., Vicario, F., et al. (2004). Advanced cancer at home: Caregiving and bereavement. *Palliative Medicine*, 18(2), 129–136.
- Goldstein, L.H., Atkins, L., Landau, S., et al. (2006). Predictors of psychological distress in carers of people with amyotrophic lateral sclerosis: A longitudinal study. *Psychological Medicine*, 36(06), 865–875.
- Grande, G.E., Farquhar, M.C., Barclay, S.I.G., et al. (2004). Caregiver bereavement outcome: Relationship with hospice at home, satisfaction with care, and home death. *Journal of Palliative Care*, 20(2), 69–77.
- Grande, G., Stajduhar, K., Aoun, S., et al. (2009a). Supporting lay carers in end-of-life care: Current gaps and future priorities. *Palliative Medicine*, 23(4), 339–344.
- Grande, G.E., Ewing, G. & National Forum for Hospice at Home (2009b). Informal carer bereavement outcome: Relation to quality of end-of-life support and achievement of preferred place of death. *Palliative Medicine*, 23(3), 248–256.
- Guest, G., MacQueen, K.M. & Namey, E. E. (2012). *Applied thematic analysis*. London: Sage.
- Hecht, M.J., Graesel, E., Tigges, S., et al. (2003). Burden of care in amyotrophic lateral sclerosis. *Palliative Medicine*, 17(4), 327–333.
- Hsieh, H.F. & Shannon, S.E. (2005). Three approaches to qualitative content analysis. *Qualitative Health Research*, 15(9), 1277–1288.
- Hudson, P.L., Trauer, T., Graham, S., et al. (2010). A systematic review of instruments related to family caregivers of palliative care patients. *Palliative Medicine*, 24(7), 656–668.
- Kissane, D.W., McKenzie, M., Bloch, S., et al. (2006). Family focused grief therapy: A randomized, controlled trial in palliative care and bereavement. *The American Journal of Psychiatry*, 163(7), 1208–1218.
- Leigh, P.N., Abrahams, S., Al-Chalabi, A., et al. (2003). The management of motor neurone disease. *Journal of Neurology, Neurosurgery, and Psychiatry*, 74(Suppl. 4), iv32–iv47.
- Lillo, P., Mioshi, E. & Hodges, J. (2012). Caregiver burden in amyotrophic lateral sclerosis is more dependent on patients' behavioral changes than physical disability: A comparative study. *BMC Neurology*, 12(1), 156.
- Locock, L. & Brown, J.B. (2010). "All in the same boat"? Patient and carer attitudes to peer support and social comparison in motor neurone disease (MND). *Social Science & Medicine*, 71(8), 1498–1505.
- Murphy, V., Felgoise, S.H., Walsh, S.M., et al. (2009). Problem solving skills predict quality of life and psychological morbidity in ALS caregivers. *Amyotrophic Lateral Sclerosis*, 10(3), 147–153.
- O'Brien, M.R., Whitehead, B., Jack, B.A., et al. (2012). The need for support services for family carers of people with motor neurone disease (MND): Views of current and former family caregivers a qualitative study. *Disability and Rehabilitation*, 34(3), 247–256.
- Oliver, D. & Aoun, S. (2013). What palliative care can do for motor neurone disease patients and their families? *European Journal of Palliative Care*, 20(6), 286–289.
- Oyebode, J.R., Smith, H.J. & Morrison, K. (2013). The personal experience of partners of individuals with motor neuron disease. *Amyotrophic Lateral Sclerosis & Frontotemporal Degeneration*, 14(1), 39–43.
- Pagnini, F., Simmons, Z., Corbo, M., et al. (2012). Amyotrophic lateral sclerosis: Time for research on psychological intervention? *Amyotrophic Lateral Sclerosis*, 13(5), 416–417.
- Schuler, T.A., Zaider, T.I., Li, Y., et al. (2014). Typology of perceived family functioning in an American sample of patients with advanced cancer. *Journal of Pain and Symptom Management*, 48(2), 281–288.
- Schulz, R. & Beach, S.R. (1999). Caregiving as a risk factor for mortality: The caregiver health effects study. *The Journal of the American Medical Association*, 282(23), 2215–2219.
- Stajduhar, K., Funk, L., Toye, C., et al. (2010). Part 1. Home-based family caregiving at the end of life: A comprehensive review of published quantitative research (1998–2008). *Palliative Medicine*, 24(6), 573–593.
- van Teijlingen, E.R., Friend, E. & Kamal, A.D. (2001). Service use and needs of people with motor neurone disease and their carers in Scotland. *Health & Social Care in the Community*, 9(6), 397–403.

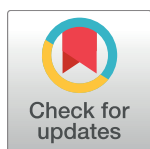
RESEARCH ARTICLE

# Bereavement support for family caregivers: The gap between guidelines and practice in palliative care

Samar M. Aoun<sup>1\*</sup>, Bruce Rumbold<sup>2</sup>, Denise Howting<sup>1</sup>, Amanda Bolleter<sup>3</sup>, Lauren J. Breen<sup>4</sup>

**1** School of Nursing, Midwifery and Paramedicine, Faculty of Health Sciences, Curtin University, Perth, Western Australia, Australia, **2** Palliative Care Unit, Department of Public Health, La Trobe University, Melbourne, Victoria, Australia, **3** WA Cancer and Palliative Care Network, Health department of WA, Perth, Western Australia, **4** School of Psychology and Speech Pathology, Faculty of Health Sciences, Curtin University, Perth, Western Australia, Australia

\* [s.aoun@curtin.edu.au](mailto:s.aoun@curtin.edu.au)



## Abstract

### Background

Standards for bereavement care propose that support should be matched to risk and need. However, studies in many countries demonstrate that palliative care services continue to adopt a generic approach in offering support to bereaved families.

### Objective

To identify patterns of bereavement support in palliative care services based upon the experience of bereaved people from a population based survey and in relation to clinical practice guidelines.

### Design

An anonymous postal survey collected information from clients of six funeral providers in four Australian states (2014–15), 6 to 24 months after the death of their family member or friend, with 1,139 responding. Responses from 506 bereaved relatives of people who had terminal illnesses were analysed. Of these, 298 had used palliative care services and 208 had not.

### Results

More people with cancer (64%) had received palliative care in comparison to other illnesses such as heart disease, dementia and organ failure (4–10%). The support for family caregivers before and after their relative's death was not considered optimal. Only 39.4% of the bereaved reported being specifically asked about their emotional/ psychological distress pre-bereavement, and just half of the bereaved perceived they had enough support from palliative care services. Half of the bereaved had a follow up contact from the service at 3–6 weeks, and a quarter had a follow-up at 6 months. Their qualitative feedback underlined the

## OPEN ACCESS

**Citation:** Aoun SM, Rumbold B, Howting D, Bolleter A, Breen LJ (2017) Bereavement support for family caregivers: The gap between guidelines and practice in palliative care. PLoS ONE 12(10): e0184750. <https://doi.org/10.1371/journal.pone.0184750>

**Editor:** Jacobus P. van Wouwe, TNO, NETHERLANDS

**Received:** May 5, 2017

**Accepted:** August 30, 2017

**Published:** October 4, 2017

**Copyright:** © 2017 Aoun et al. This is an open access article distributed under the terms of the [Creative Commons Attribution License](https://creativecommons.org/licenses/by/4.0/), which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

**Data Availability Statement:** Ethical approval specifies that all results are in aggregate form to maintain confidentiality and privacy and precludes individual level data being made publicly available. Interested and qualified researchers may send requests for data to Samar Aoun at [s.aoun@curtin.edu.au](mailto:s.aoun@curtin.edu.au).

**Funding:** The authors acknowledge the financial support of the WA Cancer and Palliative Care Network, Health Department of Western Australia. The funders had no role in study design, data



collection and analysis, decision to publish, or preparation of the manuscript.

**Competing interests:** The authors have declared that no competing interests exist.

limited helpfulness of the blanket approach to bereavement support, which was often described as “not personal” or “generic”, or “just standard practice”.

## Conclusions

Timeliness and consistency of relationship is crucial to building rapport and trust in the service’s ability to help at post-bereavement as well as a focus on the specific rather than the generic needs of the bereaved. In light of these limitations, palliative care services might do better investing their efforts principally in assessing and supporting family caregivers during the pre-bereavement period and developing community capacity and referral pathways for bereavement care. Our findings suggest that bereavement support in Australian palliative care services has only a tenuous relationship with guidelines and assessment tools, a conclusion also drawn in studies from other countries, emphasizing the international implications of our study.

## Introduction

In many countries, the most coherent approach to bereavement support is provided by palliative care services, which emphasise the care of patients with terminal illnesses and their family carers before and after the patient’s death. Policies and guidelines on standards of care propose that supports should be offered according to need [1–4]. However, despite such policies and guidelines, studies demonstrate that palliative care services in general adopt a blanket approach to supporting bereaved families regardless of risk or need [5–7]. For instance, one survey of Australian palliative care services determined that 95% (of 236 services) offered some form of bereavement support [8], with the most common types of support being a telephone call (offered by 86%), memorial service, (66%), letter (55%), anniversary card (53%), group sessions (31%), information package (5%), and informal gatherings (4%). Similar eclectic approaches appear to be taken in other countries such as the United States, Canada, the United Kingdom, and Japan [9]. A recent survey of bereavement support practice in 25 European countries (370 palliative care services) [10], showed that bereavement care was not an integral part of palliative care in a substantial number of palliative care services; more than two thirds of services did not use formal guidelines or standards to inform their bereavement care; while only about half the services employed a full time, part time or hourly bereavement coordinator or provided formal training in bereavement care.

Furthermore, where risk assessments were made, the usefulness of these assessments varied widely and often depended upon the subjective opinion of service providers or the use of non-validated screening tools [8, 11]. Approximately two-thirds of the services reported engaging in some form of bereavement risk assessment, with two-thirds relying on multidisciplinary team opinion, more than half using a formal tool to assess bereavement risk, and approximately half relying on a single staff member’s opinion (some services reported using more than one method) [8].

The situation is complex as there is no clear evidence to guide the development and allocation of cost-effective bereavement support services [8, 9, 12]. A core dilemma in making bereavement risk assessments is that much of our current evidence comes from groups of bereaved people who have self-referred, either to a community counselling service or in response to an invitation from a palliative care service [13]. The resultant focus on professional

support obscures alternative strategies that could be employed. Accordingly, we know less about bereavement experiences and needs of people who did not use palliative care services following an expected death, and considerably less again about the bereavement experiences of those for whom the death was unexpected.

We can, however, see that offering professional bereavement support, irrespective of need, to all people bereaved through deaths in palliative care is neither effective nor affordable [8, 9, 12, 14]. In Australia, 159,052 deaths were registered in 2015 [15], with the cause of death for at least two-thirds being a chronic degenerative illness. About 35,000 were in receipt of palliative care [16], but many more could have been eligible. For example, a large, retrospective cohort study in one Australian state reported that only about 60% of the population, whose dying should have been amenable to palliative care, actually received palliative care services [17].

Recognising these challenges, we embarked upon a population based survey of bereavement experiences and support needs to inform practice and develop a model of care that might apply to a whole community, not only to clients of a palliative care service. The three-tiered public health model of bereavement support we developed aligns intervention with need and is compatible with bereavement care standards and policies [12]. This model identifies three risk groups: a low risk group of bereaved people that are likely to adjust in time with appropriate support from family and friends; a moderate risk group that would benefit from grief counselling or a volunteer-led, or peer support group, to prevent the development of ongoing complications; and a high risk group that would most likely require formal support from health professionals. Empirical support for this model has been demonstrated [18]. This model challenges current palliative care bereavement provision in two ways. It suggests that the bulk of bereavement support should be located in local communities, via people's existing social networks [19], and it questions strategies for offering bereavement care that pre-dispose bereaved people to look for support in professionalised health services, such as palliative care, more than their local community.

## Aim

The aim of this study is to identify patterns of bereavement support in palliative care services based upon the experience of bereaved people from a population based survey and in relation to clinical practice guidelines.

## Objectives

The objectives are:

- To compare the profile of those who were cared for by a palliative care service with those who did not
- To compare the profile of the bereaved whose family member used palliative care services with those who did not
- To compare the bereavement support reported by the bereaved in relation to the clinical practice guidelines
- To describe which bereavement support practices were found helpful, or not helpful, by the bereaved
- To compare sources of bereavement support reported by those who did and did not receive palliative care

## Methods

Ethics approval was granted by Curtin University Human Research Ethics Committee (HR-57/2012).

An anonymous postal survey was used to collect information from clients of six funeral providers in four Australian states (2014–15), 6 to 24 months after the death of their family member or friend. A total of 6,258 study packages were delivered to the six funeral providers who agreed to participate in the study. These packages contained an invitation letter addressed from the funeral provider to the family, information sheet, the questionnaire, a list of support services for the family to use in case the participant became distressed while completing the questionnaire, and a reply-paid envelope. The funeral providers selected from their databases clients who were bereaved 6–24 months ago, attached names and address labels on the envelopes and mailed the study packages. Consent was implied by the return of the completed survey. No reminder letter was sent as the funeral providers felt it was too intrusive for the bereaved families. Clients were eligible to participate in the study if they had been bereaved by the death of a close family member or friend in the specified timeframe, were able to read, understand and write in English, and were over 18 years of age.

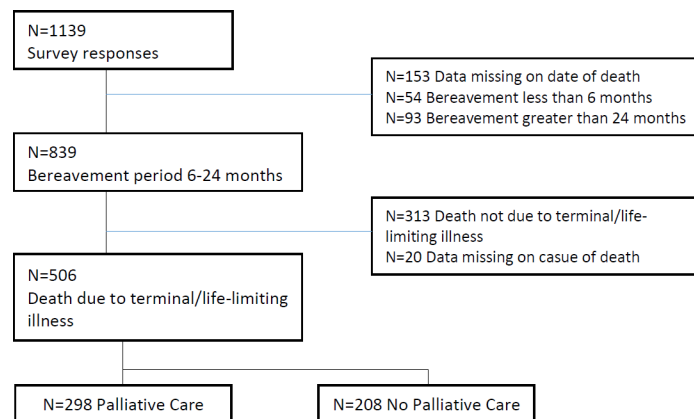
The questionnaire comprised 82 questions divided into 8 sections [20]. This present article focuses mainly on the section addressing the bereavement support received from a palliative care provider. At the start of this section, a lay definition of palliative care was provided so respondents were clear about whether they needed to complete this section: “*Palliative care is provided to patients with life-limiting/ terminal illness to ease symptoms and improve quality of life and support their families*”. Some of the questions in this section were aligned with the clinical practice guidelines for the psychosocial and bereavement support of family caregivers of palliative care patients [21]. Although these 20 guidelines were framed from a service provider perspective, we asked the questions from the bereaved people’s perspective regarding five of these guidelines (numbers 8, 12, 13, 17, 19). Questions on perceived support were adapted from the VOICES survey [22] and one question on level of caring was adapted from the Omnibus survey [23].

## Analysis

Descriptive statistics for variables were calculated: frequencies and proportions for categorical variables; means, standard deviations, medians, minimums and maximums for continuous/discrete variables. Significance testing was performed using chi-square for categorical variables, and nonparametric tests for the median for the non-Normally distributed continuous variables. Significance was set at the  $p = 0.05$  level, and analyses were conducted using IBM SPSS Statistics Version 24. The open ended responses were manually coded using an open content analysis process [24].

## Results

One thousand one hundred and thirty nine individuals completed the survey with a mean response rate of 18.1% (ranging from 13.3% to 28.6% between the six funeral providers). Those who had shorter or longer bereavement period than specified a priori; those who did not die from a terminal illness; and those who did not provide a date of death or cause of death were removed from the analysis. The sample was consequently reduced to 506 individuals. Of these, 298 had experience with palliative care services (PC) and 208 did not (NPC) (Fig 1).



**Fig 1. Flowchart of participation.**

<https://doi.org/10.1371/journal.pone.0184750.g001>

## Profile of the deceased

Half of the total deceased were female with a mean age of 77 years (SD 13.9), and those in the NPC group were slightly older (79 vs 76 years). Other characteristics that differed significantly between those who received palliative care versus those who did not were the type of terminal illness: the PC group had predominantly cancer (64.1% vs 26.4%,  $p < 0.001$ ) and the NPC group had more heart disease, dementia, lung disease and organ failure (Table 1). The PC group were significantly more likely to have an advance care plan (17.4 vs 6.1%,  $p < 0.001$ ) or an advance health directive (13.9 vs 7.1%,  $p < 0.006$ ) than the NPC group.

## Profile of the bereaved

The majority of the bereaved who responded were female (70%), had mean age of 64 years (SD 11.5), 42% were married and 44% were spouses of the person who had died. The two groups differed in some characteristics: the NPC group had more 'other relatives' who have provided care than members of the immediate family, the median length of caring was twice as long (48 vs 24 months), care was described as not hands on but still close, while more day to day hands on care was provided by the immediate family members in the PC group (Table 2).

## Experience of support pre- and post-death

The deceased received palliative care during their illness for a mean 3.7 months or median 1.0 month (range 0.03–96). Just over half of the bereaved felt that the care received by their

**Table 1. Profile of the deceased (%).**

	Total	Palliative Care		p-value
	n = 506	PC	NPC	
Gender (Female)	49.0	47.5	51.4	NS
Mean Age in years (SD)	77.2 (13.85)	76.0 (14.37)	79.0 (12.89)	0.015
Advance care plan	12.7	17.4	6.1	<0.001
Advance health directive	11.9	13.9	7.1	0.006
Cancer	48.6	64.1	26.4	<0.001
Heart disease	13.8	10.4	18.8	0.008
Dementia	12.5	8.1	18.8	0.004
Lung disease	6.1	4.0	9.1	0.006
Organ failure	11.5	7.7	16.8	0.001

<https://doi.org/10.1371/journal.pone.0184750.t001>



**Table 2. Profile of the bereaved (%).**

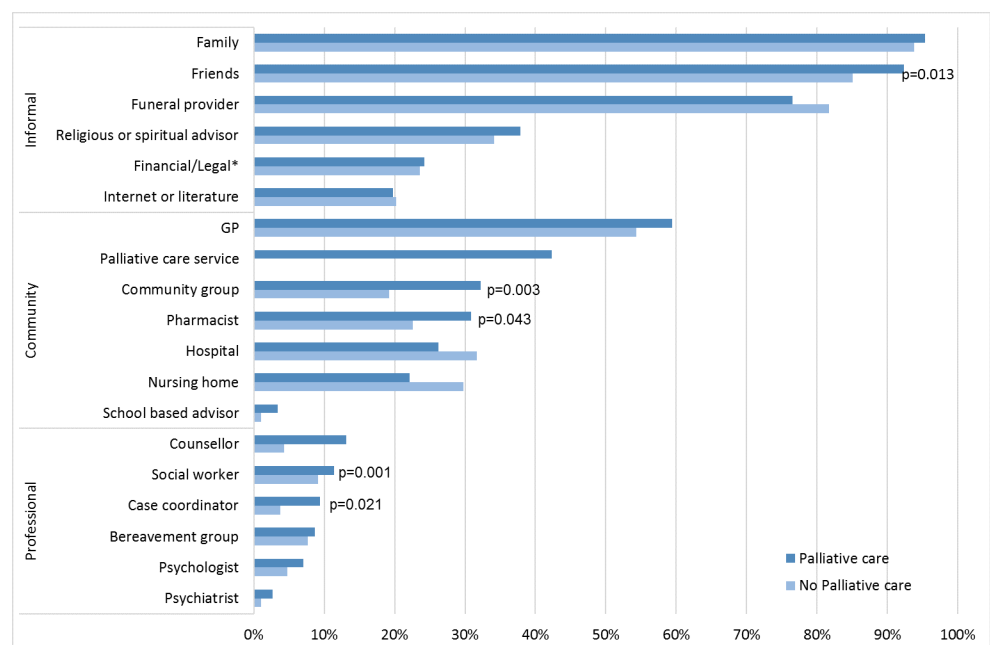
	Total	Palliative Care		p-value
	n = 506	PC	NPC	
Gender (Female)	69.8	72.7	66.2	NS
Mean Age in years (SD)	63.9 (11.46)	63.2 (11.58)	64.8 (11.26)	NS
Marital status (Married)	45.3	42.4	50.0	NS
Relationship (Spouse)	40.9	44.3	36.1	NS
Relationship (Other relative)	9.7	6.7	13.9	<0.001
Day to day hands on care	50.0	56.6	43.2	<0.001
No hands on care but still close	13.4	8.8	21.1	<0.001
Median length of care (month)	36	24	48	<0.001

<https://doi.org/10.1371/journal.pone.0184750.t002>

relative/friend from the palliative care service in the last 3 months of life was excellent (53.1%), 31.0% felt it was good, 9.5% fair, and 4.4% thought the service was poor.

Of the group who received support from palliative care providers after the death of their relative/friend, 51.2% felt they got as much help and support as they needed, a third did not feel they received enough support and 13.2% stated that they did not need support. When asked their opinion of the support they received from all sources (health and community services and networks), a higher proportion, 68.2%, felt they had received enough support, reinforcing the role of the community based networks. The two groups (PC and NPC) did not differ in their perceived support from all sources.

Fig 2 compares the sources of support for the two groups (PC and NPC). It seems that the PC group used more of all forms of support except the structured services of funeral provider, hospital and nursing home which were used more by the NPC. Professional sources, especially counsellors, were more frequently accessed by the PC group.

**Fig 2. Sources of bereavement support accessed by respondents whose care recipients received or did not receive palliative care.**

<https://doi.org/10.1371/journal.pone.0184750.g002>

## Comparison to best practice guidelines

[Table 3](#) outlines this support from the palliative care services as it relates more specifically to the best practice guidelines. The aligned survey questions were adapted to lay people. While three quarters of respondents were asked by staff how they were coping pre-death in general, only 39.4% reported receiving a needs assessment that covered the palliative care domains and more specifically were asked about the emotional and psychological domains. However 79.3% felt the staff kept them informed of the changes in the condition of the dying relative so they can be prepared for the death and 62.9% were offered information about grief and bereavement services. About half were contacted within 3–6 weeks of the death, 28.1% about 6 months after the death, and 25% at any other time.

## Differences in palliative care settings

Comparisons were undertaken between three settings: community palliative care, in-patient (79.5% hospital and 20.5% hospice/palliative care unit), and nursing homes. In terms of the quality of care received, there were significant differences ( $p = 0.001$ ) between the three settings: More respondents rated in-patient settings as excellent/good (93%) followed by community (81%), with the least being nursing homes (73%) ([Table 4](#)). There were no significant differences in the perceived bereavement support between the three settings, although there was more unmet need in nursing homes ([Table 5](#)). Community palliative care did the most bereavement follow-up at 3–6 weeks (63%) followed by in-patient (58%) with nursing homes the least (31%), and differences were significant,  $p < 0.001$  ([Table 6](#)). At the 6-months follow-up contact, the difference between community and inpatient was not pronounced (32–33%), while only 17% reported having a follow-up from nursing homes, and differences were significant,  $p = 0.003$  ([Table 7](#)).

**Table 3. Experience of support from palliative care services in relation to the best practice guidelines.**

Guidelines (Hudson et al, 2010)	Aligned questions from the survey	Percent agreeing N = 298
<i>Guideline 8: Conduct a needs assessment with the family caregiver(s). This should include psychological and physical health, social, spiritual, cultural, financial and practical elements.</i>	Did staff ask how you were coping before the death of your relative/friend?	74.6%
	Did staff ask if you had experienced any significant stress, emotional or psychological problems, before the death of your relative/friend?	39.4%
<i>Guideline 12: When death appears imminent, ensure the family caregiver(s) are aware and assess preparedness for death</i>	Did staff tell you about changes in your relative/friend's condition so that you were more prepared for his/her death?	79.3%
<i>Guideline 13: Confirm with the family caregiver(s) the type of support they may desire in the lead-up to death (e.g. last hours, days) and/or immediately after</i>	Were you offered information about grief and bereavement services that might be helpful following the death of your relative/friend?	62.9%
<i>Guideline 17: Contact the family caregiver(s) and other family members (as appropriate) to assess needs at three to six weeks post-death and adapt bereavement care plan accordingly.</i>	Did staff contact you (by phone, post or home visit) within 3–6 weeks of the death to find out how you were coping, and discuss any questions you might have had?	50.9%
<i>Guideline 19: Conduct a follow-up assessment of the family caregiver(s) and other family members (if appropriate) six months post-death.</i>	Did staff contact you (by phone, post or home visit) about 6 months after the death to find out how you were coping?	28.1%
	Did staff contact you (by phone, post or home visit) at any other time?	25.0%

<https://doi.org/10.1371/journal.pone.0184750.t003>

**Table 4. Quality of palliative care received (n = 263).**

	Excellent/Good		Fair/Poor		Don't know	
	n	%	n	%	n	%
Community Palliative Care	48	81.4	10	16.9	1	1.7
In-patient	124	93.2	8	6.0	1	0.8
Nursing home	49	73.1	15	22.4	3	4.5

F–Fisher's Exact Test (chi-square). P-value = **0.001<sup>F</sup>**

<https://doi.org/10.1371/journal.pone.0184750.t004>

## Feedback on the usefulness of the bereavement follow-ups

Respondents were provided with an opportunity to comment on their responses in terms of how this follow up support was helpful or unhelpful. One hundred and seventy seven responses were provided or 59.4% of those who received palliative care. Participants who drew comfort from the 3–6 week follow up commented that they felt secure knowing that these services were available and appreciative of being remembered by the palliative care providers. The information on bereavement services was deemed relevant by some, and the services were appreciated as per comments such as *“the phone calls I received saved me from falling to pieces”* and *“I could not have coped without it”*. Some were ambivalent about the *“friendly visit- neither helpful nor unhelpful”* or it was *“helpful, although it felt like it was their standard practice”*.

Some respondents felt negative about their experience with the 3–6 weeks follow up, apparently because they did not have any contact and felt forgotten *“Post death, non-existent”, “after my wife’s death I was all on my own. Out of sight out of mind”*; or because the contact did not lead to further help *“it was just a phone call with no help offered”, “was just a query, no help”, “would have liked more what to expect”*. A number reported that when they received a call, it was from someone they had not previously met: *“Not helpful, unspecific call from someone I had never met”, “helpful if having the same nurse. Different people at each time is adding up to the stress and suffering”*. One respondent summarised the general feeling post-bereavement that *“there was a sense that ties with the palliative care provider were abruptly cut”*.

The feedback regarding the 6-month follow up showed that some of the bereaved appreciated the contact, *“the call was helpful to talk through my feelings and thoughts”* or *“this was helpful to talk to someone away from the family”*, or the invitation to a memorial service at 6 months. However many did not receive any follow-up (72%), *“no contact so don’t know if it could have been helpful or not”* and those who did receive a contact, described the support again as generic *“only a short phone call. That was it”*. Some did not need the service because they were contented with the support they were receiving from their funeral provider (mentioned quite often) or from their informal networks or organisations such as the Leukaemia Foundation and the Cancer Council: *“No we didn’t need this service because we received bereavement counselling from [funeral provider]”; “funeral home sent card at 12 months. We were touched that they did”; “The offer of support was generic not necessarily personal but I was*

**Table 5. Bereavement support from palliative care services.**

	Enough support		Not enough support		Did not need support		Other	
	n	%	n	%	n	%	n	%
Community Palliative Care	34	55.7	17	27.9	7	11.5	3	4.9
In-patient	78	58.6	34	25.6	16	12.0	5	3.8
Nursing home	27	40.9	25	37.9	10	15.2	4	6.1

F–Fisher's Exact Test (chi-square). P-value = 0.391<sup>F</sup>

<https://doi.org/10.1371/journal.pone.0184750.t005>

**Table 6. Bereavement follow-up contact at 3–6 weeks.**

	Yes		No		Unsure	
	n	%	n	%	n	%
Community Palliative Care	37	62.7	21	35.6	1	1.7
In-patient	76	57.6	50	37.9	6	4.5
Nursing home	21	31.3	45	67.2	1	1.5

F–Fisher’s Exact Test (chi-square). P-value<0.001<sup>F</sup>

<https://doi.org/10.1371/journal.pone.0184750.t006>

receiving support via family, relatives, friends and neighbours”. In fact one respondent thought one more phone call was too many: “I really didn’t need another call. I would have thought it to be overkill. One phone call was enough”.

## Discussion

This is the first study to report on the bereavement support experience of a community based sample. Most studies specific to bereavement support in palliative care to date have relied on clinical and service based samples [5, 7, 8, 25] which exclude people who did not use palliative care. Moreover, this is the first study to recruit from funeral providers’ databases, an innovative way to engage potential providers of bereavement support and which allows comparison between those who used palliative care with those who did not.

The difference in the profiles of those who did or did not receive palliative care has important implications for service delivery as we often do not know about those who did not use the service. In this study, from the terminal illnesses that lend themselves to palliative care, more people with cancer (64%) had received palliative care in comparison to other illnesses such as heart disease, dementia and organ failure (4–10%). These non-malignant diseases are still under-represented in palliative care ten years on from the study by McNamara, Rosenwax, & Holman [26] where it was reported that less than 10% of people who died of non-malignant diseases had accessed specialist palliative care services, compared with 66% of people who died of cancer. Yet these conditions, as described in this study, have required a longer period of care, twice as long as that of malignant diseases (48 vs 24 months,  $p<0.001$ ), with more “other” relatives helping the immediate family (mainly spouses and adult children) in this prolonged period of care. Having a network of such caregivers may be a consequence of a longer disease trajectory in non-malignant diseases that allows networks of close friends and extended family to form. Burns et al [27] have pointed to this network of extended family and friends at the end of life, a network invisible to the health system but whose members also need adequate support in their role.

Those who received palliative care were 2–3 times more likely to have an advance care plan or advance health directive, albeit still in quite low proportions overall (12–13%), though a rate close to the 14% reported by an Australian study [28]. Internationally, the rate reported in the

**Table 7. Bereavement follow-up contact at 6 months.**

	Yes		No		Unsure	
	n	%	n	%	n	%
Community Palliative Care	20	33.3	40	66.7	0	0.0
In-patient	42	31.8	81	61.4	9	6.8
Nursing home	11	16.7	55	83.3	0	0.0

F–Fisher’s Exact Test (chi-square). P-value = 0.003<sup>F</sup>

<https://doi.org/10.1371/journal.pone.0184750.t007>



literature varies from 10% in American hospitals [29], to 16% in the Dutch general older population [30] and up to 25% among Swiss palliative care patients [31]. These rates are low despite studies showing positive impacts on patient quality of dying and reduction in stress and anxiety of families [32]. Many factors related to the health care professional, the patient/family and the health system are responsible for such poor end of life communication [32]. While bereavement is not addressed directly in advance care plans, it seems reasonable to assume that the existence of an advance care plan indicates some communication within the person's social network about end of life wishes. Clarity about a dying person's awareness of the situation, and an indication of preparedness, seem to be important for their family and friends in experiencing bereavement. The relationship between advance care planning and bereavement outcomes is important to explore in future studies.

The significant differences between the settings obtained in this study shed some light on the apparent discrepancy between pre- and post-death contact. Bereavement follow up tended to be common in community programs, less common in hospital programs. The site of palliative care affects follow-up, as one factor is privacy legislation. Because community palliative care providers tend to define the 'patient and family' as the unit of care, the service tends to keep better records of dependants and also has the 'right' to contact them after the death. Hospitals frequently lack adequate contact details for dependants, and are more reticent in following up because of concerns (often misplaced) regarding privacy legislation. However, in terms of the quality of care received pre-death, our results are similar to Pidgeon et al [33] who reported that family carers were likely to be less satisfied if receiving care from a community palliative care service.

Perceived bereavement support from all sources for PC and NCP groups are effectively the same, although there are differences in the patterns of support for the two groups (Fig 2). Possible explanations for the PC group's greater access to resources, including professional services, are, first, that many palliative care patients have followed a complex care pathway which mobilises a multi-disciplinary team both prior to and whilst in palliative care: that is, family and friends are introduced to a wider range of sources of support by the nature of the illness and the service structure. Secondly, palliative care is usually intentional about encouraging caregivers to look for support, whereas hospitals and nursing homes are less likely to do this. That is, palliative care recognizes the needs of caregivers and normalizes asking for support in a way that other services may not. This is supported by Bergman et al [34] who reported that bereaved caregivers who used hospice care were more likely to have access to information and services not routinely available to non-hospice users.

This is also the first study to provide an insight into the application or practice of the guidelines by palliative care services from the service users' perspective. While the majority of the bereaved (84%) felt that the care received by their relative/friend from the palliative care service was excellent to good, the support for themselves before and after their relative's death was not considered as optimal. Clearly the systematic assessment of family caregivers' support needs in the lead up to the patient's death is not given enough attention by palliative care services. Only 39.4% of the bereaved reported being specifically asked about their emotional/psychological distress pre-bereavement; rather it was a more general question about coping (75%). Furthermore, following their relative's death, just half of the bereaved perceived they had enough support from palliative care services. Half of these people had a follow up contact from the service at 3–6 weeks, and a quarter had a follow-up at 6 months. This lack of attention to members of the dying person's social network in the time prior to the death suggests that little attention is being paid to links between the pre- and post-death experience of family and friends. This link, sometimes conceptualised as preparatory or anticipatory grief, deserves further attention [35].

It is evident that supporting family caregivers, while caregiving, has benefits pre- and post-bereavement: Results of the Carer Support Needs Assessment (CSNAT) trials in Australia have showed a significant reduction in caregiver strain during the caregiving period in community palliative care [36]. In another Australian study, family caregivers of older people discharged home from hospital were significantly more prepared to provide care and reported reduced caregiver strain and distress compared to family caregivers in the control group [37]. A larger trial of CSNAT in the UK [38] found a small reduction in grief, improvements in mental and physical health post-bereavement and in the probability of death at home.

Our findings, supported by the evidence from the literature, reinforce the need for palliative care services to take action during the pre-bereavement period to effectively assess and support family caregivers. Yet the ‘window of opportunity’ for contact with caregivers to assess their grief and bereavement needs while heading to the care recipient’s impending death does not seem to be well utilised in the palliative care system, although this is the only time caregivers are likely to have face to face contact with staff [11]. Clearly, a short length of stay with a palliative care service (median of one month reported in this study) is not conducive to building rapport with the family to prepare them for the death. In the post-death period, contact with bereaved caregivers is even more difficult for services due to various barriers such as staffing, funding, and availability of service contact with them [11]. This was quite evident in this study where half of the bereaved were followed up to 6 weeks and this proportion dwindled to a quarter at 6 months. Similarly Ghesquiere et al [39] have found that hospice bereavement support only reached half of the bereaved in their study, suggesting a need to improve care access and delivery.

The qualitative feedback from bereaved people has reinforced first-hand the limited helpfulness of the blanket approach to bereavement support, where a phone call or invitation to a memorial service did not address all needs, as one size does not fit all [9]. Some of the respondents have described their service as “not personal” or “generic”, or “just standard practice”, and some clearly did not see benefits in the contact because it was not tailored to their own needs or it did not occur at the time they most needed it. More importantly, some considered it stressful to receive calls from staff members they did not know, and these were different at every contact. So it seems for some who did receive a follow up, it was generally not helpful or neutral. We don’t know if this was because assistance at this time and from this source were not helpful or that it would be helpful if the quality was better. The responses also seem to point to differing expectations of different providers. A palliative care service follow-up call can be seen as standard practice, while the equally-standard practice of an anniversary card from a funeral provider can be embraced as an expression of care. It may also be that the person-centred services of community palliative care programs elicit higher expectations of bereavement follow-up, but these are found wanting. Whatever the factors contributing to these mixed responses, palliative care services need to consider the most appropriate time to make contact post-death as timeliness is important and also who should make this contact, as consistency appears to be crucial to build rapport and trust in the ability of the service to help. The content of the call/contact should be focused on the specific needs of the bereaved. The assessment of these needs would ideally be initiated by the palliative care service in the pre-bereavement phase of care. Further research is needed to provide an evidence-based for the pre-bereavement assessment of post-bereavement risk, according to the three risk groups outlined in the public health model for bereavement support [18].

In the absence of any formal and systematic assessment of family caregivers’ needs and bereavement risks [11], palliative care services will struggle to make appropriate decisions about providing, or not providing, bereavement support, and if they do for how long [19]. A particular matter that needs to be reviewed is the common practice of assigning bereavement

care to a separate team or individual within a palliative care service. There are obvious organisational reasons for doing this, but our findings here underline the importance of having some continuity of relationship, not just continuity of service. That is, the bereavement team may need to consider including someone who was involved in the care of the person who died, or a member of the bereavement team needs to be involved in the care of the family before the death. The advantage a palliative care program has over any stand-alone bereavement program is its role in the pre-bereavement experience of dying people and their social networks. This involvement prior to the death provides opportunities both to assist family and friends in their preparations for death as well as to identify those who may be at risk of greater distress post-bereavement. To enter a bereavement care phase with a new team using generic strategies that invite bereaved people to self-refer is to fail to realise the possibilities inherent in pre-bereavement care.

There was qualitative and quantitative evidence of reliance on support from informal networks such as families, neighbours and friends, also from other services such as the funeral providers and other community based organisations (the Leukaemia Foundation and the Cancer Council), and this was reflected in 68.2% feeling they had received enough support from all community and health providers sources. In fact, we have previously reported that the majority of the bereaved in each of the 3 risk groups accessed support predominantly from family (95%) and friends (88%), followed by funeral directors (79%) and general practitioners (56%) [18], emphasized also in Fig 2. Therefore, palliative care services might do better investing their efforts principally in (a) assessing and supporting family caregivers during the pre-bereavement period and (b) developing community capacity and referral pathways for bereavement care [18, 25, 40, 41].

## Limitations

Although this is not a random sample of the general population, this sample compares well with the UK mortality follow-back survey on cancer [42] in terms of its composition: Responding bereaved females (61% UK, 70% this study), bereaved spouses (38% UK, 41% this study), bereaved sons/daughters (46% UK, 43% this study), the age of the deceased is 77 years for both studies, and proportion male in the deceased (52% UK, 51% this study). Also this sample is drawn from six funeral providers that are based in a mix of metropolitan, regional and rural areas across four Australian states. Although the low response rate in this study is in line with others who relied on postal surveys with no reminder follow up [43], those who did not respond may have had different experiences to those reported in this study. As this is a retrospective study, restricting the time since death to 6–24 months would not have been subject to a large recall bias of events [44]. While some of the wording in the survey questions is not in exact alignment to the wording in the clinical guidelines, it was necessary to adapt them to be lay friendly and we recognise that the responses may have been influenced by the interpretation of the adapted wording. Also the selection of respondents from funeral providers' database may have influenced the significant number reporting support from these providers.

## Conclusions

Although this national study is based in Australia, it has international implications as our earlier review of palliative care policies and bereavement support practices in several countries (the United States, Canada, the United Kingdom, and Japan) demonstrated similar challenges: questions over providing universal versus targeted support; a lack of clear evidence driving service delivery; informal or no risk assessment; and limited or no evaluation of services [9]. More recent evidence from Europe reinforces our finding that practice of bereavement

support in palliative care has only a tenuous relationship with guidelines and assessment tools [10]. While palliative care services offer bereavement support, this is seldom as intentional or targeted as it should be, particularly when the bereavement care phase is separated from the care and support provided prior to the death. Palliative care services should be able to provide more intentional and targeted bereavement care for the population they support. From a public health perspective, however, we have argued elsewhere that, rather than build bereavement services around those who have been in contact with a palliative care program, bereavement services should be developed to serve the whole population of bereaved people. This approach would give priority to the community support needed by most bereaved people, and ensure that professional care supplements, not replaces, the care provided by people's existing social networks [19]. We believe the findings reported here provide further support for this wider claim. Further research is needed into the attitudes and experiences that underlie the patterns of bereavement support identified in this study.

## Acknowledgments

The authors acknowledge the financial support of the WA Cancer and Palliative Care Network, Health Department of Western Australia, the cooperation of funeral providers who assisted with the recruitment of participants, and the bereaved families who agreed to complete the survey and contributed to our understanding in this field.

## Author Contributions

**Conceptualization:** Samar M. Aoun, Bruce Rumbold, Lauren J. Breen.

**Data curation:** Samar M. Aoun, Denise Howting.

**Formal analysis:** Samar M. Aoun, Denise Howting, Lauren J. Breen.

**Funding acquisition:** Samar M. Aoun, Amanda Bolleter.

**Investigation:** Samar M. Aoun, Bruce Rumbold.

**Methodology:** Samar M. Aoun, Lauren J. Breen.

**Project administration:** Samar M. Aoun.

**Resources:** Samar M. Aoun.

**Software:** Samar M. Aoun.

**Supervision:** Samar M. Aoun.

**Validation:** Samar M. Aoun, Bruce Rumbold, Denise Howting, Lauren J. Breen.

**Visualization:** Samar M. Aoun, Bruce Rumbold, Amanda Bolleter, Lauren J. Breen.

**Writing – original draft:** Samar M. Aoun, Bruce Rumbold, Denise Howting, Amanda Bolleter, Lauren J. Breen.

**Writing – review & editing:** Samar M. Aoun, Bruce Rumbold, Denise Howting, Amanda Bolleter, Lauren J. Breen.

## References

1. Hall C, Hudson P, Boughey A. Bereavement support standards for specialist palliative care services. In: Health Do, editor. Melbourne: State Government of Victoria; 2012.
2. National Hospice and Palliative Care Organization. Guidelines for bereavement care in hospice. 2 ed. Alexandria, VA: National Hospice and Palliative Care Organization; 2008.



3. Palliative Care Australia. Standards for providing quality palliative care for all Australians. Deakin West, ACT: Palliative Care Australia; 2005. Available from: <http://www.palliativecare.org.au/Portals/46/Standards%20for%20providing%20quality%20palliative%20care%20for%20all%20Australians.pdf>.
4. World Health Organization. WHO guide for effective programmes: Palliative care. Geneva: World Health Organization; 2007.
5. Abbott J, O'Connor M, Payne S. An Australian survey of palliative care and hospice bereavement services. *Australian Journal of Cancer Nursing*. 2008; 9(2):12–7.
6. Agnew A, Manktelow R, Taylor B, Jones L. Bereavement needs assessment in specialist palliative care: a review of the literature. *Palliative Medicine*. 2010; 24(1):46–59. <https://doi.org/10.1177/0269216309107013> PMID: 19762368
7. Foliat DE, Clausen M, Siljestrom C. Bereavement practices among California hospices: results of a state-wide survey. *Death Stud*. 2001; 25(5):461–7. <https://doi.org/10.1080/07481180125792> PMID: 11806414
8. Mather MA, Good PD, Cavenagh JD, Ravenscroft PJ. Survey of bereavement support provided by Australian palliative care services. *Medical Journal of Australia*. 2008; 188(4):228–30. PMID: 18279130
9. Breen LJ, Aoun SM, O'Connor M, Rumbold B. Bridging the gaps in palliative care bereavement support: an international perspective. *Death Stud*. 2014; 38(1–5):54–61. <https://doi.org/10.1080/07481187.2012.725451> PMID: 24521046
10. Guldin M, Murphy I, Keegan O, Monroe B, Lacasta-Reverte M, Benkel I. Bereavement services in palliative care in Europe: A survey study by the European Association for Palliative Care bereavement task-force. *European journal of Palliative Care*. 2015; 22(4):185–9.
11. Sealey M, O'Connor M, Aoun SM, Breen LJ. Exploring barriers to assessment of bereavement risk in palliative care: perspectives of key stakeholders. *BMC palliative care*. 2015; 14(49).
12. Aoun SM, Breen LJ, O'Connor M, Rumbold B, Nordstrom C. A public health approach to bereavement support services in palliative care. *Australian and New Zealand Journal of Public Health*. 2012; 36(1):14–6. <https://doi.org/10.1111/j.1753-6405.2012.00825.x> PMID: 22313700
13. Walter T. On bereavement: the culture of grief. Maidenhead, England: Open University Press; 1999.
14. Schut H, Stroebe M, van den Bout J, Terheggen M. The efficacy of bereavement interventions: determining who benefits. In: Stroebe M, Hansson R, Stroebe W, Schut H, editors. *Handbook of Bereavement Research: Consequences, Coping, and Care*. Washington (DC): American Psychological Association; 2001. p. 705–37.
15. Australian Bureau of Statistics. 3303.0 Causes of Death Australia. Canberra: ABS; 2015.
16. Australian Institute for Health and Welfare. Palliative care outcomes. 2016.
17. Rosenwax LK, McNamara BA. Who receives specialist palliative care in Western Australia—and who misses out. *Palliat Med*. 2006; 20(4):439–45. <https://doi.org/10.1191/0269216306pm1146oa> PMID: 16875115
18. Aoun SM, Breen LJ, Howting DA, Rumbold B, McNamara B, Hegney D. Who needs bereavement support? A population based survey of bereavement risk and support need. *PloS one*. 2015; 10(3): e0121101. <https://doi.org/10.1371/journal.pone.0121101> PMID: 25811912
19. Rumbold B, Aoun S. An assets-based approach to bereavement care. *Bereavement Care*. 2015; 34(3):99–102.
20. Aoun SM, Breen LJ, Rumbold B, Howting D. Reported experiences of bereavement support in Western Australia: a pilot study. *Aust N Z J Public Health*. 2014; 38(5):473–9. <https://doi.org/10.1111/1753-6405.12177> PMID: 24750475
21. Hudson P, Remedios C, Zordan R, Thomas K, Clifton D, Crewdson M, et al. Clinical practice guidelines for the psychosocial and bereavement support of family caregivers of palliative care patients. Melbourne: Centre for Palliative Care, St Vincent's Hospital Melbourne; 2010.
22. Office for National Statistics (UK). National Bereavement Survey (VOICES: Views of informal carer—evaluation of services). Newport, England: ONS; 2012.
23. Aoun S, McConigley R, Abernethy A, Currow DC. Caregivers of people with neurodegenerative diseases: profile and unmet needs from a population-based survey in South Australia. *Journal of palliative medicine*. 2010; 13(6):653–61. <https://doi.org/10.1089/jpm.2009.0318> PMID: 20557235
24. Hsieh HF, Shannon SE. Three approaches to qualitative content analysis. *Qualitative health research*. 2005; 15(9):1277–88. <https://doi.org/10.1177/1049732305276687> PMID: 16204405
25. Nielsen MK, Neergaard MA, Jensen AB, Vedsted P, Bro F, Guldin MB. Predictors of Complicated Grief and Depression in Bereaved Caregivers: A Nationwide Prospective Cohort Study. *J Pain Symptom Manage*. 2017; 53(3):540–50. <https://doi.org/10.1016/j.jpainsymman.2016.09.013> PMID: 28042073
26. McNamara B, Rosenwax LK, Holman CD. A method for defining and estimating the palliative care population. *J Pain Symptom Manage*. 2006; 32(1):5–12. <https://doi.org/10.1016/j.jpainsymman.2005.12.018> PMID: 16824980

27. Burns CM, Abernethy AP, Dal Grande E, Currow DC. Uncovering an invisible network of direct caregivers at the end of life: a population study. *Palliat Med.* 2013; 27(7):608–15. <https://doi.org/10.1177/0269216313483664> PMID: 23587738
28. White B, Tilse C, Wilson J, Rosenman L, Strub T, Feeney R, et al. Prevalence and predictors of advance directives in Australia. *Internal medicine journal.* 2014; 44(10):975–80. <https://doi.org/10.1111/imj.12549> PMID: 25109226
29. Auerbach AD, Katz R, Pantilat SZ, Bernacki R, Schnipper J, Kaboli P, et al. Factors associated with discussion of care plans and code status at the time of hospital admission: results from the Multicenter Hospitalist Study. *Journal of hospital medicine.* 2008; 3(6):437–45. <https://doi.org/10.1002/jhm.369> PMID: 19084893
30. Onwuteaka-Philipsen B, Kaspers P, Deeg D, Pasman H. Having and discussing preferences on forgoing treatment in the last three months of life of older people with and without an advance directive. *Palliat Med.* 2012; 26(4):656–7.
31. Pautex S, Herrmann FR, Zulian GB. Role of advance directives in palliative care units: a prospective study. *Palliat Med.* 2008; 22(7):835–41. <https://doi.org/10.1177/0269216308094336> PMID: 18718993
32. Lewis E, Cardona-Morrell M, Ong KY, Trankle SA, Hillman K. Evidence still insufficient that advance care documentation leads to engagement of healthcare professionals in end-of-life discussions: A systematic review. *Palliat Med.* 2016; 30(9):807–24. <https://doi.org/10.1177/0269216316637239> PMID: 26951066
33. Pidgeon TM, Johnson CE, Lester L, Currow D, Yates P, Allingham SF, et al. Perceptions of the care received from Australian palliative care services: A caregiver perspective. *Palliative & supportive care.* 2017:1–11.
34. Bergman EJ, Haley WE. Depressive symptoms, social network, and bereavement service utilization and preferences among spouses of former hospice patients. *Journal of palliative medicine.* 2009; 12(2):170–6. <https://doi.org/10.1089/jpm.2008.0237> PMID: 19207061
35. Stroebe M, Boerner K. Caregiving and bereavement research: Bridges over the gap. *Palliat Med.* 2015; 29(7):574–6. <https://doi.org/10.1177/0269216315585952> PMID: 26048707
36. Aoun S, Grande G, Howting D, Deas K, Toye C, Troeung L, et al. The impact of the Carer Support Needs Assessment Tool (CSNAT) in community palliative care using a stepped wedge cluster trial. *PLoS one.* 2015; 10(4).
37. Toye C, Parsons R, Slatyer S, Aoun SM, Moorin R, Osseiran-Moisson R, et al. Outcomes for family carers of a nurse-delivered hospital discharge intervention for older people (the Further Enabling Care at Home Program): Single blind randomised controlled trial. *Int J Nurs Stud.* 2016; 64:32–41. <https://doi.org/10.1016/j.ijnurstu.2016.09.012> PMID: 27684320
38. Grande GE, Austin L, Ewing G, O'Leary N, Roberts C. Assessing the impact of a Carer Support Needs Assessment Tool (CSNAT) intervention in palliative home care: a stepped wedge cluster trial. *BMJ supportive & palliative care.* 2015.
39. Ghesquiere A, Thomas J, Bruce ML. Utilization of hospice bereavement support by at-risk family members. *The American journal of hospice & palliative care.* 2016; 33(2):124–9.
40. Rumbold B, Aoun S. Bereavement and palliative care: A public health perspective. *Progress in Palliative Care.* 2014; 22(3):131–5.
41. Breen LJ, Aoun SM, Rumbold B, McNamara B, Howting DA, Mancini V. Building community capacity in bereavement support. *The American journal of hospice & palliative care.* 2017; 34(3):275–81.
42. Calanzani N, Higginson IJ, Koffman J, Gomes B. Factors Associated with Participation, Active Refusals and Reasons for Not Taking Part in a Mortality Followback Survey Evaluating End-of-Life Care. *PLoS one.* 2016; 11(1):e0146134. <https://doi.org/10.1371/journal.pone.0146134> PMID: 26745379
43. Hunt K, Shlomo N, Addington-Hall J. Participant recruitment in sensitive surveys: a comparative trial of 'opt in' versus 'opt out' approaches. *BMC Medical Research Methodology.* 2013; 13(1):3.
44. Williams BR, Woodby LL, Bailey FA, Burgio KL. Identifying and responding to ethical and methodological issues in after-death interviews with next-of-kin. *Death Stud.* 2008; 32(3):197–236. <https://doi.org/10.1080/07481180701881297> PMID: 18705168

# Effect of dignity therapy on distress and end-of-life experience in terminally ill patients: a randomised controlled trial



Harvey Max Chochinov, Linda J Kristjanson, William Breitbart, Susan McClement, Thomas F Hack, Tom Hassard, Mike Harlos

## Summary

**Background** Dignity therapy is a unique, individualised, short-term psychotherapy that was developed for patients (and their families) living with life-threatening or life-limiting illness. We investigated whether dignity therapy could mitigate distress or bolster the experience in patients nearing the end of their lives.

**Methods** Patients (aged  $\geq 18$  years) with a terminal prognosis (life expectancy  $\leq 6$  months) who were receiving palliative care in a hospital or community setting (hospice or home) in Canada, USA, and Australia were randomly assigned to dignity therapy, client-centred care, or standard palliative care in a 1:1:1 ratio. Randomisation was by use of a computer-generated table of random numbers in blocks of 30. Allocation concealment was by use of opaque sealed envelopes. The primary outcomes—reductions in various dimensions of distress before and after completion of the study—were measured with the Functional Assessment of Chronic Illness Therapy Spiritual Well-Being Scale, Patient Dignity Inventory, Hospital Anxiety and Depression Scale, items from the Structured Interview for Symptoms and Concerns, Quality of Life Scale, and modified Edmonton Symptom Assessment Scale. Secondary outcomes of self-reported end-of-life experiences were assessed in a survey that was undertaken after the completion of the study. Outcomes were assessed by research staff with whom the participant had no previous contact to avoid any possible response bias or contamination. Analyses were done on all patients with available data at baseline and at the end of the study intervention. This study is registered with ClinicalTrials.gov, number NCT00133965.

**Findings** 165 of 441 patients were assigned to dignity therapy, 140 standard palliative care, and 136 client-centred care. 108, 111, and 107 patients, respectively, were analysed. No significant differences were noted in the distress levels before and after completion of the study in the three groups. For the secondary outcomes, patients reported that dignity therapy was significantly more likely than the other two interventions to have been helpful ( $\chi^2=35.50$ ;  $df=2$ ;  $p<0.0001$ ), improve quality of life ( $\chi^2=14.52$ ;  $p=0.001$ ), increase sense of dignity ( $\chi^2=12.66$ ;  $p=0.002$ ), change how their family saw and appreciated them ( $\chi^2=33.81$ ;  $p<0.0001$ ), and be helpful to their family ( $\chi^2=33.86$ ;  $p<0.0001$ ). Dignity therapy was significantly better than client-centred care in improving spiritual wellbeing ( $\chi^2=10.35$ ;  $p=0.006$ ), and was significantly better than standard palliative care in terms of lessening sadness or depression ( $\chi^2=9.38$ ;  $p=0.009$ ); significantly more patients who had received dignity therapy reported that the study group had been satisfactory, compared with those who received standard palliative care ( $\chi^2=29.58$ ;  $p<0.0001$ ).

**Interpretation** Although the ability of dignity therapy to mitigate outright distress, such as depression, desire for death or suicidality, has yet to be proven, its benefits in terms of self-reported end-of-life experiences support its clinical application for patients nearing death.

**Funding** National Cancer Institute, National Institutes of Health.

## Introduction

Research into methods to understand and support patients who are nearing the end of their lives is increasing.<sup>1–3</sup> Dignity therapy, a unique, individualised, brief psychotherapy, was developed for the purpose of relieving distress and enhancing the end-of-life experiences of terminally ill patients. It provides these patients with an opportunity to reflect on things that matter most to them or that they would most want remembered. The therapeutic process begins with a framework of questions (panel 1) that are based on an empirical model of dignity in the terminally ill patient.<sup>4,5</sup> These conversations, guided by a trained therapist, are flexible to accommodate the patients' needs and choices about what they specifically wish to address. Dignity therapy is audiorecorded and transcribed, with an edited

version of the transcript given to patients to share or bequeath to individuals of their choice.

In a phase 1 trial with 100 terminally ill patients, 91% were satisfied with dignity therapy, and 76% reported a heightened sense of dignity, 68% an increased sense of purpose, 67% a heightened sense of meaning, 47% an increased will to live, and 81% that it had been or would be of help to their family.<sup>6</sup> Post-intervention assessments of suffering and depressive symptoms showed small, but significant improvements.<sup>6</sup> 78% of patients' family members reported that the therapy enhanced the patient's dignity, and 72% that it heightened the meaning of life for the patient; 78% said the document from the therapy session was a comfort to them in their time of grief, and 95% that they would recommend dignity therapy to other patients and their families.<sup>7</sup> We therefore investigated

*Lancet Oncol* 2011; 12: 753–62

Published Online

July 7, 2011

DOI:10.1016/S1470-

2045(11)70153-X

See [Comment](#) page 712

**Department of Psychiatry** (Prof H M Chochinov MD), **Faculty of Nursing** (Prof S McClement PhD, Prof T F Hack PhD), and **Community Health Sciences** (Prof H M Chochinov, Prof T Hassard PhD), **University of Manitoba, Winnipeg, MB, Canada**; **Manitoba Palliative Care Research Unit, CancerCare Manitoba, Winnipeg, MB, Canada** (Prof H M Chochinov, Prof S McClement); **Department of Patient Support and Family Services, CancerCare Manitoba, Winnipeg, MB, Canada** (Prof T F Hack); **Swinburne University of Technology, Melbourne, VIC, Australia** (Prof L J Kristjanson PhD); **Curtin University, Perth, WA, Australia** (Prof L J Kristjanson); **Edith Cowan University, Perth, WA, Australia** (Prof L J Kristjanson); **Department of Psychiatry and Behavioral Sciences, Memorial Sloan-Kettering Cancer Center, New York, NY, USA** (Prof W Breitbart MD); and **Winnipeg Regional Health Authority, St Boniface General Hospital, Winnipeg, MB, Canada** (Prof M Harlos MD)

Correspondence to: Prof Harvey Max Chochinov, Department of Psychiatry, University of Manitoba, CancerCare Manitoba, Palliative Care Research Unit, Room 3017, 675 McDermot Avenue, Winnipeg, Manitoba R3E 0V9, Canada. [harvey.chochinov@cancercare.mb.ca](mailto:harvey.chochinov@cancercare.mb.ca)

**Panel 1: Protocol for questions about dignity therapy**

- Tell me a little about your life history, particularly the parts that you either remember most or think are the most important? When did you feel most alive?
- Are there specific things that you would want your family to know about you, and are there particular things you would want them to remember?
- What are the most important roles you have had in life (eg, family roles, vocational roles, community-service roles)? Why were they so important to you and what do you think you accomplished in those roles?
- What are your most important accomplishments, and what do you feel most proud of?
- Are there particular things that you feel still need to be said to your loved ones or things that you would want to take the time to say once again?
- What are your hopes and dreams for your loved ones?
- What have you learned about life that you would want to pass along to others? What advice or words of guidance would you wish to pass along to your son, daughter, husband, wife, parents, or other(s)?
- Are there words or perhaps even instructions that you would like to offer your family to help prepare them for the future?
- In creating this permanent record, are there other things that you would like included?

whether this novel psychotherapeutic approach would be better than standard palliative care and client-centred care (which focused on non-generativity—ie, here and now issues) in terms of reducing psychological, existential, and spiritual distress in patients who are terminally ill.

**Methods****Participants**

Individuals were eligible for participation in the study if they had a terminal prognosis with a life expectancy of 6 months or less, according to their treating physician; were receiving palliative care in a hospital or community setting (hospice or home) through an affiliated recruitment site in Canada, USA, and Australia; were aged 18 years or older; were willing to commit to three or four contacts over about 7–10 days; and were able and willing to provide written informed consent. Patients were excluded if they were delirious or otherwise cognitively impaired (based on clinical consensus and post-randomisation Blessed Orientation Memory Concentration test),<sup>8</sup> too ill to complete the requirements of the protocol, or unable to speak and read English.

This trial was coordinated by the Manitoba Palliative Care Research Unit at CancerCare Manitoba, Winnipeg, MB, Canada. Other participating centres were Curtin University, Perth, WB, Australia, and Memorial Sloan Kettering Cancer Center, New York, NY, USA. Approval was obtained from the ethics committees of all participating centres. All participants provided written informed consent.

**Randomisation and masking**

After providing consent, all participants were randomly assigned in a 1:1:1 ratio to one of three study groups—dignity therapy, standard palliative care, or client-centred care—and asked to complete baseline psychosocial questionnaires. The study statistician used a

computer-generated table of random numbers in blocks of 30 to allocate patients. Allocation concealment was by use of opaque sealed envelopes. The envelopes were sent to each site and opened sequentially by study staff after consent had been obtained from the patient. To avoid any possible response bias or contamination, research staff with whom the participant had no previous contact administered the final psychometric battery of questionnaires. Recordings from the dignity therapy and client-centred care groups were reviewed by the study coordinator to ensure protocol adherence and prevent cross-contamination.

**Study groups**

Dignity therapy was provided by a psychologist, psychiatrist, or experienced palliative-care nurse. All therapists took part in a 3-day training workshop, led by the principal investigator (HMC). Continuous supervision of the therapy was provided by the site investigator (HMC, LJK, WB), initially for every case and then varied, until each therapist was fully competent. Group supervision, led by the principal investigator, took place every 6–8 weeks throughout the study, with videoconferencing to connect all three study sites. The principal investigator also reviewed a random sample (about one in six) of transcripts from all study sites to identify any difficulties or therapeutic improvements. Feedback from this review was offered either individually or, when more broadly applicable, within group supervision.

Patients were shown the framework of questions for dignity therapy (panel 1) and asked to consider what they might wish to speak about during their session(s); this initial introduction to, and explanation of, dignity therapy took about 30 min. Within a few days, or as soon as a second meeting could be arranged, the therapist used the question framework to help elicit patients' recollections, hopes, wishes for loved ones, lessons learned, and things they wanted remembered by those they were about to leave behind. Dignity therapy is flexible enough to accommodate participant's preferences and choices about content, but the ethos of questioning targets those things that might enhance a sense of meaning, purpose, continued sense of self, and overall sense of dignity. All therapy sessions were audiotaped; these sessions usually took about 60 min. On completion, the audiorecording was transcribed verbatim and the transcript edited, to provide a clear and readable narrative. This transcript or generativity document was returned to the patient within about 4 working days from their previous session, read to them in its entirety to ensure that no errors of omission or commission needed to be addressed (this final session usually took about 30 min). The final version of the **generativity** document was given to the patient, to be passed along to a recipient of the patient's choice, from whom follow-up data were later obtained (the effect of dignity therapy on family members given the generativity documents will be reported separately). At the conclusion

**Generativity**

or the ability to guide the next generation, encompasses how patients might find strength or comfort in knowing that they will leave behind something lasting and transcendent after death



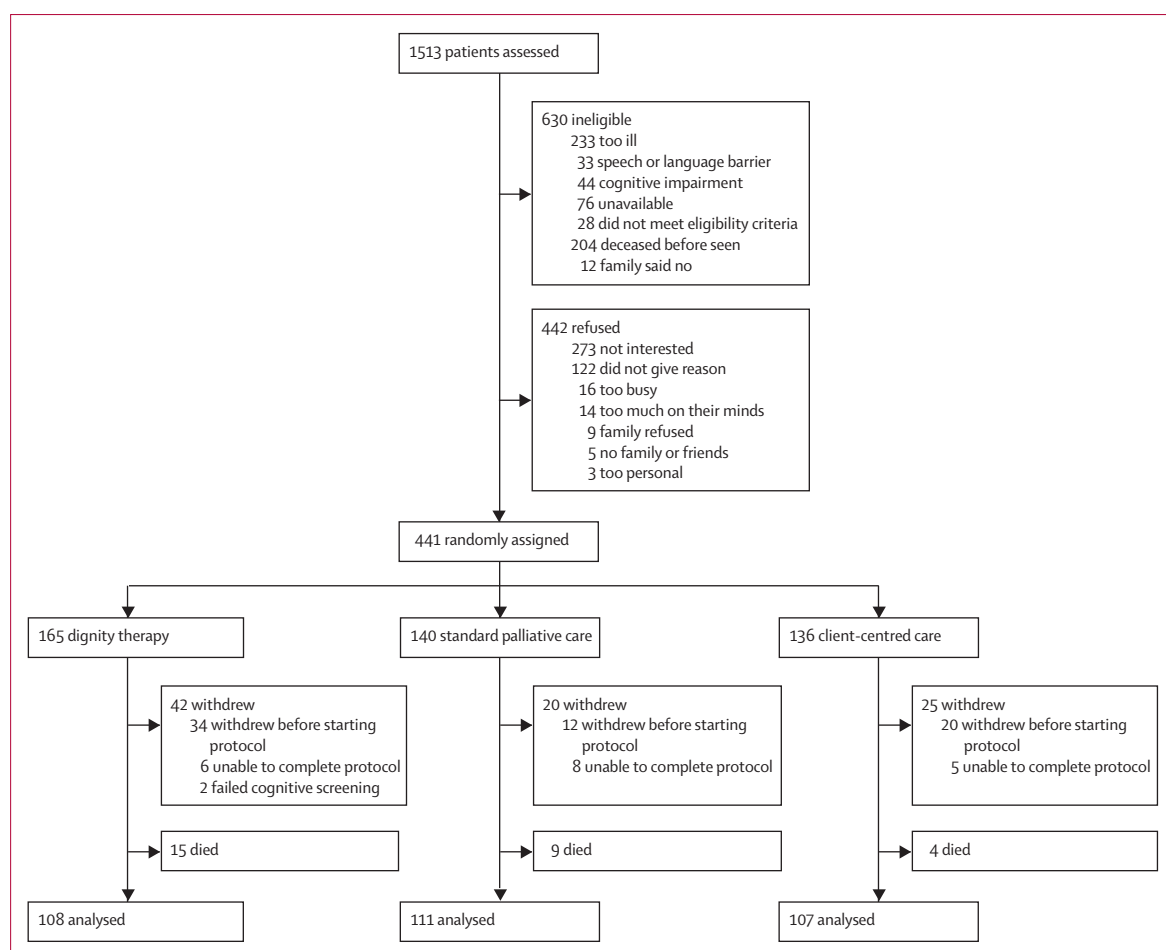


Figure: Trial profile

of this session, participants were asked to complete a psychometric follow-up battery described in the outcome measurements section below.

Client-centred care is a supportive psychotherapeutic approach, in which the research nurse therapist guides the patient through discussions that focus on here-and-now issues—ie, participants are asked about their illness, associated symptoms, and what is being done to address their distress. The content of client-centred care was kept as distinct from dignity therapy as much as possible and did not focus on issues of generativity, meaning, or purpose. If these issues were raised, the therapist did not probe for additional detail, allowing the interview to gently return to illness-related inquiry. Although these sessions were audiorecorded for purposes of auditing protocol adherence, no permanent record of these conversations was provided to the patient or the patient's family. The number of contacts for client-centred care and dignity therapy were identical, with the initial, middle, and final meetings designed to take about the same time in both groups. During the final meeting, 7–10 days later, the participants completed the psychometric battery.

Participants assigned to standard palliative care had access to the complete range of palliative-care-support services that were available to all study patients, including specialist palliative-care physicians and nurses (ie, experts in the management of pain and symptoms), social workers, chaplains, and psychologists or psychiatrists. No participating site provided a formal approach to addressing generativity issues; therefore, a programme similar to dignity therapy was not available to patients who were not randomly assigned to this group. Patients were asked to complete the battery of psychometric questionnaires as soon as they were assigned to standard palliative care and then 7–10 days later—a timeframe roughly corresponding to the time between the first and last contact in the other two study groups.

### Outcome measurements

The primary outcomes—reductions in various dimensions of distress—consisted of mean changes between baseline and end of intervention in measurements of psychosocial, spiritual, and existential distress. The primary measurements were the Palliative Performance Scale (for physical

performance; 100%=healthy, 0%=death),<sup>9</sup> Functional Assessment of Chronic Illness Therapy Spiritual Well-Being Scale (spiritual wellbeing with two subscales—meaning or peace and faith),<sup>10</sup> Patient Dignity Inventory,<sup>11</sup> and Hospital Anxiety and Depression Scale.<sup>12</sup> We also

See Online for webappendix

administered items from the Structured Interview for Symptoms and Concerns,<sup>13</sup> including dignity, desire for death, suffering, hopelessness, depression, suicidal ideation, and sense of burden to others. Additional measurements were a two-item Quality of Life Scale<sup>14</sup> and an Edmonton Symptom Assessment Scale, modified to include a will-to-live visual analogue scale.<sup>15</sup> All scales were administered at baseline and immediately after study completion. Secondary outcomes, addressing to what extent the intervention might have affected the participants' end-of-life experiences, were measured with a survey done after study completion, with detailed questions about their experience of participation in the study and how it had affected them and their family (for the questionnaire see webappendix pp 1–18).

### Statistical analysis

Analyses were done on all patients with available data at baseline and at the end of the study intervention. The data were summarised with standard descriptive measurements. Two group comparisons were undertaken—*t* tests when the data approximated a normal distribution and Mann-Whitney *U* tests when the data were ordinal. Three group comparisons were undertaken by use of ANOVA when a normal distribution was reasonably approximated and Kruskal-Wallis tests when it was not. With an  $\alpha$  set at 0.05, desired power set at 80%, and small-to-medium effect size with Cohen's value for such an effect size of 0.15, the estimated recruitment target was 120 patients per group.<sup>16</sup> The effect size here refers to the standard deviation of the group means divided by the postulated common standard deviation. With three groups, an effect size of 0.15 would imply that at least one group mean is at least a third of a standard deviation away from the other two. SPSS (version 18.0) was used for the statistical analysis.

When a significant result was obtained in a multigroup comparison, the precise nature of the group differences was investigated with Tukey's test (if ANOVA had been used) or a series of Mann-Whitney *U* tests (if the Kruskal-Wallis test had been used). Comparisons of before and after therapy were undertaken by use of the paired *t* test if the data were roughly normal or the Wilcoxon signed-rank test if they were not. All comparisons were done on a two-tailed basis. Since the number of comparisons was large and the risk of type 1 errors was increased, a probability of less than 0.01 was prespecified as the minimum acceptable designation of significance.

This study is registered with ClinicalTrials.gov, number NCT00133965.

### Role of the funding source

The funders had no role in the study design, data collection, analysis or interpretation of the data, or in writing the final report. The corresponding author had full access to all the data and final responsibility to submit for publication.

	Dignity therapy (n=108)	Standard palliative care (n=111)	Client-centred care (n=107)
Age (years)	64.2 (14.6)	66.7 (14.2)	64.3 (14.3)
PPS score	44.2 (15.2)	44.2 (15.9)	44.4 (14.4)
Sex (male)	56 (52%)	50 (45%)	55 (51%)
Marital status			
Married	60 (56%)	65 (59%)	67 (63%)
Other	48 (44%)	46 (41%)	40 (37%)
Religion*			
Catholic	31 (29%)	26 (23%)	29 (27%)
Protestant	22 (20%)	30 (27%)	16 (15%)
Jewish	17 (16%)	11 (10%)	15 (14%)
Anglican	17 (16%)	16 (14%)	18 (17%)
Other	11 (10%)	18 (16%)	14 (13%)
None	9 (8%)	10 (9%)	14 (13%)
Living arrangements			
Alone	30 (28%)	29 (26%)	27 (25%)
Spouse	37 (34%)	50 (45%)	51 (48%)
Others	18 (17%)	21 (19%)	13 (12%)
Combination	23 (21%)	11 (10%)	16 (15%)
Education			
None or primary	11 (10%)	11 (10%)	8 (7%)
High school	38 (35%)	36 (32%)	33 (31%)
College	43 (40%)	47 (42%)	45 (42%)
Postgraduate	16 (15%)	17 (15%)	20 (19%)
Terminal illness			
Primary cancer site			
Lung	15 (14%)	15 (14%)	18 (17%)
Breast	10 (9%)	8 (7%)	11 (10%)
Gastrointestinal	30 (28%)	48 (43%)	29 (27%)
Genitourinary	12 (11%)	9 (8%)	8 (2%)
Gynaecological	3 (3%)	3 (3%)	5 (5%)
Haematological	5 (5%)	6 (5%)	3 (3%)
Brain	4 (4%)	2 (2%)	3 (3%)
Other solid tumours	23 (21%)	20 (18%)	24 (22%)
Non-malignant	5 (5%)	2 (2%)	5 (5%)
Missing	1 (<1%)	1 (<1%)	1 (<1%)
Ethnic origin			
White	98 (91%)	99 (89%)	94 (88%)
Other	10 (9%)	12 (11%)	13 (12%)
Care setting			
Palliative care unit	25 (23%)	21 (22%)	24 (22%)
Inpatient palliative care service	7 (6%)	9 (9%)	13 (12%)
Outpatient or home	64 (59%)	70 (60%)	61 (57%)
Hospice	12 (11%)	11 (9%)	9 (8%)

Data are number (%) or mean (SD). PPS=Palliative Performance Scale. \*Two individuals (one in dignity therapy group and one in client-centred care group) did not provide an answer to the question about religious affiliation.

**Table 1: Demographic characteristics of patients at baseline**

	Dignity therapy (n=108)		Standard palliative care (n=111)		Client-centred care (n=107)	
	Baseline	Study completion	Baseline	Study completion	Baseline	Study completion
Not able to continue usual routines	2.47 (1.28)	2.33 (1.25)	2.70 (1.32)	2.42 (1.25)	2.51 (1.14)	2.28 (1.14)
Physically distressing symptoms	2.32 (1.29)	2.37 (1.24)	2.28 (1.21)	2.22 (1.14)	2.57 (1.33)	2.35 (1.20)
Not able to carry out important roles	1.88 (1.15)	1.88 (1.24)	2.11 (1.22)	2.05 (1.17)	2.02 (1.17)	2.02 (1.17)
No longer feeling like who I was	2.20 (1.29)	2.09 (1.27)	2.00 (1.27)	2.02 (1.15)	2.21 (1.34)	2.06 (1.26)
Not able to perform tasks of daily living	1.79 (1.08)	2.11 (1.25)	1.74 (1.15)	1.84 (1.15)	1.84 (1.11)	1.88 (1.21)
Feeling of not having control	2.15 (1.29)	2.00 (1.12)	2.08 (1.12)	2.00 (1.05)	2.15 (1.23)	2.22 (1.22)
Feeling uncertain	2.10 (1.18)	2.21 (1.22)	2.08 (1.24)	1.88 (1.09)	2.05 (1.16)	2.07 (1.23)
Not able to attend to bodily functions	1.42 (0.93)	1.82 (1.56)	1.40 (0.95)	1.64 (1.12)	1.56 (1.06)	1.61 (1.02)
Feeling anxious	1.93 (1.09)	1.90 (1.09)	1.73 (1.01)	1.80 (1.02)	1.93 (1.11)	1.92 (1.08)
Feeling of reduced privacy	1.82 (1.13)	1.84 (1.18)	1.87 (1.13)	1.73 (1.01)	1.66 (1.04)	1.83 (1.08)
Feeling a burden to others	1.90 (1.08)	1.91 (1.02)	1.99 (1.08)	1.92 (1.11)	2.07 (1.13)	2.11 (1.14)
Feeling how you look has changed	1.83 (1.12)	1.72 (1.08)	1.79 (1.11)	1.69 (1.08)	1.64 (1.04)	1.71 (0.99)
Feeling depressed	1.84 (1.12)	1.81 (1.07)	1.69 (0.95)	1.74 (0.99)	1.76 (1.00)	1.79 (1.06)
Worried about future	2.08 (1.30)	2.03 (1.26)	2.13 (1.22)	1.76 (1.03)	2.15 (1.24)	1.94 (1.17)
Not being able to think clearly	1.74 (0.93)	1.79 (1.12)	1.66 (1.04)	1.69 (0.98)	1.92 (1.12)	1.85 (1.10)
Feeling of unfinished business	1.91 (1.11)	1.79 (1.02)	1.95 (1.10)	1.86 (1.15)	2.03 (1.15)	1.91 (1.07)
Feeling life no longer has meaning or purpose	1.58 (0.98)	1.46 (0.85)	1.56 (1.05)	1.48 (0.92)	1.54 (1.01)	1.64 (1.03)
Not feeling worthwhile or valued	1.50 (0.93)	1.57 (1.15)	1.42 (0.86)	1.44 (0.91)	1.61 (1.09)	1.58 (1.06)
Feeling have not made meaningful contribution	1.33 (0.71)	1.36 (0.86)	1.30 (0.74)	1.23 (0.67)	1.39 (0.80)	1.44 (0.89)
Not feeling able to mentally fight illness	1.43 (0.84)	1.46 (0.88)	1.40 (0.79)	1.44 (0.93)	1.56 (0.90)	1.58 (0.95)
Not being able to accept things as they are	1.57 (1.02)	1.54 (0.94)	1.48 (0.86)	1.48 (0.95)	1.55 (0.91)	1.50 (0.85)
Concerns about spiritual life	1.30 (0.73)	1.37 (1.26)	1.18 (0.51)	1.23 (0.67)	1.36 (0.76)	1.24 (0.61)
Not being treated with respect	1.19 (0.59)	1.20 (0.62)	1.12 (0.40)	1.20 (0.59)	1.18 (0.53)	1.23 (0.68)
Not feeling supported by health-care providers	1.20 (0.68)	1.11 (0.42)	1.18 (0.49)	1.13 (0.49)	1.45 (0.95)	1.26 (0.76)
Not feeling supported by friends or family	1.23 (0.73)	1.08 (0.39)	1.07 (0.29)	1.14 (0.55)	1.18 (0.64)	1.23 (0.71)

Data are mean (SD). Score of 1 indicates that the treatment was not a problem, 2 a slight problem, 3 a problem, 4 a major problem, and 5 an overwhelming problem.

**Table 2: Distress measured with the Patient Dignity Inventory<sup>5</sup> at baseline and after study completion**

## Results

The figure shows the trial profile. 1513 patients were assessed for eligibility by use of a consecutive sampling approach between April, 2005, and October, 2008. After randomisation, 28 patients died, 85 withdrew because of declining health, and two failed Blessed Orientation Memory Concentration screening. These individuals were excluded from the analysis because we had either no or only incomplete data for them. Therefore, 326 participants—108 (33%) assigned to dignity therapy, 111 (34%) to standard palliative care, and 107 (33%) to client-centred care—completed the study. Recruitment was fairly even across the three study sites (Winnipeg [n=119], Perth [n=99], and New York [n=108]), with the number of participants at each site balanced across the study groups.

Table 1 shows that the baseline demographic characteristics of participants assigned to the three groups were well balanced. 161 (49%) of 326 participants were men (table 1). The mean age of the participants was 65.1 years (SD 14.4; range 22–102). 192 (59%) participants were married or living in common-law partnerships, and the rest were widowed, divorced, or not married. 138 (42%) were living with their spouse or partner, 86 (26%) were

living alone, 70 (21%) were living with children, and the rest were living with others, such as friends or other relatives. 196 (60%) were receiving home-based palliative care, 72 (22%) palliative care in an inpatient unit, 31 (10%) acute care in an inpatient unit (followed by palliative care on a non-palliative-care unit), and 27 (8%) freestanding hospice care. 68 (21%) participants were Protestant, 86 (26%) Catholic, 43 (13%) Jewish, 51 (16%) Anglican, 43 (13%) other, 33 (10%) no religious affiliation, and two individuals did not provide an answer to the question about religious affiliation. 30 (9%) individuals had less than high school education, 107 (33%) had completed high school, and 188 (58%) had completed college or postgraduate training (one individual was unable to answer). Cancer sites were gastrointestinal (108 [33%]), genitourinary (26 [8%]), lung (48 [15%]), breast (29 [9%]), gynaecological (11 [3%]), miscellaneous solid tumours (67 [21%]), haematological (14 [4%]), and brain (nine [3%]), and 12 [4%] patients had non-malignant terminal illness (ie, end-stage organ failure and amyotrophic lateral sclerosis). The mean Palliative Performance Scale rating for all participants at baseline was 44.3% (SD 15.1).

134 (41%) participants reported that they had lived with their life-limiting illness for less than 1 year, 104 (32%) for

	Dignity therapy (n=108)		Standard palliative care (n=111)		Client-centred care (n=107)	
	Baseline	Study completion	Baseline	Study completion	Baseline	Study completion
<b>Structured Interview for Symptoms and Concerns*</b>						
Loss of Dignity	0.58 (1.00)	0.67 (1.19)	0.35 (0.81)	0.51 (0.94)	0.68 (1.09)	0.47 (0.96)
Desire for Death	0.44 (0.87)	0.53 (0.88)	0.60 (1.19)	0.65 (1.29)	0.64 (1.18)	0.68 (1.18)
Sense of Suffering	1.63 (1.57)	1.34 (1.40)	1.46 (1.53)	1.41 (1.38)	1.52 (1.35)	1.46 (1.35)
Hopelessness	0.90 (1.17)	0.86 (1.27)	0.78 (1.08)	0.68 (1.31)	0.87 (1.24)	0.80 (1.17)
Depression	1.22 (1.26)	1.23 (1.23)	1.14 (1.31)	1.06 (1.38)	1.25 (1.19)	1.06 (1.25)
Suicidality	0.29 (0.72)	0.27 (0.73)	0.21 (0.66)	0.30 (1.02)	0.38 (0.86)	0.32 (0.96)
Burden to Others	1.47 (1.55)	1.33 (1.40)	1.54 (1.45)	1.24 (1.50)	1.53 (1.56)	1.43 (1.38)
<b>Edmonton Symptom Assessment Scale†</b>						
Pain	2.72 (2.79)	2.79 (2.61)	2.76 (2.80)	2.87 (2.80)	3.13 (2.84)	3.18 (2.94)
Nausea	1.52 (2.18)	1.57 (2.42)	1.62 (2.44)	1.59 (2.61)	1.30 (2.34)	1.55 (2.42)
Drowsiness	3.61 (2.96)	3.65 (2.91)	2.89 (2.71)	3.07 (2.84)	3.31 (2.92)	3.54 (2.96)
Shortness of Breath	1.98 (2.59)	2.52 (2.85)	1.70 (2.33)	1.75 (2.50)	2.35 (2.92)	2.51 (3.11)
Will to Live‡	9.08 (1.94)	8.99 (1.62)	9.05 (2.02)	8.84 (2.40)	9.00 (2.00)	9.00 (2.19)
Appetite	6.59 (3.13)	6.24 (3.04)	5.91 (3.03)	6.05 (3.18)	6.93 (2.65)	7.05 (3.00)
Level of Activity	4.59 (2.97)	4.24 (2.92)	4.59 (2.66)	4.45 (2.81)	4.80 (2.69)	4.42 (2.78)
Sense of Well Being‡	6.67 (2.67)	6.62 (2.66)	6.65 (2.64)	6.44 (2.89)	6.74 (2.62)	6.50 (2.88)
<b>Quality of life§</b>						
Rating	6.48 (2.69)	6.39 (2.54)	6.27 (2.70)	6.34 (2.47)	6.29 (2.50)	6.64 (2.55)
Satisfaction	6.34 (3.06)	6.04 (2.94)	6.10 (3.09)	6.05 (2.99)	5.83 (3.16)	6.05 (2.88)
<b>Functional Assessment of Chronic Illness Therapy (FACIT)¶ (48)</b>						
Total	34.70 (9.59)	33.97 (10.29)	33.82 (9.56)	34.44 (10.13)	32.86 (9.61)	32.43 (10.37)
FACIT-Secular (32)	24.50 (5.85)	23.88 (6.27)	23.65 (6.30)	24.04 (6.76)	23.05 (6.26)	22.80 (7.19)
FACIT-Faith (16)	10.20 (5.22)	10.09 (5.13)	10.00 (5.07)	10.40 (4.72)	9.81 (5.10)	9.63 (5.05)
<b>Hospital Anxiety and Depression Scale (HADS)   (56)</b>						
Total	11.08 (6.76)	11.45 (6.84)	11.36 (6.76)	11.39 (7.43)	12.06 (7.28)	11.76 (7.80)
HADS-Depression (28)	5.86 (3.86)	5.64 (4.07)	6.03 (4.02)	6.19 (4.21)	6.30 (4.04)	6.38 (4.30)
HADS-Anxiety (28)	5.22 (4.20)	5.81 (3.80)	5.34 (3.79)	5.20 (4.05)	5.76 (4.26)	5.38 (4.40)

The parenthesised numbers besides the scales and subscales represent the maximum scores. \*0=none, 1=minimum, 2=mild, 3=moderate, 4=strong, 5=severe, and 6=extreme. †1=no or poor, 10=worst. ‡Reverse scoring—higher scores represent better outcomes. §1=poor or not satisfied, 10=excellent or very satisfied. ¶High score indicates better spiritual wellbeing. ||High score indicates more intense depression.

**Table 3: Distress according to the Structured Interview for Symptoms and Concerns,<sup>8,9</sup> Edmonton Symptom Assessment Scale,<sup>10,11</sup> Quality of Life Scale,<sup>9,10</sup> Functional Assessment of Chronic Illness Therapy,<sup>4,5</sup> and Hospital Anxiety and Depression Scale<sup>7,12</sup> at baseline and study completion**

1–3 years, and 88 (27%) for more than 3 years. The median survival time was 110 days (IQR <56 to <235); 213 [65%] patients had died by the time the study ended; the proportion of patients surviving in each of the study groups was not significantly different between groups (62 [57%] of 108 in dignity therapy group, 78 [70%] of 111 in standard palliative care group, and 73 [68%] of 107 in client-centred care group;  $p=0.1$ ). No difference was noted in the survival times after study completion between the three groups (81 days [IQR 48–249] in dignity therapy group, 109 days [61–222] in standard palliative care group, and 128 days [67–233] in client-centred care group;  $p=0.51$ ). Age and sex distribution, Palliative Performance Scale ratings, and dropout rate (death and withdrawal because of deterioration in health), in all three study groups showed no significant differences at completion of the study.

Because no initial threshold level of distress was stipulated as an entry criterion, the sample was most noteworthy for its paucity of distress at baseline (table 2;

table 3). In most instances, patients were not greatly distressed, as indicated by the average Patient Dignity Inventory item scores and other baseline psychometric scores (table 2; table 3). Differences between before and after study measurements were calculated for Patient Dignity Inventory and Structured Interview for Symptoms and Concerns items, modified Edmonton Symptom Assessment Scale individual subscale scores, Quality of Life Scale, Functional Assessment of Chronic Illness Therapy, and Hospital Anxiety and Depression Scale; no significant differences were noted between the three groups (table 2 and table 3). The survey after the study, however, showed differences between the three study groups (table 4). Patients given dignity therapy were significantly more likely than were those in either of the two other groups to report that the treatment was helpful to them, improved their quality of life, and sense of dignity (table 4). The patients in the dignity therapy group, compared with the other study



	Dignity therapy (n=108)	Standard palliative care (n=111)	Client-centred care (n=107)	$\chi^2$ (2 df)	p value
The study group has been helpful to me	4.23 (0.64)	3.50 (1.01)	3.72 (0.89)	35.50	<0.0001*
The study group has been as helpful as any other aspect of my health care	3.63 (1.04)	3.27 (1.04)	3.12 (1.04)	6.39	0.041
The study group has improved my quality of life	3.54 (0.95)	2.96 (0.96)	2.84 (1.05)	14.52	0.001*
The study group has given me a sense of looking after unfinished business	3.35 (1.01)	2.86 (1.60)	2.93 (1.16)	6.87	0.032
The study group has improved my spiritual wellbeing	3.27 (1.09)	3.00 (1.11)	2.56 (0.98)	10.35	0.006†
The study group has lessened my sense of sadness and depression	3.11 (1.02)	2.57 (0.92)	2.65 (1.04)	9.38	0.009‡
The study group has lessened my sense of feeling a burden to others	2.81 (0.98)	2.58 (0.95)	2.53 (0.96)	2.71	0.258
The study group has made me feel more worthwhile or valued	3.38 (0.93)	3.35 (1.00)	3.02 (1.01)	3.75	0.153
The study group has made me feel like I am still me	3.81 (0.85)	3.59 (0.92)	3.40 (0.98)	5.91	0.052
The study group has given me a greater sense of having control over my life	3.02 (1.02)	3.16 (1.00)	2.81 (1.05)	2.74	0.255
The study group has helped me to accept the way things are	3.39 (1.062)	3.31 (1.01)	3.19 (1.12)	0.92	0.630
The study group has made me feel more respected and understood by others	3.16 (0.90)	3.04 (0.98)	2.79 (0.98)	3.85	0.146
The study group has made me feel that I am still able to carry out important tasks or fill an important role	3.62 (0.97)	3.48 (1.00)	3.02 (1.05)	8.96	0.011
The study group has been satisfactory	4.26 (0.63)	3.80 (0.74)	4.17 (0.56)	29.58	<0.0001‡
The study group has made me feel that life was more meaningful	3.55 (1.05)	3.19 (1.70)	3.31 (1.02)	6.73	0.035
The study group has given me a heightened sense of purpose	3.49 (1.04)	3.20 (0.98)	3.15 (1.03)	6.86	0.032
The study group has increased my sense of dignity	3.52 (1.04)	3.09 (1.02)	3.11 (0.97)	12.66	0.002*
The study group has lessened my sense of suffering	2.86 (1.04)	2.70 (1.02)	2.77 (0.98)	1.06	0.588
The study group has increased my will to live	2.94 (1.11)	2.76 (1.04)	2.92 (0.97)	1.73	0.422
The study group has or will be of help to my family	3.93 (0.80)	3.20 (1.00)	3.29 (0.99)	33.86	<0.0001*
The study group has or could change way my family sees or appreciates me	3.58 (1.01)	2.85 (1.00)	2.85 (1.04)	33.81	<0.0001*
In general, I am satisfied with my psychosocial care	4.25 (0.77)	4.14 (0.65)	3.99 (0.89)	5.97	0.051

Data are mean (SD). Score 1 is strongly disagree, 2 disagree, 3 neither agree nor disagree, 4 agree, and 5 strongly agree. NA=not applicable (detailed group comparisons only undertaken if  $p < 0.01$ ). \*Dignity therapy was significantly better than client-centred care and standard palliative care. †Dignity therapy was significantly better than client-centred care. ‡Dignity therapy was significantly better than standard palliative care.

**Table 4: Results of survey done after study completion**

groups, were also significantly more likely to report that the study treatment changed how their family saw and appreciated them, and that it had or would be of help to their family (table 4). Dignity therapy was significantly better than the client-centred care in improving spiritual wellbeing, and was significantly better than standard palliative care in terms of lessening sadness or depression; significantly more patients who had received dignity therapy reported that the study group had been satisfactory than did those who received standard palliative care (table 4).

## Discussion

Although floor effects precluded our ability to show significant differences between the study groups in terms of the primary outcomes, our secondary outcomes showed substantive benefits of dignity therapy, a novel psychotherapeutic approach, over standard palliative care and client-centred care. Patients in this group, when surveyed after the study, were significantly more likely to report benefits in terms of finding the treatment helpful, improving their quality of life, their sense of dignity, changing how their family saw or appreciated them, and helping their family than did those in the standard palliative care and client-centred care groups (panel 2).

Comprehensive palliative care requires that careful attention be paid to the physical, psychosocial, existential, and spiritual sources of end-of-life distress. Restricting the clinical focus on any of these domains to the exclusion of others does not achieve standards of palliative care as specified by WHO<sup>20</sup> and the US Institute of Medicine.<sup>21</sup> In describing factors associated with a good death, the Institute of Medicine includes preparation for death and opportunities for closure or sense of completion of the life.<sup>21</sup> Yet, although much progress has been made in our ability to achieve physical comfort for patients who are dying, few novel interventions have been designed to address the psychosocial, existential, and spiritual dimensions of end-of-life care.

Dignity therapy was developed for the purpose of lessening distress and enhancing end-of-life experience for patients nearing death. In our phase 1 trial,<sup>6</sup> nearly all patients reported that dignity therapy was helpful, whether they had disclosed substantial initial distress or not. On the basis of that finding, baseline distress was not specified as an entry criterion for the randomised controlled trial. Because the base rates of distress within our sample were quite low, it is perhaps unsurprising that we were unable to measure any significant changes in distress in the three study groups. Improvement in

**Panel 2: Research in context****Systematic review**

This study is the first randomised trial of dignity therapy. The systematic review that accompanied the application for funding of this randomised controlled trial was incorporated into two reports.<sup>17–19</sup>

**Interpretation**

Based on the findings of this study, clinicians should consider dignity therapy a viable therapeutic approach, which can enhance end-of-life experience for patients and families confronting death.

indicators of depression, will to live, and quality of life, presupposes that patients' initially consider these a problem. In the absence of such initial distress, little room exists for improvement.

Perhaps the scales we used for assessment of our primary outcome were less responsive and less sensitive to changes than were those used for the secondary outcome, whereby patients reported their experiences in the survey undertaken after study completion. The difficulty in showing end-of-life psychosocial change with self-reported measurements has been well documented. Unlike a trial of chemotherapy, in which survival time, tumour load, and overall mortality provide readily measurable and objective outcomes, defining outcomes for dignity therapy is a greater challenge. Dignity therapy was designed to positively affect the experience of people who are dying. Measurements of overt distress, with indicators of psychosocial, existential, and spiritual effect, were incorporated into the study protocol. In a recent review of quality of life measurements that are suitable for use in palliative care, none of the items could be identified as having an established responsiveness to change.<sup>22</sup> The conclusion drawn from the results of a meta-analysis is that distress before the intervention is a critical moderator of efficacy of psychosocial interventions in patients with cancer.<sup>23</sup> Murray and colleagues<sup>24</sup> have suggested that a more successful approach might be to have serial qualitative interviews. This approach, however, would not be well suited to a randomised, controlled trial.

With the lack of significant difference for the three study groups for the primary outcome measurements, we also considered whether our trial might have been underpowered. A post-hoc power calculation, based on the actual number of trial participants, determined that with 326 patients, this study had a power of 0.72–0.95 to detect an effect size of 0.15–0.30.

Patients in the dignity therapy group were significantly more likely than were those assigned to the other groups to report that this approach was helpful, improved their quality of life, enhanced their sense of dignity, and

provided benefits to their family. Although they reported their initial state of psychosocial or existential wellbeing as satisfactory, patients given dignity therapy often had an enhanced quality-of-end-of-life experience that they simply could not have expected. The effect, although difficult to assess, was often poignant and profound. For example, a 72-year-old woman with bowel cancer stated that “[dignity therapy] brought to the forefront that I have to prepare my family to the best of my ability”. A 56-year-old woman said, “Mostly I want my family to know that I’m okay with dying and they must move on”. She went on to say that “the therapy showed me I am not the cancer, I am still in here. I am so grateful for that because I lost myself....it really helped me remember who I am”.

With hindsight, the argument could be that we should have screened patients for some baseline critical distress. That might have improved the likelihood of showing differences, based on changes reported in the questionnaires before and after the intervention, in the three study groups. Our disinclination to do so, however, was based on previous findings, showing the beneficial effects of dignity therapy for nearly all patients nearing death, whether overtly distressed or not. Introduction of critical thresholds of distress for study inclusion would have excluded most patients who were most likely to participate in this study. Another consideration might have been a crossover study design, in which patients could experience and thus compare dignity therapy with other possible study treatments. However, such a design would not have been feasible, with many patients either deteriorating or dying before a comparison of the groups was possible.

Notably, of the 1513 patients who were assessed for eligibility, 42% were ineligible, mainly because they were too ill to take part in the study. Of the patients who met the eligibility criteria, half consented to participate, and nearly three-quarters of these completed the protocol. For palliative care protocols that require participants who do not have any cognitive impairment, this recruitment pattern is not at all atypical.<sup>25</sup> The nature of dignity therapy is such that only patients who are mentally capable of providing personally meaningful responses are those who will be approached to take part. Therefore, generalisability should be defined in terms of patients who maintain their cognitive capacity while they are nearing death. Although in this trial we mainly enrolled patients with cancer, the application of dignity therapy in non-cancer populations has been investigated in other studies.<sup>26</sup>

We are not aware of any instance in which patients withdrew from dignity therapy because of feeling upset or distressed. Many patients, however, indicated moments during therapy that were emotionally evocative—eg, talking about hopes or wishes for family members in anticipation of a time after their death. Mostly, however, patients appreciated the opportunity to articulate these issues. The only safety issue, which will be reported fully in a separate report of families' experiences of dignity

therapy, was that on a few occasions family members were dissatisfied with the generativity document. The family members felt the patient had become too ill to give fulsome responses, or that the answers provided a distorted image of the participant. Therefore, patients who are delirious or otherwise cognitively impaired should not be given dignity therapy.

Several studies have been done to assess dignity therapy under different circumstances. The conclusions drawn from a phase 1 Danish study<sup>27</sup> of ten health-care professionals and 20 patients was that, with minor cultural adaptations, dignity therapy was “a manageable, acceptable and relevant intervention for Danish patients admitted to palliative care”. The results of a pilot study of eight patients in rural Kentucky, USA, with end-stage cancer showed that dignity therapy could be delivered by videophone, and achieve overall benefit and high patient satisfaction.<sup>28</sup> In a phase 1 study of a cohort of 33 terminally ill patients that was done in French-speaking Canada, relevance and satisfaction were high for patients and their families.<sup>29</sup>

Despite the beneficial effects of dignity therapy, its ability to mitigate outright distress (eg, depression, desire for death or suicidality) has not yet been proven. Future research in more severely distressed patients might indeed establish its role in such circumstances. However, the purpose and potential benefit of individual psychotherapy for patients with advanced or terminal disease is not solely the symptomatic relief of stress.<sup>30</sup> Psychotherapy is also offered for the prevention of distress, promotion of wellbeing, and establishment of a sense of personal meaning and life purpose. Psychotherapeutic support can help patients cope with disappointments, process the reality of leaving behind loved ones, deal with feelings of sadness, loss, isolation, and a damaged sense of identity and personal value. It can also help patients consider personal priorities in terms of relationships, religious and spiritual beliefs, and deal with the urgency of resolving conflicts or achieve personally meaningful goals.<sup>30–33</sup> Palliative care must offer patients opportunities to engage in these complex issues, and dignity therapy is a means by which it might do so. Future research exploring the beneficial effects of dignity therapy will help to unravel the psychological, spiritual, and existential complexities for an individual facing death, and the best way to support patients with advanced and terminal illness and their families.

#### Contributors

HMC was responsible for retrieving funding for the study, overseeing the study, contributing to the data analysis, writing the initial draft of the report, and revising the report based on the recommendations of the other authors. LJK and WB were site coordinators; they oversaw patient recruitment and local staff. SM, TFH, and MH helped in the design of the protocol; they participated in monitoring the study and maintaining protocol adherence. TH oversaw the randomisation process and supervised the data analysis. All coauthors contributed to writing the final study report.

#### Conflicts of interest

We declare that we have no conflicts of interest.

#### Acknowledgments

This study is funded by the National Cancer Institute, National Institutes of Health (grant number R01CA102201). HMC is a Canada Research Chair in Palliative Care, funded by the Canadian Institutes for Health Research. We thank our research staff, including Sheila Lander, Kathy Cullihall, Beverley Cann (Winnipeg); Joanne Hale, Jenny Clarke, Sky Dawson, and Lynn Oldham (Perth); and Shannon Poppito, Wendy Lichtenthal, Amy Berg, Kimberly Pitrelli (New York).

#### References

- 1 Steinhauser KE, Christakis NA, Clipp EC, et al. Preparing for the end of life: preferences of patients, families, physicians, and other care providers. *J Pain Symptom Manage* 2001; **22**: 727–37.
- 2 Lynn J. Perspectives on care at the close of life. Serving patients who may die soon and their families: the role of hospice and other services. *JAMA* 2001; **285**: 925–32.
- 3 Emanuel L, Bennett K, Richardson VE. The dying role. *J Palliat Med* 2007; **10**: 159–68.
- 4 Chochinov HM, Hack T, McClement S, Harlos M, Kristjanson L. Dignity in the terminally ill: a developing empirical model. *Soc Sci Med* 2002; **54**: 433–43.
- 5 Chochinov HM. Dignity conserving care: a new model for palliative care. *JAMA* 2002; **287**: 2253–60.
- 6 Chochinov HM, Hack T, Hassard T, Kristjanson L, McClement S, Harlos M. Dignity therapy: a novel psychotherapeutic intervention for patients nearing death. *J Clin Oncol* 2005; **23**: 5520–25.
- 7 McClement S, Hack T, Chochinov HM, Hassard T, Kristjanson L, Harlos M. Dignity therapy: family member perspectives. *J Palliat Med* 2007; **10**: 1076–82.
- 8 Brooke P, Bullock R. Validation of a 6 item cognitive impairment test with a view to primary care usage. *Int J Geriatr Psychiatry* 1999; **14**: 936–40.
- 9 Anderson F, Downing GM, Hill J, Casorso L, Lerch N. Palliative Performance Scale (PPS): a new tool. *J Palliat Care* 1996; **12**: 5–11.
- 10 Peterman AH, Fitchett G, Brady MJ, Hernandez L, Cella D. Measuring spiritual well-being in people with cancer: the functional assessment of chronic illness therapy—spiritual well-being scale (FACIT-sp). *Ann Behav Med* 2002; **24**: 49–58.
- 11 Chochinov HM, Hassard T, McClement S, et al. The Patient Dignity Inventory: a novel way of measuring dignity related distress in palliative care. *J Pain Symptom Manage* 2008; **36**: 559–71.
- 12 Moorey S, Greer S, Watson M, et al. The factor structure and factor stability of the hospital anxiety and depression scale in patients with cancer. *Br J Psychiatry* 1991; **158**: 2555–59.
- 13 Wilson KG, Graham ID, Viola RA, et al. Structured interview assessment of symptoms and concerns in palliative care. *Can J Psychiatry* 2004; **49**: 350–58.
- 14 Graham KY, Longman AJ. Quality of life in persons with melanoma: preliminary model testing. *Cancer Nursing* 1987; **10**: 338–46.
- 15 Bruera E, Kuehn N, Miller MJ, Selmsler P, Macmillan K. The Edmonton Symptom Assessment System (ESAS): a simple method for the assessment of palliative care patients. *J Palliat Care* 1991; **7**: 6–9.
- 16 Cohen J. Statistical power analysis for the behavioral sciences (revised edition). New York: Academic Press, 1977.
- 17 Chochinov HM, Hack T, Hassard T, et al. Dignity and psychotherapeutic considerations in end of life care. *J Palliat Care* 2004; **20**: 142–43.
- 18 Chochinov HM. Dying, dignity and new horizons in palliative end-of-life care. *CA Cancer J Clin* 2006; **56**: 84–103.
- 19 Chochinov HM. Dignity therapy: final words for final days. New York: Oxford University Press (in press).
- 20 WHO. Cancer pain relief and palliative care: report of a WHO Expert Committee (technical bulletin 804). Geneva: World Health Organization, 1990.
- 21 Field MJ, Cassel CK. Approaching death: Improving care at the end of life. Washington DC: National Academy Press, 1997.
- 22 Albers G, Echteld MA, de Vet HCW, Onwuteaka-Philipsen BD, van der Linden MHM, Deliens L. Evaluation of quality-of-life measures for use in palliative care: a systematic review. *Palliat Med* 2010; **24**: 17–37.

- 23 Schneider S, Moyer A, Knapp-Oliver, Sohl S, Cannella D, Targhella V. Pre-intervention distress moderates the efficacy of psychosocial treatment for cancer patients: a meta-analysis. *J Behav Med* 2010; **33**: 1–14.
- 24 Murray SA, Kendall M, Carduff E, et al. Use of serial qualitative interviews to understand patients' evolving experiences and needs. *BMJ* 2009; **339**: b3702.
- 25 Wilson KG, Chochinov HM, McPherson CJ, et al. Desire for euthanasia or physician-assisted suicide in palliative cancer care. *Health Psychol* 2007; **26**: 314–23.
- 26 Chochinov HM, Cann B, Cullihall K, et al. Dignity therapy: a feasibility study of elders in long-term care. *J Palliat Support Care* (in press).
- 27 Houmann LJ, Rydahl-Hansen S, Chochinov HM, Kristjanson LJ, Groenvold M. Testing the feasibility of the dignity therapy interview: adaptation for the Danish culture. *BMC Palliat Care* 2010; **9**: 21.
- 28 Passik SD, Kirsh KL, Leibel S, et al. A feasibility study of dignity psychotherapy delivered via telemedicine. *Palliat Support Care* 2004; **2**: 149–55.
- 29 Gagnon P, Chochinov HM, Cochrane J, Le Moignan Moreau J, Fontaine R, Croteau L. Psychothérapie de la dignité: une intervention pour réduire la détresse psychologique chez les personnes en soins palliatifs. *Psycho-Oncologie* 2010; **4**: 169–75.
- 30 Rodin G. Individual psychotherapy for the patient with advanced disease individual psychotherapy for the patient with advanced disease. In: Chochinov HM, Breitbart W, eds. *Handbook of psychiatry and palliative medicine*. New York: Oxford University Press, 2006.
- 31 Cordova MJ, Cunningham LL, Carlson CR, Andrykowski MA. Posttraumatic growth following breast cancer: a controlled comparison study. *Health Psychol* 2001; **20**: 176–85.
- 32 Block SD. Perspectives on care at the close of life. Psychological considerations, growth, and transcendence at the end of life: the art of the possible. *JAMA* 2001; **285**: 2898–905.
- 33 Tedeschi RG, Calhoun LG. *Trauma and the transformation of suffering*. Thousand Oaks, CA: Sage Publications, 1995.