Managing and caring for individuals with amyotrophic lateral sclerosis/motor neurone disease in Norway: a qualitative study

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Abstract

Amyotrophic laterals sclerosis (ALS), also known as motor neurone disease (MND), is a neurological condition with no known cause or cure. MND causes progressive paralysis and death. While life expectancy is only 2-5 years, there are several interventions available to improve and prolong life.

The overall aim of this study is to contribute to knowledge about conditions for collaboration between family caregivers and health professionals managing and caring for individuals with ALS/MND, and there are three specific aims: i) to explore the meaning of chronicity and terminality in ALS/MND, ii) to understand more about family caregivers' work and sense of responsibility by exploring family caregivers' accounts of caring for a family member with ALS/MND, iii) to explore what challenges health professionals encounter when managing ALS/MND in primary health care in Norway.

The study draws qualitative interviews with three groups: 1) Family caregivers and persons with MND living at home, 2) Health care professionals in primary health care, and 3) Health professionals in hospitals. The three groups comprise a total of 53 qualitative interviews with 65 participants. The sample consists of eight bereaved caregivers, 17 current caregivers, five persons with ALS/MND, 18 health professionals in primary health care and 17 hospital staff.

We found that the participants emphasised chronic and terminal aspects of the condition in subtly different ways along the entire trajectory, including when recounting the trajectory in retrospect. We suggest the concept *unstable terminality* to describe this ambiguity. While ALS/MND is a fatal diagnosis, it can be contested, and contingencies and interventions can create an indefinite timeline.

We found that caregivers were engaged in five lines of care work that were often parallel or closely interconnected: i) Immediate care work was work in physical proximity to the person in need of care to replace functionality and a provide sense of safety; ii) seeking information and clarity about the disease was work to handle information about the condition, prognostic outlook and ways to manage the situation; iii) managing competing obligations was work to

accommodate other obligations than those related to the condition; iv) maintaining normality was work to provide a sense of normality, such as remaining in paid employment or sustaining a social life; v) managing external resources and assistance was work to handle and incorporate nurse assistants, friends and family and assistive devices at home. Some of the lines of work could start before a diagnosis was given, and continue throughout the trajectory. With the potential to perform care work day and night, caregivers had to continuously balance their own and others' expectations without being overwhelmed by care work.

We found that ALS/MND was usually viewed as a case that called for strong collaboration between those at home and primary health care, exceptional effort within primary health care, and planning ahead. We present five challenges health professionals reported when managing MND: i) building relationships with those at home, ii) preventing caregiver burnout and breakdown, iii) providing tailored care, iv) ensuring good working conditions in patients' homes, v) recruiting and retaining qualified nurse assistants.

We found that ALS/MND has features of both chronicity and terminality. One condition for collaboration is for the actors to have a shared understanding of the condition. We found nuances in the lines of care work carried out by family caregivers in ALS/MND and that family caregivers perform a balancing act trying to meet their own and others' expectations while not becoming overwhelmed by work. A second condition for collaboration is for the actors to have a shared understanding and appreciation of care work. We found that the health professionals reported five challenges managing ALS/MND in primary health care. The condition for collaboration is to build capacity within primary health care having available competent personnel and ability to provide them with social support in the home setting.

Sammendrag [abstract in Norwegian]

Amyotrofisk lateral sklerose (ALS) er en uhelbredelig nevrologisk sykdom uten kjent årsak. Sykdommen fører til gradvise lammelser. Forventet overlevelse er 2-5 år, men det er en rekke tiltak tilgjengelig som kan forbedre og forlenge livet, utover dette.

Målet med studien er å bidra til vilkår for bedre samarbeid mellom pårørende og helsepersonell som håndterer og gir omsorg til personer med ALS. Studien har tre delmål: i) Å utforske ALS som kronisk og terminal sykdom. ii) Å forstå mer om pårørendes arbeid og ansvarsfølelse, i ALS. iii) Å utforske hvilke utfordringer helsepersonell i kommunen møter i håndteringa av ALS.

Studien baserer seg på intervjuer med tre grupper: 1) pårørende og personer med ALS som bor hjemme, 2) helsepersonell i kommunehelsetjenesten, 3) helsepersonell på sykehus. Disse tre gruppene utgjør til sammen 53 kvalitative intervjuer med 65 deltakere. Utvalget består av åtte etterlatte, 17 aktive pårørende, fem personer med ALS, 18 helsepersonell som jobber i kommunehelsetjenesten, og 17 helsepersonell som jobber på sykehus.

Vi fant at deltakerne vektla kroniske og terminale sider ved sykdommen på ulikt vis gjennom hele sykdomsforløpet, også når de fortalte om forløpet i ettertid. Vi foreslår begrepet *ustabil terminalitet* for å beskrive denne flertydigheten. ALS er en fatal diagnose, men denne terminaliteten kan utfordres av eventualiteter og intervensjoner som skaper en åpen tidshorisont.

Vi fant at pårørende inngår i fem typer omsorgsarbeid, som kunne pågå parallelt og være tett sammenfiltret: i) umiddelbart omsorgsarbeid var arbeid i fysisk nærhet til personen som trengte omsorg, for å erstatte funksjonalitet eller gi trygghet, ii) søke informasjon og klarhet om sykdommen var arbeid som handlet om å håndtere informasjon om sykdommen, prognose og måter å håndtere situasjonen på, iii) håndtere konkurrerende forpliktelser var arbeid som handlet om å flette inn andre forpliktelser enn de direkte relatert til sykdommen i hverdagen, iv) opprettholde normalitet var arbeid for å gi en følelse av normalitet, for eksempel å være i jobb eller opprettholde et sosialt liv, v) håndtering av eksterne ressurser og assistanse var arbeid for å inkorporere helsepersonell, venner, familie og hjelpemidler i hjemmet. Noen av disse typene

arbeid kunne starte før en diagnose var gitt, og vare forløpet gjennom. Det var et potensiale for å gjøre omsorgsarbeid dag og natt, og pårørende måtte balansere deres egne og andres forventninger uten å bli overveldet av omsorgsarbeid.

I kommunehelsetjenesten fant vi at ALS vanligvis ble sett på som et tilfelle som trengte sterkt samarbeid mellom dem hjemme og kommunehelsetjenesten, ekstraordinær innsats i kommunehelsetjenesten og å kunne planlegge i forkant. I arbeidet med å møte disse idealene fortalte helsepersonell i kommunehelsetjenesten fortalte om fem utfordringer. i) Å etablere en relasjon til de hjemme, ii) Å forebygge burnout og sammenbrudd hos pårørende, iii) Å tilpasse omsorgstilbudet, iv) Sikre godt arbeidsmiljø i hjemmet, v) Å rekruttere og holde på kvalifisert personell.

Vi fant at ALS både er en kronisk og terminal sykdom. Et vilkår for samarbeid er at det er en felles forståelse av sykdommen mellom de involverte aktørene. Vi fant nyanser i typene omsorgsarbeid til pårørende i ALS, og at pårørende må balansere det å møte forventninger fra seg selv og andre, og ikke å bli overveldet av omsorgsarbeid. Et vilkår for samarbeid er at de involverte aktørene har en felles forståelse og verdsetting av ulike typer omsorgsarbeid. Vi fant fem ulike utfordringer i håndtering av ALS i kommunehelsetjenesten. Et vilkår for samarbeid er å bygge relasjoner som åpner for å forhandle om tillit og beredskap i kommunehelsetjenesten. Kommunen må kunne koordinere omsorgen raskt, samt ha tilgjengelig kvalifisert personell og å gi tilstrekkelig sosial støtte til personell som jobbet hjemme.

Contents

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All errors and mistakes in this thesis are, of course, my own.

Preface

When I applied for and subsequently embarked on this project focusing on amyotrophic lateral sclerosis or motor neurone disease I had two motivations. First, I had a clear impression that the project was important. I felt strongly that it could make a difference, and that more knowledge was needed. Secondly, it was an opportunity to do work that I was trained to do. During my previous studies I had become familiar with qualitative research on health professionals and the health services. While I was aware of the literature on illness experiences, the perspective of family caregivers was intriguingly new to me. This thesis is rooted in sociology, but I have tried to write for a multidisciplinary audience.

List of papers

Paper I

Lerum, S. V., Solbrække, K. N., Holmøy, T. & Frich, J. C. 2015. Unstable terminality: negotiating the meaning of chronicity and terminality in motor neurone disease. Sociology of Health & Illness, 37, 81-96.

Paper II

Lerum, S. V., Solbrække, K. N. & Frich, J. C. 2016. Family caregivers' accounts of caring for a family member with motor neurone disease in Norway: a qualitative study. BMC Palliative Care, 15, 1-9.

Paper III

Lerum, S. V., Solbrække, K. N., & Frich, J. C. (in review), Health care professionals' accounts of challenges managing motor neurone disease in primary health care: a qualitative study

List of abberviations

ALS: Amyotrophic lateral sclerosis

LMN: Lower motor neuron

LTMV: Long-term mechanical ventilation

MND: Motor neurone disease

PEG: Percutaneous endoscopic gastrostomy

UMN: Upper motor neuron

List of tables and figures

1. Introduction

Motor neurone disease (MND), also known as amyotrophic lateral sclerosis (ALS), causes muscle wasting, breathing and swallowing difficulties, and respiratory failure, and may involve cognitive impairment (Kiernan et al. 2011, Andersen et al. 2012). In ALS/MND the biomedical understanding is limited, attempts for a cure are experimental, and information is uncertain. ALS/MND is not curable, but there are many therapeutic interventions that can relieve symptoms and prolong and improve life. The most radical intervention in this regard is invasive ventilation via tracheostomy which has the potential to prolong life by several years, but the person is unable to move and caregivers must be present at all times. As the condition progresses, the person with ALS/MND is gradually paralysed, creating a need for large amounts of care work and many tailored assistive devices to replace functionality. As most people with ALS/MND live at home, there is often just one adult person to help out who then becomes a family caregiver. However, the workload and skill required to manage the situation can easily grow beyond the capacity of a single individual, creating a need for collaboration. This study’s focus on family caregiving and care collaboration arises from these conditions. The study has followed the diagnostic group ALS/MND through the Norwegian health service, exploring the work of caring and managing ALS/MND, using qualitative interviews with three groups: 1) family caregivers and persons with MND living at home, 2) health care professionals in primary health care, and 3) health professionals in hospitals. The sample comprise to a total of 53 qualitative interviews with 65 participants spanning family caregivers, people with ALS/MND, primary health care staff and hospital personnel.

MND and ALS are often used interchangeably. In Norway, the U.S., and in continental Europe, ALS/MND is more commonly known as amyotrophic lateral sclerosis (ALS). In the U.S., the condition is also frequently called Lou Gehrig’s disease. As the papers in this thesis are published in journals using British English, I have chosen to use the British term *MND*, for consistency.

The overall aim of this study is to contribute to knowledge about conditions for collaboration between family caregivers and health professionals managing and caring for individuals with MND. The thesis has three specific aims:

- To explore the meaning of chronicity and terminality in MND (Paper I)
- To understand more about family caregivers' work and sense of responsibility by exploring their accounts of caring for a family member with MND (Paper II)
- To explore what challenges health professionals encounter when managing MND in primary health care in Norway (Paper III)

This summary has two parts. The first part leads up to the findings, presented in the three papers (Papers I, II and III). The second part discusses these findings and the methods used.

In the following chapter on context and background I provide the conditions for the aims of this study, and how they relate to existing literature. The next chapter on theoretical perspectives continues the effort of establishing the study's aims. Additionally the theory chapter presents the analytical tools that have been used in the analysis, and the lenses to make sense and interpret the findings and the discussion of the findings. The methods chapter describes the rationale behind the empirical study and how the research was carried out. The methods chapter is followed by a presentation of the findings, with extended summaries of each paper. The text then moves on to the second part. In the chapter discussing methods, I offer a critical appraisal of what has been done. Next, when discussing findings, I contextualise the findings in relation to a broader literature within the field. In the concluding chapter, I summarise the principal claims that have been touched upon in the previous discussion chapter, crystallise the thesis's contribution and suggest some topics for further research.

2. Context and background

2.1 Motor neurone disease

My point of departure is to present MND as a medical condition.

MND has four main clinical presentations: 1) limb onset with a combination of upper and lower motor neuron (UMN and LMN) signs in the limbs; 2) bulbar onset, presenting with speech and swallowing difficulties and with limb features developing later in the course of the disease; 3) the less common primary lateral sclerosis with pure UMN involvement; and 4) progressive muscular atrophy, with pure LMN involvement (Kiernan et al. 2011:943). While initial symptoms are mild, perhaps a weakness in a hand or a foot, more and more muscles are affected as the disease unfolds. Progressively, there is a loss of functionality, and the person with MND gradually needs more assistance to perform everyday tasks. The loss of functionality can be replaced by either physical labour or technology. The fact that there are so many variations of the disease makes it more difficult to predict how the disease will unfold, and complicates care and management of the condition.

The incidence of MND in Europe is fairly stable at 2.16 per 100 000 person years (Kiernan et al. 2011). The incidence of MND in Norway was in 1994 estimated to be about 2.5 per 100 000 (Seljeseth et al. 2000). However, based on data until 2014, incidence appears to be increasing especially in age groups above 60 (Nakken et al. 2016). Improved diagnostics and aging population appears to be the main explanation, for an incidence of about 2.8 per 100 0000 in 2014, in Norway (Nakken et al. 2016). There are regional differences in incidence in MND in Norway, but these variations do not follow any clear trend (Seljeseth et al. 2000, Nakken 2016). The prevalence of MND in Norway is estimated to be about 5-7 per 100 000 (Nakken 2016). More than 90% of cases are sporadic, while familial MND accounts for approximately 5% of cases (McDermott & Shaw 2008). Peak age at onset is 58-63 years for sporadic MND and 47-52 years for familial MND (Kiernan et al. 2011). MND is slightly more common in men, with a gender ratio of 1.3:1 for men to women (Seljeseth et al. 2000). MND is relentlessly progressive – 50% of persons with MND die within 30 months of symptom onset and about 20% of patients survive between 5 and 10 years after symptom onset (Kiernan et al. 2011). The dying process usually begins with the individual slipping from sleep into a coma due to increasing hypercapnia (increased levels of carbon dioxide in the blood; Mitchell & Borasio 2007). Death in persons with MND was perceived by caregivers as being peaceful in 88% of patients in Germany, 98% in the United Kingdom, and 91% in the United States and Canada (Simmons 2005).

The condition was previously thought to affect mainly motor neurons, but there is growing evidence of cognitive symptoms present in persons with MND (Merrilees et al. 2010), implying that other neurons are affected as well. Such symptoms may initially be vague and risk being overlooked (Kiernan et al. 2011). Milder cognitive deficit estimates range from 10% to 75% in different studies, while the proportion of patients with dementia has been estimated to range from 15% to 41% (Miller et al. 2009b). The most common deficits involve executive function, either affecting language or personality, which might cause problems with judgement and impulsivity and a general deterioration in the ability to undertake routine daily tasks (Kiernan et al. 2011:945). Still, the presence of severe dementia and personality change is considered generally rare (Mitsumoto 2009).

2.1.1 Treatment

Riluzole is a disease-modifying drug which is used in the treatment of MND. In two large randomised controlled trials, riluzole extended survival by 3-6 months (Kiernan et al. 2011:948).

Many symptoms of MND can be treated (Miller et al. 2009b, Miller et al. 2009a, Andersen et al. 2012). There are assistive devices available to aid and support everyday activities, tailored to the specific context of the individual with MND. In the following I account for two interventions related to nutritional support and breathing, which largely affect the unfolding of the disease.

As the disease progresses, eating and swallowing food become increasingly difficult. This may be countered by percutaneous endoscopic gastrostomy (PEG), which involves passing a tube through the abdominal wall to deliver nutrition directly. The procedure involves mild sedation and may involve risk for persons with MND with respiratory problems and those who are in advanced stages of the disease. Clinical reviews recommend that the procedure be done earlier in the disease trajectory (Andersen et al. 2012).

Difficulty breathing is an inevitable aspect of the disease progression. To counter this development the person with MND may choose between invasive or non-invasive ventilation. Non-invasive ventilation involves using a mask to help breathing. The mask might be used at all times, or only occasionally, often during sleep. Non-invasive ventilation is generally recommended in the literature (Leigh et al. 2003, Miller et al. 2009b, Andersen et al. 2005). Invasive ventilation via tracheostomy involves the surgical placement of a tube directly into the windpipe below the vocal cords. This intervention has the potential to prolong life for several years. However, there are drawbacks to the advantage of prolonged life-expectancy. It involves an extreme lifestyle; people in the advanced stage of the disease are often unable to move and require the constant presence of caregivers. The person with MND risks losing the ability to move any muscles and becoming ‘locked in’, unable to communicate (Leigh et al. 2003). Emergency tracheostomy in respiratory failure is technically possible but represents a challenging situation from an ethical perspective. Many persons with MND make arrangements with family and caregivers regarding advance directives, should such an emergency situation occur.

From a clinical perspective the difference between invasive and non-invasive ventilation is profound. With regard to non-invasive ventilation, a mask may be administered by the person with MND or by family caregivers, with relative ease. On the other hand, invasive ventilation via tracheostomy involves the presence of skilled caregivers day and night. It is among the most advanced treatments supported by primary health care in Norway.

There were 1 282 persons registered in Norway as using invasive and non-invasive ventilation at home during 2002-2007, across an array of diagnoses. Of those, 93% were using non-invasive ventilation (Tollefsen et al. 2009). In 2007 there were 22 persons with MND living at home with tracheostomy ventilation in Norway (Tollefsen et al. 2009). There is large variation across societies regarding use of invasive ventilation (Andersen et al. 2012). In 2014 the number of persons with MND on invasive ventilation was 79, while 237 persons with MND used non-invasive ventilation, in Norway (Indrekvam et al. 2015).

In short, MND is a neurological condition whose underlying disease mechanism is not fully understood. While rapid physiological progression might be expected, there is large variation and life expectancy is difficult to predict. Furthermore, the person with MND and family may choose interventions that can both improve quality of life and prolong life expectancy. The unfolding of the disease involves decision-making about treatment and assistive devices. Managing MND involves massive amounts of equipment, advanced technical tasks and a large number of personnel. What does this mean for those involved? How do they relate to unpredictability? How do family caregivers relate to this work? These questions are central in this thesis.

2.2 Care & management

2.2.1 The concepts care, management and family caregiver

There are several ways to understand care and care work. Mol (2008:1) points out that care may be understood in contrast to cure, to make daily life more bearable, rather than to eliminate problems. Care may also be understood as a gift, which is often the case in anthropology, with accompanying expectations of reciprocity and exchange, for instance in the form of collaboration (Kleinman 2012). Care can be broadly understood as 'a concept encompassing that range of human experiences which has to do with feeling concern for and taking charge of the well-being of others' (Wærness 1984:188). 'Taking charge' implies agency in caregiving, but as Christensen points out, it is not given whether it is the caregiver or the person being cared for who is in charge of the situation (Christensen 2012). According to Wærnes, providing good care involves finding a balance between too much care and too little: 'Good caring should be performed in such a way that it, as far as possible, reinforces the self-sufficiency and independence of the receiver. The dependent is neither to be overprotected nor to be neglected' (Wærness 1984:189). To provide care is work; it takes time and energy, it requires skill and expertise, there is need of a resource pool to draw from and a motivated work force (see Corbin and Strauss 1988:118). Care, then, needs to be managed. By management or managing in this context, I think of how care work entails a plan, some goal to be attained (for instance a balance between self-sufficiency and dependence), decisions to be made (practical or ethical), problems anticipated or reactively addressed.

I use the term ‘family caregiver’ for several reasons. I understand care as an activity, which I underscore by using the term caregiver instead of ‘carer’. I began to focus on activity while working on Paper II; therefore, the term family caregiver is used consistently in Papers II and III but less so in paper I (which mainly uses carer, but also other terms). Some researchers use the term ‘next of kin’ to underscore how individuals are related, and to direct attention away from a focus on activities (Olsson et al. 2010, Olsson Ozanne et al. 2011). I use the word ‘family’ because it is descriptive; that is, the participants in this study were family members – spouses, siblings or other relations. This is the main reason for not using the broader term ‘informal caregiver’. A second reason is to underscore the normative and emotional connections between the caregiver and the cared-for. Compared to a professional or paid caregiver relationship, the family relationship has other expectations attached.

2.2.2 Family caregiving and MND

The progressive nature of MND creates changing care needs resulting in ‘a state of ongoing change and adaptation’ (King et al. 2009). For example, a person with MND might for some time benefit from a specially equipped car, enabling him or her to drive. However, as the condition progresses, it might become impossible for the person to drive at all, thus rendering the car unnecessary. Now, rather than needing a specially equipped car, there is a need for a driver. The progression of the condition causes new care needs to arise, while old ones become irrelevant. People and technologies have to work in concert to meet these changing needs.

Adding to the complex nature of MND care is the vast amounts of care that are required. In an Italian study (Chio et al. 2006), family caregivers recorded in diaries the time spent on care tasks (such as feeding, supervision, dressing, administering medication, etc.). The average time spent on caregiving was 9.5 hours a day, with care time ranging from 5 hours for persons with mild disability to 15 hours for those with severe impairment. In another study it was estimated that caregivers on average spent 15 hours a day with the person with MND and 8 hours a day providing direct help (Rabkin et al. 2009). As Rabkin and Albert note: ‘Even a good night’s sleep can make a major difference for caregivers’ (2009:302).

In addition to the changing nature of MND care and the large amounts of it, facing a condition without a cure also involves emotional losses. In an Australian study (Ray & Street 2007), family caregivers reported that the uncertainty of the progression threatened both their expectations of life and their future. Family caregivers’ basic assumptions about life were constantly challenged and had to be revised to accommodate each new loss. These authors also point out that family caregivers often avoided discussing the future with their partner, and kept emotions related to the future to themselves.

In a recent review of the literature on family caregiving in MND, a main finding is that family caregivers experience exceptional strain (Aoun et al. 2013). Receiving the diagnosis, obtaining information and access to health services, making the decision and transitioning to assisted ventilation, encountering cognitive changes in the person with MND, and caregiving and decision-making in the last months of life have all been noted to be especially difficult phases in the care trajectory (Aoun et al. 2013). However, Foley and colleagues (2014a) have argued that a life-course perspective is more sensitive to identifying challenges from the perspective of those at home, such as parenthood or age, than a standardised notion of how the disease unfolds. Further, Olsson and colleagues (Olsson Ozanne et al. 2011) point out that it appears that the dyadic relationship between the person with MND and the family caregiver has a larger effect on well-being than the physical progression of the condition.

Summarising the literature accounted for above; MND care and caregiving are characterised by rapidly changing tasks, which often involve complex interventions, such as breathing aids or PEG. In this continuously changing landscape many assistive devices require individualised adaptation. Moreover, as the literature shows, the volume of care required, with regard to labour and technology use, is vast. Facing a fatal condition that has no cure is also emotionally intense. Due to the changing needs, the use of technology and the share volume of care, it gradually becomes a necessary to have a relationship with service providers. In this context it becomes important to ask how family caregivers describe this work and how they regard their responsibility.

2.3 Norwegian health care

As the previous sections describes, persons with MND and their family caregivers have need of supportive services. What characterises the health care system the persons with MND and their family caregivers utilise and within which the health professionals involved in their care work?

Norway has a tax-based public health service, which is administered in a two-tier system (for an overview of Norwegian health services see Ringard et al. 2013). The state finances and runs specialist services, like hospitals, through regional trusts. It is the local communities, organised in municipalities that finance and run primary health care health services. An important motivation for this study is the 2012 ‘Coordination Reform’ (Norwegian Ministry of Health and Care Services 2009). This reform touches upon several core issues for this project. It aims to improve the care coordination between primary and specialised care. Further, it argues that to achieve financial sustainability in the health sector it is necessary to reallocate more advanced care and treatment tasks from specialised services (hospitals) to primary health care. MND is an example of a trajectory spanning both primary and specialised services, and where care and treatment routinely were among the most advanced forms of services sustained in primary health care. MND is a case to shed light on these core issues of the reform. This policy was aided by improved medical technology, which allowed more complex tasks to be performed outside specialised institutions. On a policy level there is broad agreement that fragmentation of services is a main challenge for the health services (Norwegian Ministry of Health and Care Services 2015, 2009). The advanced care tasks that accompany MND and the need for several professions and organisations to cooperate to carry out these tasks make the condition a case that is particularly illustrative of these policy and technology trends.

It is primary health care that provides day-to-day services to people with MND in Norway. Primary health care services are organised through the municipalities, which are the main providers of health and social services in Norway. Municipalities are relatively autonomous with their own political elections every four years. Around half of the more than 428 municipalities in Norway have fewer than 5 000 inhabitants. The population in the municipalities creates the tax-base. The smaller municipalities have a limited capacity to build a skill base among their personnel and in their institutions to target rare and resource-intensive cases such as MND. To compensate for this structural problem, and to ensure the political goal that all inhabitants of Norway have the same access to services regardless of which municipality they live in, the state refunds the municipalities for cases that are particularly resource-demanding. In MND cases, a substantial amount of the cost at the municipal level is covered by the state. The state refund provides the municipalities with financial elbow room in resource-demanding cases like MND, by limiting the toll that this spending takes on local budgets.

The relative autonomy of the municipalities allows them some freedom in the actual organisation of services, and primary health care can be organised in slightly different ways from municipality to municipality. However, in the organisational landscape of primary health care there are three physical arenas where care work often takes place: 1) the private home where the person with care needs lives, 2) various assisted living facilities, 3) nursing homes. Supporting these arenas are free-standing professionals, such as occupational therapists, physical therapists, general practitioners and the like. In addition, there are often collaborative units, such as health professionals with designated coordinative duties and offices with coordinative functions (*Bestillerkontor*, or Purchasing unit/Local procurement unit).

Vabø (Vabø 2012) has noted that the introduction of such purchaser-provider models represents a shift in home services. It represents transparency, but the accompanying strict focus on tasks may contribute to fragmentation of services because the system may encourage institutions to off-load responsibilities, providing less flexibility to individually tailor services. An example of an attempt to compensate for fragmentation in primary health care is the individual care plan, which was introduced by law in 2001 (Bjerkan et al. 2011). The individual care plan has an administrative intention, which sets it apart from the clinical focus of patient records. Individual care plans attempt to facilitate better collaboration across organisational boundaries by using tools such as meetings and having a designated professional responsible for each plan. The plans are designed for persons with complex and chronic care needs (Bjerkan et al. 2011). However, even though this kind of care is a legal entitlement, there are fewer of the individual care plans than the targeted patient group would suggest. Bjerkan and colleagues point out that it is not clear who is responsible for the plans, and they conclude that the plans have not been successful at contributing to improve multidisciplinary cooperation.

In Norway there are specialised MND multidisciplinary teams in hospitals, although some Norwegian hospitals organise their MND follow-up without any clear team structure. Multidisciplinary clinics are recommended in international guidelines on MND care (Miller et al. 2009b, Andersen et al. 2012). In Norway, the number of people within a hospital catchment area is larger than most municipalities. Hence, hospital staff may to a larger extent than health care professionals in primary health care, accumulate experience across MND cases. Hospitals provide the MND diagnosis and initiate interventions such as breathing aids or PEG. The day-to-day follow-up of these interventions is managed by primary health care.

Hospitals are in a position to accumulate experience with MND cases. Most municipalities are not in such a position, and some may expect no MND cases at all. Consequently, from a care perspective, while hospitals may have the necessary competence, they are not to perform the care work at home where the person with MND lives. Primary health care, on the other hand, has to do the job, but struggles to acquire the competence. The health services in general, and primary health care in particular, represent a complex organisational landscape that struggles with fragmentation. Given this contextual backdrop, primary health care has to manage fragmentation and to accommodate a clear political intention of performing more advanced tasks. How does this context influence the quality of MND care? What demands and challenges is primary health care facing when trying to provide MND care?

2.4 Summary of context and background

This study, then, follows the condition MND across the health services, from those at home, to primary health care and providers in hospitals. Despite extensive research, there is no curative treatment for MND, but other interventions may improve quality of life and prolong life expectancy. For the health services, MND is an extreme case. The tasks required to sustain a person with MND in an advanced state are at the limit of what primary health care is capable of, making it necessary to find a way to collaborate.

3. Theoretical perspectives

In this chapter I position the thesis theoretically. The aim of this presentation is to establish the tools to necessary understand the analytic process for each paper, and findings in the findings chapter. Some additional theory is presented in the discussion chapter, to establish concepts to discuss across the papers.

This thesis is influenced by the American symbolic interactionist tradition. One of this tradition's most famous advocates, Herbert Blumer (1969a:2), formulated the following three premises: 1) Humans act towards things on the basis of the meanings the things have for them; 2) The meaning of things is derived from the social interaction one has with one's fellows; 3) These meanings are handled and modified through an interpretative process used by the person in dealing with the things she encounters. Hence, meaning is both reproduced and altered by actors.

Following this perspective the meaning of MND is not inherent to the condition. Nor is it a result of the pure inner world of an isolated individual; we run into trouble if we understand MND entirely differently from those around us. Instead, according to the interactionist tradition, the meaning of MND should be seen as embedded in interaction, in culture – in what people have said and written about it, and our own interaction with this information. Hence, MND does not have a fixed meaning. MND might be perceived as a punishment from God, as something to be ashamed of and to hide from others. Another interpretation would be that MND strikes randomly, nothing to be ashamed of, but rather a situation where sharing experiences would allow for people to exchange of tips and tricks to make a difficult situation less so. We cannot randomly ascribe meaning to MND; we would stand corrected by arguments from other people and the condition itself. Culture – in this case Norwegian culture, which is in slow but constant movement – contains a repertoire of symbols to use in negotiating of the meaning of MND. By 'symbols' I do not mean religion or that something is symbolic, as in being empty of meaning. Quite the contrary, symbols are objects, gestures, speech sounds, stories, memories – in a broad sense, elements with the capacity to signal something to others, to represent something (Joas & Knöbl 2009:128).

3.1 Work and illness trajectories

There are two core theoretical concepts for this thesis. The first concept is work, and the second is illness trajectory. I devote a section to each of these.

I find work to be a sensitising concept (Blumer 1969b). Blumer argues that a sensitising concept is a concept that offers a hunch that suggests a direction to look in and what to look for. The reason for having less rigidly defined concepts derives from the ontological status of social practice, which is, in part, inconsistent, contradictive, and highly variable. Concepts that are either too definitive or too narrowly defined run the risk of excluding natural variation of the same pattern, interaction, or event. Blumer uses such terms as ‘the institution’ and ‘the social’ as examples of sensitising concepts. Viewing work as a sensitising concept, it is a tool that allows us to interpret this complexity. I understand Strauss to use work in such a way. For instance, in his book on medical work (Strauss et al. 1985), he mentions at least 22 types of work: safety work, machine work, and articulation work or sentimental work, to mention but a few. These are concepts that provide research guidance on where to look and how to understand interaction.

Further, when we term something work, other things happen too. The term ‘work’ points to the fact that an effort is being made, that there is a purpose to the activity (in the interactional sense of meaning outlined above), that it requires competence and skill, that you can get better at it and learn from others, that you can get tired from it. Work is closely related to managing constraints. By referring to an activity as a type of work, then, is to *recognise* it, in the double sense of *describing* as well as *respecting* it. A prime example is care work. By referring to the activity of providing care as care work, we ascribe the properties mentioned above to it, which is a typical feminist move (see Waerness 1984). This is in contrast to understanding care as an effortless activity that does not require skill and is taken for granted. It is important to use the double analytic meaning of work to both describe and to show respect if we wish to understand more about the work of family caregivers in MND.

My normative position, which is heavily inspired by this symbolic interactionist tradition, is that the participants in this study are skilled at what they do. Such skills run the risk of being ignored, even by the people who perform them. I have attempted to identify and articulate this experienced-based competence, for instance by labelling it work (paper II). Another example is when I explore the meaning of chronicity and terminality in MND (paper I).

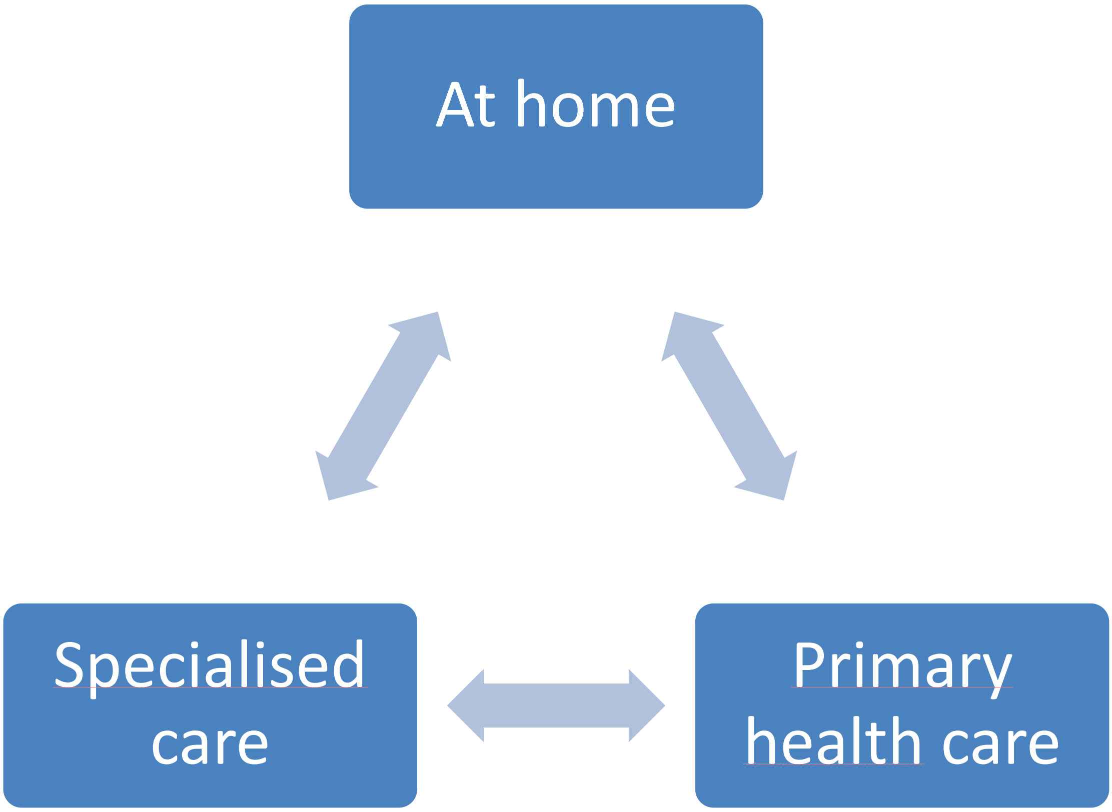
The other central theoretical concept for this thesis is *illness trajectory*. Anselm Strauss and colleagues developed this classic concept to sociologically explore and understand the unfolding of illness (Corbin & Strauss 1988, Strauss et al. 1985). The concept includes not just the physical unfolding of illness but all tasks, work, personnel, technology, organisations, and overall welfare regimes that shape the course of the illness. Importantly, in this landscape, it is rare that one single actor defines what will happen. Rather, interaction or negotiation between several actors shapes the trajectory, for instance the interaction between the condition and available treatments. Illness trajectories might seem obvious, but the concept represents a sharp contrast to understanding illness as a process that runs its course undisturbed, or in cases where the physician can introduce a ‘silver bullet’ such as anti-biotics to effectively stop the ailment.

Strauss developed the concept of illness trajectories studying hospitals (Strauss et al. 1985). His study focused on the patient in a clinical setting. However, people who are chronically ill spend most of their time at home, and Strauss also contributed to a study focusing on persons with illness and family caregivers who lived their lives at home (Corbin & Strauss 1988). Corbin and Strauss (1988) point out three lines of work performed by the person with the illness and family caregivers. The first is illness work, which is work directed at the illness. The second line of work is biographical work, which is work to accommodate a life with chronic illness into one’s sense of time, self, and body. The third line of work is everyday work, which is work not directly related to illness, such as housekeeping, occupational work, raising children or the like. These lines of work conceptualise how family caregivers shape and interact with the illness trajectory. They play a central part in exploring family caregivers’ work, which is the second aim of this thesis.

3.2 The MND context – communities of practice

The concept of illness trajectories has generated an abundance of research, but also critique. Davina Allen and colleagues (2004) point out several shortcomings: Illness trajectories often focus on contingencies arising from unexpected turns of the illness or from medical interventions. According to Allen and colleagues these contingencies remain contingencies and are not theoretically conceptualised (Allen et al. 2004:1011-1012). I have tried to compensate for this under-theorised aspect of illness trajectories by introducing communities of practice as a way of theorizing how illness trajectories are shaped by contingencies. The figure below illustrates how I understand the landscape in which illness trajectories play out (Lerum & Frich 2012):

Figure 1: The MND context as a community of practice



I view the overall MND context, the three boxes taken together, as a community of practice (Wenger 2011). A community of practice is a central arena for interaction that contributes to meaning-making. Communities of practice have three main characteristics: domain, community and practice. 1) Domain refers to shared domain of interest, such as handling MND. 2) Community refers to engaging in joint activities and discussions, helping each other, and sharing information; in short, interaction. At home this might involve using internet resources or contacting others who are living with MND. In primary health care this might involve contacting colleagues managing MND in other vicinities to see what they do and to share experiences. 3) Practice and specifically a community of practice, refers to a community of practitioners, and not a community of, for instance, interests. This means that the community members develop a shared repertoire of resources, experiences, stories, tools, and ways of addressing recurring problems. For instance, nurses may share stories about cases in their practice during lunch, as a type of sustained practice-based interaction. In hospitals there are several communities of practice, depending on the patients and tasks care providers are involved with. Communities of practice cut across formal organisational structures, and thus may be seen as a network of colleagues (Wenger 2011). A community of practice may seek to formalise, for instance as a patient association. However, this is not a requirement for a community of practice, and those who chose not to be a member of a patient association are still embedded in the same practice. To become a member of a community of practice you need to take part and commit to the domain, community and practice in that particular case.

Within the overall community of practice around MND, there are three subsystems – at home, specialised care and primary health care. There is a loose coupling between the subsystems and the overall community of practice. Each subsystem develops its own informal culture, which informs meaning-making and categories. This informal culture 'relates to how the members within a subsystem create their own definitions of the work they do, and they act based on these definitions' (Lerum & Frich 2012:35). This is not to say that the differences between the subsystems are absolute; rather, the point is that the subsystems do not necessarily share a unified understanding of MND. This assessment follows Bowker and Star (Bowker & Star 2000) who argue that classifications and categories are naturalised within a community and gradually become taken for granted.

In the case of MND the illness trajectory cuts across the subsystems in the community of practice. Each subsystem consists of actors who contribute to shape the trajectory. Specialised clinics in hospitals provide diagnosis and initiate interventions. Primary health care sustains care and treatment initiated by specialised care. Most of the work, however, takes place at home where the person with MND and their family live. These subsystems must collaborate in order for the illness trajectory not to break down.

3.3 Illness experiences

To understand the interaction between health professionals and family caregivers managing an illness trajectory, I find the concepts of lines of work and communities of practice to be nuanced and effective theoretical tools. To theoretically conceptualise the experience of illness, I turn to narrative theory and the concept of ‘biographical disruption’. I find these approaches important to thinking about the meaning of illness, including whether to view MND as a chronic or terminal illness.

3.3.1 Meaning and narrative

According to Polkinghorne (1988), creating narratives is a fundamental human cognitive quality that entails ‘noting the contributions that actions and events make to a particular outcome and then [configuring] these parts into a whole episode’ (Polkinghorne 1988:6). A story, then, is something an actor imposes on the world, to create meaning and to give a sense of orderliness to events and everyday life. To create a narrative is to subjectively impose causality or explanation on what happens in the world, drawing on a very broad range of factors (personal, relational, traditional, cultural, subjective, objective and so on). From a symbolic interactionist perspective, creating narratives is an interactional endeavour that involves reusing culturally available symbols and adapting them to the individual context. A condition for creating narratives is that we have to construct them based on insufficient information. If absolute knowledge of our surroundings was a condition for action, this would lead to an inability to act. Creating stories to make sense of one's life despite a lack of information is a necessary and fundamental human activity.

Arthur Kleinman has noted how narratives play a particularly important role when people are facing illness. The process of meaning-making in chronic illness involves transforming illness from a wild, disordered and natural occurrence into a more or less domesticated, mythologised and ritually controlled happening; hence, it is a cultural process (Kleinman 1988:48). Culturalising illness is something all sick people do, and health professionals play an important role in facilitating that effort, for example through prognostication. Creating meaning, from this perspective, involves translating the sickness, the natural process in the body, into an understandable or cultural event.

Brown and Addington-Hall (2008) interviewed people with MND and explored narratives in the MND context. They found four narratives in MND. The preserving narrative focuses on actively pursuing potential opportunities to increase the chance of survival, for instance by pursuing options outside the traditional health services. The sustaining narrative is an approach that focuses on the positive and on encountering challenges one by one, day by day. This approach emphasises what is achieved, rather than what is no longer possible. The enduring narrative, on the other hand, tells about quiet suffering, where neither life nor death was a welcome option. The enduring narrative provides a way to live through a difficult situation: there is little to be done, one has only to endure. Finally, the fracturing narrative tells of loss and breakdown. There is a shattering of the future, for instance not seeing one's own children grow up or having to abandon a professional career.

Such narrative typologies are not meant to be boxes to put people's experiences into; rather, they are to be seen as tools to help each person make sense of his or her own situation. One person may simultaneously draw on several of the narratives, or move between them, when creating his or her own personal story. An individual's narrative is unique. In the MND context, Jeppesen underscores the importance of narrative's ability to express difference and variation in illness experiences, a feature that makes narrative an effective and nuanced mode of self-presentation (Jeppesen 2013).

3.3.2 Biographical disruption

Biographical disruption (Bury 1982) offers another perspective from which to understand chronic illness, and has laid the groundwork for a longstanding sociological interest in illness experiences. Interviewing people with rheumatoid arthritis, Bury noted how the illness resulted in 1) ‘the disruption of taken-for-granted assumptions and behaviours; the breaching of common-sense boundaries’ (Bury 1982:169). As normal activities ‘become deliberately conscious activities’ (Bury 1982:175), the illness disrupts underlying foundations for reciprocity and social relationships, as well as material and practical affairs. 2) The disruption also involves a ‘fundamental re-thinking of the person’s biography and self-concept’. 3) In response to the altered situation caused by the disruption, people mobilise resources, of a personal, material, social or other kind (Bury 1982:169). This last point is important, as it points to Bury’s sensitivity to social class and the resources embedded in class position. Bury has suggested that biographical disruption is better understood as a heuristic than a formal concept (Locock & Ziébland 2015). I use biographical disruption as a heuristic, a cognitive short-cut for summarising, understanding and increasing my sensitivity to a complex phenomenon: the management of large unexpected life events.

This approach arose in the late 1970s, a time when new technology such as in asthma inhalers or insulin for diabetes began converting fatal conditions into chronic illness (Locock & Ziébland 2015). As a response to these developments, several studies called for a shift of attention from the causes to the consequences of illness. William’s ‘narrative re-construction’ (O’Brien et al. 2012), and Charmaz’s ‘loss of self’ (Charmaz 1983), are examples of concepts born out of this context. The shift from causes to consequences also refocused sociologists’ attention from the *patient* in a clinical setting to the *person* living a life outside of health services (Locock & Ziébland 2015). This was the context that gave rise to the concept of biographical disruption (Bury 1982). The conversion of fatal conditions into chronic ones using technology is central to this thesis’ exploration of the meaning of chronicity and terminality in MND.

Biographical disruption draws on narrative to make sense of the disruption. Gay Becker has pointed to the importance of narrative and plot disruptive events. She has expanded the biographical disruption approach by exploring many different forms of unexpected life events, such as divorce or infertility (Becker 1997). Her point of departure is that order is a condition for something to disrupt, and that order and predictability are cultural products. In Western societies our cultural expectations of order are strong, and as these expectations grow, crisis and chaos gain disruptive momentum. Faced with a disruption, a person enters a liminal phase in which they try to figure out what to do. This neither-nor phase, or limbo, may be short or long, depending on the situation. The limbo is ended by finding a solution or reconnecting to a stable familiar or altered situation.

Biographical disruption is a widely cited concept, but it has also been challenged and adapted, as in Becker's example above. Age has turned out to be an important factor in whether chronic illness actually is disrupting (Locock & Ziébland 2015). There are phases of life where illness is expected and thus not considered a disruption. This has been conceptualised as biographical flow or continuity (Locock & Ziébland 2015, Pound et al. 1998). Carricaburu and Pierret (1995) have expanded biographical disruption by adding the concept biographical reinforcement, pointing out that chronic illness may also strengthen identity and group belonging. Adamson and Donovan (2005) have used biographical disruption as a concept that is relevant for family caregivers. A key issue is that family caregivers may feel that no one else could care for their relatives as well as they can. This may create a moral imperative to perform care work. When faced with severe illness, then, family caregivers may also experience disruption of an everyday that was taken for granted, requiring them to fundamentally rethink their own biography and to mobilise and respond to the altered situation. Further, biographical disruption has also been expanded from describing individuals to include dyads. Aasbø and colleagues (2016) point out that spouses who may have a shared biographical project may find that sense of 'biographical we' is disrupted by chronic illness. Maintaining the biographical we, then, is a reciprocal affair between the person with illness and the family caregiver, which requires family caregivers to make 'great efforts to maintain the routines of an ordinary everyday life and sense of continuity in their relationships, despite the progressive illness trajectory of their partners' (Aasbø et al. 2016).

In a key study for this thesis, biographical disruption has been explored in the context of MND (Locock et al. 2009). Locock and colleagues coined the term ‘biographical abruption’ to set the MND experience apart from ‘normal’ chronic illness (Locock et al. 2009). Biographical abruption refers to how the MND diagnosis was understood as a death sentence. The diagnosis was experienced as an ‘existential shock’ (Brown 2003) because it denied the person a future rather than simply forcing them to fundamentally rethink it. This created a notion of ‘a sense of life over but not over, a sense of living death’ (Locock et al. 2009:1049). Importantly, Locock and colleagues note how people with MND mobilise and are able to create strategies for maintaining a sense of normality. Examples include holding on to aspects of normality, such as driving; creating new normality, such as taking up creative writing; and creating a heightened normality, a strategy in which vacations often played an important role. Even though facing MND was devastating, some people were still able to find new meaning, such as when personal relationships grew closer as the illness progressed. A single individual might ‘straddle’ different categories, from utter despair to having a sense of being able to live with the condition (Locock et al. 2009:1051-1055). The work of Locock and colleagues built upon Brown’s (2003) notion of MND being ‘astride’ terminal and chronic categories (2003:216). We explore this ambiguous approach to MND and what it represents in Paper I.

The tradition following biographical disruption, then, sets MND apart from ‘normal’ chronic illness; it is a terminal illness that shares many features of typical chronic illness (Locock et al. 2009). How does this ambiguity play out at home and translate into the health services? The advances in medical technology that initiated a new tradition in the 1970s by turning fatal illnesses into chronic ones are continuing today, effecting more conditions.

3.4 Classification

Illness experiences are different in chronic and terminal illness; so too is the health services’ management of these different categories. These two categories are the core of the first sub-aim, namely to explore the meaning of chronicity and terminality in MND. But what do they entail? Taking a symbolic interactionist approach Bowker and Star (2000) have studied classification as a form of meaning-making. They point out that we have to classify and categorise our surroundings; this is the basic way we manage our everyday. Otherwise we would be overwhelmed by the complexity of our surroundings, which would result in an inability to act. When there is a mutual understanding of a category, the categorisation becomes effective as a means of coordinating action between people. Importantly, a category ‘valorizes some point of view and silences another’ (Bowker & Star 2000:5). Hence, it is important to reflect on the categorisations we use and the consequences they have. This is especially salient when facing a condition like MND, as categories may contribute to a difficult situation becoming more or less so.

Classification systems follow a logic of purity. They utilise consistent principles in their operation, producing mutually exclusive categories and creating a complete system of the worlds they describe (Bowker & Star 2000:10-11). Still following Bowker and Star, a core argument of Paper 1, is that practice is not pure. Therefore, when used in real life, categories must be worked on, ignored or adapted. It is the practical use of the categories *chronic* and *terminal* that we explore in Paper I.

3.4.1 Prognosis

An important tool for making sense of and classifying a trajectory is prognosis, which points to what will happen in the future. A prognosis may relate to statistical probability, but there is a subjective dimension to prognostication as well (Christakis 1999:xvii-xviii). When a patient or a doctor fears a given diagnosis, it is often because of its prognosis, because of what is going to happen. Hence, the meaning of a diagnosis comes from the prognosis. Christakis (1999:31) argues that having a sense of what to expect, getting to know the prognosis, is important for any seriously ill person. This allows them to make the most effective use of emotional, fiscal, and temporal resources and to gain a sense of control over their life. Paradoxically, then, a prognosis is both feared and desired.

Another subjective aspect of prognosis relates to its susceptibility to being shaped. The doctor, the person with the illness or some other actor may choose one intervention over another or no intervention at all, any of which decisions may change the prognosis considerably. This interactional character adds to the complexity of prognostication. Interaction around prognosis may be utilised in several ways by physicians, for example in the interest of 'fostering compliance, cultivating hope, managing expectations, relieving anxiety, and engendering confidence' (Christakis 1999:42).

Hence, the meaning of prognosis is not given. In some cases, as in MND, prognostic certainty may mean losing hope but gaining control. On the other hand, uncertainty may give hope, for instance of a slow progression. An uncertain prognosis from a doctor might also legitimise not knowing what will happen, potentially helping those living with the condition to have a greater acceptance of uncertainty (Christakis 1999:59-60). Thus, another paradox in prognostication relates to ideals about control and hope. In the case of MND accuracy is related to control, which is often desired. Conversely, ambiguity may relate to hope, which also is desirable. The paradox rests in cultivating both hope and control at the same time.

The many paradoxes of prognosis, such as being feared and desired or offering accuracy and control or ambiguity and hope, contribute to the complex meaning-making in the MND context.

3.5 Collabroation

An important condition for this thesis is that the amount and complexity of work that has to be done calls for more than a single individual to perform it. Hence, several people must work together coordinating their tasks and work effort. Coordination of work tasks is fundamental to the health services, and there is a large literature with an array of concepts and operationalisations (see Nolte & McKee 2008, Van Houdt et al. 2013, Lerum & Frich 2012). As an umbrella term I use the concept of collaboration. However, I also draw on more specific theories to operationalise this. Two theorists are of particular importance in this regard: the first is Henry Mintzberg, and his concept of 'coordinating mechanisms', and the second is Jody Gittell, and her concept of 'relational coordination'.

Mintzberg (1989), sees organisations to consist of different types of constellations of different types of coordinating mechanisms. Hence, coordination mechanisms are the build-bricks which constitute an organisation, of which Mintzberg specifies six distinctive types (1989:101): 1) Mutual adjustment and 2) direct supervision are mechanisms that rely on the interaction of individuals, such as talking during work. The main difference between the two is that direct supervision implies a clear hierarchy, while mutual adjustment does not. The four remaining coordinating mechanisms are forms of standardisation. 3) Standardisation of work process coordinates by specifying the work and the tasks of the personnel. 4) Standardisation of output coordinates by specifying the results of the work, ideally leaving it to the personnel involved to specify the work process. 5) Standardisation of skills and of knowledge relate to formalisation of personnel training, hence the need for the personnel performing the work to internalise the coordination. This type of training is typically the way surgeons coordinate work. 6) Standardisation of norms also relates to internalisation, but of values and beliefs rather than of skills.

Jody Gittell (2002) coined the term ‘relational coordination’. She defines relational coordination as ‘the coordination of work through relationships of shared goals, shared knowledge and mutual respect’ (Gittell 2009:13). Coordinating through relationships means coordinating between roles rather than between tasks. With these three ideals in place, participants manage to see beyond their individual effort and to contribute to the overall work process. Hence, instead of implementing coordinating mechanisms that reduce the need for interaction, such as standardisation of work processes, the intention is to facilitate interaction among the participants (Gittell 2002:1408-1409). According to Gittell, relational coordination is meaningful under certain conditions such as task interdependence, high degree of uncertainty and time constraint. If such conditions are lacking, other modes of coordination are likely to be more effective.

4. Methods and data

In this methods section I describe how I planned the project, and how this turned out in practice. In the later chapter “Discussion of methods” I take a retrospective perspective, attempting to assess the process.

4.1 Choice of method

I used qualitative interviews as the tool to generate data for this project. There were a priori, normative and practical reasons for this. An a priori condition was limited prior research about care collaboration for people with MND, especially in Norway. This called for an exploration of practice, and hence, a qualitative approach. My normative position is that the actors in the sub-systems are experienced and skilled at what they do, which is to handle MND. I wanted to understand their world and their lives, so why not ask them? (Kvale & Brinkmann 2009). I wanted to identify and articulate experience-based competence. Further, due to the non-standardised nature of MND there was limited documentation about how MND was handled. Because we were exploring collaboration, many of the activities we expected the participants to talk about would be difficult to observe: care work would take place in different locations, in different subsystems and at different times. Finally, in dealing with death and dying we expected to find lot of emotional and biographical work among the participants and interviews provide a method of directly accessing experiences and meaning (Polkinghorne 1988, Kvale & Brinkmann 2009). Interviewing was a method that was able to provide us with an overview of peoples’ experiences while allowing us to inquire about the informal culture within the sub-systems and to access meaning and experiences among the participants.

There are several methodological approaches to choose from when conducting qualitative research. I committed to Kvale and Brinkman as the standard of reference for the methods chapter. There were three reasons for this: The first is their focus on interviewing, which has been the main mode of inquiry for the thesis. The second is their explicit sensitivity to context when conducting qualitative research. They stress the craft aspect of qualitative research and the concept of phronesis, encouraging the qualitative researcher's continuous commitment to validity and ethical responsibility. Hence, deductive reasoning or formal theories may become insensitive to the relevance of local particularities, for instance when interviewing, when building a purposive sample or when reporting findings. Rather than having a once-and-for-all checklist, the qualitative researcher needs to continuously be attentive to ethics and validity throughout his or her research, from initiation to after the findings are published. The third reason is their non-fixed approach to analysis; I did not want to decide on the analytic approach in advance, but to decide on the analytic strategy when I had more insight into the data in hand.

I have also used other theorists of qualitative methods to support Kvale and Brinkman. In addition to viewing validity as craft (Kvale 1995, Kvale & Brinkmann 2009), I use Malterud's thoughts on reflexivity and validity as norms for discussing these issues. Justesen and Mik-Meyer (2012) stress the importance of a coherent epistemological position in a thesis. I find their focus on epistemology too dominating to use their approach as the core reference in this thesis, but their book was the main reason for my explicit mentioning of being influenced by symbolic interactionism.

4.2 Sample and recruitment

The design of this study and accompanying sample was strongly informed by the community of practice and sub-system theory in MND care (see theory chapter, and also Lerum & Frich 2012). Hence, it was necessary with participants from 1) those living with MND at home, 2) primary health care and 3) hospitals. Within each sub-system the recruitment rationale was to seek variation, and several sampling strategies were used (see Kuper et al. 2008 for overview).

Across these three groups, a total of 53 interviews were conducted with 65 participants. The final sample consisted of eight bereaved caregivers, 17 current caregivers, five persons with MND, 18 health professionals in primary health care and 17 hospital staff. The three groups in the sample are presented in the table below:

Table 1: Overview of interviews

	Pre-project with bereaved caregivers	Hospital X	Hospital Y	Hospital Z	Total
In the home	8	5	5	5	23
Primary health care staff		5	5	5	15
Hospital staff		5	5	5	15
Total	8	15	15	15	53

The large sample size reflects a commitment to exploring health services delivery rather than in-depth knowledge of illness experiences or the perspectives of health professionals. Within each of the three subsystems I sought to vary the background of the participants to capture a wide range of perspectives, information and experiences (Kuper et al. 2008). Preconceived criteria in this regard was to focus on three different hospital catchment areas, participants from rural and urban areas, of varying ages, genders and professional backgrounds. Before I started actively recruiting I understood that MND is rare and I assumed that in many cases I would be forced to make a convenience sample, as there were few people with a relevant MND background.

Defining good collaboration is also a matter of power, of setting one's own definition of what constitutes successful collaboration (Lerum & Frich 2012). This theoretical insight informed the order of the interviews. For each site, I decided to first interviewed caregivers, followed by health professionals in corresponding hospitals and primary health care. In this way I intended to gain knowledge from the family caregivers first, which allowed me to bring these experiences along for the interviews with health professionals, instead of the other way around. I saw the health professionals as the more powerful actors and wanted to use the family caregivers' experiences as leverage in these encounters.

4.2.1 Sampling those at home

I decided to interview bereaved family caregivers, before moving on to interview current family caregivers. I assumed such a pre-project would fine-tune the interview guide and interview technique and give me a sense of the trajectory from beginning to end. There were also ethical reasons for this. I interviewed bereaved family caregivers first because I thought these participants had acquired some distance from the illness trajectory, and that recounting past experiences would be less painful for them than for the active family caregivers who might be in the midst of a chaotic and intense situation.

Health professionals already known to the persons with MND approached the bereaved family caregivers, the persons with MND and the current family caregivers. These health professionals, all MND coordinators in hospitals, briefly explained the purpose of the study and asked for permission to pass on contact information to me. In one case health professionals shaped the sample, where an MND coordinator argued that the potential participant 'was in too poor shape to participate'. I did not pursue to recruit this individual. I provided further information over the telephone, scheduled interviews, and asked for written informed consent when we met face-to-face. I stressed that participation was voluntarily, that confidentiality was guaranteed, and that participants could withdraw from the interview and the research at any time and without explanation.

As there was little prior research about living with MND at home in Norway, I intended to include typical experiences of living with MND and the health services. Following a typical case sampling logic, I wanted to aim for most of the participants to be in the age group where MND was most prevalent. I also tried to keep the gender ratio around 50/50, as well as distribute the participants across rural and urban areas, and seeking variation with regard to educational background. The sample aimed to purposely shed light on family caregivers' experiences with MND in Norway.

Preparing the project I had found less literature on family caregivers in MND compared to persons with MND. Further, I hypothesised that as the condition progressed the family caregiver had to take on increasingly more tasks to keep the everyday afloat. Hence, initially I focused exclusively on family caregivers when sampling. However, as the recruitment carried on, this approach met obstacles. In some cases the family caregiver would not be able to attend the interview, due to heavy care duties. They expressed strong motivation to participate in the research, but if they had to do it without the person with MND they would not be able to. Facing this situation I made two arguments. First, I was interested in the home as system, and those with MND lived there too. Including persons with MND did not fundamentally breach the design of the study. Second, I found it very likely that an important characteristic of a proportion of the population of family caregivers in MND, was to be strongly tied to the person with MND and the commitment to care work. So strongly tied, that participating in research without the person with MND, would exclude their participation. This characteristic, which I was not aware of initially, made it important to include family caregivers who strongly expressed to participate together with the person with MND. This was in line with the principle of seeking variation within the sub-systems.

With regard to the sample of those living with MND at home, I recruited family caregivers and those living with MND through health professionals who already knew them. Additionally, I decided to exclude participants well known in the media or public sphere. There had already been studies of how MND experiences are represented in books and media (Holmøy and Frich 2006). These authors note how some illness experiences portrayed in the media may occur as superior. I found this to be a reason to focus on those who for some reason have not had the opportunity to express their experiences publicly. Further, I had read Knut Dybwik's research project (2011), particularly concerned with LTMV. To some extent his research covered the long term aspect of MND. My perspective was that to live with MND, without LTMV, was the most common experience. Additionally, this was the experience where there was least existing research available in a Norwegian context.

To get a high response rate my strategy was to be flexible as to when and where the interviews were to take place. Hence, I deliberately asked the participants at home about their preferred location for being interviewed. I suggested that the interview could take place at home where they lived, a public place, or at my own office; where ever the participants felt more comfortable. I planned for, and suggested, that the interviews where to last about 45 to 60 minutes.

For the pre-study with bereaved family caregivers, I contacted 10. Two of these were not able to participate, and the final sample consisted of 8 interviews. The two cases of not wanting to participate related to whether they felt it too demanding, and too early to talk about the experience. Among the current family caregivers the response rate was high. All contacted participants in hospital X wanted to participate. In the hospital Y case, one participant did not want participate, expressing a desire not wanting to prioritise research in this hectic time. Another participant from hospital Y did not answer the telephone when I called. All participants connected to hospital Z wanted to participate.

The final sample is presented in the table below, which I provide further comments on in the chapter “Discussion of methods”.

Table 2: Characteristics of persons with MND and caregivers interviewed (n =30)

Group	
Bereaved family caregiver	8
Current family caregiver	17
Persons with MND	5
Age	
20-29	2
30-39	2
40-49	7
50-59	11
60-69	6
70-79	2
Gender	
Male	12
Female	18
Educational status	
> 3 years of higher education	12
< 3 years of higher education	6
No higher education	12

4.2.2 Sampling health professionals participants

At first it was not obvious to me what sort of participants in primary health care who would to a largest extent shed light on MND management within this sub-system. I wanted participants which had actually coordinated services, hence, having responsibilities beyond face-to-face care. Committed to the aim of exploring service collaboration, I decided that an exclusion criterion was if the health care professionals had exclusive face-to-face care experience with people with MND. To identify potential participants with coordination experience, I decided on the inclusion criterion of their informal reputation of handling MND in their local environment. I assumed that this informal inclusion criterion would provide participants in a role where they had to have contact with different institutions within primary health care, and to keep in contact with hospitals and with those at home. This position established them as key participants for understanding care work collaboration. These participants would be in a mediating role between those at home and the primary health care system. The goal was to identify a critical case sample, as the participants would have positional and reputational features that predict them to be especially information-rich and to shed light on issues within primary health care and collaboration (Kuper et al. 2008).

I sought variation by including different municipalities across three hospital catchment areas. I telephoned the main reception of each municipality and briefly explained the research project. After this initial contact I was usually directed to a manager in the health services in the municipality, who then again knew which person with the most MND knowledge in the organisation. All the municipalities and health care professionals I contacted agreed to participate.

To recruit participants from hospitals I got additional permission from hospital management to interview staff. As there are very few professionals involved in MND care in each hospital, the permissions are not in the appendix to ensure confidentiality (see paragraph on ethics). Two of the hospitals in the sample had multidisciplinary MND clinics, where I recruited all the active members of the teams. The teams were based in the neurology departments. In the third hospital, care for people with MND was organised through the general neurology department, but without any clear team structure. Here I decided on snowball sampling to identify the staff with the most MND experience at this hospital. In all three hospitals all health professionals I contacted were willing to participate in research. The participants from the hospitals were the same people who provided services to the family caregivers and persons with MND.

Table 3: Characteristics of health professionals interviewed (N=35)

Primary health care (n = 18)	
Occupational therapist	3
Cancer coordinating nurse	3
Hospice nurse	4
Nurse with leadership and administrative duties	8
Hospital staff (n = 17)	
Neurologist	4
Nutritionist	1
Physiotherapist	3
Social worker	1
Nurse	6
Occupational therapist	1
Pulmonologist	1
Total	35

All in all, the sample is large for a qualitative project. There were several reasons for this. First, to explore conditions for collaboration between family caregivers and health professionals, I wanted to establish conditions that were not particular to a specific site. A large sample allowed identification of several instances of the same phenomenon. Further, an explorative study runs the risk of some interviews being less relevant and of lower quality. Having a higher number of interviews was a way to compensate for this.

4.3 Interviews and transcription

I conducted the interviews in 2011-2012, with interview guides aimed at each subsystem (see appendix). I noticed how the literature strongly warned against using too abstract and researcher driven questions (Kvale and Brinkmann 2009, Braun and Clarke 2006). Even though I was interested in care coordination, a theoretically inspired question like: “Do you coordinate care with coordination mechanisms such as “mutual adjustment” or do you rely on more standardized approaches?” would in the interview situation generate weak data, poorly utilizing the knowledge and vocabulary of the participants. Following Kvale and Brinkmann’s differentiation between “researcher questions” and “interviewer questions” (Kvale and Brinkmann 2009:132) a better question to explore care coordination would be to ask a family caregiver: “How do you contact primary health care?”. This type of question would to a larger extent enable the interview to be an arena for co-exploring what went on within a subsystem managing MND.

I involved the supervisors in the development of the interview guides. They read and discussed several interview guide drafts. The written guides needed to be brief, to be meaningful and accessible in live interview situations. Hence, not all relevant thoughts about the interviews could be written there. To me the backstage talk with the supervisors about the interview guides and interview technique, were as important as the written guides. These talks about interviews and interviewing drilled me in orally discussing the project, which was useful in an academic environment where most of the activity relates to writing text. It was also a way of sharing and agreeing upon the aims and intensions of the overall project, and the purpose of the interviews.

When creating the interview guides the goal was for the guides to be a tool for generating detailed accounts of what happened within each subsystem. I wanted to have concrete and direct questions that every participant in each subsystem would be able to answer. This was important as the recruitment strategy sought participants with varying backgrounds.

For example, I would ask those at home about psychological and health related aspects of their own health, as the condition or case progressed. These were questions that all of them could answer; all were competent to say something about their own health. Further, by asking about changing sleeping patterns, I thought such experiences would create breaches in the flow of everyday, which led to talk about how the everyday was managed at home. How we manage our everyday is often based on tacit and taken for granted knowledge, difficult to talk about in a very standardised or abstract way. Still, it was the everyday I wanted the questions to evoke; for those living with MND at home and those working with MND professionally, alike. Some topics in the interviews were of a sensitive nature. For those at home death and dying could be difficult to talk about. For health professionals I assumed they could seek to avoid talking about their own work in negative ways. One, among several tactics in this regard, was to ask indirect questions. For instance, using third person, “How do you think others manage, this or that?” and then consider whether to directly ask “How do you do it?”

I wanted to facilitate a conversation which allowed the participants room to talk about issues I had not thought about myself in advance. I wanted the questions to be aimed in such a way that the participants would be able to talk about what they, themselves, saw as the most important issues. What mattered for the family caregivers?

Hence, while planning the interviews my intention was to get as detailed an account as possible about everyday aspects in MND care and management. I assumed that this interview approach would lead to rich data with ample potential for many different forms of analyses; not rigidly binding me to a particular theoretical position or a strict predefined research question.

4.3.1 Interviewing those at home

In advance I assumed the face-to-face interviews with persons with MND and family caregivers would be a meaningful, but challenging task in the course of the project. MND is a devastating experience, and this was reflected in the interviews. I prepared the following opening of an interview with someone at home: Introducing the interview, I specified that we were to talk about MND, from the first time they noticed anything strange, to their encounters with the health services and what had happened up until the day of the interview. Additionally I wanted to discuss thoughts about the future. I decided to have the interview guide in my lap and explained that it contained an overview of the themes the project was meant to cover. I pointed out that in most cases those topics were thoroughly covered in the conversation without my having to use the guide as anything more than a checklist. This combination of telling about the trajectory chronologically, combined with a semi-structured checklist was very similar and inspired by the strategy Locock and colleagues used to explore MND (Locock et al. 2009). I finally asked if the participant wanted to be interviewed and reminded them that they could decline to be interviewed without giving a reason.

As several of the current caregivers had difficulty leaving their home due to care work, most of those interviews took place in the home; some, however, preferred public places. I planned for only one interview per day; to have plenty of time should unforeseen things happen. This turned out to be a good strategy, as some interviews taking place at home lasted for several hours and included lunch and snacks. Due to long travel time, in some cases I was forced to make exceptions of the rule of one interview per day, for practical reasons. In a few occasions I had two interviews in course of a day. Most interviews lasted about 45 minutes.

In the interviews the interviewee sometimes compared the interview to other more familiar situations. Some saw me as an expert, like a health professional, and asked questions about MND. In most cases I argued that health professionals would have better answers than me, and I specified who they should ask. Others were frustrated about services and vented this frustration. In most cases I allowed for such stories to unfold without interruption. The nature of the topic could make the research interview resemble a therapeutic session. I have neither the training nor the knowledge to be a therapist; nor do I have the capacity to follow up a therapeutic relationship. Following Fog (2004), it was important for me not to pose as a therapist, and I continuously tried to critically self-reflect upon my role during interviews. Consequently, on one occasion I had a telephone conversation with a participant after an interview, and we agreed that the participant should contacted the health services to get further follow-up.

My background allowed me to clearly communicate that I was not in a position to influence access to health services, and that I could ensure anonymity. It was not uncommon for the participants to ask me if I had any personal connection to MND. I answered that I had neither close friends nor relatives currently living with or having lived with the condition. It is possible that my lack of personal ties to MND gave the participants room to define what they saw as important and what was at stake.

4.3.2 Interviewing health professionals

My approach to the interview with health professionals had several similarities to interviewing those at home. During the interview talk, I aimed the questions in the same way, and my ideal was to generate talk about the everyday at the work place. Generally, I wanted to explore how MND was managed, from the respective health professional’s point of view. When preparing for interviews with health professionals I had a more theme- on not so much a chronological based approach, compared to the interviews with those at home.

The few open conflicts that appeared in my material mainly had to do with the relationship between those living with MND and primary health care. This was reflected in the interviews with health professionals in primary health care. Some of these participants used the research interview as an opportunity to vent frustration. Others were cautious, explicitly noting that they withheld certain details. Some participants obviously knew that conflicts did occur and were happy to note that this was not the case in their location. For some participants my research interview seemed to represent an opportunity to promote the importance of the work they did. For instance, at one hospice I had scheduled an interview with the manager. When I arrived the entire staff was physically present in her office and was included in the sample. The one-to-one interview became a focus group. This is in contrast to another location where I discussed the project at length with a leader, who then assigned and scheduled the interview with a particular nurse who coordinated care.

4.3.3 Transcription

Transcribing is hard and time-consuming work. I wrote 1,450 pages with 1.5 line spacing using 12-point Times New Roman font. When transcribing I slowed down the audio tapes, which allowed my writing to keep up with the conversation. In the particular software I used, the slower speed also lowered the pitch of the voices, making them sound darker and sadder. It was at times painful to sit hour after hour transcribing in detail the suffering experienced by some of the family caregivers. For instance, due to the lowered speed, the natural pauses in the conversations were artificially prolonged. A one-second pause live became roughly a 1.5-second pause in the recording. This sounds trivial, but it changed the social meaning of the pause, making the recording sound gloomier than it actually was in the interview. After some time I got more used to this effect and I also listened to the recording at the true speed to check that I was interpreting the tone of the interview correctly. Despite these challenges, there were also benefits to transcribing the interviews myself. I found it particularly helpful to write analytic memos in parallel with the transcription, which also served as a meaningful break from the transcription work.

I transcribed the interviews shortly after they took place. However, Kvale and Brinkmann note that moving from oral to written language is an interpretative act that raises both practical and principal questions (Kvale & Brinkmann 2009:177), which need to be addressed. I used a fairly strict verbatim style. I did not correct wording that sounded good orally but awkward in writing. I included pauses as well as the many hesitant beginnings and unfinished sentences where the participants were unsure of what to say. Given my analysis' focus on meaning rather than linguistics, the transcripts were perhaps too close to oral language. The verbatim style would perhaps be confusing to other researchers who were not there during the interviews. Because I conducted the interviews and wrote the transcripts, the transcriptions provided me with cues to remember the atmosphere and emotion of each interview. The transcription style helped me re-contextualise the data back to the dialogic face-to-face interview.

There was one important systematic exception to my commitment to writing exactly what was said. Kvale and Brinkmann encourage using transcripts as a way to improve interview technique (Kvale & Brinkmann 2009:89-90), which I did. When following this advice I found my interviews to have a better flow when I used phatic and non-verbal cues to encourage communication, including saying 'mhm' and nodding my head. These cues did not obstruct the oral conversation, quite the contrary. In writing, however, my almost constant use of cues appeared confusing, apparently interrupting several times per sentence. In the written transcripts I generally omitted these cues.

4.4 Analysis

In this section I will present an overview of the methodological principles I chose to lean on during the analysis in the three papers. I will then turn to a detailed account of the analytic process of each paper.

4.4.1 General conditions for analysing qualitative data

Several authors argue that there is no single “right” approach to qualitative analysis (Coffey and Athkinson 1996, Kvale and Brinkmann 2009). However, to tailor the analytic strategy to my data I reviewed some central authors in qualitative analysis (Coffey and Athkinson 1996, Kvale and Brinkmann 2009, Malterud 2001a, 2001b, 2011, 2012, Tjora 2012, Braun and Clarke 2006, Corbin and Strauss 1990 and Saldana 2009). A common trait in this literature is for the researcher to write and code text committed to the detail of the data, to avoid for the researcher having to rely on personal memory to account for the data. Another, and complementing trait is also to write texts committed to the ‘analyst narrative’ of the research. This is the narrative that compellingly illustrates the story the researcher is telling about the data, which goes beyond description and makes an argument in relation to the research question (Braun and Clarke 2006:93). The analyst narrative is to the core of what qualitative analysis is. Braun and Clarke (2006) goes so far as to argue that if the themes, categories and questions asked in the interview are very similar to the presentation of results in the final paper, this is a sign of weak analysis by the researcher: “Some of the worst examples of ‘thematic’ analysis we have read have simply used the questions put to participants as the ‘themes’ identified in the ‘analysis’ – although in such instance, no analysis has really been done at all!” (Braun and Clarke 2006:85-86).

An important point of departure for my analysis was the amount of data. Having 1 450 pages of transcript I worried that to initially code all of the material, as suggested in systematic text-condensation (Malterud 2012) and in thematic analysis (Braun and Clarke 2006), would take too long and that the material was too large to code it in a thorough manner. This would weaken the validity of the analysis. Malterud underscores the importance of not having a too large data material, warning that this undermines the researcher’s ability to do a proper analysis (Malterud 2011).

Facing the large amount of data I needed an approach which allowed to focus on specific parts of the data, and legitimately define large proportions of the data as less relevant for the analysis. At this point I did find Braun and Clarkes discussion (2006) useful. They argue to separate between a bottum up strategy or inductive analysis, on the one hand, where analysis is in large part driven by the data. And on the other hand, deductive or theoretical informed analysis driven by the researcher's theoretical or analytic interest (Braun and Clarke 2006). Most qualitative data anlyses are somewhere in between these two positions. Still, in this project theory had come to play a decicive role in the analytic work. That is, I used theory to narrow down my focus, and to provide a more detailed analysis of certain aspects across the data.

This being said, I have simultaneously been aware of the dangers of anecdotalism in qualitative analysis, what Silverman describes as: "…where research reports appear to tell entertaining stories or anecdotes but fail to convince the reader of their scientific credibility" (Silverman 2010:432). Braun and Clarke also warn against anecdotalism as: " … where one or a few instance of a phenomenon are reified into a pattern or theme, when it or they are actually idiosyncratic" (Braun and Clarke 2006:95).

In brief, anecdotalism involes an analytic strategy of "cherry picking"; to include excerpts of data which support the research question or claim, and to leave out data which do not. This is always a danger dealing with large qualitative data sets. The data are so rich in detail that excerpts to support a claim may always be found. Hence, a theoretically driven analysis always needs to have 'checks and balances' not to degenerate into anecdotalism. What, then, may work as an antidote against such wrongful interpretation? Several authors underscore the importance of "playing the devil's advocate", attempting to falsify and suggest alternative interpretations of claims (Kvale and Brinkmann 2009:238), to examining deviant cases in the data (Silverman 2010:281-282). Coffey and Athkinson also describe the strategy of actively seeking out "negative" findings as a common method to seek validation in qualitative data analsysis (Coffey and Athkinson 1996:46). I will account for how I tried to balance between theory and data for each paper below.

Finally, in the outset of an analysis the claim or question which leads on the analytic work might be weakly grounded or plain out wrong. Silverman points out that in a qualitative research project the researcher cannot know if the design and research question is a good fit to the data, before analysis has started (Silverman 2010:212-222). He goes so far as to argue that sticking with an original idea, design or approach to the project, may "…be a sign of inadequate data analysis rather tha demonstrating a welcome consistency" (Silverman 2010:222). Hence, willingness to adjust the research question or analytic approach during analysis is, if well informed, something which may strengthen validity.

4.4.2 The three papers

Having completed the interviews, I sketched a plan with three article ideas, which I presented and discussed with the supervisors. The first question was 'What is MND?' (Paper I). The second question was 'How to manage MND?' (Paper II). The third question related to conflicts appearing in the primary health care data, with the theme: 'The home-health service boundary' (Paper III).

Paper I

In the final and published paper I, I present much of the theory which guided the analytic process, but there was little room to elaborate on the procedural aspects of the analysis. I compensate for this in the following.

In the fall of 2012 I had completed most of the interviews, and was eager to work throughout the data. At the time, I attended the British Sociological Association's conference in medical sociology 'Medsoc' which took place in Leicester, that year. During the conference I sat in on a presentation about planning a good death. While I listened (to Kristian Pollock) I thought that in the MND case it would be difficult to plan a good death, because you always could consider initiating LTMV, postponing the process indefinitely. If so, the intent of the situation would no longer be to die. What would happen then?

On that connection I made up the term "chronimal", as in combining *chroni*c and ter*minal*. In a text I have dated 09.09.2012, and I emailed to the supervisors, was a very early formulation of what later became the concept "unstable terminality":

> *The claim is that the tension between urgency (terminal illness) and routine (chronic illness) represents something new; chronimal illness poses challenges separate or independent from the other two understandings of illness.*

In the ensuing meeting with the supervisors they were supportive of the idea, and encouraged me to pursue it further. I had begun the analysis on a relative small part of the data. I had formulated a claim, that MND represented something new beyond chronic and terminal, and unclear tension between the two. The following analytic work was similar to what Silverman suggested; I discussed my theoretical claim against a steadily increasing part of the data (Silverman 2010:280).

An important drive in the work to apply the concept on more data was the supervisors, and Trygve Holmøy, the last co-author. Throughout the analytic process I regularly presented drafts to them which accounted for more and more of the data. I account for this process in the paragraph below.

I was intrigued by the initial concept of "chronimality", I saw it to highlight a social constructivist principle: that what is defined as real, become real in their consequences. Initially, the idea had been generated from data from family caregivers, but what about the other sub-systems? Could they also change the definition of what MND represented? I discussed what the overarching research question for this analysis should be with the supervisors, and I proposed "What is MND?". This question was inspired by the book Body Multiple (Mol 2002), which explores the condition atherosclerosis, and how it is understood in different ways by different health professionals. I thought the same might was the case in MND. Another theoretical underpinning for that question was Andrew Abbott (Abbott 1988), I wanted to find out if different professions tried to view or define MND in particular ways, presumably relevant for their own jurisdiction.

I decided to write a text where I presented excerpts from the transcripts with direct and indirect descriptions of MND in the interviews with family caregivers, persons with MND and health professionals. As I read the transcripts I focused on different "versions" of MND, such as functional or practical challenge, existential challenge, respiratory challenge or nutritional challenge. I expanded this list as I read and wrote my way through the transcripts. Besides focusing on different versions of MND I also paid attention to differences in the sense of time and progression of MND expressed in the transcripts.

I wrote this using Word. Each heading in the document was a code identifying a participant. I actively used the "navigation pane" functionality in Word, and the headings were easily available in the left margin. Below each heading the text followed the form of a findings section in an article. I presented a quote, and under it I commented on what MND was in the excerpt above. When writing I consulted Saldana's "The Coding Manual for Qualitative researchers" (2009). Two types of coding techniques guided the process. Saldana describes "holistic coding" (Saldana 2009:118-120), as an approach where there is no maximum length restriction for the data excerpt, relevant for labelling vignettes or episodes in the data. Further, he points out that this is an applicable approach when the researcher already has a general idea of what to investigate in the data, and a time-saving method facing massive amounts of data. This form of coding fitted well with my theoretically driven analysis (I had a general idea of what to investigate), and my data material was large. The other was "versus coding" (Saldana 2009:93-97), as I read the transcripts I tried to identify and juxtapose inconsistencies and contradictions (such as MND being fast or slow, urgent or routine). These two techniques led my attention to longer excerpts of data, and excerpts containing a contradiction or inconsistency.

The writing in this analytic phase would be labelled as interpretative or latent analysis by Braun and Clarke (2006), in the sense that I focused on underlying ideas and assumptions in the data. The writing, such as the use of holistic and versus-coding techniques, was strongly theoretically informed. I had reviewed some of the qualitative literature on how people with MND experience the condition (Locock et al 2009, Locock and Brown 2010, Locock et al 2012, Mazanderani et al. 2012), where persons with MND appeared to have shifting and ambivalent responses to the impact of the condition. My analysis entailed to look for such ambivalence also among family caregivers and health professionals in the MND context.

This text described above served as a resource base for another more condensed text I presented to the supervisors. In this condensed text I aimed to account for the variation in the data which I had analysed thus far. Hence, I wrote the underlying analytic work, while the supervisors read a condensed version of this, typically between 20 and 30 pages. This may be seen as a parallel process where I wrote one text exploring the details in each interview, and another summarizing this.

In one of the supervisor meetings we agreed to adjust the research question, instead of broadly account for “What is MND?”, we decided to narrow down the focus to account for one core concept, what after several revisions became unstable terminality. One reason for this was that I discovered that Abbott’s theory of professions (Abbott 1988) was more appropriately applied on a macro or institutional level than to the day-to-day interactions in my data. This meant that coding I had done focusing on different “versions” of MND became less central, while the coding I had done focusing on expressions of time and progression in MND became the centre of attention. Contributing factors to the adjustment of the research question was discussions with the supervisors, as well as the arrival of the final co-author of the paper, Trygve Holmøy. He was involved in the process when the concept was fairly well established, and it was an important drive for the analysis that he, as an active neurologist, recognized it from a clinical perspective. The adjustment of the research questions meant that the continuing analytic work had two aims, to account for the boundaries and consequences of the concept.

Having a set theoretical perspective, such as the concept unstable terminality, the analysis runs the risk of the researcher only notices those aspects in the data that can be seen through their theoretical lens (Kvale and Brinkmann 2009:238). Hence, it was necessary to avoid such theoretically shaped anecdotalism. For instance, the versus-coding technique could lead to an over-focus on inconsistencies. Therefore, I actively sought out participants and excerpts of data that did not contain tensions or inconsistencies. For instance, after the aim was adjusted I went back to the original transcripts, especially working with interviews which initially had I seen to portray little tensions. This was a way to seek negative examples of unstable terminality. Importantly, such negative examples are seldom considered a problem in qualitative analysis. Seeking out deviant examples and describing them may enhance the analysis, capturing and illustrating to a larger extent the variation in the data (Silverman 2010:281-282). In this analysis deviant examples were important to show the boundaries of the concept in question. Variation in the data material, then, is not to be seen as a disturbance in qualitative analysis, but rather, when accounted for, offers further insights in the social phenomenon in question.

During the analysis I came across several negative examples of the concept, which I account for in the final paper. For instance, most of the interviews at home described MND as a devastating and disruptive event. However, there was variation to the degree of disruption, and a few hardly talked about disruptive experiences at all. The presentation of how those at home manage MND contains a continuum from more to less disruptive experiences. The quotes I presented in the final paper are deliberately picked to illustrate these varying positions. These quotes are examples for my analyst narrative committed to account for the variation in the material. It is the narrative, not the quotes, which defines how to understand variation in the material, from a strongly disruptive experience and being stuck in limbo as a consequence of unstable terminality, towards less disruptive experiences, almost to the extent of biographical continuity or flow.

Importantly, the number of quotes or participants experiencing different types of disruptive experiences is not essential here. To illustrate the variation in the material it is necessary to identify the most radical positions. These are often uncommon in the material, occurring in few instances. Hence, for me to be able to articulate an analyst narrative accounting for variation, I had to make an interpretive act where I found the uncommon positions in the material.

Paper II

The research question leading the way in paper II was "How to manage MND?". I started coding for "coordination work" of the different ways the participants articulated the coordination between the home and the health services, primary health care and hospitals, alike. This was strongly inspired by Strauss's concept of articulation work; the coordination of all the different types of tasks and work which shapes a trajectory (Strauss et al 1985:151). The ambition, then, was to develop subtypes of articulation work across sub-systems. Strauss's original work took place solely within a hospital (Strauss et al. 1985), and Davina Allen and colleagues (Allen et al. 2004) had critizised Strauss's work as lacking theoretical conceptualization of what happened outside the hospital in illness trajectories. I imagined these subtypes to resemble Mintzberg's coordination mechanisms (Mintzberg 1989:101, see theory chapter), and categorical work (Bowker and Star 2000:310), the articulation work of meaning rather than practical tasks.

I used these theoretical lenses to write a text with direct and indirect descriptions of how those at home coordinated services, and I planned to go on with this exercise with the data from primary health care and hospitals, consecutively. However, this strategy did not resonate well with the data. I, again, aimed to write one text accounting for details in the data, and another summarizing this to present to the supervisors. I regularly met with the supervisors, and I struggled to present a summarized version of my analytic work to them. They helped me to change analytic strategy. The problem I had encountered, which I presented and solved together with the supervisors, is presented below.

The theoretically informed holistic coding approach I had used previously did not, in this case, separate between relevant and irrelevant excerpts of transcripts in an effective way. Far too large proportions of each interview were relevant. I changed tactics to get an overview of the material. Instead of coding actual excerpts I read through each interview and wrote a summary in my own words, using fewer actual data excerpts. Each summary was focused on the family caregiver's relationship towards: 1) The person with MND, 2) primary health care, 3) the hospital and 4) other activities not directly related to MND. Having conducted these exercises I consulted the supervisors. While the initial idea was to explore coordination of care across sub-systems, we decided to adjust the focus to focus on what went on within the subsystem of those at home. The rationale behind this decision was that the question of "How to manage MND?" needed far closer analysis of the data, and it was necessary to limit the amount of data to analyse to ensure validity.

Narrowing down the aim towards the home, this thematic was strikingly similar to the book "Unending work and care: managing chronic illness at home" (Corbin and Strauss 1988), where Corbin and Strauss conceptualize the management of chronic illness along three lines of work. These being illness work, biographical work and everyday work. I wanted to point to the specific types of work in the MND case. This was the reverse ambition of Corbin and Strauss. They sought out people across and array of different diagnoses to see what was common across diagnoses.

Drawing on Silverman (2010:280), I made a detailed analysis of four interviews, and developed seven lines of work based on them. I then applied these to the rest of the material, writing two texts in parallel. In one text I coded for the different lines of work, seeking confirmation and disconfirmation of the claims I had made, actively seeking deviant examples. While I increased the amount of data, I also wrote on the lines of work themselves, trying to reflect the variation and the detail in the underlying coding. In a similar fashion as in paper I, I discussed this condensed version with the supervisors. In developing the typology of lines of work, each line had to have sufficient internal homogeneity, as well as external heterogeneity (Kluge 2000). The underlying coding led to several revisions of the lines of work, and the final paper suggests five lines of work in the MND context.

Presenting the data in the final paper I chose excerpts of data which vividly illustrated the line of work in question, and from different participants showing the breadth of the material.

Paper III

While underlying ideas for the first two papers were theoretically driven, the third paper was initiated by issues occurring in the data. I had expected the health professionals to be relatively unaffected in their talk about how they managed MND (Hughes 1958:81-84), and most health professionals were. In some interviews, however, this was not the case. These health care professionals stood out as they appeared desperate and emotionally tolled by the situation. These interviews entailed conflicts about the relationship between those living at home with MND and primary health care. Hence, the aim leading this analysis was to explore the relationship between those at home and primary health care, from a primary health care perspective. Contrary to the other two papers which focused on all three sub-systems from the outset, paper III related to primary health care exclusively, from the start.

To explore this issue, I started to write down the timelines of the MND trajectories in the interviews where I had identified conflict. These interviews contained descriptions of one or a few persons with MND and their relatives. I then moved on to write timelines of the other interviews, which contained one or two MND trajectories, but where there was no explicit conflict. Finally, I wrote timelines of the MND cases from the interviews with participants having managed multiple MND cases. Among these participants some of the MND cases were very briefly described, not providing the same level of detail as in the interviews with only a few MND cases.

I wanted to find similarities and differences of the MND cases across the participants. I used the previous writing where the MND trajectories were accounted for, to write summaries of each participant. These summaries were headed ‘general background’, ‘types of relationship in the story’ (between those at home and primary health care) and ‘conditions for the relationship’. As I explored the types of and conditions for relationships in the timelines, I wrote a text in parallel where I tried to account for the variation in these relationships with a typology. For this typology I used vocabulary from narrative theory (Polkinghorne 1988, Frank 1995). I presented six typical narratives MND represented for primary health care professionals: 1) *The casual narrative* was the underlying default understanding of any case in primary care, focusing on standardized coordination and cost containment (this was uncommon in MND and mainly used for contrast). 2) *The exceptional narrative* presented MND as a great challenge in need of utilizing the maximum potential of the primary care system, and the health professionals succeeded in this. 3) *The cautious narrative* was when health care professionals thought that something unfortunate would happen in the near future, and there was need to be on alert for sudden turn of events. 4) *The breakdown narrative* entailed insurmountable problems with care provision. 5) *The entrepreneurial narrative* was when MND was an argument for creating system change in primary health care, for instance building new institutions. 6) *The advanced routine narrative* was when primary health care provided services which were unimaginable and far more advanced compared to the past. Overall, I thought these types of narratives to illustrate how MND was on the verge of Norwegian primary health care’s capacity. Further, how some municipalities, perhaps related to the Coordination Reform, had managed to build new institutions to handle more advanced care tasks.

As with the other papers, I regularly discussed the developments in the analysis with the supervisors. As the work progressed, it became clear that suggestion for a typology had several short-comings and needed further refinement. For instance, the stories were not mutually excluding. On a more detailed level it was not obvious that this was a narrative typology, and perhaps better understood as “scenarios” or “cases”. Together with the supervisors, we decided to narrow down the aim for the analysis. Instead of accounting for the relationship between those at home and primary health care in general, we decided to focus on challenges occurring in this relationship. Hence, the new aim was to explore what challenges health professionals encounter when managing MND in primary health care. I then coded for challenges appearing across the summaries of each participant and the typology of narratives. Additionally, based on the narratives I went back to check the original transcripts. This resulted in a new manuscript, more similar to the final paper, which then was revised several times in dialogue with the supervisors. The quotes presented in the final paper were chosen because they occurred in several cases and in several places and participants underscored that they were distressing.

4.5 Translation

I generally wrote all texts in English. The transcripts were in Norwegian and were not translated until late in the process, close to submitting a paper. I did this to remain committed to the original intention of the data, not translating until necessary.

4.6 Ethics

The study was submitted to the Regional Committees for Medical and Health Research Ethics in Norway (ref: 2010/3334). Because the project focuses on care management and health services rather than health and disease directly, they found the project exempt from review. The project was approved by the Norwegian Social Science Data Services (ref: 26498/3/KS).

In qualitative and explorative projects, ethical responsibility follows the researcher throughout the entire project, and continues after publication (Widerberg 2001, Kvale & Brinkmann 2009). Hence, due to the phronetic nature of ethics, most of my ethical considerations are integrated into the different sections of the summary rather than being addressed separately in this section. Still, some issues should be explicitly mentioned here.

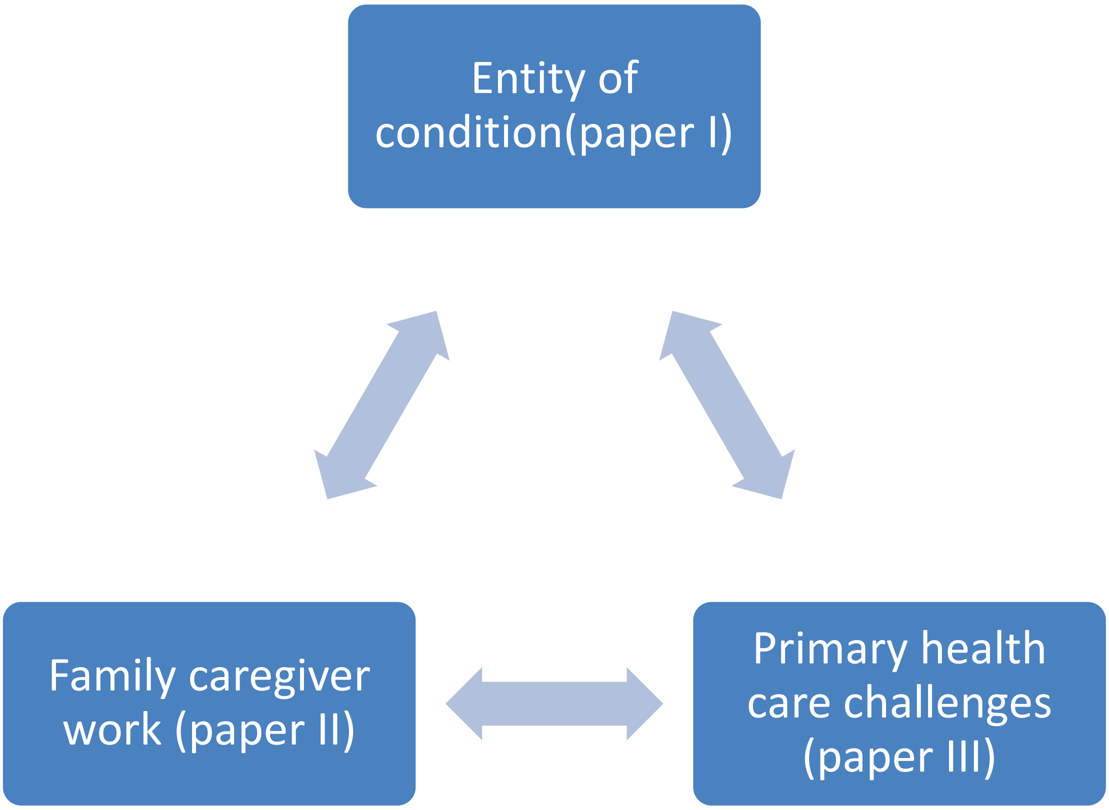
To ensure confidentiality and anonymity I have altered details about the participants, and some information has been omitted. During the interview process, my impression was that some of the participants saw this research project as a long-awaited opportunity to broadcast their experiences. Some made comments about contact they had with local newspapers or on social media. On the other hand, some participants had stories they were less willing to share, for instance relating to disagreement within the family about how to manage MND, and anonymity was a condition for their participation.

Due to the small size of the MND community of practice, there is an issue of indirect recognition of the participants. For instance, specifying which hospitals were included or identifying some of the participants could lead to indirect recognition of other participants. Family caregivers and some primary health care staff especially underscored the importance of confidentiality. Anonymity was also a formal condition in all participants’ written informed consent (see appendix).

5. The findings

The findings in this PhD consist of three papers. Paper I is based on data from all the interviews in the study and explores the meaning of MND. Paper II relies on interviews with family caregivers and focuses their care work. Paper III explores challenges in care organisation for MND in primary health care, drawing on interviews with primary health care staff. The focus of each paper is illustrated in the figure below, and a summary of each paper follows.

Figure 2: The relationship between the three papers



5.1 Paper I

Lerum, Sverre Vigeland, Kari Nyheim Solbrække, Trygve Holmøy and Jan C. Frich (2015). 'Unstable terminality: negotiating the meaning of chronicity and terminality in motor neurone disease.' Sociology of Health & Illness 37(1): 81-96.

This paper explores the meaning of two common concepts: chronicity and terminality, understood respectively as an indefinite or definite closure of an illness trajectory. As a consequence of improved health services and medical technology the distinction between chronicity and terminality has become increasingly vague and is sometimes ambiguous.

The study draws on 53 qualitative interviews with 65 participants who are persons with MND, family caregivers, primary health care staff and staff in multidisciplinary MND clinics in hospitals. In the interviews the participants emphasised chronic and terminal aspects in subtly different ways along the entire illness trajectory and when recounting the trajectory in retrospect. Our main finding, then, was that the meaning of MND is unstable in both time and space.

We suggest the concept *unstable terminality* to describe this ambiguity. While MND is a fatal diagnosis, it can be contested, and contingencies and interventions can create an indefinite timeline. Hospitals tackled the instability with pre-scheduled consultations allowing them to avoid giving an explicit prognosis. Although this system gives the hospitals considerable robustness, a potential weakness is the tendency to lean too heavily on this passive prognosis and provide persons with MND and their caregivers with less information about the long-term outlook. Care needs were changing and personalised, and in primary health care skilled and experienced personnel needed specific training for each case. The ability to procure proper equipment and to train personnel in primary health care was dependent on prognostic information, which was required to organise their efforts and was challenged by the unpredictability in unstable terminality. When receiving the diagnosis persons with MND and family caregivers often understood MND as a terminal condition, there was no cure. 'Galloping' was a metaphor used several times by family caregivers, comparing MND to a wild horse, strong, fast and difficult to control. However, if the trajectory prolonged, the situation became more stable, characterised by enduring and vast amounts of care work. Hence, there was a shift of focus towards maintaining routines, eating health foods and getting enough sleep and exercise. These were typical features of chronic rather than a terminal illness. Even though the care work stabilised in this way, the person with MND became gradually weaker, so terminality persisted. Some family caregivers experienced what we understand as living with chronic and terminal illness simultaneously, which made it more challenging to find sustainable strategies for family caregivers.

5.2 Paper II

Lerum, Sverre Vigeland, Kari Nyheim Solbrække and Jan C. Frich (2016), 'Family caregivers' accounts of caring for a family member with motor neurone disease in Norway: a qualitative study' BMC Palliative Care 15:22

This paper explores family caregivers' accounts of caring for a family member with MND, drawing on 23 interviews with 25 participants, including 17 current and eight bereaved family caregivers. MND care is demanding, complex, and involves a variety of care tasks. Family members may experience significant and enduring strain.

We found that caregivers were engaged in five lines of care work that were often parallel or closely interconnected: i) Immediate care work was work in physical proximity to the person in need of care, to replace of functionality and a sense of safety; ii) Seeking information and clarity about the disease was work to handle information about the condition, prognostic outlook and ways to manage the situation; iii) Managing competing obligations was work to accommodate other obligations than those related to MND; iv) Maintaining normality was work to provide a sense of normality, such as to remain in paid employment or sustain a social life; v) Managing external resources and assistance was work to handle and incorporate nurse assistants, friends and family and assistive devices at home. Some of the lines of work could start before a diagnosis was given and carry on throughout the illness trajectory.

The total work load of the lines of work could become overwhelming, and striking a balance between them was a main project for caregivers. Care work had a symbolic and moral meaning for caregivers, and was associated with self-worth and respect from others. We found that family caregivers seem to compromise their own needs in everyday life while caring for a family member. With the potential to perform care work day and night, caregivers had to continuously balance their own and others' expectations without being overwhelmed by care work. How family caregivers prioritised different lines of work changed continuously during the course of the trajectory. Their priorities were shaped by their interactions with the person with MND, available assistive devices, the development of the condition, and utilisation of paid care.

Disagreement about work could draw on time and energy for family caregivers. Being in a changing and potentially chaotic situation may compromise family caregivers’ capacity to utilise supportive services. Using the lines of work as a framework to assess caregivers’ preferences and priorities, health professionals can tailor assistance and support to family members caring for persons with MND.

5.3 Paper III

Lerum, Sverre Vigeland, Kari Nyheim Solbrække, Jan C. Frich (submitted manuscript), 'Health care professionals' accounts of challenges in managing motor neurone disease in primary health care: a qualitative study'

Motor neurone disease (MND) is a progressive neurological disease causing muscle wasting, gradual paralysis, and respiratory failure, with a life expectancy of 2–4 years. In order to better understand how MND is managed in the community, we conducted a qualitative study to explore the challenges health care professionals encounter when managing MND in primary health care.

Based on data from 15 semi-structured interviews with primary health care professionals in Norway, we found that MND is viewed as a condition that requires exceptional effort and detailed planning.

Health care professionals reported five main challenges in managing MND in primary health care: 1) building relationships with those giving and receiving care in the home; 2) preventing caregiver burnout and breakdown; 3) providing tailored care; 4) ensuring good working conditions in patients' homes; and 5) recruiting and retaining qualified nursing assistants. Health care professionals reported needing working conditions that allow them to tailor their approach to the personal, emotional, and existential nature of care preferences of those living with MND. However, people with MND and their families were sometimes perceived by health care professionals to prefer a strictly task-focused relationship with care providers. Such relationships limited the health care professionals' control over the MND trajectory and their capacity to prevent family caregiver burnout and breakdown. Adequate resources, along with training and support of nursing assistants, may increase the continuity of nursing assistants. Responsiveness to patient and family needs may enhance collaboration and promote tailored primary care and support for patients with MND and their families.

6. Discussion of methods

In this chapter I look back on the project and the methodological choices I made, as accounted for in the previous chapter on methods and data.

6.1 Reflexivity

In line with the interactionist approach my point of departure is that we, through our experience and cultural competence, are able to communicate, interpret and understand others. Kvale and Brinkmann note that the researcher as a person, and the accompanying social and cultural competence, create a most sensitive instrument to investigate meaning (2009:171). What Malterud (2001b) labels reflexivity in qualitative research is not achieved by eliminating – or worse, ignoring – researcher effect, but by accounting for and sharing aspects of this.

6.1.1 Interviewing those at home

Age shaped the interviews. At 28-29 years old at the time of the interviews, I was considerably younger than most of the people I interviewed. From a life-course perspective it is obvious that there were several phases of life that we did not share. Most participants were in their 50s and 60s. Several participants mentioned that they had looked forward to sharing retirement together, or pointed out that it was not long ago their children had moved out of their home. While I obviously understood these experiences, I think that we did not dwell on these topics to the same extent as if I had been the same age and had had similar experiences.

Like age, gender norms shape illness experiences (Ahlsen 2013), and gender creates expectations. In a loosely structured interview setting such expectations colour the content. For instance, I did not plan to ask about sexuality in the interview guide, and this was not something I actively pursued in the interviews, as I felt it would be intruding when I had not signalled this as a central thematic in advance or when obtaining informed consent. Still, some of the men in the sample commented on their loss of a sex life, while none of the women touched upon this theme. My impression was that I, as a man, to some extent was expected to be more interested in technical aspects of care than a female interviewer might be, and to minimise small talk. Perhaps with a female interviewer the threshold for initiating explorations of intimate matters between spouses would be lower. I do not think such gender expectations are absolute or unchallengeable. In some interviews I downplayed the importance of technology, if that theme appeared overly dominant to me.

When planning the interviews, I assumed the bereaved caregivers would be in a less vulnerable situation than the current caregivers. The interviews with bereaved caregivers were meant to serve the purpose of and allowing me to gain both insight of the topic and interview experience. This was, in my view, a satisfactory strategy, with some exceptions. In retrospect, some of the interviews with bereaved family caregivers seemed more emotionally intense than many of the interviews with the current caregivers. There might be several explanations for this. I might have gotten used to hearing dramatic stories over time. Further, when recounting the caregiving experience, bereaved caregivers might be more likely to remember dramatic events that stood out. In my estimation, the interviews with the current family caregivers often had more detail about non-dramatic everyday events to create meaning here and now. Additionally, exhaustion occurs among family caregivers, and the last few months of an MND trajectory are particularly straining (Aoun et al. 2013). Exhaustion and intense periods during the trajectory may exclude research participation. However, family caregivers might feel that it is important to recount such experiences in retrospect, in which case a research interview might serve as an opportunity for reflection.

I experienced a peculiar phenomenon when encountering suffering in the interviews. The stronger the suffering, the larger the difference this PhD could make seemed to me. For me, this phenomenon was a way to contain and enter suffering in the interviews, as well as to create meaning of suffering in the research project as a whole. This way of making sense of suffering was also mentioned by the participants as a motivation for their participation. I think this view of suffering, and my willingness to enter it shaped the interviews. There is, however, no need for suffering to be meaningful; illness may also be nothing but chaotic (Frank 1995). Meaninglessness may also be intensely present in biographical abruption (Locock et al. 2009). My impression was that through the questions I asked and the interest in the conversation that I attempted to show, I appeared as a person who was willing to listen to uncomfortable stories. My general impression of the interviews was that they were meaningful events for me and the participants, and the project was an opportunity for their experience to make a difference for others.

6.1.2 Interviewing health professionals

In the interviews with health professionals, professional background was a prominent factor, making age and gender less explicitly so.

Due to a misunderstanding at the start of one interview, a health professional at a hospital assumed that I was a physician. The participant made friendly jokes about the narrow-mindedness of medical doctors. This, and the general tone of the interview, changed when I explained my background. My impression is that my background as sociologist positioned me as a novice in these interviews. As an outsider I could ask questions too obvious for health professionals, and the participants took their time to explain in detail. I was sometimes ‘questioned back’ about what practice was like in other hospitals, and I was asked for my opinion and advice. In retrospect I view my role as having been a balance between an outsider and an expert on MND.

The design and order of the interviews invited me to use the experiences from those at home as a resource to challenge the health professionals. In the actual interviews with health professionals, the word ‘challenge’ hardly describes the use of my prior interview experiences. Rather, I felt the prior interviews were a way of connecting with the health professionals and increasing my understanding of their world and their interactions with persons with MND and family caregivers. There was one general exception to this. At the hospitals there was, with few exceptions, a general opinion that those at home often were in surprisingly good spirits, given the severity of the condition. I routinely probed more deeply into this issue, asking about the atmosphere and the mood at consultations. My impression was that many of those at home, when outside the hospital, did not feel as spirited as the comments from hospital staff would suggest.

Some persons with MND and family caregivers were critical when recounting their experiences with the hospital, but most participants had a positive impression of this contact.

6.2 Internal validity

A broad definition of internal validity is that ‘a method investigates what it is intended to investigate’ (Kvale 1995:22). One challenge for the type of research this thesis represents is that we do not have direct access to what we are researching. Experiences, lines of work, or the meaning of illness are only available through interpretation. For instance, a photograph may represent a physical reality, but the meaning of a family being together on Christmas Eve is only available through interpretation (see Lian 2012). Ensuring and strengthening internal validity may be seen as a continual process across the research project involving several tactics: for instance, establishing a suitable design for the research question, identifying the right method to answer the research question, checking for researcher effects, checking the meaning of the extreme cases in the material, looking for negative evidence, getting feedback from new participants, checking the transcript translation from oral to written language, creating systems for self-reflection, and so on (Kvale 1995:27). Following this craft approach, this summary has already addressed many issues of internal validity.

6.2.1 Recruitment and sample

A critical note regarding the study design in general, would be that I could have defined a broader universe for the study as illustrated in the study by Cappellato and colleagues (Cappellato et al. 2015). When studying MND care, in addition to those at home, primary health care, and hospitals, they included a subsystem of social services. This additional sub-system was made up of social workers in the community and regional directors of health and social services.

Another consideration of the general study design, is that increased care work at home requires renegotiation of work-family balance, and employers were often mentioned in the interviews with those at home as parties influencing these negotiations. Hence, employers could have been included as yet another subsystem in the overall MND community of practice.

Including additional sub-systems would have provided more nodes in the complex network of actors that needed to be coordinated in each case. Such a study would have provided an even broader perspective on which institutions and professions which have to work in concert, in order to support an MND trajectory. A trade of in such a design would be that fewer cases could have been included. This would mean fewer municipalities included, and less geographic variation in the sample. Increased attention to fewer cases might have increased the internal validity of how people handle MND. However, such a strategy could also have limited the variation to the exploration experiences of services across multiple settings.

The participants in primary health care appeared clinically updated, generally knowledgeable and confident managing MND. This reflects our findings and runs counter to literature, where lack of knowledge about MND has been reported among primary health care staff (Brown 2003, Brown et al. 2005, Hughes et al. 2005). This being said, using another sampling strategy for primary health care, for instance snowball sampling through the family caregivers instead of through the municipalities directly, would perhaps reveal a less flattering picture of primary health care.

With regard to the sample of health professionals in hospitals, I have two critical comments in retrospect. First, in the hospitals with MND teams, there were more people involved in the follow-up than those health professionals formally in the team. If I had used less strict inclusion criteria, and used snow-ball recruitment I assume some respiratory physicians would have been included in the sample. This increased variation would to a larger extent have captured the management of MND in the hospitals. Second, interviewing all the members of the teams meant that some of the content in the interviews overlapped. In retrospect, I could have chosen to interview fewer of the members in the teams, and more health professionals involved in MND outside the teams. I think that the sample from hospital Z, where there was no clear team structure and I used snow-ball recruitment, to a larger extent accounts for the hospital follow-up of people with MND, compared to the sample of hospital X and Y.

6.2.2 Interviews

An important issue influencing the interviews with those at home was whether the person with MND was present. Going into the interviews I expected family caregivers to be more frank about difficult experiences relating to the person with MND when I interviewed them alone. This is the same reason Taylor chose to interview people with MND and their partners separately in a study on MND and sexuality (Taylor and de Vocht 2011). She assumed people with MND and their partners "… might not have secrets as such, but might speak of their fears or frustrations to a nurse, preferring not to share these with their partner quite so explicitly for fear of causing further distress" (Taylor and de Vocht 2011:1578). This happened in some cases (see also the discussion on the distinction between bereaved and current family caregivers below). However, I also experienced that family caregivers, or the person with MND, seemed to use the research interview as an opportunity to raise difficult issues. For instance, one family caregiver, in more direct language than I had anticipated, questioned the person with MND repeatedly about thoughts of dying and how to spend the time they had left together. Another couple discussed the strategies they had to manage disagreements and the importance of disagreeing and negotiating as a way to maintain normality. To me it appeared that the research interview also could be an occasion facilitating talk about fears and frustration, not just couching them. The interviews with both family caregivers and the person with MND present also displayed different types of work of seeking information and clarity about the disease, for instance family caregivers buffering information about prognosis that they assumed to be damaging in some way. In some interviews either the person with MND or the family caregiver took the lead, talking for most of the interview. Taylor and de Vocht not: "Joint interviews can result in particular insights that are not achievable in individual interviews because they provide a window into the couple's world of shared experiences and meanings. This does not mean that joint interviews are necessarily superior, just that they produce different data" (Taylor and de Vocht 2011:1584). All in all, the presence of some persons with MND in the interview situation enriched the variation in the material, hence, supporting the recruitment strategy of seeking variation within sub-systems. The proximity and reciprocal aspect of caregiving of MND is underscored in "immediate care work", that much of the care the family caregiver performed, had to happen in the proximity of the person with MND. This line of work was one way the inclusion of persons with MND coloured the findings. As some of the examples above indicate, it was not given that family caregivers are more likely to self-censor about difficult experiences when they are interviewed alone.

In primary health care my interviews were understood in various ways. For some it was a welcome opportunity to reflect on a difficult task or a chance to promote their work. For others it was a less welcome occasion and a situation that could generate the risk of publishing sensitive information. The variation meant that the interviews compensated for each other, providing a more balanced picture: The cautious interviews presented their ideals in MND care, while the interviews venting frustration described challenges.

6.2.3 Analysis

There is discrepancy between the questions asked in the interview guides and the research questions and analyses presented in the final papers. This discrepancy, however, is what Braun and Clarke view as a sign of good quality qualitative analysis. They note that if the resemblance between the questions, themes and categories in the interview guide are the same as in the final paper, no analysis has taken place (Braun and Clarke 2006).

I had planned for the interview guides to provide detailed data about MND at home, through asking questions about the everyday. In retrospect this approach generated ample opportunity for analysis. One example was to ask about sleep and sleeping patterns for those at home. Sleepless nights and such compromises on basic needs were indicators for how the participants defined the situation. In an emergency no or little sleep is acceptable; while in a sustainable everyday sleep needs to be part of the routine. Hence, questions about sleep turned out decisive for the concept unstable terminality. Family caregivers could compromise on their sleep in a terminal situation, but little sleep was difficulty if MND represented a chronic condition. In this sense psychological and health related questions turned out to be helpful means to create interviews to shed light on social, cultural, ethical or coordinative aspects of MND care and management.

In retrospect, I will highlight in two ways that the supervisors played an important role in the analytic process. First, they were central discussion partners in large decisions during the analysis, such as adjustment of aim or research question. Second, as we regularly met and I produced text summarizing my work for each meeting, the supervisors were central in making sure that there was progression in the analytic work. They encouraged me to make adjustments in my analytic strategy, when I was not advancing properly.

The large data set provided the opportunity to increase internal validation by comparing within and between subsystems. It also served as a reservoir in which to seek out contradictions and to expand on the claims I made. The findings could have benefited from more active member validation (Kvale & Brinkmann 2009:214); for example, they could have been presented in focus groups and the resulting feedback could have been incorporated in the study. Papers II and III especially would have benefited from such an approach, but this proved difficult due to time concerns. Member validation could have spurred those at home to think of other lines of work relevant for them, or specific tactics they used managing the work. Similarly, health care professionals in primary care could have suggested other challenges, or other solutions to them, than what appear in the papers.

6.3 External validity

In general medical journals, there is ongoing debate about the status of qualitative research and its capacity to inform clinical practice (Greenhalgh et al. 2016, Loder et al. 2016). This debate is linked to how valid findings based on qualitative data are outside the immediate context in which the research took place. Is qualitative research generalisable, and if so, in what way? And connected to this, are the findings in this thesis relevant for clinical practice?

Generalisation is also a subject of debate within the qualitative methodology literature. Nadim (2015) points to three positions in this field: 1) the rejection of generalisability (see Lincoln & Guba 2000); 2) a middle position arguing that generalisability in qualitative research is possible, but of a fundamentally different kind than quantitative approaches; 3) that qualitative research is generalisable in the same way as quantitative research. The first position, to reject generalisability, is difficult to hold in my view. But qualitative research aiming at generating hypotheses rather than claims is one example. Most qualitative studies at least implicitly communicate an ambition to have something to say outside their own immediate study context, even though this ambition may be vaguely stated and unclear (Nadim 2015, Payne & Williams 2005). The findings in this thesis belong to the second and third positions above, and I will account for these positions below.

The middle position, that qualitative research generalises in a distinct way, has several adaptations. Malterud (2001b), for instance, prefers the term transferability to generalisability to highlight the different underlying logics in qualitative and quantitative research. Transferability, then, is an argumentative form of generalisation, highlighting the need to explicitly account for the aspects of the sample, context and findings that have relevance beyond the study context. This has similarities to Payne and Williams's concept of moderatum generalisation (Payne & Williams 2005), which requires the qualitative researcher to take responsibility for specifying what other contexts the findings might be relevant in. The third position, which claims that qualitative findings are generalisable in the same way that quantitative findings are, is more radical and less common. This position maintains that qualitative research based on empirical rather than theoretical findings with non-probability samples may form the basis for general conclusions, as in quantitative survey research. This last position underscores the importance of a strategic sample for drawing conclusions (Nadim 2015:135).

Drawing on this literature, four questions arise when discussing generalisability of qualitative findings: 1) *What is the ontological status of the object being claimed generalisable* (Payne and Williams 2005:306-307)? Is it a theory or a perspective, for instance a lens to untangle a complex social situation? Is it a social artefact, such as the prevalence of a given attitude? 2) *What is the 'fit' between the sample in the study and the 'receiving' sample to be generalised to* (Nadim 2015:136)? Is a sample exclusively consisting of women relevant for men? 3) *What is the 'fit' between the context of the study, and the 'receiving' context?* Context is central to Malterud's concept of transferability (Malterud 2011:22); for example, the similarity in patient-doctor relationships across time and space is a context with potential for generalisation. 4) *Who is responsible for the generalisation?* Payne and Williams underscore that the researcher has the most insight and is thus in a primary position to take responsibility for how and where a finding is valid outside the research context (Payne and Williams 2005).

Kvale and Brinkmann take a more pluralistic position regarding who should be responsible for generalisation. They point to how participants may use research as an opportunity to reflect on self-understanding. In this context the participant may provide *member validation* (Kvale & Brinkmann 2009:214*)*. In the general public sphere, Kvale and Brinkmann point to critical common sense understanding of research findings. This context provides *audience validation*. Finally, the research community, having a theoretical understanding, may contextualise findings in the research literature, providing *peer validation* (Kvale & Brinkmann 2009:214). In this view, the claim of generalisation to a given context may be endorsed or refused by members, audience or peers.

I will now turn to discuss these questions of ontological status, sample, context and responsibility of generalisation in regard to the findings in this thesis.

Generally, the three papers in this thesis are written for an international audience, and as such they claim to provide a meaningful contribution internationally. However, in line with the argumentative types of generalisation sketched out above, the relevance of a finding does not depend on findings being identical in the receiving context and the original context studied, it is not necessarily a correspondence generalisation claim. Qualitative findings may also be meaningful because the provide contrasts, inspiration, or negative or positive comparison in the receiving context.

In Paper I 'the what' to generalise is the theoretical concept unstable terminality. This concept claims to provide a lens through which to understand a complex social phenomenon for those in the MND community of practice. My claim is that the sampling strategy's focus on variation and typicality strengthens the external validity of the concept. I will return to the importance of context and unstable terminality in the discussion of the findings. Briefly, it seems that the availability of mechanical ventilation and cultural acceptance of this intervention plays an important role in making unstable terminality a relevant concept for MND. With regard to responsibility of generalisation, the concept can be grasped by and tested among family caregivers, people with MND and associated health professionals according to whether or not they recognise this type of ambiguity in their own situation. This kind of member validation

(Kvale and Brinkmann 2009:214) can also be done with people handling diagnoses other than MND that have similar trajectories and treatments.

The findings in Paper II provide a theoretical lens through which to understand a complex social phenomenon, namely care work. Again, the sample combines strategies for variation and typicality and aims at being broadly relevant for people living in this situation. With regard to context, the theoretical nature of the findings makes them less bound by context. The lines of work to interpret family caregiver work are not necessarily tied to the Norwegian welfare state. The lines of work may be judged as relevant or irrelevant to inform practice by family caregivers, persons with MND and associated health professionals, and to diagnoses other than MND. As noted in the section on internal validity in qualitative analysis, in Paper II I wanted to limit the visibility of theory in the paper in the interest of communicating more clearly with a health science audience. For instance, with a stronger commitment to theory I would rather have labelled the finding 'seeking information and clarity about the disease' the shorter 'information work'. Moreover, I would also have given examples of the different tactics the participants used to manage unwelcome information about a condition without a cure, to deal with excess information, to distinguish reliable from unreliable information and so on. Even though the analytic process involved a great deal of theory, I tried to present the findings as a thematic analysis to increase the potential for member validation.

While Paper II was an attempt to move away from a theory-driven notion of what a finding in qualitative research should be, Paper III goes even further in this direction. The data are reports of subjective experiences of health professionals managing MND in primary health care in the form of descriptions of challenges they encounter. Other actors involved in the same care, for instance those at home, may describe these situations differently, and perhaps not as challenges at all. Still, following a symbolic interactionist approach, the way a situation is defined has real consequences, and this subjective basis is what primary health care use as their knowledge base to make decisions. In this sense the findings in Paper III move in the direction of Nadim's third position of generalisability, relating to the empirical prevalence of the subjective experience of those challenge descriptions. On this note, the purposive sample of 18 participants in primary health care is a key factor contributing to generalisability. These participants were critical (Kuper et al. 2008) or elite (Tansey 2007) participants in the sense that they were selected based on positional and reputational criteria of being especially knowledgeable in MND management in primary health care. In addition to providing a critical case sample, this sampling strategy was combined with another strategy to identify participants in varying contexts, different hospital catchment areas and urban and rural areas. The combination of such sampling strategies allows for stronger claims regarding the empirical occurrences of the findings across Norway. Compared to the findings in Papers I and II, the findings in Paper III are more strongly embedded in the context of the generously financed public Norwegian welfare state. This welfare context informs what is perceived as ideals for care. Further, the challenges depend on the ideals in the sense that a challenge is to not meet the ideals. However, even if these findings, from a theoretical perspective, are context bound and as such less generalisable to other settings, paradoxically, the number of potential readers is likely higher than for the other papers.

7. Discussion of findings

In this chapter I discuss findings in the three papers, and explore how their individual empirical contributions may comprise a larger whole when put together. First, I discuss how the unpredictability inherent in the concept unstable terminality (Paper I) may be understood. I conclude that there will always be uncertainties related to the prognosis, as decisions made by the person living with the illness shape outcomes in MND. Second, I turn to the MND trajectory (Paper I) and compare it to other more common illness trajectories in the health services. I conclude that MND is an atypical case with characteristics that challenge the institutional structure in primary health care. Third, I discuss the lines of work involved in MND family caregiving, especially information work (Paper II), and family caregivers who become expert caregivers (Allen 2000). My findings suggest that family caregivers with both too little and too much information are at risk for exhaustion. Finally, I discuss coordinating mechanisms in primary health care and point out how MND goes against the default mode of collaboration. Difficulties experienced by health professionals in primary health care may be understood in this perspective, as they have to put in extra effort to compensate for this collaborative mode.

7.1 Prognostic uncertainty in MND

The continuous development of medical technology may change and challenge established notions of a given condition, for instance how we treat and act on it (Hofmann 2001). Consequently, conditions for and experiences of illness trajectories are gradually changing. A key finding in this thesis is the concept unstable terminality (Paper I), pointing to whether a condition remains fatal despite the efforts of those at home, primary health care and hospitals, and the technology, skill and resources they have available.

The unpredictability of MND is a recognised problem for clinicians and in research. Elamin and colleagues (2015) have addressed this challenge and proposed a prognostic tool. By scoring whether a person with MND has a bulbar or respiratory onset or not, whether there is executive dysfunction or not, and measuring the pace of physical decline (using the Revised ALS

Functional Rating Scale, ALSFRS-R), this tool predicts whether the person with MND has a poor prognosis (defined as death within 25 months of symptom onset) or a good prognosis (defined as survival of at least 50 months or more from symptom onset). Does this or any such tool challenge the unpredictability implied in unstable terminality?

As noted in the theory section on prognosis, there are both biomedical and subjective aspects to prognosis (see Christakis 1999). While the physical unfolding of a disease may be predictable, the illness trajectory, as an assembly of interacting actors (these may be humans, technology, the condition and so on), may not be. Prognosis involves peoples' subjective judgements. An important consideration in this regard is that most of the sample validating the prognostic tool is from Ireland (Elamin et al. 2015), where long-term invasive ventilation is not readily available (Foley et al. 2014a). This intervention may prolong life expectancy considerably. The decision of whether or not to implement invasive ventilation is better understood through a psychological, social or cultural lens, rather than from a purely biomedical perspective. Unstable terminality does not seem to be a fitting description for the experiences of persons with MND in that study (Foley et al. 2014b), where MND appears to be a pure terminal category. Based on Irish data, Foley and colleagues describe MND as involving multidimensional and unremitting loss: people never regain what was lost, and biographical repair or new types of normality do not appear (Foley et al. 2014b). Foley and colleagues' findings explicitly challenge Locock and colleagues (2009), who find traces of 'normal' chronic illness in MND, which is supported by data from Britain.

The other part of the sample to validate the prognostic tool is from Italy (Elamin et al. 2015). An Italian study by Cappellato and colleagues (2015) supports the concept of unstable terminality with its discussion of the implications of various definitions of MND as curable or treatable among services providers in Italy. The study points out that in Italy, legislation guarantees rehabilitation for people with temporary or non-progressive functional limitations, yet this is not granted for people with degenerative conditions. Cappellato and colleagues argue that health professionals may legitimately interpret symptoms in the person with MND in contrasting ways with regard to the legislation entitling rehabilitation services. Thus, because the condition can be classified as both chronic and terminal, unstable terminality (Paper I) finds support in studies based on British (Locock et al. 2009, Brown 2003) and Italian data (Cappellato et al. 2015), but not Irish data (Foley et al. 2014b).

Implications of the unstable terminality in a certain phase of MND (Paper I) also seem to be apparent in American data (Rabkin et al. 2009). Rabkin and colleagues point to how family caregivers were significantly more depressed in cases where the person with MND had planned invasive ventilation than in cases where the person with MND did not make such plans. However, this difference was lost over time (Rabkin et al. 2009:454). They note how the depressed pre-invasive ventilation family caregivers made major life changes as the moment of installing invasive ventilation approached. One family caregiver got divorced and in another case, the person with MND moved to a nursing home. Rabkin and colleagues conclude: 'these caregivers accommodated to the idea of the patient's indefinite survival by making major changes in the circumstances that had generated their distress. The implication for clinicians is to suggest to severely distressed caregivers that some kind of significant change is indicated both for their benefit and that of the patient' (454). Using Becker's (1997) notion of plot, as we did in Paper I, it seems that the family caregivers in Rabkin and colleagues' study managed to break out of limbo and establish a sustainable strategy in the face of an uncertain future.

Asking for a prognosis is not just a query about 'how long do I have?', but also about 'what will happen?' (Murray et al. 2005). Even though the prevalence of long-term mechanical ventilation for people with MND is low in Norway (Tollefsen et al. 2009), it is increasing (Indrekvam et al. 2015). Living on long-term invasive respiration is a question of reframing a terminal condition as a chronic disability (Paper I). Considering 'What will happen?' was what we found to be a component of information work (Paper II). An important finding in this regard is that a key feature in predicting the prognosis of MND is the decision whether or not to use invasive mechanical intervention (Paper I). To respect the autonomy of those living with the illness, health professionals must strike a delicate balancing between describing the consequences of such an intervention and advising the patient what to do. The issue of social support for nurse assistants supporting patients who choose invasive mechanical ventilation is important in this regard (Paper III). Hence, a core insight from the concept of unstable terminality and the ambiguity and unpredictability that it entails, is that a technical solution is not necessarily required to eliminate the ambiguity. Rather, unpredictability must be integrated in the organisation of MND care. This decision-making is not desirable to fixate, in order to respect the autonomy of those living with the illness.

As pointed out in the theory chapter on meaning and narratives, there are studies of how people with chronic illness make sense of their lives in general (Kleinman 1988, Frank 1995), and those with MND specifically (Brown & Addington-Hall 2008, Locock et al. 2009). Unpredictability is inherent in MND, and health professionals must be capable of managing this. Having established that prognostication is not a purely technical exercise, we turn to the question of how health professionals facilitate prognostication and meaning-making for those with illness and family caregivers. We have found that MND shifts between chronic and terminal framings, which raises the issue of what narratives health professionals, especially those in multidisciplinary MND clinics, aim to facilitate, as well as what tactics the health professionals use to secure the autonomy of those living with the illness and family caregivers. This approach would mean changing the focus of the narrative analysis from those living with illness to the health professionals facilitating their care.

7.2 Classification, collaboration and trajectories

How, then, does the unpredictability in unstable terminality fit the organisational landscape of the Norwegian health services? Viewed from above, the organisational landscape in the health services may be seen as a series of pigeonholes, where each hole is an available option in a case being managed by a health professional. In primary health care available options would typically be to initiate home services, to move a frail elderly to a nursing home or to provide assistive devices. A physician in a hospital would have various laboratory tests, treatments, or referrals to other specialists as some available options. Classification is crucial for creating order and clarity. The challenges in primary health care described in Paper III, including difficulties providing tailored care, problems with ensuring good working conditions at home, and difficulties recruiting and retaining qualified personnel, have no readily available pigeonhole that fits the particular case of MND. This lack of fit may reflect the ambiguous classification of MND (Paper

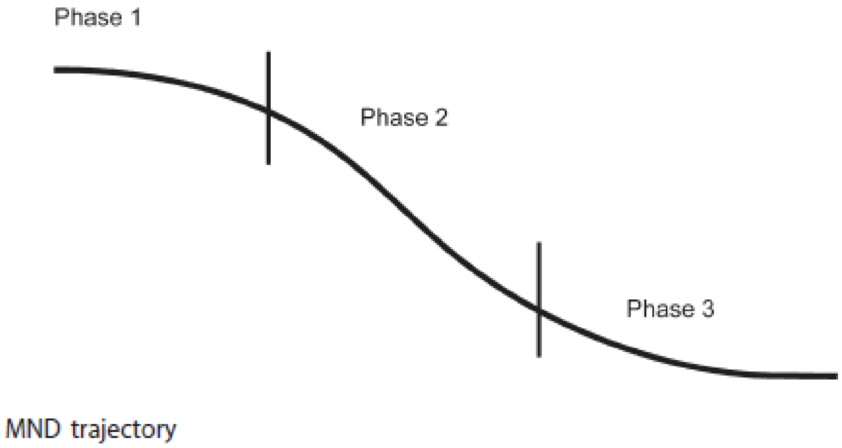
I), as well as family caregivers' difficulties in managing external resources such as the health services (Paper II). For family caregivers, it might be frustrating to do this work if there are no obvious institutions to help out with the challenges that arise. The lack of fit in the MND case raises the question of how MND compares to other conditions.

7.2.1 Trajectories

Comparison may involve an element of valuing something more or less than something else. A key issue in this regard is that what constitutes good collaboration of care is a matter of perspective (Lerum & Frich 2012): what might be a sufficient solution for a physician might not be one for an economist or those living with illness. These actors may have differing opinions of how to define optimal or sufficient solutions. This problem of normativity has also been raised in connection with Strauss's concept of illness trajectories (Corbin & Strauss 1988, Strauss et al. 1985). Allen and colleagues (Allen et al. 2004), have noted that Strauss, when presenting illness trajectories, seldom comments on normativity and perspective, and that a trajectory may be problematic from one point of view and not another (Allen et al. 2004:1011-1012). The overall aim of this summary is to contribute to knowledge about conditions for collaboration between family caregivers and health professionals managing MND, each of whom approaches MND from a different perspective.

Differing opinions become especially salient when an illness trajectory cuts through several subsystems. What is desirable, beneficial or manageable within one subsystem may not be so within another. Adding to the complexity in this regard is the atypical trajectory MND represents. As underscored in Paper I, the ambiguous classification of MND and the shifting impact this has on the work of family caregivers (Paper II) and services provided (Paper III), may create challenges for care collaboration. A key finding in Paper I was the MND trajectory:

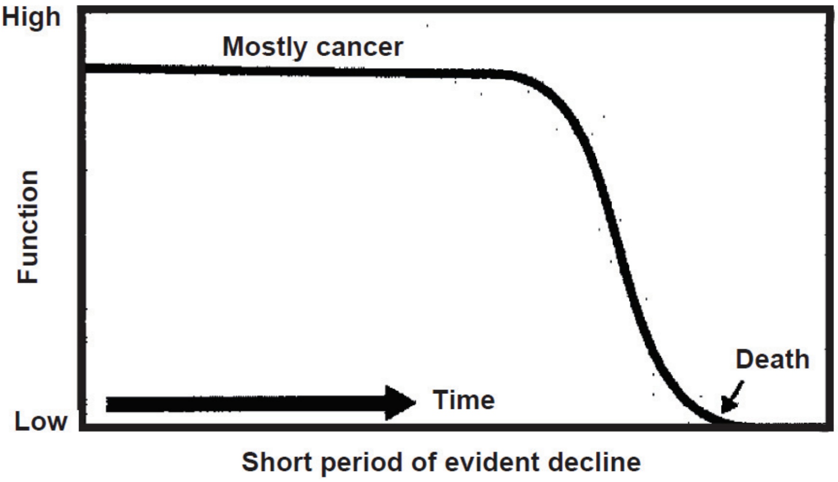
Figure 3: MND trajectory (Paper I)



The figure above does of course not fully do justice to the variation in MND illness trajectories, and it is important to mention that MND trajectories may have well-coordinated services.

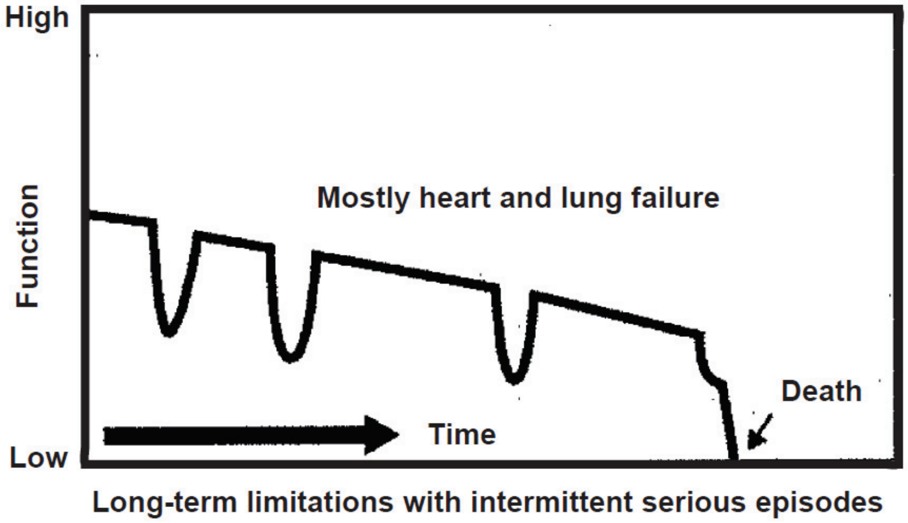
To discuss the atypical MND trajectory, it is useful to contrast it with more common trajectories, presented in the three figures below:

Figure 4:A trajectory with short and evident decline, typically cancer.



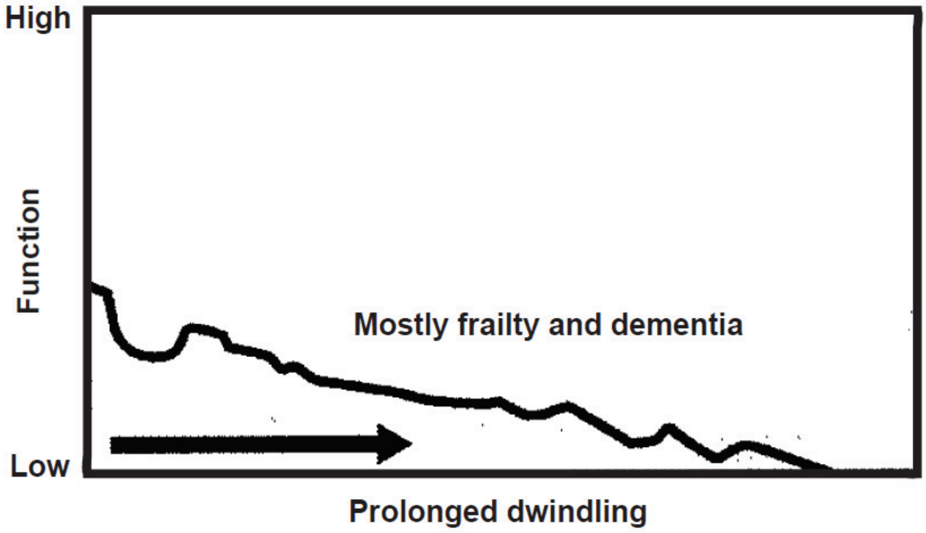
Adapted from (Lynn & Adamson 2003).

Figure 5:A long-term trajectory with serious intermittent episodes.



Adapted from (Lynn & Adamson 2003).

Figure 6: A trajectory with prolonged dwindling.



Adapted from (Lynn & Adamson 2003).

The three trajectories presented in the above figures have different properties. The first trajectory often has a clear diagnosis, typically cancer, and a short period of evident decline. The second trajectory is long-term, characterised by intermittent serious episodes. This may typically include conditions like organ failure in heart conditions or chronic obstructive pulmonary disease (COPD). The third trajectory involves prolonged dwindling, typical cases being old age, a general fragile state and dementia (see also Murray et al. 2005). These are common trajectories in the health services, which are all dependent on interaction between those at home, primary health care and hospitals.

7.2.2 Trajectories with short and evident decline

In contrast to their preparedness for dealing with MND, the health services are well adapted to the first trajectory above, which typically involves cancer. In these cases there is a period of active, or even aggressive, treatment before there is a relatively clear-cut transition to a palliative phase. These properties of the trajectory are reflected in the surrounding institutions: there are specialised clinics that provide treatment and other specialised clinics that provide palliative care. The predictability of the trajectory allows for standardised follow-up or well-defined categories, and there are specialised institutions available depending on the various outcomes of the trajectory (worsening or remission). Hence, even the undesired outcome of the cancer being fatal involves specialised care. From the perspective of those living with the illness, the cancer trajectory above is undesirable and difficult to control. From the point of view of health professionals, however, the health services are prepared for such trajectories. The available specialised institutions give health professionals a sense of control, even though the trajectory takes an inevitable course.

In MND trajectories the specialised institution is replaced by the multidisciplinary team located in the hospitals, which is where diagnosing and initiation of interventions takes place. As pointed out in Paper I, the teams are typically relying on a passive prognosis, standardising consultations every three months. While specialised palliative care is readily available for most cancer conditions, it is less commonly available for MND (Aoun et al. 2012). This might be related to the rarity of MND as condition, which undermines the effort to establish specialised institutions with the targeted clinical competence. Unstable terminality and the associated lack of predictability (Paper I) are two striking differences to trajectories with short and evident decline. One attempt to compensate and build capacity to manage this unpredictability involved building relationships between health professionals and those at home (Paper III). The unpredictability of MND also challenges family caregivers who are balancing efforts to control overwhelming amounts of care work on the one hand, and not to compromise their own and others' expectations on the other (Paper II).

7.2.3 Long-term trajectories with serious intermittent episodes

This kind of trajectory involves reoccurring acute exacerbations, each with the potential to be deadly; examples include conditions such as heart failure or COPD. A similarity to the MND trajectory relates to unpredictability (see Paper I). In heart conditions or COPD, the difficulty lies in predicting which exacerbation will ultimately be fatal. This unpredictability is to some extent incorporated in the division of labour between primary health care and specialised services. The acuteness of the exacerbation makes a clear criterion for hospital admission. Moreover, primary health care is geared towards handling the more stable phases that follow acute episodes. The shifts between acute and stable phases of the trajectory are difficult to handle, but do not fundamentally challenge the institutional structure of the health services. From a health professional perspective, the unpredictability of serious intermittent episodes is manageable on a system level. From the point of view of those at home, however, the reoccurring acute episodes disrupt everyday life, and life expectancy is difficult to predict. The issue of family caregiver burnout (Paper III) may be relevant in such cases. It is imaginable that this trajectory, too, may create a sense of unstable terminality (Paper I) for those at home. In MND health professionals also experience unstable terminality (Paper I), which might not be the case for health professionals handling heart conditions or COPD.

Contrary to a trajectory with a short and evident decline, neither trajectories with serious intermittent episodes nor MND offer a clear-cut passage to a palliative phase. For instance, for people with heart conditions, common types of death may be sudden death or death in an acute setting in a hospital. In the case of MND, however, it is difficult to facilitate the preference of dying in an institutional setting such as a hospice (Paper I).

Serious episodes in the trajectory create situations that make the use of specialised services imperative. The smoother progression of MND makes it less clear when to decide when such services are needed. As found in Paper I, the hospitals in the sample in this study countered this vagueness with prescheduled consultations, often every three months. In this sense the hospitals were mimicking a trajectory with intermittent serious episodes. In primary health care, however, the differences between organ failure and MND went beyond what is captured in this model. MND is a relatively rare diagnosis compared to heart failure or COPD, and we found that health professionals in primary health care struggled to recruit and retain qualified nurse assistants (Paper III). Additionally, from a primary health care perspective, a person with a heart condition returning from hospital would be expected to have improved or to have remained in a stable phase. A person with MND returning from the multidisciplinary clinic would likely be worse, and there would be additional requests to acquire more advanced and unfamiliar technical equipment. As far as primary health care is concerned, hospital admission would involve more work, rather than providing relief. During a serious intermittent episode, the actual intervention and change is implemented in specialised services; primary health care's role is to maintain the regimen and rehabilitation. Hence, in this division of labour, hospitals represent change and flexibility, while primary health care represents stability. The division of labour is less clear cut in the MND trajectory. Many interventions, including most of those associated with continual change and adaptation (King et al. 2009) in phase 2 (Paper I), are managed within primary health care, without much assistance from hospitals. Primary health care, then, has to handle a dynamic and changing trajectory in the when dealing with MND rather than the usual stable phase. One of the challenges we found in Paper III, namely the difficulty of providing tailored care, may be related to the dynamic and changing nature of the trajectory of MND. This challenge may also relate to the fact that the institutions which constitute primary health care do not normally manage such dynamic and changing trajectories.

7.2.4 Trajectories with prolonged dwindling

These trajectories are characterised by a 'prolonged gradual decline' (Murray et al. 2005:1007). Typically they involve 'patients [who] may lose weight and functional capacity and then succumb to minor physical events or daily social "hassles" that may in themselves seem trivial but, occurring in combination with declining reserves, can prove fatal' (Murray 2005:1008). The comparison to advanced MND in what is described as phase 3 (Paper I), where trivial events may have fatal consequences, is obvious. Even though trajectories with prolonged dwindling are unpredictable, they are common in primary health care (as opposed to MND's rarity), and there is a standardised option to manage these trajectories, which involves increasingly more help offered at home followed by a gradual phasing into a nursing home.

A significant discrepancy in this regard relates to a life-course perspective. In MND peak onset is between 58 and 63 years old (Kiernan et al. 2011). As noted in the section on biographical disruption, age is an important factor in whether chronic illness is experienced as disruptive (Locock and Ziebland 2015). Further, Foley and colleagues (2014a) use a life-course perspective to criticize international recommendations (Aoun et al. 2013, Andersen et al. 2012), which point to specific episodes in the trajectory as indications to initiate interventions. For Foley and colleagues, situations such as missing out on parenthood and attempts to shelter children from disruptive events are more salient when interpreting the needs of those at home. A life-course perspective may explain why many people living with MND find nursing homes highly undesirable and view them as a last resort.

Furthermore, as pointed out in Paper III, it can be challenging for primary health care to sustain the advanced type of care needed for MND within a private home. We found that health professionals in primary health care reported difficult working conditions in the homes of people with MND (Paper III). This was particularly true for people with MND in advanced stages, and was connected to difficulty finding competent personnel. In this regard, our findings support the notion of primary health care being between a rock and a hard place (Dybwik et al. 2011), being asked to perform tasks and follow-up on trajectories that are perhaps beyond what the institutional infrastructure is capable of.

There is also a clear difference between these dwindling trajectories and MND with regard to different phases. People with MND are not characterised by sustained periods of frailty, but rather by mild symptoms in phase 1 and rapid changes in phase 2 (Paper I).

7.2.5 Atypicality, institutional fit and the potential for building competency

The comparison between MND and more common illness trajectories underscores MND’s status as an atypical case. Trajectories with short and evident decline, typically cancer, may be more predictable, with a clear palliative phase. Even though terminal cancer is a devastating experience, the trajectory fits the institutional landscape in the health services. Trajectories with serious intermittent episodes, such as MND, are unpredictable. However, this unpredictability does not challenge the institutional structure of the health services. The division of labour between primary health care and hospitals is designed to cope with acute exacerbations. Trajectories with prolonged dwindling are also unpredictable, but so common that a standardised option has been developed in the health services, namely the nursing home. A main difference between dwindling trajectories and MND is the life-course perspective.

Atypicality and rarity set MND apart from all of the other aforementioned trajectories. With regard to atypicality, this is reflected in how all the other trajectories have features which fit the institutional landscape to a larger degree than MND. Additionally, rarity limits the opportunities health professionals have to accumulate clinical experience, as there are few cases to draw on. Hence, the main challenges in the health services when managing MND trajectories are lack of institutional fit and limited opportunity to accumulate experience.

The challenge of accumulating experience is countered by the multidisciplinary MND teams found in hospitals. A property, perhaps unintended, of these teams is their function as knowledge banks that help build competency. The rarity of the condition is compensated for by the larger catchment area of the hospital, meaning that health professionals in multidisciplinary clinics may accumulate experience across MND cases. This knowledge bank aspect contributes to more informed clinical follow-up of persons with MND and their family caregivers. The potential this knowledge bank has for other health professionals is often underestimated. The day-to-day needs of people with MND and their family caregivers are mainly supported by primary health care (Papers I and III). For this reason, the connection between the multidisciplinary clinics and primary health care is especially salient. The findings suggest that a practical implication of the knowledge bank status is that primary health care professionals can come to the clinic and participate in consultations to learn more about MND across cases. In this perspective an additional reason for people with MND to visit multidisciplinary clinics, besides getting help with their own situation, would be to contribute to a larger knowledge bank that other people with MND and health professionals may benefit from. Taking this one step further, multidisciplinary clinics offer the potential to facilitate participation in research. When facing a condition without a cure, there is a struggle to create meaning; and research may play a role in creating such meaning. As the respondent ‘Sandra’ in Robinson and Hunter (1998) points out:

> *It helps to know that there is so much research going on to find a cure for MND [British for ALS], and all those scientists are trying as hard as they can. I never want anyone else to suffer like my husband did. I know the answer didn’t come in time for him, but now we know what it is and what we are up against, I will do all I can to help the doctors and scientists. (Robinson & Hunter 1998:132)*

Compared to Norway, Denmark seems to have pursued the potential of such a knowledge bank further. Most persons with MND in Denmark attend the specialist hospital RehabiliteringsCenter for Muskelsvind (RCMF, Jeppesen 2013); there is no such centralised clinic for MND treatment in Norway. At the hospital level Norway appears to have a decentralised model of specialised MND care compared to Denmark. This can, of course, be explained by the radically different geographical realities in the two countries. Still, when managing rare conditions there are several benefits associated with a centralised approach. Jeppesen (2013:78) points out the potential of these larger centralised clinics to generate broader and deeper knowledge, as well as the potential of health professionals' expertise to influence the personal encounter with the person with MND. Clinics also have the potential to establish registers and to manage archives and data for research.

7.3 The lines of work

Glaser & Strauss (1971:177-178) make a distinction between substantive and formal theory. Substantive theory is informed by the empirical field in which the study takes place, while formal theory is developed on a more conceptual level across fields and studies. The distinction between substantive and formal theory is not absolute, but rather a matter of degree of abstraction. Corbin and Strauss's (1988) attempt to formulate a rather formal theory of chronic illness, results in a high level of abstraction. They continue to use illness trajectory as a concept for inquiry, and, as presented in Paper II, they view all chronic illness as requiring those affected to take part in three lines of work: illness work, biographical work and everyday work.

Again following Strauss, a condition for developing a theory is that it has explanatory power, is relevant, is grounded in data, and that is modifiable (Glaser & Strauss 1971:177). Paper II may be seen as an exercise designed to generate substantive theory about care work in MND and as a contribution to the reformulation of 20-year-old formal theory about chronic illness.

We understood immediate care work as involving work in physical proximity to the person in need of care with the aim of replacing functionality or providing a sense of safety (Paper II). In the classic study by Strauss and colleagues (1985) hospital staff are involved in five lines of work when shaping illness trajectories: 1) machine work relating to the relationship between social actors and technology; 2) safety work relating to checking and monitoring patients, personnel and environment, 3) comfort work relating to alleviating the physical discomfort of illness, and 4) sentimental work focusing on the psychological aspects of being ill. Machines and concerns about safety and physical and psychological comfort are often integral parts of immediate care work (Paper II). Hence, family caregivers' immediate care work at home may entail several or all of these hospital lines of work. This suggests that the work hospital staff does in hospital wards bears a striking resemblance to what family caregivers do at home, illustrating the competence and support needed to perform such work.

A fifth type of work in hospitals is articulation work (Strauss et al. 1985). Articulation work is the coordination of all the different types of tasks and work that shape a trajectory. Often it is the physician who has a blueprint of this plan, but it is the head nurse who carries it out. Without articulation work both the organisation and the illness trajectory would break down (Strauss et al. 1985:151). Articulation work, then, is work of a higher order, meta-work needed to manage the other lines of work. Obviously, family caregivers engage in articulation work. Moreover, the line of work used to manage external resources and assistance (Paper II) may be seen as a sub-type of articulation work, namely the work to integrate health services into the bustle of everyday care at home. An important issue in this regard is how the lines of work are entangled in one another. For example, the articulation work of managing external resources may have consequences for biographical work. This notion is supported by Foley and colleagues (2014c), who point out that resignation towards MND was an important reason for utilisation of services for people with MND.

7.3.1 Information work and expert caregiving

One striking difference between Corbin and Strauss's (1988) original theory on family caregiving, presented in the theory section above, and current theories on managing illness is the access to information. In Paper II I label this 'seeking information and clarity', which I subsequently abbreviate as 'information work'. As pointed out in Paper II, information work is a term that bundles the different strategies and tactics people with MND and their family caregivers use to process information about the condition and their situation. It is not unusual for persons with MND to use family caregivers to screen information that is frightening, unreliable, overwhelming and so on (Paper II, see also O'Brien 2004). Jeppesen (2013:62-63), also gives examples of what may be interpreted as different information work tactics used by people with MND, including reluctance to relate to the future, insistence on stopping information exchange with health professionals, and withholding of information about individual development of the condition. Information work also relates to handling the problem of excess of information (see Abbott 2014). MND is poorly understood, meaning that much of the information available about the condition may be wrong or not properly validated. Separating good from bad information becomes increasingly difficult as the amount of information increases.

MND is fearsome, uncommon and unstable (Paper I), and information work (Paper II) may shape the classification of the condition. That MND is uncommon makes information work more salient. As discussed above, we have readily available images to help us make sense of cancer, heart disease or dementia. Cancer survivors appear regularly in the media and share their experiences. In most families there is or has been someone with dementia; we often know this condition first-hand. These cultural images of illness shape how we classify them. There is an abundance of information about MND as well. But because MND is uncommon and less frequently in the media, processing this information is a more active process and requires more demanding work than simply using the images already available to us.

One important source of information for family caregivers is health professionals. Consequently, information work to deny, avoid or shelter oneself from unpleasant and unwelcome information suggests an explanation as to why health professionals struggle building relationships with those at home (Paper III). On a positive note, family caregivers may gain resources through information work that allow them to reshape their role. Hence, information work is a type of work that may represent a way for family caregivers to manage or escape from the limbo that the disruption of MND creates (see Paper I).

Davina Allen (2000) points out roles family caregivers may have in an adult hospital ward, roles that also are useful for understanding interaction between family caregivers and the health services in general. Allen draws on Rosenthal and colleagues (1980), who suggest that family caregivers are cast in three roles: the visitor, the worker and the patient. The family caregiver as visitor only performs comfort work and lets the health professionals define and perform what other work needs to be done. The caregiver as worker goes beyond the visitor role by performing tasks such as those related to intimate tending and bodily excretions. Similar to the visitor, the worker also refrains from challenging decisions or the workflow of the nurses. The family caregiver as patient has broken down under the stress of caregiving and needs attention from health professionals. Hence, none of these roles challenges the authority of the health professionals to any great extent.

Allen (2000) expands this caregiver typology by adding the role of the expert carer. The expert carer has a well-established relationship with the person with the illness, and Allen stresses the strong feeling of responsibility that expert caregivers feel for their loved ones. Handing over the care work to others challenged the expert caregivers' sense of control over the caring process. If necessary, the expert carer will challenge decisions made by health professionals. Hence, negotiations arise about whose knowledge and authority over the patient will prevail: the family caregiver's or the health professional's. Such negotiations play out against a highly-charged emotional backdrop of expert caregivers' strong sense of moral responsibility for the cared-for. Allen notes that expert caregivers may create problems with coordinating services. She notes how it is difficult for expert caregivers to interact with health personnel without being perceived as being critical. Hence, a hypothesis is that family caregivers in an expert role may receive less support from health professionals than those in more compliant family caregiver roles such as the worker, visitor or patient, often becoming isolated.

Due to health professionals' limited experience with MND, family caregivers may have more knowledge about the condition than health professionals. Information work (paper II), may lead family caregivers to become expert caregivers, claiming control over health professionals in the division of labor (Allen 2000).

Expert caregiving and information work are important with regard to the available resources for family caregivers to escape from the limbo created by the MND diagnosis (Paper I). On the one hand, we can imagine that family caregivers with little information about MND and the prognostic outlook might risk becoming passive, stuck in immediate care work (Paper II), and thus finding a sustainable role would be difficult. On the other hand, it might also be problematic for expert family caregivers with a lot of information to utilise health services. Family caregivers who become experts run the risk of easily identifying lack of knowledge among health professionals. This could lead to distrust of and lack of respect for the health services, which might, in turn, increase the family caregiver's sense of commitment to immediate care work (Paper II). We found (paper III) that health professionals in primary health care feared family caregivers to break down. Hence, from a clinical perspective both caregivers with information work tactics involving denial and refraining from gathering information and expert caregivers with much information may experience difficulty in collaborating with health services.

7.4 Coordinating MND

To manage the ongoing change and adaption (King et al 2010) associated with phase 2 of the trajectory (see Paper I), there is need of a flexible coordinating mechanism, such as mutual adaptation or direct supervision (Mintzberg 1989). However, flexible coordinating mechanisms are time and energy consuming. Vabø (2012) notes that in new public management (NPM) standardisation of output is often the preferred coordinating mechanism. Focusing on the output allows for discretion in the actual performance of the task. However, Vabø's analyses suggest that home services in primary health care are characterised by a standardisation of input and regulation. This is an effective coordinating mechanism for stable and simple tasks and an optimal coordinating mechanism when the goal is to allocate as much of the workforce as possible to actual task performance, rather than coordination between tasks. However, it is a burdensome mode of coordination when the tasks are complex, in need of constant adjustment, and require discretion in performing them. Standardising input makes it difficult to implement new tasks, as this involves developing an entire new protocol and routine. From this coordination perspective, primary health care is geared towards stable task performance. MND, then, is an atypical case of care because the tasks change faster and they are more advanced.

As noted in the theory chapter, Gittell (2002, 2009) argues that relational coordination is more effective when there is high task interdependence, a high degree of uncertainty and time constraints. Relational coordination relates to facilitating interaction in order to establish shared goals, shared knowledge and mutual respect. The papers in this thesis make important contributions to the goal of establishing collaboration. Paper I contributes to establishing shared goals by illustrating the importance of having a shared understanding of the condition, as this cannot be taken for granted. Paper II underscores the importance of having a shared understanding of care work and appreciation of this work. In this way, both Papers I and II provide conditions for facilitating collaboration, contributing to establishing shared goals and knowledge. This collaboration could otherwise be based on conditions that remain unspoken, private and non-unified. The task-focused relationships described in Paper III point in the direction of Gittell's third condition, mutual respect, and the need for respect when performing advanced care work at home.

8. Conclusion

The overall aim of this study was to contribute knowledge about conditions for collaboration between family caregivers and health professionals managing and caring for individuals with MND.

The first sub-aim was to explore the meaning of chronicity and terminality in MND (Paper I). We found that MND has features of both chronic and terminal illness. A condition for collaboration is for the actors to have a shared understanding of the condition. For instance, collaboration may break down if health professionals suggest initiating palliative care when the family wants to pursue a life on invasive ventilation (or vice versa).

The second sub-aim was to understand more about family caregivers' work and sense of responsibility by exploring their accounts of caring for a family member with MND (Paper II). We found nuances in the lines of care work of family caregivers in MND, and that family caregivers perform a balancing act in trying to meet expectations from self and others while not being overwhelmed by work. A condition for collaboration is for the actors to have a shared understanding and appreciation of care work. For instance, collaboration may break down if health professionals do not recognise the intimacy, fearfulness and intrinsic value that family caregivers ascribe to different types of care work.

The third sub-aim was to explore what challenges health professionals encounter when managing MND in primary health care (Paper III). We found that health professionals reported five challenges managing MND. Conditions for collaboration are the construction of relationships allowing for negotiation of trust and capacity within primary health care to coordinate care, the availability of competent personnel, and the ability to provide personnel with social support in the home setting. For instance, collaboration may break down if health professionals do not build local knowledge about the situation at home and use this knowledge to mobilise the primary health care system to provide tailored care.

MND is a complex trajectory and may be experienced as an overwhelming condition. Family caregivers as well as primary health care run the risk of breaking down while trying to manage it.

Collaboration is necessary to avoid breakdown. The conditions above are meant to aid such collaboration. They are dynamic conditions intended to enhance collaboration, not causal conditions for optimal care.

8.1 Implications for practice

8.1.1 Family caregivers

There are several implications for practice to be drawn from the findings of this thesis that are relevant for family caregivers:

- Family caregivers in MND face the challenge of finding a sustainable role managing a condition that has features of both chronic and terminal illness.
- The findings in this thesis suggest that social support should routinely be offered to family caregivers in MND throughout the trajectory and beyond, for instance in the form of a psychologist or other professional. This third party may contribute with counselling on how to prioritise lines of work at home, how to deal with limbo and establish a sustainable caregiver role, and tactics for using health services.
- Family caregivers need two plans, one with a short-term, and another with a long-term outlook. These plans must be carried out simultaneously and should not be mutually exclusive. It is important to describe the current situation and determine how long it can be sustained.

8.1.2 Health professionals

There are several implications for practice to be drawn from the findings of this thesis that are relevant for health professionals:

- Recognition that the transition from non-palliative to palliative care is ambiguous in MND (Paper I), and that ongoing dialogue between different actors is necessary to coordinate care.

- Lines of work (Paper II) may be used by health professionals as tools to broaden the scope of interpreting family caregiver needs in MND. This approach may aid health professionals in building relationships with those at home, as well as in strengthening capacity within the primary health care system (Paper III).
- Awareness of challenges commonly experienced by health professionals in primary health care in MND (Paper III) may enhance collaboration between health professionals and those at home, as well as within the primary health care system.
- MND is an uncommon condition, which limits the opportunities health professionals have to accumulate experience, especially in primary health care. Multidisciplinary MND clinics in hospitals have the potential to be knowledge banks, providing an arena for the exchange of health professionals between hospitals and primary health care.
- The atypicality of the MND trajectory creates a lack of fit in the institutional structure of primary health care, making it imperative for institutions to cooperate in ways that are out of the ordinary.

8.2 Future research

In this thesis, the home, primary health care and specialised services have been the focus.

- The most central technology contributing to unstable terminality in MND is invasive ventilation. This is not a new technology, having been in use since the 1950s. Future research could use a historical approach to understand the use and non-use of such life-supporting technology.
- Studies of people with MND on LTMV and their families may further explore the concept unstable terminality, especially relating to those choosing to turn of the respiratory support.

- There is need for studies exploring how class and gender may influence tactics and division of labour along lines of work managing MND.
- There is a need to further explore whether expert caregivers are a risk factor for family caregiver exhaustion.
- There is a need for qualitative exploration going beyond cross-sectional interviews of how family caregivers in MND cope.
- Information work tactics could be further explored in conditions other than MND.
- There is a role for theory in untangling the complex social phenomenon that occurs in conditions like MND. Research that builds theory may contribute to better understanding, more precise interventions, and fewer futile attempts to improve a difficult situation.
- Further studies should explore what narratives health professionals facilitate in MND and other conditions. This is especially related to doctors, neurologists and others in specialist multidisciplinary MND clinics.

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10. Appendix

10.1 Interview guide at home

Personalia

Sivilstatus og relasjon til den med ALS. Alder og arbeid. Bosted og boforhold.

Om ALS

Første symptomer. Diagnostisering. Informasjonsinnhenting. Hvordan går det nå?

Hjemmet

Hva slags tilpassninger er det snakk om?

Hvordan vil man ha det? Vil man være hjemme eller i institusjon?

Forventninger. Hvordan hadde du håpet det skulle være, om du kunne få det helt som du ville?

Hva får du hjelp til?

Personlighetsforandringer.

Stemning i hjemmet.

Kommunehelsetjenesten

Hva slags relasjon? Kommer de hjem? Hva slags kontaktpunkt har du?

Hva er viktigst? Hjemmesykepleie. Fastlege. Tekniske hjelpemidler. Individuell plan.

Forventninger. Rutiner

NAV og hjelpemiddelsentral

Hvordan er det med dem? Har de forståelse?

Alternativ behandling

Relevant? Hvorfor? Hvordan kom dere i kontakt?

Sykehuset

En typisk konsultasjon.

Hvordan er stemninga?

Forventninger.

Hva er du opptatt av å få svar på der?

Hva slags kontakt har du? Bare under konsultasjoner, eller ellers også?

Hvem i teamet synes du er greiest at er der?

Pårørenderollen

Har du noen ALS-frie rom?

Har du en hobby?

Spurte du noen om hjelp om det?

Får du sove om natta?

Hvem passet på deg da?

Hvordan gikk det med helsa di?

Vil du si at du er en annen i dag, enn du var før du ble kjent med denne sykdommen?

Til slutt

Hvordan vil du oppsummere utviklinga?

Hvilke institusjoner har vært viktige?

Hva ville du gitt i råd til noen andre i en liknende situasjon?

Er det noe viktig du føler at du ikke har fått sagt?

10.2 Interview guide primary health care

Hva synes du er god ALS omsorg?

Hva ser du for deg er en bra rolle for deg å ha i så måte?

Arbeidsfellesskap og din rolle

Hva er dine oppgaver omkring ALS?

Har du kolleger med samme erfaring som deg?

Kan du fortelle om de viktigste som du samarbeider med?

Er det noe spesielt du er særlig fornøyd med? Noe du savner?

Om sykehuset

Fortell hvordan du opplever møte med sykehuset. Har du vært med en pasient noen gang?

Spør du dem om råd? Er de tilgjengelige?

Hvordan er stemninga der?

Hvem synes du er greiest at er der i teamet?

Om kommunehelsetjenesten

Hjemmetjeneste

Sykehjem

Bestillerkontor

Nav

Hjelpemiddelsentral

Fastlege

Hjemmet

Fortell litt om hvordan det er å jobbe i et ALS-hjem.

Hvilke oppgaver er det du gjør? Praktisk? Eksistensielt? Psykologisk?

Hvordan forholder du deg til resten av familien?

Hvis pasienten og pårørende vil forskjellige ting, hva gjør du da?

Å trenge seg på. Hvordan er det å arbeide i noens hjem? Når mener du det er riktig å intervenere i en familie?

Hvilke tilpasninger mener du det er viktig å gjøre i hjemmet for at du skal få gjort jobben din skikkelig?

Hvordan håper du det skal være?

Hvordan er stemninga i huset?

Har du erfaringer med kognitiv svikt?

Hvordan mener du at en kan ta vare på pårørende?

ALS som sykdom

Hvis du skal fortelle til noen som ikke visste noen ting hva ALS er, hva ville du lagt særlig vekt på?

Hvordan prate om døden? Det er ingen kur. Hva er det som er målet med arbeidet da?

Tidsorisont. Hvor langt fram kan du som helsepersonell se? Hvordan forholder dette seg til hvor langt fram pasienter og pårørende ser? Hva er passe informasjon synes du?

Til slutt

Hvis du skulle gitt et råd til noen som skulle begynne i din jobb. Hva ville vært de viktigste tingene?

Av alle de forskjellige som er med å jobber med ALS. Hvem vil du si er viktigst?

Er det noe viktig du føler at du ikke har fått sagt?

Personalia

10.3 Interview guide hospital

Presentasjon av prosjektet

Hva synes du er god ALS omsorg?

Hva tenker du et ALS team på et sykehus kan bidra med?

Og hva tenker du om din rolle i et slikt team?

Har du noen spørsmål før vi starter?

Teamet og din rolle

Hvordan synes du et ALS team skal fungere? Hva er det dere tilbyr og til hvem?

Kan du fortelle om en typisk konsultasjon?

Hva er dine oppgaver teamet?

Er det noe du opplever som spesielt givende med å jobbe med ALS? Er det noe som er spesielt vanskelig? Når er du spesielt fornøyd etter en konsultasjon?

Hva har du lært om ALS av å jobbe i dette teamet? Hva er viktigst, erfaringene fra jobb, eller utdanning og tidsskrifter?

Er det noe du savner i teamet? En type kompetanse, eller yrkesgruppe?

Er det noen ting dere ikke er opptatt av? Har dere en slags grense for hva dere er opptatt av her?

Prater dere om at man noen ganger ikke skal gi behandling?

Hvem, utenfor teamet, er det dere jobber tettest med? Hvilke avdelinger samarbeider dere med, og om hva?

Hvem er det du og dere jobber med utenfor sykehuset?

Pasient og pårørende

Fortell litt om noen typiske pasienter. Fortell om en flink pasient du har vært borti en gang.

Fortell litt om noen typiske pårørende. Hva forventer du av pårørende? Fortell om en særlig god pårørende du har vært borti?

Hvis du skulle fortelle om ALS til noen for første gang, hva ville du lagt vekt på?

Hvordan opplever du møtet med pasienter og pårørende? Er det noe som går inn på deg?

Har du erfaring med om pårørende og pasienter ikke er enige?

Hvordan ser du for deg at pasienten og pårørende har det hjemme hos seg selv? Er det neon spesielle hjemmeforhold du biter deg merke i?

Hva vil du si er et typisk pasientforløp i deres team? Hva tenker du på i ulike faser av forløpet? Hvordan skiller et slikt typisk forløp seg fra noen av de siste forløpene og konsultasjonene du hadde?

Noen pasienter og pårørende sier i blant 'det er ikke lett å ta imot hjelp'. Hvorfor tror du de sier det? Hva tror du er det vanskelige?

Kommunehelsetjeneste

Fortell om den typiske kontakten du har med kommunehelsetjenesten. Hva slags kontakt har du? Og med hvem?

Når er du fornøyd etter å ha vært i kontakt med kommunehelsetjenesten? Hva skal de gjøre for at din jobb skal bli grei? Hva forventer du av dem?

Hvis du skulle forklart hva ALS er til en i kommunehelsetjenesten, hva ville du lagt vekt på da?

Til slutt

Føler du at du får gjort det du egentlig skal gjøre? Hva er det egentlige? Og hva er det som eventuelt kommer i veien?

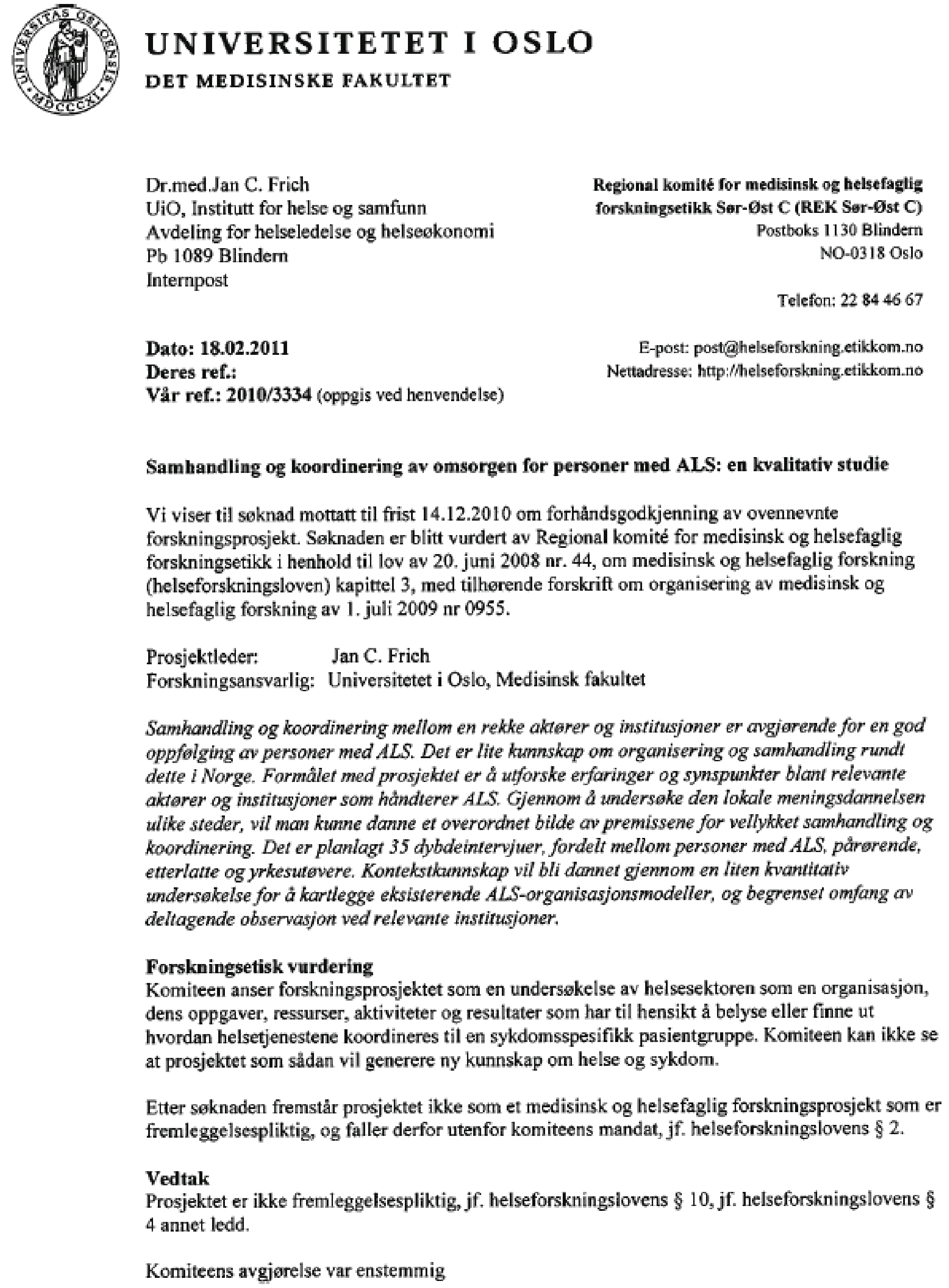
Hva ser du som de viktigste tingene å jobbe med framover knyttet til ALS? Hva er utfordringene?

Hvis du er i et selskap, hvordan ville du forklart om ALS og jobben din da?

Er det noe viktig du føler at du ikke har fått sagt?

10.4 Ethical approval – NSD – REK

10.4.1 Exempt from review from the Regional Ethics Committee



UNIVERSITETET I OSLO
DET MEDISINSKE FAKULTET

Dr.med.Jan C. Frich
UiO, Institutt for helse og samfunn
Avdeling for helseledelse og helseøkonomi
Pb 1089 Blindern
Internpost

Regional komité for medisinsk og helsefaglig
forskningsetikk Sør-Øst C (REK Sør-Øst C)
Postboks 1130 Blindern
NO-0318 Oslo

Telefon: 22 84 46 67

E-post: post@helseforskning.etikkom.no
Nettadresse: http://helseforskning.etikkom.no

Dato: 18.02.2011
Deres ref.:
Vår ref.: 2010/3334 (oppgis ved henvendelse)

Samhandling og koordinering av omsorgen for personer med ALS: en kvalitativ studie

Vi viser til søknad mottatt til frist 14.12.2010 om forhåndsgodkjenning av ovennevnte forskningsprosjekt. Søknaden er blitt vurdert av Regional komité for medisinsk og helsefaglig forskningsetikk i henhold til lov av 20. juni 2008 nr. 44, om medisinsk og helsefaglig forskning (helseforskningsloven) kapittel 3, med tilhørende forskrift om organisering av medisinsk og helsefaglig forskning av 1. juli 2009 nr 0955.

Prosjektleder: Jan C. Frich
Forskningsansvarlig: Universitetet i Oslo, Medisinsk fakultet

Samhandling og koordinering mellom en rekke aktører og institusjoner er avgjørende for en god oppfølging av personer med ALS. Det er lite kunnskap om organisering og samhandling rundt dette i Norge. Formålet med prosjektet er å utforske erfaringer og synspunkter blant relevante aktører og institusjoner som håndterer ALS. Gjennom å undersøke den lokale meningsdannelsen ulike steder, vil man kunne danne et overordnet bilde av premissene for vellykket samhandling og koordinering. Det er planlagt 35 dybdeintervjuer, fordelt mellom personer med ALS, pårørende, etterlatte og yrkesutøvere. Kontekstkunnskap vil bli dannet gjennom en liten kvantitativ undersøkelse for å kartlegge eksisterende ALS-organisasjonsmodeller, og begrenset omfang av deltagende observasjon ved relevante institusjoner.

Forskningsetisk vurdering
Komiteen anser forskningsprosjektet som en undersøkelse av helsesektoren som en organisasjon, dens oppgaver, ressurser, aktiviteter og resultater som har til hensikt å belyse eller finne ut hvordan helsetjenestene koordineres til en sykdomsspesifikk pasientgruppe. Komiteen kan ikke se at prosjektet som sådan vil generere ny kunnskap om helse og sykdom.

Etter søknaden fremstår prosjektet ikke som et medisinsk og helsefaglig forskningsprosjekt som er fremleggelsespliktig, og faller derfor utenfor komiteens mandat, jf. helseforskningslovens § 2.

Vedtak
Prosjektet er ikke fremleggelsespliktig, jf. helseforskningslovens § 10, jf. helseforskningslovens § 4 annet ledd.

Komiteens avgjørelse var enstemmig

Komiteens vedtak kan påklages til Den nasjonale forskningsetiske komité for medisin og helsefag, jf. Forvaltningslovens § 28 flg. Eventuell klage sendes til REK Sør-Øst. Klagefristen er tre uker fra mottak av dette brevet.

Med vennlig hilsen

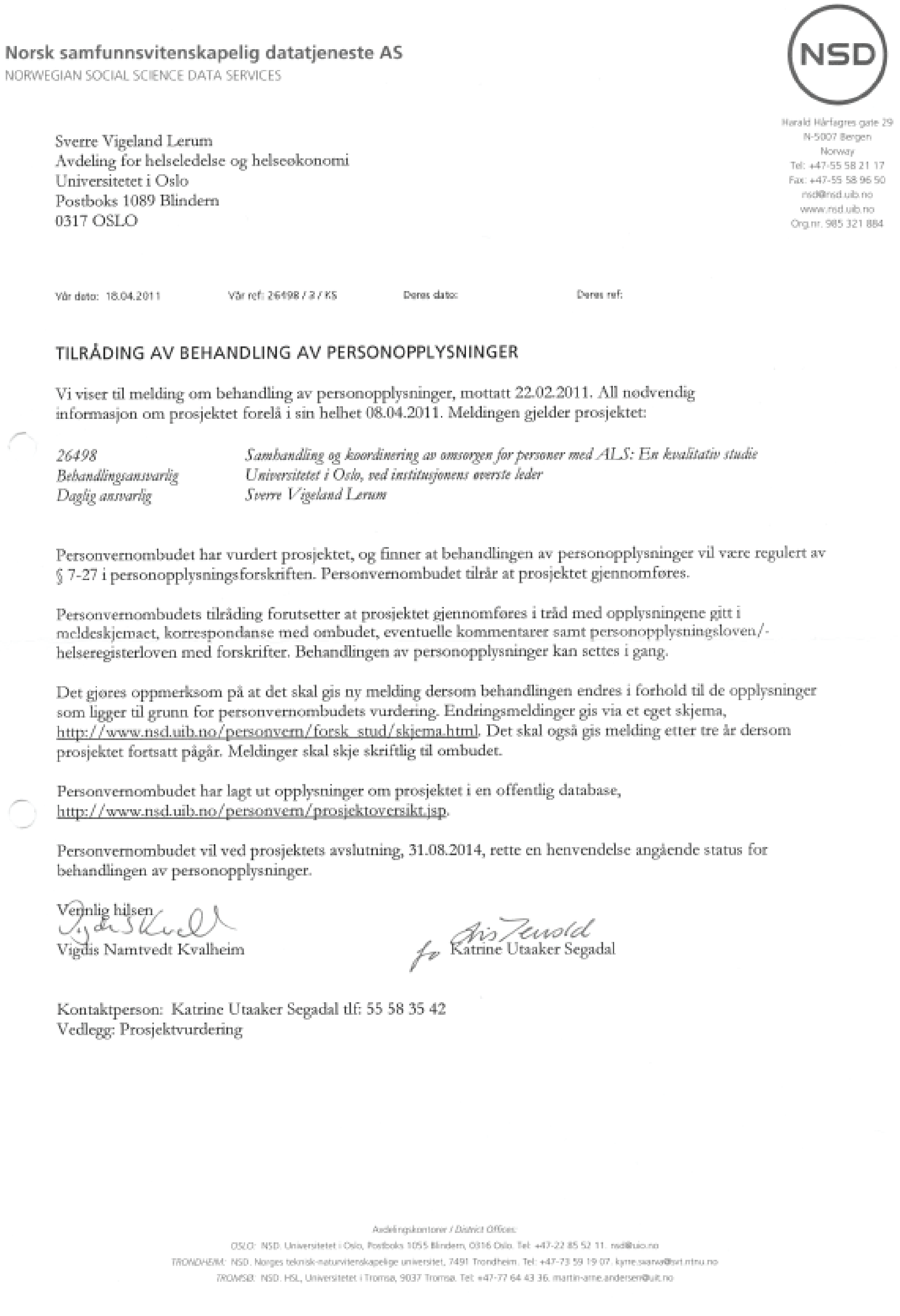
Arvid Heiberg (sign.)
professor dr. med.
leder

Øyvind Grønlie Olsen
Øyvind Grønlie Olsen
jurist/ rådgiver

Kopi: Universitetsdirektøren, universitetsdirektørens kontor, Pb 1072 Blindern, internpost

Vi ber om at alle henvendelser sendes inn via vår saksportal: http://helseforskning.etikkom.no eller på e-post til: post@helseforskning.etikkom.no. Vennligst oppgi vårt saksnummer/referansenummer i korrespondansen

10.4.2 Approval from Norwegian Centre for Research Data



Norsk samfunnsvitenskapelig datatjeneste AS
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Vår dato: 18.04.2011 Vår ref: 26498 / 3 / KS Deres dato: Deres ref:

TILRÅDING AV BEHANDLING AV PERSONOPPLYSNINGER

Vi viser til melding om behandling av personopplysninger, mottatt 22.02.2011. All nødvendig informasjon om prosjektet forelå i sin helhet 08.04.2011. Meldingen gjelder prosjektet:

26498	*Samhandling og koordinering av omsorgen for personer med ALS: En kvalitativ studie*
Behandlingsansvarlig	*Universitetet i Oslo, ved institusjonens øverste leder*
Daglig ansvarlig	*Sverre Vigeland Lerum*

Personvernombudet har vurdert prosjektet, og finner at behandlingen av personopplysninger vil være regulert av § 7-27 i personopplysningsforskriften. Personvernombudet tilrår at prosjektet gjennomføres.

Personvernombudets tilråding forutsetter at prosjektet gjennomføres i tråd med opplysningene gitt i meldeskjemaet, korrespondanse med ombudet, eventuelle kommentarer samt personopplysningsloven/-helseregisterloven med forskrifter. Behandlingen av personopplysninger kan settes i gang.

Det gjøres oppmerksom på at det skal gis ny melding dersom behandlingen endres i forhold til de opplysninger som ligger til grunn for personvernombudets vurdering. Endringsmeldinger gis via et eget skjema, http://www.nsd.uib.no/personvern/forsk_stud/skjema.html. Det skal også gis melding etter tre år dersom prosjektet fortsatt pågår. Meldinger skal skje skriftlig til ombudet.

Personvernombudet har lagt ut opplysninger om prosjektet i en offentlig database, http://www.nsd.uib.no/personvern/prosjektoversikt.jsp.

Personvernombudet vil ved prosjektets avslutning, 31.08.2014, rette en henvendelse angående status for behandlingen av personopplysninger.

Vennlig hilsen

Vigdis Namtvedt Kvalheim

for Katrine Utaaker Segadal

Kontaktperson: Katrine Utaaker Segadal tlf: 55 58 35 42
Vedlegg: Prosjektvurdering

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