Social Science & Medicine 360 (2024) 117345

Contents lists available at ScienceDirect

Social Science & Medicine

journal homepage: www.elsevier.com/locate/socscimed

The impact of home mechanical ventilation on the time and manner of death for those with Motor neurone disease (MND): A qualitative study of bereaved family members

Glenys Caswell [a,*], Eleanor Wilson [b]

[a] independent scholar., Stapleford, Nottingham NG9 7EY, UK
[b] School of Health Sciences, University of Nottingham, UK

ARTICLE INFO

Handling editor: Medical Sociology Office

Keywords:
Motor neurone disease (MND)
Amyotrophic lateral sclerosis (ALS)
Ventilation
Dying
End-of-life
Time of death
Vigil

ABSTRACT

Motor neurone disease (MND) is a progressive neurodegenerative disorder which is ultimately terminal. It causes muscle weakness which can lead to the need for assistance in breathing, for some with the disease. This paper draws on qualitative research using semi-structured interviews with 32 people bereaved by the death of a family member with MND who was dependent on home mechanical ventilation, from across the United Kingdom. Interviews explored how the end-of-life of a person who had used non-invasive ventilation to assist their breathing was experienced by participants, who had cared about, and for them. Four themes are used to examine the impact of dependent ventilation technology on the experience of dying on the part of bereaved family members. Themes are: accompanied dying, planned withdrawal of ventilation, blurred time of death, time post-death. The perception and experience of time was a key component across all four themes. Ventilator technology played a critical role in sustaining life, but it could also contribute to a complex dynamic where the realities of death were mediated or obscured. This raises ethical, emotional, and existential considerations, both for the individuals receiving ventilator support and their families, as well as for healthcare professionals involved in end-of-life care.

1. Introduction

Motor Neurone Disease (MND), also known as Amyotrophic Lateral Sclerosis (ALS), is a progressive neurodegenerative disorder that leads to muscle weakness, atrophy, and eventually paralysis. Initial symptoms can include muscle twitching, fatigue, weight loss and numbness in the extremities. As the disease progresses and muscles weaken, people can lose the ability walk, move, swallow and speak. Approximately 20–40% of people living with MND can also be affected by cognitive impairments (Leigh et al., 2003). The condition affects up to 5000 adults in the UK, there is no cure (MND Association, 2022). MND often causes the progressive weakening of the muscles involved in breathing, with respiratory failure as a common cause of death (Masrori and Van Damme, 2020). In such cases, home mechanical ventilation can be used to support respiratory function. In the UK non-invasive ventilation (NIV) is primarily used, although a small proportion of patients may have invasive ventilation through a tracheostomy tube in the neck(Palmer et al., 2020a). NIV consists of a mask attached by a tube to a ventilator machine. The ventilator is usually a portable device that runs on electricity, by battery, or both. Home mechanical ventilation supports breathing by improving the natural flow of air each time the person takes a breath. There are a range of different types of masks, which usually cover the nose and mouth, or just the nose (Motor Neurone Disease Association, 2022). Ventilator settings and masks are adjusted to suit the individual. Ventilation can significantly improve quality of life for people living with MND by alleviating respiratory distress and enhancing overall comfort (Bourke et al., 2006). There is also evidence that by addressing respiratory insufficiency, ventilation can contribute to extended life expectancy, but cannot prolong it indefinitely (O`Brien et al., 2019; Palmer et al., 2020b). This allows patients to remain at home, in familiar surroundings and with their support networks.

Ventilation in MND can have significant impacts on decision making at the end-of-life, which can be nuanced for patients, families, and the health professionals involved in their care (Faull et al., 2014, ; Phelps et al., 2022: Phelps, Regen et al. 2022;Wilson et al., 2024) . Faull's work showed that health professionals find the withdrawal of ventilation at the patient's request to be ethically and morally challenging, despite being legally sound (Faull et al., 2014; Phelps et al., 2017). For patients

---

* Corresponding author.
E-mail addresses: glenyscaswell@gmail.com (G. Caswell), Eleanor.wilson@nottingham.ac.uk (E. Wilson).

https://doi.org/10.1016/j.socscimed.2024.117345
Received 29 January 2024; Received in revised form 9 September 2024; Accepted 12 September 2024

and families, a lack of clear understanding of options about ventilation use at the end of life may lead to a reluctance to make decisions in advance (Wilson et al., 2024).

As respiratory function deteriorates patients become dependent on their ventilation, meaning they are unable to breathe comfortably without it (Mercadante and Al-Husinat, 2023). They may be able to tolerate short breaks to tend to hygiene, but if the ventilation were removed for a longer period, it would result in their death. In the UK, and many other countries, ventilation is considered a treatment, and as such can be refused at any time, indeed to continue with that treatment against the patient's wishes would constitute a criminal offence (Mental Capacity Act 2005, General Medcial Council, 2010, Association for Palliative Medicine of Great Britain and Ireland, 2015). Given the progressive nature of MND, care-planning discussions become essential. These conversations involve decisions about the continuation or withdrawal of ventilation based on the individual's wishes and values (Oliver, 2004; Faull and Oliver, 2016; Mercadante and Al-Husinat, 2023).

Little is known about people experiences of the impact of technologies, such as ventilation, on the dying process. Efstathiou et al.'s (2020) systematic review provides some insight into the withdrawal of ventilation in the intensive Care Unit, suggesting that being able to be present at the time off death can have positive impacts for family members. A scoping review by (Wilson et al., (2022) draws together the available literature more specifically for MND and emphasises the paucity of knowledge on dying when ventilation is a factor. The review highlights that there is a small body of clinically focused work illustrating the physiological changes and management of dying, with particular emphasis on withdrawal of ventilation (Kühnlein et al., 2008; LeBon and Fisher, 2011, Dreyer et al., 2012; Kettemann et al., 2017Kettemann, Funke et al. 2017; Faull and Wenzel, 2020, Messer et al., 2020). Yet what dying is like and how it is experienced by family members in these circumstances has yet to be clearly documented(Wilson et al., 2022).

Bereaved family members often describe a mix of emotions during the final moments of a relative's life, including sorrow, relief, and a sense of closure (Hamer, 2021). Importance is placed on a cultural script in the UK which carries the expectation that families will maintain a vigil beside a family member who is dying, irrespective of how the person is dying (Caswell et al., 2022). Family members, and sometimes close friends, make an effort to accompany the person even though they are often unsure how long this vigil may last or how to behave at the bedside. It is not unusual to miss the moment when the person dies, whether through absence from the bedside for some reason or through failing to realise that the person for whom they are keeping vigil has died (Caswell et al., 2022 Donnelly and Battley, 2010; Seale, 1998).

The use of technology can also affect the experiences of family members when a person is dying. People in a persistent vegetative state, for example, can appear as if they are alive, with good facial colour and the appearance of breathing independently. However, it is the life support system that is keeping their body breathing and the blood circulating (Kitzinger and Kitzinger, 2014a, 2014b). Such supportive treatments may be withdrawn in order to allow a person who will never recover to die, and the timing of this may be managed so as to facilitate organ donation. The death of the body may thus be delayed to maintain the organs in a suitable condition to be transplanted into another body and save a life. Technologies can also be used to defer the moment when someone dies, making the identification of when, or even whether, someone has died more complex. Research on the use of defibrillators in public spaces discovered that they are sometimes used to obscure the fact of a death from bystanders and move the death into the private location of an ambulance. The timing of the dying is thus difficult for bystanders to discern (Timmons et al., 2010).

The boundaries between living and dying are thus sometimes opaque and lack clarity, making it difficult to ascribe a precise time to a death. The moment of death is perceived as significant, even if not easily identifiable, and the usual boundaries of time are often suspended when someone is dying. The concept of a time of death, an identifiable moment at which someone dies, is a social construction (Caswell, 2024). Time itself is also a complex construct. It is measured using technologies created by humans, some of which, such as the months and hours, are loosely based upon events in the natural world, such as the seasons and the rising and setting sun. Other ways of measuring time, such as the week, are inventions which are not based on anything in the natural world. Assigning a time to 'the when' of a person's death is therefore not straightforward, and it is made more complex by the application of technologies (Caswell, 2024).

For an individual who is experiencing the death of someone to whom they are close time often appears to behave in an unusual way, unrelated to the time as shown on a clock (Sellen et al., 2021). The present may seem to expand or contract, for time becomes fluid in the presence of death and grief, it can slow down, speed up, and even appear to stop. During the period of dying, the people around them have no control over the process, nor over when the dying ends and they have to begin to engage with their person no longer being alive (Caswell, 2024; Sellen et al., 2021). Time is inextricably linked with human experience, including dying, so that to understand human experience it is necessary to examine its temporal elements (Adam, 2004). Despite the integral relationship of dying and time, there is a lack of research exploring the ways in which time is experienced vis-a-vis dying. In this paper we make a unique contribution to the literature.

The main aim of the paper is to explore the impact which the use of non-invasive ventilation technology by people with MND has on the experience of their dying on the part of bereaved family members. The secondary aim is to examine the embeddedness of time within that experience.

2. Research study

2.1. Study design

The study employed a qualitative methodology to explore patients' and family members' experiences of end-of-life decisions about continuing or withdrawing home mechanical ventilation. Findings from the wider study data, including interview topic guides, have been published elsewhere (Wilson and Turner, 2024). Data reported in this paper were generated by interviews with bereaved family members of patients with MND who were dependent on their ventilation and subsequently died, either with ventilation in place, or once it had been withdrawn. Bereaved family members were asked to reflect on their experiences of care for someone with MND and the subsequent death of that person.

2.2. Setting

This study primarily took place remotely, as adjustments were made due to the ongoing impacts of Covid-19, allowing the inclusion of participants from across the UK.

Ethical approval

Ethical approval for the study was granted by the London-Dulwich Research Ethics Committee for the NHS Health Research Authority in April 2021 (REC ref: 21/PR/0252).

2.3. Data collection

Participants were eligible if they had been involved in the care of someone with MND who had been dependent on ventilation, either dying with it in place or as a result of withdrawal. Family members were those who had been bereaved since the December 31, 2015 and at least 8 weeks prior to study involvement. Participants could self-refer to the study having seen it promoted via the Motor Neurone Disease Association website, newsletters or other forums. Recruitment also took place via clinical teams at five NHS Trusts and two hospices, where bereaved family members were identified and approached by a healthcare professional known to them. Once given, or sent, the participant information sheet they could contact the researcher directly if they wished to take part or discuss the study further.

Interviews were carried out by EW, who is an experienced qualitative researcher with experience in end-of-life care research and MND. They were conducted via an approved video conferencing platform, telephone, email or (when suitable) in-person. Participants could choose which medium they preferred. Interviews were conducted between May 2021 and November 2023. An interview schedule was used as a guide, but all interviews were tailored to the participant and their chosen medium of communication. The study design and interview schedule were developed in collaboration with a Project Advisory Group, made up of clinicians with experience in MND, palliative care, and respiratory management as well as a bereaved family member as a Patient and Public Involvement (PPI) participant. The Project Advisory Group and a researcher mentor gave support to EW throughout the duration of the study.

Participants predominantly gave consent via an online consent form, but some used a written consent form and one chose to have their verbal consent audio recorded. This flexibility in design allowed participants to respond in the way most appropriate for them. With permission, all interviews were audio recorded using an external digital recording device. Notes were also made after each interview.

2.4. Data analysis

All interviews were transcribed and anonymised, and ranged in length from 40 to 126 min. A thematic analysis was used, to enable the identification of key concepts contained within the data set and to capture both recurring experiences and those which differed. The process started with an initial process of open inductive coding by EW and supported by the use of NVivo14© (Jackson and Bazeley, 2019). Once initial codes were identified these were reviewed and further explored by EW and GC. Decisions about data to be included in the paper were made collaboratively by both authors in discussion. This iterative process was used to explore, identify and discuss themes, as well as reframing and reorganising them (Bazeley, 2021). It was during this process that the themes for this paper were drawn out, enabling us to understand the complexity of the dying process and the nature of its course in the context of ventilator dependence. The theme titles were reviewed and refined during the peer review process. Pseudonyms are used in the presentation of findings in order to protect the anonymity of participants.

3. Findings

Thirty-six bereaved family members took part in 30 individual and three joint interviews, discussing 32 different people who died with MND having been dependent on their ventilation. Twenty were conducted using an online video platform, eight on the telephone, four in-person (in the participant's home), and one via email correspondence. Participants were predominantly female (n = 30, 83%), and spouses of the people they talked about. However, six daughters, two sons and two parents also took part. Of the 32 people discussed 19 died at home, 10 in a hospice and three in hospital. They had known of their MND diagnosis from between 2 months and 8 years. Of the 32 patients reported on, 15 died with their ventilation in place and 17 had it withdrawn.

Findings are presented using four themes as a lens through which to explore the experiences of bereaved family members with regard to the time when their relative died. The people with MND all utilised non-invasive assisted ventilation as they needed support in order to breathe. The patients discussed by their bereaved family member were considered to be dependent on their ventilation, meaning they could only tolerate short periods without it. Removal of their ventilation at this stage in their illness would result in their death. As their respiratory condition deteriorated, many had gradually increased their ventilation use over an extended period of time, sometime years. Their families and friends therefore became accustomed to seeing them in the mask.

3.1. Accompanied dying

Participants talked about the ways in which they had accompanied their dying relatives and, in some cases, the ways in which they planned for the last days and hours of their relative's life. For some people with MND dependent on ventilation and their families, however, it was possible to take a proactive approach to planning for the final hours of their lives. This could happen either as part of a planned withdrawal of ventilation or once it was clear the person with MND had shifted into the dying phase.

Elizabeth's husband planned when his mask would be removed, and on the day before this was scheduled to occur, they arranged for members of the extended family to come and say goodbye.

> *Yeah, we had a party. When he'd made his [decision], the night before it was going to happen I told everybody to come, all the family to come, grandchildren and everything, and we all sat and drank rum punches and had fish and chips from the local fish and chip shop. So everybody said their goodbyes* (Elizabeth).

Mary also planned a party at which family members and the person with MND's closest friend would have the opportunity to see him for the last time and to say goodbye. Mary said:

> *They all came here for the day and we had like a big party kind of atmosphere and it was just a real happy day all together and even though [he] was on the machine on his bed everybody came and had special time with [him] and the family all decided they'd want to say goodbye to him then. So they did. And we had lots of photographs taken and laughs and it was a real happy day*. (Mary)

When Victoria was told by the hospice nurse caring for her mother that she was likely to die soon, she called her sisters and they all came to be with their mother. Victoria said:

> *So my sisters come round and then we said right OK we'll take it in turns and somebody always be with mum, you always want to hope that somebody can be there [don't] you?* (Victoria)

Other families were also keen that their relative should be accompanied through the last hours of their lives, and family members gathered to ensure that this should happen. Time was spent talking, being together, laughing and joking, eating, holding the person with MND's hand, reading to them, opening the doors to let in the sunshine. Evelyn, for example, said 'the whole family came in and we had a picnic on his legs and we smiled and we joked and we talked about things [like] he could hear us'.

Iris spoke about being unsure what to do when sitting with her husband who was dying. She said:

> *So when [he] had been diagnosed in that summer when we were away he'd sent me an article … about death and what it meant to quantum physics. And I'd kept it open on my phone ever since and so I read it back to him while he was dying. Because I didn't know what to say or do and I read that hearing is the last thing to go and I wanted him to hear that we were with him.* (Iris)

The timing of the vigil was important. If it went on for too long it could exhaust family members. Victoria's mother was dying for a longer period than the health professionals had expected, and Victoria said, 'And then a week later, of course you're exhausted, I was literally an hour and a half of sleep a day'.

Two participants spoke about how their relative died whilst alone. Stacey's husband died when she had left him alone, sleeping. Kate's husband had not wanted her to see him die, nor to see his body after his death but she seemed to feel a social pressure to do this when staff invited her to accompany her husband's dying. She said of this time:

> *Even then at the end-of-life he'd always said I don't want you to see me die. And they called me in and said oh we think you'd better come in and I didn't want to say no then, you know what I mean … Oh I don't want to come in and see my husband's last breath or whatever. So I went in and that was difficult because I could see he'd gone and they'd taken the mask off. I'd seen him for like four months with a mask on 24/7, so that was really difficult and I couldn't sit with him after that. I said 'well he didn't want me to see him dead, so I'm not sitting in a room with him when he's not here'.* (Kate)

Most participants wished to spend time with their dying relative. Others, however, like Mary and Elizabeth, described an active process of forward planning, so that friends and members of the wider family could spend time with the dying person and have the opportunity to say their goodbyes.

3.2. *Planned withdrawal of ventilation*

Most family members who took part in a research interview were aware that their relative was approaching the end of their life. One key factor in how that period of time was experienced was whether or not there were plans put in place and then implemented for the cessation of assisted ventilation and the removal of the mask. For a planned removal to happen it was necessary that the person with MND and their family were supported by healthcare professionals who were both knowledgeable about and skilled in the care of people with MND, who were also able to make an accurate prognosis of when their patient was coming to the end-of-life. It was also helpful if the person with MND had either expressed their wishes in advance or was able to do so as they reached the end of their life.

In a joint interview, for example, Pamela and David described how their son and his wife had discussed with the respiratory consultant about the best way to manage the removal of his mask. Timing was thus paramount, as was the need they felt for their son to be without pain or suffering. When the time came, in accordance with his wishes, their son was sedated before his mask was removed, and he then died more quickly than the consultant had expected. David said:

> He was completely comfortable, he was deeply peaceful and unaware, and so they were just letting nature take its course at that point … he was totally up for what was happening before it began and it was his choice which is so important in terms of personal dignity. (David)

For Pamela and David, the planned removal of their son's ventilation and mask enabled them to ensure that he was comfortable and peaceful when he died. It also meant that the manner of his death was in accordance with the person that he was and his choices were respected.

Olivia said that her mother 'couldn't take it (mask) off herself, but it was taken off at her request when she was ready.' Her brother, Sam, who took part in a separate interview, said that their mother had asked for her mask to be removed with the express intention of being able to say goodbye to her family before she died. Professionals suggested that it might be a good idea to do this before they began to administer medications to assist with adverse symptoms, as they were not sure how long an interval there would be between removal of the mask and their mother's death. She was admitted to hospice and, after checking with her that she still wished for this to occur, her mask was removed.

> It wasn't very easy to talk to mum when she had the ventilator on … yes she took it off and then they were like oh and then we'll just play it by ear, whatever, if you're feeling OK we'll just keep it off. (Sam)

It was several hours later when she died, and during most of that time, due to medication keeping her comfortable, she was able to relax her breathing and chat with her family. Olivia said:

> it was nice in a way to have this mask off and to be able to have a conversation, not be worrying or feeling, you feel a bit guilty when she's got her mask off and [without medication she would be] suffering just to have a conversation with you. (Olivia)

The time which it took for the person with MND to die once their ventilation had been removed, and participants' perception of time, varied. Sarah said of her husband that 'he slipped away immediately, just as he knew he would.' Mary had a similar experience, saying 'They took his mask off and as I said within 2 min he had passed away.' Penny commented on how peaceful her relative appeared, saying '(husband) took two or three breaths, shallow breaths and died, and that was the end of it. He looked very peaceful.'

For Kate, however, there was some doubt as to when her husband died, whether it was when his mask was removed or if he had died before then. She said:

> *I always wonder had he died before, has he died already and they've just, but no they said they went to turn him and they said we think you'd better come in. And they said 'are we OK to take the mask off now?' and I said 'yeah'. So then they took the mask off and he just went. So yeah, part of me feels like he'd already gone … I think he might have died earlier than that … I felt and I said to my mum I think he was, he'd already passed away, which is fine, I'm fine with that.* (Kate)

Kate's experience highlights the difficulty in being sure exactly when a person has died, and the part which the removal of their assisted ventilation mask may, or may not, play in that process. Claire and her family were left alone with her father after the mask had been removed. The time which passed before he died 'felt like it was really quick, but they said it was about an hour or two but felt like 15 min' Lesley was also uncertain about the passage of time once her husband's mask had been removed, saying 'I think it took and my memory may be wrong, but the records may say, but it seemed to take an hour, possibly two before he actually passed away'.

In most cases where there was a planned removal of the mask it was a healthcare professional who performed the task. In two cases members of the family were asked if they would like to take the mask off. Lesley experienced this as 'something I wanted to do as a sort of caring' for her husband, but Susan perceived the suggestion differently. She said, 'I can remember feeling really shocked, thinking no. And then I said no … Do you want to kill them? That's what it felt like to me.' It seemed to be too onerous to ask her to remove her husband's final life sustaining measure.

The removal of the mask was, in most cases, a precursor to the death of the person with MND, but it was also an opportunity for their family to see their face without covering, as well as to have the opportunity to say goodbye. Pamela noted that it was 'the first time that we'd seen him without his mask for weeks and weeks'. Owen felt that the removal of the mask 'made her look a bit more like (herself)'. Seeing the person without their mask could, however, also be a difficult experience, as described by Claire:

> *But I guess the thing that we didn't really think about was what his face was going to look like under that mask, because he hadn't taken if off for so long. And obviously, he'd lost a lot of weight, but for some reason didn't quite notice it in his face because he'd had the mask on as a distraction. So I think that was quite shocking when they took it off.* (Claire)

3.3. *Blurred time of death*

Several participants described how their relative died with the ventilation mask still in place, and how this obscured the fact of their death and blurred time of death. There were a number of factors identified in the data which we noted to have influenced this, including an unwillingness to engage with planning for the future on the part of either professionals or people with MND and their close families; a lack of recognition that the person was imminently dying; a rapid decline; the provision of care by health professionals who have less experience of caring for people with MND.

The lack of forward planning did leave some participants with the impression that their relative was not necessarily about to die. Fiona, for example, said:

> I didn't expect him to actually die. I suppose that's probably always true isn't it? … And then I went upstairs when the carer arrived and I heard the ventilator being in a different beep that I hadn't heard before. And we went in, and as I say I think the carer realised that he'd died. But I didn't realise - it was probably shock and things … and I think it was because he'd still been breathing but with the ventilator on and it was a signal to say he wasn't breathing anymore even though the ventilator was still working. (Fiona)

Stacey thought that her husband had died but was uncertain whether it would be all right to remove the ventilator before the doctor came to see him.

> *So, the doctor came, yes, and pronounced him dead and they said that, they took ten o'clock, but I mean it could have been a bit earlier because of the ventilator, don't know, but he never, ever talked about not having the ventilator, that never came into any conversation, because I don't think either of us thought he'd reach that stage, to be honest* (Stacey).

The topic of the removal of the mask in a planned way had not been discussed, and Stacey thought it likely that her husband did not realise how far along the dying trajectory he was. There was also a degree of uncertainty about the mask and when it would be appropriate to remove it.

Victoria was aware that the assisted ventilation could make it appear that her mother was still breathing even when she was not. She said:

> *She (hospice nurse) said it can be difficult to know if somebody's passed because of the air being pushed in can make it look like they're still breathing which it does for sure … yeah and when I took it off I could tell straightaway, I think mum had actually been passed away a good half an hour … it wasn't that she'd just gone, she'd definitely been gone a while* (Victoria).

Dylan, however, had not realised that his mother had died, until a hospice nurse visited to make sure she was comfortable:

> *He actually realised she had passed away. We hadn't realised. Because she was still on the [ventilator] the nurse said you won't tell if she's passed, and they said she'd passed quite a long time, an hour or two. So she passed very peacefully* (Dylan).

Retention of the mask left some participants with an enduring impression that their relative was kept alive by the ventilator longer than was perhaps desirable. Grace, for example, said:

> I just feel that last week when his oxygen was between 44 and 75, if someone had said take [the mask] off while he's asleep he might have gone in his sleep more peacefully than he did …. Are they not telling people that's what's going to happen? If your partner's on a ventilator, or your mum, dad, whoever is on a ventilator, that is just going to keep them going. Even though they're dying that's going to keep them going longer … He's obviously dying, but why is he still alive with [saturation levels] of that? And literally we took it off and, because I said to the girls do I take it off and they said do what you think is right mum. And I took it off and he went really quickly. (Grace)

A nurse from their local hospice talked to Owen about her judgement that:

> *It's time to end it really because it was explained that it was the ventilator, it was only the ventilator that was keeping her alive at that point, in terms of take the ventilator away and she's gone. It's just a machine keeping her alive.* (Owen)

The mask being in place and the noise of the ventilator made it difficult for participants to be sure when or ever whether, their relative had died. Some participants felt that the ventilator had kept their person breathing beyond the breakdown of other bodily functions, potentially continuing the person's suffering for longer than was necessary.

> *I stayed [at the hospice] overnight that week and I mean he was gone. He wasn't there. And that suddenly, his skin had all broken down. And it was just horrific. So if he'd gone a week sooner it would have been better. I think for that week the ventilation was keeping him alive artificially. That's how I felt. So when it was put to us that they were trying to withdraw it, I had no question about it, no doubt. That was what had to be done.* (Angela)

Once the futility of the treatment was recognised and family members thought that their relative's suffering was being prolonged, removal of the ventilation was welcomed. When the removal of the mask occurred on the cusp of death it was not often clear if the person had died prior to withdrawal or once withdrawal took place, again blurring the time of death and shifting understanding of when 'death' occurred for these families.

3.4. Time post-death

As noted in the previous sections the presence of ventilation and a mask could obscure the time of death. Seeing their relative without the mask was of benefit to some and distressing for others. Evelyn referred to this as '*… you're shielded by a mask, the full impact of what is underneath is not available to you*', until the mask is removed, either just prior to, or after, death. As relatives often did not feel comfortable, and were sometimes told not to remove the mask until a health professional verified the death, the time post-death was often obfuscated. When asked who removed her husband's mask Emma said:

> *I did, because, what happened is I didn't even know when [he] passed away. I mean I was holding him but then one of the ambulance people said that, he said you can take the mask off now, his heart stopped five minutes ago. So that's when I took the mask off.* (Emma)

The usual practice after a death in the UK is for the death to be verified by a suitably qualified health professional and then to be certified by a doctor. The process of certification is a legal requirement which must be completed before the death can be registered and arrangements made for the funeral, if there is going to be one. Most participants had spent time both caring for, and caring about, their relative with MND who died. Neither form of caring stops at death and most participants talked about some of the different ways in which they continued to enact their care after the person had died.

Kate did not sit with her husband after his death, as that had been his wish. Several participants did, however, spend time sitting with their deceased relative. For example, Sarah said, 'Once they confirmed his death they left me alone with him until I was ready.' Carol said that, after her wife had died, 'I sat with her for 10 min and talked to her, you know, as if she was there.'

Part of the process of preparing the body of the deceased person for the next stage of the transition is to wash them. Mary was offered the opportunity to help wash her husband's body, but she preferred not to do so, although some participants did wash their relative. Julia washed her husband's body before sitting with him, saying 'after he'd died we cleaned him and washed him as well … and I was able to stay there as long as I wanted with him.'

Emma's husband died at home. The nurse offered to wash him, but Emma preferred to do this herself. Lauren also made the decision to care for her husband after his death and to involve their children in this process to support their grieving. The family and a close friend took on preparation of his body, as well as the ongoing care of his body and the funeral, without the input of a funeral director. Lauren experienced this as special and as a way of caring for her husband. She said:

> *So we washed him. We were like oh, that's really quick and easy. … you can care for someone in death as you would in life and people said well wasn't it hard … He was still, he was still him and it was still his body and it was still warm and it felt really special. … To me that was really important rather than sending him off to strangers to be put in a fridge for two weeks … We wrapped him and all the rest of it. … And then the burial itself, we put him in our van which we'd planned and we like took him to the burial ground and we had a celebrant there but we carried him ourselves.* (Lauren)

4. Discussion

Participants in the study were all bereaved family members, who drew on their experiences and memories in order to offer their perspective on the death of their relative, who used home mechanical ventilation. Time, and the ways in which participants experienced and perceived it, formed a common feature across all four themes. It was key to processes of decision making with regard to both the planned and unplanned withdrawal of assisted ventilation. Dying of MND while dependent on assisted ventilation is unusual as it appears to offer a choice between retaining the ventilation and waiting for death to occur, or taking charge of the process and planning for the removal of the mask and the end-of-life. Planning for the withdrawal of ventilation and removal of the mask creates an opportunity for a person with MND and those around them to say goodbye, in a way that they choose. This is what some participants did, opting to arrange for people to come and see them before they died and to be able to celebrate their life while they still lived it. This was not a viable option for everyone, some people preferred to wait and see what would happen, and others were not fully aware of the withdrawal process in order to make an informed decision about it (Wilson et al., 2024).

That some participants supported relatives who made the decision to stop treatment and thus avoid a potentially lengthened progress towards death highlights the debate that exists between the prolongation of life versus the prolongation of dying. Ventilator technology can obscure the physical manifestations of the dying process and blur time of death. Individuals on ventilators may appear more stable, so that the traditional signs of imminent death, such as changes in skin colour or respiratory patterns, may be less apparent. The use of ventilators can also alter the perception of death, potentially leading to a sense of distance from the natural progression of life's end. The artificial support provided by technology may create a buffer that separates the individual, their families and friends, and healthcare professionals from the immediate, visceral experience of death.

Families may experience a form of anticipatory grief when the person is on a ventilator, aware that their time together is limited (Dekker, 2022). The uncertainty surrounding the timing of death, coupled with the visible signs of life-sustaining measures, can make the grieving process complex and emotionally challenging. As Mary described, the death of the person with MND leaves their family and friends grief stricken, but their pain was balanced against the wish for suffering to end.

As other studies have found, technology can obscure the natural course of death, appearing to defer it (Kitzinger and Kitzinger, 2014a, 2014b; Timmermans, 2005; Timmons et al., 2010). This has a number of effects, including that it can make it challenging for families to discern when it is ethically and emotionally appropriate to transition to end-of-life care. As the study shows people with MND who are wearing a ventilation mask can appear to be breathing, even when the breath and the movement are caused by the functioning of the ventilator. This can make it hard for their family to be sure when they die and makes the moment of mask removal significant and memorable (Kitzinger and Kitzinger, 2014a, 2014b). Discussions between healthcare professionals and the person with MND and their close family about how the dying might occur can play a useful role by acknowledging the limitations of technology (Wilson et al., 2024). This study has shown that further exploration between health professionals and family members about the impact of the mask itself and the change in facial features could also be useful in preparing family members.

Time was a key component of all the experiences which participants spoke about in relation to the death of their person with MND. As the person approached the end of their life, participants experienced the pressure of the social norm to spend time in vigil with their relative, no matter how long that period of time was. Timely recognition that dying is likely to occur within the next days or hours can be difficult, even for clinicians. (Colquhoun-Flannery et al., 2024). However, when someone is known to be dying of a terminal illness, such as MND, the length of time over which a vigil can take place is potentially extended (Caswell et al., 2022).

Being there when someone dies is considered socially important (Caswell et al., 2022; Donnelly and Battley, 2010), and the concept of dying alone is considered socially undesirable (Caswell, 2022). Maintaining a vigil alongside a person who is dying is a common cultural script in the UK, understood by families and encouraged by healthcare professionals. Experiences during the Covid-19 pandemic, when families were prevented from spending time with dying family members as a way of controlling the spread of the infection, highlighted the perceived importance of the deathbed vigil (Anderson-Shaw and Zar, 2020).

The concept of the vigil suggests a period of passive waiting for death during which family members may talk, hold the dying person's hand, eat, laugh or sit quietly (Caswell et al., 2022). Findings from this study suggest that planned withdrawal of ventilation allows for more active goodbyes, which can concentrate the vigil and shift the emphasis to a more celebratory event. When the use of ventilation blurs the time of death this has the potential to disrupt the vigil, by obscuring whether or not someone is present when the person died. We have reported how, for some participants, there was uncertainty about the time when their relative died. This uncertainty was impacted by the length of time that was spent wearing a ventilation mask, the timing of the mask's removal, and the timeframe between removal of the mask and the person's death. When bereaved participants spoke about sitting with the person with MND as they were dying their perception of time was untethered to time as measured by clocks. Like previous research participants who had experienced the death of someone about whom they cared, they felt that time was fluid, rather than passing in a regular, linear fashion (Caswell, 2024). Their timescape was composed of myriad temporal features that were an integral part of the experience of their person dying. These temporal features included such things as the tempo, or pace and intensity of activities; the patterns and sequences of timing; and the time frames within which their experiences occurred. Such features are largely taken for granted and unremarked; individuals are used to time behaving in strange ways, so rarely comment upon it (Adam, 2004).

The usual temporal landscape is shifted in the post-death period, too, and it is not unusual for people who have just experienced a bereavement to spend time with the body of the person who died, and this is not unique to the participants in this study. However, the facial changes covered screened by the mask were highlighted as an important feature of the death period. Spending time with their relative's body helps, perhaps, to cushion the abruptness of the death, which can be reinforced when the person's body is removed too swiftly and their bereaved relative is left alone. A rather more unusual choice was that made by Lauren to care for her husband's body after his death. There can often be a limited understanding that there is no legal requirement for families to use the services of a funeral director (Natural Death Centre).

4.1. Strengths and limitations

This study was carefully designed to allow bereaved family members to participate in the way that they felt best suited them. We recognise that this will have had some impact on the nature of the data, but not necessarily the quality. However, encouraging and adapting research tools is vital in end-of-life care research and it is important to hear the voices of the bereaved, whatever the medium of data collection (Wilson and Turner, 2024). Being able to self-refer to the study was a strong indicator that this was an important and suitable topic of study, this would have also negated any gatekeeping issues that may have naturally been in place for those identified by healthcare professionals. The time since bereavement varied from nine weeks to over five years, which could affect the accuracy of recalling specific details like dates and recounting experiences. Due to the nature of the topic and the small population size we were not able to sample for ethnicity in any way and we recognise that this may impact the cultural breadth and applicability of the data. The sample was predominantly women, but this is in line with care roles more broadly (AgeUK, 2019) and, for carers of people with MND, in particular those who chose more complex interventions, such as ventilation (Greenaway et al., 2015).

5. Conclusion

Across the four themes findings from this study have shown how the temporal landscape of dying is affected by the technological interface of home mechanical ventilation when people are dying of MND. Being present during the final moments of life can lead to profound moments of connection between the dying individual and their family. By optimising respiratory function, ventilation may offer a bridge, allowing patients and their families to navigate the dying process with a greater sense of control and reduced physical distress.

However, ventilator technology, while playing a crucial role in sustaining life, can also contribute to a complex dynamic where the realities of death may be obscured or mediated. This was shown to challenge participant's understanding and experience of the time of death and posed ethical, emotional, and existential considerations. This study has shown that dependence on the use of assisted breathing technology, and its planned removal, grants the time to plan active goodbyes. In this way the experiences of people who have been bereaved by the death of someone with MND are unusual and contribute to our understanding of the impact of time at the end-of-life, as well as increasing knowledge about the impact which technology may have on the end-of-life.

Ethical approval

Ethical approval for the study was granted by the London-Dulwich Research Ethics Committee for the NHS Health Research Authority in April 2021 (REC ref: 21/PR/0252).

CRediT authorship contribution statement

Glenys Caswell: Writing – review & editing, Writing – original draft, Formal analysis. **Eleanor Wilson:** Writing – review & editing, Writing – original draft, Project administration, Methodology, Investigation, Funding acquisition, Formal analysis, Data curation, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data availability

The authors do not have permission to share data.

Acknowledgements

the research on which this paper is based was funded by a University of Nottingham Anne McLaren Research Fellowship.

References

Adam, B., 2004. Time. Polity Press.

AgeUK, 2019. Breaking point: the social care burden on women. AgeUK. https://www.ageuk.org.uk/our-impact/campaigning/care-in-crisis/breaking-point-report/.

Anderson-Shaw, L.K., Zar, F.A., 2020. COVID-19, moral conflict, distress, and dying alone. Bioethical Inquiry. https://doi.org/10.1007/s11673-020-10040-9.

Association for Palliative Medicine of Great Britain and Ireland (AMP), 2015. Fareham, Association for Palliative Medicine of Great Britain and Ireland (AMP).

Bazeley, P., 2021. Qualitative Data Analysis: Practical Strategies. SAGE, London.

Bourke, S.C., Tomlinson, M., Williams, T.L., Bullock, R.E., Shaw, P.J., Gibson, G.J., 2006. Effects of non-invasive ventilation on survival and quality of life in patients with amyotrophic lateral sclerosis: a randomised controlled trial. Lancet Neurol. 5 (2), 140–147.

Caswell, G., 2022. *Dying alone: challenging assumptions*. Palgrave Macmillan, London.

Caswell, G., 2024. *Time of death: a sociological exploration*. Emerald, Bingley.

Caswell, G., Wilson, E., Turner, N., Pollock, K., 2022. It's not like in the films': bereaved people's experiences of the deathbed vigil. OMEGA - Journal of Death and Dying. Online first. https://doi.org/10.1177/00302228221133413.

Colquhoun-Flannery, E., Goodwin, D., Walshe, C., 2024. How clinicians recognise people who are dying: an integrative review. Int. J. Nurs. Stud. 151 https://doi.org/10.1016/j.ijnurstu.2023.104666.

Dekker, N., 2022. Anticipatory grief in dementia: an ethnographic study of loss and connection. Cult. Med. Psychiatr. 47, 701–721. https://doi.org/10.1007/s11013-022-09792-3.

Donnelly, S., Battley, J., 2010. Relatives' experiences of the moment of death in a tertiary referral hospital. Mortality 15 (1), 81–100. https://doi.org/10.1080/13576270903537641.

Dreyer, P., Felding, M., Sønderskov Klitnæs, C., Kirkegård Lorenzen, C., 2012. Withdrawal of invasive home mechanical ventilation in patients with advanced amyotrophic lateral sclerosis: ten years of Danish experience. J. Palliat. Med. 15 (2), 205–209.

Efstathiou, N., Vanderspank-Wright, B., Vandyk, A., Al-Janabi, M., Daham, Z., Sarti, A., et al., 2020. Terminal withdrawal of mechanical ventilation in adult intensive care units: a systematic review and narrative synthesis of perceptions, experiences and practices. Palliative medicine 34 (9), 1140–1164.

Faull, C., Oliver, D., 2016. Withdrawal of ventilation at the request of a patient with motor neurone disease: guidance for professionals. BMJ Support. Palliat. Care 6 (2), 144–146.https://doi.org/10.1136/bmjspcare-2019-002170.

Faull, C., Rowe Haynes, C., Oliver, D., 2014. Issues for palliative medicine doctors surrounding the withdrawal of non-invasive ventilation at the request of a patient with motor neurone disease: a scoping study. BMJ Support. Palliat. Care 4, 43–49.

Faull, C., Wenzel, D., 2020. Mechanical ventilation withdrawal in motor neuron disease: an evaluation of practice. BMJ Support. Palliat. Care: bmjspcare-2019-002170.

General Medcial Council, 2010. Treatment and care towards the end of life: good practice in decision making. https://www.gmc-uk.org/ethical-guidance/ethical-guidance-for-doctors/treatment-and-care-towards-the-end-of-life/guidance.

Greenaway, L.P., Martin, N.H., Lawrence, V., Janssen, A., Al-Chalabi, A., Leigh, P.N., et al., 2015. Accepting or declining non-invasive ventilation or gastrostomy in amyotrophic lateral sclerosis: patients' perspectives. J. Neurol. 262 (4), 1002–1013. https://doi.org/10.1007/s00415-015-7665-z.

Hamer, J., 2021. *Why it's OK to feel relieved when someone dies*. Marie Curie. https://www.mariecurie.org.uk/talkabout/articles/relieved-when-someone-dies/313426.

Jackson, K., Bazeley, P., 2019. Qualitative Data Analysis with NVivo. SAGE, London.

Kettemann, D., Funke, A., Maier, A., Rosseau, S., Meyer, R., Spittel, S., Munch, C., Meyer, T., 2017. Clinical characteristics and course of dying in patients with amyotrophic lateral sclerosis withdrawing from long-term ventilation. Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration 18 (1–2), 53–59.

Kitzinger, C., Kitzinger, J., 2014a. 'This in-between': how families talk about death in relation to severe brain injury and disorders of consciousness. In: Van Brussel, L., Carpentier, N. (Eds.), The Social Construction of Death. Palgrave MacMillan, pp. 239–258.

Kitzinger, C., Kitzinger, J., 2014b. Withdrawing artificial nutrition and hydration from minimally conscious and vegetative patients: family perspectives. J. Med. Ethics 41, 157–160. https://doi.org/10.1136/medethics-2013-101799.

Kühnlein, P., Kübler, A., Raubold, S., Worrell, M., Kurt, A., Gdynia, H.-J., Sperfeld, A.-D., Ludolph, A.C., 2008. Palliative care and circumstances of dying in German ALS patients using non-invasive ventilation. Amyotroph Lateral Scler. : official publication of the World Federation of Neurology Research Group on Motor Neuron Diseases 9 (2), 91–98.

LeBon, B., Fisher, S., 2011. Case report: maintaining and withdrawing long-term invasive ventilation in a patient with MND/ALS in a home setting. Palliat. Med. 25 (3), 262–265.

Leigh, P.N., Abrahams, S., Al-Chalabi, A., Ampong, M.-A., Goldstein, L.H., Johnson, J., et al., 2003. The management of motor neurone disease. J. Neurol. Neurosurg. Psychiatr. 74 (Suppl. 4), iv32–iv47. https://doi.org/10.1136/jnnp.74.suppl_4.iv32.

Masrori, P., Van Damme, P., 2020. Amyotrophic lateral sclerosis: a clinical review. Eur. J. Neurol. 27 (10), 1918–1929.

Mental Capacity Act, 2005. Mental capacity Act (england and wales). https://www.legislation.gov.uk/ukpga/2005/9/contents,Legislation.gov.uk.

Mercadante, S., Al-Husinat, L.i., 2023. Palliative care in amyotrophic lateral sclerosis. J. Pain Symptom Manag.E485-E499.

Messer, B., Armstrong, A., Doris, T., Williams, T., 2020. Requested withdrawal of mechanical ventilation in six patients with motor neuron disease. BMJ Support. Palliat. Care bmjspcare-2017-001464.https://doi.org/10.1136/bmjspcare-2017-001464.

MND Association, 2022. Living with motor nuerone disease. Northampton, MND Association 1–9.

Motor Neurone Disease Association, 2022. Ventilation for Motor Neurone Disease -8B, vol. 12. https://dbsy278t81889.cloudfront.net/app/uploads/2017/05/19135932/08b-ventilation-for-motor-neurone-disease.pdf.

Natural Death Centre (nd). Family led & DIY funerals. http://www.naturaldeath.org.uk/index.php?page=diy-funerals.

O'Brien, D., Stavroulakis, T., Baxter, S., Norman, P., Bianchi, S., Elliott, M., Johnson, M., Clowes, M., Garcia-Sánchez, A., Hobson, E., McDermott, C., 2019. The optimisation of noninvasive ventilation in amyotrophic lateral sclerosis: a systematic review. Eur. Respir. J. 54 (3), 1900261.

Oliver, D., 2004. Ventilation in motor neuron disease: difficult decisions in difficult circumstances. Amyotroph Lateral Scler. 5 (1), 6–8.

Palmer, J., Kathiresan, B., Messer, B., Ramsay, M., 2020a. Does tracheostomy ventilation in MND in the UK lead to improved survival? ERJ Open Research 6 (Suppl. 4), 1.

Palmer, J., Messer, B., Ramsay, M., 2020b. Tracheostomy ventilation in motor neurone disease: a snapshot of UK practice. Amyotrophic Lateral Sclerosis and Frontotemporal Degeneration 6 (52).

Phelps, K., Regen, E., McDermott, C., Oliver, D., Faull, C.. Withdrawal of assisted ventilation at the patient's request in MND: a retrospective exploration of the ethical and legal issues concerning relatives, nurses and allied health care professionals. https://doi.org/10.1101/2022.03.14.22271768.

Phelps, K., Regen, E., Oliver, D., McDermott, C., Faull, C., 2017. Withdrawal of ventilation at the patient's request in MND: a retrospective exploration of the ethical and legal issues that have arisen for doctors in the UK. BMJ Support. Palliat. Care 7 (2), 189–196.

Seale, C., 1998. Constructing Death: the Sociology of Dying and Bereavement. Cambridge University Press.

Sellen, K., McGovern, M., MacGregor, E., Halleran, L., Ly, L., 2021. The *Time Moving* exhibit: exploring perceptions of time in end-of-life experiences. Health Expect. https://doi.org/10.1111/hex.13379.

Timmermans, S., 2005. Death brokering: constructing culturally appropriate deaths. Sociol. Health Illness 27 (7), 993–1013. https://doi.org/10.1111/j.1467-9566.2005.00467.x.

Timmons, S., Crosbie, B., Harrison-Paul, R., 2010. Displacement of death in public space by lay people using the automated external defibrillator. Health Place 16, 365–370. https://doi.org/10.1016/j.healthplace.2009.11.008.

Wilson, E., Lee, J.-S., Wenzel, D., Faull, C., 2022. The Use of Mechanical Ventilation Support at the End of Life in Motor Neurone Disease/Amyotrophic Lateral Sclerosis: A Scoping Review. Brain Sciences 12 (9), 1162.

Wilson, E., Turner, N., 2024. Expanding Qualitative Interviewing for Studies Involving Adults With Different Communication Needs: Reflections on Research With People Living With Motor Neurone Disease. International Journal of Qualitative Methods 23, 16094069241251537. https://doi.org/10.1177/1609406924125153.

Wilson, E., Palmer, J., Armstrong, A., Messer, B., Presswood, E., Faull, C., 2024. 2024/05/02). End of life decision making when home mechanical ventilation is used to sustain breathing in Motor Neurone Disease: patient and family perspectives. BMC Palliative Care 23 (1), 115. https://doi.org/10.1186/s12904-024-01443-1.