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## Moving together towards participation

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## SUMMARY

This thesis provides insight into the illness and care experiences of people with neuromuscular diseases (NMDs). **Chapter 1** opens with the story of Eric, who was diagnosed with ALS. His story shows the broad impact this disease has on a life, as it affects his identity, his relationship with friends and family, his job, his house, the health care, et cetera. This thesis tried to put these experiences in a broader perspective. These experiences should be seen in the context of various societal transitions concerning health and health care that took place over the last couple of decades. These transitions pertain to improvements made for the benefit of general public health (such as sewers systems and vaccination programs) and developments in health care, which have led to a reduction in acute diseases and an increase of chronic diseases (for example diabetes and some forms of cancer). Because of this, there are relatively more people living with a disability than there have been in the past. These transformations have implications for our understanding of health and illness. No longer can health be defined as 'the absence of an illness', as living with a chronic illness does not mean that person is actually permanently ill. Despite their chronic illness, people are often very well able to participate in society - sometimes with use of assistive devices. In this new definition of health, societal participation is an important factor that is integrated in health care more often. An increasing amount of health care interventions focuses on the improvement of (societal) participation of people with a disability or chronic illness. Societal participation can be understood as the ability to participate in daily activities, such as the house hold, job, personal care et cetera. Rehabilitation care is a specialism that focuses precisely on this participation.

In line with the transition toward more participation, the patient's role has also changed over the last decades. The doctor is no longer seen as 'the' authority and has become less paternalistic. Instead, concepts such as 'shared decision making' have become more important. In this new relationship, experiences and values of patients have become more central in the decision making process. These experiences can be formulated in terms of care needs, but also illness experiences can be valuable to integrate in health care. Although an increasing amount of research has been executed on the illness and care experiences of people in general, relatively little is known about the illness and care experiences of people with neuromuscular diseases.

Therefore, the main research question of this thesis is:

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

This thesis consists of two parts. Part A looks into the illness experiences and Part B focuses on care experiences of people with neuromuscular diseases. The articles in this thesis are based on two studies. The first study is a multi-centred randomised controlled trial study, called the FACTS-2-NMD study. Aim of this study was to reduce fatigue in people with NMDs and increase their quality of life. The study consisted of three patient groups: one group followed a cognitive behavioural therapy (CBT), a second group followed an exercise therapy (ET) and the third group was the control group, receiving regular care. The three patient groups included in this study consisted of people with post-polio syndrome (PPS), people with facioscapulohumeral muscular dystrophy (FSHD) and people with amyotrophic lateral sclerosis (ALS). In this study, not only the patient perspective was taken into account, but also the experiences of the involved health care professionals (physiotherapists and psychologists).

The second study is called the ALS Case Manager study. In this study, patients and their informal caregivers were included. Using a case manager, this study aimed to increase ALS patients' quality of life and to reduce the care burden of their informal caregivers.

### **Part A: Illness Experiences**

Part A of this thesis starts with a review of what is already known about illness experiences of people with neuromuscular diseases, and, more specifically, people with PPS, FSHD and ALS. Most studies on NMDs concentrate on biomedical issues. However, research on illness experiences of people suffering from NMDs is increasing. In order to provide an overview of this research a systematic literature review was carried out of the existing knowledge about peoples experiences of living with a NMD. This could help professionals to better understand the care needs of people with NMDs. The analysis of the included articles shows that relatively many articles focus on the illness experiences of people with ALS. In the literature, less attention is paid to the experiences of people with PPS. And, remarkably, none of the articles looked into the illness experiences of people with FSHD. Moreover, it is noteworthy that most studies focus on the experience of the physical and mental state of people (such as pain, fatigue and other symptoms), while less attention is paid to the social consequences of being disabled. Some articles do look at concepts of illness acceptance, quality of life and, for ALS, end of life decisions. But social consequences could also include the loss of dependence, changing identity or changing relationships.

**Chapter 3** gives insight into the illness experiences of people with FSHD, as only little is known about the illness experiences of this patient group. This study shows the versatility of illness experiences of people with FSHD. Several aspects play a role in the illness experiences. These aspects can be divided into intra- and extra-individual aspects. Intra-individual aspects originate from or operate within a person. This includes aspects such as coping and lifestyle changes. Extra-individual factors concern aspects

introduced from outside the person, such as care, medicines or rehabilitation, assistive devices and the physical and social environment. The data analysis shows that the illness of people with FSHD relates to both intra- as well as extra-individual factors. Intra-individual factors concern the emotions evoked when hearing the diagnosis, integrating the disabilities into one's life, handling fatigue, wrestling with heredity and progeny, and the changing relationship with one's partner. Extra-individual aspects include reaction of the social environment to the diagnosis, postponing the use of assistive devices, and giving up work. This study shows that illness experiences reach beyond the physical aspects. A better understanding of the individual illness experiences, cognitions, and social context of people with FSHD can give health professionals tools to improve their care. It could lead to a more holistic, patient-centred health care.

**Chapter 4** elaborates on one specific aspect of illness experiences of people with FSHD: fatigue. Reason to focus on this aspect is the considerable impact of fatigue on peoples' lives (which is why it was also a central concept in the FACTS-2-NMD study). This chapter gives a detailed description of the nuances of fatigue for people with FSHD. Patients experience various types of fatigue, and describe that these types should be handled in different ways. Patients distinguish physical from mental fatigue, the former being a result of weak muscles, physical overexertion or powerlessness and stress. But patients also experience fear of getting fatigue, which is what they call mental fatigue. This mental fatigue can lead to reluctance in performing activities, as people are afraid of getting (physical) fatigue. Most of the time patients do not know the actual cause of their fatigue, which makes it hard to deal with. The experienced fatigue of patients has a large impact on their the ability to participate, being able to sustain social contacts and hence their quality of life. For this reason patients try to adopt many strategies and adapt to the constantly changing situations. Thus, the study confirms the importance of taking fatigue into account in rehabilitation care.

**Chapter 5** concerns a narrative analysis of illness experiences of people with PPS. It looks into the way in which people with PPS experience their body by analysing illness narratives. The study shows that people with PPS experience their body in different ways. This difference can be summarised as '*being* a body' versus '*having* a body'. In the latter definition, the body is experienced as an object, almost seen as separate from the 'self'. In the former definition, by contrast, the body is part of one's 'self', one's personal identity. This is called an associated body relatedness (versus a dissociated body relatedness). The narratives show that the associated and dissociated body relatedness each asks for a different health care approach. In case of a dissociated body experience there seems to be more need for focus on the body; how can this body be fixed so that the patient will feel better. In case of an associated body experience the patients seems to need a more holistic approach in health care, an approach in which aspects such as social or psychological impact of the disease should also be

addressed. This study provides insights into the behaviours, needs, and struggles of people with PPS. Studying these relational body-selves can support healthcare professionals to better understand the needs and preferences of people with PPS and aid them in finding the right approach for treatment and support.

## **Part B: Care Experiences**

Part B focuses on the care experiences of people with neuromuscular diseases by looking into three different settings. Firstly, the interventions of the FACTS-2-NMD study are evaluated. Secondly, the experiences of people with ALS and the experiences of their informal caregivers are discussed. And, finally, the experiences of participants with taking part in the FACTS-2-NMD study are elaborated upon.

In **chapter 6**, the experiences within the FACTS-2-PPS trial are discussed (this study focuses specifically on people with PPS). The effects of the interventions have also been studied in a quantitative study. This study showed that patients who followed the intervention did not improve significantly with regard to either their quality of life or fatigue compared to the control group. This held true for the exercise therapy (ET) as well as the cognitive behavioural therapy (CBT). The results of the qualitative study described in chapter 6 give further insight into the care experiences of the participants. Some patients experienced a short term enhanced endurance and a better use of energy during the day. However, in general patients did not experience a long-lasting reduction of fatigue from the CBT or ET. Mainly patients of the CBT, but also some patients of the ET described an increase of self-esteem and self-acceptance. As a result, patients were sometimes better able to perform physical activities during the day. In contrast to the CBT, the ET was in general perceived by the patients as an intensive therapy, which was difficult to fit into their daily routine. Therapists of both the CBT and the ET struggled with a low intrinsic motivation of the patients in the study. This made it sometimes difficult for the therapists to follow the protocol. Confirming the negative quantitative study outcome, the qualitative results did not demonstrate lasting effects on fatigue. Patients did, however, experience some benefits regarding self-esteem and acceptance of their disease. This study shows that it is of great importance to work with feasible interventions: they should fit the patients' needs on both a practical (fit into their daily routine) and mental (fit their need for support) level.

In **chapter 7**, the care experiences of people with ALS and their informal caregivers with the ALS case manager are described. A case manager is someone who has regular contact with the patient and his or her informal caregiver (in this study, the spouses of the patients) and provides them with advice issues such as the use of assistive devices. This trial was also studied with quantitative research methods. The outcome of this quantitative study showed neither significant increase of quality of life of patients with ALS nor a significant decrease of the care burden of the spouses. The involved

professionals did, however, get the impression that the patients and their spouses were satisfied with the intervention. Therefore, an additional qualitative study was executed to explore the experiences of patients and their spouses with the ALS case manager intervention. The results of this qualitative study show that not all patients felt the need for a case manager. Certain factors played a role in their need for a case manager. Especially patients with a large social network did not experience much additional value of the case manager. Other aspects include the adequacy of usual care, the rate of disease progression, and personal factors of patients and their spouses. Aspect of the case manager care that patients and spouses do value are the time for consultation, the house calls and the pro-active approach of the case manager. Besides, patients with ALS and caregivers appreciated the emotional support, whereas professionals did not mention the importance of emotional support by the case manager. This study shows that the context of the patient plays an important role in the care needs of people. This also shows the importance of patient-centred care, where this context can be taken into account when discussing the care needs of the patient.

In chapter 6 and 7, specific attention is paid to the care experiences of patients and other relevant stakeholders. One can also evaluate the experiences with the trial in general (instead of the specific interventions). Every year, many clinical trials are executed, which all ask for the commitment of patients (and sometimes other stakeholders). The outcomes of these trials are based on the participation of the patients; they need to execute the interventions and fill out questionnaires. Little attention is paid toward how the patients experience being a participant in such a trial and how they handle the interventions and questionnaires. **Chapter 8** presents a study that provides insight into the experiences of people with PPS, FSHD and ALS with the FACTS-2-NMD trial study. The study shows that participants are sometimes disappointed about the questionnaires: they experienced some questions as painful, patronizing or frustrating. Besides, some participants point out that they did not fill out the questionnaires entirely honest, as they were afraid that if they would, they would be excluded from the trial (e.g. questions about their ability to walk without devices). This could be relevant for the results of the quantitative study. Besides, involving patients in the design of a trial could contribute to the feasibility of a trial intervention.

In the last chapter, **chapter 9**, a reflection on the results is given from a sociological perspective. The main findings provide several general insights into the characteristics of the illness and care experiences of people with NMDs: these illness experiences affect many aspects of the lives of patients, quality of life is influenced by the interaction between intra- and extra-individual factors, experiences (and care needs) are context-bound, and care innovations require patient participation and dialogue with other stakeholders. These findings fit the 'social model' of health and illness, which is in line with the 'international classification of functioning, disability and health' (ICF). The ICF

resembles the social rather than the medical model, and it fits better with our findings than the traditional medical model. In the social model the disability (resulting from an impairment) is no longer a personal feature, but rather a disability that should be considered a relationship between a person and their environment. Almost contrary to the social model is the 'medical model', which is characterised by the metaphor of the body as a machine: if a part is broken, the doctor needs to repair it. In this medical model, an illness or disability is seen as an individual problem. From the social model, the meaning of a disease such as ALS, FSHD or PPS is not so much determined by its biomedical etiology, but rather by the consequences of the disease in a certain context. In other words, not so much the disease itself determines the disability, but rather the context in which people live, with all its inclusion and exclusion mechanisms. From this 'disability studies' perspective, a disability is seen as a complex biopsychosocial phenomenon.

The findings of this thesis and the impact of NMDs on the lives of people can be better understood from this disability studies perspective. The disability studies perspective has several characteristics. Firstly, it emphasises that a disability is life-long and affects many different aspects of peoples' lives. This fits the illness experiences described in this thesis. Besides, much research on disability studies uses a 'cross-disability approach', meaning that patients with different diagnosis are included in one research study. This approach does not depart from a specific diagnosis, as it is not so much the diagnosis that determines the experiences and needs of patients, but rather the social context. This thesis also used a cross disability perspective, as it included people with different diagnoses. Still, the findings of this thesis show that, despite the similarities, there are also some differences between the experiences of the three illnesses. In the context of a fatal disease, such as ALS, certain specific themes are of higher importance than in the context of genetic disease, such as FSHD. A last essential characteristic of the disability studies perspective is its emancipatory character. This can be explained by the way in which disability studies originally started, namely as an activist movement led by people with handicaps. The most famous slogan of the disability studies perspective is '*Nothing about us, without us!*', which reflects this emancipatory character very well. However, it turns out to be difficult to apply: in research as well as healthcare practice, often the patient voice is still overheard.

The findings of this thesis result in some practical recommendations for rehabilitation care. The concept of 'patient-centred care' has already been mentioned several times in this summary. It is a concept that is getting more popular in health care practice. Patient-centred care is defined as 'respecting people for their knowledge and understanding of their own experience, their own clinical condition, their experience of the illness and how it impacts their life'. This definition clearly shows the importance of the experiences of patients and of the meaning they give to their illness. By explicitly paying attention

to this meaning and their experiences, healthcare professionals can get an even better understanding of patients' care needs than merely enquiring as to the circumstances in which a patient lives. These illness experiences can give insight into how patients experience their body and how the illness affects their personal identity and are therefore recommended to be taken into account.