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Unstable terminality: negotiating the meaning of chronicity and terminality in motor neurone disease

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Abstract   This paper explores the meaning of chronicity and terminality in motor neurone disease (MND), also known as amyotrophic lateral sclerosis (ALS). There is no known cause or cure for MND, and expected survival is 2–5 years, but several interventions may improve or prolong life. This study draws on qualitative interview data with health professionals in hospitals and primary care, and family carers, in Norway. The actors emphasised chronic and terminal aspects in subtly different ways along the entire illness trajectory, also when recounting the trajectory in retrospect. As a consequence of improved health services and medical technology the distinction between chronicity and terminality has become more vague and sometimes ambiguous. We suggest the concept unstable terminality to describe this ambiguity. While MND is a fatal diagnosis; it may be contested, as contingencies and interventions create an indefinite time scope. The instability creates challenges for primary care which is dependent on prognostic information to organise their effort; hospitals tackle the instability by pre-scheduled consultations allowing for avoidance of an explicit prognosis. Some carers experienced what we understand as a disruption within the disruption, living with chronic and terminal illness simultaneously, which made the limbo phase more challenging to overcome.

Keywords: unstable terminality, motor neurone disease, limbo, prognosis, ambiguity

Introduction

In this paper we explore the meaning of two common concepts in serious illness; chronicity and terminality, understood as an indefinite or definite closure of an illness trajectory. As a consequence of improved health services and medical technology the distinction between the concepts has become more vague and sometimes ambiguous. The particular case to illustrate this is motor neurone disease (MND), also known as amyotrophic lateral sclerosis (ALS), which several researchers have noted to lie beyond chronic or terminal illness (Brown 2003, Locock *et al.* 2009). This paper conceptualises MND as a phenomenon shifting between

chronicity and terminality, and suggests the term 'unstable terminality' to capture this ambiguity.

We rely mainly on interviews with family carers and health professionals, but unstable terminality is not exclusive to any perspective. The concept is an attempt to summarise the at times fragmented and ambiguous categorisations of the condition. The unstable meaning of MND is closely related to the illness trajectory, which we, in line with Strauss (Corbin and Strauss 1988, Strauss *et al.* 1985), view as not only the physiological unfolding of the illness, but the total organisation of work accompanying it. The trajectory is shaped by the overall framework of the welfare state, health services and available medical technology, but also family caregivers, persons with MND, paid carers, health professionals and the illness as physical entity. Illness trajectories have a negotiated or interactional character; so too unstable terminality and the meaning of MND.

Background

Chronicity and terminality
The World Health Organisation (WHO) defines chronic conditions as 'health problems that persist across time and require some degree of health care management' (WHO 2002a: 6). Diabetes and chronic obstructive pulmonary disease (COPD) are typical examples of chronic conditions that cannot be fully cured, but to some extent can be controlled and accommodated into the life of the chronically ill. Chronic conditions are frequently labelled as 'long-term', but the time span is seldom specified in formal definitions of chronicity.

Terminal conditions are considered incurable, and expected survival is usually less than 3–6 months. Key features of palliative care for terminal conditions are to affirm life, and neither to hasten nor postpone death. Palliative care should 'be applied as early as possible in the course of any chronic, ultimately fatal illness' (WHO 2002b: 83), but the document does not specify life expectancy.

There is a vague boundary between chronicity and terminality that invites a negotiation of meaning. A major difference between these concepts relates to distinguishable value rationalities. In a chronic case it makes sense to establish sustainable routines, and adapt to the current situation in a way which allows for a better, or at least some, future. Typical chronicity is on display in *The Logic of Care* (Mol 2008), showing various actors' work to manage diabetes. By closely monitoring blood sugar levels, a person with diabetes may stabilise the condition, and the work allows for a somewhat predictable and desirable future. In a terminal case, where the future is often predictable with unwelcome clarity and undesirability, managing the illness will involve attempts to create meaning in a short-term perspective. Hence, prognosis establishes an essential difference between chronicity and terminality; a tension between an indefinite or definitive closure of the trajectory. If the person with diabetes also has a fatal brain tumour, one may reconsider the rationality of meticulous management of blood sugar levels.

The medical literature
In the medical literature MND is described as an incurable and fatal neurodegenerative disease. The loss of motor neurones leads to progressive muscle weakness and death. Considerable progress has been made in understanding the underlying biological processes, but there is still no known cause, except for a 10–15 per cent proportion having a familial version (among recent clinical reviews are Kiernan *et al.* 2011; Andersen *et al.* 2012). MND is relatively uncommon, with an annual incidence of roughly two in 100,000. Life expectancy is about 2–5 years, but the individual variation is large, and a small proportion may live for a decade or more.

Interventions that replace vital bodily functions may prolong and improve quality of life, such as invasive and non-invasive breathing aids, often referred to as long-term mechanical ventilation (LTMV), as well as nutritional support through percutaneous endoscopic gastrostomy (PEG). In some cases, persons with MND develop signs of frontotemporal dementia, indicating that not only motor neurons are affected. While initial symptoms may be mild, mundane activities such as dressing, eating or visiting the toilet become increasingly difficult without help as the disease progresses. Importantly, the loss of function must be compensated for by labour or technology, calling for complex and substantial amounts of physical work. As the diagnosis is fatal, emotional work is essential. There is a potential for almost unlimited care, and persons with advanced MND are in need of round-the-clock care. The toll of MND care is usually beyond the capacity of a single individual.

The Norwegian healthcare system

There are three main arenas in which chronic and terminal conditions in Norway are managed: the hospital, primary healthcare in the community, and the private home (Lerum and Frich 2012). Hospital services and primary care are subsidised at the point of delivery due to the tax-based financing of the healthcare system. A person with MND, for example, is entitled to paid care and technical equipment, with few direct financial costs. The most knowledge-intensive tasks are performed at the hospital, such as establishing and communicating the diagnosis, introduction of PEG, and breathing aids. The municipalities are responsible for providing care and health services in private and nursing homes. A precinct usually covers a demographic area with none or a few persons with MND.

Theoretical framework

This article draws on theoretical concepts such as illness trajectories (Corbin and Strauss 1988, Strauss *et al.* 1985), but also classification (Bowker and Star 2000), prognosis (Christakis 1999), and disruptive experiences (Bury 1982, Becker 1997).

Bowker and Star (2000) underscore that humans have a tendency to impose order on the world through categories and classification systems. Classifications enable people to make sense of what goes on, and may also serve to coordinate action. Nevertheless, people routinely ignore classification principles, acknowledging that social practice is more complex than classification systems may allow for, or there may be disagreement or ambivalence about how to classify an ambiguous phenomenon. Local adaption and modification of abstract categories are necessary to fit classification systems with everyday realities (Bowker and Star 2000).

Bowker and Star (2000) borrow the concept of 'social worlds' from Strauss (1993) in order to handle the discrepancy between categories and practice. A social world, or community of practice, is a unit of analysis which cuts across formal organisations and institutions. In our context, there are three social subworlds in MND care: the hospital, primary care and the private home where persons live with MND. To become a member of any of these social subworlds it is necessary to learn the respective classifications developed locally. Within each of the social subworlds categories are naturalised, and as categories are used again and again 'you forget the strange and contingent nature of its categories seen from the outside' (Bowker and Star 2000: 294).

We argue that MND may be perceived as an ambiguous phenomenon, and that it is not always obvious whether it should be classified as chronic or terminal. Moreover, categorisations in this regard may not only differ between subworlds, but may also have an ambiguous character within them. Shifting categorisations make a substantial difference in the negotiations

among respective actors shaping a trajectory; one needs to take into account that the disease may be an unstable category. The prognosis represents an important tool in such negotiations.

Christakis (1999) underlines that prognostication is a moral act in the sense that it not only describes, but to some extent shapes the future. Hence, 'the patient's request for a prognosis is a query not only about what the doctor thinks will happen, but also about what actions the doctor [and others] can take to avoid, assure, or modify the predicted outcome' (Christakis 1999: 191). Prognosis influences people's expectations of illness trajectories, and may also cause frustration if health professionals do not agree. Christakis documents that doctors not only find prognostication difficult, but may also be unwilling to give a prognosis. It is unpleasant for doctors to convey bad news, and a prognosis may give rise to conflict in the relationship between doctor and patient. A poor prognosis may deprive the person living with MND of hope. Furthermore, if the prognosis turns out to be wrong, the mistake is not only embarrassing, revealing a doctor's lack of insight, but also threatening to the profession's legitimacy. Even if the prognostic outlook is poor, people may still ask what the future will bring for practical reasons, or to gain a sense of control and hope. When handling prognostic paradoxes and the uncomfortable position of being responsible for predictions and guidance, physicians tend to be overly optimistic and make active use of uncertainty and vagueness (Christakis 1999). Prognosis plays a key role in shaping the trajectory as it attempts to provide time frames, and legitimises the use of categories such as chronicity and terminality. Moreover, prognosis is important for individuals' sense-making of the disruption, and the following limbo, that illness may represent.

Disruption alludes to something being thrown into disorder or broken apart, and the term has been used to characterise illness experiences (Bury 1982). The concept is a part of a sociological tradition with a broader understanding of the impact of illness, beyond the strictly biomedical (see Bury 1982, Charmaz 1983, Williams 1984).

Disruption has been used more generally to explore life events; for instance, biographical disruption may be used to describe and understand informal carers' experiences of caring for an older family member (Adamson and Donovan 2005). The term reflects a cultural assumption of continuity or order, and there may be stages in life where such continuity is not assumed (Pound *et al.* 1998, Faircloth *et al.* 2004). According to Becker (1997) 'plot' is an important concept for managing disruptive events. Faced with a new and threatening situation, the person may need to assess what to do in a short-term and long-term perspective. Before decisions have been made, the individual is in a liminal phase or limbo; a state of neither subscribing to an old role nor being recast in a new. To escape limbo the individual has to mobilise resources, for instance of a personal, social, medical or economic nature. Available cultural categories and an understanding of the prognosis are such resources, which informs notions about typical roles and behaviour in the face of chronic and terminal illness. Limbo may be managed in many different ways, Becker (1997) points out that '[p]eople whose lives are disrupted are often thrust into a limbo-like state for long periods of time, sometimes even for life'. The basic plot, then, is that of disruption, liminality or limbo, and a reconnection to either an altered or familiar situation or role.

Methods

In 2011–2012 the first author (SVL) interviewed carers, persons with MND and health professionals in hospitals and primary care (Table 1). The sample centres on family caregivers and health professionals, as the study had a focus on their role and care coordination. A total of 53 interviews were conducted with 65 participants. The sample consists of eight bereaved carers,

17 active carers, five persons with MND, 18 health professionals in primary care and 17 hospital staff, which are specified below.

Sample and recruitment
We recruited from three sites; each site consisted of a hospital, with the corresponding primary care, and people living with MND. By recruiting from three different hospitals we sought insight into general features of MND trajectories, independent of local particularities.

With regard to those living with MND, the bereaved carers were interviewed in an early phase of the project to obtain a sense of the trajectory from beginning to end. The remaining 16 of those interviews comprise 22 participants, since some carers wanted to bring the person with MND or family members along for the interview. Several participants had lived with MND for years, while others had been diagnosed a few months earlier. Most interviews took place in the carers' homes, some in public places as chosen by the participants.

In accordance with ethical guidelines in Norway, those living with MND were approached by health professionals they already knew. The health professionals, MND coordinators in hospitals, briefly explained the purpose of the study and asked for permission to pass on contact information to a researcher (SVL), who gave further information and handled written informed consent.

The hospital staff included were the same persons providing hospital services for those interviewed who lived with the condition. Using health professionals to recruit participants creates a possibility for sample selection bias. We used temporal and pragmatic sampling criteria. In one of the hospitals all persons with MND coming for routine consultation on a particular day were asked to participate. In another hospital, there was a new coordinator wanting to introduce herself to all persons enlisted with MND, who presented the research project in the same telephone call. In these cases the researcher selected participants to maximise variation with regard to gender, family situation and place of residence in a rural or urban area. However, health professionals were discretionally involved in shaping the sample in one case, where the MND coordinator argued that the potential participant 'was in too poor shape to participate', which was respected.

In primary care, general enquiries were made asking if those concerned were currently caring for any persons with MND, or if they had prior experience of this. Contrary to the hospital staff, the primary care sample was recruited independently of whether or not they provided care to the same people interviewed who lived with the condition. Interviews were conducted with persons, often nurses, with a coordinating role. Persons with strictly paid carer experience were excluded. One interview at a hospice unit comprised four participants, as the entire staff present wanted to participate. These interviews cover both urban and rural areas, precincts with extensive and no prior MND experience, as well as seasoned and novice coordinators.

Table 1 *Overview of interviews*

	Pre-project	*Hospital A*	*Hospital B*	*Hospital C*	*Total*
Living with MND (informal carers and persons with MND)	8	5	5	5	23
Primary care staff		5	5	5	15
Hospital staff		5	5	5	15
Total	8	15	15	15	53

At two hospitals the role of MND coordinator was shared between two nurses, who preferred to be interviewed together. One important difference among the hospitals related to internal organisation of MND care. Two hospitals had multidisciplinary MND teams, where active members were interviewed. One hospital dealt with MND in the general neurological department, where chain referral was used to identify participants most involved. Health professionals with varying MND experience and professional background were interviewed. All hospitals were located in larger Norwegian cities; some were university hospitals and some provided care for rural areas.

Data collection
Most interviews lasted between one and two hours, and were transcribed verbatim. All participants were encouraged to elaborate on their understanding of MND and their experiences with MND care. The interviews with bereaved and current carers had a narrative approach, and the participants were encouraged to talk about their MND experiences in their own words, for as long as they wanted, starting from the first time they noticed anything 'strange'. The latter part of the interviews had a thematic approach ensuring that pre-defined topics such as the role of the carer, of primary care and of the hospital were reflected upon.

The interviews with health professionals focused on specific topics, for example, their perception of a typical person with MND, characteristics of good MND care, typical relation to primary care or the home, and what tasks were considered relevant for particular professions.

Analysis
As data collection proceeded, some authors (SVL, KNS, JCF) routinely met to discuss transcripts and emerging themes. Analytical memos were circulated focusing on different aspects of the study. One of the themes we decided to pursue further was: 'What is MND?' There were empirical and theoretical reasons for this. Empirically we observed that MND represented different things for different actors, and that the entity could change over time. Our perspective was the entire care chain and carers in particular, and the literature on individual MND experiences was essential in this analytic strategy. The prognosis could be perceived with 'unwelcome clarity' or as 'helpful practical guidance' (Locock *et al*. 2012), and such ambivalence was also found to be central in MND and peer support (Locock and Brown 2010). Individuals with MND could experience tensions between support and distress when sharing illness experiences (Mazanderani *et al*. 2012). Brown (2003) noted that the condition seemed to straddle a classification as either chronic or terminal. Following this line of thinking Locock *et al*. (2009) coined the term 'biographical abruption', to set MND apart from 'normal' chronic illness. Abruption suggests that MND represents a breaking off of life, 'the sense of living death, a life over but not over' (Locock *et al*. 2009). The study also identified typical chronic illness features of MND experiences, such as biographical disruption and even biographical repair or flow. The literature on individual experiences spurred our interest towards the nature of the underlying phenomenon generating such shifting responses, and how health services and carers related to this.

On this note, *The Body Multiple* (Mol 2002) sensitised us to the multiplicity of presumably fixed disease entities. The analytic work led us to an array of categories such as MND as existential challenge, MND as functional challenge, MND as economic challenge, MND as acute, MND as chronic, MND as terminal, and so on. However, in our eyes the most significant discovery of our study did not concern the differences between these categories but the unstable relationship between them, in particular between 'chronic' and 'terminal'. To further focus the analysis, sociological theory relating to categorisation (Bowker and Star 2000), prognosis (Christakis 1999), and disruption (Becker 1997) was incorporated, which led to presenting

'unstable terminality' as a concept to illustrate choices and dilemmas in illness trajectories. In the subsequent phase an experienced MND clinician (TH) joined the study team to contribute to the analysis, and in the writing phase. All quotes have been translated from Norwegian to English by the authors.

Findings

The trajectory

A key issue in unstable terminality was a tension between declining functionality and the meaning ascribed to this, which one coordinative nurse struggled with:

> They [persons with MND] have a disease which can't be cured, and that's why you call it palliative. But still, it isn't like that, as I'm saying, they don't have those kinds of problems. They are perhaps well functioning, and for a long time in a job. Then they start to loose more and more functionality, and need more and more help. When are you to call it a palliative phase? Are they that all the time? Are they becoming palliative after a while? It's all a bit vague. (MND coordinator in Hospital A)

We view 'palliative' to categorise the disease as terminal, but the nurse found it difficult to apply this category. With regard to functionality we found MND to have three phases (Figure 1), which roughly described the loss of functionality, but whether each phase represented chronicity or terminality was not given.

Phase 1 is the time around when diagnosis is given. In this period the symptoms are often relatively mild. However, resonating with our own material, Brown (2003) described how receiving the news of the diagnosis often represented an 'existential shock'. After some time, in phase 2, a common trait is an ongoing change and decline. Even though the underlying progression is continuous and smooth, the effects are often perceived as incremental. Becker (1997) notes how constantly shifting bodily experiences undermine the efforts to repair disruption. Importantly, when adapting to changes and repairing disruption, negotiations about chronicity and terminality are essential. When functionality reaches an exceedingly low level, in phase 3, the ongoing change aspect diminishes. In this phase technology may replace vital bodily functions. The choice of whether or not to introduce such technology strongly influences the actors' use of categories such as chronic or terminal.
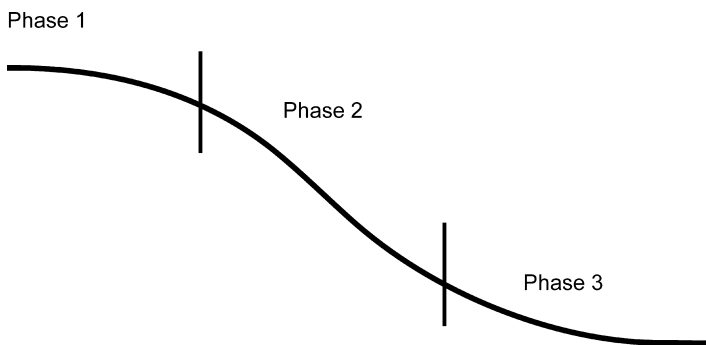

Figure 1 *MND trajectory*

The somewhat fixed properties of the overall MND trajectory are the three phases, and the shape of the curve. The time span of the trajectory and the phases may vary from a few months to several years, and making a precise prognosis is challenging even for experienced doctors. There are several versions of MND, and some have a steep downward curve and a clear terminal unfolding. On the other hand, very slowly progressing variations may be clear-cut chronic illness. Our claim is that between the extreme trajectories there is a median pace to the progression, partly determined by the use of technology, which challenges the use of categories such as chronic and terminal.

The social subworlds

We view hospitals, primary care and the homes of carers and persons with MND as social subworlds, membership of which allows for learning the local meaning of MND. The analysis demonstrates how the participants made continual assessments of whether a chronic or terminal framing should be used. The actors within the respective social subworlds emphasise these aspects in subtly different ways, making for a potentially fragmented understanding of the challenge at hand. Our main finding, then, is that in this context the meaning of MND is unstable, both in time and space. Findings from hospitals, primary care and private homes are presented in the following.

Instability and prognosis in the hospital

The MND coordinators 'I' and 'K' from hospital C told a story about breathing aids that demonstrates the instability of terminality. The story described a person with MND and a carer who perceived MND as a terminal condition, and who had agreed on advance directives not to use invasive respiratory technology or to resuscitate in the case of acute respiratory emergency.

I:　　　　　　[T]he MND patient developed respiratory failure in the ambulance. The patient and the carer had very clearly rejected long-term mechanical ventilation [LTMV]. The wife said to the ambulance driver: 'Don't stop, we are going in [to the hospital], we are not stopping to resuscitate'. And they [the health professionals in the ambulance] listened to her and didn't stop. She felt such respect for that, precisely because they had been through that [advance directives]. The patient used a mask, but did not want a hole in the throat. He didn't want it. Because, she said later, it had been very important for her that they had listened to her decision. Later the TV documentary was broadcast [about an individual with MND living on LTMV], and then she called me, and said 'Was it the right thing to do?'

S:　　　　　　Mmm, mmm

K:　　　　　　I see, then she started to . . . [question whether to resuscitate]. Yes.

I:　　　　　　Yes, you know. After . . .

K:　　　　　　Because I was involved in admitting [the patient when he got to the hospital] . . .

I:　　　　　　Yes, I know . . . down in the emergency room . . .

K:　　　　　　Yes, exactly. So she started to think about it then . . .

I:　　　　　　Yes, then she started to think about it. Mmm . . .

K:　　　　　　But it was the right thing to do.

I:　　　　　　Yes, it was absolutely right.

K:　　　　　　Absolutely.

The person with MND had chosen not to live with LTMV. However, this decision was not accepted without question, which happened several times. First, to arrive at the advance directives was no easy matter to begin with. Second, the decision not to resuscitate had to be repeated to the health professionals in the ambulance, which was respected. Third, the decision was challenged retrospectively by a documentary about a person living on LTMV. Death did not settle the category, and as the bereaved carer struggled, she sought help and support from health professionals at the hospital. From the carer's perspective, the terminality was not taken for granted, and was even challenged. Negotiation of meaning, as in this case, illustrates that the terminal category can be unstable, and may remain uncertain in retrospect.

One may view the story from the perspective of the storytellers, the health professionals, who needed to reassure each other about the decision that was made. Such interaction is a consequence of the unstable terminal category. Also, in less dramatic circumstances, the hospital staff was explicitly concerned with the instability of MND:

> This is the peculiar thing about MND – that they [the persons with MND and carers] are told that it goes quickly. 'We [the person with MND and the carer] are going to cope, because this goes fast'. And then they don't know. We have had a lot of discussion about that, with some of them: 'If I just knew how long it was, then I would be able to cope'. You know. 'If I had known there were three months left, or three years, that would have been two very different things'. And it is not always that we [the health professionals] can tell them whether it will be three months or three years. [. . .] I remember, [. . .] one carer who was so tired, but then it was like: 'Maybe it is the last summer we have together, I just have to hold it together a little longer'. (MND coordinator in hospital A)

All hospitals made pre-scheduled consultations, usually every three months. Even though the coordinative nurses organised this, the pre-scheduling was to some extent taken for granted and neither the hospital staff nor the person with MND had to deliberately discuss when to have the next consultation. This was an implicit form of prognostication, allowing the health professionals to avoid addressing the prognosis explicitly. Hence, health professionals organised their way out of the prognostic dilemma by 'being one step ahead', simultaneously avoiding the need to explicitly address the future. This gave some flexibility in conveying the explicit prognosis to the particular individual with MND. There were at least two types of explicit prognostication: one type was used among health professionals, while the other was used for persons with MND and their carers. One of the neurologists commented on prognostication as follows:

> At the same time it is difficult, because . . . some people argue that the patient should know everything, and get all the information at once, you know, by painting a picture of everything at once. But . . . we do that in a way. They [the persons with MND and their carers] learn that this is a disease with no cure and that it is lethal. But I don't think you should beat them to the ground with that every time, that this is terrible. We have to try to . . . when they have no hope of getting well, then at least we have to try to help support them so that they manage to get the most out of the time they have left. (Neurologist from hospital B)

This quote reflects a distinction between the internal and the communicated prognosis; in this case there was a professional understanding of MND as terminal, which was couched in less direct terms when communicating the prognosis. When providing a prognosis to persons with MND and carers, health professionals were wary that this could be damaging. In general the

approach among hospital staff was that prognostic information had to be timed, a strategy easily accomplished within the pre-scheduling. This meant continual and cautious probing of whether or not the person with MND and their carer were 'ready', especially when discussing LTMV which was considered to be the most sensitive issue. Moreover, health professionals encouraged the person to 'live here and now', and stressed the uncertainty and vagueness of the future.

Health professionals underlined how the scheduled consultations helped them to become familiar with the person with MND and to build trust. Furthermore, they typically noted that 'we don't give this type of attention to any other diagnoses'. The arrangement of pre-scheduled consultations allowed for quite a lot of discretion, and large variation in practice. At one end of the spectrum a doctor stressed how measuring lung capacity at every consultation, to monitor the decline, was a form of 'torture' only to confirm the inevitable and expected progression. Another doctor underscored the importance of performing exactly the same measurements as a necessity. Doctors had very different approaches when weighing between uncertainty and control in preparing persons with MND and their carers for the future. The way care was organised provided considerable robustness to handle the instability of MND. A potential weakness, however, was the tendency to lean too heavily on the passive prognosis, and provide persons with MND and their carers with less information about the long-term outlook.

Prognostic dependency in primary health care

The primary healthcare services delivered paid care and technical aids to the homes of persons with MND. Typically, the paid carers needed the appropriate skills to handle technical equipment such as respiratory aids and computerised communication technology. Many such skills were MND-specific and personalised and paid carers had to receive specific training in each case. In order to procure the proper equipment in a timely manner, and to train personnel, the primary healthcare services needed a relatively accurate prognosis. Labelling the illness as chronic or terminal had a strong impact on resource allocation. This was a major institutional difference between primary health care and hospitals, as hospitals utilised the same personnel irrespective of how the illness unfolded:

> It has just escalated, it just becomes more and more [care work] . . . and someone [from primary care] was at the hospital and talked with them. And [the primary care officials] said: 'We are to do everything in our power for him [the person with MND] to remain and receive good care at home'. [. . .] then the view was that he [the person with MND] didn't have so much time left. [. . .] And now the entire problem changed. Suddenly, now he has got the mask, and he breathes. He even breathes without the mask, sometimes. (Primary care coordinating nurse associated with hospital C)

In this excerpt, the actors reclassified the illness from terminal to chronic. The terminal category had triggered the same system of care as that provided for people with terminal cancer, which involved a palliative team and substantial amounts of paid care, day and night. When the breathing mask was introduced, the illness shifted towards chronicity, making the care regimen of abundance difficult to sustain; the staffing costs were high and the specialist palliative personnel were needed elsewhere.

Phase 2, with rapidly changing needs, was particularly challenging for primary health care. A major concern was to be 'one step ahead', which for the coordinating nurse meant establishing contact and raising awareness about MND among the local occupational therapists, physical therapists, palliative nurses, computer communication specialists and several others. One typical challenge was that resources were usually provided retrospective to the care need.

Experienced coordinating nurses managed to bypass this formal obstacle by using prognosis, arguing for a rapidly declining functionality. Another difficult problem was that of prognostic uncertainty. There was a considerable chance that the person with MND would die, rendering for instance the hired carers unnecessary.

Managing expectations was frequently an important issue for primary care participants, especially those who had handled several MND cases. One such coordinating nurse summarised her experiences as follows:

> I think you have to be very open about what you [primary care] are able to offer, and what you cannot [stressing 'not'] offer. So that they [person with MND and carer] understand the perspective [of primary care], so that you don't make promises you can't keep. (Primary care coordinating nurse associated with hospital A)

Unfulfilled expectations were a frequent source of conflict among actors, especially regarding whether care should be delivered within a private home or an institution. The primary care services had a strong wish to avoid such conflicts. The instability between chronicity and terminality made them ambivalent towards making long-term plans. Discrepancies in prognostic outlook between those living with MND and primary care were quite common.

In addition to prognostic discrepancies, another issue was that of persons with MND and their carers refusing to discuss prognosis at all:

> Many [persons with MND and carers] struggle against technical aids and everything, and they feel that it is a pathologisation of it all. That is of course a challenge. To persuade them of the need to get help, before they are necessarily totally dependent on it. (Primary care coordinating nurse associated with hospital B)

Due to the individual variation in MND, primary care professionals were dependent on social interaction with the persons with MND and their carers to get a sense of the prognosis in that particular case. From a primary care perspective, it was not uncommon that the poor prognosis was feared and avoided by the persons with MND and their carers. The lack of an accurate prognosis and a clear overview of the situation meant that primary care professionals had less time for preparations, or made wrong preparations, often resulting in what in their view were suboptimal last-minute solutions. The ambiguity of MND made it difficult for primary health care professionals to plan, as they continuously had to make trade-offs relating to allocation of personnel.

Unstable terminality at home

Typically, in carers' accounts of the illness trajectory, MND was initially categorised as terminal. As the trajectory unfolded, the terminality, and the expectations that followed it, was contested. An additional chronic aspect of the illness trajectory became evident, but even if the time frame was uncertain, carers often suspected death might unexpectedly occur. Due to the ambiguous chronic-terminal situation, carers had to manage what appeared as a disruption within the disruption, living with chronicity and terminality simultaneously. While some carers managed to live with these disruptions, and to escape from the limbo phase, others had difficulties reconciling with the 'double' disruption, and remained in limbo.

We will elaborate on the ambiguity of MND as experienced by carers in the home setting, and present a case involving a dual-career middle-class couple. After the husband was diagnosed some years ago, the husband and wife were no longer in paid work and both received disability pension. In the interview, the wife summed up the last few years as follows:

> To start with, you have to accept that the person is going to die, and then you are supposed to manage the functional loss [. . .] And I'm thinking, with the kind of strain we are having, I'm feeling that . . . I'm thinking that my body . . . becomes . . . I get a few reactions [from the body], which aren't too good. And it isn't that easy for others to help with that. [. . .] I'm thinking that I'm so extremely tired of being reasonable, with regard to sleep, and food, and exercise, and . . . to be reasonable, you know, just so that the body doesn't [break down] . . . 'Cause I'm feeling that I'm compromising on everything, I have had this stress in my body all the time, for several years now. I have had to take this into account to such an extreme degree. [. . .] It is very tiring. (Carer associated with hospital A)

The carer pointed out that the initial phase was characterised by disruption, shock and existential issues. MND was perceived and framed as a terminal condition, and she experienced disruption related to the rapid functional decline of her husband and changes in everyday life. Many of the participants, both carers and health professionals, underscored that MND develops fast. 'Galloping' was a metaphor participants used several times. Hence, they portrayed MND as a wild horse which was strong, fast and difficult to control. The above story challenges this galloping horse metaphor, as the carer experienced MND as enduring and relatively stable. Being faced with stable but vast amounts of care work, the challenge for the carer shifted from managing rapid existential and functional changes to maintaining routines, such as getting enough sleep, exercise and healthy food. As the terminal frame is contested, the carer experienced another disruption within the disruption; new meanings have to be negotiated and new approaches established. Even though the illness has lasted longer than first expected, thus gravitating towards chronicity, terminality remains. Several carers shared the experience that MND is both fast and slow, thus representing an ambiguous condition lying between terminality and chronicity.

A bereaved carer in her early sixties reflected on the last phase of her husband's MND trajectory. For the last few months of the husband's life she kept a diary, which she partly comments on and reads aloud in the interview:

> There was one [paid carer] who came at 11 p.m. and helped him to bed. And then he [the husband] got help, because of stomach ache, until 11.50 p.m. I have, evidently, written from 02.10 a.m., turning and twisting and moving and things like that. At 03.10 a.m., move the legs. At 04.00 a.m., scratching under the arm. At 04.10 a.m., turning and moving about. At 06.00 a.m., there was a cord that snapped, and a bit of this and that. And then he visited the bathroom at 06.30, and at 07.40 came the seven o'clock shift and did some cleaning and tidying. [. . .] That was what the nights were like. (Bereaved carer)

Towards the end of the illness trajectory wife experienced that the amount of care work escalated. Getting out of the apartment to shop for groceries became increasingly difficult, and she experienced a growing sense of isolation. Doing care work day and night is inevitably only possible on a short-term basis. Importantly, though the challenge did not disappear but endured, carers struggled to re-categorise the condition. The event expected to end the limbo was the death of the loved one, and neither the present nor the future represented a preferable solution. In this case, both of them viewed the condition as terminal and the carer's description of her late husband's experience resonated with biographical abruption. Consequentially, the notion that it would soon be over contributed to the decision to refrain from more paid care and other types of help.

Living in limbo could last for years, as illustrated by the story of a daughter caring for her father. The father was diagnosed more than ten years ago, and even though the situation at the

time of the interview was relatively stable, there had been several exacerbations leading to hospitalisation over the years. Still caring for her father, she looked back on the last ten years or so:

> I want to take care of dad. That is the best thing for my dad. I'm thinking about that time when he was very ill, when we almost lost him in 2000 something [does not recall exactly]. Then I just thought ahhh [sigh of relief], I am so glad that I sacrificed myself, in order to be together with dad. Because, then there was no more [if he died], you know. [...] It is such a good thing that I have had the opportunity to be together with him so much, you know. And then he survived. And now we get even more [time together]. [...] And I am thinking that I had better contribute or get to be with him as long as possible. (Carer associated with hospital B)

The carer had been hospitalised with exhaustion several times. In a terminal trajectory, the illness is not expected to last very long, allowing for a massive effort for a limited period of time. However, in a chronic case the indefinite time span calls for sustainable routines. The carer had tried for the entire illness trajectory to find some sort of permanent solution to the disruption. Several carers who felt stuck in limbo conveyed the experience of terminality and fear of being left alone.

A few carers did not feel they were stuck in limbo, for instance some spouses framed MND as a chronic condition and agreed on moving the person with MND to a nursing home. One example was a bereaved carer who continued to work throughout the illness trajectory. In contrast to most carers she expressed a clear sense of being prepared for how the illness would develop. Among the resources she underlined in repairing disruptions was her previous experience as a paid carer:

> I did picture to myself what I did in the house for that lady I tended to, back then. I was there from 08.00 to 16.00, cleaned for her, gave her food ... I was her arms and legs within her home, you know ... So I have thought that perhaps there was a meaning to it, that I was to [do that once again] ... You start thinking a bit like that. (Bereaved carer)

Besides the sense of being prepared, the carer's husband already had health problems and lived on disability benefit prior to the onset of MND. The deterioration in his health was therefore not surprising, although MND was an unexpected additional diagnosis, and a sense of biographical continuity may describe this situation.

Discussion

We have shown that MND continuously shifts between being understood as chronic and terminal along the entire illness trajectory, also when recounting the trajectory in retrospect. The meaning of MND seems fundamentally unstable. We suggest using the term 'unstable terminality' to denote the ambiguity of MND. Terminality implies a fundamental understanding of the illness as a definitive temporary challenge, as opposed to an indefinite time span. In this perspective, terminality may be perceived as a negotiated agreement on what the entity at hand represents. The terminality may be picked apart, attacked, contested and to some degree reversed. Partly, this is due to physiological contingencies. Additionally, technology may replace vital body functions, further contributing to an indefinite time span.

In our study the unstable terminality was handled in a variety of ways. The hospitals have established a system to manage MND with a strong implicit prognostic assumption of rapid decline. The approach is productive in the sense that it allows health professionals to avoid making a prognosis if they feel it necessary or uncomfortable. In comparison, the primary care services did not have a system of care independent of disease progression. They were dependent on explicit prognostic information, and more vulnerable to the unstable terminality. Some carers experienced what we understand as a disruption within the disruption, living with chronic and terminal illness simultaneously. This made the limbo phase more challenging to handle and overcome. This study raises the issue of whether prognostication was effectively utilised to handle the limbo phase.

Other terms could have been used to describe the phenomenon of unstable terminality. For instance 'frail' or 'fragile terminality' would highlight the settlement of the negotiations leading to a terminal categorisation. Other options considered were 'urgent chronicity', 'enduring acuteness', 'fluid terminality', 'shifting terminality' or 'chronic terminality'. However, we found unstable terminality to be the most descriptive term, as it underscores the potential shift away from a terminal categorisation, not just a prolongation of a terminal phase.

We would like to mention two concrete challenges posed by unstable terminality. First, unclear categorisation of chronicity and terminality may complicate access to hospice and palliative care (O'Brien *et al.* 2012). Second, health professionals are legally obligated to resuscitate people with chronic illness, but not necessarily those in a terminal phase. Unstable terminality makes the illness problematic to define in advance directives.

Unstable terminality expands current categories. Can this concept and its accompanying properties be generalisable to other diagnoses? A very specific aspect of MND is the possibility to replace vital body functions with technology, without any other prospect than further decline of muscle strength and increased need for care. In most other potentially lethal conditions, including cancer, declining vital functions can either not be replaced, or there is a fair chance of long-term remission or even cure. Thus, organ transplantation is usually not performed unless it is likely that the patient will recover to lead an independent life. A perhaps more comparable case is COPD, although slower progression and lack of pareses set it apart. Other neurological conditions, such as multiple sclerosis and Parkinson's disease, may also provide a degree of relevant comparison, although they are less acute and life-saving technology is rarely indicated.

Advances in medical technology which blur the boundary between terminal and chronic might make the concept of unstable terminality relevant to other conditions. The concept might be ahead of its time, as improved medical interventions allow people to survive with very severe conditions for an indefinite time. In this regard, unstable terminality may be understood as a consequence of modernity and progress. Such advances raise new challenges for care work, ethics and understanding illness experiences. The added complexity and increased care burden accompanying unstable terminality challenge the rationale behind some of the major trends in healthcare services today, such as moving complex and intensive tasks out of hospitals into the home, and general deskilling to deliver services more cheaply.

Unstable terminality could be further explored. Coordination of care relating to an ambiguous phenomenon complicates reliance on standardised clinical pathways, as it implicates the coordination of meaning, or what Bowker and Star (2000) label categorical work. Moreover, unstable terminality poses several challenges to the household of the person living with MND, for instance with regard to staffing there is often just one person, the carer. Another such theme is co-construction of illness narratives, and interaction between spouses. The issue of disruption within disruption and being 'stuck in limbo' could perhaps be further explored in the case of parents caring for (unstable) terminally ill children. As many of the consequences

of unstable terminality depend on how it is handled within the household, interaction between primary care and the home should be further explored.

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