

**“AS YOU CAN SEE, WE PLOD ALONG”**

**NARRATIVES OF LIVING WITH MOTOR  
NEURONE DISEASE IN WALES**

This thesis is submitted at Cardiff University for the  
award of the degree of Doctor of Philosophy

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**DECLARATION**

This work has not been submitted in substance for any other degree or award at this or any other university or place of learning, nor is being submitted concurrently in candidature for any degree or other award.

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## Abstract

This dissertation is the outcome of a 25-month long narrative inquiry-based study on experiences of living with motor neurone disease (MND). MND is an adult onset, incurable, neurodegenerative condition that is characterised by loss of voluntary muscle movement as a result of destruction of motor neurones, leading gradually to partial or complete paralysis and eventually to death. The aim of this study was to explore the experiences of people living with MND. Following a narrative inquiry methodology the focus of the study was to explore how specific people live in their local contexts. Data were collected through the use of multiple semi-structured interviews with people with MND and some of their partners. The findings illustrate the unique ways in which people with MND experience the disease and make sense of their life. The seven people who participated in this study were trying to construct a notion of normality in their everyday life, in the midst of what were sometimes perceived as difficult or even abnormal circumstances. Through seeking and trying out different solutions to the challenges they were facing, participants were trying to create a life they could describe as good. The findings highlight the intersubjective nature of illness experiences. For the three couples who participated in the study the experience of being in a long-term relationship was a vital part of who they were, and how they experienced MND. The findings also underline the importance of exploring the experiential knowledge that people living with MND develop through managing the disease and incorporating it in various ways in their daily life. This knowledge can help create a kind of life that people living with MND feel is worth living.

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**LIST OF ABBREVIATIONS**

AACD	Alternative and augmentative communication devices
ALS	Amyotrophic lateral sclerosis
DoH	Department of Health
EMG	Electromyography
GP	General practitioner
NIPPV	Non-invasive positive pressure ventilation
MND	Motor neurone disease
MNDA	Motor neurone disease association
NHS	National health service
PBP	Progressive bulbar palsy
PEG	Percutaneous endoscopic gastrostomy
PLS	Primary lateral sclerosis
PMA	Progressive muscular atrophy
RIG	Radiologically-inserted gastrostomy
UK	United Kingdom
USA	United States of America

## KEY TO TRANSCRIPTION SYMBOLS

Square brackets [] indicate ..... Text added by the researcher  
 within the quotation (for clarity  
 or to offer information on non-  
 verbal elements).

A series of three full stops  
 within brackets (...) indicates ..... Text that has been removed.

A series of three full stops ... indicates a .... Pause.

Double quotation marks “”  
 in block quotations indicate ..... Quotations within the quotation.

Single quotation marks ‘’ in  
 in-the-text quotations indicate ..... Quotations within the quotation.

**LIST OF DEFINITIONS**

- Amyotrophic lateral sclerosis..... A subtype of motor neurone disease.
- Dysarthria ..... Difficulty with the production of oral speech.
- Dysphagia ..... Difficulty with swallowing.
- Gastrostomy..... An opening through the abdominal wall and into the stomach, through which people with motor neurone disease sometimes receive food, water, and medication.
- Hypercapnic coma..... A condition in which the level of carbon dioxide (CO<sub>2</sub>) in the blood is too high and vital body functions cannot be maintained.
- Lightwriter ..... A text-to-speech device that synthesises voice from text input.
- Motor neurone disease ..... An adult onset, incurable, progressive, neurodegenerative condition of unknown aetiology, which can lead to partial or complete paralysis.

Percutaneous endoscopic

gastrostomy ..... The opening of a gastrostomy and  
insertion of feeding tube through the  
abdominal wall.

Primary lateral sclerosis ..... A subtype of motor neurone disease.

Progressive bulbar palsy ..... A subtype of motor neurone disease.

Progressive muscular atrophy ..... A subtype of motor neurone disease.

## 1. Introduction

### 1.1 Background

Health in an absolute sense is often unattainable. Advances in healthcare, increased life expectancy, and an associated increased prevalence of disability and chronic illness have led to many people needing to learn how to live with the ongoing effects of chronic illness or disability (Mol, 2006a). Motor neurone disease (MND), also known as amyotrophic lateral sclerosis or Lou Gehrig's disease, is an incurable, neurodegenerative disease of unknown aetiology. People with MND are "*not just sick, but inexplicably sick*", to use Wikan's words (2000, p.215). The average survival expectancy after diagnosis is three to five years, which can however be up to thirty or more years depending on the type of MND. MND can lead to complete paralysis of voluntary muscles, affecting functions such as walking, eating, talking, and breathing. The voices of people living with MND often remain unheard in research although their knowledge about how they live with MND in their local contexts could offer valuable insights into the experience of MND.

MND is often represented in an emotive way. A recent awareness campaign by the British motor neurone disease association (MNDA) presented the disease as a body snatcher; it hits suddenly and unexpectedly, leaving you powerless, trapped in a dead body (MNDA, 2010). An older campaign by the American MND association depicted MND as a fast travelling bullet, unstoppable and bound to kill, unless some new form of

defence stops it (The Amyotrophic Lateral Sclerosis Association, 2008). Sontag (1991) pointed out the fact that chronic illness, and especially incurable illness, is often given names that belong to something else, turning illness into a metaphor. These metaphors are usually laden with negative value. Illness is turned into guilt, pain, shame, death, loneliness and existential angst. MND is turned into a murder weapon. In the absence of a cure and with no hope for recovery, studies exploring the experience of living with MND often emphasise loss and suffering, presenting narratives of tragedy and implying the impossibility of a happy ending. In these stories, the central characters cannot live a good life, ever after.

A good life, however, can refer to “*possible worlds and possible selves worth striving for*” (Mattingly, 2008, p.95). People with MND often need to learn how to combine a life with an incurable disease that leads to paralysis and reduced life expectancy, and a life that is worth living. There is currently a gap between the biomedical knowledge on disease processes and the lived experience of living with MND (Brown, 2003; Brown, Lattimer, & Tudball, 2005; Hughes, Sinha et al., 2005; Kleinman & Hanna, 2008). Furthermore, the literature suggests that the needs of people living with MND are not always effectively met (Brown, Lattimer, & Tudball, 2005; Hughes, Sinha et al., 2005; van Teijlingen, Friend, & Kamal, 2001). In a comprehensive study of service use and needs of people living with MND in Scotland, van Teijlingen, Friend and Kamal (2001) found that only 25% of the informants felt that statutory services completely responded to their needs.

There has been limited research exploring the experiences of people living with MND (see for example, Cox, 1992; Gysels, Shipman, & Higginson, 2008a; Hughes, Sinha et al., 2005; Robinson & Hunter, 1998). Writing on the process of learning how to live with an illness, Manderson (2011) states that

In the face of illness, people search for a diagnosis; to name a condition is to begin to control it. The second step is to search for a cure, taking advantage of the proliferation of health services available from complementary and alternative modalities and cosmopolitan medicine (p.242).

What happens after these two first stages, and when people with MND realise there is no cure available, is unique to each person. People living with MND require access to a wide range of healthcare services and their care needs change gradually but constantly. Individuals, their families and friends, and healthcare professionals embody unique belief systems and have their own ideas of how life is to be lived, what needs to be done, how, and why (Toombs, 1992). These different perspectives are in constant negotiation in the context of daily life with everybody involved in a dynamic dialogue, trying to establish a common ground for understanding what is good and what has to be avoided (Letiche, 2008).

Currently there is not enough knowledge on how individuals themselves experience life with the disease. This study focuses on the following identified gaps in knowledge:

1. The limited information regarding daily life experiences of people living<sup>1</sup> with MND.
2. The lack of knowledge on how people make sense of their life with the illness.

## **1.2 Research questions**

The main research questions are the following:

1. How do people experience life in the context of MND?
2. How do people living with MND make sense of their life?

## **1.3 Aim and objectives**

The overall aim of this study is to add to existing research by exploring experiences of living in the context of MND. The specific objectives are:

1. To explore the experiences of daily life from the standpoint of seven people (three couples and one person participating by herself) living with MND in Wales.
2. To foreground<sup>2</sup> how these people make sense of their life in the context of living with MND.

Although at the outset of the study the objective was to explore the individual experiences of the seven participants, in the process of the study this objective was modified, as a result, it is more accurate to describe participants as being three couples and one person (the latter, while part of a

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<sup>1</sup> The phrase ‘people living with MND’ is used in this study to refer to people diagnosed with the disease and their partners.

<sup>2</sup> The word ‘foreground’ as used in this thesis reflects Mattingly’s (2010) use of the word and refers to a process whereby participants’ experiences are brought to the fore and become the main focus of the study.

couple she chose to participate in the study without her partner). This is discussed in more detail in paragraphs 5.2, 5.3.1.2 and 12.3.2.

#### **1.4 Significance**

This study aims at illuminating experiences of daily life from the standpoint of people with MND and their partners. The knowledge acquired through this research has the potential to further health professionals' understanding of MND, thus leading to improved quality of healthcare. Due to limited information and relevant knowledge base health professionals often make assumptions about the impact of MND on people's daily life. The production of knowledge on individuals' experiences of living with MND is necessary for the development of healthcare practices that can respond to the needs of people living with MND. Hughes, Sinha et al. (2005) recommend that developing an understanding of the condition and its impact on people's life is necessary in order to inform healthcare practices. More specifically, the knowledge produced through this study has the potential to:

- Enhance understanding on the meaning and process of daily life in the context of MND.
- Enhance understanding on how individuals make sense of their life with the illness, which can contribute to existing knowledge on desirable outcomes of healthcare.

### **1.5 Theoretical standpoint**

In this paragraph, I offer a brief outline of my theoretical standpoint. As an occupational therapist with an underpinning knowledge of occupational science, I have a particular interest on how people live their life. My professional and disciplinary background means that I am attuned to issues such as the structure of daily life or the enactment of social roles through occupations. The exploration of these issues, however, was not the main aim of this study. Rather, I wanted to explore daily life with a disability from a broader perspective, focussing on how people make sense of their life in their own local context, in the presence of a disability. Therefore, in this thesis I needed to build theoretically on my professional background in order to broaden the understanding of living with a disability.

This study draws from and intends to contribute to knowledge development in a variety of fields, which all come together under the rubric of disability studies. Disability studies is an inherently transdisciplinary field, whose aim is to produce knowledge about disability, often placing disabled people at the centre of the inquiry by exploring how disability is constructed, imposed or enacted (Shakespeare, 2006). This thesis is specifically based in this broad theoretical field.

My theoretical standpoint was further refined by the use of postmodernism. Ideas about the diffusion of power in society and its operation through and on bodies, and ideas about the heterarchical organisation of knowledge were crucial for my understanding of the participants' stories and the construction of their narratives. The two main

theorists I drew from were Foucault and Lyotard. Foucault (1994a, 1994c) developed a postmodern theory of biopolitics, dealing with power and how it is exercised on people but also by people. In biopolitics, people can be both acting subjects, who exercise power, and objects on whom power is exercised. Using biopolitics as an analytical lens enabled close attention to people and how disability was not only enacted but also constructed within their local contexts.

In order for the analysis to be sensitive to the personal experiences of the participants, and at the same time situate them within a broader context, my theoretical standpoint was further influenced by Lyotard and in particular his ideas on knowledge legitimation. According to Lyotard (1984), there is no single truth, no grand narrative waiting to be explored, but knowledge is heterarchical. This enabled me to listen to participants' stories and rather than try to defend or justify them, I engaged in a process of description and explanation.

## **1.6 Organisation of thesis**

This thesis consists of twelve chapters. Chapter 1 presents the research aim and the background to it and outlines the theoretical framework that guided this thesis, while chapter 2 foregrounds my own involvement in data production and construction of narratives. Chapter 3 presents a review of the relevant literature. In order to give a comprehensive overview of existing knowledge relevant to the research questions, the review is focused on two broad types of literature: biomedical knowledge and illness experiences.

Chapter 4 discusses the use of narratives in this thesis and chapter 5 gives a detailed account of the processes I followed in the study, and justifies them. Chapter 6 introduces the four narrative chapters. Chapters 7-10 present the participants' narratives, whereas in chapter 11 I discuss the findings in relation to the objectives of the study. Finally, chapter 12 draws the main conclusions of the study and presents the implications of the study findings for healthcare practice. Chapter 12 also discusses the implications of the study for the development of research methodologies sensitive to capturing illness experiences and practices of care.

The use of technical language has been kept to a minimum throughout this thesis. Where it was necessary, some terms have been used and these are explained where they occur in the text. Some terms appear more often as they relate to experiences several of the participants discussed. These terms are compiled and explained in Appendix A.

Throughout the thesis I use the first pronoun to refer to myself, the researcher. This is not a stylistic choice but an epistemological one, wanting to acknowledge my role in the construction of the data and the narratives. Using Papadimitriou's (2008a) words, the *I* used in this thesis is the "*I of the beholder*" (p.216), who co-produced and analysed the data, and wrote the narratives.

## 2. Establishing the researcher's *I*

The narratives presented in this study were produced from the interaction between the study participants and myself. Because of the interactive nature of the methodology, I use the first personal pronoun *I* to refer to myself, the researcher, throughout this thesis, pointing attention to my involvement in the construction of the narratives. For this reason, the narratives would be incomplete if I did not present my *I* and my personal motivation for this study.

As an occupational therapist working first in Greece, then in Japan, and finally in the United Kingdom, I was no stranger to incurable, progressive diseases and the ways people make sense of them, at least during their interactions with healthcare professionals. Guided by the specific circumstances of each person I would interact with, I would offer some equipment, arrange for some adaptations, perhaps link in with other relevant services, and organise some sort of support; the situation often seemed manageable and in some strange way straightforward to me. I would interact with people for a brief time, catching a glimpse of their daily life, listen to the difficulties they faced and I would come up with some kind of solution, some way to make life just a little bit better. I was, however, largely unaware of what was going on outside of the brief interactions we had and I could not know how these people experienced everyday life.

I became acutely aware of this knowledge gap when I had to deal with disease in my own everyday life. My mother was affected by the bulbar form of MND. The first sign, dysarthria, appeared in early 2008, and

soon after the rest of her body became weaker and her movements slower. She lived with MND for three years during which time myself, my brother, and my father cared for her at home. While the study on how people with incurable diseases was conceived before my mother's diagnosis, it acquired a moral urgency when MND presented itself in my life. Questions about what is a good life, and conversely what is not a good one, became important and real.

After the first year, progression was fast; my mother's oral speech was gone first and then her arms became weaker, followed by the breathing muscles and legs. She could hold a pen or type on a mobile phone until a couple of months before she died in late 2010, so she could still communicate. Between weakening limbs, a tongue that was not doing what it was supposed to, and various devices, from ventilation machines to suction devices, where was my mother? How do all these different parts come together to constitute a body, and a self, and how is this body experienced, as Merleau-Ponty (1962/2002) asked.

I still have tens of thousands of words typed in text messages that we exchanged in the years of silence. It happened so gradually, yet so fast, that I cannot remember when was the last time I heard her voice. Perhaps it was in a phone call when unable to understand what she was saying, I had asked her to repeat a word a couple of times. Or perhaps it was when she last visited me in Wales and I was translating back and forth between her and my then partner.

With the speech gone and with the arms getting weaker, soon to be followed by breathing and legs, we (my father, brother, and myself) were all at a loss. Family and friends were there to help, each in their own way, but we were all struggling with one big question, and in particular my mother and the immediate family: what now? What can be done now to make life better, to lead to a positive outcome, and what that positive outcome might look like? Much as I like happy endings, I cannot say we found satisfactory answers to these questions. We each had our own understanding of the situation and our own ways of dealing with it. For my mother, a positive outcome was to keep her body intact, which meant no PEG. This was hardly a positive outcome for me, as I was afraid it would lead to malnutrition and eventually starvation.

As my mother's condition progressed, we had to constantly adjust to giving a bit more, but not too much, help. First, it was enough to help her cut her food. Then somebody had to carry her plate to the dining room. Later on, somebody had to be there with her always because of the fear of choking (I had to perform the Heimlich manoeuvre twice). When she could no longer eat solid food, initially she would just drink a high-energy food supplement and mix other supplements in it. Later on somebody else had to do the mixing. Then, as the muscles in her forearms and the hands became weaker, it was about positioning the mug in her hands. When arm and shoulder musculature was too weak to help her lift her arms so that she could reach her mouth, we would first flex her forearms and slightly elevate her arms and then place the mug in her hands. Weakened neck musculature

later on meant that two people were needed during meal times; one to help with the mug and positioning of her arms and forearms and another to protect her head by holding one hand behind it to prevent it from suddenly falling backwards (collars and chairs with neck support were not comfortable or suitable).

None of this was pleasant, desired or even anticipated. However, this was not a story of suffering, although suffering was certainly part of it, sometimes more than others. Instead, this was a story of living with MND, making the best one can, trying to construct the best life one can under the circumstances. It was a relational good we were trying to construct, while the good life remained elusive.

This personal experience was an inspiration to explore how people with MND experience life. Having lived with my mother through part of her experiences with this disease, I could see, and later on read in the literature, how the experience of living with MND is often not understood, not only by health professionals, but also by the immediate social network. Partly this is because of the unique nature of the disease. MND is incurable, often progresses fast and often leads to speech impairment and resulting communication problems (Eisen, 2009). In other words, people with MND often do not get the chance to share their story. The limited understanding of the experience of living with MND, is also partly because of the different vantage points that different people occupy, so that it becomes difficult to talk about *the* experience of living with MND and instead it is more accurate to focus on a multitude of experiences. I often found that what I thought was

best for my mother, or how I thought she experienced life, were my own interpretations of the situation and her own interpretation would be different. My motivation for this study was to help some people with MND get their voice heard and have their experiences listened.

## **2.1 Proximity**

Proximity has for a long time been a contested issue in qualitative research. In this study, proximity was perhaps further complicated because of its rather unilateral nature. On the surface, and from the standpoint of the research participants, I was far removed from the experiences I was exploring. In categories such as age, ethnicity, and health status, I could not identify with the research participants, neither could they with me. However, my own personal experiences led to the construction of a perhaps unilateral proximity, as I did not disclose my experiences to the participants, for the reasons outlined in paragraph 5.6.1. Not disclosing my experiences was a very difficult decision and it is discussed in more detail in paragraph 12.6.

Starting off with data collection a few months after my mother's death, I was afraid that the door to a participant's home would open and in there sitting in the front room all propped up and smiling, speaking silently with her eyes, would be my mother. This did not happen. Perhaps the distance (the study was conducted in Wales, UK, while my mother lived in Greece) and the different language created a safety buffer and made distancing in my involvement easier.

These, however, were superficial differences. Perhaps more crucial for my distancing was that it was not possible to escape the uniqueness of each participant, and how essentially different they all were. As Throop (2010) emphasised, homologous experiences do not necessarily lead to a shared understanding about the meaning of these experiences. On the other side of the door, there were people, each unique, each within a specific context, each of them living with MND in their own way, different to everybody else. On the other side of the door, I encountered the singularity of experience, as I confronted the “*impenetrability of others’ and our [my] own subjective lives*” (Throop, 2010, p.771). Living in the same village or in different countries, everybody has their own way of living life, with or without illness. Perhaps when somebody has to live with an illness such as MND- incurable, progressive and terminal-, often leading to partial or complete paralysis, the unique nature of the way one wants to live their life acquires an urgency that was not there before. But, what that way is, what it should look like and what decisions might lead to it, are deeply individual issues. As Jackson (2013) reflected, understanding another person entails “*the loss of the illusion that one’s own particular worldview holds true for everyone*” (p. 11).

My *I* represents my own particular worldview and is not an authoritative *I*. My *I* does not assume knowledge, does not share the experiences of the participants in this study and cannot assume or guess what their daily life looks like. Instead, my *I* tries to approach the

experiences of the research participants and sometimes struggles to make sense of them.

While I was near the research participants, in the sense that I could relate to some of their experiences (Jacobsen, 2007), the proximity was cursory and I cannot claim I achieved empathy if it refers to feeling what somebody else feels (Fainzang, 2007). I was ‘between one and one another’ to borrow Jackson’s (2012) phrase. Relating emotionally to the experiences of the participants did not mean that I could understand or feel what they were feeling.

Proximity can sometimes lead to an illusion of understanding other people’s experiences if they are homologous to one’s one, thereby “*reducing the irreducibility of another’s self-experience to the self-sameness of my [one’s] own being*” (Throop, 2010, p.777). Sometimes during the research process, I became aware of signs that I might have been doing that, as described, for example, in paragraph 10.5. The nature of the experiences the participants had were removed from my own experiences as a young man, working full-time, and having in the past been a part-time carer for my mother. Essentially, our experiences were all different in their uniqueness.

Dwyer and Buckle (2009) argue that researchers can never be fully neutral and they need to be aware of their own position in terms of what they are exploring. I reflected on my insider-outsider position throughout the study in order to illustrate my own influence on the findings. The main function of proximity in this study was the establishment of a sense of moral urgency. Moral urgency means that exploring the experiences of the

research participants was urgent, not in terms of time but in terms of morality and establishing how they make sense of their lives. Proximity, to the degree it was present, enabled me to see how important it was for the research participants to be able to construct a life that followed their own, unique notion of what was good and what was desired. Similar to other researchers, who have studied and written about experiences that are close to them (for example, Berger, 2013; Kleinman & Hanna, 2008; Kleinman & van der Geest, 2009; Verwey, 2010), I could relate not only on an intellectual level, but also emotionally, to the experiences shared by the participants, without however assuming that I could feel what they felt.

### **3. Literature review**

#### **3.1 Introduction**

The aim of this chapter is to provide background knowledge on what is currently known about the experiences of people living with MND. Existing literature on this subject can be divided in two main categories: 1) literature concerning scientific knowledge about the disease and how it can affect people, and includes diagnosis and symptoms, quality of life, and pathogenesis of the disease, and 2) literature presenting how MND is actually experienced by people. The first category takes MND as its focal point, while the second focuses on people.

The following paragraph explains the choice to present literature from both these categories, rather than just concentrate on illness experiences, which is the focus of this study. The subsequent paragraphs present and review the literature.

#### **3.2 Illness experience and disease process**

When discussing illness and disability experiences, researchers often talk of a physical body and a lived or phenomenological body to point to the different ways that illness can be experienced. The lived body is essentially one's identity, linked to the surrounding social, cultural, physical, and other environments and to one's previous experiences. The physical body is the biological body, disconnected from the surrounding socio-cultural environment. Several authors are moving beyond the lived body/physical body divide and acknowledge the body in all its complexity. Echoing the

medical philosopher Annemarie Mol's (2006a) argument that people have multiple bodies, research conducted by Longhurst (2001) and Papadimitriou (2008b), for example, takes into account multiple dimensions of the body.

Furthermore, the medical anthropologist Byron Good (1994) states that

Disease occurs not only in the body- in the sense of an ontological order in the great chain of being- but in time, in place, in history, and in the context of lived experience and the social world. Its effect is on the body in the world! (p.133).

In other words, disease is not only a process but also a unique experience (Good & Good, 2000).

This study acknowledges that people live within historical, political, and social contexts that influence the experience of living with MND. It also recognises that people engage with the world around them through their physical body and therefore how MND affects the body is important. According to Frank (1995), bodies are not only illness' experiential terrain, but also the means through which this experience is shaped and communicated. The following paragraphs present literature on the disease process and on the experience of MND. The last paragraph of the chapter summarises the literature.

### **3.3 Motor neurone disease**

MND was first described by the French neurologist Charcot in 1874 and is an adult onset, progressive, neurodegenerative condition leading gradually to partial or complete paralysis, including loss of speech (Eisen, 2009). The majority of MND cases are classified as sporadic with no related family

history of the disease, while gene mutations have been discovered in 5-10% of the affected population (familial type). The four main types of MND are shown in Table 2.1, with amyotrophic lateral sclerosis (ALS) being the most common form presented in about 70% of people with MND. Progressive bulbar palsy (PBP) accounts for approximately 25% of all diagnosed cases and often progresses to the ALS type within the course of a few years. Primary lateral sclerosis (PLS) is a rare subtype, with only 1% of people diagnosed with MND presenting with it, while progressive muscular atrophy (PMA) is diagnosed in 5% of the cases (Kiernan et al., 2011). For the purpose of this study these types will be referred to collectively as MND, with specification offered when necessary.

**Table 3.1 Classification of MND types**

<b>Type</b>	<b>Symptoms</b>
Amyotrophic lateral sclerosis	Upper and lower motor neurone symptoms. Affects bulbar and limb/trunk musculature.
Progressive bulbar palsy <sup>3</sup>	Upper and lower motor neurone symptoms. Affects bulbar musculature.
Progressive muscular atrophy	Lower motor neurone symptoms.

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<sup>3</sup> Bulbar palsy refers to impairment as a result of lesion to cranial nerves IX, X and XII. Muscles involved in swallowing, speech production, and breathing may be affected (Talbot et al., 2010).

	Affects limb/trunk musculature.  Rarely, affects brain stem innervated muscles.
Primary lateral sclerosis	Upper motor neurone symptoms.  Affects limb/trunk and/or bulbar musculature.

Source: Adapted from Francis, Bach, & DeLisa (1999, p.952).

### 3.3.1 *Diagnosis and epidemiology*

The current lack of biomarkers for MND means that the diagnosis is still mainly clinical and cannot be confirmed through laboratory tests (Turner, Kiernan, & Talbot, 2009). The commonly accepted El Escorial diagnostic criteria are shown in Table 2.2. Research directed towards establishing protein biomarkers could provide an early and relatively straightforward diagnostic method (Henrik & Bowser, 2008).

**Table 3.2 Revised El Escorial criteria for the diagnosis of MND**

Clinically Definite ALS	Defined on clinical evidence alone by the presence of upper motor neurone, as well as lower motor neurone signs, in three [body] regions.
Clinically Probable ALS	Defined on clinical evidence alone

	<p>by upper motor neurone and lower motor neurone signs in at least two regions with some upper motor neurone signs necessarily rostral to the lower motor neurone signs.</p>
<p>Clinically Probable - Laboratory-supported ALS</p>	<p>Defined when clinical signs of upper motor neurone and lower motor neurone dysfunction are in only one region, or when upper motor neurone signs alone are present in one region, and lower motor neurone signs evidenced by electromyography (EMG) are present in at least two limbs, with proper application of neuroimaging and clinical laboratory protocols to exclude other causes.</p>
<p>Clinically Possible ALS</p>	<p>Defined when clinical signs of upper motor neurone and lower motor neurone dysfunction are found together in only one region or upper motor neurone signs are found alone in two or more regions; or when lower motor neurone signs are found rostral to upper motor neurone signs</p>

	<p>and the diagnosis of Clinically Probable - Laboratory-supported ALS cannot be proven by evidence on clinical grounds in conjunction with electrodiagnostic, neurophysiologic, neuroimaging or clinical laboratory studies. Other diagnoses must have been excluded to accept a diagnosis of clinically possible ALS.</p>
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Source: Adapted from the World Federation of Neurology archives (2010).

Average incidence of MND is estimated to be between 1.5 and 3 per 100,000 people per year and appears to be relatively similar across the population and across countries (Alonso, Logroscino, Jick, & Hernan, 2009; Logroscino et al., 2010; Pradas et al., 2013), although some differences have been observed (Imam et al., 2010). The highest incidence occurs over 55 years of age and average life expectancy after diagnosis is two to five years, although one fifth of people survive beyond five years and some much longer (Eisen, 2009). This has led to some confusion whether MND ought to be described as a terminal or as a chronic disease (Small & Rhodes, 2000). Qureshi et al. (2009) reviewed survival and rate of functional decline as documented in efficacy trials performed between 1990 and 2008 and

concluded that the overall survival of MND is improving, attributing this to improvements in symptomatic care.

Lifetime risk is 1 in 472 for women and 1 in 350 for men making it not such a rare disease. Due to the increase in the life expectancy of the general population prevalence of MND is bound to increase in the coming years (Talbot & Marsden, 2008).

### *3.3.2 Symptoms and disease management*

The course of the disease is highly individualised and comparisons cannot be easily made between individuals with regards to disease progression, affected areas and survival estimates. Disease progression appears to be positively correlated with bulbar onset and with older age, but there is conflicting evidence on other possible factors, such as diagnostic delay (Chio et al., 2009). It has been argued that respiratory function tests can be used to predict survival of people living with MND (Baumann et al., 2010). Similarly, the different types of MND could potentially be assigned distinct disease progression rates (Talman, Forbes, & Mathers, 2009).

MND affects both upper and lower motor neurones, resulting in extended symptomatology (Table 2.3). As muscle groups become weaker and waste away, activities such as writing, walking, lifting, swallowing, coughing, talking, and breathing become difficult and eventually cannot be carried out. Mental capacities are usually, although not always, left intact and people are aware of the unfolding condition.

**Table 3.3. Clinical signs of motor neurone degeneration**

<b>Upper motor neurone manifestations</b>	<b>Lower motor neurone manifestations</b>	<b>Unusual manifestations</b>
Moderate weakness	Severe weakness	Dementia
Hyperreflexia	Hyporeflexia	Autonomic involvement
Pathologic reflexes	Muscle atrophy	Pain
Pseudobulbar effect	Fasciculations	Bladder urgency/ incontinence
Spasticity	Muscle cramps	Sensory symptoms
Loss of dexterity	Muscle hypotonicity	
Slowed movements		

Source: Adapted from Jackson & Rosenfeld (2001, p.336).

Eventually, the muscles controlling breathing become affected and death often occurs due to respiratory failure. Symptoms that can develop in the course of the disease include dysarthria (Appendix A), dysphagia (Appendix A), spasticity, sleep disturbances, pain, emotional lability, fatigue, constipation, cognitive impairment, depression, and olfactory disorders (Corcia & Meininger, 2008; Hawkes et al., 1998; Lou, 2008; Portet,

Cadilhac, Touchon, & Camu, 2001; Tomik & Guilloff, 2010). These symptoms can be managed to a degree through drugs or specialist therapies, such as kinesiotherapy and pulmonary physiotherapy (Corcia & Meininger, 2008). Management of saliva can be challenging and often unsuccessful. As the musculature around the mouth and in the oral cavity becomes weaker managing saliva becomes difficult, resulting in drooling or pooling of saliva in the mouth and leading to frequent choking episodes as people cannot swallow the accumulated secretions (Hadjikoutis, Eccles, & Wiles, 2000).

At some stage in the disease progression people often require medical procedures and equipment to manage issues, such as limited calorific intake due to dysphagia, compromised breathing ability, and reduced mobility (Langmore, Kasarskis, Manca, & Olney, 2009; Miller et al., 2009). Gastrostomy (percutaneous endoscopic gastrostomy, PEG, or radiologically-inserted gastrostomy, RIG, Appendix A) and non-invasive positive pressure ventilation (NIPPV) (Appendix A) are often suggested by healthcare professionals although not all patients want to use them or can benefit from their use (Allen et al., 2013; Jackson et al., 2006; Langmore et al., 2009). NIPPV requires the attachment of a mask to the mouth and this can be challenging when saliva management is an issue, as often people need to let saliva drool out of the mouth to avoid pooling and choking. The procedures mentioned above, combined with the prescription of the drug Riluzole and the use of antioxidant agents such as vitamin E or the co-enzyme Q10 (Orrell, Lane, & Ross, 2009; Rosenfeld & Ellis, 2008) are the most commonly used strategies in the management of MND.

In order to manage with disease, various clinical guidelines have been developed and systematic reviews of the efficacy of management techniques have been undertaken (Lechtzin, 2009; Leigh & Wijesekera, 2010; Miller et al., 2009). The literature suggests that for these to be effective, they need to be adjusted to individual circumstances (King, 2005; Small & Rhodes, 2000). Literature also suggests that healthcare for people with MND is best provided in specialised multidisciplinary clinics (Aridegbe et al., 2013) but even in countries that these do exist, they are not easily accessible by all people (Mayadev et al., 2008; Ng, Khan, & Mathers, 2009).

### *3.3.3 Terminal stage and causes of death*

In advanced stages of MND the use of NIPPV is not adequate for respiration and people can experience respiratory insufficiency. A small percentage of people with MND, estimated at about 3% in the USA, choose to use long-term invasive mechanical ventilation through tracheostomy, a procedure that can prolong life while the condition progresses (Rabkin et al., 2006). Most people, however, do not wish to be intubated because of the possibility for them to reach a 'locked in' state, where they will not have control of any of their muscle groups (Rabkin et al., 2006).

Medical interventions in the last stages of the disease focus on making people comfortable, through the use of appropriate medication (Borasio & Voltz, 1997). Research suggests that the most common cause of death in MND are respiratory complications attributed to pneumonia

(bronchopneumonia or aspiration pneumonia), or to terminal stage respiratory failure, which can lead to hypercapnic coma (Kurian, Forbes, Colvile, & Swingler, 2009; Talbot et al., 2010).

### *3.3.4 Main research directions in MND*

#### *3.3.4.1 Pathogenesis and emerging pathways to treatment*

There is currently no cure for MND, with the only approved drug for the treatment of the disease, Riluzole, showing only moderate effect, prolonging median survival by two to three months (Miller, Mitchell, & Moore, 2012; Mitchell, O'Brien, & Joshi, 2006; Zoccolella, Begi, Plagano, & Fraddosio, 2007). Researchers believe that unravelling the pathogenetic mechanisms of the disease may offer pathways to a cure (Bedlack, Traynor, & Cudkowicz, 2007).

At this stage, the mechanisms of pathogenesis are not known for either the familial or the sporadic form of the disease. Factors that seem to play a role in the pathogenesis of MND include genetic factors, oxidative stress, glutamatergic toxicity, and damage to certain proteins (Rothstein, 2009; Shaw, 1999). Approximately 5 to 10% of MND cases have a family history and are classified as familial. Altogether genetic mutations that play a role in the development of these cases have been identified in 30% of these cases, with mutations in genes responsible for the expression of the proteins SOD1 and fused in sarcoma (FUS) being the most common ones. However, the mechanisms of pathogenesis are not yet clear (Polymenidou & Cleveland, 2008). Mutations in FUS protein (Deng et al., 2010) and

accumulation of the protein TDP-43 (Rothstein, 2009) have recently been linked to destruction of motor neurones in cases of both familial and sporadic origin, offering a connecting link between the two types of the disease (van Damme & Robberecht, 2009).

Several researchers argue that the aetiology of MND is complex, with a combination of genetic, environmental, and possibly lifestyle factors playing a role (Bedlack, Traynor, & Cudkowicz, 2007; Burvill, 2009; Cookson & Shaw, 1999; Gallo et al., 2009; Goodall & Morrison, 2006; Morahan, Yu, Trent, & Pamphlett, 2007; Okamoto, Kihira, Kondo, & Kobashi, 2009; Veldink, van den Berg, & Wokke, 2004). It has been suggested that occupation might be a risk factor and although conclusive evidence does not exist, the following groups might be at a higher risk of developing MND: veterinarians and other health workers, athletes, hairdressers, power-production plant workers, and electrical and military workers (Sutedja et al., 2009).

Basic research on the pathways of pathogenesis has led to developments in research on disease-modifying therapies, although no substance has been approved yet (Bedlack, Traynor, & Cudkowicz, 2007; Goodall & Morrison, 2006; Orrell, 2010; Orrell, Lane, & Ross, 2009). Some of the substances which are being examined as potential therapeutic means include the insulin-like growth factor I (Orrell, 2010), lithium carbonate (Leigh & Wijesekera, 2010), and antioxidants (Orrell, Lane, & Ross, 2009).

#### 3.3.4.2 *Quality of life in MND*

Quality of life indices have been used extensively in MND research, exploring overall perspectives of well-being, and the presence of depressive symptoms amongst people with MND and family carers. While there is no uniformity in how the concept of ‘quality of life’ is defined in the literature, the World Health Organisation conceptualises it as

A broad ranging concept affected in a complex way by the person’s physical health, psychological state, level of independence, social relationships, personal beliefs and their relationship to salient features of their environment (The WHOQOL Group, 1998, p.1570).

There is an extensive body of literature that suggests a weak, if any, correlation between physical impairment, or length of disease, and quality of life or depressive symptoms (Bromberg, 2008; Cardol et al., 1996; Ganzini, Johnston, & Hoffman, 1999; Goldstein, Atkins, & Leigh, 2002; Kubler et al., 2005; Nelson et al., 2003; Nygren & Askmark, 2006). This is so even in the case of severely disabled people who may be using ventilatory support or tube feeding (Kubler et al., 2005; Neudert, Wasner, & Borasio, 2004).

Quality of sleep (Cardol et al., 1996) and a sense of purpose (Bromberg, 2008; Cardol et al., 1996; Plahuta et al., 2002) appear to be stronger indicators of quality of life in MND rather than the severity of physical symptoms. Possible reasons for this lack of correlation of physical function and quality of life may be the lack of pain in MND, and the availability of palliative care to ameliorate some of the disease effects, such as the use of NIPVV and tube feeding.

Another important parameter is the fact that ideas about quality of life are not stable. When developing a disabling condition, people often reprioritise, changing their perspective of life and sometimes finding satisfaction through ways not previously contemplated (Bromberg, 2008). People around them, even close relatives might not be aware of this process. Olsson, Markhede, Strang and Persson's (2010) study, for example, highlights how the next of kin can sometimes rate the well-being of people diagnosed with MND lower than the people with MND themselves.

### *3.3.5 Summary of section*

This section presented an overview of the effects of MND on the physical body. It also offered an overview of the directions of current research. There is a substantial body of research on MND focusing on mechanisms of pathogenesis, pathways to treatment, and quality of life. Several hypotheses as to the aetiology of MND are being currently tested and these may lead to a cure. Until this happens, MND remains an incurable, progressive neurodegenerative disease that can lead to complete muscle paralysis. Furthermore, research shows only a weak, if any, correlation between quality of life for people with MND and physical impairment, suggesting that there are other factors, beyond physical ability and function, that are important to people with MND and that make life worth living.

## **3.4 Living with MND**

The aim of this section is to present literature on the lived experience of MND that complements knowledge on the biomedical parameters of the

disease. This section focuses on three areas that are prominent in the literature; experiences of living with MND, experiences of services and lay and professional perspectives on MND.

#### *3.4.1 Literature review methods*

The review was performed in a systematic way and followed the process for review of qualitative studies developed by the Centre of Reviews and Dissemination (CRD, 2009). The aim of this review was to provide an answer to the question of what is known about people's experiences of living with MND.

Articles were identified through electronic searches that were carried out between May 2011 and May, 2012. MEDLINE, CINAHL Plus and PsychInfo databases were used for the literature search, with the keywords 'ALS' or 'amyotrophic lateral sclerosis' or 'motor neuron\* disease' or 'MND' and 'experience\*' or 'qualitative'. All search fields (for example, title, keywords, abstract, etc.) were selected in all three databases. The basic search function was used in Medline and PsychInfo, with the 'related terms' search function activated. The advanced search function was used in CINAHL Plus, utilising the Boolean/Phrase search mode. The search and selection process is presented in Appendix B. The articles that were identified through the literature search are presented in Appendix C.

The bibliographical details of the articles that were identified through the searches were inputted into a Microsoft Word document. The details included the title, the keywords, and the abstract, where available.

Appropriateness for inclusion was judged by these elements. Common reasons for exclusion were: a focus on effectiveness of interventions; a focus on MND symptoms; a focus exclusively on carers' perspectives; and not being research based. Following appraisal, data were extracted in accordance with Thomas and Harden's (2008) methods for developing a thematic synthesis. Following Thomas and Harden (2008), these data were the analyses and interpretations of the researchers rather than participants' quotations, which were considered to be raw data. Segments of text under the headings of 'results' and 'findings' were treated as data and pasted onto a Microsoft Word document. The discussion section of all articles was also reviewed, as sometimes it included results. Finally, data were complemented by other sources that were identified through hand searches of references lists and literature searches on specific authors.

#### *3.4.2 Experiences of living with MND*

A diagnosis of MND represents a major change in people's lives. It has been described as an existential shock (Brown, 2003). Questions about how one should live his or her life, what choices to take and how to set priorities are part of the process of adapting to living with MND (Bolmsjö, 2001). Different people manage the disease in different ways. In effect, people develop their own explanations about illness and how it makes sense in the context of their lives, as has been evidenced by ethnographic studies on chronic illness (see for example, Good, 1994; Hunt & Arar, 2001).

In their exploration of the lived experience of MND Robinson and Hunter (1998) highlighted how the participants in their study were coping with a constantly changing situation; as the condition progressed different adaptations were necessary to enable them to go on with life as they wished. Robinson and Hunter (1998) drew their data from the stories of over three hundred people with MND that were written for and stored at the John Bevan MND Research Unit at Brunel University (the research unit has ceased to exist). These findings echo stories of living with MND written from a lay perspective (for example, Kaye, 1994; Sackett & Sakel, 2011).

As a result of MND, the image that people had of themselves prior to being diagnosed with MND, the idea of who they are and who they want to be can change. People living with MND modify their notion of a ‘desired self’, i.e. how they want to live their life and who they want to be (King, Duke, & O’Connor, 2009). They engage in a continuous process of adapting to ongoing change. As King, Duke and O’Connor’s (2009) study highlighted, people living with MND take decisions that will enable them to keep on living and maintain a sense of self and well-being in the face of constant change and loss of physical abilities. To do this, participants in their study engaged in what the authors described as a “*distinct cyclic decision-making pattern addressing ‘ongoing change and adaptation’*” (King, Duke, & O’Connor, 2009, p.752).

This constant change and adaptation to the progressive nature of MND may explain why people with MND appear to talk about and make sense of their life in various and diverse ways. Studies carried out by Brown

and Addington-Hall (2008) and by Locock, Ziebland and Dunelow (2009) illustrate this diversity. For some of the participants in these studies, life appeared to be over. They were in effect experiencing what Locock, Ziebland and Dunelow (2009) described as ‘biographical abruption’. Biographical abruption resonates with the chaos storyline of illness as described by Frank (1995). In chaos narratives people cannot make any sense of the unfolding situation and they feel powerless and not in control of their lives, a condition which sometimes leads to hopelessness (Plahuta et al., 2002). Some other participants felt an overwhelming sense of difficulties to come, disrupting their plans and priorities and leading to a life that is fractured (Brown & Addington-Hall, 2008). Living life to the full and enjoying what is available, or modifying one’s priorities, were also part of some people’s life, a process that Locock, Ziebland and Dunelow (2009) referred to as ‘biographical repair’.

One important element of people’s experience of living with MND is that they develop knowledge that is specific to their situation (Robinson & Hunter, 1998). In a way, they need to learn how to live with MND as this represents a major change compared to their life prior to MND. This process of learning how to live with MND can be described as a process of developing experiential knowledge. Experiential knowledge is experience-based, and gives people insights on how to incorporate MND in the course of their daily life, guiding their choices on which strategies work well and which do not (Stewart, Abidi, & Finley, 2010). As Pols (2011) stated, experiential knowledge refers to “*having knowledge about how to live with*

*a chronic disease on a daily basis*” (p.200). People with MND need to navigate an uncertain future; their symptoms keep progressing, new ones emerge and there is no definitive information about prognosis. Faced with too much, too little, too technical or ill-timed information, people with MND often concentrate on the here and now and how they can live with MND on a day-to-day basis (Cobb & Hamera, 1986; Lemoignan & Ells, 2010; McNaughton, Light, & Groszyk, 2001; O’Brien, Whitehead, Jack, & Mitchell, 2011; Sundling, Ekman, Weinberg, & Klefbeck, 2009; Vesey, Leslie, & Exley, 2008). They prefer to deal with issues as they arise since information given too far in advance might reveal a frightening future.

People with MND are faced with a body they cannot control, a body they cannot rely on or trust (Brown & Addinton-Hall, 2008; Locock, Ziebland, & Dumelow, 2009). Their abilities change all the time and things that were possible one day may not be possible the next. With changing abilities comes a moving threshold from acceptable to not acceptable levels of dependence; from loss of independence with toileting, to use of PEG, use of NIPPV or loss of speech. People with MND are afraid of losing those functions associated with dignity and communication as these are deeply connected to their identity (Lemoignan & Ells, 2010; Vesey, Leslie, & Exley, 2008). In Brott, Hocking and Paddy’s (2007) study, the main disruptions participants were experiencing were related to their activities and the social roles these were associated with; as their body became increasingly paralysed people found it hard to engage in meaningful activities and maintain valued social roles.

While the physical body cannot be trusted, or exactly because of that, people with MND try to take control over their life and they do that in many ways. From adapting activities to reflect current levels of function to changing their diet and from becoming experts on MND to making the best of what they have, people with MND try to effect control over their lives. This may be done in a pragmatic way as people develop an awareness of what they can and what they cannot do and focus on what is possible (Brown & Addinton-Hall, 2008; Brott, Hocking, & Paddy, 2007; Foley, O'Mahoney, & Hardiman, 2007). Limited mobility, for example, can affect expression of intimacy, but people can modify the associated activities (Taylor, 2011).

In seeking to take control over their life, many people use various pieces of equipment. These can include, but are not limited to, NIPPV, PEG, and alternative and augmentative communication devices (AACD). Equipment comes with certain benefits and certain drawbacks and people with MND often accept the equipment when the perceived benefits outweigh the inconvenience from the incurred changes on daily routines (King, Duke, & O'Connor, 2009; Sundling et al., 2009). Sometimes equipment can be perceived to be too technical and complex (McNaughton, Light, & Groszyk, 2001; Murphy, 2004). The use of NIPPV, for example, is not always seen favourably because it can upset other people, or because of too overt associations with disease and dependence. Also, the mask (depending on length of use and type of mask) can cause sores on the nose

and around the mouth. On the other hand, NIPPV offers the benefit of restoring sleep and days with more energy (Sundling et al., 2009).

Early reliance on equipment can be perceived as contributing to functional deterioration, or as a definitive sign that the threshold from independence to dependence has been traversed (Murphy, 2004). The decision to use equipment appears to be guided by both the perceived benefits of such equipment and the extent to which use of the equipment contributes to the person's own view of their life. It would seem that people with MND make decisions that will enable them to retain their sense of identity while at the same time addressing some of the effects of the condition on their daily life.

#### *3.4.3 Experiences of services*

Studies specifically on MND point to a dissatisfaction with available services, to limited knowledge about the disease, and to a divergence in perspectives between healthcare professionals and people living with the disease. People with MND report that despite professionals' best efforts sometimes services do not meet their needs effectively. This is mainly attributed to inadequate knowledge of MND and the organisation of services (Brown, Lattimer, & Tudball, 2005; Hughes, Sinha et al., 2005). The early and the late stages of the disease process in particular are especially critical stages as they represent important changes in the life of the patients and the people around them. These stages are reflected in healthcare by diagnosis and palliative care. Satisfaction levels with provided services during these

stages are not always high (O'Brien, Whitehead, Jack, & Mitchell, 2011; Pavey, Allen-Collinson, & Pavey, 2013; Whitehead, O'Brien, Jack, & Mitchell, 2012). Participants in a study by O'Brien et al. (2011) reported diagnostic delays and misreferrals, while participants in several studies (O'Brien et al., 2011; Robinson & Hunter, 1998; Small & Rhodes, 2000) expressed dissatisfaction with not only the level (too much or too little) but also the content of the information provided during the communication of diagnosis. Some people reported receiving information that was unrealistically optimistic or unjustifiably negative. Participants in the same studies also reported uncertainty regarding end-of-life care, stating that staff in residential care services was not knowledgeable about the condition. A study by Whitehead et al. (2012) rendered similar findings, with participants expressing a strong wish to die at home.

In O'Brien's (2004) study on healthcare professionals' knowledge of MND, 57% of professionals felt that their current level of knowledge was inadequate. This sometimes leads to health professionals giving contradictory advice, even on issues such as progression of the condition, average life expectancy, and efficacy of services (Talbot & Marsden, 2008). It should be noted that sampling was performed via an MND clinic, and thus respondents were likely to have come in contact and worked with people living with MND. Brown, Lattimer and Tudball's (2005) investigation of service providers and services users' views of MND services rendered similar results.

Professionals' knowledge about MND and the organisation and coordination of services have been reported as problematic areas in studies on service use and needs of people with MND in Scotland (van Teijlingen, Friend, & Kamal, 2001) and in England (Hughes, Sinha et al., 2005). Informants in an Australian study on satisfaction of services among people with neurodegenerative conditions also highlighted structure and provision of services as problematic areas (McCabe, Roberts, & Firth, 2008). On the other hand, specialist multidisciplinary clinics are generally seen in a positive way, perhaps due to the specialised knowledge professionals have (O'Brien et al., 2011). However, attending these clinics can be tiring and sometimes intimidating due to the involvement of many professionals at the same time (O'Brien et al., 2011).

Knowledge about MND and service structures are of course interconnected; services can be appropriately and effectively organised to respond to the needs of people living with MND only when professionals have detailed knowledge on MND and understand the associated needs. Through their experiences, people with MND develop their own understanding of what MND is and what services they require. In her exploration of experiences of care in MND, Brown (2003) concluded that people with MND and service professionals operate from different standpoints, based on the value structure they adopt as being valid. The scientific standpoint adopted by professionals often leads them to focus on functional assessments, adaptations, equipment or other changes that will render a functional outcome. People living with MND, however, often focus

on their lived experience of MND or, in other words, on what it means for them to live with MND in their own local contexts.

Cox's (1992) findings are similar to Brown's (2003), to the extent that both studies agree that service users and service professionals have different perspectives on MND and what ought to be done about it. In Cox's study, people living with MND focused more on practical solutions and physical needs, while healthcare professionals prioritised the coordination of the different aspects of MND care. However, people with MND often report coordination of services as a problematic area and they find services to be fragmented and not always responsive to their needs (Brown, 2003; Hughes, Sinha et al., 2005).

#### *3.4.4 Lay and professional perspectives on MND*

The reported dissatisfaction with services can partly be explained by the evidenced divergence of perspectives between healthcare professionals and people living with ongoing illness. Numerous studies illustrate the fact that people living with ongoing illness or disability and healthcare professionals often have different perspectives about the disease and focus on different aspects when considering care and management options (Emanuel, Fairclough, Daniels, & Claridge, 1996; Frank, 1995; Good, 1994; Montgomery & Fahey, 2001; Slevin et al., 1990; van der Waal, Capsarie, & Lako, 1996). A Cochrane review on multidisciplinary care (Ng, Khan, & Mathers, 2009) for people with MND also demonstrated a divergence of perspectives between healthcare professionals and people living with MND.

Hunt and Mattingly (1998) referred to this multitude of perspectives as ‘diverse rationalities’ and ‘multiple realities’. These terms mean that people living with a disease and healthcare professionals may use different systems of knowledge to help them make sense of what a particular disease is about, and so they may reach different explanations for it. Robinson and Hunter (1998) discussed how participants in their study had developed their own ways of making sense of MND and its impact on them, and their perspective did not always coincide with that of healthcare professionals. These findings agree with those from an earlier study by Cox (1992). Cox’s study also suggested a divergence of perspective between people with MND and their main carer.

Caron-Flinterman, Boerse and Bunders’ (2005) study on the potential contribution of service users to biomedical research highlighted the importance of experiential knowledge. People living with an illness often develop a wealth of information as a result of their lived experience. They construct strategies to cope with it in their daily life, and ways to make sense of it. This knowledge is not always valued in health research, where the scientific knowledge of professionals takes precedence over the knowledge of people living with an illness (Frank, 1995). This can be problematic because research suggests that health care providers sometimes struggle to choose what is the best, or the desired outcome of an intervention (Rodriguez & Young, 2006). The reason for this is a dichotomy between quality of life-based and physiology-based goals, or in other words between the biomedical and the lived perspective and experience of illness.

Each perspective and each experience are equally true and valid within the lifeworld where they have been developed (Frank, 1995; Mol, 1998).

#### *3.4.5 Summary of section*

This section highlighted the complexity of the experience of living with MND. Just like the disease progresses and changes all the time, the experience of living with MND is dynamic and people adapt to it continuously. Furthermore, this section showed how people with MND develop knowledge through living with the disease and develop their own ways of living with it. These ways can range from learning how to avoid or treat sores from the NIPPV mask, to experimenting with ways to express intimacy. Exploring the experiential knowledge that enables people to keep on with their daily life can offer insights about the kinds of care that are most useful.

#### *3.4.6 Critique of the literature*

The studies reviewed contain rich information on living with MND, bringing the perspectives of people with MND to the foreground. The results of the studies reviewed offered similar information, to some extent. This is probably a fabricated data saturation that can be attributed to the research design that most of the studies followed, i.e. interview-based studies where participants were interviewed once. Designs based on single interviews cannot always capture the processes of ongoing change that are present in living with MND as they offer a snapshot of experience.

Sampling was another area that weaknesses were observed in several of the studies. While size sample in qualitative research is guided by saturation rather than by numbers, several studies did not specify how saturation was reached. Furthermore, issues of bias were often left unaddressed, as was often the researcher's position.

### **3.5 Chapter summary**

There is an important and ever growing body of research on the pathogenetic mechanisms of MND and on pathways to finding a cure. There is also a wealth of information on the management of some of the symptoms of MND. Furthermore, people living with MND often develop a wealth of information about the disease as a result of their lived experience. They construct strategies to cope with MND in their daily life and ways to make sense of it. The way people make sense of their life with MND influences their needs, their choice of services, and the way they negotiate illness and life with it (Brown & Addington-Hall, 2008; King, 2005). This points to a need to listen to personal experiences of living with MND.

Knowledge on how people with MND experience life with the disease is currently limited (Brown, 2003; Brown, Lattimer, & Tudball, 2005; Hughes, Sinha et al., 2005; O'Brien, 2004). This has an impact on service provision (Brown, Lattimer, & Tudball, 2005; Foley, Timonen, & Hardiman, 2012). Literature suggests that the needs of people living with MND are not always effectively met (Brown, Lattimer, & Tudball, 2005; Hughes, Sinha et al., 2005; Tanaka et al., 2010; van Teijlingen, Friend, &

Kamal, 2001). How life is experienced, how people make sense of MND, what their priorities are, and how they work towards them is not clear.

## **4. Use of narratives in research**

### **4.1 Introduction**

The literature review revealed a wealth of biomedical knowledge on MND. This knowledge concerns facts and possibilities that guide choices and illustrates how MND *can* progress. Knowledge about how people with MND experience life and how they make sense of it, in their own contexts, is not explored in much depth in the literature. It was this experiential knowledge, i.e. focusing on how people understand and view their life with MND, that I wanted to explore in this study. I selected a qualitative research design because I sought to focus on the individual and the subjective nature of the experience of living with MND.

There are several qualitative research designs, each of them suited to particular research questions. A narrative inquiry design, focusing on the personal, unique narratives of the participants was considered as the most appropriate design for this study, allowing an in depth exploration of how individual people make sense of their life. This chapter illustrates the use of narratives in research on illness experiences before discussing how narratives are used in this study.

### **4.2 Definition of narrative**

Narratives can refer to the stories people say or enact about aspects of their life, and to the interpretation and representation of these stories by a researcher (Polkinghorne, 1988). The Merriam Webster (2013) dictionary

gives three definitions for narrative: a) something that is narrated, a story, or an account, b) the art or practice of narration, and c) the representation in art of an event or story.

While narratives can be conceptualised as speech events (Labov & Waletzky, 1967/1997), the point of departure for the use of narratives in this thesis is the concept of meaning. People make sense of their life in different ways, finding ways to connect the past with the present and projecting their self into the future (Ricoeur, 1980). Doing so requires the construction of stories that give meaning to a person's life so that life is experienced as a connected whole rather than as a multitude of fragments in time and space (Ricoeur, 1980). These stories can be relayed through different means; they can be enacted, written, narrated in their entirety or in snippets or in any other way that is available and resonates with a person's life. According to Chase (2005), a narrative can be written or oral and can be short and topical, take the form of an extended analysis of an aspect of one's life or be presented as a story of one's whole life. For Hydén and Antelius (2010)

Stories are actually embodied in the gestures, the linguistic, para-linguistic, non-verbal and other physical artifacts that are used as resources in telling and listening to a story (p.590).

Similarly, Mattingly (1998a; 2010) and Alsaker (Alsaker, Bongaard, & Josephsson, 2009) describe narratives as enacted performances that people engage in order to create meaning and make sense out of life events.

### 4.3 Use of narratives in studies on illness experiences

Narratives are being increasingly used in research on illness experiences. The ways they are defined and used are as diverse as the experiences they describe. Some researchers use narratives to explore the experiences of people who live with illness or disability (for example, Charmaz, 1991; Cole, 2004; Kirmayer, 2000; Kleinman, 1988; Robinson & Hunter, 1998). Some other researchers use narratives to share their own personal stories of living with illness, through an autobiographical genre (for example, Frank, 2002; Murphy, 1990; Wikan, 2000) and others use narratives to explore encounters between healthcare professionals and people living with illness (for example, Good, 1994; Hurwitz, 2000; Mattingly, 1998a; 2010; Park, 2008). Some studies on experiences of people living with MND (for example, Brown & Addington-Hall, 2008; Locock, Ziebland, & Dumelow, 2009) have also used narrative methodology in order to bring to the fore participants' stories.

Gareth Williams (1984) was one of the first researchers who explicitly referred to narratives in the context of health and illness. Williams (1984) conducted semi-structured interviews with thirty people with rheumatoid arthritis looking for explanatory models of illness that would answer the question "*why do you think you got arthritis?*". Data pointed to a process whereby participants discussed their changing relationship to the world and constructed pasts that could lead to their lived presents, so as to make it possible for the disease to emerge. In the opening paragraph of Williams' (1984) article on the genesis of chronic illness, Bill, one of the

study participants, breaks out asking: “*how the hell have I come to be like this*” (p.175). Bill’s question points to the need to construct intelligible stories, leading from the past to the present and perhaps into the future. Williams’ (1984) statement that “[*disease*] *assaults the taken for granted world and demands explanation*” (p.197) reminds us of the fundamental human need for meaning.

The process whereby one’s life story is disrupted and the lived past does not lead to a previously envisaged future, has been defined by Bury (1982) as ‘biographical disruption’. The thematic thread that was leading from the past to an unfolding present and through to a projected future is ruptured and new meanings and links need to be established so that people can once again make sense of their life (Charmaz, 1991). Narratives about living with an illness can provide a means for people to engage in meaning-making (Frank, 1995; Kleinman, 1988). Stories do this by providing the basis for people to share emotions, thoughts, and construct new possibilities by projecting themselves into the future (Frank, 1995). A narrative presupposes a past, a present and a future, all meaningfully connected (Ricoeur, 1980). For Frank (2002), “*stories give lives legibility; when shaped as narratives, lives come from somewhere and are going somewhere*” (p.5). Narratives of lives lived with an illness help us understand one’s life in context (Frank, 1995; 1998).

Drawing from personal experience and from empirical material, Frank (1995) presented three storylines of illness narratives. Rather than being thought of as fixed categories, these storylines can be more accurately

conceptualised as points between which stories of illness constantly gravitate. These storylines are ‘restitution’, ‘chaos’, and ‘quest’. Restitution stems from the human needs for safety and control that lead to a desire to know that ‘all will be fine’. Life-threatening disease or disability ruptures life in a fundamental way, creating a schism between an experienced past and an uncertain future. The first reaction of people may be to believe that things will go back to normal and their sense of identity will not be threatened. The emergence of what Mol (2008) referred to as the ‘logic of choice’, has led to the construction of the patient-consumer who views healthcare as a product. In that context, disease is merely a disruption of the normal rhythm of life and the main problem lies in finding the right way or product to deal with it, so that health can be reinstated. Often this is possible; when it is not, as is the case with MND, other stories need to be told.

Chaos narratives refer to these stories where not only is restitution not conceivable, but also the sense of identity has been affected in such a deep way that meaning making is not possible. The thread that was connecting a person’s story from the past, to the present and on to a projected future has been sheared. For Frank (1995), a chaos storyline is a non-narrative as a person living it cannot engage in a process of narration and meaning making. It is only when the person is out of this chaos narrative that she or he can reflect back and construct the story.

The final storyline described by Frank (1995) is the quest narrative. In quest narratives the narrator engages in a dialogical relationship with

illness, which is seen neither as an external element nor as an all-destructive force. Instead, in quest narratives illness is viewed as part of the person's sense of identity and emphasis is placed on learning to live with it. Frank (1995) draws parallels between quest narratives and the heroic journey as described by Joseph Campbell (1968) in his study on the construction of the 'hero'. According to Campbell (1968), essential to the construction of the hero is a journey, which unfolds in three main stages: departure, initiation, and return. All of these can have their equivalent in stories of illness, with departure being the stage where the person first notices symptoms or consults a specialist; initiation being the stage where it becomes clear that return to previous normality is not likely; and return being the stage where a person is transformed through living with an illness. There is a crucial difference, however, between the 'hero's journey' as described by Campbell (1968) and stories of illness; people do not choose to embark on a life with illness. Illness is presented to them and leaves them no choice.

Illness narratives are stories about life (Frank, 1995; Kleinman, 1988). Sometimes these stories can be dominated by illness, while some other times illness is just in the background, as a possibility or as a lived normality. Narratives of living with an illness are often about restructuring experience in such a way that the narrator can make sense of it, or gain control over it (Frank, 1995; Jackson, 1998; Mattingly, 2010). By constructing a story, people construct themselves as actors; they can take decisions and exercise some control over their life. But they also construct themselves as what Jackson (1998) called 'sufferers'; sufferers not

necessarily in the sense of being subjected to something painful or unpleasant, but in a metaphorical one of not having control of certain elements of their lives. In life, and in the stories people construct about their life, people oscillate between being an actor and a sufferer, between having control and having to accept loss of control.

#### **4.4 Background to research design**

This study followed a narrative inquiry design. The focus was on creating narratives about people and how they live with MND. These narratives were based on the stories shared by the participants, but they were ultimately interpreted and written by myself, the researcher. Riessman (2001), and Clandinin and Connelly (2000), amongst others, have noted that there is an increasing attention to narratives in the context of research. Arguments in favour of the use of narratives have emanated from many and diverse fields, such as education (Clandinin & Connelly, 2000), medical anthropology (Mattingly, 1998a; 2000; 2010), social sciences (Frank, 1995; Franzosi, 1998; Murphy, 1990) and medicine (Hurwitz, 2000; Kleinman, 1988). The thread that connects studies that have a narrative focus is an attention to experience and how people make sense of it. This section discusses how narratives are conceptualised in this study. After presenting the features of narratives as used in this study and discussing the authorship of the narratives, this section concludes with a justification of the choice of a narrative-based design.

#### 4.4.1 *Features of narratives*

Narratives, according to Mattingly (1998a), have three distinctive features. They are event-centred, experience-centred, and create new experiences. Aristotle (335 B.C./ 1967), in *Poetics*, said that a narrative has a beginning, a middle, and an end. Many scholars agree that narratives have a temporal structure, in the form of start and end points, with events unfolding in between. Polkinghorne (1995) argues that these events are organised thematically, through a plot. A plot is the organising theme that structures the story and makes it function as a unified whole rather than as disjointed events. Plot, for Ricoeur (1980) is “*the intelligible whole that governs a succession of events in any story*” (p.171). The existence of a plot is a vital difference between lives as lived and lives as narrated. From the perspective of the individual who is going through it, lived experience lacks plot, as the effects, or underlying reasons, of the unfolding events are not known. In other words, when we live an experience, we do not know what the end will be, neither do we always have an overview of the factors that led to that experience. The organisation of these events and experiences into a story that makes some sense comes only after the events have been lived.

Mattingly (1994) described the process of producing a plot as ‘*emplotment*’. The distinction between plot and *emplotment* is a useful one because it reminds us that plot is not inherent in the narrative but produced by the narrator, or by the audience. The existence of a plot does not imply that narratives run smoothly in a cause-effect way, neither does it assume the existence of an end beyond which further stories are no more possible.

Narratives can be fragmented and disconnected, and the plot may be hard to detect.

Narratives presuppose the existence of a narrator and an audience, who participate in the sharing of the story, interpret, and shape it accordingly. Narrator and audience give to the story its meaning and construct its plot. Chase (2005) refers to narratives as interactive performances, while Good (1994) states that “*in order to constitute narrative, the story must be appropriated by a reader or an audience*” (p.143). For Riessman (2001), “*storytelling is a relational activity that gathers others to listen and empathise*” (p.696). Not only does the narrator transform an experience and construct new experiences through telling a story, but the audience give their own interpretations to the events narrated and draw their own conclusions. In other words, they develop their own plots.

Narratives, according to Smith and Sparkes (2008) “*are not natural [...] but are social creations*” (p.18). Narratives are produced within specific cultural and social settings, and to some extent they reveal something about the circumstances within which they were created. For Chase (2005), narratives are socially situated events, “*produced in this particular setting, for this particular audience, for these particular purposes*” (p.657). In her monograph ‘Venus on wheels’, Gelya Frank (2000) chose to tell the story of Diane DeVries, a woman born with the congenital disorder tetra-amelia (underdeveloped or missing upper and lower limbs), by situating both women in space, time and culture, and

relaying several events from both women's lives. Writing for an academic journal she adopted a different style, with Frank's role in the construction of the narrative being put in the background and her interpretations being foregrounded (Frank, 1984). Had DeVries told the story it would have had yet another focus, as the standpoint of the narrator would have been different.

The constant refocusing of stories depending on the standpoint of the narrator becomes even more obvious when a story is narrated by many different people. Seeking to understand why a particular technology failed, Latour (1996) studied the stories that all people related to this technology had to share; his informants included a long list of engineers, bureaucrats, representatives of the public, and scientists. The product of this study is a book that is half a novel and half a scientific report. Halfway through it, Latour's alter ego, Norbert, a young sociologist, is exasperated by the multitude of the different stories that each informant gives, despite all starting from the same externally observable and verifiable facts. It was not only the existence of a multitude of stories that was so challenging for Norbert. It was the fact that none of these stories took precedence over the others that made it impossible to find one *true* answer to why the technology failed, as all answers were true in their own right. All narrators were telling a story that made sense to them. This multitude of stories that Latour (1996) observed points to another feature of narratives: they are concerned about individuals and their roles in stories. The focus is not on the events but on how they are interpreted and experienced by the narrators, as similar events

can mean different things to different people (Frank, 1995; Mattingly, 1998a; 2000).

Summing up, narratives in this study are dynamic. Rather than concerning one truth, or a grand narrative, their remit and focus is with individuals and with multiple interpretations of stories (Rodriguez, 2002). Following Eco (1989), narratives in this study are viewed as ‘opera aperta’ (i.e. open works): they are open to multiple interpretations, constructed by different narrators for different readers. All interpretations are *true*, to the extent that they are meaningful for a narrator and/or a reader. Borrowing from the Personal Narrative Group (cited in Riessman, 2001, p.704), it is “truths” rather than “the truth” that narratives in this study explore, illustrating the various ways through which people with MND make sense of their lives. The following paragraph discusses my role, as the researcher, in the authorship of the narratives.

#### *4.4.2 Whose narrative? Issues of authorship*

Storytelling is an intersubjective experience, happening between a narrator and his or her audience, and its value lies in that relationship rather than on an accurate reproduction of events (Jackson, 1998). Narrators transform an experience and construct new experiences through telling a story and audiences give their own interpretations to the events narrated and draw their own conclusions.

In this study, narratives were approached from a postmodern perspective, acknowledging the existence of multiple perspectives and

voices, all interconnected and equally valid (Lyotard, 1984). As Bakhtin (1995a) but also Lyotard (1984) remind us, all these perspectives are equally valid, although some may exercise more power than others. Narratives however are at some point actually constructed, told and sometimes written by one individual, who may or may not be a researcher. This implies that by virtue of being a narrator, one person's voice assumes authority to select, present and explain lived life in a particular way. In a paper discussing authorship of research narratives, Josselson (2011) asks "*whose narrative is it in narrative research?*" (p.33). In other words, whom does the narrative belong to and whom is it about? Josselson (2011) argues that the narrative belongs to the researcher and is about an experience, rather than about particular persons.

During interviews, I engaged in a dialogical relationship with the participants, with the focus being to generate rich stories that describe the experience of living with MND. Through my approach to data collection and analysis and writing-up, I sought to acknowledge and respect the intersubjectivity between the participants' voices and mine (Chase, 2005), viewing us all as "*two [or three] active participants who jointly construct narrative and meaning*" (Riessman, 2008, p.23). The products of the analysis were the reconstructed stories of the participants, seen through several theoretical lenses. The various methodological and epistemological decisions (for example, theories that were used in the analysis process, questions asked during interviews and questions not asked etc.) were mine. I analysed the stories from a postmodern epistemological stance, believing

that all stories are valid, and potentially incomplete or fragmented. My decisions, thoughts and feelings became part of these stories, and they are presented throughout the methodology and the findings chapters.

I acknowledge that the products of this study, the narratives, were written by myself and it was me who made decisions on what to include and what to leave out. However, in contrast to Josselson (2011), I believe that these narratives refer to particular people's experiences. Although the narratives presented in this thesis might hold meaning of greater significance for other people as well, the point of departure for this study were specific people and their experiences and these were therefore foregrounded during the writing up of the narratives.

#### *4.4.3 Narrative inquiry in this study*

The focus of this study was on perspectives and experiences of daily life of people living with MND and their partners. This study sought to construct and present multiple narratives, unique to each participant, and not on developing common themes across the experiences of the participants. I selected a narrative inquiry design because of its sensitivity to multiple realities of living with an illness.

The selected design allowed me to focus on stories about lives lived with an illness, focusing on how people make sense of their experiences, rather than foregrounding illness as such (Garro & Mattingly, 2000). It was not an aim of this study to represent or reproduce an objectively observable reality, but to create meaning through the production of narratives. In this

study, narratives were not considered as stories about events that were merely extracted by the people who narrate them; rather, they were co-constructed events, developing from the interaction between listener and narrators. Following Mattingly (2010), narratives in this study accept and even foreground the “*unfinished, idiosyncratic, unpredictable, suspenseful qualities of life*” (p.44). These stories open up the possibility for alternative explanations and interpretations of experiences.

#### **4.5 Chapter summary**

This chapter presented the background to the research design. Narratives were defined, and their use in studies on illness experiences was discussed, before detailing how narratives are conceptualised in this study. The role of the researcher in creating these narratives was also discussed.

Summing up, this study was guided by a narrative inquiry design that allowed the exploration of the unique ways that people live and make sense of their life. Narratives were conceptualised as occurring from the interaction between the research participants and myself. The focal point of narratives as used in this study was with specific people and how they experienced their life. Narratives revealed not only what these people were doing to the world, their actions, but also what the world did to them and how they interacted with the world around them (Mattingly, 1998a).

## 5. Methodology

### 5.1 Introduction

Seeing all worldviews not as theories of knowledge about the world but as existential means of achieving viable ways of living in and with the world (Jackson, 2012, p.123).

This study followed a narrative inquiry design focusing on experiences of daily life of people living with MND and their partners. The aim of the study was to explore the experiences of the participants and how they make sense of their life. The research methods detailed in this chapter aimed at creating what Dossa (2009) called “*a paradigm of telling and listening*” (p.26). Some of these methods, and especially data collection, were developed in the course of the study and through the interaction with the participants, and the rationale for this will be explained both in this chapter and in the findings chapters.

In this study, participants were seen as “*subjects-who-know*” (Pols, 2005, p.204). Through the methods detailed in this chapter, I aimed at creating an environment where participants could share their stories and I could be attuned to these stories and listen to them. Jackson’s (2012) stipulation that introduces this chapter guided the design of this study in so far as my interest was not in producing a general theory about the experience of living with MND, but rather in exploring how specific people live in their local contexts. The use of a narrative inquiry design enabled the emergence of multiple stories, unique to the participants of this study. The development of these unique stories was important, as the literature review

revealed that there is limited knowledge on how people who live with MND experience life in their local contexts.

Methodologically, the study was based on Polkinghorne's theory of narrative inquiry. Polkinghorne (1988; 1995) described two types of methodologies for narrative research; 'paradigmatic analysis' and 'narrative analysis'. This study was guided by the narrative analysis methodology, where the emphasis is on constructing narratives based on the stories shared by the participants. Rather than breaking stories into themes, the purpose of the analysis was to interpret them and construct them into coherent narratives.

The narrative inquiry design underpinned all methodological decisions. Participants were seen as collaborators in the construction of narratives, while at the same time it was recognised that I would develop the final interpretation in the form of the findings. Recruitment was carried out with the specific aim to engage with participants who could share rich stories, while data were collected through semi-structured interviews in order to enable participants to share what was important to them. Data analysis was sensitive to the complex nature of the experience and the many levels of interpretation, and it was closely linked to data collection so I could collect more data as the analysis progressed.

## **5.2 Sampling and recruitment**

Participants were selected with the aim of collecting rich data on living with MND. This richness was thought of as being inherent in the different stories

shared by the participants and their different experiences rather than to their external characteristics, such as age or time since diagnosis. For this reason, criterion rather than maximum variation sampling was used (Patton, 2002).

Prospective participants had to meet the following inclusion criteria:

1. a) Be diagnosed with MND ***or***
  - b) Be partners or significant others (with the relation extending to prior to the onset of the disease) to someone who has been diagnosed with MND and has consented to participate in the study.
2. Be over 18 years of age.
3. Not live in residential care settings.
4. Be able to engage in dialogue in English and communicate thoughts with or without the help of AACD, personal assistants or other means.

The final criterion was very important because the experiences of people with PBP are often not heard in research due to the speech impairments associated with this subtype of MND (Billinghurst, 2001). Pols (2005) stated that in research “*to have a perspective, one needs language*” (p.205), and people who talk little or not at all are often not represented in studies on illness experiences. In this study, the interview, and the ways to capture it, were modified to accommodate potential difficulties with oral speech. The strategies used are described in paragraph 4.3.1.1.

Recruitment of participants took place through the Motor Neurone Disease Association (MNDA) and through an online discussion forum for people with MND. Previous qualitative studies about MND have used similar recruitment routes (Brown & Addington-Hall, 2008). Recruitment commenced after the appropriate ethics committees (Research Ethics Committee of the School of Healthcare Studies, Cardiff University, and Research Department, MNDA) had approved the study protocol.

Information packages were forwarded to potential participants through the regional care coordinator of the MNDA. These packages included an invitation letter (Appendix D), an information sheet (Appendix F), a reply slip and a pre-stamped envelope. These packages were distributed in support network meetings attended by people with MND and their families or friends. To avoid the possibility of perceived coercion to participate, potential participants were invited to contact the researcher directly for more information should they be interested in participating in the study, rather than be expected to disclose their intentions during the meeting. In order to facilitate communication, a choice of communication means was offered, including an email address, a telephone number, a mobile phone number for text messaging, and a postal address. This was deemed to be important, as people living with MND often experience speech impairment and diminished motor skills.

The invitation letter to the study was also posted on an online discussion forum for people with MND (Appendix E). This particular discussion forum was chosen as many people with MND and their families

used it. The invitation letter contained identical information to the hard copy invitation and prospective informants could contact me through any of the means described above. Furthermore, the discussion forum offered a personal messaging facility and prospective informants were invited to use that function as well.

The inclusion criteria and the recruitment strategy were developed with the explicit aim to *hear* the stories of people living with MND. By using a two-route recruitment strategy, one based on electronic communication and one based on face-to-face meetings, I aimed to access a variety of people. Especially through the online forum, I aimed to reach people who may have limited support networks or who, for various reasons (for example, living in a rural area or not having access to transportation) could not attend support network meetings. Furthermore, while the inclusion criteria were specific to the aims of the study, they were broad enough so as not to unduly exclude people. However, as highlighted by Wiklund-Gustin (2010), it was inevitable that some stories would not be heard.

Aiming to explore how people live and make sense of their daily life, I decided to focus the study on people who live primarily at home rather than at residential care settings. The main reason for this choice was that most people with MND live at home and wish to continue to do so even in the later stages of the disease (Whitehead et al., 2012). Another group that I was concerned whether they might be excluded were young and middle age adults with family commitments, as they need to consider not only their own needs but also those of their children. Through the electronic

recruitment route, a woman with MND who was a single mother was initially recruited in the study, but the data were not analysed and she was not included in the study, for reasons explained in paragraph 12.5.2.

Seven people participated in the study; four people with MND and the partners of three of them. The partners were also informal carers. Due to the nature of data collection, which is explained in more detail in paragraphs 5.3.1.2 and 12.3.2, it is more accurate to describe participants as being three couples and one person, who while part of a couple she chose to participate in the study without her partner.

All participants were recruited through the MNDA recruitment route. The question of ‘how many participants’ is often asked in research and this study was no exception. In answering this question, I was guided by my focus on individual experiences. Qualitative research literature suggests that recruitment is often driven by the data and is deemed complete when the data are saturated and no new information is forthcoming. Sandelowski (1995) refers to this process as data-based sample size. While the overall principle of data-based sample size was followed in this study, it had to be modified. As experiences of illness are unique to each individual (Kleinman, 1988), overall data saturation can never be achieved. This would imply the existence of a finite number of experiences and ways to make sense of life with MND. Instead of saturation, the concept of ‘substantial difference’ or ‘variation’ was used. Variation referred not to participants’ characteristics such as age, or time since diagnosis, but to the experiences shared through data collection. Recruitment was deemed to be complete,

when the stories shared by the participants were substantially different from each other, highlighting varied ways of making sense of living with MND.

### **5.3 Data collection**

The term data collection as used in this study refers to the production of representations of experience. Data were collected over a period of approximately 25 months through the use of multiple (four to six) semi-structured interviews and observations with people with MND and some of their partners. The participants in the study participated in twenty three interviews, which lasted between one to two hours each resulting in more than 1,000 pages of data in total (transcribed interviews and field notes from observations). The following paragraphs explain and detail the processes that were followed.

#### *5.3.1 The interview*

In order to facilitate an in depth exploration of participants' stories and give participants the space to shape data collection, I used a semi-structured interview format. Interviews focused on experiences of daily life and also on how people made sense of their life. According to Kvale (2007), the semi-structured interview format is an appropriate method to explore experiences from the perspective of research participants as it enables them to focus on what is important to them. I treated the interview as an opportunity for the production of rich stories. The interview was a "*discursive accomplishment*" (Riessman, 2008, p. 23), in which the participants and myself were active collaborators.

Through the use of open-ended questions, I aimed at eliciting stories about the experience of living with MND and at the same time letting the participants free to narrate any stories they perceived as relevant and important for their experience (Miller & Crabtree, 1999). Following Holstein and Gubrium's (1995) discussion of the interview as an occasion where meaning is constructed through the interaction between interviewer and interviewee, participants and myself jointly constructed stories during the interviews. In order to accomplish this, I created the circumstances for extensive narration, as Riessman (2008) advises. This entailed giving up control of the interview and exercising minimal direction. I would ask questions about issues that interested me, but without having a set of answers in mind. Often a question would elicit a long story, intertwining past experiences, reflections on living with MND, and childhood memories. Rather than deciding myself what was important, I let the participants decide what was relevant to their story and let them tell it uninterrupted, unless it was for clarifying questions. Participants shared important stories about their life and their role in it, and all this information was very useful in reconstructing their stories.

While I sought to create a story with the participants, reaching a consensus between the interviewees and myself was not a goal of the interview. Following Fontana and Frey's (2000) discussion of the postmodern interview, I acknowledged and respected all different perspectives accepting the presence of more than one story and more than one interpretation for each story. For example, a participant discussed a

particular medical procedure in many instances throughout data collection, often in different ways. Sometimes it was referred to as something positive, sometimes as something negative, and sometimes she was ambivalent about it. Rather than trying to reach a definitive answer, I accepted all these stories as equally true to this participant.

Data collection took place in each participant's house. Multiple interviews were conducted with each participant, with each interview lasting approximately one to two hours. Each interview built upon the preceding ones, going into more depth until a storyline emerged and a narrative was constructed. A digital dictaphone was used to audio record all interviews with the participants' consent. Data collection continued for a period of 25 months but not everybody was involved for the duration of this time. Individual circumstances, such as disease progress, medical appointments and availability for interviews determined how often I would meet with participants and therefore how long the participation would last.

Before the first interview, I met and informed each participant about the research process. Consent forms (Appendix G) were signed during the initial interview that occurred one week or more after the initial meeting. The first meeting was also used to establish rapport between researcher and participants and to develop an overview of each participant's narrative. The establishment of rapport was particularly important as I was inviting people to share deeply personal thoughts, feelings, and experiences about their life.

Each initial interview started with a general question. This allowed the participants to decide what is important and what story they wanted to

tell (Chase, 2005). While each initial question was slightly different, they were all variations of the following question: “*can you please tell me what it is like to live with motor neurone disease?*”. This question often led to a long story that touched upon several experiences. Issues that often featured in participants’ stories included the diagnosis; negotiating daily life with MND; encounters with healthcare professionals; and explanatory models of MND. Consequent questions focused on developing a deeper understanding of the unfolding story, and I would also ask some clarifying questions, for example about the timeline of the events narrated or their significance for the participants (Appendix H).

Subsequent interviews started with a brief summing up of the preceding interview and proceeded with clarifying questions, seeking to further develop the emerging story. I asked questions with the aim of both confirming that story, but also of challenging it by exploring the possibility of alternative stories. Towards the conclusion of data collection with each informant he or she was invited to summarise the emerging story for the researcher. This was done in several ways: some provided a basic timeline during an interview, outlining some main events, while others provided a new story, going into more depth in some of the storylines explored in preceding interviews.

Data collection was guided by the concept of theoretical saturation, modified for the specific circumstances of this study. Theoretical saturation is defined as a stage in the research process when there is no more new information forthcoming (Murphy et al., 1998). As the aim of the study was

to explore how people make sense of their life with MND and how they experience it, rather than to draw comparisons between people or construct an overall explanatory theory, in this study saturation was applied to each participant or couple of participants separately rather than to all of the participants as if they were one entity. The decision to discontinue data collection with a participant was taken when I believed I had developed an understanding of how they made sense of their life and I had adequate data to support that understanding and justify my interpretations. Having adequate data referred not only to data quantity (how many interviews) but also to the quality of the data (how rich they were in information) (O'Reilly & Parker, 2013). The following two sections present two particular issues with interviewing in this study.

#### *5.3.1.1 Dealing with the effects of speech impairment*

Some of the participants spoke with what has been described as “*broken and vicarious voices*” (Hydén, 2008, p.36) due to bulbar symptoms. While they were able to create meaning out of their life, communicating that meaning was not always easy. Four participants presented with dysarthria and one of them used a lightwriter, which is a portable machine looking like a word processor that turns text into speech (Appendix A). Three participants had dysarthria but their speech was still intelligible although sometimes laborious and slow, requiring frequent breaks.

Adaptations were necessary in order to enable participation. The following strategies, as suggested by Philpin, Jordan and Warring (2005)

and King (2005), were used when interviewing people who experienced communication difficulties:

1. Participants were given as much time as necessary in order to communicate.
2. When a participant used a lightwriter, I refrained from talking while the participant was typing, as also advised by Wengraf (2001). While this may appear to be common conversational practice it can be quite challenging at the beginning. Depending on the length of the speech typed and the typing speed, this process can lead to a silence of between a few seconds to a minute or more. Initially, I had the urge to jump in and guess what participants wanted to say or finish their sentence (the lightwriter sometimes would produce speech word by word and then repeat the completed utterance at the end). Instead of interrupting them, I attended to the process of speech input instead. The participant would signify that the utterance was complete by switching off the machine or by looking up to me from the screen. Adjustments to this process are discussed in paragraph 7.6.
3. The text-to-speech equipment of one participant spoke with an American accent that I sometimes found hard to understand and I would ask the participant for confirmation of my understanding.

4. While interviewing the three people who had dysarthria but whose speech was still intelligible, I sometimes had to ask clarifying questions to ensure accurate understanding and recording. This happened mostly during the first interview while I was getting used to their speech pattern.
5. During interviews with the three participants who had dysarthria but were not using AACD, the microphone sensitivity of the dictaphone was set to maximum because the sound volume of the speech was often low.
6. Partners would often assist the person with MND in telling their story. This is further discussed in the next paragraph.

Moreover, following Hydén and Antelius' (2010) stipulation that

Stories are actually embodied in the gestures, the linguistic, para-linguistic, non-verbal and other physical artifacts that are used as resources in telling and listening to a story (p.590),

I paid particular attention to the non-verbal elements of the communication, such as gestures, gaze, pointing, and blinking and also to para-linguistic elements such as grunting, groaning, and sighing. The para-linguistic elements were captured through the recording while the non-verbal elements were recorded in the field notes after the interview.

### *5.3.1.2 The rationale for carrying out joint interviews*

The term ‘joint interviews’ is used to refer to interviews that include more than two participants (Seymour, Dix, & Eardley, 1995). In this study, I use the term to refer to an interview between one researcher and two people who share an experience. This use of the term is inclusive of joint interviews referred to by other terms, such as ‘multivocal occasion’ (Holstein & Gubrium, 1995) or ‘couple interview’ (Bjørnholt & Farstad, 2012). In joint interviews, the two participants are interviewed at the same time, together (Morgan, Ataie, Carder, & Hoffman, 2013). This requirement to interview participants together differentiates joint interviews from multi-perspective interviews, because in the latter research participants are not necessarily interviewed together, as Kendall et al. (2010) demonstrated.

As discussed previously, participants in this study were four people with MND and three of their partners. When early on in the data collection process I suggested that the three couples be interviewed separately, this suggestion was not met well by informants. They expressed a strong desire to be interviewed together because they viewed living with MND as a shared experience. Consequently, the three couples were interviewed jointly to enable the exploration of the co-construction of illness experiences (Morris, 2001; Radcliffe, Lowton, & Morgan, 2013). There were several reasons that led to the decision to carry out joint interviews with these three couples, ranging from practical to ethical. This section will explain the rationale that led to that decision and the impact joint interviews had on the data collected.

Some of the participants in this study were people who lived with serious impairments; some could not speak without the use of communication aids, several could not walk or use their upper limbs and most used wheelchairs. Also, some participants needed a carer next to them at most times to help with saliva management or body repositioning. In that context, I recognised that it could be stressful to separate the couple and I therefore respected participants' request to carry out joint interviews.

Furthermore, due to the speech difficulties experienced by some of the participants, the partners often acted as the animators of the story narrated by the persons with MND, assisting them in telling their story. This meant that on some occasions the partners had to narrate their own story but also the story of the person with MND. Often it was a joint story that the couple had discussed and agreed upon and it was relayed during the interview process. Sometimes though there was divergence of perspective, and in these cases I had to ask clarifying questions to ensure it was clear which was the story of the person with MND and which the story of the partner, if the latter was animating both.

Carrying out joint interviews presented several benefits but also several challenges. Often the partners would expand on short answers given by the person with MND, and then the person with MND would signal approval or disapproval. On many occasions, either of the partners would remind the other about experiences and occurrences and by rekindling each other's memory they would offer a richer story, something that has been observed by other researchers as well (Bjørnholt & Farstad, 2013; King,

2005; Philpin, Jordan, & Warring, 2005). Joint interviews also helped overcome some of the communication challenges due to speech impairment. The partner would sometimes animate an entire story or parts of it, especially if it was a story previously agreed by the couple. Joint interviews also enabled a valuable insight into the relationship of the couple and how they made sense of MND within that context, which was a big part of their life, as suggested by all three couples' assertion that living with MND was a shared experience.

As interviews were joint, the individual perspectives of the couple may have not been as clearly communicated as they might have been through individual interviews (Kendall et al., 2010). This was a considerable concern of mine especially during the first interviews. Going back to the aims of my study, I was reminded that I wanted to explore how people make sense of living with MND in their own context. For these three couples, the experience of being in a relationship lasting over 30 years was a vital part of who they were and how they experienced MND. Therefore, I decided that seeking an *individual* perspective from these participants would be contrary to the aims of the study, removing participants from their local context. Their experience was essentially intersubjective and data collection had to be sensitive to this, and appreciate and respect this. Other researchers (Cort, Monroe, & Oliviere, 2004; Taylor & de Vocht, 2011) have also stressed the importance of interviewing couples together when studying the effects of serious illness, as the “*coupled relationship will influence how the course of the illness is negotiated by the patient*” (Taylor & de Vocht, 2011, p.1576).

Similarly, Holstein and Gubrium (1995) stress the importance of constructing the interview as a multivocal occasion with more than one participants present, when this can contribute to the meaning making process of the interview.

Individual perspectives were still expressed through the joint interviews, but sometimes they were not easy to discern. Often when partners of people with MND would animate the story, they would at the same time include their own interpretation of it, thus leading to intertwined stories, one entering the other. In these cases, I had to be careful to establish if they both agreed with the interpretation by asking clarifying questions.

### *5.3.2 Observation and field notes*

Congruent with the understanding of narratives as embodied and enacted practices and not only as oral speech (Alsaker, Bongaard, & Josephsson, 2009; Hydén, & Antelius, 2010; Mattingly, 2010), alongside the interviews I engaged in observations of the setting and the unfolding interactions between the interview participants, including myself. This was done in order to enrich the data with all the nuances, feelings, and interactions that could not be captured by the audio recorded interview (Clandinin & Connelly, 2000). This was particularly important in this study because several of the participants had a speech impairment.

Following Philpin et al. (2005) and King's (2005) advice on how to address the effects of speech impairment in interview-based studies, I was careful to observe all aspects of the interaction, such as location (for example, room of the house), positioning of participants and myself in

relation to each other, time, the interaction between the participants (for example, instances when a partner had to care for the person with MND), and the non-verbal elements of the interview, such as gestures and smiles.

These would be written down as soon as possible after the interview as advised by Patton (2002), often in the form of brief bullet points while on the train returning from an interview in order to capture as much information as possible. This first draft of the field notes started with a description of the interaction, including time, place, and participants. Then, I would revisit the notes later on the same or the following day and rewrite them adding more detail and also expanding upon my feelings and reactions. These field notes were part of the data and were used in two main ways. Firstly, while listening to the recorded interviews, I often noticed periods of silence. The field notes enabled me to enrich the transcribed interviews with what was going on during these silences. For example, knowing about the gestures a participant was making during the interview enabled me to interpret an interaction more fully rather than if I was only depending on the recorded material. Secondly, the notes would help me be aware of my own role and impact on the data, and my decision making process during data collection. This was important for the writing of the methodology section of this thesis, but also to ensure the rigour of the study by keeping detailed field notes that could be used as an audit trail.

Several reasons led me to the decision to write down notes after rather than during the interview. As four of the participants presented with varying degrees of dysarthria, it was important to stay focused on the

unfolding interaction in order to ensure accurate understanding. Beukelman, Fager and Nordness (2011) report that increased attention allocation to the speech of people with dysarthria increases understanding. Furthermore, note keeping during the interview would create an even bigger distance between the participants and me, in that it would be a constant reminder that I was *researching* them (Clandinin & Connelly, 2000). While they were aware that this was a research interview and I reminded them of this at the beginning of each interview, I did not want to replicate traditional power dynamics by assuming the role of the observer and allocating the role of the observed to the participants. This would be incongruent with my understanding of the interview as a dialogical occurrence where meaning is co-created.

#### **5.4 Data management**

Data in this study were audio and textual, in the form of audio recorded and transcribed interviews and in the form of field notes. Each interview was audio recorded using a digital audio recorder and then it was transcribed verbatim. Field notes were written on a laptop computer as Microsoft Word files. After each interview, both the audio and the textual data were stored onto a password protected personal computer and cross-linked to the qualitative data analysis software package NVivo 8.

Transcription was the first level of analysis (Riessman, 2008); I therefore decided it was important to carry it out myself rather than assigning the task to a research assistant. Doing this enabled me to develop a deeper understanding of the interaction and contextualise the field notes

for each interview. Following Beukelman, Fager and Nordness' (2011) recommendation, during transcription I listened very carefully, in slow speed (-70% or -60%), especially when informants experienced difficulties with oral speech, in order to mitigate the effects of dysarthria to intelligibility of speech. Transcription included non-verbal communication, such as pauses or laughter, as these were important elements of the interaction and were necessary for the analysis (Kvale & Brinkmann, 2009; MacLean, Meyer, & Estable, 2004).

Several punctuation symbols are used in the transcribed text. Square brackets are used to denote text added by myself within a quotation (for clarity or to offer information on non-verbal elements). A series of three full stops within brackets is used to denote text that has been removed. A series of three full stops indicates a pause. Double quotation marks in block quotations and single quotation marks in in-the-text quotations indicate quotations within the quotation; for example, "*Rhian might say 'let's try it this way' like*" (see also key to transcription symbols, p.xvi).

The audio files, the transcribed text files, and the field notes were backed up on an external hard disk that was kept at a secure place. All interviews and field notes were organised by date in participant specific folders. In order to ensure confidentiality, the folder and file names were identified only by a participant number and the date. Participant identifying information was kept in a different location from the data.

## 5.5 Data analysis

The purpose of the analysis was to develop narratives on how specific people make sense of life with MND. Constructing a theory or developing general themes was not an aim of this study and therefore the narratives were not compared to each other and no cross-case analysis took place. Data analysis focused on the following areas in order to reconstruct the narratives of the participants:

1. Actions, choices, and experiences related to the narrated events.
  
2. The varieties of knowledge that guided participants' understandings of life with MND, and how these different varieties of knowledge were enacted in daily life.
  
3. The enactment of practices of care in everyday life.

Methodologically, the analysis was based on Polkinghorne's (1988; 1995) narrative analysis method. I decided this was an appropriate method because I wanted to develop narratives for each participant in order to explore in depth their understanding of their life with MND. In narrative analysis, happenings are drawn together and integrated into a temporally organised whole (Ricoeur, 1980). These happenings are connected through a thematic thread, i.e. the plot (Mattingly, 1998a; Polkinghorne, 1995). The plot developed through an active engagement between the data and theory.

During the first stage of data analysis, data were read a number of times and important events and incidents, as defined by the informants, were identified. This led to the construction of some initial, possible storylines. In the second stage I used theory in order to interpret the story and further inform the plots.

In order to achieve the intended focus on experiences of daily life and enactment of practices of care, analysis was informed by a phenomenological approach, as described by Jackson (2010; 2012; 2013) and Mattingly (2009; 2013) and by an enacted approach as described by Alsaker (Alsaker, Bongaard, & Josephsson, 2009; Alsaker & Josephsson, 2011) and Mattingly (1998a; 1998b; 2010). This offered a vantage point from which to view participants both as *sufferers* and as *actors*. The interaction between the two approaches enabled a nuanced understanding of participants as actors who can sometimes use power and subject themselves to different practices of care and sometimes they are subjected to power.

The analytical process I used included seven steps, organised in three stages. The first stage consisted of three steps, focusing on understanding the participants' stories. These first three steps were descriptive, aiming to develop initial plots. The second stage was interpretative, aiming to produce meaning by engaging the data in critical discussion with relevant theories. The third stage related to the interactive nature between data collection and analysis and the writing up of the narratives. Data analysis was closely linked with data collection so that new data were collected if there were gaps in the analysis and more data were

deemed to be necessary. This back and forth process between data collection and data analysis is referred to as iterative, and it is often chosen in narrative analysis as it enables the researcher to engage in further data collection as the analysis dictates (Riessman, 2008).

The three stages of data analysis did not progress in a linear way but rather there was a dialectic movement between all three. This movement is depicted in Figure 4.1. During data analysis, I looked at the complete set of interviews and field notes for each participant or pair of participants as a whole, rather than as separate items.

### **Stage 1**

**1. Initial reading.** In this step, I focused on the entire interview and read it several times, highlighting significant events within the stories. Also, in this step, I coded the data. I performed descriptive coding using the software package Nvivo 8, aiming to illuminate the main issues discussed during the interview, rather than aiming to develop these codes into themes. These codes were relatively broad and contained utterances that referred to a common issue discussed by participants (Saldaña, 2012). For example, all stories about the PEG were coded under one code. These codes were not exclusive and one excerpt often was coded under more than one code. For example, a discussion about attending a social activity was coded both under 'socialising' and under 'getting on with life'. As my interest in doing this coding was to identify main storylines, or emerging plots, in the data, I did not try to join codes into categories or arrange them hierarchically.

## **2. Initial potential links between my pre-understanding and narrated**

**events.** The aim of this step was to examine how my pre-understanding influenced the emerging plots. Throughout the study, I sought to be aware of my own preconceptions and rather than suspend them, I wanted to include these in the analysis. During this step, I engaged in what Ricoeur (1984) describes as a process of distanciation. What I was distanced from was not my preconceptions but the narrated events themselves. By taking a distance from the narrated events and looking at them as stories that hold some kind of meaning for the participants, I became cognisant of my impact on the data (Dreyer & Pedersen, 2009). One of the informants, for example, mentioned a particular process several times, and often in different ways. Rereading the data, it emerged that the participant actually held conflicting opinions about the procedure, sometimes discussing it as something positive and sometimes as something negative. As data collection progressed, this appeared to be common among other participants as well, who appeared to express seemingly conflicting feelings or opinions about some aspects of their life. Realising that participants could narrate seemingly contradictory stories was an important point in data analysis, as up to that point I rather naively expected to collect stories that if not smooth, they would at least reflect some sort of linear structure of meaning making. Instead, I had to attune myself to look for meaning in what at first appeared to be inconsistencies. It was in these inconsistencies that it was most evident how the participants made sense of their life on a daily basis. Meaning making

was continuous and unfolding before my eyes as people were embracing all the different perspectives and elements of their life. This step of the analysis enabled me to focus on what was important for the participants, rather than on what I thought was important.

**3. Reconstructing a storyline.** During step 3, I reconstructed the stories shared by the participants, aiming to understand them. The codes from step 1, as informed by my own reflections from step 2, led to the development of emerging plots, or storylines, outlining the main elements of the participants' stories. In order to develop these plots, I explored questions such as: "what is important in the daily life of this participant?", "how are the participants taking decisions about their daily life?", "how are they managing their care?", and "how is MND evident in their daily life?". These questions were developed as I became immersed in the data and answers to these questions led to an initial plot that connected meaningfully the narrated events into a story. During this step, I also referred back to my field notes to answer some of the questions I was asking of the data. Stories at this stage were more descriptive than analytical, and were informed by the coding performed during the first stage of the analysis. Also, during this step, I discussed the emerging plots in relation to the social, political, and physical context within which they were constructed, since according to Ricoeur (1984) narratives need to be understood within their context. For example, the health and social care services accessible to the participants

played an important role in several, although not all, of the participants' stories.

## **Stage 2.**

**4. Bringing in theory to interpret the story.** Up to step three I had constructed mostly descriptive stories, to a large extent replicating the information directly shared by the informants and my own reflections. While the basis of the emerging narrative was still the story shared by the participants, I decided to further analyse this story following Ricoeur's (1991) assertion that it is through interpretation and critical discussion with theory that narratives reveal their meaning. During this step of the analysis, I started to interpret the stories, using the theoretical framework underlying this study. Following Clandinin and Connelly's (2000) suggestion, I was asking the stories "*questions of meaning and social significance*" (p.130). Data were discussed critically in relation to theory, while theory was used to fill the meaning-gaps in the emerging story and offer an explanation of the story. In order to select the theory I went back to the aims of the study and the questions I wanted to explore. The various theories that were used are presented in the relevant findings chapters. My choice of certain theories does not exclude the relevance of alternative theories to the stories of the participants. Indeed, the stories could be seen and analysed through the lens of several theories, including psychological theories of control and agency,

or theories about suffering. Using these theories would have resulted in different stories, and therefore, a different study.

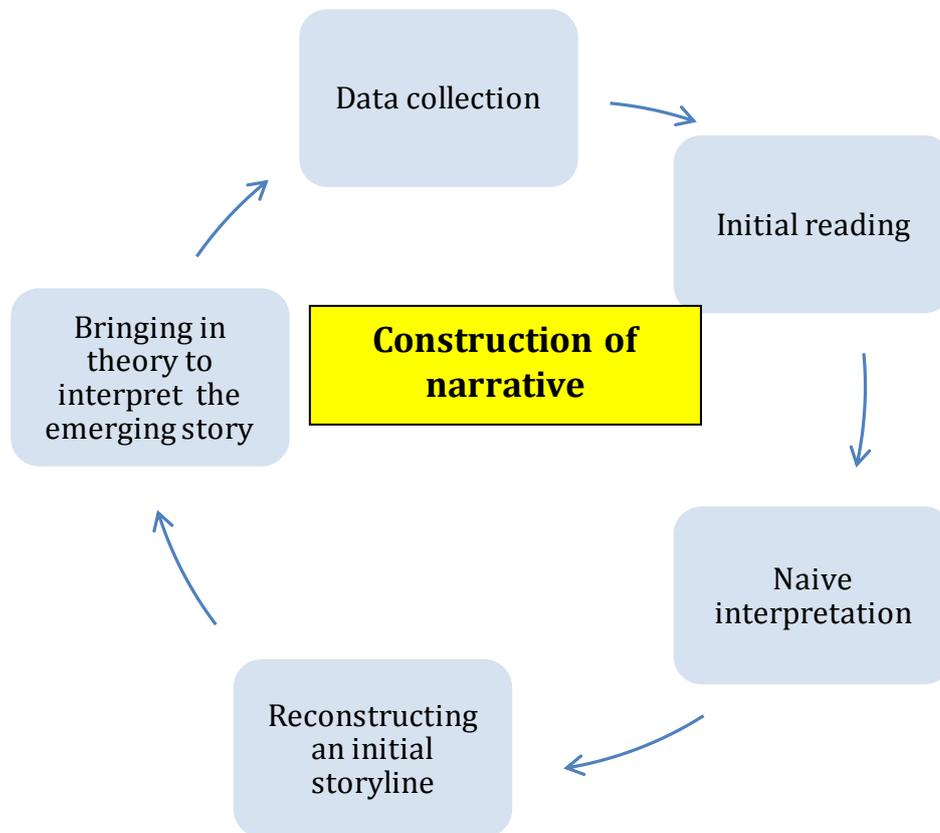
### **Stage 3.**

**5. Further data collection.** This step consisted of going back to the field to collect further data in order to enrich the emerging narrative. During the analysis, I often realised that I did not have enough information about a particular aspect of the participant's life and I would arrange to meet him or her again for further data collection. Rather than a weakness of the design, this was one of its strengths because it enabled me to return to the participants several times seeking more data, which sometimes was in the form of explanations or clarifications. During this step, I fed back to the participants, in order to see that I had understood their perspective on their story. During this step, I also sought data that offered alternative interpretations to the ones reached during the preceding stages of the analysis process.

**6. Reiterate process.** The data analysis / data collection iterative process was repeated several times for each participant until a meaningful narrative was reconstructed. This narrative was meaningful in relation to the events narrated and to the theories used to construct it.

**7. Reconstruct narratives.** These narratives were the end result of this study and were unique for each participant or couple of participants. Although unique these narratives are not exhaustive. Congruent with my postmodern conceptualisation of knowledge as fragmented and multifocal, these stories represent one of the ways people made sense of their life and one of the possible ways that I could interpret these stories. Other stories remain possible.

**Figure 5.1. Process of data analysis**



## 5.6 Quality of the study

The starting point of the discussion about the quality of this study is the fact that it resulted in unique narratives. As described in the section on data analysis, these narratives were developed through an interaction between the participants, the researcher, data, and theory. Use of different theories, different participants or researcher, or a different interaction between myself and the participants of this study, would all result in quite different narratives.

Quality in this study, therefore, does not refer to a notion of generalisability of the findings to a wider population. Neither does it refer to the reproducibility of the findings, if other researchers were to replicate the methodological steps that I followed. Instead, quality refers to depth and richness of data, and coherence of the final narratives (Creswell, 2013). I strove to produce data that would help me address the research aims and develop narratives that would illustrate the participants' experiences. The decisions taken and the processes used during the study, like reflection for example, cannot be presented as a separate neat category but they infuse the entire thesis, and especially this chapter and the findings, guiding the reader through my decision making process.

Several researchers have discussed different sets of quality criteria used in qualitative research (Kvale & Brinkmann, 2009; Morse et al., 2002; Patton, 2002) and there is an equally active discussion about the need, or even the appropriateness of using specific quality criteria in qualitative

research (Denzin & Lincoln, 2005; Sandelowski & Barroso, 2002). Cognisant of this debate, I use the concept of methodological rigour to refer to the specific processes that were used to enhance the quality of data collection and data analysis. Ensuring methodological rigour was important in order to achieve depth and richness in the data and produce coherent narratives. The two criteria that explain some of the decisions taken to ensure methodological rigour are dependability and credibility (Clandinin & Connelly, 2000; Patton, 2002).

Dependability is an indicator of whether the findings are actually based on the data. The iterative design of the study, whereby data collection and analysis were closely linked, ensured that data collected could inform the developing narratives in an interactive process “*between what is known and what one needs to know*” (Morse et al., 2002, p.18). Dependability was further ensured through maintaining a detailed audit trail to enable reproducibility of the research processes (Krefting, 1991), but not of the findings, as each story of life with an illness is different. The aim of the audit trail was to document my decisions and choices, making the research process transparent. I maintained an audit trail through detailed description of the entire research process and recording of theoretical, methodological, and analytic decisions. This information was recorded in a notebook, which informed the writing of the methodology of this study.

Credibility refers to the appropriateness and comprehensiveness of the research methods, including the data collection and analysis methods and the role of the researcher. This criterion ensures that the participants’

experiences are described thoroughly. One method to enhance credibility of this study was through the relatively long-term nature of the study. Meeting and interviewing participants over a period of several months facilitated the development of trust in our relationship. People felt freer to share personal experiences after the second or third meeting, and they also had time to reflect and enrich their stories in subsequent meetings. Another way to ensure credibility was through the interviewing process. I re-framed questions or expanded them on different occasions in order to receive rich information. I also often fed back to the participants, or reflected on the data to ensure that data were captured accurately (Krefting, 1991).

Furthermore, accurate and appropriate recording, transcription, and handling of the data were vital for the establishment of credibility. Interviews were recorded electronically and were transcribed verbatim, with paralinguistic communication, such as sighs and laughter, being included in the transcribed text. Detailed field notes were also maintained, which recorded non-verbal communication, such as smiling and pointing. In order for rigour to be evaluated more fully by the reader, the following paragraph outlines my roles in the study.

#### *5.6.1 The roles of the researcher*

As discussed throughout this chapter, my personal thoughts and feelings were part of the research process. I reflected on every step of the research process and I was consciously trying to be aware of my decision making process in order to illustrate my own influence on the research process, and

ultimately, on the findings (Finlay, 2002). My feelings and thoughts were recorded as part of the field notes and were included in data analysis and writing up of the findings, in order to make my contributions to the study clear. This paragraph discusses some specific issues relating to how I positioned myself within the study, in order for the reader to be able to make judgments about the credibility of the study. My role in the study is further discussed in chapter 5 and in the findings chapters (7-10).

I hold several roles as an academic, occupational therapist, and student, and could be 'storied' (Clandinin & Connelly, 2000) in different ways by the participants within their stories. During the study I intentionally emphasised my role as a student, as that was the capacity in which I met the participants and carried out this study. I am not a native speaker of English, nor acculturated in the UK and there was a large age gap (20 years or more) between the participants and myself.

These identities of mine may have positioned me partly as an outsider, influencing the study in several ways. I had to be particularly aware of the social context and do extensive research on specific issues that came up as part of the stories (for example, ideas about retirement and old age or cultural values associated with male carers). I also had to be aware that just as I had to get accustomed to the speech patterns of some of the participants, they in turn had to get used to my speech pattern. I found that perhaps due to being perceived as an outsider, participants were volunteering information about cultural context and background to their

everyday life as also observed by Manderson, Bennett and Andajani-Sutjahjo (2006).

I positioned myself as an outsider to the participants' experience of MND approaching this experience as a newcomer. However, I did have personal experience of MND as my mother lived with and died of this disease. The decision to not disclose this information to participants was taken after intensive deliberation and was based mainly on the following reasons:

1. Sharing my personal experience with the participants would position me differently within the study (perhaps as a friend, or as a colleague in disease, or as a former carer) and I was concerned this could radically change my role from being a researcher to a role I could not control. Having had personal experience of MND did not make me any more knowledgeable about the lived experiences and the stories of the participants, as these stories are unique for each individual. I was, therefore, honestly approaching the experience of each participant as a newcomer to it.

2. Disclosing my personal experience would require sharing how my story ended (with the death of my mother). I was aware that some participants, especially those in the early stages of the disease, might not be prepared for this. Some participants explicitly said during data collection that they did not want to receive too much information

about the progress of the disease. Sharing such information could have been perceived by participants as a projection of their own future and this could have caused them unnecessary distress.

3. Sharing this information could also evoke to me painful memories and feelings, thus running the risk of foregrounding my story rather than the participants' story. While I diligently explored my feelings and how these affected the research process throughout the study, revealing these to the participants would recast all participants in different roles and focus would potentially shift to me, as Berger (2013) warned when researchers include their personal experiences in data collection. I wanted to avoid this.

## **5.7 Ethical considerations**

This study was approved by the Research Ethics Committee of the School of Healthcare Studies, Cardiff University. Research procedures commenced after approval had been granted. The study also received clearance by the research department of the MNDA enabling me to distribute recruitment packages through the MNDA regional care adviser.

In order to achieve informed consent, potential participants received an information sheet (Appendix F), which contained full information regarding the research project, before they consented to participate. The explanatory statement outlined the nature of involvement, the procedures of the study (including handling of the data), potential benefits, and issues of confidentiality (Gracey, 2003; Kvale & Brinkmann, 2009; Seal & Barnard,

1999). Participants were given at least one week (no maximum limit was set) to think about their participation and sign the consent forms (Appendix G) if they wished to take part in the study.

Informed consent did not finish with the signing of the form but was an ongoing process, as advised by Katz and Fox (2004). During the data collection phase, I discussed with the informants questions about the nature of the research, the potential uses of the data and strategies to ensure confidentiality throughout the research process. Informants were free to withdraw any time they wished. I kept reminding participants that neither the study nor myself were associated with the MNDA and their participation in the study was in no way related to any services or support they may be accessing through that association. To stress that point and ensure that potential participants were not put under stress to participate, I did not attend social events and support groups organised by the MNDA.

In order to ensure confidentiality, all identifying information, including the audio files and the transcribed material, were kept in a safe, locked space. Pseudonyms were used for all participants and some of the details of their lives, like for example exact time since diagnosis, were altered if there was concern that participants could be identified through these particular details.

I was aware that data collection could be a challenging process for some of the participants. This was due to various reasons, including:

- Low physical stamina or concentration levels.
- The need to carry out personal care tasks.

- Compromised breathing.
- Discomfort when talking about sensitive issues.

I took every measure necessary to accommodate the needs of the informants, as suggested by relevant literature (Locock, Ziebland, & Dumelow, 2009), including:

- Frequent breaks as and when required.
- Some data were collected through email.
- Participants were encouraged to only share information that they felt comfortable sharing.

I was aware that the use of joint interviews might cause tensions in the relationship between the couple, especially if there was a discordance of opinion, or if partners shared information in front of each other for the first time (Bottorff et al., 2005; Gysels, Shipman, & Higginson, 2008b). Participants in this study seemed to be within their comfort zone during the interviews and even though discordances occurred several times, this did not appear to cause tension. Although it is not possible to know if tension arose after the end of the interview and my departure, the relatively long term nature of the study gave participants some opportunities to discuss such tensions.

I was also aware of the fact that this study and its findings would not necessarily lead to any benefits to the participants personally in terms of the healthcare available to them and I clearly explained this to participants. Research suggests that despite these challenges people with MND are keen to participate in research that will produce knowledge on the disease

(Gysels, Shipman, & Higginson, 2008a) and they are eager to impart their experiential knowledge of the disease (Locock, Ziebland, & Dumelow, 2009). Participants in this study were no exception to this.

Furthermore, I acknowledged that talking about living with an illness can be stressful or upsetting. I was attentive to any potential distress that participation in the study could cause and when necessary reminded participants that they could take a break, discontinue the interview or not answer a question. Due to the recruitment route this study followed, all participants had access to support services through the MNDA or through peer support groups and these could also provide emotional support.

Finally, taking the decision to not disclose about my personal experience was a very difficult one. My main concern was the wellbeing of the participants and I wanted to avoid any action that could cause distress to them. If I started over again, I would take the same decision. This, however, was not a perfect decision, and it presented me with a number of ethical dilemmas, which are discussed in paragraph 12.6.

## **5.8 Chapter summary**

This chapter presented the methodological processes used in this study. A narrative inquiry design was used to facilitate in depth story-telling by the participants. I sought to create a robust methodological framework which would be sensitive to the needs of the research participants and which could develop organically in the course of the study. Through data collection and analysis, I strove to co-create, together with the participants, rich data on their experiences and then analyse these data. The outcome of data analysis

were the four narratives that are presented in chapters 7-10. These narratives are prefaced by chapter 6, which acts as an introduction to the findings.

## **6. Introduction to the findings**

In the four chapters that follow (7-10) I aim to highlight how specific people make sense of life with MND in their local contexts. Seven people participated in the study; three people diagnosed with MND participated together with their spouses as couples, and one person participated by herself but she was not single. The people who participated are presented, using pseudonyms, in Table 6.1. Three more people had expressed interest to participate, but this unfortunately was not possible due to repeated hospitalisations, death or unsuitability of available data (this is discussed in detail in paragraph 12.5.2). I also had several conversations, through email or in person, with the care coordinator for the MNDA in Wales, so that I could understand better the wider context of services, as this formed the background to the experiences of some of the participants.

In the following chapters, I focus on experiences of everyday life and the enactment of practices of care, paying particular attention to the actions, choices, and desires of specific people who live with MND. Although I focus on specific people, I situate these narratives within a socio-political or theoretical context, details of which are incorporated in each of the narratives.

This study does not offer categories on how people make sense of living with MND or general themes. The four narratives presented in the following four chapters are by no means exhaustive of how people with MND make sense of their life. Many stories remain unheard. The narratives

in this study illustrate the unique ways some people make sense of MND and how this sense is enacted in their everyday life through decisions, interactions with other people or everyday activities. They show possibilities and variation, and highlight the uniqueness of the experience of living with MND, and the importance of listening to people and respecting their experiential knowledge.

The narratives presented in the findings are essentially unique, reflecting idiosyncratic regimes of living with MND. I wanted to avoid turning participants to “*familiar strangers*” (Mattingly, 2010, p. 235). Like Mattingly (2010), I “*offer portraits of individual lives (or, more accurately, segments of lives) and not merely typical social events or discursive formations*” (p.235). This, however, does not mean that the narratives provide a comprehensive account of the participants’ experience of living with MND. More accurately they offer one possible interpretation of how specific people make sense of living with MND, in their local context and in a specific timeframe, where they foreground some parts of their experience while some others are not given as much attention.

Consequently, the narratives that follow in the next four chapters do not make any claims for *the* experience of living with MND, but rather highlight different ways that people experience MND. Being unique, however, does not make these narratives relevant only to the participants. These narratives represent how people living with a progressive, incurable and terminal disease make decisions on how to live their life. While, as Jackson (2002) maintains, people cannot always determine the course of

their life, the stories they share can perhaps be a way for them to shape the meaning of their experiences.

The narratives presented in this study foreground what people desire, what they hope for, what they are afraid of and what makes them angry. These narratives highlight the complexity of living in the knowledge that the disease will only progress and will not go away. These narratives have something to teach us about what it means to live with a serious illness or disability.

These narratives could of course have been presented in many different ways, highlighting different issues. In chapter 7, for example, I chose to focus on what it means to construct a good life and perform self-management. Another potential storyline could have been one on independence, or on interdependence. Similarly, in chapter 9, I focus on the experience of caring in MND, as this was a prominent issue with that couple of participants. However, they also spoke about anger about having to live with MND; this could have been a different narrative. My main criterion for choosing which storyline to bring to the fore was data-based; I chose the storylines for which I had the richest data, so that I could present a detailed and plausible narrative. Chapters 7-10 present the unique narratives of the participants. Each of these chapters finishes with a reflection on the methodology of this study and my involvement in the production of the narratives.

Chapter 7 presents a narrative that focuses on the desire to construct a good life, and the practices that the participants mobilised in order to

achieve this. The chapter draws theoretically from Foucault and his ideas about biopolitics and more specifically the use of technologies of self (see for example, Foucault, 1994a; 2010). It focuses on the construction of a good life through the use of technologies of the self and describes what this looks like in the life of one specific couple. The chapter ends with a reflection on interviewing people who cannot talk.

Chapter 8 highlights the disempowerment that people living with MND can sometimes feel. The couple whose story provides the empirical basis for this chapter felt lost within the new world of MND that they involuntarily had to inhabit. Drawing theoretically from postmodern ideas about knowledge (Lyotard, 1984) and Bakhtin's notion of heteroglossia (Bakhtin, 1995a), chapter 8 foregrounds issues of power and disempowerment in the experience of living with MND. The chapter ends with a discussion on the use of joint interviews to explore shared experiences.

Chapter 9 focuses on the practice of care and its characteristics. More specifically, drawing from literature on care and caring (from medical anthropology, disability studies, and nursing), this chapter explores the performance and meanings of care in MND from the perspective of the person who offers care. The chapter situates the participants' experiences within the context of care in Wales and finishes with a reflection on my feelings of losing control during interviewing.

Chapter 10 highlights the experience of disability and the desire of people with MND to live their life without letting the disease affect them too much. To illustrate this, I draw on critical disability studies to discuss the importance of the body in experiences of illness, and highlight the contextual and transactional nature of disability as experienced by one of the participants. At the end of the chapter, I reflect on the interview as an interactive performance and I discuss my role and potential influence in the production of the narrative.

In Chapter 11, I discuss how the narratives presented in chapters 7-10 offer a response to the objectives of this study and I illustrate how participants in this study were experimenting with a notion of normality, not defined by illness, but shaped by everyday life and all its exigencies.

**Table 6.1 List of participants**

<b>Participants</b>	<b>Age</b>	<b>Diagnosis and main symptoms</b>
<p><b>Rhian and Gwyn</b></p> <p>Married couple. Rhian has MND.</p>	Early seventies.	Bulbar onset amyotrophic lateral sclerosis. Rhian uses a power wheelchair, a PEG and a lightwriter. She can use her head and one of her arms and her husband is her main carer.
<p><b>Dave and Marion</b></p> <p>Married couple. Dave has MND.</p>	Late fifties.	Amyotrophic lateral sclerosis. Dave uses a manual wheelchair inside and a power one outside. His legs are weak and he gets tired when talking for a long time.
<p><b>Gareth and Maggie</b></p> <p>Married couple. Gareth has MND.</p>	Early seventies.	Progressive muscular atrophy. Recently started using a wheelchair outside although still walks inside the house. Gareth still uses his arms, but range of motion in the shoulder is diminishing and cannot always lift arms high.
<p><b>Arleen</b></p> <p>Participated by herself, without her partner.</p>	Early fifties.	Primary lateral sclerosis. She uses a power wheelchair outside. Her limbs are weak and she gets tired when talking for a long time. She experiences dysarthria.

## 7. “But we got a good life, Rhian and I”

Rhian is in her early 70s and has been married to Gwyn for more than 40 years. MND has been present in their daily life for a big part of these years. According to most scales of functional independence, Rhian is severely disabled. She has some control of the muscles in her left arm and hand, neck, and face but uses assistive equipment in order to communicate, transfer, and eat. When I met her, Rhian was using the following equipment: power wheelchair, lightwriter, and nightly PEG feed. She was also receiving twice-daily visits from carers who helped with dressing in the morning and with some housework in the afternoon.

However, rather than viewing themselves as powerless in the face of a progressive, incurable disease, Rhian and Gwyn create their own solutions, working out different possibilities. Using literature on the morality of care (Kleinman & van der Geest, 2009; Mattingly, 2008) and Foucault’s (1994a; 1994b) work on technologies of the self, this chapter aims to illustrate how people with MND enact care through the construction of solutions that work in the context of their life. By putting the person in a powerful position where he or she can experiment with practices of daily life and select those that are necessary for the construction of the life the person wants to live, Foucault’s ideas helped keep Rhian and Gwyn as central characters in their story, rather than letting constructs such as culture, disease or healthcare services assume central role.

## 7.1 Setting the scene; The diagnosis

After raising two children and taking care of her family, Rhian decided it was time to fulfil her desire for further education. And so, in her late forties, after the children had moved out of the family home she enrolled at a local university to study sociology. At the time, Rhian was working as a seamstress at a local factory and Gwyn, her husband, was also working at another factory as a manual worker. As she later explained, it was not a need to pursue a more financially rewarding career that carried her. What motivated her was her desire to learn more, or as Gwyn put it “*to satisfy her inquisitive mind*”. However, during Rhian’s first year at university something was not quite right. Her legs would feel weak and climbing up and down steps in local buses was sometimes difficult. One evening, her daughter called and as they were chatting she asked “*are you alright mum?*”. She sounded different. Living with her, Gwyn had not noticed the progression into dysarthria.

Gwyn: Her voice went first. Slurred voice. Like, I didn’t know, because I lived [with] her. But my daughter noticed it. Cause, she does not come here very often. She said, “you are not talking very nice mum, what’s the matter?”. So, we know something was wrong.

This first observation that something was not quite right led to a series of diagnostic tests and a long quest for a diagnosis.

Gwyn: ...The first...the first three or four years...we used to go to the hospital. And we didn’t know what the hell it was. Because Rhian’s voice was going, we didn’t know what it was. We went to the doctor and he sent us out to see a specialist, every six months...so, we go down there, first time we go down there, get from the car park into the surgery, force a couple of needles in her arm, take them off her arm, “thank you very much, bugger off...come see us in

next six months”. Six months go by, we were back, same thing. “Aw, you got feeling in your arm, I got the electrical impulse in your arm are not working properly”, but that’s all he ever did like. And scan, big scan.

Eventually the diagnosis of MND came. Rhian was informed matter-of-factly about estimated life expectancy (four to five years) and was told that there is no cure. Rhian and Gwyn experienced the process of diagnosis as stressful and upsetting, reflecting the findings of a study by Pavey, Allen-Collinson and Pavey (2013) on experiences of diagnosis delivery in MND.

As Gwyn said:

Yes, I was still at work. It’s a big shock, like, you know, especially when they give you the pamphlet, and they say you are goner, twelve years, whatever it is.

The first few months after the diagnosis were difficult, with both of them trying to get over the shock of the diagnosis and figure out what their life would be like. Rhian had to retire on health grounds from the factory and discontinue her studies. Gwyn kept on working full time but, with the support of his employer, he would drive the 10 miles back home during the day if there was an emergency such as a fall. They both recounted the first few months as very stressful, which led for the first time in their life together to arguments, mostly focused on ways to manage living with MND.

But then something changed. As they put it:

Gwyn:…And then you say well, whatever time we got left…use it best way you can.

Rhian: There is no alternative.

Gwyn: Well, the only alternative is to vegetate isn’t it? And, uhh, and let it get on top of you like. To let it get on top of you is going to destroy you.

According to Thorne, Paterson and Russell (2003), the decision to assume control over an illness often comes early on, when people realise the chronicity of the situation and acknowledge that the disease will be a feature of their life. And so, Rhian and Gwyn *accepted* MND into their life.

During our meetings they often laughed as they recounted stories of loss, thus, turning MND into just another part of daily life rather than a focal point dominating their lives. In other words, while what they described could be interpreted as loss and suffering (for example, a story about an unsuccessful transfer from the commode onto the bed which led to a fall), they did not view their life in that way. For them, it is about making the best of what one has and living and hoping, rather than experiencing loss.

Gwyn: A week last Saturday it was. Get her out off the bath, her feet sled on the floor (...) I don't have no bare [free] hands so she drops on the floor. Now, she is sitting on the floor now, she is, and I'm thinking, "how can I pick her up" like. Because, you can't get between her arms to lift her, you can't lift her up by the elbows, so we seem to be in a quandary, which way can I do it like (...) We had a laugh about that like. Can't go, "oh, oh oh, can't do it". What am I gonna do like. Try this, it doesn't work, trying that it doesn't work, but something has got to work anyway.

Rhian:...ateed.

Gwyn: Attitude? We have a positive attitude. Hard word to spell that one, attitude.

Rhian: ateedude. [laughter].

Gwyn: Yeah, we laugh, don't we?

Rhian and Gwyn create their own solutions, working out different possibilities. In order to do this they had to learn how to manage the disease and fit it into their lives.

## 7.2 Self-management or managing a self

Living with MND is an everyday reality for many people, who while coping with the effects of various losses brought upon by MND they also strive to live a good life. According to Mattingly (2008) a good life can refer to “*possible worlds and possible selves worth striving for*” (p.95). Rather than focusing on loss, the focus could be on what makes people go on with their lives and construct a self and a life worth striving and living for. The main issue then becomes, how can one live with MND? Dudley Clendinen, an American journalist diagnosed with MND in his early 70s, discussed his desire to live a ‘good, short life’ (Clendinen, 2011); a life allowing him to maintain his notion of selfhood without being dominated by medical technologies. A good short life is about combining a *short* life with a *good* life. In other words, a life lived with an incurable disease that leads to paralysis and reduced life expectancy, and a life that one can still define as good and that is worth living.

Rhian and Gwyn engage in an active process of accepting MND, with the ultimate goal to construct a good life. They have created a present where life is good and imagine a future where this will continue to be so, despite the difficulties. These difficulties, such as the time it takes to do the morning routine, are there and acknowledged but do not play such a big role in their narrative. To do this, they mobilise certain practices in order to manage themselves and their lives. These practices of managing oneself are related but distinct to self-management as the term is commonly used in healthcare literature.

Self-management of chronic conditions often refers to the use of skills to monitor and manage disease (Schulman-Green et al., 2012; Willems, 2000). It has been defined as

The ability of the individual, in conjunction with family, community, and healthcare professionals, to manage symptoms, treatments, lifestyle changes, and psychosocial, cultural, and spiritual consequences of health conditions (Richard & Shea, 2011, p.261).

Self-management often involves the migration of technologies of diagnosis and care from the clinic to the home (Willems, 2010). Devices, such as a PEG feed for example, enable people with MND to take an active role in their care. Such technologies need to be used in a certain way towards specific goals. In the case of the PEG feed, the goal is adequate nutritional intake through a gastrostomy. This can have multiple medical benefits, including decreased risk for aspiration pneumonia, and adequate hydration. It also frees up time because eating can be a tiring and long process due to paralysis of the muscles necessary for chewing and swallowing. As Willems (2000) points out, self-management technologies “*aim to increase the autonomy of patients by defining the active role they should play in the course of their disease*” (p.27). The focus of self-management is on a disease and how to best monitor and address its symptoms, leading to adaptations in daily life (Willems, 2000).

While Rhian and Gwyn engage in various practices of self-management, these practices occur within a broader framework where their life and their ideas about how to live their life take precedence over managing the disease (Schulman-Green et al., 2012). As the following

paragraphs illustrate, they discuss, decide and perform practices that enable the construction of a life with which they are satisfied. Sometimes, in order to achieve this, the disease has to be managed, but sometimes the disease remains in the background, or is managed in unconventional ways that nonetheless make sense within the context of their life.

### **7.3 “We never lived in each other’s pockets before, so why now?”; Between needs and desires**

Towards the end of the interview Rhian appears to be uncomfortable and acts as if she wants to say something. She protracts head as if elongating neck and Gwyn starts scratching her neck. Then Rhian signals it’s not that what she wants. Gwyn then wipes her mouth. We resume conversation.

These laconic notes were written after the second meeting with Rhian and Gwyn. Rhian has been living with MND for several years, proudly saying that only Stephen Hawking (University of Cambridge professor of physics who has been living with MND for over 40 years) has had it for longer. Comparing themselves with other couples they know, Rhian and Gwyn attribute their long and happy marriage mainly to four reasons: they are not “naggers”; they each maintain their own interests and keep a little bit of individuality within the marriage; they take life as it comes, and finally they love each other. So, they have learnt to live with MND (and are still learning), but they do not live for MND or despite it. In the interview excerpt below, Gwyn describes a part their morning routine:

[MND is] too life changing, you see. So...everything...awkward. Cleaning the teeth in the morning is awkward. Before she could [do] it alright, now it’s very difficult, now we get to fight [Rhian laughs] to clean her teeth in the morning like, you know. I want to do

it for her, she wants to do it herself. If I do it for her, it's another step down the road, isn't it. So, I'm putting toothbrush, toothpaste in her hand and she can't get it up there, in her mouth, like, around [Rhian smiles] and of course she is fighting me because she wants to do it herself, and I am fighting her because "let's get on with it for Chrissake, we can't have all day on it". So, each step down is a step away from what it was before, isn't it. So, that's what she is fighting against, and I understand that....you don't want to...you want to do it yourself and [I] tell you, "you can't do it yourself"...you see you are getting further and further down the road. Rhian is totally dependent on me now, but she didn't want to be dependent on me. **Obviously** [emphasis by Gwyn]. And I wouldn't like to be dependent on her, like.

Gwyn is frustrated, or perhaps not really frustrated, but he wants to get the morning chores done so they can both get on with their day. He is eager to help his wife of 40 years with her morning routine, such as brushing her teeth. But if he does that, what will be left for Rhian to do? Care is often conceptualised as something that is given by a less ill to a more ill person; a process constructed between two or more people, or a person and a system; a burden; and a way of being for carers (Fine, 2007; Held, 2006). Irrespective of the approach or conceptual framework adopted, linked with care is a notion of *need*; somebody needs something, because of lack of resources or inability to use existing resources. For Kleinman and van der Geest (2009), one aspect of care is a process whereby "*one person completes another one*" (p.159). The main question then becomes: what can be done to best support people to engage in this inevitable process of care, where one will always give more than the other; more attention, more concern, more help.

Antonucci, Akiyama and Takahashi (2004) found that couples in later life engage in interaction in their daily life, helping each other to carry out activities, in a way of pooling resources, as has also been reported by others (for example, van Nes, Jonsson, Abma, & Deeg, 2013). Through their co-constructed practices of care, Gwyn and Rhian oscillate between what they need and what they want and between what Jackson (2012) referred to as ‘one and one another’. Gwyn and Rhian want to retain their individuality while negotiating how much one needs to give and the other needs to take, in the process of managing their life with MND.

Gwyn: When we first got started courting, didn't think it would work out because she is so quiet, so I thought "bugger", but it seems to work alright. I think that two people who likes all the same things it gets bloody boring you know. [We are] two people who like different things, like, she likes to read I like to go walking, she will sit down and read, I don't like [to sit down and read]. I go walking, or whatever. You can still live a separate life as such like. I go training five times a week, she has two hours on her own. She will sit here, read her paper.

Rhian: We never lived in each other's pockets before, so why now?

The excerpt ends with a question. This is not a question in search for an answer, but a rhetoric question, directed to an unseen interlocutor, society, challenging a discourse of care as burden and need. Rhian and Gwyn paint a more nuanced picture, where their everyday practices of care follow a pattern that predated the entrance of MND in their lives, constantly oscillating between what they need and what they desire. Both Rhian and Gwyn want to remain active agents in their lives, but as Jackson (2010) reminds us “*one can only be one's own person to the extent that one belongs to a wider context than the self*” (p.137). For Rhian and Gwyn, that context

is a relationship they have constructed over the years they have been together.

Gwyn: Sometimes she absolutely hated me...absolutely hated me. But I had to help her to get on the toilet, do things for her. It must have been very very difficult for her and to have to allow me to do that for her (...). It's hard for me to doing it for her, but it must be harder for her to allow me to do it for her. Do you understand what I mean? "I got let him hold me now, I will let him wipe me", you know, whatever it is, "and I don't want that to happen and I don't want him to do that now" (...) But you got to, like (...) a lot of people don't, can't cope with it. They put them in homes or whatever it is and, you know (...) bugger off, I got a life to live on my own, like. But we got a good life, Rhian and I, we live a good life. I do what I do, she does what she does. She likes to read her paper, she likes to watch telly. Still in charge of the house. I can't get her to bed before half past eleven, quarter to midnight [laughter, by Rhian and myself] (...). So, she is still in charge of the house, I got to do what I am told anyway [laughter]. Even though she can't talk, she is still bullying me all the time.

Rereading the transcripts of the interviews with Rhian and Gwyn, it was striking how they both maintained their individuality despite spending most of their time together. Privacy, Jackson argues (2012) "*should not be equated with individuality*" (p.11). Conversely, lack of privacy should not be equated with lack of individuality either. The use of joint interviews with Rhian and Gwyn foregrounded that individuality through the different ways they were approaching their shared experience of living with MND. The stories they were sharing together revealed a continuous oscillation between being actors and being acted upon (Jackson, 2002), illustrating one of the main characteristics of intersubjectivity, that of the constantly changing nature of human agency. People are in constant motion between positions where they have control over elements of their life and positions where they

have to accept loss of control. This negotiation involves experimentations, trying to find out what works.

Gwyn: Yes, different problems come up all the time, like. We work together, can we do this, can we do that like. I was putting her in the bath every Sunday but can't do this anymore, can't lift her up you know, so I wash her in bedroom like (...). I can't do that anymore, we'll try it this way. Sometimes it works, sometimes it doesn't. You change and adapt all the time. (...) and if I don't know what to do, Rhian might say "let's try it this way" like. We are trying this way, and "no, I can't do that". Like I, I got to lift her from this way, I can't do it from that way. I don't know why, but I can't.

Rhian and Gwyn enact their acceptance of MND into their life through specific actions than take them to places that they had not anticipated. They are "*together-in-the-world*" through "*acting-together-in-the-world*" (Honkasalo, 2009, p.58), and this is often expressed through practices of care. The following paragraph offers a critical discussion of current conceptualisations of care and their relevance to Rhian and Gwyn's life.

#### **7.4 On care**

In disability studies informed by the social model of disability, care has been associated with a role of people as passive recipients of care, which can perpetuate dependency and detract attention from the political dimension of disability (Kröger, 2009). It has been argued that care can signify oppression or patronising attitudes and has been associated with dependency, as Hughes, McKie, Hopkins and Watson (2005) discussed in an overview of the uses of 'care' in the disability movement and in feminist literature. Indeed, care practices have often been disempowering and controlling, constructing disabled people as *objects of care* (Beckett, 2007;

Watson et al., 2004) and locating them in the place of a passive receiver and as a burden (see for example, Awad & Lakshmi-Voruganti, 2008). In disability studies care is thus often seen as a practice of normalisation that contributes to the construction of a disabled rather than a desired self.

However, care does not need to be disempowering and construct disabled people as objects, as Rhian and Gwyn's story illustrates. Recent conceptualisations of care place more emphasis on the notion of interdependence over independence (Shakespeare, 2006; Struhkamp, Mol, & Swierstra, 2009). By challenging the notion of passivity and dependency, the emergence of interdependence has reconstituted disabled people as active participants in the process of care, leading to a critical examination of the concept of care in relation to disability beyond the binary of dependency/ independence (Kröger, 2009; Fine & Glendinning, 2005).

Dependency and independence are parts of the same continuum and people move between the two extremes. Letiche (2008) referred to care as being *in-between* or *being-two* since it cannot be defined by a single process, but it is always situation-bound. People need to make decisions not only regarding how much care they want to receive, but what kind of care and how that fits in with their life. These decisions are rarely straightforward and often involve a negotiation between which outcome is the best or most desired one (Struhkamp, Mol, & Swierstra, 2009). Rhian does not want Gwyn to brush her teeth for her although it would save them both energy and time. Brushing her teeth however is one of the few self-care

activities Rhian can perform and she is intent to keep doing it, despite the effort.

People, whether disabled or not, often need to care for themselves, for their own body, or manage their care. They are free<sup>4</sup> to choose those practices of care that are suitable for them; those practices of care that will lead to the construction of a self they envisage for themselves. As Rhian stated, “*I know best*”. She knows best what she needs and what works for her. Rhian wants assistance for a *lived* body, not for a body-as-a-tool. In effect, she wants to be able to engage in a process of caring for herself where she will be able to make the decisions that are right for her. In a way, she wants to be able to manage how her body will be cared for, assuming control of the process.

Rhian and Gwyn’s narrative is a collaborative one; rather than Rhian feeling dependent upon Gwyn, they work together as a team, towards shared outcomes (Radcliffe, Lowton, & Morgan, 2013). This does not mean that they necessarily always agree, but that they collaborate. The following section discusses in more detail Rhian and Gwyn’s acting-together-in-the-world (often through caring), in order to construct a good life.

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<sup>4</sup> The notion of freedom as used here needs to be qualified. People can choose practices of care that are suitable for them, but these choices happen within specific discourses of care, disability, and knowledge legitimation. Due to power and knowledge asymmetry, people often think that professionals know best and thus they sometimes delegate (or are expected to delegate) these choices to professional expertise. This is further discussed in chapter 8.

### **7.5 Gastrostomy and hairclips; Trying out possibilities**

The lack of biomarkers of MND complicates the process of surveillance, as does the lack of accurate prognostic factors. Motor neurone disease cannot yet be isolated and observed, although the development of more refined neuroimaging techniques may change this in the future (Kiernan et al., 2011). This inability to observe the disease per se does not mean that MND is not subjected to the gaze of scientific knowledge. While its actual presence in the body cannot be located and observed, its effects on the body appear in many ways and thus the focus of scientific knowledge shifts from isolation of the disease towards its manifestations, and ways to manage them. Gastrostomy, for example, is a technology that impacts certain changes on the people who use it. By creating an alternative entrance into the stomach, bypassing the mouth, it enables continued nutrition.

Foucault's (1994a) 'care of the self' is an important theoretical concept used to understand how people make sense of their life and how they look after themselves, taking whatever decisions are necessary so that they can construct a life with which they are satisfied. People subject themselves to certain technologies in order to produce a desired version of themselves (Frank & Jones, 2003). This can mean a self that conforms to certain social, ethical or political standards, a body that carries out desired or expected activities or a series of other practices, or technologies, pertaining to the care of the self. Technologies of the self

Permit individuals to effect (...) a certain number of operations on their own bodies and souls, thoughts, conduct, and way of being. So as to transform themselves in

order to attain a certain state of happiness, purity (...) perfection (Foucault, 1994a, p.225).

People can choose from an array of technologies of the self, which are possible within a certain system of discourses. These choices are essentially experiments with games of truth (Foucault, 1994b), in the sense that they reflect perceptions on what is considered as a valid (desired) or not valid (not desired) outcome. These decisions involve constant negotiations between what people need and what they want. Through engaging in certain technologies, people can actively work towards the construction of a desired version of themselves and in the process think of themselves as active agents (Frank & Jones, 2003). Of course, different people have different ideas about which technology is a good one, and why. As a case in point, a systematic review suggested that people living with MND sometimes choose to not have gastrostomy to facilitate nutritional intake even though the procedure could prolong their life (Katzberg & Benatar, 2011).

To care for one self requires close attention to the kind of self one needs and wants to care for. In the following excerpt, Rhian and Gwyn discuss about the time Rhian had a PEG inserted. Healthcare professionals often recommend that the PEG procedure is performed early on in the disease process before the respiratory function is compromised (Talbot et al., 2010). PEG was introduced into clinical practice around 1980 (Kurien, McAlindon, Westaby, & Sanders, 2010) and guidelines for its use in MND were published in 1999 and again in 2009 by the American Academy of Neurology (Miller et al., 1999; 2009), and in 2007 by the European ALS consortium (Andersen et al., 2007). It is now fairly common practice in

MND, although this does not mean that all people with MND view it in a positive way.

Gwyn: No, Rhian didn't want it, no.

Dikaios: Why not?

Gwyn: Because it is another step down the road, you see. Each step you lose you going further down under (...) [Rhian starts synthesising answer]. So having the PEG was another step down the road, She didn't want that, she wanted to stay as she was. I felt that she didn't need it, I said "don't do it, she don't need it. She is okay as she is, right". He says [the doctor], he insisted that she should have it. Because when she was in the hospital for a couple of days, she didn't have anything to eat, because it wasn't mashed or anything, and of course they weren't feeding her either (...) So the doctor said "well she will starve to death, you got to have a PEG" like. For she blamed me for having had the PEG put in but it wasn't my fault, I didn't want it in the first place. But best thing that ever happened to her.

Rhian: I felt it was too life changing.

Rhian and Gwyn are taking care of themselves in small or bigger ways, and they continuously modify their perceptions of what needs to be done in order to construct a good life. They choose some ways to live, engaging in what Mol, Moser and Pols (2010) described as *tinkering*, or exercising control when and where they can in order to experiment with what is possible and what is desirable. In other words, in choosing different technologies of the self, they engage in truth games about who they are and who they want to be. A PEG, with its "*fusion of technology with the organic*" (Manderson, 2011, p.58), can act to highlight "*the departure of the individual from normatively able bodies*" (Manderson, 2011, p.86). PEG is an aberration; bodies do not have holes in the abdomen, with tubes coming out of them (Appendix A, Figure A.1). A PEG signifies a radical change of what the bodies are supposed to look like. As the excerpt above illustrates

however, these ideas are not stable but change over time. While initially against the idea of having a PEG, Rhian's perception of it eventually changed.

Rhian recently had a new PEG tube inserted, using a less invasive technique than the one she went through several years earlier. While this was initially seen as a positive development, Rhian and Gwyn were not satisfied with the end connector of the outside lead of the PEG, which was soon found to be leaking. As this was made with a hard plastic component placed inside a softer tube, any pressure change caused, for example by gas, would cause the inside part to be dislocated and food to drip out. When they reported this problem to the nurse, she said nobody else has complained and the tube is fine. But as Gwyn said:

I am the one who has to clean up the mess, not her. I am the one who lies down thinking, "what's happening, will I find a mess in the morning?", and Rhian getting all soiled. And it smells too.

Rhian agreed: "*it's smelly, glue-like*". Instead of waiting for a solution that was not forthcoming, they decided to experiment.

Gwyn: Look at that... look at this [showing the PEG tube]. Come on [I move closer]. You see that flap there was on top of there, but it was leaking all the time. Not now, we taped it up, put a hairclip on it, tied it on there with the cord. Now I can put her to bed and put the feed on and it won't fall out. Before it was falling out...the feed was still going, it messed on the floor, all over Rhian, everywhere like. When the nurse saw that she said "that's the only one we got"...well, it's rubbish, absolutely crap...they couldn't change it. So I had to adapt that now. So, the idea of this [hair]clip is hers [Rhian's], and the idea of the cord is mine...[both laughing]. It can't come out, you see. It works. The other one it doesn't work (...). And you tell these people, "it's not working, it's making a mess everywhere" like. And they come and say "that's the only one we got, we can't do

anything else” like. But I couldn’t go to sleep in the night because I was afraid it was falling out all the time (...). Now, it’s working perfectly fine, we got no problem at all.

“*We got no problem at all*” means that they found a solution that worked for them and there was no more food dripping out of the tube. They enacted what Pols (2011) framed as ‘patient knowledge’. This knowledge is developed through their everyday aesthetic experience of living with MND. For Gwyn and Rhian, food dripping was *bad* because it smelled bad, it was hard to clean from the carpet and soiled Rhian’s clothes in the middle of the night. So, the solution was to find a way to stop food from dripping out of the tube so they could both have an uninterrupted night’s sleep, without worrying about potential mess in the morning. Uninterrupted sleep was the good they were trying to achieve.

Many strategies for managing some of the impairments of the disease have been developed, several of them based on high technology, such as gaze control systems for computers, text to speech software and hardware, ventilation machines, and power wheelchairs. These technologies are effected not upon a disease but upon a life. While some of the more biomedical technologies may be recommended as a way to address specific symptoms of the disease (for example, PEG to address dysphagia), technologies of the self are ultimately being chosen, implemented and evaluated in the context of a person’s life (King, Duke, & O’Connor, 2009; Murphy, 2004; Sundling et al., 2009). Sometimes, improvised solutions developed in local contexts are what people need. In making these choices and engaging in these technologies of the self that will facilitate the

construction of a desired life, sometimes Rhian and Gwyn made their own experimentations to fill the gaps that could not be filled by established professional knowledge and practices alone.

### **7.6 Reflections on interviewing people who cannot talk**

When I started this study, I was aware that some of the potential participants would experience speech impairment, or they might not be able to communicate verbally. Wanting to enable the participation of all potential participants, I stated in the information sheet that interviews could be performed and captured in various formats. Carrying out joint interviews was one of the ways that people were enabled to participate. In this paragraph, I reflect on carrying out interviews where a participant cannot talk.

Rhian used a lightwriter machine to communicate. This involved typing into the machine words or phrases, and then these were voiced by the machine. It was a slow process, requiring a few seconds for a single word and sometimes over a minute for a sentence. Time was not an issue and I was happy to wait. In fact, it was a rare opportunity to have some time to reflect during the interview and take time to think.

However, there was another issue I had to learn to adjust to and that was silence. I was not prepared for numerous short, or not so short, silences during interviews. These were not nervous silences, full with meanings and innuendos; they were functional silences, required so that Rhian could participate in the interview. Had it just been Rhian and myself participating in the interviews perhaps I would not have found the silences as hard to

manage. The presence of Gwyn however complicated things, for me. In the first couple of interviews I was torn between what I thought were conflicting requirements; one was to maintain silence waiting for Rhian's response and the other was to engage in conversation with Gwyn. Soon I realised that what I was trying to do was to impose a tempo in Rhian and Gwyn's interactions. As soon as I relaxed, stopped trying to control the situation and let the interview take the natural rhythm of Rhian and Gwyn's daily interactions, silence stopped being a problem. Rhian was happy to interject, through a move of the hand or some sound, if she wanted us to stop talking and listen to her, and Gwyn was aware when Rhian had stopped typing into the lightwriter.

## **7.7 Chapter summary**

Rhian and Gwyn engage in various practices in order to construct a life they are satisfied with, a good life. Rather than adjusting their life to accommodate the disease and its symptoms, they decided to enact practices of care and mobilise technologies of the self that would enable them to construct what they called "*a good life*". Concepts, for instance care, and practices such as using the PEG, were continuously being tinkered until they could be accommodated into their lives.

This tinkering requires an understanding of the idiosyncratic regimes and practices that can facilitate the production of a tolerable present or an ideal future, in combination with biomedical practices. This tinkering is the result of a constant negotiation between what is medically needed and what people desire in order for them to live a good life that *includes* disability and

continued illness (Mol, 2006b). In other words, a negotiation between their specific, experiential knowledge, and the more standardised and generalisable medical knowledge that can present facts and offer possibilities that guide choices (Berg, Meulen, & van den Burg, 2001). Rhian and Gwyn trust their knowledge and keep on experimenting until they construct practices of care that work in the context of their life. As Gwyn said: “*So plod along, as best as you can like. As long as you can do it, you can do it*”. Rather than feeling dominated by MND, Rhian and Gwyn take decisions, carry out everyday activities, hope, despair, love, feel angry and feel happy, amongst many other dimensions of their life.

## 8. “It is frustrating”; Trying to be a good recipient of services

Dave and Marion have been married for more than 30 years and they live in an urban area in south Wales. When I met them they were in their late fifties and Dave had recently retired from his job as an electrician, on health grounds. At the time of the first interview, Dave could still walk short distances and drive but he soon had to start using a wheelchair both indoors and outdoors and he stopped driving. His hands and arms also became progressively weaker so within a few months after he had enrolled in the study he only had some limited movement in his right arm. He could use the movement to turn the pages of the newspaper or move the arm across his body, but that movement was not fine enough to allow him to use the TV remote control and his arm was not strong enough to support him during transfers.

Being relatively recently diagnosed, Dave and Marion were trying to find their way around the health and social care systems. Their view appeared to be that however flawed it may be, there is a system and the system will sort things out. Dave and Marion felt that the decision making power was held by the system, not by them, and they were seeking and following the recommendations of various professionals. They were “*chasing, chasing, chasing*” as Marion pointed out during our interviews; chasing advice, chasing people, chasing time. Also, they were being chased by the disease, which was progressing at a faster pace than they could adapt to. The resulting angst permeated all our conversations. Drawing from literature on biopolitics (Foucault 1994a; 1994b; 2010), Bakhtin’s (1995a;

1995b) discussion of heteroglossia, and theory on knowledge legitimation (Lyotard, 1984), in this chapter I discuss how while trying to live with MND and navigate the health and social care systems, Dave and Marion became disempowered and felt they were losing control over the disease and their lives.

### **8.1 “Gets you a bit frustrated...what’s happening...”; Being a good patient**

An avid driver, Dave first noted that something was not quite right during driving. There were times when the pedals felt tighter than usual, or changing gears was difficult. Eventually, Dave was diagnosed with MND after having experienced symptoms for two years. Diagnostic delays are common in MND (Kraemer, Buerger, & Belit, 2010; O’Brien, Whitehead, Jack, & Mitchell, 2011; Shook & Piro, 2009; Zoccolella et al., 2006). Misreferrals, lack of knowledge about MND by professionals, and non-specificity of symptoms can lead to a long diagnostic process (O’Brien, Whitehead, Jack, & Mitchell, 2011; Pavey, Allen-Collinson, & Pavey, 2013), which can be experienced as “*highly stressful, distressing and profoundly upsetting*” (Pavey, Allen-Collinson, & Pavey, 2013, no page number).

The general practitioner (GP) first thought it was arthritis as Dave’s mother had it, but when tests were negative he was referred to a neurologist. The appointment took a long time to be arranged and because the clinical signs were not conclusive, Dave had to be re-examined four months later. Dave and Marion believe that the neurologist knew what the diagnosis was,

but he was waiting for the results of some tests in order to completely rule out other possible diagnoses. At that stage they were already experiencing problems that affected their daily life (Dave had developed a drop foot due to muscle weakness), and they were waiting for guidance from the professionals in order to take action.

This early encounter with representatives of the healthcare system set the tone for their story of living with MND. Dave and Marion tried to be good patients by adjusting their life to accommodate MND. For them, being a good patient meant seeking and following advice by health and social care professionals without questioning it (Jadad, Rizo, & Enkin, 2003) and adjusting their life to accommodate that advice. It was also about seeing possibilities for solutions that might work in the context of one's life. When I first met them, Dave and Marion were in the middle of major adaptations being carried out on their semi-detached house. They were also waiting for delivery of an adapted car that would accommodate a power wheelchair and they were considering putting a care package (Appendix A) in place. At the time of the first interview Dave could still walk short distances but he could not climb stairs. In the following excerpt, Dave and Marion discuss about the effect not having a stairlift has had on their life.

Marion:...It's very difficult. It is stopping Dave, I think, having what he needs, you know, now. I mean, at the moment he is still getting up out of the chair okay, but they have given us these risers...

Dikaios: How you are coping at the moment then...?

Dave: With the stairs? I don't go upstairs at all. We've moved the bed down in the dining room and I sleep, well, we both sleep, in there.

Marion: We sleep downstairs, yes. Cause he was trying, with one stick, holding on to the stairs and then one stick.

His good leg is the right leg and he was getting up the stairs, but then he was getting more and more tired...and...

Dave: ...One point is the amount I can lift this leg. I can sort of lift like curbs, but I can't lift the height of the step...so, that's stopped me from going upstairs.

Marion: ...It's a shame, I mean, I mean if they could put in a lift or a stairlift temporarily, that would have been the answer (...). And I think it's just all the delays really, in getting everybody together, making a decision.

Dave: Gets you a bit frustrated...what's happening...

Dikaos: There are many people involved, aren't there?

Dave and Marion: Yes.

Dave: You know, at least if you ask the specialist nurse for something it happens very quickly, you know. But when it goes to the council, that's what makes it slow.

Marion: But he had to refer Dave again, because the first time...

Dave: The first occupational therapist shut the case...

Marion: She shut the case...apparently they do that (...).

Dave: It's the coordination, I think.

Dave and Marion were frustrated that the services were appearing to be slow to respond to their needs, and they did not have a complete understanding of why that was happening. Their frustration with the processes followed by the health and social care systems was even more evident later on during the same interview. While talking about the delays in getting a specially adapted car that would accommodate Dave's power wheelchair they expressed how that made them feel.

Marion said: It's strange. All these things mean chasing, chasing, chasing (...).

Dave:...It's annoying...

Marion: It is frustrating, yes.

The help Dave and Marion wanted from the health and social care services was either too slow to materialise, or required too much effort on their part. While they were trying to be good patients, by being clear about their needs and by following advice for example, they were frustrated by the delays in

getting things done through the health and social care systems. Being at the early stages of living with MND, they needed guidance on how to use these services. On a number of occasions they both stated that they would not have minded paying for some services or equipment themselves but they would have needed someone to tell them what is needed.

Marion: If we had known before how much stress this would have been I think we would have tried to do it ourselves, you know, all of it. Because we could have done it a lot quicker. It would have been a lot more money, but you know, when you are not waiting then, all the stress really from everything, you know, it would have been quicker, wouldn't it (...).

These delays resulted in major disruptions in their daily life, such as the couple moving their bedroom into the downstairs dining room. Dave and Marion found themselves in a situation where they did not feel knowledgeable enough to make decisions about their life with MND and act upon them. At the same time however, they were increasingly realising that the professionals upon whom they depended could not respond to their needs in a satisfactory way. The view that the professionals know best seemed to pervade their story, although the professionals did not, and perhaps could not, know what practices of care Dave and Marion wanted to enact in their daily life. Even when they doubted the usefulness of what was happening to them (for example, with regard to the adaptations), they still felt obliged in some way to go along with what the professionals were saying, yet still being aware that the professionals did not have the answers or solutions they were looking for. The following paragraphs illustrate the various ways Dave and Marion became disempowered while trying to be

good patients.

## 8.2 Illness as a heteroglossic world

When Dave was initially diagnosed, the healthcare professionals recommended installing a stairlift. However, and before any action was taken, they reconsidered and felt he may not be able to use a stairlift as the MND progresses. Consequently, a recommendation for a through-floor lift was made and during most of the data collection, Dave and Marion were liaising about the lift with the city council and relevant professionals, including an occupational therapist, a physiotherapist, a surveyor, and a contractor.

Marion: Well, the surveyor came to do the drawings and he has to do them on the computer. He said he would submit that to the council. I said, "can you order the lift now?", he said "no", they have to have an official order from the council for the lift.

Dave: If they get the official order they start the manufacture of the lift, but they will also then be coming into the house for a site meeting with the builder they are using to do the holes. Probably when it comes to that they will give us a definite day when to come in and do it.

Marion: Because we did ask...because we have to do the bathroom ourselves, so we did ask if we could start the work there, and the occupational therapist said, "no you'd better wait until the lift is approved first", because it affects where the bathroom door will be and everything in that way. So we thought we could save a bit of time. Once they have this meeting we will ask them if we can get our own contractor then...at least to order things. Because we might have to wait for him as well to do everything. But before, when we had the other contractor, the one who's gone into liquidation (...) we can't use him now so we got to find somebody else to do it. The other problem that we have is that the project officer went on holiday...everything...he was on holiday in July and then he went off again. I think he is back this week.

Trying to deal with all these different professionals took up a lot of effort and time from both Dave and Marion. As Dave's facial and respiratory muscles and his tongue got weaker, Marion had to do most of the talking on the phone and chasing up different services, explaining their needs. Their experiences point to the heteroglossic nature of illness and of the health and social care systems constructed around illness. Heteroglossia refers to the presence of "*another's speech in another's language*" (Bakhtin, 1995a). In the case of MND, the presence of multiple perspectives constitute a heteroglossic world comprising the voices of individuals living with the illness and the voices of the health and social care providers, but also those of family and friends among others. These multiple discourses are interconnected and are grounded in the diverse cultural discourses operative in every society. The different ways people talk and thereby represent the world around them, express their realities and the way they live in the world. In an analogy with the textual construction of a novel where heroes are situated in interactions initiated by the author and act within preset boundaries, social actors operate within a set of cultural discourses.

The way people make sense of their life with MND influences their needs, their choice of services, and the way they negotiate their illness. Individuals, their families and friends, and health and social care professionals have their own ideas of how life is to be lived, what needs to be done, how, and why. They create what Kleinman (1988) called the different personal and interpersonal meanings of the disease and of life with it. These different perspectives are in constant negotiation in the context of

daily life with everybody involved in a dynamic dialogue, trying to establish a common ground for understanding what is *good* and what has to be avoided (Letiche, 2008). The process of healthcare is not about always curing and restoring. After all, there might not be any cure available. The process of healthcare is about understanding and negotiating the positions of everybody involved. This understanding is achieved little by little, through synthesising the different voices and making sense of the heteroglossic world of illness (Good, 1994).

In the context of MND, multiple discourses exist: official and vernacular, explicit and implicit, scientific and narrative and so forth. People experience living with MND in different ways. Findings from Hogden, Greenfield, Nugus and Kiernan's (2012) study suggest that people living with MND often focus on living in the present and make decisions that have a direct impact on their daily life, whereas health professionals focus on the future. The frustration experienced by Dave and Marion regarding the delays having a stairlift or through-floor lift installed could in part be caused by these different priorities. At some point during our conversations, Marion said "*at least they could install a temporary stairlift or something*", expressing their need to get something done. The excerpt below illustrates how an activity that many people take for granted in this case becomes a challenge.

Marion: We are still waiting for the shower to be put in.

That will have a huge difference really.

Dave: It will make a big difference.

Marion: Dave will be able to sit down and be able to wash himself then, rather than me...doing it between us. But it's very hard, you know....

Dave: In the kitchen.

Marion: And it's coming up to the winter now, so I'm hoping...

Dave: ...Hopefully it will be done then...it will be warmer.

Dave had to strip wash in the kitchen for several months while the major adaptations application was being processed. Although he was still able to stand and could also use his hands, Dave could not walk up and down the stairs and thus could not access the bathroom, which was upstairs. Dave and Marion had to enact a practice of care, strip washing in the kitchen, which was not ideal for either of them.

Other areas of their daily life were addressed more expediently by health and social care systems. For example, Dave was referred to a gastroenterologist for a PEG evaluation before any signs of dysphagia developed. He was also referred to a pulmonologist as soon as the first signs of compromised respiratory function were present. While they both commented on the efficiency of services to deal with these life-threatening symptoms, they were wondering why other areas affected by MND, such as using the shower, were not dealt with the same efficiency, especially since time is a very precious commodity for people with MND and their families. Arguably this could be due to the structural differences in the way in which acute and community healthcare services have developed, but delays in services such as the installation of adaptations can threaten possibilities for participation in daily life (Johansson, Josephsson, & Lilja, 2009). As Marion pointed out: *“To do it later, it's too late sometimes”*. Unfortunately, this turned out to be true, as the through-floor lift and the accessible bathroom

were completed just weeks before Dave died.

Literature (for example, Aujoulat, Luminet, & Deccache, 2007) shows that limited access to the physical environment can lead to a feeling of loss of control. When people are no longer able to access and use all areas of their house this can be frustrating and disempowering, and it limits the possibilities people have for action (Johansson, Josephsson, & Lilja, 2009). Dave could not access the first floor of the house where the bedroom and the bathroom were and this had caused a major disruption in his and Marion's life. The lack of access to space was further complicated by a loss of control over time. A recurring theme in our interviews was the delay in getting major adaptations done in their house. Month after month there would be another unexpected delay, and appointments with the social services and the contractors would take a long time to arrange. Being chased by time in a very pragmatic way, they were chasing up in turn the services responsible for these adaptations. What they did not know however, was that it was futile for them to do so as the average duration of major adaptations in Wales, from application to completion, was 386 days in 2012 (National Assembly for Wales, 2012). While these data are available through the National Assembly for Wales, this timeframe was an unknown factor within their story. With an average prognosis of three to five years survival after diagnosis, MND is not a condition that gives people the luxury of time.

The reality that Dave had an incurable condition, that the timing of death was not only unknown but unknowable and the fact that the

professionals knew that the adaptations would take a long time could be unsettling for all involved. With so many different people involved in the process of the house adaptations, it was getting more and more difficult to differentiate between their own needs and the professionals' decisions. Aujoulat, Marcolongo, Bonadiman and Deccache (2008) describe empowerment in the context of living with an illness as a process of 'holding on' and 'letting go', requiring the integration of

Different and sometimes conflicting aspects of one's self in order to develop a renewed and valuable sense of self, by differentiating one's self from illness on the one hand, and by integrating illness and illness-driven boundaries as being part of a reconciled self on the other hand (p.1236).

However, what should one hold on to and what should one let go off, are not easy decisions, especially when there are different priorities and different varieties of knowledge to be considered. Differentiating between one's self and the illness can be very difficult, especially at the initial stages of MND, when people often find they need to do major changes in their life. The following paragraph discusses how these different varieties of knowledge were experienced by Dave and Marion.

### **8.3 Who knows best; Knowledge legitimization**

The previous paragraph illustrated how Dave and Marion had to negotiate different priorities and positions in their daily experience of living with MND. This negotiation was even more challenging when they felt that their wishes on how they wanted to live with MND were sidelined in favour of decisions taken based on professional knowledge. This was especially noted

in the numerous discussions we had regarding their sleeping arrangements. At the initial stages of the negotiations regarding the house adaptations, before the decision to apply for the through-floor lift was taken, a healthcare professional suggested converting their front room downstairs into a toilet and a bedroom. Marion was concerned they would not be able to fit a double bed or two single beds in there.

Marion: And they [health and social care professionals] say, “well, Dave can sleep there because he would probably need a special bed quite soon”, and then I said “well, I would need a single bed then...Oh, you will have to sleep somewhere else” and I said “no, that’s not possible”, because...

Dave: ... We want to stay together...

Marion: We want to stay together, but apart from that he does sometimes need me in the night, so...it’s this sort of adaptability...I can’t understand.

They went on to explain that Dave sometimes has problems breathing, because his diaphragm is weakened. Later on, when considering applying for a through-floor lift, the occupational therapist initially was reluctant because of restricted space to turn the wheelchair upon exit from the lift on the first floor.

Marion: We’ve thought of ways of making it bigger from the other rooms, so Dave can turn the wheelchair. So, we just meet hopefully with the engineer and the surveyor from the council, whether they can talk together...

Dave: All they are whining about is six inches, which is nothing really. The engineer can find a way around it, you know. So...

Establishing communication in health and social care calls for a reconceptualisation of what counts as valid knowledge in the context of illness and disability. Lyotard (1984) framed the issue of knowledge

legitimation in epistemological terms, discussing the mechanisms involved in the process of deciding what constitutes knowledge and what does not. Different kinds of knowledge employ different criteria to establish legitimacy. For example, the scientific knowledge about the impact of a disease that a healthcare professional has mastered and the narrative knowledge developed through experience of the lived body that people living with illness have, are not mutually exclusive and neither are they opposing forms of knowledge (Kleinman & Hanna, 2008). They are both valid and important in a person's life. Dave and Marion felt that they had to constantly defend their opinions and wishes against those of the professionals with whom they were liaising. The following excerpt illustrates how they sometimes favoured the scientific knowledge over their own wishes about how to live their life.

Marion: But we find, with this as well, the more equipment you have, you know, it's...

Dave: It's cluttering the house up...you know, because it's not a big house (...). All I am hoping is that all the work in the house and everything gets done before Christmas....otherwise it won't be a very good Christmas.

Marion: Cause we find too when the family comes, my daughter and son-in-law (...) they have two children. Well, we don't have enough room now, because there are so many other things. So they stayed and the children stayed with someone else. It's sort of stopping the family staying together. Cause [we are] using the other room as a bedroom, they come in here basically [living room] (...). My daughter is wondering how I'm going to cook Christmas dinner [laughter].

Dave and Marion were quite clear about not finding the various pieces of equipment helpful. Not only were they not helpful, but also they took up so much space that there was no space left for their grandchildren to stay over.

And yet they kept all that equipment. However, this was not necessarily as much of a free option as it might appear. Their choice to keep the equipment, even though they were not using it, could be explained by the perceived higher legitimacy of scientific knowledge as expressed through the recommendations made by professionals over their own experiential knowledge.

In his discussion on the acquisition of knowledge, Latour (2007) remarked that knowledge is not produced solely by an external subject, but also by and through interaction with the object of the inquiry, meaning that no single voice or way of knowing is an authoritative one in its own right. For Marion and Dave, this means that the knowledge they had developed through living with MND was as valuable as the knowledge the various professionals have about managing the symptoms of MND, or the knowledge that the various contracted builders had about the right way to carry out alterations in their property. However, Marion and Dave did not think so. Considering knowledge as authoritative can lead to problems having a direct impact on people, as the following excerpt illustrates.

Marion: The one thing with the contractors when they knocked the two rooms into one they made a new doorframe, but uhm, they weren't very good at all, they didn't have sort of, a level, to level it out. It wasn't square. So, when another man came then to fit the door, he couldn't fit the door because the frame wasn't square, and they had already plastered everything as well, so he had to redo it as best as he could, but...very, very poor really, because they don't employ skilled people. That, that was a bit of a stress, stressful (...). We did ask if we could have a friend fit the door ourselves, and they said "no, you can't". If the council are contracted to do something they got to use their own people, which was a shame.

When social services do major adaptations on private properties, the property owners often receive a grant and are responsible to choose and pay contractors (Welsh Government, 2009). It was not clear what arrangements were put in place for the adaptations at Dave and Marion's house. In their story, their knowledge was not acknowledged and respected, which led to a feeling of disempowerment. The following paragraph discusses this interplay between knowledge and power.

#### **8.4 Power/knowledge**

The antithesis to the technologies of the self discussed in chapter 7 are the technologies of power that governed Dave and Marion's daily life. These technologies "*determine the conduct of individuals and submit them to certain ends or domination, an objectivation of the subject*" (Foucault, 1994a, p.225). It is not an either or choice between technologies of self and of power. They are both inescapable. The issue is how they relate to each other and which dominate. The dialogical relationship between the two determines how people's lives are governed (Foucault, 1994c).

Dave and Marion evidently made some choices and engaged in various practices of care. They actively sought solutions that worked in the context of their life. They decided, for example, to do adaptations in their house, ordered a new car and accepted help from paid carers. They tried to create possibilities for action, despite the difficult circumstances they were facing; the disease was progressing quickly, and services were slow to respond to their needs.

When things did not go as anticipated, like when the major adaptations were delayed (delayed both from Dave and Marion's perspective of limited knowledge about the procedure and in respect to the couple's effort to live in the present, in the knowledge that time was limited), they could not devise a different course of action. While they were trying to find out other solutions that worked for them, they did so on the terms of the health and social care system. Dave and Marion were lost in what was seen as a maze made up by procedures introduced by the representatives of health and social care systems. Rather than making choices based on their own perceptions of what is good and desired, they followed professional advice, not being "*capable of choosing which truth games and technologies to be subjected to*" (Frank & Jones, 2003, p.184) and thus delegating the responsibility for the care of the self to professionals.

Jackson's (2013) reflections on migrant imaginaries, and the difficulties encountered by people, who out of choice or not, have to learn to navigate new environments are relevant here:

Perhaps the worst fate that can befall any human being is to be stripped of the power to play any part in deciding the course of his or her life, to be rendered passive before impersonal forces he or she cannot comprehend and with which he or she cannot negotiate (Jackson, 2013, p.149).

Even though Dave and Marion had not changed physical environments, and still inhabited the country they were born in, they were recent migrants in illness, just learning to live with it. They had to learn to navigate a new environment, one where disability and illness were not only the focal point,

but also the domain of professional power. Within that environment they felt they had limited power to make their own decisions.

If power for Foucault (1994c) is diffused in ideologies and practices that govern daily life, then where should one look to find it? How can power be located and how can it be shared out? Although not explicitly stated, this was a source of angst for Dave and Marion. They wanted the power to act and do what they thought was best but did not know how to get that power. So, instead they decided to “*powerfully participate in the discourse that defines them as weak*” (Holstein & Gubrium, 2005. p. 491). They were relinquishing the disease and its management on to professionals, passing it on to the people who they thought knew how to deal with it.

Dave and Marion were perhaps looking for power in the wrong place. They were looking for it in institutions and systems such as hospitals and social services and their representatives. These are of course all locations where power operates. However, that power is not transferable but it is inextricably linked to systems of knowledge, and more particularly to systems of codified knowledge, such as those embodied by the various professionals Dave and Marion came in contact with. It is more accurate to talk about power/knowledge, as the two are always connected; one operates through the other (Foucault, 1994c). By entrusting all hopes to professional knowledge, Dave and Marion were objectified by that very knowledge and were dominated by its power.

Knowledge is not always a means of domination and neither does possession of knowledge necessarily lead to power. The two are dynamic

and inherently contextual. Knowledge and power are neither bad or good. Although power might be located anywhere, it is constructed through knowledge that is perceived as legitimate and hence *true*. In turn, access to this knowledge gives access to power. The knowledge possessed by professionals is generally perceived as valid and true and professionals have the power to make recommendations, suggestions, and decisions that can have a direct impact on how people live their life. Dave and Marion could not get the adaptations they wanted unless a professional approved them. Through the system of power and knowledge that they represented, professionals could decide whether Dave could once again take showers or keep on strip washing in the kitchen.

Power/knowledge, however, is not absolute, but relational and dynamic. Brott, Hocking and Paddy (2007) reported that some of the participants in their study felt a sense of control in rejecting equipment recommended by professionals. Dave and Marion did not have to believe that the professionals' knowledge was better than their own. They could have resisted the domination of professionals over their life, for example, by rejecting the portable hoist which was rarely used and instead cluttered up their living room or by deciding to move to a different house in order to avoid a long wait for adaptations. However, they lacked the knowledge to make this and other decisions. Although they were "*chasing, chasing, chasing*" they were doing so in a maze, without a compass and without a map. They knew that the prognosis was not very good, but how much time they had left was unknowable. They knew that MND is a serious disease,

but they did not know what symptoms to expect and what the end would look like. They knew that health and social services could provide for their needs, but they did not know the processes and time frames involved.

Dave and Marion appeared to be caught between what Mol (2008) described a 'logic of care' and a 'logic of choice'. The logic of care focuses on finding the best solutions to suit individual people, accepting that they might need to live with ongoing frailty. In contrast, in the logic of choice people are turned into what Mold (2011) called patient-consumers and the emphasis shifts "*to the rights of individuals within increasingly marketized services*" (p.509). People are expected to make the choices that are right for them and assume responsibility for the outcomes of their choices. Often, however, people want someone to care for them and construct these choices collaboratively, either because they are too preoccupied with the process of adapting to an everyday life complicated by the effects of illness, or because they do not feel competent enough to make these choices (Mol, 2008).

The feeling of disempowerment was particularly pronounced during two particular stages of their life with MND. The first instance was when they decided to use paid carers. Initially, their adult children helped out, but:

Dave: They couldn't come in the morning you see, only in the evenings to put me in bed. They are all in work.

Marion: They still come round but we decided to have the carers instead. They [the children] were a lot better, Dikaios. They were able to hold him up. I found sometimes difficult to lift his legs alone all the time, but they would always lift him..

Dave: When they were doing that I could lift myself a bit. That's when I decided for the carers, I was finding it a bit difficult.

However, having carers helping out was not as easy as they had anticipated.

Marion: It's been a bit difficult because it is different people coming a lot (...) but Dave, did fall across the bed, the carer didn't hold him...what happened was, you weren't in the middle of the bed when they helped him up and he slid across, and both of them didn't know what to do. I helped get, you know, get him back on the bed lying down, and then we both helped him get up (...). Sometimes we felt that they don't look at the care package [Appendix A] (...). Another time they put him into bed but they didn't allow sort of [how] high the bed was...

Dave: ...Hit my head on top of the bed boards.

Marion: ...And, uhm, they keep asking Dave to lift his feet or his arms (...) so, I don't find that very good really on that level.

Dave: There was that girl who thought I had a stroke.

Marion: Because Dave fell on the bed that one time the care agency insisted he had a hoist, you know, a full hoist. But he doesn't really need that. We got it here, just in case.

Not only were there many different carers, but they also had varying degrees of understanding MND and Dave's needs. However, due to the nature of service delivery, Marion and Dave experienced a lack of control over their care package, even when they were not satisfied with the services they were receiving. This situation lasted for a couple of months until Dave was admitted to the emergency room when he experienced breathing difficulties. He died from respiratory failure within three days of admission.

Marion recognised that talking about end-of-life care is not easy and can be upsetting, but in retrospect she would have liked to have been given the opportunity to discuss it. Neither of them knew what to expect, which made the situation frightening. Marion reported that they had not realised this was the terminal stage of the illness until the day Dave died, when a health professional clearly communicated it to her.

Marion: It would be nice to be prepared really more for that, and to know that stage really, what you could expect. Because nobody likes to talk about it really. Perhaps for Dave it would have been difficult. But I think, because when we were in hospital he was quite frightened at all these things happening. He had oxygen and then of course because he couldn't breath properly, he had a machine to expell the carbon monoxide. He didn't know any of this and all the time he kept saying, "I can't breath, I can't breath". But then I found myself running around trying to get a doctor, you know. And the medical staff do not always know enough about MND, with the breathing side to know what's going on really. That's quite a stress really.

The feeling of disempowerment stayed on even after Dave died. Marion told me that social services had contacted her to see if she would be willing to give back the through-floor lift. Not wanting to go through her experience with health and social services once again, and seeing herself as an ageing person who might be using the lift in the near future, she decided to keep the lift, but complained of the insensitivity of the services.

### **8.5 A note on interviewing; Shared experiences**

People living with an illness and their partners may feel angry, happy, embarrassed, tired, confused, content or any other of an endless list of emotions and feelings. Rather than being individual, these experiences are shared. This does not mean that both partners experience the same thing and interpret it in the same way. These experiences are shared because they are co-constructed and they would be something else, something different if one of the partners was not part of the experience. Jackson (1998) and Arendt (1998) remind us that human experience needs to be explored in its intersubjective context. The way Dave and Marion were sharing experiences

and feelings was evident from how they engaged with the interview, completing each other's sentences and building upon each other's points. In the following excerpt, they share their frustration about having to pay for their newly ordered, specially adapted car, without having actually received it.

Dave: You sign for it electronically, they've given me a code to type in to say that I accept the car, that's the time when you start paying for it.  
 Dikaios: And they collect money already?  
 Dave and Marion: They collect money already.  
 Marion: Already four weeks they've taken. But they give it to you back. I don't see why they do it.  
 Dave: I don't see why they don't wait until it comes in.  
 Marion: It's strange. All these things mean chasing, chasing, chasing...It's...  
 Dave:...It's annoying...  
 Marion: It is frustrating, yes.

Often they would complement each other and even sometimes prompt each other to share information. In the following excerpt, Marion prompts Dave to tell me more about their new bed supplied by social services. Although Marion started off the story by saying that they got a new bed, she then invites Dave into the conversation with her suggestion to "*tell Dikaios*".

Marion: You are okay at the moment, but...sort of...more symptoms...and knowing the best way to treat everything really (...) he had a profile bed, one of those with the air mattresses, but that was okay for a while. But Dave finds it a bit more uncomfortable now, because you sleep on your side, don't you. Tell Dikaios how you get out...you were hoping it would be a bit better to get out.  
 Dave: Well, I was....it doesn't go to 90 degrees, it's a bit short of...I can get up so far but it's this last little bit, they have to pull me to get me upright.  
 Marion: They don't do an attachment they have on a normal mattress (...) and the cot sides, which Dave finds good to

pull on, but they don't go right down, so they dig in your knees.

Dave: So, when I sit at the side of the bed they dig in in the back of the leg.

Marion: But I think the ones in the hospital would go straight down. I think they see what they can do, you know, but we'll see (...). We had the district nurses and they gave us some slip sheets which are quite smooth (...).

Dave: To move...when I get in the bed to get in the right position.

After I had transcribed the excerpt above I noticed how well Dave and Marion were coordinating their contributions to the story, filling in the gaps and prompting each other to share more information. During the course of the interview, they would negotiate between them who would say what by directly asking each other, as in the excerpt above, or by introducing a subject and then looking at each other to decide who would share the story. Dave and Marion were keen to get their story heard and often they produced long stories, filling in for each other. Even when not verbally communicated, their frustration with the services and feeling of disempowerment were evident from their long stories, where a single question would often result in a thirty-minute-long, or more, narration.

## **8.6 Chapter summary**

Dave and Marion were relying on professional help, seeking solutions that would address some of the problems they were facing. To some extent, they relinquished the disease and its control to professionals. This sometimes led to Dave and Marion feeling frustrated and disempowered, perhaps because they felt the professionals were not meeting their needs. Professionals and

people living with MND operate in different contexts, with different agendas (Hogden et al., 2012; Ng, Khan, & Mathers, 2009). People with MND often focus on the present, seeking some sort of help that will enable them to live a better life, for example, taking a shower, or being able to sleep together again. Professionals on the other hand focus on the future, and while they might seek to be client-centred, they are restricted by organisational policies. When this dissonance is not clearly communicated, it can lead to confusion and disappointment with services.

This chapter was about how one couple made sense of their story, not necessarily about what actually happened in their interactions with the representatives of health and social care systems. The latter cannot and need not be verified. What matters is the way people interpret things and therefore it was important to get the couple's interpretation of events. In Dave and Marion's life, the disease was structured as an inescapable discourse, which structured their daily life. Dave and Marion felt disempowered and while they constantly tried to be good patients, they felt that the health and the social care systems failed them. They tried to accommodate the factual scientific knowledge embodied by the various health and social care professionals through their everyday practices.

Being a good patient requires a constant negotiation with health and social care professionals. This revealed a tension between Dave and Marion's lived experiences and desires, and the structure and delivery of health and social care services. Long waiting times, inconsistencies of

approach, and just too many people and services to liaise with, led to a feeling of disempowerment and loss of control over their life.

## 9. “He needs someone here, to sort him out”

Maggie and Gareth are both in their early 70s and have been married for more than forty years. Gareth was diagnosed with MND nine years before our first meeting. Initially, they were told that he only had two to three years to live, but when five years passed Maggie started researching about the disease and began suspecting that Gareth had progressive muscular atrophy (PMA), which is a more slowly-moving variant of MND. This was subsequently confirmed in the MND clinic.

Gareth uses a wheelchair for mobilising outdoors and a walking stick or a wheelchair when moving indoors. He also needs help with moving into and out of the wheelchair. Over the course of our meetings, he reported that his arms and hands were becoming increasingly weaker and less dexterous and that he was finding a variety of everyday activities challenging. From combing his hair, cleaning his ears and brushing his teeth to eating and drinking, and from gardening to driving, his repertoire of activities was either becoming narrower or changes were required in the ways he was carrying out these activities.

Partly, these everyday activities are facilitated through Maggie who helps Gareth with toileting, bathing, dressing, getting up from the bed and getting into bed, eating, and transferring. She is there to offer help when Gareth cannot do something, or when he finds an activity challenging. She also carries out all those daily life activities that are necessary, such as paying the bills, driving, attending to the garden, arranging medical

appointments, and food shopping. In brief, she carries out what Kleinman and van der Geest (2009) called the ‘technical/practical’ constituents of caring. She does more than that though, as she is also engaged emotionally in the process of caring for her partner. In this chapter, I focus on the performance and meanings of care in MND from the perspective of the person who offers care. Using literature on care and caring (mainly from medical anthropology and nursing), I describe how Maggie makes sense of living with MND as Gareth’s carer. Although she is not the one diagnosed with MND, the experience of the disease is part of her daily life and in that sense not only Gareth but Maggie also lives with MND.

## **9.1 The context of care**

Before discussing Maggie’s experiences of care, it is useful to describe the context within which these experiences take place. Maggie is one of more than 370,000 carers in Wales (Welsh Government, 2013), who perform the majority of care in the community (Welsh Government, 2007). Statistics show that over 100,000 of these care for more than 50 hours a week (Welsh Government, 2013). Both the percentage of the population in Wales offering care and the absolute number of carers are rising.

The government documents dealing with care use the term ‘carer’ to refer to the person offering care. The terms ‘care’ and ‘carer’ are used in a very specific way in the policy and strategy documents. Care is used to refer to these activities carried out by friends or relatives of a person who due to health or social circumstances requires short or long-term assistance in the

community. Consequently, the term carer does not include all persons offering care but solely people who care for a relative or friend in the community without remuneration (Heaton, 1999). Paid carers, health and social care professionals, and volunteers are not included in this definition. Heaton (1999) observed that until approximately the early 1990s, the official documents made a distinction between informal carers (the current 'carers') and family members. Gradually, an expectation was constructed that care is to be offered by family members or friends, and informal carers and family members were subsumed under the rubric of carers. Such care, provided in the community by the community has often been regarded as an indication of increased power for people who receive or who provide care, as they can employ care within their own environments, in ways that are best suited to their needs (Fine, 2007).

Alternative interpretations of care exist. For Heaton (1999), the emphasis of care, as it is defined above, in health and social care structures indicates "*the devolution and extension of the [medical] gaze*" (p.773). Rather than giving more power to people, the process of care as conceptualised in official documents offers the ground for a different *modus operandi* of the medical gaze, through the carers who act as its agents. Furthermore, through their delineation as a specific group, carers are also subjected to surveillance and are the subject of policies in their own right (for example, in Department of Health, 2008).

Whether care is a tool for the extension of the medical gaze or a means to greater autonomy makes little difference in the daily life of

Maggie and Gareth. What is important to them is not the motivations behind the increased emphasis in care, but the practices associated with this emphasis. The Department of Health (DoH, 2008) recognises the importance of carers as they offer the majority of care. The fact that carers are not paid represents savings for health and social care services who in the absence of carers would have to put other arrangements in place, such as moving people to nursing homes or providing more intensive home care packages than the ones currently available.

The Carers Strategy for Wales 2013 (Welsh Government, 2013) document states that “*no carer can be expected to care 24/7 for 365 days a year*” (p.32), and the Department of Health (2008) states that one of its priorities is for carers to be able “*to have a life of their own outside of their caring role*” (p.17). Services to offer help to carers have been developed, such as respite care (Appendix A) or day care centres. Carers can request an assessment for their specific needs as carers (DoH, 1995), and the outcomes of this assessment might instigate the provision of services.

## **9.2 Defining care**

Care can be viewed as a process and a practice, and has been discussed in terms of a logic (Mol, 2008) or in terms of an ethic (Tronto, 1993) or even multiple ethics (Held, 2006). For Tronto (1993)

Care [first] implies a reaching out to something other than the self: it is neither self-referring nor self-absorbing. Second, care implicitly suggests that it will lead to some type of action (p.102).

As an analytical category it was developed by feminist scholars and first examined within the family, focusing on issues of care vis-a-vis unpaid labour distribution and responsibility (Daly, 2002). Several authors (for example, Fine, 2007; Held, 2006) argue that care refers to two interconnected yet distinct domains. In the first domain, care refers to actual practices that are carried out, either as a response to a process of concern or as a response to an identified need. These attitudes and practices require the presence of a caregiver (i.e. a person who offers care) and a care recipient (i.e. a person who receives care) (Mol, Moser, & Pols, 2010). In the second domain, care is used to indicate a mental disposition or an emotional attachment, which leads to a process of concern, worry and attentiveness.

Research on care in MND has highlighted the importance of the emotional aspects of caring (for example, Ray & Street, 2005; 2007). Caring is not only about doing things for or with other people and offering help. Caring is also about concern about another person and as such it is an inherently intersubjective practice. In their study on the experience of caring for people with MND, Herz, McKinnon and Butow (2006) found that the emotional elements of care were more challenging than its practical side. The two cannot be separated and the practical elements of caring intertwine with the emotional. For Tronto (1993), the process of care can be broken down into four distinct phases, starting from a concern (caring about) and progressing to making plans (taking care of), engaging in care giving practices (care-giving) and finally anticipating a response (care-receiving) that has the potential to modify the process.

Care can be viewed as an interaction between two people, although the relative roles of these are not always clear (for example, is the recipient a passive actor, or can he or she play an active role in the interaction?). Chattoo and Ahmad (2008) argue that in literature on care people involved in relationships of care are often explored in their individuality rather as two integral elements of an intersubjective experience. Although this chapter focuses on Maggie's experience, where possible, the voices of both Maggie and Gareth are included to highlight the shared nature of the experience of care.

Literature on caregiving in general, and on caregiving in the context of MND more specifically, often focuses on the process of care as a problem, where caregiving is framed as burden (for example, Chio et al., 2006; Hecht et al., 2003; Miyashita et al., 2009). This reflects one significant aspect of the experience of caregiving. Indeed, many people who have caring responsibilities feel tired, emotionally exhausted and mourn for the life they feel has been taken away from them. Care is not easy. Writing about her experience of caring for her husband who had MND, medical anthropologist Martine Verwey (2010) described the tensions she was experiencing: "*I know that one can love another person very deeply and at the same time, out of desperation, want to kill that person*" (p.38). However, as Verwey (2010) and Kleinman (2009) have argued, caregiving can also be the natural progression of a loving relationship when something does not go right, like when one is diagnosed with MND and as such it is a deeply intersubjective experience. In her monograph on the construction of hope in

the context of long term, serious illness, Mattingly (2010) described how one family practiced care:

So, we pair up the slow person with someone else in the family, a cousin, a sister or brother, something like that, and they become a kind of twin. They help the one who is slow, like my boy. You could say it's their family job (Mattingly, 2010, p.22).

Care can be viewed as a process of twining, involving “*protection and care*”, and “*self-sacrifice*” (Mattingly, 2010, p.23). The following sections describe how Maggie experiences the process of caregiving.

### **9.3 Becoming a carer**

Maggie and Gareth had anticipated a long retirement where they could enjoy time travelling and generally have a good time. They would play golf, spend more time with their grandchildren and friends, and maybe renovate the house. The diagnosis of MND changed that anticipated future by introducing many uncertainties and one certainty into their life. The one certainty was that Gareth had a progressive disease that would affect his ability to carry out everyday activities. This one certainty however belied the many uncertainties Maggie and Gareth were experiencing; although they knew how MND *can* affect somebody, they did not know, and had no way of knowing, exactly how Gareth would be affected, or how fast. This made it hard to make plans, as Maggie explained:

Gareth just thinks these are the cards that we have been dealt and we've got to deal with it [Gareth nods] (...) All the plans that you've made all through your marriage, the children are grown up, are off your hands, retirement, you know...but of course that all came to...you go from having

a wide horizon to having one that is very close to you. You don't think much further ahead than, uhm, a month or two really, if that...we are quite happy, contentment I think the word is. We are quite content. Well, you got to be, don't you? What can you do...you can't fight against it (...) therefore you have to accept it and get on with it.

As the condition progressed and Gareth's muscles became weaker, Maggie had to step in and help. When Gareth first started needing help with some activities, Maggie and Gareth did not sit down to discuss it, or make special arrangements, or consider other options. Maggie had not planned to become a carer. She did it because it was there to be done, as Kleinman (2009) reflected on his own experience of becoming a carer. There are perhaps two main reasons why this happened. For the first few years after diagnosis Gareth could still carry out by himself most activities so that there was no major change in their daily life. Having lived together for more than forty years they had learnt to help each other and it felt natural for Maggie to start offering help as a way of adapting to a changing context. Changes were introduced little by little and it was not until several years later that Maggie realised that she had become a full-time carer. A second reason is that at the start of the disease, Maggie and Gareth thought that theirs would be a short illness trajectory. When Gareth was diagnosed they were given the standard prognosis for the ALS form of MND and the possibility of a slower progression was not discussed.

Maggie: Yes, yeah. And, uhm, and then of course as soon as we got this diagnosis I gave up work, more or less straight away because we thought it was going to be, you know, Gareth wasn't going to be around for very long. So I gave up work to look after him and that's what I do. And

that [is how] our day is filled then, with me looking after him (...).

Gareth kept on working for one year after he was diagnosed and then he retired as he felt he could not cope with the demands any longer. Maggie decided to also stop working and spend time with Gareth, as they both thought they would have only another two to three years together, based on the prognosis they were given. However, Gareth was consequently diagnosed with a slow progressing form of MND, which necessitated a more extended period of caregiving than they had anticipated.

## 9.4 Being a carer

### 9.4.1 *The content of care*

*“He needs someone here, to sort him out”*

Maggie: No, he needs someone here, to sort him out (...) to dress him, to dry him out, to dress him for bed.

Gareth: I don't mind that at all, she has a nice touch.

For Maggie and Gareth, caring entails all these activities that Gareth finds difficult to perform by himself. Maggie needs to “*sort him out*”, which can mean helping him to dress, wash, dry out, clean his ears, but also undress, go to the toilet, eat and drink, and get up when he falls on the floor, and other personal activities of daily living. To some extent these activities appear to happen automatically as an inherent part of a long-term relationship. Our meetings and discussions often took place around the kitchen table and I could see how Maggie would place a glass of water just in the right position between Gareth's hands, or help him wear his glasses, or stand behind his chair and hold it steady as he was getting up. These were

activities that felt natural and were performed without much thinking. They were done because they had to be done. This however does not mean that care activities are introduced into a couple's life without any impact.

Maggie: Yes, it's just been a very gradual...don't really notice these things happening until all of a sudden, "oh, I hadn't noticed that before". So we now got to a stage where I got to help you to dress, wash...he can manage the shaving himself. Cut his nails...you know, things that we take for granted. Going to the toilet, [asking Gareth] you can't manage that on your own any more, can you?

Care has many dimensions. There is the quantitative dimension that considers to what extent someone is dependent upon someone else. This dimension is operationalised through functioning rating scales, such as the revised ALS functioning rating scale (ALSFRS-R) (Kaufmann et al., 2005) or the combined assessment of function and survival (CAFS) (Berry et al., 2013). Issues of concern for this dimension are whether Gareth can perform certain activities, such as eating or going to the toilet, and whether he needs help to do so.

Care, however, also has a qualitative dimension, which considers not how much help Gareth needs, but what kind of help. As Struhkamp, Mol and Swierstra (2009) observed, there are variations in dependence and independence and in the help that people need in order to carry out certain activities. To say that Gareth needs help with some activities, or is dependent upon Maggie, does not give information on exactly what it is that Maggie needs to do to help Gareth. For some activities, it is enough for Maggie just to be there and offer minimal assistance, like for example with eating. She needs to serve the food and cut it up, but then Gareth can eat by

himself. When he is in the living room reading the newspaper, Maggie just needs to be around in case he needs to go to the toilet or have some water. For some other activities, like cleaning Gareth after he goes to the toilet, more assistance is needed.

Maggie and Gareth live in the countryside and they enjoy going out. Soon after our first meeting, Gareth's legs became weak and he was finding walking difficult. Gareth started using a wheelchair when outdoors enabling them to go for walks again. They both commented on what a positive influence being able to go out had in their lives. However, the wheelchair has to be put into the car and out of the car. It needs to be lifted, collapsed and pushed open again. And it has to be propelled. Gareth cannot self-propel so Maggie has to do that and although she enjoys being able to go out for walks with Gareth she finds all these accompanying tasks tiring.

Maggie and Gareth often receive help from friends. In their study on the social support networks for people living with MND, Ray and Street (2005) noted that people over 60 years of age had strong and diverse sources of community support. Maggie and Gareth have lived in the same neighbourhood for over 40 years and they have an extended network of friends who live locally; some of them they met through sports activities, others through work and others through being in the same community for a long time. They also have three grown up children who support them, although they do not live locally. Friends and family help with rides to hospital appointments, carrying luggage when travelling or helping to lift Gareth when he has a fall. They also offer moral support just by being there

and caring about Maggie and Gareth. Sometimes they also come up with ideas to make daily life a little bit easier.

Maggie: One of our friends has made a special thing for Gareth when he holds his beer glass in the pub [she demonstrates it- a rubber wrap around for the pint glass, with handles for the hands to go through]. As long as it's not full right up to the top Gareth can lift it.

Gareth: If it's full up to the top I can drink the first sip from the table.

Maggie: It's made out of...I think it is the inner tube of a tyre and it's all stitched, look. You can't get anything like this. So there is one that stays down the pub and we have one at home if we go somewhere else.

Gareth: Best invention ever, that.

Activities, such as transferring or going to the toilet, described above are an important part of care, especially because they cannot be avoided but have to be carried out somehow. As Maggie found out however, other activities are also important and she eventually had to take on activities that used to be part of Gareth's role in their relationship.

Maggie: I have to do all the driving now. I see to the garden. Gareth used to love gardening. He used to grow championship vegetables, didn't you?

Gareth: I used to take most of the ...

Maggie: ...Prizes in the locals shows.

Gareth: Prizes in the local shows.

Maggie: Massive great leeks. Huge. But of course he can't do any of that now.

Gareth: About five years ago, I stuck a fork in the ground and couldn't get it out. That was my last move in the garden.

Maggie: I have to pay all the bills. Just do everything that most husbands do, plus what a wife does, all the cooking and cleaning. It has made a big difference.

Gareth: We've changed now. I do the beds and the ironing [he laughs].

Maggie: Yes...so....life has changed...drastically.

*“Life has changed drastically”* means that the roles they had learned to perform and the positions they occupied in their relationship had to be shifted. Fraser (1989) argues that care is located within local contexts and is part of peoples’ lives and their structures of interdependence. Through four decades of shared living, Maggie and Gareth had established a repertoire of activities and constructed a social identity that was closely related to these activities; Gareth was the avid gardener and prize-winning vegetable grower; Maggie was the stay-at-home mum running a household that included three children, and later on several grandchildren.

Maggie: Gareth was, well, full time job working, keeping the business going. And he was a sportsman, so he wasn’t home an awful lot. So, I’m, I’m a nurturing type of person, so I enjoyed bringing up the children, and that’s the old fashioned marriage that we had.

Maggie’s role never entailed gardening or paying the bills; those were Gareth’s responsibilities. As a result of Gareth’s increasing muscle weakness, Maggie had to take on these new activities and add them to her repertoire. These new activities were in addition to the other, more easily recognisable caring responsibilities for activities such as going to the toilet. The aim of care is not only to look after a physical body but also to look after a social identity and the accompanied activities. Furthermore, it is to ensure that necessary activities are reallocated so they are still carried out. In the process of doing this Maggie’s identity had to be modified to incorporate new roles and new meanings, as also found by Hughes, Locock and Ziebland (2013). Maggie and Gareth live in a semi-rural area and they

need to drive to get to most places. This was something Gareth always did. Now that he can no longer drive, Maggie had to take on this activity.

As they both age and MND progresses, Maggie has to offer help for a growing number of activities. So far these are incorporated into her daily life and she can carry out activities such as driving, paying the bills, and propelling the wheelchair without much trouble. But what will happen in the future?

Maggie: So, we [are] just gradually taking on board more and more...I help him as much as I possibly can. When I get to the stage where I can't do any more then we'll have to make some inquiries to see if we can get some help in.

Care can be tiring and time consuming and Maggie worries that she might not be able to care for Gareth in the future, because she might be weaker due to age and because Gareth's needs might be greater due to a combination of progressing age and MND. There are potential solutions and Maggie knows that she needs to "*make some inquiries*" to find out what kind of support might be available or suitable; respite care, a care package, arrangements with family and friends to offer more support, or perhaps some equipment are all potential solutions. What will be needed will partly depend on how MND will progress in the future.

#### 9.4.2 *The timing of care*

*"He needs someone here, to sort him out"*

*"He needs someone here"*, as Maggie said, points to two different dimensions of care; its content but also its timing. As described in the paragraph above, Gareth needs help with some activities, such as eating and

transferring, due to increasing muscle weakness. Maggie and Gareth know what needs to be done and how. What they do not, and cannot, know is when Gareth will require care. While some aspects of care, such as help with getting out of bed, dressing, or undressing and being put to bed, happen at relatively fixed times, other activities are not as predictable. Going to the toilet, having a fall, or wanting a glass of water cannot be planned. Although the content of the help might be known and sometimes minimal, someone has to be available to help Gareth. As the following quote illustrates, care requirements constantly change:

Maggie: Now, going to the toilet is getting difficult. So, whether there are any aids about that we can have to help Ken...clean himself up. I mean, I'll do it, but he doesn't like it.

Gareth: Difficult now because I am losing the feeling in all my fingers, you know.

Maggie: The same with cleaning his ears out. You don't realise, do you, when you...

Dikaios: No, because you need really fine movement.

Maggie: Yes, yes. And he's got none of that, so..I am gonna have to get in touch with the occupational therapy and ask them to suggest something.

Gareth: You take for granted and then it suddenly don't happen.

Maggie: Suddenly it's taken away and it's not likely to come back.

Gareth: Crazy world.

This adds to the general unpredictability experienced by Maggie. The realities of care mean that it is hard to make short-term plans, which depend to some extent on care responsibilities that cannot always be planned in advance.

Unless people with MND have difficulty with breathing or managing saliva, which can be frightening and potentially dangerous, they usually do

not require somebody to be present at all times. Gareth has no problem managing his saliva, breathes without the use of any equipment and does not experience discomfort or difficulties in doing so. In theory, he could spend part of the day by himself. However, the help he needs while sometimes minimal, is necessary. If, for example, he needs to go to the toilet, someone has to be there to help him get up from the chair and clean himself after he uses the toilet. In other words it is not so much that Gareth needs a lot of help with all activities, but that the help he needs cannot be planned in advance. Care is unpredictable, as is life. Worrying that Gareth might need her while out of the house, Maggie tends to stay at home.

Maggie: The children keep saying, “take up painting again mum”, so, I can’t be bothered at the moment. But, uh, you know, I will...one day...I used to quite enjoy that. I used to go twice a week. But then it got that I was really worried all the time that I was out. I was concerned that Gareth was going to fall out of bed, or get up (...) I was always worried that he would fall down the stairs, that was the worst thing (...) And...but now we got the stairlift which is marvelous, and he doesn’t have to cross the top of the stairs to go to the toilet when he is upstairs (...) And...so that’s a lot better...

Going out for medical appointments or shopping means leaving Gareth alone. But, as she said:

I leave Gareth in bed, because I know he is safe when in bed. So, I mean, I was an hour and a half waiting in the doctor’s surgery which is very annoying, but I know he is safe because if I go out I lock the door. He is quite happy lying in bed, aren’t you (Gareth: Yeah) until I come back. But I wouldn’t leave him if I knew he would be wandering around.

Maggie and Gareth have developed their own ways to deal with the short-term unpredictability of care in the context of daily life where sometimes

she has to go out for a while. Earlier in this chapter, I discussed how Maggie feels that it is hard to make long-term plans because of Gareth's diagnosis and the fact that they cannot know how MND will progress, or how fast. The kind of help that Gareth needs changes as the disease progresses. To give one example, increasing weakness in his hands and arms that developed over the course of a few months during our meetings meant that he ultimately needed help combing his hair and brushing his teeth. Although these changes happen relatively slowly and thus allow Maggie and Gareth to adjust to a new content of care, nobody can predict what kind of care will be needed in the future. Whether Gareth will be able to walk, whether he will need NIPPV, and whether Maggie will need to help with it and when that might be, are questions that cannot be answered. In these cases, both the content and the timing of the care are unknown and become a source of uncertainty.

Maggie is worried about the future and how they will be able to cope, in the face of advancing age and progressing impairment. They have a supporting network of close friends who offer help, but several of them are in their 70s and deal with ill health themselves. They also receive support from social services who have come up with solutions for several of the problems they face, or that they might face in the future. Despite these sources of support, the changes that MND and the associated caring responsibilities have brought on Maggie's and Gareth's life are major. The following excerpt reflects the anxiety Maggie feels about the future.

Maggie: But the storytelling group, I think you are coming to a stage where it is harder and harder for you to go to the storytelling group, haven't you?

Gareth: No. I enjoy going to it, I just have to be careful. Can't go to any venues where there are steps.

Maggie: Yes, it's getting more difficult. There will come a stage when you won't be able to go to the storytelling group.

Gareth: As far as there are no steps.

Maggie: Yes, I know that, but there will come a stage when you can't walk at all.

Gareth: Yes, but I can sit in a chair, so I don't have to.

Maggie: Yes, but I will still have to get you in and out of the car, Gareth. It isn't going to be that straightforward, is it.

Gareth: I don't know, I can't tell.

Maggie: So, we don't know. But he does enjoy it.

Gareth: Yes.

In the excerpt above Maggie and Gareth discuss Gareth's involvement in a storytelling group that Gareth has been involved in for many years. Going to the storytelling group is something that he enjoys a lot and is also a source of support as he regularly goes out for meals with other people from the group. Maggie also enjoys attending the group and thinks it is important for Gareth to keep on going. However, Maggie anticipates future difficulties and is reluctant to make any plans regarding Gareth's continuing involvement in the group and participation in storytelling events out of town, while Gareth focuses more on his present experiences.

People living with an illness and their partners can experience illness in different ways because their roles are different. Even when they carry out an activity together, they can ascribe different meanings to that activity (van Nes et al., 2012; van Nes, Runge, & Jonsson, 2009). Maggie sometimes has

to accept loss of control as she does not know what the future will look like and this can be frustrating. While Gareth does not think about a future that is unknown, Maggie anticipates difficulties to come and does not know how they will cope with these new, but yet unknown, challenges. The unpredictability of the future has been highlighted as a source of stress in research on the experiences of partners of people with MND (Oyebode, Smith, & Morrison, 2013). Ray and Street (2007) stated that

Continually facing the physical evidence of degeneration and not knowing what to expect next, increased the loss of certainty and predictability in life and the capacity for short and long term planning (p.38).

Both Maggie and Gareth expressed the different ways they made sense of living with an illness but in the end they arrived at a shared understanding that they did not really know how the disease would progress and whether Gareth would be able to still participate in the storytelling group in the future.

Several times during our conversations Maggie used the metaphor of the horizon to express how she felt about the future.

As you grow older your horizon gets narrower, and then something happens and the horizon suddenly gets very narrow.

A narrow horizon is an horizon that does not allow much scope for dreaming and many diversions from the immediate reality. A narrow horizon refers to a future where possibilities turn into uncertainties. If an imaginative horizon for Crapanzano (2004) refers to an ability to imagine what might lie beyond the present, what possibilities might emerge in the future and what these might look like, Maggie's horizon is dominated by

MND and she cannot construct a notion of the future that she is satisfied with.

A possibility refers to something that a person wants to do, something that is possible. Maggie wanted to go travelling after retirement and before Gareth's diagnosis this was a possibility that Maggie and Gareth felt they were in control of. After the diagnosis, what was previously a possibility became an uncertainty. Whether or not they would be able to travel was not viewed as something they had control over, but as an uncertainty caused by the MND and its symptoms. Maggie's horizon is not narrow because the range of possibilities is smaller, but because she does not feel she is in control of these possibilities.

## **9.5 Reflections on dealing with heteroglossia**

The standpoint and perspectives of people living with an illness and their partners do not always coincide. Sometimes they experience illness in different ways because their roles are different. The use of joint interviews allows for the different perspectives and voices to be heard (Bjørnholt & Farstad, 2012). Although I was intellectually prepared for participants to be expressing contrasting views, perhaps disagreeing with each other, in interviews, I felt uncomfortable the first times it happened. When Maggie and Gareth were discussing about his involvement in the storytelling group, I remember how awkward I felt, and yet unable to intervene in a meaningful way.

After I had listened to the first interview a few times, I realised that what made me uncomfortable was not so much the differing perspectives expressed by Maggie and Gareth as the realisation that the interview was performed in a heterarchic way, with power distributed between the three participants, one of them being me. The two research participants could, and did, have conversations without my direct involvement. In theory, this was something I was trying to achieve, as I wanted to share control of the interview (Ziebland, 2013). When I realised that it was the heterarchy of the interview that made me feel awkward through my lack of control over the unfolding situation, I was able to relinquish the need for control and take part in the conversation without assuming full responsibility for it.

Maggie and Gareth's conversations offered rich data illustrating the mechanisms of heteroglossia. Maggie would often focus on the future, anticipating difficulties to come that it was impossible to plan for, whereas Gareth would focus more on the present. The foregrounding of the heteroglossic nature of illness experiences through the use of joint interviews highlighted the contextual nature of care practices. Letiche (2008) referred to care as 'being in-between' or 'being-two' because it cannot be defined by a single process but it is always situation-bound and depends on who delivers it and who receives it; in effect the meaning of care is co-constructed by the people who are involved in it. The foregrounding of heteroglossia also highlighted to me that letting go of power during interviews requires acknowledging and respecting the power

of the research participants to guide the interview where they feel they need to.

## **9.6 Chapter summary**

Maggie and Gareth have lived together for many years, taking care of each other in many different ways. Living with MND has necessitated a change in their repertoire of activities. Maggie had to take on new roles, such as driving, and let go of others such as work. She also had to learn to care for Gareth in new ways, helping him carry out activities that he used to do by himself. The process of care was not only about doing things for or with Gareth, but also about worrying about him, and worrying about the future. One of the main changes brought about by MND in their common life was the introduction of a pervasive sense of uncertainty. For Maggie and Gareth, the future was no longer theirs to make plans, but it was dependent on what kind of care would be needed.

Care can be disruptive and exhausting, both emotionally and physically. However, framing care as something negative, to be avoided or alleviated, can disregard the broader context of care. Difficult as it is, for Maggie and Gareth, care developed within the context of a life course within relations of interdependence and love.

### 10. “I got to be me first and my illness second”

Arleen is a woman in her early fifties. She lives in an urban area and takes pride in her house, which she keeps in an immaculate condition. Arleen has a big social circle and enjoys spending time with friends and family and entertaining people at home. She is also well-travelled and we often talked about some of the places she has been to, which she always described in vivid detail. She is in a long-term relationship and when data collection started Arleen and her partner each maintained their own house. This was done out of choice as Arleen values her independence and she wanted to keep her own living space rather than move in with somebody. Arleen was diagnosed with MND two years prior to her participation in this study. The process of diagnosis was pretty straightforward for her, as she described in an email message:

Late 2004: I began to notice a lack of fine dexterity with my right hand; this was magnified in the work place, as many of the tasks demanded precise movements.

January 2005: I initially sought the advice of an Orthopaedic Surgeon, thinking it was a trapped nerve! X-rays & MRI [magnetic resonance imaging] scan of C-spine [cervical spine] & shoulder, no problem, was advised to “see” a Neurologist. Routine Neurological tests plus lumbar puncture, & brain MRI.

August 2005: Diagnosed with PLS/MND [primary lateral sclerosis subtype of MND].

Arleen believes that the main reason why she did not face problems such as delay with the process of diagnosis, or with access to services later on, was because she had in depth knowledge of the modus operandi of the health and social care system due to her professional role within those systems.

Arleen continued working for some time after diagnosis but she eventually retired on medical terms when dysarthria started posing considerable communication challenges.

MND threatened to encroach upon Arleen's control over her life. During our conversations she explained how she tried to make those choices that would enable her to continue living her life the way she wanted to. What was at stake for Arleen in her effort to take control of her life was her sense of identity, which she felt was threatened by MND.

In this chapter, I highlight Arleen's desire to live her life without letting MND affect her too much. To illustrate this I draw on critical disability studies and particularly on the concepts of recognition and body, as they relate to Arleen's narrative. This chapter concludes with a discussion of my own involvement in the construction of this narrative.

### **10.1 Arleen's storylines**

Arleen: Yes...I think too much, I put problems when there are no problems.

Dikaios: You said before how you find solutions, there is always a way around things. Have there been any things that you had to stop doing, or that you had to ask help from somebody?

Arleen: Uhm, I don't go shopping on my own, I always go with somebody, but I go around the supermarket on my own. And you can always ask somebody to do something. I don't go to the local shops anymore because they are on a really steep hill. So, there are things that I don't do. I don't use public transport anymore. Because I always used to either walk or catch a bus into town because the parking is too expensive. But now we have to park. Uhm, I think to be happy and to still be positive you mustn't dwell on these things.

The excerpt above highlights the two main storylines that started to emerge from the conversations with Arleen. The one storyline revolved around the body and how Arleen experienced the changes brought about by MND. Borrowing from Leder (1990), this storyline is about the body's 'dys-appearance', or in other words about the corporeal experience of MND through the problematic appearance of the body. The other storyline is about the transactional nature of disability, and the possibility to transcend it through changes in one's life, such as stop using public transport or shopping locally. Initially I thought these two storylines were conflicting and I was trying to see which one was more *true* to Arleen. I had to remind myself that I was not looking for a definitive truth, but for different plots that combined together would tell a story of living with MND. In this chapter, I illustrate how these two seemingly contrasting storylines complemented each other in Arleen's story.

## **10.2 On dysarthria and other corporeal experiences of MND**

Arleen has dysarthria. As a result of this, she speaks slowly and some consonants are not articulated clearly. At Arleen's request our initial interactions took place via email and when we first met in person I remember being nervous, worrying I might not be able to understand her leading to a potentially awkward situation, especially since it was me who had suggested that meeting in person might facilitate data collection. However, Arleen's speech was entirely intelligible and over the course of the 18 months of her involvement in the study, I cannot recall a time when I

could not understand what she was saying. This might partly be because I am sensitive to altered speech patterns, both through my previous clinical experience as an occupational therapist, and also through my personal experiences as my mother developed dysarthria early on in the process of her illness. It might also be because of the nature and location of our conversations; we would often begin with me asking a question and waiting for an answer, with no particular time pressures. Our conversations took place in Arleen's living room, which was a quiet room, with no distractions. In different situations, Arleen was more aware of the effects of dysarthria.

Arleen: The frustrating thing is I can't interact very quickly with any situation. I can't jump into a conversation, even though I still go out and socialise. Even going out for a meal I cannot eat and talk at the same time anymore and I can't move as quickly as I used to. Uhm, yeah, that's what's really frustrating me, the actual time thing.

Arleen is made aware of the ways MND affects her life through the changes brought about in the way she interacts with the world around her; not being able to talk as fast as she used to becomes problematic when it affects her ability to enact sociality. Social interactions have a tempo and dysarthria threatens to disrupt that tempo by imposing a speed limit. It is not only dysarthria, however. As Arleen explains, many activities seem to be taking more time:

Arleen: Uhm, I get really angry and frustrated with myself when I can't do something. But, there is always more than one way to do something, so...uhm, to stay independent I have to make changes. When I find out I can't do something, I don't stop doing it, I find another way to do it. Like, cleaning the windows. I can't balance to stretch out to the top. So, I get a pole with a duster on it and I can still do

the windows. Uhm, I am not happy that everything is a lot slower, but there is nothing I can do about that.

It is not so much the fact that she cannot balance on top of a stool; it is the fact that she cannot use her body in a way that was familiar to do something that was commonplace in her repertoire of everyday activities. What matters for Arleen is not the actual speed but the degree to which this seems to be altered. In other words, it is the perception of change.

Arleen's body is somehow altered as a result of the MND. As motor neurones degenerate, muscles atrophy leading to reduced strength in different muscle groups. Changes can be imperceptible until they reach a threshold over which the weakness cannot be counterbalanced by other muscles. From that point on, degenerated motor neurones translate into perceptible changes such as slurred speech, or difficulty cleaning the windows. Leder (1990) remarked that in disability the body *dys*-appears; it makes its presence felt when the way people engage with the world changes. For Arleen it was a changed speech pattern that signalled that her body was changing and her relationship with the world around her was affected. From the background where it was located, her body emerged into the foreground of consciousness and the way it was experienced changed.

As Arleen declares, she can still do all the things she used to do before; she had to modify some of the activities, for example, by using the car more than she did before, or allowing more time to carry out activities. This does not imply that one way of being or doing is better than the other.

What is at stake here is a phenomenological change of the lived-body, rather than a moral statement about a preferred way of being.

As an analytical category, the body has for a long time been conceptualised in two distinct ways; as lived and as physical. The focus of disability studies based on the social model of disability has been with the social and political environment, and more specifically with the construction of disability as oppression by a disabling society. The social model of disability has thus been constructed as a discourse of resistance to what has been perceived to be a normalising biomedical discourse that supports rather than resists the social construction of disability. But what began as a discourse of resistance has now produced its own normalising discourse that decrees what is right and what is wrong (Shildrick, 2005). Marks (1999), discussed some of the problems of the “*either/or models of disability*” (p.611) demonstrating that they are inadequate as their focus is too narrow, while more recently Siebers (2008) discussed the re-evaluation of the importance of corporeality within disability studies. Several authors are moving beyond the lived body/physical body divide and acknowledge the body in all its complexity. Work by Longhurst (2001) and Papadimitriou (2008b), for example, take into account multiple dimensions of the body.

As Arleen’s experiences show, this bifurcation can be problematic. MND might in part be located within the physical body of a person, but for it to manifest as a problem it often needs to affect a *lived* body, a body-in-the-world. The body has a central role in Merleau-Ponty’s (1962/2002) phenomenology of perception, where he perceives the body as the main

means we have to make sense of the world; through our body we are constantly engaged in a dynamic dialogue with our surroundings. Several researchers have followed on from his ideas and explored the centrality of the body in the experience of disability (see for example, Seymour, 1998; Cole, 2004; 2009), and also the process of re-embodiment after disability (Papadimitriou, 2008b).

### **10.3 “Someone with my *condition*”; Being misrecognised**

Arleen lives in a spacious flat, in a relatively central location, close to shops and a big park. Her front door, which is only used by her, is on the ground floor, but the living space is one flight of stairs up on the first floor. When I first met Arleen she had just received a power wheelchair and she was looking forward to going shopping again by herself. However, as she does not have any space in the entrance of her flat to store the wheelchair and it is not possible to move it up and down the stairs to the main living space, she keeps it in the garage. The garage is accessed through an uneven pathway that can be quite slippery when wet and especially when there are leaves on the path. Recognising this as a problem, she wanted to have handrails installed for part of the pathway, but this was not as straightforward as she had hoped, as she described in an email message:

My garage is at the front of my property & the garage occupies the ground floor, the front door is at the back & accessed via a pathway on a gradual incline consisting of a number of steps some metres apart. Even though it is a public footpath it only houses 12 properties & where the steps are situated is a blind spot not overlooked by any properties or the car park/road. On dark mornings (I usually left the house before 07.15) I began to lose my confidence

on the steps. I applied to the Council for a hand rail to be installed, I was told that the waiting list for the “District” Physiotherapist would be anything from 3 to 6 months, & with my disease it properly would not be worth it. I was told that it would have been futile for me to go on the waiting list for a district Physiotherapist assessment for outside hand-rails as the waiting list was too long for someone with my “condition” (...) This [is] one of many “like” examples. All this makes me so sad & very frustrated not only for me but for others who get wrongly categorised.

In Arleen’s story, someone with *her* condition is recognised as someone who has a few months to live and services cannot help her. In the story shared Arleen *is* the MND. The boundaries between MND and personhood blur when important decisions concerning everyday life are taken on the basis of a pathological entity alone without regard to the specificities of the context and the person. Arleen was sad and frustrated for being wrongly categorised, for being *mis*recognised not as Arleen but as *someone with MND*, having her individuality and personal needs ignored in favour of a group identity.

A diagnosis of MND, or other serious, incurable diseases, often leads to an overwhelming focus on pathology and limitations. This focus on a perceived lack of ability might stem from an existential angst guarding off against disease, as Sontag (1991) observed in her analysis of the negative metaphors surrounding AIDS and cancer. It might also stem from the dominating metaphor of medical practices as a fight against disease, which is seen as a body foreign to the person who is ill. In the case of a person with an incurable disease, someone with Arleen’s condition -with *her* condition- this fight is lost even before it begins.

These metaphors and their effects are not always easy to discern. During our first couple of interviews, Arleen would state how lucky she felt for knowing how the health and social care systems work and having privileged access to them through her professional network. Gradually, through sharing stories about her interactions with these systems she came to a different conclusion, as illustrated through the following email excerpt.

I realise that the system is under a considerable financial strain, but my frustration & concern is that even Social & Health Professionals only see MND as a disease with a rapid progression & a short lifespan.

According to the philosopher Nancy Fraser (2000)

What requires recognition is not group-specific identity but the status of individual group members as full partners in social interaction. Misrecognition, accordingly, does not mean the depreciation and deformation of group identity, but social subordination - in the sense of being prevented from participating as a peer in social life (p.113).

Being constructed as just another case of MND, Arleen was given a group identity that had little to do with either her needs or with how she viewed herself. Rather than an abstract sense of exclusion, Arleen felt she was denied her chance to participate on a very practical level, by being told it is futile to apply for adaptations, although it was of course her right to proceed with the application and she was aware of this. But who is to blame for this situation? The professional who advised Arleen against having the handrails installed might have in fact be trying to be helpful by giving her a realistic estimate of how long the adaptations would take to be carried out (advice, which as I described in chapter 8, is sometimes needed). His or her intention might have been to help Arleen make an informed decision as to whether

she wants to wait for six months for an assessment, given the average life expectancy of a person with MND. In fact, following that conversation, Arleen decided to get the handrails installed privately. However, rather than feeling that she was helped, she felt like she was treated as another representative case.

#### **10.4 On the transactional nature of disability**

With no accessible route to her garage, Arleen was disabled. She was disabled by an interaction between a body affected by MND, a physical terrain and a service that was too slow to respond to her needs. A change in any of these elements could eliminate the disability that Arleen experienced; while changing the body was not an option, changing the physical terrain, either through a publicly funded service or privately, would enable her to access the garage again. So, Arleen took action:

I was then transferred back to the Council switchboard, who asked which department I wanted, where I explained my predicament again, & was given the number of a company which specialized in erecting rails for people with mobility problems, the work was completed with the costs paid for by me. (email excerpt).

However, there is a small segment at the beginning of the path that is directly in front of another person's property and handrails could not be fitted there. So, Arleen still needs someone to help her from her front door to the start of the handrails so that she can get to the garage and on to the wheelchair. Her solution to that is to move into a bungalow in order to be able to use the wheelchair without help and not to depend on someone being

there when she wants to go out. If she moves into a bungalow she will have her own drive, with the car parked in front of her front door, and the wheelchair will be stored in an accessible location. But doing so will mean that she will need to give up a piece of independence (being near the town centre) to gain another piece of independence (being able to access the wheelchair and her car without help), echoing the constant negotiation between desires and needs. The location of her current flat is very convenient as she can easily get taxis into town or go for walks in her wheelchair at the nearby park. Moving into a bungalow might mean that she will need to live a bit further out of town.

Dikaios: And then you also said buying a house is a necessity to make your life easier, and that's coming out as an important theme as well, to make life easier. How do you mean life is easier, or what makes life easier for you?

Arleen: So, I will be able to get to the car on my own, and drive around. It won't just be easier, it will be nicer, it will be more pleasurable. I will have a garden to sit it [in]. And [now] everytime the post comes I got to go up and down the stairs...

A good life for Arleen would be a life in which she would be able, among other activities, to 1) get the post without having to go up and down stairs, and 2) get to the car or the power wheelchair even if she is by herself. In other words, a good life is a life where the environment is modified in order to eliminate obstacles, such as stairs and slippery pathways, so that she can carry out the activities she wants to.

Disability has been conceptualised in many ways, including as a social construct, as something missing or something in excess in a body or

in terms of barriers and facilitators in the environment (Shakespeare, 2006). Disability can be all these but much more as well; it refers to an individual's life, not just a body and not just an abstract society, but a specific life that one person leads, within a specific context (Siebers, 2008). Stone (2013) describes disability as a transactional condition, emerging at the intersection of people and their contexts, whether physical, cultural, political or of any other nature.

Disability, in other words, does not reside in an individual's body as something that someone may have, but is defined as the loss of opportunity resulting from discrimination (p.96).

Rather than being an individual characteristic, located in the body, disability emerges when the match between people and their environment is not ideal, thus leading to restrictions in participation. The body is of course an important dimension. However, the body alone is not enough to construct a disabling situation, as Arleen's story illustrates. Some time after the incident with the handrails, she contacted again the social services. This time her request was about a door control system. The following excerpt is from an email she sent me:

Once again I contacted Social Services, for a door entry system, as by the time I could safely go downstairs, any callers would have left, they accessed [assessed] that as I had no falls on the stairs at this time they could not help, but if my circumstances/condition changed. (I can only assume they meant break my neck while rushing to answer the door) I paid for my own.

Once again, Arleen took action to modify the environment around her. While it is not possible to know the actual response of the social services, or

their rationale for their response, Arleen felt her needs were not recognised. She was either too unhealthy for the handrails, or too healthy for the door control system. Something happened in that interaction she had with the services that made her feel she was seen not only as a disabled person but as someone who could not be helped. In a way the speed of her progressing condition was not in match with the services offered; too quick for the handrails, too slow for the door control system.

Arleen, however, was not a novice in the health and social care system. She knew how it worked, as she was part of it for several years, having worked both in a clinical and an administrative position. This gave her what she described as an advantageous vantage point, knowing what she could expect from the system and what she would be better off pursuing by herself to address some of the disabling situations she was facing. More importantly, she refused to let herself be disabled by processes that could be controlled.

Arleen: I think I got to be me first and my illness second. So, I just live life as I would if I was totally fit. But with the realisation that I can't move as fast or whatever. So...as for buying a house, that's a necessity. And it will make my life easier. I still try to be as normal as possible for my partner, so he doesn't lose out on his life as well. Because, he can't live his life through me, so.

Dikaios: This realisation that you say, that it is you, with the added realisation that the illness is there...how did you reach that realisation?

Arleen: It took a long time, but I am still me. People are defined by the things they do, and they are not defined because they are ill. It's like saying I cannot do this because I am a woman. I would never think that. I would never think I could not do something just because I was a woman. I just think, I think I would be really, really depressed if I wasn't

thinking I have to live my life. Like, when you texted me I peeled a bowl of potatoes because it was an extra ten minutes in my morning. So, I did something, yes. When I was first diagnosed I waited for something to happen, but life is too short. Yes, it did take some time until it clicked, but it did, and I am glad it did.

### **10.5 Reflections on interviewing as an interactive performance**

Interviewing Arleen, and analysing the transcripts, looking for emerging storylines, was not an easy process for me. Nothing seemed to be jumping out of the data and it all seemed so normal. So, I kept looking more and more trying to find out where exactly MND was hidden in between the stories Arleen was sharing about her life. During one of our interviews, sitting down in the living room, I asked: “*How do you cope with the stairs now?*”. I was wondering whether Arleen had any problems with going up and down the stairs, as I, mistakenly, thought that this had been reported as an issue in a previous interview. Arleen interpreted the question in a different way, as reflected in her answer: “*Oh, the postman has only just been so I didn’t go downstairs*”.

I suddenly realised that my question could be heard as a judgment. Perhaps the timing of the question, perhaps the way I asked it, perhaps for no reason at all, a connection was made between the uncollected mail remaining at the foot of the steps and my question. I quickly explained that I was not referring to the mail: “*No, I meant, because before you said that going from the property to the garage through the stairs, the steps, it was a bit tricky*”.

Yet, is that what I meant? Could it be that I could not believe that Arleen's life was as normal as she was describing it and I was constantly seeking confirmation of the presence of the progressing, disabling, terminal disease I believed was somewhere there? Even the very phrasing of this question, repeated from my field notes, is revealing. What was it that I believed was there, and where was 'there'? Despite my personal experiences and my research training, perhaps I was confused between the different realities signified by the concept of a biomedical entity, a disease, and by the experience of it in somebody's life. I caught myself localising MND into somebody's life, ascribing meanings that were not there. It was as if I insisted that something must be wrong, normality in the face of MND is not normal. So, I kept looking for something that I had constructed in my head; I was looking for an experience as misrecognised as Arleen felt when she was thought of as *someone with her condition*.

This incident made me particularly aware of my role as an interlocutor in the interviews. As Alsaker, Bongaard and Josephsson (2009) state, the way researchers position themselves during interviews, the types of questions they ask, and how they ask these questions, can have an effect on how participants tell their story, or what story they share. During the interview process, I was consciously aware that normality is not a universal standard, but consists of individualised and localised practices, unique to specific people rather than anonymous aggregates.

Annemarie Mol (2006a) referred to atherosclerosis as being more than one but less than many. This can be applied to any disease, including

MND. MND is enacted through many practices, in many different settings, by different people. It involves a body, and a society in dialogue between them (Shakespeare, 2006). But at the same time it all comes together in the life of a specific person and all the seemingly heterogeneous practices become part of this life, in all their inconsistencies. It is only within the context of a particular life, within a local context, that a narrative tracing the meaning of a disease into a person's life can be constructed.

### **10.6 Chapter summary**

In her narrative, Arleen enacts her guiding belief of "*I got to be me first and my illness second*". MND, and the disabling situations associated with it, are not a given reality for Arleen, but a challenge. Through the choices she makes she seeks to transcend disability and maintain a sense of who she is. For Arleen, transcending disability does not mean denying its presence or limiting her activities. It is about modifying the environment in such a way so that it does not pose obstacles to her participation in her repertoire of everyday activities. Arleen tries to construct a life that she enjoys and a self that she desires, despite, or because, of the unpredictability of the prognosis of MND.

## **11. The ab/normality of living with MND**

In this chapter, I present how the findings offer a response to the objectives of this study, bridging the gaps in knowledge identified in chapter 1. My interviews with the participants over 25 months produced rich data and afforded me an opportunity to:

1. Explore the experiences of daily life from the standpoint of seven people (three couples and one person participating by herself) living with MND in Wales.
2. Foreground how these people make sense of their life in the context of living with MND.

I started the study with some knowledge of MND, both experiential through my personal involvement, and scientific through my professional training. I was, however, aware that each individual was unique and I was consciously aware of the need to be open to that uniqueness throughout the research process. Sometimes I had to remind myself to hold back my assumptions and listen to the unfolding story, as happened with Arleen (see paragraph 10.5), in order to construct a narrative illustrating how a particular person, or couple, experienced MND and made sense of their life.

In constructing their own unique stories, participants demonstrated elements of all three storylines (restitution, chaos and quest) described by Frank (1995). The focus of most participants was on living as best as they could in the presence of MND. Despite the fact that all participants were indeed unique in their experiences of MND and how they made sense of

their life, there were some commonalities. I am reluctant to call these commonalities themes, as the experiences people described do not form neat groupings. Neither are they grand, explanatory narratives, pulling the different, divergent stories together into a harmonious whole. They are ways of understanding life with MND, each of them true to different participants for different reasons, and to different degrees. These different ways may not always be combined together into a model illustrating the experience of living with MND, but they can offer a glimpse into how some people experience life with a progressive, degenerative, incurable, and terminal disease.

I have called the ways that the participants in this study made sense of life with MND “*constructing a subject or producing an object*” and “*whatever works; enacting normality*”. They are not exclusive and are among many other different practices that the research participants enacted. If, as Mattingly (2009) reflected, “*we find ourselves in the unsure position of being situated among several possible plots, and all at once*” (p.246), the only certainty is the normality of ambiguity. The following paragraphs illustrate how research participants experienced MND in their everyday life, how they made sense of themselves and what was happening to them and how they engaged in those practices that would lead to what they perceived to be a good outcome, in the face of a challenging present and an unknown future.

### 11.1 Constructing a subject, or producing an object

*“It got to be me first and my illness second”* said Arleen in one of our interviews. She wanted to be Arleen, a woman who happens to have MND, rather than a representative MND case. All participants echoed that desire to retain an identity that was not subsumed by MND. Sometimes people talked about how they were trying to maintain that identity; some other times they were discussing how they were working towards establishing it; and some other times they were talking about how they felt they had lost it.

Whether it is because of an altered physical body, a changed body image or changed interactions with one’s surroundings, the onset of serious illness can lead to a re-evaluation of how people relate to themselves and what their desired self looks like, and how it can be achieved. The people whose narratives are presented in this thesis experienced this in varying degrees.

Some of them were clear that MND came second, after an identity that was solidified prior to the emergence of their illness. When Arleen said that she puts herself first and her illness second, she did not mean this literally; she did not experience being two different people, one with and one without MND, always giving precedence to her healthy version. What was at stake for her was the maintenance of an identity that while it included MND was not dominated by it.

Several authors have explored the identity changes precipitated by the onset of illness. Drawing from her personal experience of living with cancer, essayist Susan Sontag (1991) wrote about the difficulty in

maintaining one's identity when faced with negative metaphors about illness, where disease can be seen as punishment, or as dirty or ugly. Also motivated by his encounter with cancer, literary critic Anatole Broyard (1992) foregrounded the positive metaphors of illness that can act in a liberating way, allowing a sharpening of one's focus only on those things that matter. For many people, their experiences lie somewhere in between, while they try to make sense of what is going on.

Pavey, Allen-Collinson and Pavey (2013) discussed how *“developing MND means learning to live with irremovable loss as a constant reality, mourning for those losses and accepting life in the present”* (p.5.1). They refer to this process as ‘chronic sadness’, echoing ‘chronic sorrow’, a term introduced by Olshansky (1962). The research participants in the present study of course did not experience MND as a pleasant development in their lives. They did have to learn to live with continuous changes, even irrevocable losses some times, as Pavey, Allen-Collinson and Pavey (2013) discuss. However, saying that they lived in a state of ‘chronic sadness’ would not be accurate, and perhaps would not be fair to their experiences.

For the seven people who participated in this study MND was a major part of their lives, but still only a part. While they were in varying degrees angered at the diagnosis and at times felt hopeless, afraid, confused, exhausted or sad, other times they were happy, optimistic, and grateful for what they had and what they could enjoy. Below the surface of their daily

interactions, it was just as likely to be an undercurrent of sadness, anger or determination.

Diverse as the experiences of living with MND were, power was central in participants' stories. Access to power, whether in the form of social, cultural health (Dubbin, Shang, & Shim, 2013), knowledge or financial capital influenced the construction of participants as subjects, i.e. bearers of knowledge/power within a specific discourse. The participants were oscillating between being subjects and being objects and between exercising power and being the locus where power was exercised; they were at the same time both actors and sufferers (Arendt, 1998), or in other words both active and passive agents in their life. The boundaries between being a subject and being an object were fluid and negotiable.

Subjectivity was not a given but it was actively constructed by the participants. In their own ways, participants were seeking to create their own versions of Campbell's (1968) 'hero', and take a central role on how they lived their life and managed MND. Becoming a subject, or hero, however, with power to take those choices that felt right in the context of their life, was a precarious process that did not always have the anticipated results. All participants were in varying degrees caught in the constant oscillation between being a subject and being an object, where people can be both actors and sufferers. Dave and Marion for example, in subjecting themselves to the operations of the health and social care systems felt they had lost their power and had become objects. They felt that decisions on what technologies to be subjected to were beyond their control. They spent

what was to be their last Christmas together, although of course they had no way of knowing this, in a house so full with medical equipment and right in the middle of adaptations that were going on for several months, that their grandchildren could not stay with them.

Returning to Arleen, in one of our last interviews she shared that she had decided to donate her body for neurological research (Text box 11.1).

**Text box 11.1 Arleen's end of life bequest***My end of life gift*

I recently received a condensed result synopsis from the “XXX” research project.

This together with reading the article, “XXX” of XXX, led me to reflect on when I discussed my wishes with family & loved ones, I was amazed that my wish to donate my body to research raised the most questions, & even opposition.

It took many hours of carefully selected debate to reassure my family that my body would be treated with respect & hopefully it would contribute something positive, & help gain an insight to this disease.

This was not my only hurdle; I also had to find a centre to “take me” as there are geographical and other practical matters to consider.

The XXX centre @ the XXX institution, accepted me.

Consent forms witnessed, Family Spouse/Partner acknowledgement signed, I requested 3 copies;

- 1 for my Hospital records.
- 1 for my GP records
- 1 for my Solicitor.

I also asked for an electronic copy which I emailed to family and loved ones.

I realise that this is a very sensitive, personal & possibly even a taboo subject, but I would not wish this disease on anyone, so for me this is my *“Donation” to a world free from MND.*

To do so she had to assume power not only over her body, but over the many people that surrounded her; her family, her partner, and her friends, and also institutions such as the health and legal systems. She was at the same time a subject, asserting her power and control over herself, and an object, that would be subjected to the operations of scientific research. Through this act she thought that although powerless herself to help find a cure for MND, she could help other people work toward a cure by donating her body to science. Her body, marked by MND, was her power.

## **11.2 Whatever works; Enacting normality**

Despite, or perhaps of, the ambiguity they were experiencing, the people whose stories are presented in this thesis wanted to live a normal life. They did not refer to an abstract notion of normality, but a tangible, aesthetic experience that encompassed MND but was not necessarily governed by it. They all had different perceptions of what this life would look like and varied access to power to achieve that normality through a variety of means. That normality was a *good* they were working toward, but it was not defined a priori but only within the ever-changing context of the participants' life. Life might be subjunctive, with uncertain endings to different stories, but participants in this study were trying out different means to different endings.

Whether normality referred to being able to go the gym every morning (Rhian and Gwyn), take a shower in the bathroom, rather than having to strip wash in the kitchen (Dave and Marion), plan for the future

(Maggie and Gareth), or retain a sense of autonomy (Arleen), research participants mobilised those practices that were available to them to try and reach that normality or justify its unattainability. Often their perceptions of normality changed and procedures such as a PEG would move from being abnormal and thus undesired, to being normal and desired.

They had to work toward that normality within a context of what they experienced as abnormality. Their body, or that of their partners, was changing as a result of the MND. As muscles were getting weaker, activities such as talking, eating, walking, writing, and driving became difficult and had to be modified somehow. More than the physical dimension of the body, their experience of having a body was changing and sometimes this was further complicated by having to undergo invasive processes such as the PEG. That abnormality presented itself as a new condition that people had to live with and make sense of.

The abnormality of the situation extended beyond the body. Research participants had to learn to navigate new environments, such as health and social services, and use devices such as lightwriters, PEG feeds, and power wheelchairs. Some of the participants had to adjust to having professional carers coming into their homes a few times a day, and they all had to deal with planning for outings by, for example, checking accessibility of different locations.

De Certeau (1988) argues that people implement different 'strategies' and 'tactics' in their efforts to 'make do', or enact everyday life. What is at stake for people is, de Certeau (1988) argues, their autonomy and

power to engage in those practices that they deem to be desirable, or needed.

Strategies for de Certeau (1988) refer to

The calculation (or manipulation) of power relationships that becomes possible as soon as a subject with will and power (...) can be isolated. It postulates a place that can be delimited as its own and serve as a base from which relations with an exteriority (...) can be managed (p.36).

Conversely, tactics are determined, for de Certeau, by “*the absence of a proper locus*” (de Certeau, 1988, p.37). Mobilising tactics requires use of somebody else’s space and power in a subversive way toward achieving autonomy.

The narratives presented in this thesis point to something else, however. They point to practices that have elements of both strategies and tactics, but are neither. While their everyday life was localised in a particular locus, a house, participants in this study had to *borrow* power from other people, notably health and social care professionals. They tried to enact normality through small actions that often were not planned and did not aim at appropriation or manipulation of power, but can rather be viewed as experiments. These were experiments in normality and like all experiments some were successful and some were not.

Experiments often take place in a laboratory. For Latour and Woolgar (1986) who carried out an anthropological study in its culture, the laboratory is a space where scientific facts are constructed, through many mechanisms, including the production of artefacts, such as articles, social interactions, and the manipulation of objects. The types of construction that were of interest to the participants of this study did not take place in a

science laboratory. Mattingly (2013) introduced the metaphor of a moral laboratory as an imaginary illustrating how people try to create a normal life for themselves and for those they care about. The moral laboratory for the participants of this study was the locus of their everyday life itself and normality was what they were trying to achieve.

In these experiments, people living with MND were both researchers and researched upon, subjecting themselves to different practices and measuring the outcomes against their notion of normality. They were both actors and sufferers, to use Arendt's (1998) terms, subjecting themselves to different procedures, or letting themselves to be subjected. The outcome was never certain. Will the new silky sheets, for example, help Dave to change position in bed, or will they be so slippery he cannot support himself? Will the hairclip hold the PEG tube in place, or will Rhian and Gwyn face another morning of cleaning spilled food on the bed linen and carpet? Or perhaps, will their action jeopardise their relationships with healthcare professionals? Will having a power wheelchair enable Arleen to go out by herself?

These experiments were often not planned in advance, but were developed as particular problems arose. Borrowing from Criado and Callén (2013), these experiments were the product of a vulnerability of established practices. In other words, something was not working right and had to be modified. When Rhian and Gwyn realised their new PEG connector did not work as it was supposed to, leaking food on the beddings and on to the carpet, they were not sure what to do to fix this problem. What they did

know however was that this was a problem and it needed fixing. Their first reaction was to seek advice from a healthcare professional that had expertise in dealing with issues such as the one they were facing. When a solution was not forthcoming, they decided to see if they could address the problem by themselves. Using a piece of cord and a hairclip they improvised a solution that worked. They had no way of knowing how the story would turn out; their experiment aimed to establish efficacy of their method. As things turned out, a few months later they decided to change again. While the cord and the hairclip were effective, they required Gwyn to put them in place together every evening and remove them every morning. Eventually, they decided to use a different connector for the PEG feed, one that was made of soft plastic and this eventually addressed the problem in a more satisfactory way.

Research participants had to experiment with everyday activities and sometimes modify them. Echoing Mattingly's (2013) reflection on the moral laboratory, these modifications did not happen somewhere outside of everyday life, in a space reserved for people with MND, but rather "*they were accomplished as the expected or the normative, becomes subject to experiment*" (p. 22). It was a notion of normality, not defined by illness, but shaped by everyday life and all its exigencies with which participants in this study experimented. Their experiments were about being able to go for a walk, eat, participate in social gatherings or be able to share a bed with one's partner.

The narratives presented in this thesis are unique to the participants. The uniqueness and the importance of these narratives do not lie in the fact that these people lived with MND, but on the ways that specific people make sense of illness and mobilise different practices to enact normality on their everyday life in the face of a progressive, degenerative, incurable, and terminal disease. These narratives are not unique because of the presence of MND, but because of the uniqueness of the individuals who embody them.

### **11.3 Chapter summary**

The people who participated in this study enacted many stories and the narratives produced reflect this diversity. Research participants were trying to construct a notion of normality in their everyday life, in the midst of what were sometimes perceived as abnormal circumstances. Using the imaginary of a laboratory, in this case the moral laboratory of everyday life, the participants made sense of life with MND as an experiment, seeking and trying out different solutions to the challenges they were facing, with the ultimate goal to enact normality. In doing so, participants were in a constant oscillation between using power and being subjected to power; this was illustrated by their various choices and actions.

## **12. Conclusions**

In chapters 7-10 I presented the four unique narratives of the research participants and in chapter 11 I discussed how these narratives offer a response to the objectives of this study. In this concluding chapter I present the main contribution of this thesis to knowledge development. I discuss the implications of the findings for healthcare practice, focusing on the importance of experiential knowledge and of everyday life, as these were highlighted through the narratives.

In this chapter, I also return to the methodology and discuss how it was developed in the course of the study. I discuss the implications of this study for the advancement of qualitative research methodologies and the implications for future research. The chapter concludes with a discussion of some of the limitations of the study.

### **12.1 Contribution to knowledge development**

The findings of this study contribute to ongoing discussions in disability studies, in particular in relation to subjectivity. The four narratives presented provide rich information on how the participants exercised power, or were subjected to power. In particular, the research reported in this thesis produced findings that contribute to the development of knowledge in the following areas:

1. This thesis demonstrated that people living with MND may develop experiential knowledge that can be applied in their local contexts.

2. This thesis demonstrated that people living with MND can try out different ways, guided by their experiential knowledge, to construct a life they feel is worth living.
3. This thesis demonstrated that living with an illness can be a fundamentally intersubjective experience.
4. Methodologically, this thesis developed and implemented joint interviews as a data collection method sensitive to intersubjective experiences.

## **12.2 Implications for healthcare practice**

The main implications of the findings for healthcare practice are the following:

1. Exploring and respecting the practical knowledge that people with MND develop, can offer insights on appropriate care and potentially lead to more effective care that can respond to the unique needs experienced by specific people in their local contexts.
2. It is important to offer integrated health and social care services for people with MND because these two areas intertwine. It is significant for the quality of care that people MND receive that the various professionals involved in their care collaborate closely.
3. Assessment practices in healthcare need to take into account the challenges faced by people with MND in their everyday life, in their local contexts.

The intention to effect some change that can be recognised as good lies at the heart of healthcare practices. This good can pertain to a quick discharge,

a speedy rehabilitation process, a higher score in a certain assessment or the prolongation of life. People enter the healthcare process with a story about a particular complaint and how it affects their life, often leading to isolation of the problem, diagnosis, and treatment of the specific condition (Latimer, 2008). This presents several challenges, as Annemarie Mol (2006b) discussed. Questions such as, what is a good life, or what to do when health or function cannot be restored or maintained, do not have a definitive answer. The *good* desired by people living with an illness cannot be defined a priori, but only within the context of their life. MND, being an incurable and progressive disease, demands a healthcare that understands the specific challenges faced by people who live with this disease and works towards a notion of good, keeping in mind Mol's (2006b) question "*which good should we strive after, if health is beyond the horizon?*" (p.406).

Research has shown that people living with ongoing illness or disability and healthcare professionals often have different perspectives regarding care and treatment options (Emanuel et al., 1996; Montgomery & Fahey, 2001; Slevin et al., 1990; van der Waal, Capsarie, & Lako, 1996). It is important that a common ground for understanding is established so that health professionals and people living with an illness refer to the same thing when they talk about right decisions regarding care or improving quality of life.

This study did not aim to explore either the good life, or healthcare experiences, but these were often part of the experiences about which participants wanted to talk. Naturally, participants reported both positive

and negative experiences and some issues were shared by all of the participants (for example, delay of services to respond or limited knowledge on MND by healthcare professionals), but the way these were interpreted by each participant was different. Two main areas of particular relevance to healthcare that emerged from the narratives are the importance of experiential knowledge and the importance of everyday life.

### *12.2.1 Importance of experiential knowledge*

Healthcare can be viewed as a

Knowledge-based system. It draws on different types of knowledge- scientific knowledge about biological processes, epidemiological knowledge about patterns of disease and risk factors, and clinical knowledge about how to treat a medical problem (Ziebland & Coulter, 2013, p.1).

As Ziebland and Coulter (2013) argue however, there is also a fourth type of knowledge, which concerns how people themselves experience illness. This type of knowledge refers to the knowledge developed through living with an illness. Healthcare professionals can access this knowledge only through getting to know how people “*care and repair*”, as Criado and Callén (2013, p.1) put it, i.e. how they deal with problems as they arise in their daily life. Participants in this study, as was illustrated through the narratives, had developed various solutions to deal with problems they faced in their everyday life. From dealing with a PEG that was leaking, to navigating what often seemed an unhelpful health and social care system, participants had developed a knowledge of which solutions worked and when, and which did not.

While people with progressive, incurable conditions such as MND may not emerge at the other side of the healthcare process as *cured*, they can still try to construct a desired life, one that they feel is the right one for them and the people they care for. This requires a constant negotiation between what is medically needed and what is wanted by people in order for them to live a life that *includes* disability and continued illness (Mol, 2006b). In other words, a negotiation between the experiential knowledge of the person whose body is changing, and the more standardised, generalised scientific knowledge that can present facts and offer possibilities that guide choices. This can lead to what Dubbin, Chang and Shim (2013) called patient-centred care, which encompasses

- 1) A provider understanding the patient within his/her biopsychosocial context; 2) shared understanding of the clinical condition; and 3) sharing power and responsibility (p.113).

Such a notion of patient-centred or person-centred care calls for an effort to integrate what a body needs medically with what people think is best in the context of their life (Berg, Meulen, & van den Burg, 2001). I call this a *togetherness of perspective*. This togetherness of perspective can facilitate an understanding of the idiosyncratic regimes and practices that can facilitate the production of a tolerable present or an ideal future, in combination with biomedical practices (Ferzacca, 2000). This is especially important in the context of a progressive, incurable illness because of a perceived time pressure to construct that ideal future.

### 12.2.2 *Importance of everyday life*

Sharing stories of illness can be empowering (Frank, 1998) and lead to different ways of relating to illness, both for service providers and service users. Stories can also be viewed as an instance of ‘care of the self’ (Foucault, 1994a), in the sense that they enable individuals to provide their own interpretations of their lives and diseases. In doing so, people not only can make sense of their life, but they can also challenge existing discourses of disability and illness.

The people whose stories appear in this thesis were taking decisions, carrying out everyday activities, hoping, despairing, loving, feeling angry and feeling happy amongst many other dimensions of their lives. They demonstrated that the popular discourse of MND as a tragedy offers an inadequate explanation of the experiences of people with MND. But, why is it so important to know about these experiences and explore in more depth the different dimensions of these experiences? What is at stake?

The answer is *recognition* (Fraser, 2000). Writing about the feeling of belonging and feeling at home, anthropologist Michael Jackson (2012) stated that “*I guess home is somewhere where you’re recognised*” (p.91). If people only feel at-home when they are recognised for who they are as individuals rather than representatives of a particular group, what does it mean for people with progressive, incurable conditions to have their experiences reduced to a discourse of loss, tragedy, and catastrophe? Through an autoethnographical article about her mother, Taylor (2010) offered a compelling account of the misrecognition afforded to people with

dementia. Misrecognition constructs people as impersonalised objects or passive agents in their lives, reducing their experience to loss and tragedy, not only for them but also for their family.

Rhian and Gwyn, Marion and Dave, Maggie and Gareth, and Arleen constructed their own version of a good, short life on a daily basis, moving from a theoretical understanding of illness to an experiential account. Their stories offered information that moves us from an abstract conceptualisation of illness to seeing illness as an aesthetic experience constructed on a daily basis through the practice of everyday life. The narratives presented in this study illustrate the importance of everyday life and the need for it to be an integral part of healthcare practices.

### **12.3 Implications for the development of research methodologies**

The main implication of this study for the development of research methodologies is that it illustrated the importance of using joint interviews to explore intersubjective experiences. This section describes the development of the methodology and its usefulness in research on illness experiences.

#### *12.3.1 Reflections on the development of the methodology*

When I was designing this study, I was aware of the need to develop a methodology that would be sensitive to the experiences of the participants. I wanted the data collection to be as open to the participants' experiences as possible, and allow them to project the story, or the versions of the story, they wanted to, in the way they wanted. Whether participants were

comfortable with, or indeed needed, long silences in order to think or rest, or whether I had to contribute stories about my own everyday life, the style of each interview was different and it was shaped through the interaction itself. The fact that I met several times with the research participants enabled us to co-construct an interview style that felt comfortable and enabled the sharing of experiences.

Of particular note is the fact that all interviews with the couples that took part in the study were joint interviews. At the beginning of the study, I had not anticipated that all couples would want to be interviewed together. Thinking that they might be reluctant to state this in front of their partner, I offered to take that decision myself on some occasions, and in the first few interviews always reminded them of that option. Upon reflecting however, it was clear that they wanted to be interviewed together as their relationship was part of the story they wanted to share. There were several practical reasons as well, which were discussed in detail in paragraphs 4.3.1.1 and 4.3.1.2. Here, I want to discuss the methodological implications of using joint interviews. In particular I focus on joint interviews as a method that allows the exploration of the intersubjective nature of illness experiences.

### *12.3.2 Illness as an intersubjective experience*

Gysels, Shipman and Higginson (2008b) argue that the choice for joint or one-to-one interviews should be guided by the aims of the study and by the preferences of the participants. In their study, they found that several of the participants (couples, in which one partner was either diagnosed with cancer

or receiving palliative care) objected to being interviewed separately, and the researcher had to accommodate this. Mattingly (1998a) discussed how in the context of joint interviews people can make sense of experiences together, because they get to interpret experiences and convey meaning not only to themselves but also to somebody else with whom they are sharing these experiences.

Human experience, Jackson (1998; 2012) reminds us, is intersubjective. Intersubjectivity refers to how humans share experiences and how they depend on each other to construct and to make sense of these experiences (Jackson, 1998; 2012). While one possible outcome of intersubjectivity might be mutual understanding, it is a process of “*trading places*” (Duranti, 2010, p.6) that exemplifies intersubjectivity. In essence, intersubjectivity is about a sharing of individuals’ lifeworlds, which is a prerequisite for human interaction (Husserl, 1922/2002). Rather than being independent, autonomous subjects with total control over their lives, people are linked to other people with emotional, biological, social, financial, and a multitude of other ties (Arendt, 1998). This network of human interconnections provides the foundations for human experience (Husserl, 1922/2002).

For the three couples who participated in this study, the experience of being in a long-term relationship was a vital part of who they were, and how they experienced MND. Antonucci, Akiyama and Takahashi, (2009) argue that couples in later life often engage in interactions in their daily life, leading to what van Nes calls ‘intertwined occupations’ (van Nes, Runge, &

Jonsson, 2009). Seeking an individual perspective from these participants would be contrary to the aims of the study, removing participants from the actual context within which they made sense of life with MND. Their experience was essentially intersubjective and data collection had to be sensitive to this and appreciate and respect this (Sakellariou, Boniface, & Brown, 2013a). Often the participants would complement each other and even sometimes prompt each other to share information.

People living with an illness and their partners might feel angry, embarrassed, tired, confused, content or any other of an endless list of emotions and feelings. Rather than being individual, these experiences are often co-constructed and shared between people. These experiences would be something else, something different if they were individual (Husserl, 1922/2002). This does not mean that people experience the same thing and interpret it in the same way, but that they have access to each other's lifeworld and they can perhaps understand it (Husserl, 1922/2002).

Disease can alter the way people relate to each other and to the world around them. In the case of a couple, disease can change relationships when, for example, one partner has to take additional responsibilities as a carer (Kleinman, 2009; 2010), or it can require new ways of structuring and sharing experiences when a person develops physical disability or speech impairment. People with MND might lose speech altogether and need to use AACD, adopting a new voice. They might also develop paralysis leading them to require assistance with many daily activities, such as going out.

Social activities such as cooking, or sharing meals with a partner can be affected (Sakellariou, Boniface, & Brown, 2013b).

In the following excerpt, Gwyn describes how Rhian, his wife, has started eating again after receiving nutrition exclusively through a PEG for several years.

Now she started to eat again. She can't eat meat, but she can eat almost anything else. I am having dinner, she wants a bit of it all the time, you know. So, she eats pasta. I was doing a poached egg with cheese in it, mash it up, she eats that. Mashed potatoes, she would eat that. Anything, like. Yesterday we had cream cake, she had that [Rhian laughs] and then she had my half of it [more laughter].

The story above was narrated by Gwyn, but Rhian actively participated in it by nodding acceptance and by laughing, sharing emotion and confirmation of the story relayed by her husband, while she was taking a rest from typing on her lightwriter. This narration by Gwyn, enriched by Rhian's participation, offered a glimpse of a daily experience shared between the couple. What could have been a story about Rhian's PEG, is seen under a new light through the joint interview; Rhian and Gwyn had not been able to share a meal for a long time, during which she was fed exclusively through a PEG.

Towards the end of the excerpt Gwyn teases Rhian by saying "*she had my half of it*" and Rhian laughs, thus communicating their joy at this newly reestablished joint experience. Rather than merely the actual fact that Rhian is reducing her dependency on the PEG and is able to eat some foods, what is communicated on this occasion is a couple's joy at being able to share a meal. The use of joint interviews in this study highlighted the various ways the participants approached their shared experience of living

with a progressive, incurable condition. Using joint interviews enabled me to access emotions, such as joy, which might have been hard to capture through a different data collection method.

#### **12.4 Implications for future research**

Naturally, this study opens up more questions than it answers. Some of the questions that stem from the findings include the following:

1. What are the mechanisms through which people develop experiential knowledge?
2. How can this experiential knowledge translate to health and social care practice?
3. How can carers be best supported in their role?
4. What are the knowledge gaps of health and social care professionals and how can they be addressed?

As an immediate first step after this study, I would like to contribute to the development of a participatory action research study aiming to produce comprehensive care guidelines for people with MND. This study demonstrated how people can create their own solutions to the problems they encounter in their everyday life with MND. A participatory action research study could further build upon this knowledge and inform guidelines for care and consequently healthcare practices.

## 12.5 Limitations

### 12.5.1 *Stories heard*

From the nature of its design, this study required a lengthy data collection phase. I met with participants several times over a period of several months to two years and meetings would sometimes last two hours or more. I acknowledge that several people could not invest this amount of time, or had other priorities and I discuss the implications of this in the following paragraph.

To enable and facilitate the participation of the people who volunteered to participate, I was open to the use of several communication methods. Furthermore, as I discussed in the methodology and findings chapters, control of the interview was shared between the research participants and myself to the extent that was possible. While it was still *my* study, conceived and written by me, I did not enter the interviews with specific questions or a very structured agenda; this might have helped participants to relax and view our meetings as occasions where they could talk about their experiences any way they wanted, rather than having to present predetermined versions of those experiences. All of the people who did participate remained in the study until its conclusion.

Morse and Field (1995) stated that the use of recording devices can make people self-conscious of what they say, thus engaging in a process of self-censorship. To overcome this potential limitation, the device selected in this study was a small, unobtrusive digital recorder with a sensitive microphone and when possible it was placed out of immediate sight. The

participants were informed every time that the interview would be recorded and that they could request for the recorder to be switched off at any time, but this was not requested by any of the participants. Because the data collection required numerous meetings and took place over a relatively long period of time, the establishment of trust enabled communication and sharing of experiences, and the presence of the digital recorder was not perceived as problematic.

The research participants were unique people in all their characteristics, including the exact diagnosis of the four people who had MND. As I discussed in paragraph 2.3, MND is an umbrella term encompassing several variants of the disease and out of coincidence each of the four participants who had MND, had been diagnosed with a different variant. Remaining consistent with the design and the conceptual framework of this study, I did not aim to develop a model or theory about living with MND, or to examine presentation or frequency of particular experiences. Rather than being a limitation, the small number of participants and the variety in the exact diagnoses was one of the strengths of the study. Concentrating on a small number of people, I was able to collect in depth data about their experiences over a relatively long period of time and accommodate the needs and wishes of the participants as to the frequency and timing of our meetings. The narratives developed from these interactions focused on how these people experienced MND and made sense of their life. These narratives are not *representative* of any population and do not treat the research participants as exemplary cases of the experience of

living with MND. The strength of this study lies in its ability to present the uniqueness of illness experiences and to illustrate specific practices of everyday life that people enact in the context of MND.

### *12.5.2 Stories unheard*

Through the interviews, and some email messages, I collected rich data that allowed the development of the narratives presented. Most of the participants could either talk, used a lightwriter device, or had a partner who could help with communication. Research participants also lived in a radius of 30 miles from where I lived, which meant that most interactions were face-to-face enabling non-verbal communication as well as verbal. One potential participant however lived by herself in a different part of the country, could not talk, and communicated through an eye-track interface. In an eye-track communication interface, the user tracks letters with her or his eye movement on a computer screen, often mounted onto a wheelchair, and selects letters through blinking or staring. This process has to be repeated for every letter, in order to write words and sentences, so it can be a time-consuming and tiring process.

Initially, wanting to facilitate the participation of all potential participants, I decided to allow data collection solely via electronic means, such as email and electronic blogs that this participant was using. In taking this decision, I was guided by the fact that there is some literature supporting the use of internet-based media to collect stories on illness experiences (for example, Mazanderani & Powell, 2013). That potential

participant invited me to use her public diary, published in her blog, where she described her daily experiences of living with MND, and she agreed to answer questions via email. The blog contained a wealth of information, usually several entries for every month, over a period of three years, charting not only the progress of MND but also the changes in her life. At first, I thought this might work, and the data might be rich enough to allow the construction of that person's narrative. This however did not happen. While the data were indeed rich, they did not seem to be amenable to narrative analysis. The data depicted one stable self that was not constructed through a dialectic relationship with an interviewer. Furthermore, the focus of the blog was to depict the progression of MND, focusing on the illness itself rather than on experiences of everyday life. I explained these reasons to the potential participant and we agreed that the data would not be included in the study.

While the story of that particular individual was still heard as it was and still is in the public domain, through this incident I became more aware of an issue I thought had been addressed through my research design, and especially the sampling and the data collection methods. That issue referred to whose stories remained unheard. The invitation to the study was circulated through a popular online forum for people with MND, and through the local branch of the MNDA. However, all participants were recruited through the MNDA route, and consequently all participants were to some extent involved with and/or supported by the MNDA. I can only speculate that many people with MND are overwhelmed with other

requirements of everyday life and taking part in a study as lengthy and time-consuming as this, toward uncertain benefits, might not be a priority. The people involved with MNDA, through regular social gatherings or other ways, were perhaps more keen to share their experiences.

Initially, I thought that a main difference would be disease progression, with people in more advanced stages being less inclined to participate, and that most of the participants would be people in the early stages of MND. However, in terms of disease progression, participants were at various stages, from having lived with MND for a few decades, to having been diagnosed a year before data collection started. Several of them had to undergo medical procedures during the data collection phase, and all of them were using some sort of equipment to address symptoms of the disease. The main difference between the people who participated and the people who did not participate was perhaps a willingness to share their experiences and an ability to invest the time to do so.

## **12.6 Revisiting my *I***

In chapter 2, I presented how I was positioned within this study. My *I* was a complex one; mainly that of a researcher, but also an occupational therapist and an occupational scientist. While these professional and academic backgrounds were not highlighted in this thesis, they were in the background and influenced what I saw and what I heard during data collection. I was also positioned in a more personal way within the study, as my mother lived with and died of MND.

Embarking on a study on experiences of living with MND was not an easy decision. I took a break of several months after my mother died before starting data collection, wanting to protect myself. Once I did start data collection however, it became obvious very quickly that everybody's experiences were unique. In a way, this reflected the objectives of the study, which rather than looking for a generic or generalisable MND experience, were focused on how specific people live with MND. The emphasis was on people's lives, of which MND was a part; sometimes a big part, but still only one part. This uniqueness in the experiences I explored meant that data collection was not a constant reminder of my mother's illness.

I am not trying to say that this study was an easy experience for me. It was not so much that I saw my mother in any one of the participants, but several of the participants' experiences resonated with mine, especially in relation to caring. After each interview I would write down my feelings, thoughts and memories as they would come to the surface of my consciousness. This helped me be aware of what I brought to data analysis, and how I saw the data.

Finishing the writing of this thesis, I feel I need to raise one more issue of importance pertaining to my decision to position myself as a researcher, without disclosing my own experience with MND. I will not repeat the reasons that led me to this decision, but I will reflect on them from where I currently am, more than a year after completion of data collection. My main concern was the wellbeing of the participants and I did not want to cause undue distress to them. I realise that had I revealed my

own experiences, the participants would have presented a different version of their story, a different truth. It would not have necessarily been richer, deeper or better, but it would have been different. Revealing my experiences in order to create a different story with the participants would have given too much information on a possible, yet unwanted, future. My own story ended with the death of my mother. Would participants be seeing their own future in my story? That was a risk I was not prepared to take.

I am confident I took the right decision. Yet, the use of this adjective does not sound appropriate in this sentence. It is probably more accurate to say that I am confident I took what I thought to be the right decision for other people. And this is what makes me uncomfortable: I took a decision to protect the wellbeing of people that I did not really know that well. I was making assumptions about what might and what might not cause them distress. However, consulting with them before taking the decision would have meant exposing them to my story, again leaving them with no choice. In other words, I could either tell them or not tell them, and the decision had to be mine. There could be no “would you like to hear my story?” as this would just introduce my story in our interactions. I decided to not tell them, and I would do the same again. But this creates several ethical dilemmas.

Taking this decision reminded me very sharply that I have control over information flow in the study. Although I wanted data collection to be as interactive as possible, some decisions, like the one about non-disclosure, were taken solely by me. This made me feel quite uncomfortable because

whatever my decision would be, I had to base it on my assumptions about the potential impact on participants.

The other ethical dilemma is about disclosure of my personal story in future publications and how this will be dealt with. While this study has been completed, the original reasons for my non-disclosure still stand. The decision I have taken at this stage, in order to be able to share the published findings with the participants, is to publish the stories of the participants as unique narratives, separately from a methodological article which will explore the ethical dilemmas surrounding non-disclosure of personal experiences in research studies.

## **12.7 Conclusion**

The aim of this thesis was to present how seven people living with MND experienced and made sense of their life. Through the narratives presented in this thesis, I tried to follow van Dongen's (1998) advice and pass on my understanding about other people's lives, acknowledging my own role in the process of both producing and telling these narratives. Borrowing from Zaner (2004), in these narratives I tried to be faithful to what the participants' stories were about. I did this by focusing on issues that were of importance to them. The narratives that were produced from my interactions with the participants illustrate the different ways that people incorporate MND in their daily lives. Central in the stories shared by all participants were a desire to maintain some sort of control in the face of an incurable disease and a desire to live a normal life. The narratives illustrate the unique

ways that participants perceived and enacted these desires in their local contexts, working towards the construction of a tolerable present, or an ideal future. In order to achieve this, they experimented with various practices of care with the ultimate goal to construct a life they could recognise as good.

As Gwyn said

But we have a system working and it works for us, innit.  
Works for us (...) As you can see, we plod along. It's better  
to smile about something than to cry about something, innit.

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## **APPENDIX A**

### **Glossary**

**Care package** refers to an array of services that are put in place in the United Kingdom, often by social and health care services, to assist people who wish to continue living in their own home despite illness or disability. In the case of MND, a care package might include daily visits from paid carers at different times of the day (up to four times daily) who can offer help with getting up from bed, dressing, washing, or getting ready to go to bed. Paid carers are not allowed to lift people and therefore, if people with MND are unable to get out of bed by themselves, they either need to install a hoist, or a friend or family member needs to be available to help them get out of bed. A care package might also include the provision of equipment, adaptations in one's residence, and access to day-care centres or respite care.

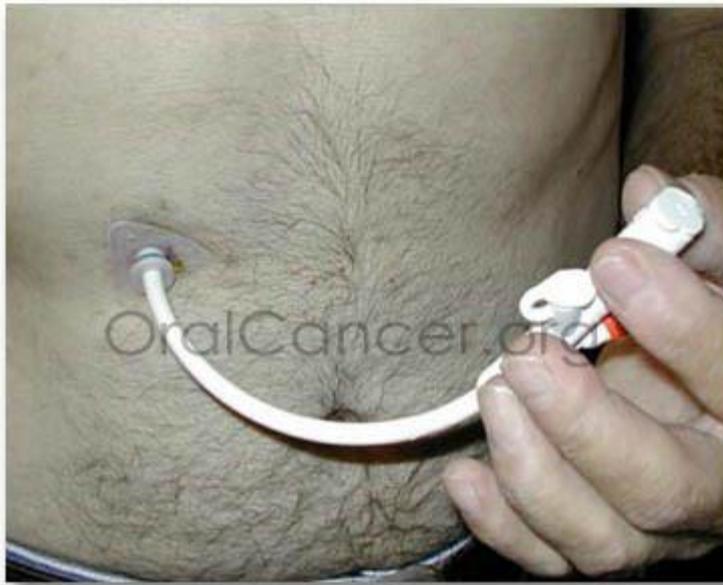
**Dysarthria** is difficulty with the production of oral speech. It is sometimes the initial presenting symptom of MND, and most people with MND will develop dysarthria to some degree. It is caused by hypertonia or hypotonia (increased and decreased muscle tone, respectively) affecting the tongue, the palate, the jaw, the larynx or the lips. Initially, dysarthria might be presented as nasal speech, as hypophonia (low speech volume, leading to whispering) or as difficulty pronouncing some consonants. Sometimes people with dysarthria are thought of as being inebriated. In advanced stages dysarthria can lead to inability to produce oral speech (Tomik & Guilloff, 2010).

**Dysphagia** is difficulty with any of the four phases of swallowing. These four phases are 1) the oral preparation where the food is formed into a bolus through chewing, 2) the oral phase where the bolus is transferred by the tongue towards the pharynx, 3) the pharyngeal phase where the bolus is directed towards the oesophagus and not allowed to enter either the larynx or the nasal cavity, and 4) the oesophageal stage where the bolus passes through the oesophagus and into the stomach. The first two phases are voluntary, while the last two are under involuntary neuromuscular control. People with bulbar onset of MND often experience dysphagia early on in the disease progress, but most people with MND will develop dysphagia to some degree. Dysphagia can initially be presented as difficulty with chewing, leakage of food or saliva from the mouth, or choking episodes. At the initial stages it can be managed with careful selection of food (for example, single consistency foods and fluid thickeners). At more advanced stages the management of dysphagia, and the associated weight loss, can be addressed by intake of food through a gastrostomy (Talbot et al., 2010).

**Gastrostomy** is an opening through the abdominal wall and into the stomach. It is often recommended to people who experience dysphagia in order to prevent weight loss. The two main ways it is performed are the percutaneous endoscopic gastrostomy (PEG) and the radiologically-inserted gastrostomy (RIG). These two terms refer to both the way gastrostomy is performed and to the tube that is inserted through the opening (gastrostomy) into the stomach. Figure A.1 shows a gastrostomy tube. Food, water, and

some medication in liquid or liquidised form can be inserted through the tube, thus ensuring the person receives adequate nutrition and hydration (Kurien et al., 2010).

**Figure A.1 Gastrostomy tube**

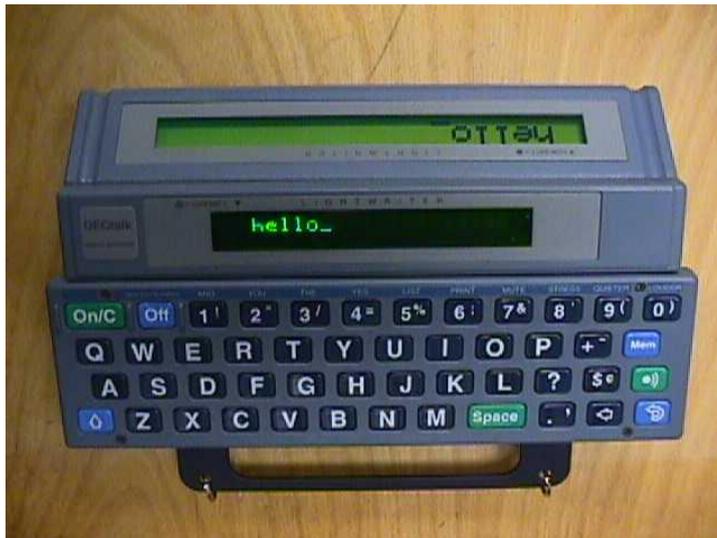


The Oral Cancer Foundation.

[http://www.oralcancerfoundation.org/dental/tube\\_feeding.htm](http://www.oralcancerfoundation.org/dental/tube_feeding.htm)

**Lightwriter<sup>TM</sup>** is a text-to-speech device that synthesises voice from text input. It looks like an electronic typewriter, with a small screen attached to a keyboard (Figure A.2). The user types each word on the keyboard and when the sentence is complete it is produced as oral speech that can be heard through the built-in speakers (Talbot et al., 2010).

**Figure A.2 Lightwriter**



University of Washington, Department of Speech & Hearing Science.

Augmentative and Alternative Communication

[http://depts.washington.edu/augcomm/images\\_aac/tc\\_lightwriter\\_display.jp](http://depts.washington.edu/augcomm/images_aac/tc_lightwriter_display.jp)

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**Motor neurone disease (MND)** is an adult onset, incurable, progressive, neurodegenerative condition that is characterised by the wasting of voluntary muscles secondary to destruction of motor neurones leading gradually to partial or complete paralysis, including loss of speech (Eisen, 2009). The different variants of the disease are presented in chapter 2. In this study, MND is used as an umbrella term referring to all variants.

**Non-invasive positive pressure ventilation (NIPPV)** is a type of mechanical ventilation used by people with MND when breathing becomes difficult. It works by

Delivering air to the chest at a higher pressure than that normally achieved during breathing. This is done by fitting a tightly sealed mask over the nose (Talbot & Marsden, 2008, p.65-66).

The NIPPV interface consists of the main device that generates the pressure, the mask that is fitted over the nose and a tube that connects the mask to the machine. While early models of NIPPV were quite noisy, more recent ones have greatly improved noise levels. There are different types of masks that people can use depending on their preferences and shape of their face. Masks can be sometimes uncomfortable and may even cause skin breakdown. Also, air leakage is a common problem and the mask often needs to be adjusted a few times in order to achieve a good fit.

**Respite care** is defined as

The temporary physical, emotional or social care of a dependent person in order to provide relief from caregiving to the primary care provider (Gilmour, 2002, p.546).

Respite care can be provided at the person's home, at a day centre or at a residential care service and it "*is based on the assumption that temporary relief from caregiving will relieve caregiver burden*" (Gilmour, 2002, p.546). Gilmour (2002) argues that the assumption that respite care offers relief from caregiving responsibilities might be too simplistic, as it does not take into account the concerns carers might have about the quality and appropriateness of services offered.

## **APPENDIX B**

### **Literature review search process**

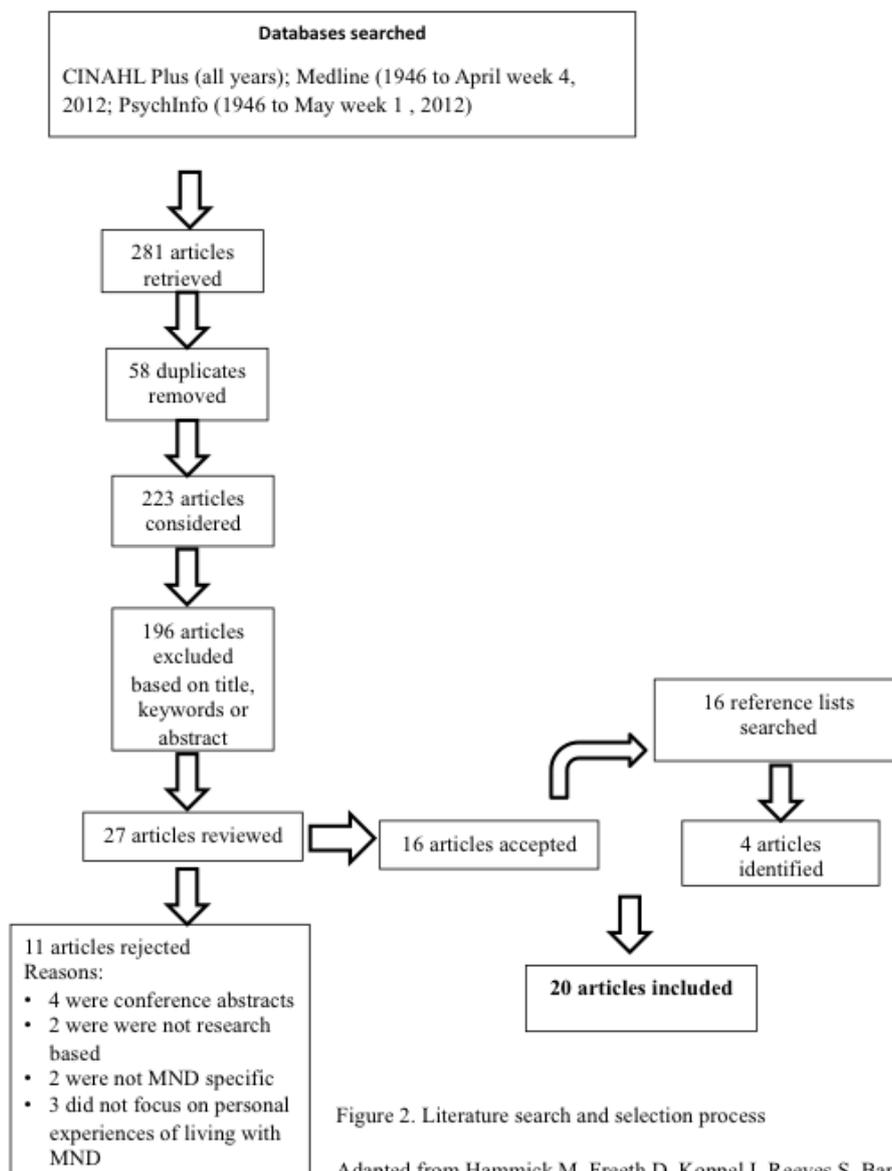


Figure 2. Literature search and selection process

Adapted from Hammick M, Freeth D, Koppel I, Reeves S, Barr H. 2007. A best evidence systematic review of interprofessional education: BEME Guide No. 9. *Med Tech* 29(8): 735–751.

## **APPENDIX C**

### **Articles identified through literature search**

<b>Author s/ Year</b>	<b>Loca tion</b>	<b>N</b>	<b>Aim</b>	<b>Design</b>	<b>Findings/ Recommendations</b>
Cobb and Hamera, 1986	USA	2	To explore the effect of the illness on participants' relationships with family, friends and the healthcare system	Case study approach	Social relationships undergo radical change. Dissatisfaction with professional services. Importance of dialogue between lay and professional perspectives on MND is highlighted
Cox, 1992	UK	10 people with MND, 10 carers and 8 occupational therapists	To explore the everyday needs of people with MND	Semi structured interviews. Exact design unclear	Importance of physical needs and practical solutions
Bolmsjö, 2001	Sweden	7 people with MND	To investigate existential issues in palliative care	Semi structured interviews. Exact design unclear.	Importance of the need to be respected and of the relevance of existential issues was highlighted
McNaughton, Light and Groszyk, 2001	USA	7 people living with MND (data from 5 were analysed)	To explore employment experiences of people who use AACD	Qualitative design, with use of a focus group (internet based discussion group)	Participants described several barriers to employment. The importance of identifying appropriate AACD was highlighted
Bolmsjö and Hermeré	Sweden	8 people living with MND and 8	To compare needs as expressed by	Qualitative design, with use of	The two groups perceived needs in different ways and

n, 2001		informal carers	people living with MND and their informal carers	interviews	had different responses to MND
Brown, 2003	UK	6 people living with MND, 6 family carers, and 9 professionals	To explore professional and lay values of care in MND	Hermeneutic phenomenology	The three groups perceived care in different ways and focused on different aspects of it. Listening to people's voice is important in order to construct the care they need
Murphy, 2004	UK	15 people living with MND and 13 communication partners (spouses, friends or relatives)	To explore perceptions of using AACD	Qualitative design, with use of video recordings, narratives, and field notes	Use of AACD was not as beneficial as anticipated. The main reasons for this were the complexity of equipment and the social distance that it creates (i.e. inflection is not possible)

Hughes, McKie et al., 2005	UK	9 people living with MND, 5 family carers and 15 professionals	To explore participants' experiences of living with MND, their experiences of services and suggestions for change	Qualitative design with use of semi structured interviews	Professionals' understanding of MND needs to be improved. People living with MND need more information regarding the process of the disease but also about therapies, management strategies and equipment
Brott, Hocking and Paddy, 2007	New Zealand	7 people living with MND	To elucidate the experience of engaging in day to day activities from the experience of people who live with MND	In depth interviews, guided by a phenomenological design	Participants experienced MND in terms of changes in participation in daily life. As the condition progressed, changes in their levels of engagement in activities resulted to the loss of valued social roles
Foley, O'Mahony and Hardiman, 2007	Ireland	5 people living with MND	To explore meaning of quality of life and explore the influence of healthcare on perceived wellbeing	In depth interviews, guided by a phenomenological design	Findings highlight the importance of faith, search for control, dignity, desire to maintain identity and family. Recommendation to consider how people

					with MND adapt to change as the disease progresses
Brown and Addington-Hall, 2008	UK	13 people living with MND	To explore participants' experiences of living with MND and how they talk about living and coping with the disease	Longitudinal narrative case studies	People experience MND in different ways, sharing storylines described as sustaining, preserving, enduring and fracturing. Stories help individuals, their families and healthcare professionals understand what it is like to live with MND
Vesey, Leslie and Exley, 2008	USA	7 people living with MND	To explore the decision making process regarding PEG	Qualitative design, with use of semi structured interviews	Participants felt that they had no control over the decision, as this was dictated by the medical condition. It is suggested that clear information about the process can increase the perceived control and involvement of patients in the process
Locock, Ziebland, & Dumelo	UK	35 people living with MND	To explore how people construct their accounts	Narrative interviews	People experience MND in different ways; as a death sentence

w, 2009			of living with MND and what they say about their life		(biographical abruption), as a major disruption to their life (disruption) or as a challenge that they try to make sense of (repair)
King, Duke, & O'Connor, 2009	Australia	25 people living with MND	To develop a model explicating the dimensions of living with MND	Grounded theory based on symbolic interactionism	People living with MND make decisions as their circumstances change. They need to adapt to an evolving situation and to diminishing physical abilities
Sundling et al., 2009	Sweden	7 people living with MND and 8 family carers	To explore the experience of non-invasive ventilation	Qualitative design, semi structured interviews	The use of ventilation was perceived favourably, after an initial period of adjustment, as it had a positive effect on daily life
Lemoignan and Ellis, 2010	Canada	9 people living with MND	To explore the decision making process regarding initiation of assisted ventilation	In depth interviews, guided by a phenomenological design	The decision making process is influenced by many factors, and participants valued autonomy in the decision making process
O'Brien et al., 2011	UK	24 people with MND and 28	To explore perspectives on the	Narrative interviews	Participants reported diagnostic delays and failure by

		carers (10 of them former)	process of diagnosis		healthcare professionals to recognise early symptoms. Delivery of diagnosis and support immediately afterwards was often unsatisfactory
Taylor, 2011	UK	13 people with MND and 10 partners	To explore the meaning of sexuality for people with MND	In depth interviews guided by a phenomenological design	Sexuality is an important, yet overlooked aspect of people's lives. None of the informants had been given the opportunity to discuss with an occupational therapist the use of assistive equipment to enable expression of sexuality
Whitehead et al., 2012	UK	24 people with MND and 18 carers	To explore the experiences of people with MND and their carers during the final stages of the disease process (and during the bereavement period)	Narrative interviews	Needs during the final stage of the disease process are not adequately met. Issues that are highlighted include: care burden, and its impact on carers and on people with MND, and limited use of advance care planning tools

## **APPENDIX D**

### **Invitation to the study**

Dikaios Sakellariou  
Department of Occupational Therapy  
School of Healthcare Studies  
Cardiff University  
Ty Dewi Sant, Heath Park Campus  
Cardiff CF14 4XN

**Email:** sakellarioud@cardiff.ac.uk

Phone numbers

**Mobile:** XXXXX

**Office:** 02920-687793

### **Invitation to participate in a study**

**Title of the study:** Living in the context of motor neurone disease

You are being invited to take part in a research study regarding experiences and perspectives of living with motor neurone disease. The aim of this study is to explore individuals' experiences and thoughts on living with motor neurone disease.

One way to learn more about experiences of living with an illness is for people to tell stories about their lives. We can do this in written format or by other means if you are experiencing difficulties talking. If you decide to take part, we will meet two or three times over a period of a few months so that you can share your experiences. It is hoped that the results will contribute to the production of knowledge on how life is experienced in the context of motor neurone disease. This knowledge may further health professionals'

understanding of motor neurone disease, thus leading to improved quality of healthcare.

You can find more detailed information in the attached information sheet. Please feel free to contact me with any questions you may have or to further discuss the implications of your participation in this study.

If you are interested in participating in the study please complete your details in the reply slip and return it using the pre-stamped envelope provided or contact me using the contact details above.

Sincerely,

Dikaios Sakellariou

## **APPENDIX E**

### **Invitation to the study (online form)**

Dikaios Sakellariou  
Department of Occupational Therapy  
School of Healthcare Studies  
Cardiff University  
Ty Dewi Sant, Heath Park Campus  
Cardiff CF14 4XN

**Email:** sakellarioud@cardiff.ac.uk

Phone number

**Office:** 02920-687793

### **Invitation to participate in a study**

**Title of the study:** Living with motor neurone disease

You are being invited to take part in a research study regarding experiences and perspectives of living with motor neurone disease. The aim of this study is to explore individuals' experiences and thoughts on living with motor neurone disease.

One way to learn more about experiences of living with an illness is for people to tell stories about their lives. We can do this in written format or by other means if you are experiencing difficulties talking. If you decide to take part, we will meet two or three times over a period of a few months so that you can share your experiences. It is hoped that the results will contribute to the production of knowledge on how life is experienced in the context of motor neurone disease. This knowledge may further health professionals'

understanding of motor neurone disease, thus leading to improved quality of healthcare.

Please feel free to contact me with any questions you may have or to further discuss the implications of your participation in this study.

If you are interested in participating in the study please let me know and I will send you the complete participant's information sheet.

Sincerely,

Dikaios Sakellariou

**APPENDIX F**

**Information Sheet**



## **Information sheet**

You are being invited to take part in a research study. You have been contacted to participate because you either have been diagnosed with motor neurone disease, or you are a partner or significant other to someone who has been diagnosed with motor neurone disease.

The title of the study is “Living in the context of motor neurone disease”. To help you decide whether or not you wish to take part, I would like to give you some information about the research and what your participation will involve. Please take your time to read this information and to discuss it with other people if you wish. If there is anything that you wish to discuss with me, please contact me; my contact details are on the last page.

### **The Researcher**

My name is Dikaios and I am conducting this research as part of studies leading to a Doctor of Philosophy (PhD) research degree award at Cardiff University, School of Healthcare Studies, Department of Occupational Therapy, where I also work as a lecturer.

### **What is the aim of this study**

The aim of this study is to explore individuals’ experiences and perspectives of life with motor neurone disease. To do this I want to listen to people’s stories about their life. Both people living with motor neurone disease and their partners or significant others are invited to participate.

### **What are the possible benefits of the study?**

It is hoped that the results will contribute to the production of knowledge on how daily life is experienced in the context of motor neurone disease. This knowledge has the potential to further health professionals’ understanding of motor neurone disease, thus leading to improved quality of healthcare. However there is no direct therapeutic benefit to you in taking part.

### **Do I have to take part?**

No, it is entirely up to you whether you would like to take part or not. If you do decide to take part, you may withdraw at any time and without explaining the reason. In all our conversations you may choose what you talk about and may choose not to answer a certain question if you do not wish to. Also if you answer a certain question or give some information

which later you decide you do not want included, you can tell me and I will delete it from the record.

### **What does the study involve?**

One way to learn more about experiences of life with an illness is for people to talk and tell stories about their lives. This is part of what is known as qualitative research, which is based on the understanding that everybody's experiences are unique and that the best way to learn more about this is to talk to people.

We will need to meet, probably two or three times over a period of a few months (approximately six (6) to nine (9) months, but this will be negotiated on an individual basis).

You can choose the place and time that you would like us to meet in. The interview will be audio- or video recorded with your permission. If you communicate through the written speech, detailed notes will be taken and written notes may be scanned onto a personal computer. Each meeting will last approximately one hour. All names of people, including your own, will be changed.

### **Will my participation in this study be kept confidential?**

Your name will not be used in any of the documents and you will be able to choose a pseudonym. Any identifying data will be altered, replaced with a code or erased and I will not pass any of the data to any third party. Following the conclusion of the research project, the audio-recorded material will be destroyed but the anonymous transcribed interviews will be kept for some time in a safe, locked place before being also destroyed.

### **What are the possible disadvantages and risks of taking part?**

I will make every effort so that you will not experience any distress throughout the interview phase. However, it is acknowledged that the issue under exploration is a sensitive one. If you feel uncomfortable with any question, you do not need to feel you have to answer it and if you wish can stop the interview. Your participation is voluntary and you have the right to stop at any time, without providing any explanation.

### **What happens if I withdraw from the research?**

You can withdraw from the study at any time without providing any justification. If during the research you decide not to continue, you can decide whether the information you have given already will be used or not in the study. Participation in this study is not related in any way to any health and social care services that you may be using and these will not be affected in any way.

**What will happen to the results of the study?**

The results of the study will form part of a thesis that I will submit to qualify for a PhD degree. In addition research papers on the topic may be published in professional journals and presented at professional conferences. You will not be identified in any report/publication unless you have given explicit consent.

**Who has reviewed the study?**

The study has been reviewed and approved by the Research Ethics Committee of the School of Healthcare Studies, Cardiff University.

**What next?**

Please feel free to contact me with any questions you may have or to further discuss the implications of your participation in this study. If you decide to take part in the study please let me know through phone, email or by returning the reply slip using the pre-stamped envelope provided.

**Contact details**

Dikaios Sakellariou  
Department of Occupational Therapy  
School of Healthcare Studies  
Cardiff University  
Ty Dewi Sant, Heath Park Campus  
Cardiff CF14 4XN

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**Phone numbers**

**Mobile:** XXXXX

**Office:** 02920-687793

## **APPENDIX G**

### **Consent form**



## PARTICIPANT CONSENT FORM

**Title of the study:** Living in the context of motor neurone disease

**Name of the researcher:** Dikaios Sakellariou

*Please initial box*

- I confirm I have read and understood the information sheet, dated ..... , for the above study and have had the opportunity to ask questions and to have had these answered to my full satisfaction.
  
- I understand that interview sessions may be audio recorded, and I consent to it.
  
- I understand that my participation is voluntary and that I am free to withdraw at anytime without giving any reason, and without any penalty whatsoever.
  
- I agree to direct quotes to be used in publications. I understand this will not breach confidentiality.
  
- I understand that my identity will not be disclosed to anyone and all information about me will be securely stored.
  
- I agree to take part in this study.

Name of participant.....  
.....

Signature ..... Date  
.....

Name of witness (Researcher) .....

Signature ..... Date  
.....

## **APPENDIX H**

### **Examples of questions**

Each initial interview with participants opened up with a variation of the following question:

*Can you please tell me what it is like to live with motor neurone disease?*

This usually led to a long narration. My subsequent questions were a combination of clarifying questions, and questions around the following issues:

1. Social network, including family, friends and neighbours.
2. Support network, which could include people mentioned under point 1 above, but also paid carers, community nurses and volunteers from the Motor Neurone Disease Association.
3. Daily life; how participants spent or would like to spend their time.
4. Special events, such as holidays and celebrations.
5. Experiences of health and social care, since these were part of several participants' stories.
6. Physical environment and access to it.
7. Communication. In particular, how this was carried out when speech impairment was present, and how it affected social interactions.
8. Caring. This was explored extensively with all participants as it was present in all stories. A specific issue around caring explored with the three couples, was how it was structured and negotiated within the couple.

The following are some sample questions on these issues:

1. Are you always aware you have MND?
2. Has MND affected your relationship with your partner? If yes, how?
3. If you need help, whom can you ask?
4. Can you tell me about the impact MND has had on your family?
5. Can you tell me about your friends and how you socialise with them?
6. Can you describe a typical day?
7. Can you tell me about a holiday you went on recently?
8. How has your perception of MND changed since being diagnosed?