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# Dying with motor neurone disease, what can we learn from family caregivers?

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## Abstract

**Background** Increasingly, people with neurodegenerative illness are cared for at home until close to death. Yet, discussing the reality of dying remains a social taboo.

**Objective** To examine the ways, family caregivers of people living with motor neurone disease (MND) experienced the dying of their relative and to identify how health practitioners can better prepare families for end-of-life care.

**Design** Secondary analysis was undertaken on data sets generated from two longitudinal qualitative studies employing similar data collection and analysis methods. Combining data sets increased participant numbers in a low incidence disease group.

**Setting and participants** Primary studies were undertaken with family caregivers in England and Australia. Interview and observational data were collected mostly in home. Participants who discussed dying and death formed the sample for secondary analysis.

**Results** Combined data revealed four major themes: planning for end of life, unexpected dying, dignity in the dying body and positive end to MND. Despite short survival predictions, discussions among family members about dying were often sporadic and linked to loss of hope. Effective planning for death assisted caregivers to manage the final degenerative processes of dying. When plans were not effectively communicated or enacted, capacity to preserve personhood was reduced.

**Discussion and Conclusion** Returning death and dying to social discourse will raise the level of community awareness and normalize conversations about end-of-life care. Strategies for ongoing, effective communication that facilitates advance care planning among patients, their families and practitioners are essential to improve dying and death for people with MND and their family caregivers.

Trends indicate that increasingly people dying with motor neurone disease (MND), also known as amyotrophic lateral sclerosis (ALS), are being cared for at home until near their death.[1] While westernized health-care systems expect that families will provide care in chronic illness, specific support to enable family caregivers of people with non-cancer diagnoses, to manage the dying process, is under resourced.[2] Consequently, little is known about how family caregivers of people with neurodegenerative disorders construct the dying process and the death.[3] This paper draws on the findings from two parallel studies to discuss ways in which family caregivers talk about dying and death and the influences that shape their understanding of end-of-life care in MND.

## Background

Western society has been described as a death-avoiding society whose focus is health and lifestyle changes to increase longevity.[4] Until the advent of the hospice movement, most doctors focused on cure and construed death as failure, dying patients were isolated and health practitioners had difficulty talking about or communicating with dying patients and their relatives.[5] Death had become increasing medicalized, and doctors practised formalized deception, withholding terminal diagnoses from patients and families.[6] Kubler-Ross'[7] work brought some social acceptability to discussions about death and dying and enabled inherent psychosocial issues to be recognized among health practitioners and those intimately involved in care of the dying. While violent and untimely death is frequently portrayed in modern media, personal reality of dying has been described as the last great taboo of modern society.[8]

Despite the generalized reluctance to discuss dying and death, the death-avoiding culture is changing. Lobby groups and media representations concerning euthanasia and assisted suicide have awakened public interest in choices about the manner of dying and desire for control over the process. In recent years, three bills have been presented to the UK parliament calling for legalization concerning assisted dying.[9] Meanwhile, the Australian Parliament remains divided over the relevance of the euthanasia debate.[10] Despite the hesitancy of either Government to endorse any assisted measures to control the timing of death, both have legislation in place to support advance care planning including the provision of a substitute decision maker.[11,12]

Over the last decade, palliative care in England and Australia has gained a higher public profile providing more opportunities to talk about dying and death. In Australia, the health-promoting palliative care approach utilized the elements of the Ottawa Charter to increase conversations about death and dying in community and mainstream health care.[13,14] The gold standards framework (GSF) developed by Thomas[15] is integral to the National Health Service End of Life Strategy in England. Embedded in the GSF are opportunities for patients and families to discuss preferred place of care and death, as well as facilitating advance care planning (ACP).[16,17] While ACP provides directions for medical treatment, it more importantly provides an avenue for social discussion about dying and death among relatives and close friends.[18,19] People are taking opportunities to make choices about care, gain control over end of life, decide where to live while dying and avoid unwanted treatments.[20,21] Yet, for some, this new autonomy with its implied choices has created a level of uncertainty especially when death is viewed as 'existentially frightening and ambiguous'.[22 p.993] People diagnosed with MND and their caregivers often fear the dying process.[23,24] These fears are linked with choking to death, pain, loss of body control and breathlessness.[25]

MND describes a group of phenotypes that result from combinations of progressive neurodegeneration in lower and upper motor neurons. Onset is most frequently in the upper or lower limbs in 75% of cases, with the majority of the remainder presenting with bulbar onset.[26] The rate of progression of disease varies between phenotypes with certain phenotypes having a better prognosis. However, individual survival is

difficult to predict.[27] Symptoms such as fasciculations, muscle wasting, increasing weakness and loss of function (including mobility, speech and swallowing), and occasionally frontotemporal lobe disturbances, cause significant physical, emotional and social challenges, culminating in an untimely death.[28,29]

Family caregivers of people with MND usually remain central to the provision of care throughout the illness trajectory including at least the early period of dying. They become attuned to their relative's continual loss of function and grapple with the sometimes daily readjustments to care requirements.[30] However, timely access to supportive care resources is often impeded when people with MND and their families have difficulty accepting the future impact of the disease.[31,32] Many caregivers find it hard to talk about the emotional burden and non-finite physical and relational losses experienced in day-to-day caring, much less talking about dying with MND.[33,34]

## Aim

This study sought to examine the different ways family caregivers of people living with MND, constructed dying and the death event of their relative. Given the neurologically progressive, degenerative nature of MND, we wanted to know about their planning for and experiences of dying and death, to enable health and social practitioners to develop better interventions to support family caregivers providing end-of-life care.

## Methods

Secondary data analysis enables existing data to be used to answer a related research question not asked in the original study[35] when permission to reuse data has been provided as part of the consent process. This process enables the researchers to pursue emergent questions at a later stage. Comparable data sets had been generated when longitudinal qualitative studies employing similar methods were undertaken by the authors, in Australia and England. The opportunity to combine data sets increased participant numbers in a disease group where incidence is approximately 2.2 per 100 000 and the loss of participants from longitudinal studies among people with MND is common.[36–38]

The primary studies were designed to elucidate family caregivers' experiences of providing care for a relative with MND. Family caregivers participated in in-depth, semi-structured and conversational interviews at 3–4 month intervals for 18 months. For some, this included the end of life.[32,34] Audio-recorded interviews were conducted mostly in home, and field notes were made during each interaction. Secondary data analysis of our own and each others' interview transcripts revealed that both studies contained sufficient quality data to address the research question.[39]

## Sample

Based on disease incidence figures, the MND Associations of England and Victoria, Australia, estimate they are in contact with 66 and 78% of people diagnosed with MND, respectively. Recruitment of participants through MND Associations has been successful in other studies, prompting us to use this method of convenience sampling for our primary studies. Family caregivers volunteered to participate in response to invitations issued through the MND Associations. Potential participants, whose family member had a confirmed diagnosis of MND and were willing to continue with the project, contacted the researchers who explained the project in more detail and answered questions. Despite loss of potential participants through sudden health decline, 18 family caregivers from Australia and 11 from England participated in the original studies conducted between 2003 and 2006.

Caregivers who consented to a final interview after their relative's death were sampled for secondary analysis. All caregivers who were followed through to the bereavement phase in both our primary studies (except for one who was the person's daughter) were the partners of the person with MND. The ratio of male

caregivers to female caregivers is consistent with the incidence figures for MND; male-to-female ratio of 1.4 : 1.[40]

Ethics approval for the projects was granted by the respective universities and the MND Associations. Data were cleaned and coded by the primary researcher to remove any identifying information, before being shared with the other researcher.

### Data collection and analysis

Supplementary analysis is one of five types of secondary analysis described by Heaton.[39] 'Supplementary analysis involves a more in-depth focus on an emergent issue that was not addressed by the primary research'.[39 p.41] Neither of the primary studies had analysed data concerning family caregiver's experiences of death; yet, 13 family caregivers from the combined studies discussed the dying process and/or the death of their relative. Applying supplementary analysis to the data sets enabled the investigation of caregivers' construction of the dying process and the death event for people with MND.

The symbols E or A were assigned to each caregiver's data to identify the country of origin and to enable comparisons to be made. NVivo software was used to manage the data and to generate themes. A process of description and conceptual ordering[41] allowed data to be categorized and the constructions of the dying process and death event to be identified as they emerged from each caregiver's story. Data analysis including concept generation was achieved through face-to-face meetings at MND Symposia and on-going email correspondence. Sadly, Janice Brown died suddenly while we were in the final stages of analysis. Our combined work to date enabled the generation of this paper. However, some specific details such as demographics were no longer available.

## Results

In the broadly similar socio-cultural structure of Australia and England, family caregivers in both countries expressed comparable experiences of their relative's death. Their construction of the dying process varied in relation to their preparation for and place of death. In all English cases, the person died in hospital or hospice. In four of the Australian cases, the person died at home while the remainder died in a hospital or hospice. More importantly, despite the emotional and physical toll of the caregiving experience,[42] in eleven of the thirteen cases, the person with MND was cared for at home until the last days of life.

Analysis of combined data revealed four major themes: planning for end of life, unexpected dying, dignity in the dying body and positive end to MND.

### Planning for end-of-life care

Construction of the dying process and death as a planned event was variable across the data from both countries. Discussions about care and acceptance or rejection of specific symptom management options were evident in all cases. Even though the life expectancy for people with MND is 3–5 years from diagnosis to death,[29] discussions concerning dying or death only appeared sporadically in our data. Specifically, conversations between the caregiver and the dying person about end of life were only reported by three caregivers from England and four from Australia.

Consistent with the 'phenomenon of silence' among cancer patients and their families,[43] some MND caregivers suggested that discussions about the dying process or the death event were associated with loss of hope, negative attitudes or seen as unnecessary. In two cases, caregivers described a covert understanding that MND was life limiting, but that dying and death were not subjects for discussion until the last hours of life.

> 'We knew yes, but we never discussed it'. (E4)

> 'I think we were in denial for quite some while you know, we knew it was coming, but we didn't plan anything about it'. (A3)

Initiating conversations was constrained by the person with MND's unwillingness and the discomfort of family members when such

matters were raised. Three people rejected the notion that they were dying, precluding family members from knowing their wishes (A5; E3; E5).

> '[name] had it in his head that he had 8 years to live and then he would just die. He never accepted it right from the beginning. He was in a time warp, he fought all the way. In the end, we [teenage children and wife] made the decision to keep him comfortable [as opposed to antibiotic therapy and ventilation]'. (A5)

> 'His idea was that he was not going to give in to it …even when things about voluntary euthanasia came up, we never discussed it … he wasn't supposed to die yet'. (E5)

In cases where conversations about dying and death had occurred, positive experiences were reported. Plans had been made, and caregivers were able to achieve some sense of comfort.

> 'He told us everything he wanted; he always had done from the end of last year. He said you know I want this, I don't want that'. (E2)

> 'We just decided that we did not want to get involved with hospitals, he just wanted to be here … it was a conscious thing'. (A2)

However, making plans did not always lead to positive outcomes. Formalized plans in the form of two 'not for resuscitation' orders (A2, E4) and two advance care plans (E2, E3) were reported. However, despite these efforts to plan for death, problems arose when plans were not communicated effectively. E3 described her husband's unexpected death from respiratory failure as 'horrific' as she related the struggle she had to ensure his wishes were respected.

> 'He wanted them to do everything they possibly could … I had to fight to have his wishes respected. The nurse said they couldn't look after him because he wanted to be resuscitated.

> The ambulance crew resuscitated him … they tried their best. I said I know you tried and I'm really glad because if he had been [in the hospital], they wouldn't have even tried'. (E3)

Breaches of the patient wishes and minimal support from health-care workers were reported by family caregivers in two other cases where the person was dying at home (E4, A2). In E4's case, paid caregivers intervened inappropriately in the dying process. The 'not for resuscitation' order and the family caregiver's wishes were ignored. The family caregiver felt powerless to change the situation. The ensuing chaos left the caregiver with regrets about the undignified nature of his wife's death.

> E 4: I couldn't [make them stop] 'stop it, don't give her artificial resuscitation' but…

> Interviewer: Why couldn't you do that if you… if that agreement had been made?

> E4: Well she'd already started, you know, but those carers were in a hell of state and hysterical, well one of them was hysterical … a green no resuscitation and that ought to be in big letters and told to all the carers. That's the one disappointment; I would have loved her to go peacefully.

### Unexpected dying

Negative experiences were discussed by caregivers who were unprepared for the rapid deterioration and sudden death of their relative. Living with a neurodegenerative illness raised expectations that the person would eventually die of respiratory failure.[25] However, death also occurred from co-morbidities and could happen quite suddenly. Caregiver A6's mother had a short stay in hospital for a chest infection and was recovering well. As A6 was preparing to bring her home, she received a telephone call to say her mother had died.

> 'I am devastated, she was fine and we had plans to take her on holiday. We had renovated our house expecting her to live with us for the next few years'. (A6)

The unexpected nature of E5's partner's death, while in hospital, was contrary to her understanding of the illness trajectory (she imagined him becoming bedbound and needing increasing care) and deprived her of being at his side in the death event.

> 'I wasn't there with him, he died alone, the end was quick, unexpected, it shocked me, I was unprepared. He wasn't going to die; it wasn't part of the plan'. (E5)

Half the caregivers stated that they did not realize that end of life was near or they were

unprepared for the symptoms of dying. While caregivers recognized death as the end of suffering, the death experience was a shock with separation difficulties. One caregiver described her reaction to death that occurred during her husband's third fall for the day.

> 'It was not like I expected, I thought he would sleep more, go into a coma … it made me angry. He fell and hit his head... the neighbours said you'd better get an ambulance... I knew they (ambulance crew) weren't going to get him back. My biggest regret was that I was running around getting all the things for the hospital when he was dying on the floor'. (E3)

Another caregiver identified that the unexpected nature of death made it more difficult for her to reorganize her social world post-care giving. Her spouse, who required full care including ventilation support, had attended respite for 1 week on a 6-week rotation. Despite being interviewed 2 months after this death, in her mind caregiving persisted.

> 'He died so unexpectedly with no warning … I feel like he is on respite and will be coming home soon'. (A4)

On the other hand, half the caregivers were prepared for the person's death and the 'reality that the battle was over'. These caregivers constructed the dying process in a more positive manner, interacting with their partner and accepting their need to pass away. After being hospitalized for an infection at the percutaneous endoscopic gastrostomy (PEG) site that did not respond to treatment, this person's family recognized he was in the process of dying. His partner recalls:

> 'We sat with him day and night, he was groaning and restless. We gave him permission to die: close your eyes and fly away and be at peace'. (A5)

### Dignity in the dying body

Dignity in dying has been associated with autonomy, self-worth and control over the body.[44] Muscle wasting, loss of mobility, eating difficulties and communication challenges had already isolated many patients and their caregivers from their social networks.[45] As MND progressed, the indignity of 'having an accident' (faecal incontinence) or feeding or saliva issues had a detrimental effect on the person.

> 'I got the point when [name] couldn't eat, he tried to swallow and he would sick it up, he couldn't talk ... I'd cuddle him a lot and stroke him...so many times he said he didn't want to live'. (E1)

> 'After prune juice, he scoured for all the next day oh and he had an accident and that demoralized him. On Saturday prior to his death he had around the PEG a great spill of food came out. When I went to put the food in the tube, bright bile came out straightaway, gushed out and I said I don't like the look of that'. (A1)

Dignity found in their own home away from the reactions of others was important in the later stages of neurodegeneration.

> 'It was much better to have him here amongst his own things and his own people. When he was having trouble breathing, it was agonising to watch ... some friends don't cope very well with illness, they don't know how to relate'. (A8)

### Positive end to MND

Caregivers were able to construct death as the final part of the MND disease process. Despite having a variety of experiences of the dying process, caregivers reflected that while their loss of their partner was extremely significant; 'there is a huge space there that [person] occupied' (A4); they did not want them to continue to suffer with MND or go on facing the continual losses.

> 'I wouldn't have liked her to get any worse … the body was not worth having, I wouldn't have wished that [MND] on anyone'. (E4)

Hospital staff played a part in creating a positive end-of-life experience for family caregivers.

> 'They put [name] in a little ward on the side and gave us privacy. I could hold him in my arms, he had morphine drip in his leg and he just went like that'. (E1)

In two cases where the person died at home, caregivers described feelings of calmness and peace, taking their time to say goodbye.

> 'We were prepared for him to go as he wanted. He died peacefully in the chair … we took it in turns to sit with the body, we knew it was a shell and that he wasn't there'. (A2)

Although in one case, finding the person dead was a surprise, the trajectory to date and the caregiver's knowledge of MND enabled her to construct death as a positive event.

> 'I just went into [name] room and there he was ... the expression on his face indicated to me that he was peaceful, he stayed at home with us for 24 hours'. (A4)

## Discussion

The studies suggest the increasing trend towards end-of-life care at home emerging in recent years can be both challenging and rewarding for family caregivers of people with MND. Key findings indicated the reality of their experience of the dying process and death event was dependent on knowledge of the progression of MND, preparation and planning for death, knowledge of the dying process, ability to preserve dignity in the face of unpredictable neurodegeneration and expectations in relation to the death event. 'How people die remains in the memory of those that live on',[46 p.185] and impacts on their ability to readjust in bereavement.[47] If the caregiver's experience of their relative's dying with MND was unplanned, unexpected or characterized by conflict about decision making, then negative constructs were perpetuated into the bereavement phase. Yet, when caregivers reflected on the dying experience and said 'we got it right', they lived on with the positive affirmations of caregiving that stretched through the ravages of MND to a positive death experience.

Dignity is not a new concept in end-of-life care.[48] Our studies showed the importance of dignity in dying gained further significance as caregivers continued their struggle to manage the on-going losses consistent with MND. Dignity was invested in personhood and managing the continuing loss of control over body function.[49] Dignity was also about relationships, socially threatened by degenerative processes of MND such as the loss of speech or swallowing. Relationships that existed before caregiving began laid the foundation on which caregivers constructed dignity in terms of place, control and having a body that functioned in a socially acceptable manner.[50] As bodily control is progressively lost, some relatives and friends have difficulty accepting the physical decline and support networks decrease leaving the person and the family caregiver more isolated.[51] Despite activities designed to encourage discussion around dying, end-of-life choices and care options, both English and Australian societies relegate talk about death and dying to particular topics mediated through medical and media discourses. The effect of this avoidance means family caregivers and people living with MND are often not well prepared for challenges they face during the dying trajectory, nor are they well-supported by their social networks who have difficulty confronting functional loss and conversations that may include dying.[45] Public education as well as specific patient and family education is necessary, so that decisions concerning end-of-life care in MND are based on relevant, clearly understood, accurate information.

Our data indicated that where people were well informed, some positive outcomes resulted from planning for end-of-life care. However, similar to Gardner and Kramer's[48] findings, discussion about end of life was mediated by caregivers' perceptions of the patients' willingness to engage in this type of conversation. Preparation for the physiological process of dying and death is often avoided because of the emotional challenges associated with this conceptual reality.[52,53] People and/or their families have difficulty coming to terms with the reality of the life-limiting nature of MND, especially when it most often occurs at a socially unacceptable time of life. The average age of onset for women and men is 57 and 55 years of age, respectively.[26] A time of life when people in western societies expect to have independent children and be at the peak of their working life, perhaps contemplating an active retirement. Additionally, the speed and often unpredictable

nature of degeneration experienced in MND complicates family caregivers' capacity to manage the losses witnessed in their relative as the disease progressively destroys functionality and impedes socialization associated with critical day-to-day activities such as communication and communal eating.

As demonstrated in our studies, families who are hesitant to confront the neurodegenerative reality of MND may also have difficulty discussing and planning for death. Some people with MND are known to avoid face-to-face meetings with other people with MND because it is too hard to confront the reality of another person whose MND has progressed further than their own.[54] Yet, as family caregivers are likely to be providing care at home when their relative is dying, they need to have some knowledge about the dying process and what dying might look like. While health practitioners might consider they have discussed the prospect of death with patients, in practice, conversations about death often assume rather than detail the dying process.

Consequently, some caregivers found that the death they prepared for was not the death their relative experienced, compounding their emotional burden. Other caregivers indicated a level of preparedness for dying and death based on knowledge of the neurodegenerative processes of MND. From this and their own desire for a 'good' death, they had constructed images of dying characterized by increasing dependency, coma and death. Emerging out of their feelings of vulnerability and the need for safety and security amidst, the ambiguous nature of degenerative disease, dying and death was constructed as peaceful and well managed.[21] Unfortunately, in some cases, discrepancies occurred between perceptions of the dying process and actuality. Emotions from anger to profound disappointment increased already significant levels of emotional labour experienced by family caregivers.[34] Where discontinuity between expectations of dying and death occur in degenerative neurological diseases, there is an increased need for interventions to develop coping strategies and provide on-going support for family caregivers living-on after providing care.

Oliver[25] suggests that in MND, end-of-life discussions need to begin early in the disease progression and continue through the course of the disease. These conversations have important implications for not only physical management of the dying process, but also for decisions about preferred place of death, family and friends who should be present, and whether family want to provide care during the final phase. Open communication and advance care planning (ACP) have been shown to reduce negative grief-related symptoms such as sadness, apathy, guilt and depression among family caregivers.[52] Yet, poor outcomes are still occurring, despite ACP being incorporated into mainstream health policy in both countries.[55] Carefully orchestrated discussions over time can enable people with MND and their family members to reconstruct normality to include dying as part of life and enable the development of a sense of control over care in an uncontrollable disease. In countries such as England and Australia, ACP provides scope for regularizing discussions about planning and preparation for death as well as reconstructing hope based on dignity and protection of rights.[11,12,19] While it is recognized that the person and their family have to be emotionally ready to have these conversations, the often unpredictable, degenerative nature of MND[25] increases the need to facilitate preparation for dying in a timely manner.

Initiating conversations about planning for on-going care needs and end-of-life care requires complex communication skills and experience. It is important to be able to recognize cues from patients and/or family members and to look for opportunities presented by changes in health status. However, initiating and continuing conversations about dying may be difficult for health and social care workers who have been socialized in an age where the expectation in health care is for curative outcomes. Additionally, MND is an illness for which there is currently no known cure, necessitating a social reconstruction of the meaning of hope based on understanding the support available to promote

quality of life in dying.[19] Therefore, health and social care practitioners need advanced communication skills and mentoring to facilitate ongoing conversations that build trust and enhance the coping skills necessary to undertake planning for dying and death.[11] An inclusive process of effective communication about shared values and goals between the person, family and health and social care practitioners will build emotional readiness to prepare for dying and death.[56] Alternatively, if these conversations do not take place in an effective manner and there is little or no planning for dying and death, the likelihood of people with MND being able to die in their preferred place (usually at home) with the support needed to die with dignity is reduced. Further research is needed to explore the notion of emotional readiness among health and social care practitioners, the dying person and family members.

## Limitations

Although the data were collected from similar studies in two countries, the sample size was limited. However, family caregivers' constructions of death shared characteristics across the Australian and English data.

Limited resources for these studies precluded interviewing those who did not speak English. Further work is needed to examine cultural variations concerning the death experience in MND and other neurodegenerative conditions.

## Conclusions

Caregivers in this study constructed the dying process and the death event in positive and negative terms according to their knowledge of MND, their ability to provide dignified care amidst the ravages of neurodegeneration and their preparedness for the dying process and death. Returning death and dying to social discourse is an important step in normalizing end-of-life care, gaining a sense of control and maintaining support systems. Continuing to develop strategies for on-going, effective communication that facilitates advance care planning discussion among patients, their families and practitioners is an essential part of planning for death and reconstructing life after caregiving. Advanced communication skills for health and social care practitioners and strategies to provide effective community education will improve dying and the death for people with MND and their family caregivers.

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## Author contributions

RR and JB were responsible for the study conception, design, data collection, analysis and the initial work on the paper. JB did not particulate past this point due to her untimely death. RR and AS were responsible for drafting the final paper and critical revisions for important intellectual content. AS mentored the project.

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## Conflict of interest statement

No conflict of interest has been declared by the authors.

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