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## Moving together towards participation

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**General introduction**

**1**

## **“You have to keep living your life”**

*Having been diagnosed with ALS, just over a year ago, Eric<sup>1</sup> is still not used to it. Eric had recently retired when he received the diagnosis. He still undertook some counselling jobs, but wanted to cut down on them. He had big plans for his retirement, but everything fell apart on hearing the diagnosis. It also came as a real shock to his wife, children and grandchildren. The period after the diagnosis was difficult: ‘There were so many things we had to take care of, both me and my wife. So many things you have to think about and have to process.’ He worried, for instance, about whether his wife could still live in their house after his death, he thought about his funeral, whether or not he wanted to accept ventilation and the process of becoming dependent on a voice computer. ‘I talked about all this with my GP. I said to him, I want you to help me [die] when the time is right. It was important to me to discuss that with my GP. And once I discussed it, it was out of my system, I don’t want to think about it for now.’*

*Eric noticed that many things changed and that they keep on changing: ‘You have to keep living your life. That’s what I told my wife, we have to keep living our life like we did, but now we have to take one step back every now and then. Things have to change, we need to slow down, I can’t help that (...) At the same time I can see how a lot of things have already changed. When I just look at my wife... She got very protective... Too protective (...) I’m not used to that. Rationally I get it, but emotionally I find it very hard. And it’s not just my wife, of course, with friends I have the same problem. Everybody wants to take care of you, friends and family, we were almost cuddled to death. And of course that’s very nice of them, but we almost choked in it... it was oppressing sometimes. We discussed that with our friends and agreed that we can talk about me for just 15 minutes at a time (...).’*

*The constant decrease in strength and possibilities is hard for Eric. He wants to play football with his grandson, but can’t walk without a walker or cane. He loved to have dinner parties with his wife and friends, but now he cannot sit for more than two hours. After that he is exhausted and needs to rest, often even the following day. He cannot do many things, although he still wants to. These ‘normal’ things he misses the most. ‘Mentally, there is nothing wrong, so you still want to do everything, but you can’t. This really drives me mad sometimes. It’s so frustrating. Only people with ALS really understand the meaning of term ‘powerlessness’...’ Still, Eric tries to live life to the maximum, within his possibilities. ‘Everything I can, I do. I go to the supermarket (...) I’m a member of several clubs (...) You have to do the things you still can.’ He still enjoys contact with family and friends. ‘Those are the things that make your life worth living. You don’t have a life if you only focus on the deterioration. You have to make it worthwhile, and you’re the only one who can.’*

*His constant muscle decline also affects his autonomy. Since quite recently, Eric has needed help from a professional caregiver to get out of bed and showered in the morning. ‘I still have to figure out how to*

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1 Eric is a fictitious name

*handle the constant intrusion in my life. I find that very difficult. We used to be, and to a certain level still are, very independent people. And now suddenly you have to accept strangers in your house and even in your bedroom and bathroom. That is just so unlike us.'*

Eric told me his story a couple of years ago in an interview that I held for this dissertation. He was one of the first people with amyotrophic lateral sclerosis (ALS) that I interviewed. His interview was one of the interviews that really stuck in my mind; it showed that ALS is so much more than 'just' a neuromuscular disease. Eric could articulate very well how ALS affected not only his body, but also his relationships with his wife, family, friends, his perspective on the future, his privacy, his identity; in fact, his entire life.

As a sociologist, I wanted to interpret the story of Eric, and the other patients<sup>1</sup> that I interviewed, in a broader context. How do these stories fit into the current health care system? What do these stories tell us about how society handles people with a chronic disease and disabilities? This dissertation tries to give an insight into this broader context of the illness experiences of people with neuromuscular diseases. ALS is one example of such a neuromuscular disease, but there are more examples. In this dissertation we focus, as well as on ALS, on post-polio syndrome (PPS) and facioscapulohumeral muscular dystrophy (FSHD).

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1 For the sake of readability, we decided to use the term 'patients'. What we mean by this is 'people with ALS/FSHD/PPS'.

# CONTEXT

Health and health care have been (and still are) in transformation due to changes in society and demographics. A first change is the significant shift that has taken place in the disease burden since the second half of the twentieth century. There has been a move from predominantly acute, life-threatening infectious diseases to chronic conditions (1; 2). Moreover, life expectancy is increasing, and chronic conditions are more prevalent in an ageing population (1). These changes have consequences for our health care. Nettleton (2013) summarises transition process. Due to the chronic nature of many diseases, people no longer receive treatment to cure them, but care to improve their quality of life. This implies that society no longer sees people with chronic illnesses or disabilities merely as patients, but rather as people *with* a chronic disease or disability, who want to keep participating in daily life (1) This participation has become a great good, not only in daily life, but also in health care. We will elaborate on the consequences of this transition in the context of health and health care.

## **Redefinition of health**

The shift in focus from acute diseases to chronic disease, as described above, has led to an international discussion about the definition of 'health'. The WHO defines health as 'a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity' (3). The term 'complete' wellbeing is criticised in a society with such a large number of chronic diseases (2). It seems no longer tenable or desirable to define people with chronic diseases as 'ill' or 'sick' (2). Being categorised as 'ill' or 'sick', places people in a role as being sick (the 'sick role') where they no longer have the duty, nor the right, to fully participate in society (4). This is an undesirable situation, as people with chronic diseases or disabilities are often still able to participate in society, albeit with support. This movement towards participation has been recognised by the WHO, which published the International Classification of Functioning, Disability and Health (ICF) in 2001 (5). The ICF framework focusses more on the functioning (and factors influencing the functioning) of people with chronic diseases and disabilities rather than on the biomedical aspects of the illness. Participation is important in this functioning and is described as 'involvement in a life situation' (5). The aim of participation is to improve quality of life.

This shift from 'patient' (someone who is more or less excluded from society) towards 'person' (someone who can participate) has consequences for the health care system. There has been a movement from curing patients (as this is not possible for people with a chronic disease or disability) to improving peoples' functional and psychological abilities in order to improve their quality of life (5; 6). An increasing number of health care programmes are aimed at improving people's ability to participate in daily life

activities (7). Within these health care programmes, the broader definition of health has become more central; as shown by the fact that these days rehabilitation programmes for many conditions much more often embrace a patient-centred perspective – including the mental and social well-being – rather than a purely biomedical perspective (7). In line with these developments, research into effective rehabilitation programmes has increased in recent decades (Pubmed returns 271 studies on the effectiveness of rehabilitation programmes in 1995, and 1634 in 2015).

### **Participation as goal in rehabilitation**

Nowadays, people with chronic diseases are no longer seen solely as patients (although this might be part of their identity), but rather as people with a disease or disability, wanting to participate in all aspects of daily life (8). This transition towards a more active role for people with a chronic disease or disability goes hand in hand with the changing role of doctors: doctors are no longer the only authority in consultations; the patient themselves also has a say in the treatment process (1). The rise of information technology facilitates this process towards a less hierarchical relationship between doctor and patient.

In light of these transformations, it is not surprising that the perspectives of patients have been increasingly acknowledged as important in health care improvement and research (9-11). More and more studies show that patient experiences can improve health care practice. A study by Visse et al. showed that taking care experiences into account can lead to opportunities for humanisation in health care practice. This implies a more dialogical and patient-centred means of health care (12). In addition to the improvement of health care, patient participation can also help to improve future patient involvement in (scientific) research. Teunissen et al. created a pocket guide, including criteria for good patient participation in scientific health research (11). These examples show that by listening to the needs and wishes of patients, another perspective is taken into account in health care, which can help to improve health care practice.

It is not only patient experiences with health care that can improve practice. The illness experiences, (that is, how patients experience the process of having a disease) of people with a disease or disability can also help to improve health care. Problems, general experiences with the disease and the needs of patients can help health care professionals to understand the broader impact of a disease and, correspondingly, what issues should be addressed in what way, what kind of support might be needed and how this could be arranged. An overall understanding of illness experiences is therefore of great value for health care improvement (13). In addition to studying illness, experiences can give insights into how people handle their disease; some experience their disease as part of themselves and their identity, whereas others describe the illness as being a separate part of their life (14). Insight into how people handle their diseases

and disabilities can help health care professionals to better align their care with the needs of the persons concerned (13; 15; 16).

The practice of this patient involvement in scientific research is not entirely new; it has been developing since the beginning of this century. An increasing number of studies involving patient perspectives have been made since then, in the context of rheumatism, acquired brain injury, asthma, and neuromuscular diseases, amongst others (9-11; 17). Including patient perspectives in health care research did not begin spontaneously; it needed efforts from researchers to create partnerships between the research community and patient communities (18; 19). This dissertation builds on such a partnership between researchers and patients, and more specifically patients with neuromuscular diseases. In 2006 an agenda-setting study was started to gain insight into the research priorities for patients with neuromuscular diseases (NMDs) (17; 18), improving the quality of life of people with NMDs through physical training or cognitive behavioural therapy. This qualitative research is part of that trial programme, focussing on the illness and care experiences of people with neuromuscular diseases, as so far little is known about the illness and care experiences of this patient group. This is a rather special feature of this larger trial study, as including a qualitative study in a randomised controlled trial is still relatively rare (20).

## NEUROMUSCULAR DISEASES

As this dissertation focusses on people with neuromuscular diseases (NMDs), it is relevant to elaborate on the nature of these diseases, and on the type of diseases on which we specifically focus. What these different types of NMDs have in common, is that muscle function is impaired and declines over time (21). There are many different types of NMDs which may be genetic or acquired, and may affect the motor neurons on the spinal cord, such as motor neuron disease or spinal muscular atrophy, the nerves, such as hereditary motor and sensory neuropathies, the neuromuscular junction, such as myasthenia gravis or the muscles, such as muscular dystrophy. None of the NMDs are curable, therefore rehabilitation management is the mainstay of treatment (21).

Most NMDs involve muscle weakness and fatigue, and the impact on psychosocial function may result in the avoidance of physical activity, and consequently enhance the decline in muscle function. To preserve functioning at the highest achievable level, two distinctly different therapeutic approaches can be followed: exercise therapy to maintain functional capacity, or a cognitive-behavioural approach (CBT) to stimulate an active life-style yet avoid excessive physical strain. Both eventually aim to improve daily life activities and quality of life (20; 22; 23).

There are around 600 different types of NMDs and although they all have some common characteristics, there are also great differences between the specific diagnoses, considering, for example, the age of onset, the muscles involved and the rate of progression (24) and therefore, it is not possible to study NMDs as a single group. In 2005 a national meeting of neurologists, rehabilitation physicians and patient union representatives was initiated by the Dutch Interuniversity Support Centre for Neuromuscular Research (ISNO) in the Netherlands to prioritise the neuromuscular diseases to be studied in the trial. The selection criteria were that the diseases should have a large impact on rehabilitation care, vary in age of onset, rate of progression, and pathophysiology. Taking into account the established expertise of the research groups involved, amyotrophic lateral sclerosis (ALS), post-polio syndrome (PPS) and facioscapulohumeral muscular dystrophy (FSHD) were selected from the wide spectrum of NMDs (24). These three NMDs are central in this dissertation, and therefore, a brief description of all three diseases will be given in the next section.

ALS is a disease characterised by the progressive degeneration of motor neurons in the brain and the spinal cord, leading to muscle weakness. The median age of onset of ALS is the mid-fifties. Initial manifestations are weakness of limbs, or weakness in the bulbar region causing speech abnormalities, swallowing difficulties and facial weakness. Muscle weakness progresses and the patient eventually becomes paralysed. Approximately 50% of patients die within three years of onset, usually due to respiratory failure (25). Symptoms presented in the early stages of ALS may vary, and typically result from a combination of lower motor neuron loss (atrophy, weakness) and upper neuron motor loss (spasticity, pathological reflexes) (23; 26). Muscle weakness caused by ALS can also be a result of the avoidance of physical activity, sometimes leading to cardiovascular deconditioning and disuse weakness (23; 27). If the reduced level of activity persists, further deconditioning can develop, and muscle and joint tightness may lead to contractures and pain. All these aspects of the disease hamper daily activities (23; 27).

PPS also acts on the motor neurons, but progression is very slow. PPS only occurs in people with a history of acute poliomyelitis. Poliomyelitis is an acute viral disease that attacks the motor neurones. Poliomyelitis often occurs in children under the age of five, and usually there is partial and sometimes complete recovery. However, many people with a history of poliomyelitis report late onset neuromuscular symptoms and a decline in functional abilities (20). These late symptoms are referred to as post-poliomyelitis syndrome (PPS) and include symptoms such as new or increased muscle weakness, abnormal muscle fatigability, generalised fatigue, muscle atrophy, muscle and joint pain, muscle cramps and cold intolerance (20; 28). All these symptoms negatively affect the quality of life of people with PPS. PPS symptoms usually start 30 to 40 years after the acute polio (29). After recovery from the acute stage, patients were encouraged to push themselves physically to limit the impact of their impairments upon their lives (30).

FSHD is an inherited autosomal dominant disease with an onset of symptoms between six and twenty years of age, and is included as the model of a slow progressing muscular dystrophy. It is characterised by muscle weakness in the upper limbs and face, but the lower limbs can also be affected. Severe fatigue is a frequent symptom in FSHD (31; 32). Loss of muscle strength contributes to fatigue through a lower level of physical activity (31). Fatigue and physical inactivity determine social dependency and loss of participation.

## RESEARCH AIM AND DESIGN

### **Research question**

As pointed out, studying patient perspectives of health care programmes and illness experiences enables health care professionals to understand the effort that people with disabilities and chronic diseases have to make to participate in daily life, and can help to improve health care (9; 12). The illness and care experiences have already been described for many diseases, however, so far, little is known about the illness and care experiences of people with neuromuscular diseases (NMDs). This dissertation thus aims to provide insight into these experiences of patients with NMDs. The main research question of this dissertation is therefore:

What characterises the illness and care experiences of people with neuromuscular diseases and how can these contribute to health care innovation?

### **Design**

We used a responsive evaluation approach in this dissertation, because this approach enables an understanding of multiple stakeholder perspectives, in our case the experiences of people with NMDs, and their health professionals.

#### *Responsive evaluation*

Robert Stake was the first to introduce an evaluation approach in which the user perspective was structurally embedded (33; 34): the responsive evaluation. Until then, evaluation studies were focused merely on the effectiveness of programmes and interventions, and were executed by an assessor in consultation with the funding bodies (34). Stake wanted evaluation to be meaningful for all stakeholders (34). In his opinion, there was a need for qualitative, contextualised data to reveal the situated and cultural nature of programmes. Stake drastically changed evaluation, by including multiple stakeholders (33; 35). A complete insight into the quality of a programme can only be reached by questioning all relevant stakeholders (in health care, these can be patients, health care professionals, spouses et cetera) (35). The aim of a responsive

evaluation is to understand a programme, intervention or situation from the perspective of all involved stakeholders (36), and therefore, it is not important to 'check' whether a programme ticks some boxes; it is much more important to identify different stakeholder experiences of the programme (37).

This evaluation approach was expanded by Guba and Lincoln, who emphasised the importance of not only consulting multiple stakeholders, but conducting evaluation research with the active involvement of all stakeholders (38). Abma elaborated on the responsive approach through a social-critical and hermeneutic-dialogical framework (39-41). Within such a critical framework, evaluation should actively steer towards the inclusion of marginalised voices. A measurement orientation is one-sided since only the plans and intentions of policymakers are used to judge a programme. Participants are only taken seriously for the information they are able to provide. They are approached instrumentally. This does not lead to the acceptance and implementation of evaluation data and results. Within responsive evaluation, deliberative dialogue striving towards consensus is pertinent. The evaluator is not only an anthropologist as Stake describes, but also a caring researcher trying to actively attend to marginalised voices and the balancing of power hierarchies (42). This is a value-based dimension related to social justice and democracy.

Guba and Lincoln (1989) used the term "negotiating" to denote the interactions between stakeholders. Abma and Widdershoven (2006) refer to it as 'dialogue' (43). The central features of dialogue are openness, respect, inclusion and engagement. The value of involving different stakeholders is multidimensional. First, it gives a more complete impression of the value of an intervention. Secondly, it gives all stakeholder a voice (36; 43). In many evaluation studies the opinion of the 'users' (e.g. patients, students, employees), the people whose lives are affected by the intervention, is not heard (9; 43). The users can be, or become, a marginalised group by excluding them from programme development and evaluation. In a responsive evaluation approach, the aim is to also include potentially marginalised groups, which can lead to the empowerment of patients or patient groups (42; 44). Abma & Widdershoven (2006) structured the responsive evaluation process in health research as follows:

- Creating social conditions for research
- Collecting data (e.g. by using in-depth interviews)
- Facilitating a dialogue with homogeneous stakeholder groups
- Facilitating a dialogue with heterogeneous stakeholder groups

Compared to Stakes' approach of responsive evaluation, this approach is much more focused on the dialogue within and between different stakeholder groups. The

main belief in this approach is that people's perspectives are exchanged and refined through a process where the researcher aims to generate a 'shared construction' of the perspectives of participants (12).

### *Narrative approaches*

Abma's version of responsive evaluation is grounded within narrative psychology (45-47). Narrative psychology helps to understand how people make sense of and give meaning to their own identity and life context. Subjective experiences are crucial in people's narratives. By describing their lives with an illness, people gain some distance from the events, enabling them to review and re-evaluate the situation (48). The beauty of narratives is that they are personal and social at the same time. They are personal, as they are told by an individual from their individual background, experiences, knowledge and view. They are lived experiences and therefore only experienced by the storyteller. At the same time narratives are shaped by society. The shape of the telling is formed by all the rhetorical expectations that the storyteller has been internalising (14).

When looking at *illness* narratives, a wealth of information is given by the storyteller about how a disease is experienced and handled. This is not only done explicitly, but also very much implicitly in the way people tell their story, their choice of words, the structure of the story; there is much hidden about how people experience a disease and see their role as a person living with that disease (14). By listening and reading between the lines much can be learned about the meaning people attribute to situations and how they see their own role and identity; the meaning of having a disease and how it affects their identity (or identities), the way their body has been experienced since the disease or disability, and how this influences ways of coping with the disease or disability (14). This shows that narratives provide much more than just the spoken words; they tell a multi-faceted story which can be studied and analysed. By analysing these stories, a deeper understanding can be gained about how people experience disease or disability and, correspondingly, how health care can be attuned to their needs.

### **Setting**

The research setting was formed by two research projects that were executed between 2008 and 2015. Chapters 3, 4, 5, 7 and 8 are based on the **Fitness And Cognitive Behavioural Therapies for Fatigue and Activities in Neuro-Muscular Disease** (FACTS-2-NMD) study programme. This programme consisted of three separate multicentre randomised controlled trials (RCTs) in which the effectiveness of exercise therapy and cognitive behavioural therapy on fatigue, daily activities and health-related quality of life was studied in patients with facioscapulohumeral muscular dystrophy (FSHD), post-polio syndrome (PPS) and amyotrophic lateral sclerosis (20; 22; 23).

Several university hospitals collaborated in this research programme, and with general hospitals and rehabilitation centres throughout the country. Various interventions were offered to patients in these RCTs. For each disease (FSHD, ALS and PPS) one group of patients was randomised into a cognitive behavioural therapy, one group into an exercise therapy and one received the usual care as a control group (20; 22; 23). Both the exercise therapy and the cognitive behavioural therapy had a duration of four months. The primary purpose of the FACTS-2-NMD study was to investigate the effect of both interventions in reducing fatigue and improving activities and quality of life (20; 22; 23). Outcomes were assessed quantitatively with questionnaires (for ALS, FSHD and PPS) and with physical measurements (for FSHD and PPS), such as submaximal exercise tests (20; 22; 23; 49). All outcome assessments were obtained in the three University Medical Centres which were each responsible for one of the trials; the Academic Medical Centre Amsterdam for PPS, the University Medical Centre Utrecht for ALS and the Radboud University Medical Centre Nijmegen for FSHD. In addition to these quantitative measurements, patients and health care professionals were interviewed about their experiences with the intervention. This qualitative study was performed by the department of Medical Humanities at the VU University Medical Centre and the results are included in this dissertation.

This dissertation is also based on another trial, which focused on the added value of case management to multidisciplinary ALS care (Chapter 6). This trial was a cluster randomised controlled trial (RCT) involving 43 Dutch multidisciplinary ALS teams (clusters). The aim of this trial was to improve the quality of life of the patients with ALS and to reduce the strain of the primary informal caregiver. In this trial, case management was performed at the individual patient level. During the twelve month intervention period, case management was provided by two experienced occupational therapists, specialised in ALS care and trained in client-centred practice. The case manager's role was to be attentive to the needs of the patients and to stay in close contact with the ALS care team. The case manager provided patients with all the information required to allow individual choices about how their needs would be met. The case manager started the intervention by visiting patients at home at study entry and subsequently every three months. Between visits, contact was possible by telephone, e-mail, or in writing (50). The outcomes of this cluster RCT were primarily measured through quantitative research methods; questionnaires were used, measuring the quality of life of the patients and caregiver strain of the informal caregiver (50). In addition to the quantitative study, interviews were held with patients, their informal caregivers and the health care professionals to evaluate the experiences of these patients with the case management intervention. These interviews were used as the data for this dissertation.

## RESEARCH METHODS

Both studies (ALS study and FACTS-2-NMD study) followed a responsive design, and consisted of individual in-depth interviews and focus groups. This section briefly describes the sampling, overall methods of data collection and analysis, and briefly highlights the main characteristics of both studies. Precise descriptions of the methods are provided in the individual chapters.

### **Sampling**

As described in this dissertation, patients, spouses and health care professionals were interviewed for the studies. The sampling of all these patients proceeded in a similar manner. The sampling of our respondents was determined by the sampling of the larger RCTs. All respondents were patients, spouses or health care professionals involved in one of the RCTs. This recruitment strategy had several advantages. Firstly, by recruiting patients in this way, we were certain that we only included diagnosed patients who were over 18 years of age and under the supervision of a rehabilitation specialist. In this way, we recruited patients who had experienced limitations from their disease, increasing the likelihood that they would be able to talk about their illness experiences as well as care experiences. Secondly, it was relatively easy to involve all relevant stakeholders. Experience shows that including health care professionals in a study can be difficult, due to their busy schedule, but as all health care professionals were now involved in the trial setting, they easily gave consent for the interviews, as they recognised the importance of this evaluation.

All respondents were recruited via the university medical centres involved. They were first approached by the independent investigator or the rehabilitation specialist of the specific medical centre. This principle investigator approached all patients included in the RCT to ask them to participate in the qualitative study. Patients who agreed were then called by one of the researchers from our research team. During this phone call, patients were given further information about the qualitative study and were asked to participate in an interview, or, for the ALS case manager, a trial, together with their informal caregiver. If willing to participate, an appointment was made for the interview.

The principle investigators of the university medical centres also facilitated our research team in getting contact with the health care professionals involved in the trial. This included physiotherapists and psychologists for the FACTS-2-NMD study, and case managers and rehabilitation specialists for the ALS case management trial. These health care professionals were contacted by our research team and asked to participate in an interview about their expectations and experiences of the trial. If willing to participate, an appointment was made for the interview.

## Data collection

Data was collected from patients, spouses and health care professionals, through semi-structured in-depth interviews. The duration of the interviews differed slightly between patients and professionals. Interviews with professionals lasted for 45 minutes to an hour and interviews with patients lasted for an hour to an hour and a half (with exceptions of interviews of two hours). Interview guides setting out topics were developed for both studies and different interview guides were used for the various respondents (patients, spouses and health care professionals). The interview guides were based on literature and discussed in the research team. The interviews were conducted in various places, chosen by the respondents. All interviews with patients and spouses took place at their homes. The interviews with health care professionals took place at the rehabilitation centre.

For quality reasons, the aim of the FACTS-2-NMD study was to conduct two interviews with all patients and health care professionals. The first interview focused on the motivation to take part in the trial and the expectations of the intervention. This interview was held prior to the start of the intervention. The second interview was held after finishing the intervention. This interview focused on the experience with the intervention. In both interviews the illness experiences of the patients were addressed. In the ALS case management trial, one interview was held with each patient and one (separate) interview with the informal caregiver.

For triangulation and quality purposes, various homogeneous as well as heterogeneous focus groups were held with patients, informal caregivers and health care professionals (51). Homogeneous focus groups consisted of people with shared perspectives or similar backgrounds, whereas heterogeneous focus groups consisted of people with different background or perspectives. The aim of the focus groups was to validate and deepen the findings of the interviews (51; 52). A protocol was developed for each focus group. Each focus group lasted for about two hours.

All interviews and focus groups were, after consent, audio-recorded and transcribed. All respondents (of the interviews and focus groups) received a summary of the interview/focus group by e-mail. They were asked whether they confirmed the reports or had any additional remarks to make (member check) (53). The aim of the member check was to improve the credibility of our study (52).

## Data analysis

The process of data analysis ran synchronously with the process of data collection. This is known as the iterative process of a qualitative study (54). The value of this process is that data collection can continue to the point where saturation is reached, meaning that no new codes or themes emerge from the additional interviews (55). In addition,

the iterative process enables the researcher to make minor adjustments to the topic list, which increases the quality of the interviews and the gathered data (56).

Data analysis varied according to the aim of the research project. Most studies involved an inductive thematic analysis of the interviews and focus groups (57). The analysis was performed following a three-step model of open, axial and selective coding (58). In this way, we could stay close to the text, developing a conceptual or coding scheme from there. The thematic analyses were all executed using computerised techniques and MAXqda software. One chapter presents the outcome of a narrative analysis. In this analysis, the researcher focused on the structure and wording that communicated meaning (12).

## OUTLINE

This dissertation consists of two parts. Part A presents four chapters on the *illness experiences* of people with NMDs. Chapter 2 provides an overview of what is already known about the illness experiences of people with NMDs. It provides an insight into the existing literature on illness experiences of people with ALS, FSHD and PPS. This systematic review was the starting point of the other chapters in Part A, as it showed that relatively little is known about the illness experiences of people with FSHD and PPS. Two chapters in Part A thus present the illness experiences of people with FSHD. Chapter 3 pays attention to the broad illness experiences of people with FSHD, and Chapter 4 looks at the aspect of fatigue in people with FSHD in more detail. Chapter 5 presents a narrative study of people with PPS. It gives insights into the lived experiences of people, rather than the described and explicated experiences.

Part B focuses on the *care experiences* of people with NMDs. Chapter 6 describes, from different perspectives, the value of a case manager for people with ALS. This chapter includes the perspective of the patient, the spousal caregiver and the professionals. In Chapter 7 the care experiences of the FACTS-2-NMD study are described. This chapter focuses on the group of patients with PPS and the health care professionals who performed the interventions (psychologists and physiotherapists). Part B ends with a presentation of experiences at a meta-level. Chapter 8 describes the experiences of patients with the FACTS-2-NMD study.

This dissertation ends with a general discussion that integrates the findings of the earlier chapters and answers the central research questions.

Between the chapters, three field notes are presented. These field notes show important steps for the researcher in the research process. They provide an insight into the reflection process of the researcher and show how some difficulties were experienced and handled.

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