

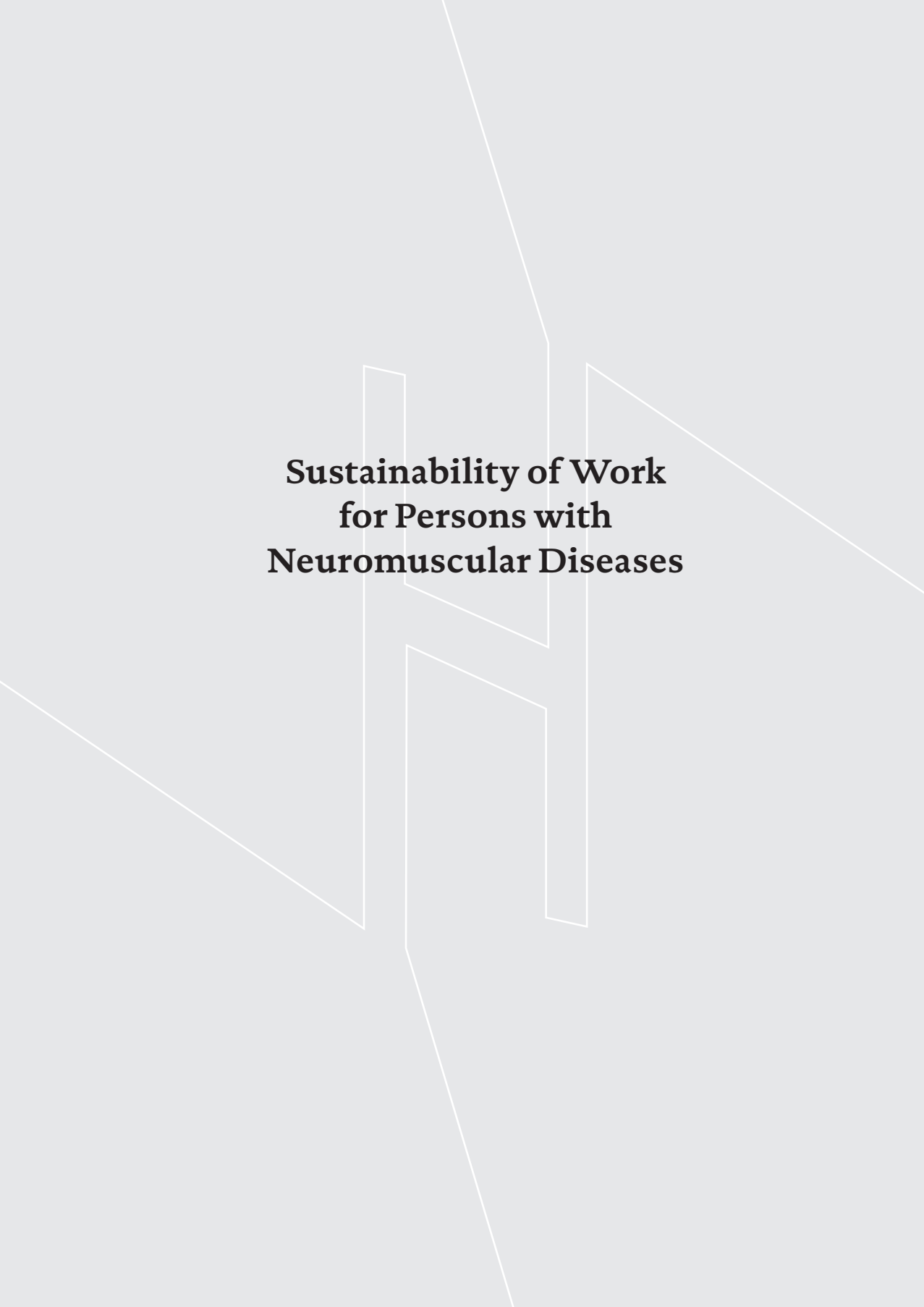
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**Sustainability of Work  
for Persons with  
Neuromuscular Diseases**

This thesis is a result of cooperation between the Radboud University Medical Centre, Scientific Institute for Quality of Health Care and the Neuromuscular Centre Nijmegen, and the HAN University of Applied Sciences, Department Occupation & Health.

With special thanks to Prof. Dr. R.A.B. Oostendorp, former first promotor.

The work presented in this thesis was financially supported by the HAN University of Applied Sciences, Faculty Health & Social Affairs and a grant from “Zestor arbeidsmarkt en opleidingsfonds hbo” The Hague, The Netherlands (grant no. PR0510-B).

The printing of this thesis was financially supported by HAN University of Applied sciences, Institute Allied Health Professionals, and Van Kuyk Uitgevers.

*Cover* Harry van Kuyk, bewerkt door Frederik de Wal  
*Lay-out* studio frederik de wal, Schelluinen  
*Print* NPN Drukkers, Breda  
*Uitgeverij* Van Kuyk Uitgevers, Ooij  
ISBN-10 978-90-800610-3-3

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CHAPTER 2 © Disability and Rehabilitation – Informa UK Ltd  
CHAPTER 3 © 2010 The Author. doi: 10.2340/16501977-0482  
© 2010 Foundation of Rehabilitation Medicine.  
ISSN 1650-1977  
CHAPTER 4 © 2012 by the American Congress of Rehabilitation  
Medicine – Elsevier  
CHAPTER 5 © Springer Science+Business Media New York 2013

**Sustainability of Work for Persons  
with Neuromuscular Diseases**

Proefschrift

ter verkrijging van de graad van doctor  
aan de Radboud Universiteit Nijmegen  
op gezag van de rector magnificus, prof. mr. S.C.J.J. Kortmann,  
volgens het besluit van het college van decanen  
in het openbaar te verdedigen op

woensdag 13 november 2013

om 12.30 uur precies

door

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Geboren op 27 april 1955  
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‘OVER DE LENGTE VAN JE LEVEN HEB JE NIETS TE ZEGGEN,  
DE BREEDTE VUL JE ZELF IN.’

HARRY VAN KUYK





# **CHAPTER I**

## **General introduction**



## Introduction

*“Occupation is as necessary to life as food and drink. Every human being should have both physical and mental occupations. All should have occupations which they enjoy, or hobbies. Sick minds, sick bodies and sick souls may be healed thru occupation”<sup>[1]</sup>.*

The aim of this dissertation is to gain insight into the extent of employment issues, and challenges persons with a slowly progressive type of Neuromuscular Disease (NMD) encounter. For people with NMD employment is not as assured as for healthy workers. The impact of NMD on employment participation and challenges in job retention has been investigated only sporadically. At the start of this dissertation, a search of medical and psychological databases on employment focused on three main types of neuromuscular diseases i.e. facioscapulo-humeral muscular dystrophy (FSHD), hereditary motor and sensory neuropathy (HMSN) and myotonic dystrophy (MD), revealed five studies:<sup>[2-6]</sup>. This small quantity of studies shows that little attention has been given to employment participation of persons with these three types of NMD. To find factors that influence job retention in persons with NMD a systematic review (SR) and a secondary analysis on data of a large cohort of persons with the above mentioned types of NMD were conducted. The impact of the professional expertise of occupational therapists, a client-centred health profession concerned with promoting health and well-being through occupation<sup>[7]</sup>, in the prevention of job loss in these specific client groups was studied. Furthermore, perceptions about gainful employment in persons with NMD who continuously have to deal with changes in their health conditions as a result of the progressive nature of the disease were studied.

### The nature of NMD and consequences for employment participation

#### *Impairments in function and structure and manifestations of neuromuscular diseases*

There are many different NMDs with a diversity of symptoms, etiology, progression, prognosis, and treatment<sup>[8]</sup>. Neuromuscular diseases can be classified anatomically, depending on their localization in the peripheral nervous system. Four categories can be distinguished<sup>[9]</sup>:

- 1) **Motor neuron disorders (MND)** such as amyotrophic lateral sclerosis (ALS) and spinal muscular atrophy (SMA) which may involve motor neurons in the spinal cord, and ultimately weaken the muscles (number 1 in figure 1);
- 2) **Motor nerve root disorders and peripheral neuropathies** such as *hereditary motor and sensory neuropathy (HMSN)\**, and Guillain Barré syndrome, that not only affect motor but also sensory nerves (number 2 in figure 1);
- 3) **Neuromuscular transmission disorders** in which the neuromuscular junction may also be directly involved in disease such as myasthenia gravis (number 3 in figure 1); and

\*The study focuses on three large groups of people with NMD printed in Italics.

- 4) **Muscle disorders** such as Duchenne muscular dystrophy, *myotonic dystrophy (MD)*, *facioscapulo-humeral muscular dystrophy (FSHD)* and limb-girdle muscular dystrophies (LGD) (number 4 in figure 1). Some of these conditions are slowly progressive and involve impairments in functions such as muscle weakness, sensory loss, pain, and fatigue in varying combinations. Some are fast progressive, such as ALS. Other types of NMD, like MD, are in fact multisystem disorders.

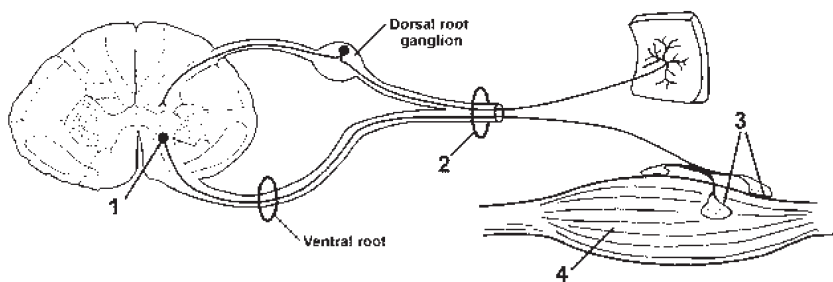


Figure 1. Anatomical classification of neuromuscular disorders <sup>[10]</sup>

### *Epidemiology*

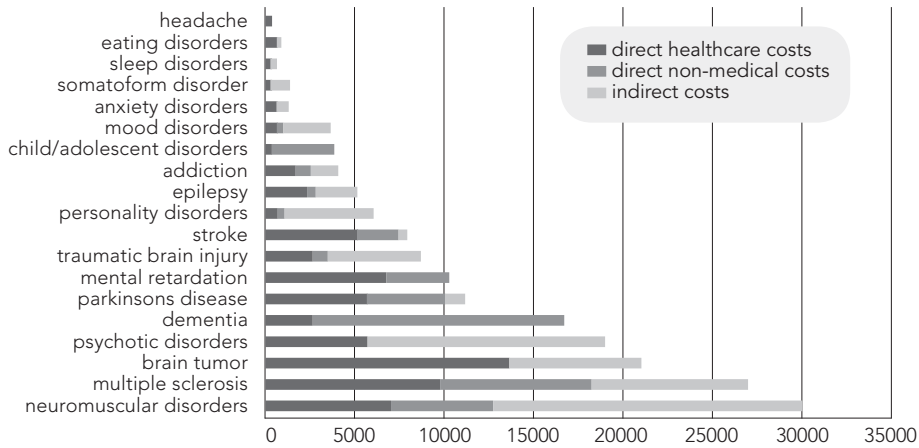
In the Netherlands the most recent investigated prevalence of FSHD is 1 in 20,000; of HMSN 1 in 1,000; and of MD 1 in 8,000 <sup>[8,9,11]</sup>. The majority of hereditary NMDs are rare. A survey of the world literature on population frequencies of children and adults with various inherited NMDs resulted in 1991 in a conservative estimate of 1 in 3500 of the population to have a disabling NMD <sup>[12]</sup>. Most likely this prevalence has increased due to the increased longevity of persons with NMDs.

### *Consequences for daily activities and employment participation*

The impairments in various functions and structures may result in a variety of limitations in activities and restrictions in participation. Due to the progression and early onset of NMD, compared to i.e. Parkinson's disease, the relative costs for health care and cure are large <sup>[13]</sup>: see figure 2.

### *Neuromuscular Diseases (NMD)*

The current study investigates the impact of the slow process of deterioration on employment participation of three different neuromuscular genetically well defined, homogeneous, and large client groups with relatively common neuromuscular disorders including: a myopathy, namely FSHD; a neuropathy, HMSN; and a multisystem disorder, MD.



**Figure 2.** Direct and indirect costs of chronic diseases <sup>[13]</sup>

#### *Facioscapulo-humeral muscular dystrophy (FSHD)*

FSHD is an autosomal dominantly inherited myopathy characterized by asymmetric, progressive weakness and wasting of muscles, starting in the face and shoulder girdle. In the majority of patients the disease is progressive, and muscles of the upper arms, foot extensors and abdomen become affected as well. Over time, the pelvic girdle and upper leg muscles, and eventually the lower arm muscles may become weak. Approximately twenty per cent of the persons carrying the gene remain mildly or non-affected (non-penetrant carriers). The rate and extent of disease progression shows large inter-individual variability, even within families in which all patients share the same mutation of their DNA. The median age of onset is thought to be around 17 years, but with a wide range, varying at the most extreme from infancy to the seventh decade <sup>[11]</sup>. The clinical variability of the disease is mainly related to the rate and extent of the progression. The pattern in which muscles become clinically affected in the course of the disease is rather uniform and recognizable.

#### *Hereditary motor and sensory neuropathy (HMSN)*

HMSN is a peripheral neuropathy characterized by slowly progressive distal wasting and sensory loss, in addition to loss of reflexes in the muscles of the legs (and occasionally involving the arms). It is the most common genetic disorder affecting the peripheral nervous system. HMSN is transmitted most often as an autosomal dominant trait. Onset is usually in the second to fourth decade of life. This condition has been divided into two subtypes, HMSN types I and II. HMSN I is associated with abnormal nerve conduction velocities and nerve hypertrophy, features not seen in HMSN II <sup>[14]</sup>.

*Myotonic dystrophy (MD)*

MD is a hereditary chronic systemic disease with wide variability in clinical expression, both within families and between families. MD is a multisystem disorder with involvement of many organs, leading to impairments in the muscular, respiratory, cardiac, central nervous, endocrine and ocular functions and structures [15,16].

Typical symptoms of the disease include myotonia and progressive loss of muscle strength, usually distal to proximal, and weakness of facial and anterior neck muscles. Other symptoms include cataracts, frontal baldness, dysarthria, fatigue and daytime somnolence. Also cognitive decline, including lower levels of concentration, lack of initiative and specific personality traits such as an avoidant personality, is related to MD [17-19]. Compared to other neuromuscular diseases, persons with MD tend to have the gravest functional disability and the greatest dependence on others for activities of daily living [20]. They also have the lowest social participation, most psychosocial problems and poorest psychosocial well-being. The educational level of persons with MD is also significantly lower than that of the FSHD and HMSN group [21].

This study is needed because:

1. Studies on employment issues for persons with slow progressive adult type of NMD are scarce.
2. Occupational therapists working in the health care system perceive barriers in supporting job retention for clients with NMD.
3. There are no published studies describing the issue of employment retention from the perspective of persons with NMD.

Below, a case study is presented to illustrate the uncertainty about job retention by a person with NMD and challenges for the occupational therapist when, in shared decision-making with the client, job retention is chosen as the main occupational therapy treatment goal. The case study demonstrates challenges in client-centred care for the occupational therapist employed in a health care setting where daily practice is influenced by legislation. Dilemmas encountered by occupational therapists in enabling clients to remain employed are mirrored in the description of Bernadette in Box 1, the starting point of this research project.

**Box 1.** Case description of a client with a progressive Neuromuscular Disease (NMD) prioritizing job retention beyond other occupational performance areas.

**The case of Bernadette**

Bernadette was 42 years of age when she was referred to the occupational therapy department by a neurologist after diagnosis with a possible neuromuscular disease. In the first stages of such a neuromuscular disease it may not be possible to make a definitive diagnosis. The reason for referral was difficulty writing due to paralysis

of the intrinsic muscles of her right dominant hand. After a short analysis, the occupational therapist decided to solve the most tangible problem directly during Bernadette's first visit by making a custom made orthosis supporting her fingers and pen to enable her to write. At the second visit, a semi-structured interview with the Canadian Occupational Performance Measure (COPM) <sup>[22]</sup> was conducted and revealed that Bernadette identified her work as a head nurse in a nursing home as her top priority. She wanted support to carry out her job as long as possible. Also some self-care activities (such as brushing her teeth and applying make-up), biking, and finding alternatives for her leisure activity (guitar playing) needed attention. The occupational therapist, under health insurance policy, was only allowed to address Bernadette's restrictions in occupational performance in regards to domestic, self-care and leisure activities, not problems related to her employment which were covered by a different jurisdiction. Support from the occupational therapist to facilitate the performance of domestic tasks, however, was not Bernadette's priority. At home, her partner and a household worker took care of all the daily household activities.

Although a clear diagnosis had not been made, her physical condition deteriorated over the following months and private costs were rising as Bernadette was trying to maintain her goal of remaining employed. Lack of assistive products became a threat to Bernadette's ability to continue working. Her supportive partner bought her a car with an automatic transmission to enable her to commute but did not investigate relevant driving rules and regulations. Without a clear prognosis, the Dutch Employee Insurances Implementing Agency (DEIIA) (in Dutch: UWV [Uitvoering Werknemers Verzekeringen]) could not justify the costs of assistive products. Thus based on Bernadette's current mobility restriction, determined by the insurance physician, it might be decided by the DEIIA what would be the cheapest and most adequate option to solve her mobility problem to go to work.

Setting aside the issue of funding resources, the occupational therapist worked with Bernadette to provide her with recommendations for the work place to facilitate job retention. To be able to observe and analyse the problems at work, the occupational therapist decided to combine a home visit with a visit to Bernadette's work place where recommendations in relation to Bernadette's work as a head nurse as well as self-care issues at work were provided. One example was suggesting a skirt and stay-up stockings in order to make bathroom visits easier for her as one of the main preconditions of Bernadette staying at work was that there was no acceptance of staff support for private activities. Bernadette was able and willing to negotiate with her employer herself and because the employer understood the difficulties of the uncertain prognosis, the costs for the assistive products she needed, such as a shower-toilet was paid for by the institution where she worked. The employer also agreed to pay for the increased personal assistant time so that reports and minutes were typed for her. An advantage for the employer was that some assistive products could also be used by the residents of the nursing home, thus the costs could be covered by the budget of the nursing home.



## **Challenges for job retention of the case study at micro, meso and macro level**

### *At micro or individual level*

The occupational therapist enabled Bernadette to overcome occupational issues by finding strategies to stay engaged in the occupations of everyday life, her job in particular. Bernadette was able to explain her situation and negotiate with her employer about the solutions needed to solve the problems she encountered at work, after consultation of the occupational therapist, who coached her and stimulated self-management and self-efficacy. The relationship between Bernadette and her employer was good. He was willing to support her remaining at work by providing assistive products, environmental redesign and personal assistance. These provisions were paid for by the general resources of the organization that employed Bernadette.

### *At meso or organizational level*

From the perspective of occupational therapists all daily occupations of a client can be addressed integrally by the occupational therapist, employment included. Indeed Bernadette was empowered at an individual level to cope with her progressive employment issues. The occupational therapist experienced barriers in care delivery by health insurance policy which did not permit intervention in work issues. Occupational therapy theoretically includes the currently underutilized area of 'paid work' but occupational therapists working in health care setting are prohibited from providing integrated care and using a participatory approach due to governmental rules and regulations <sup>[23]</sup>.

### *At macro or societal level*

This case illustrates the obstacles to providing adequate support to clients. Examples are: the uncertain diagnosis and prognosis of the disease, lengthy bureaucratic procedures, lack of clear information for the client, partner, and employer about regulations and opportunities and compensation arrangements, and barriers in cooperation between health professionals and occupational health services due to divided governmental financial resources. The approach of health professionals is diagnosis-related and client centred, whereas 'occupational health services' approach clients from an organizational perspective with governmental regulations in mind. Based on Bernadette's diagnosis, i.e., increasing limitations, the DEIIA may decide not to provide assistive products, but to provide Bernadette with a disability benefit. The DEIIA may make such decisions on the basis of identified severity of limitations at one moment in time related to the working environment. Exploring possibilities with the employer to find adaptations in tasks and to solve problems related to the expected increase of limitations is then taken into account. However, in the case of slow progressive physical deteriorations, a prognosis regarding the increase in impairments and limitations in activities is difficult. The insurance physician will monitor the decline and periodically invite the patient for a new rating of physical performance.

### **Comparison of the International Classification of Functioning, Disability and Health (ICF) and the Canadian Model of Occupational Performance & Enablement (CMOP-E)**

In 2001, the World Health Organization (WHO) published the *International Classification of Functioning, Disability and Health* (ICF) <sup>[24]</sup>. The ICF contains a graphic representation of health (the ICF scheme) containing disease/disorder and three domains of human functioning with the contextual factors (environmental and personal factors) which can influence health. The following terms can be used to describe functioning problems (disability): impairments, limitations in activities, and restrictions in participation without setting a relationship between these levels, for instance that impairment reduction might enable participation or occupational engagement.

The ICF has a goodness of fit with occupational therapy models of practice such as the Canadian Model of Occupational Performance & Enablement (CMOP-E). The ICF defines activity, as the “*The execution of a task or action by an individual*” <sup>[25]</sup>, much the same way as the Taxonomy of Occupational Performance (TCOP) defines it: “*Activity is a set of tasks with a specific end point or outcome that is greater than that of any constituent task*” <sup>[26]</sup>. In the ICF, activity is conceptually viewed as representing health at the individual level, and a problem at this level is labelled an activity limitation. In the ICF, participation is defined as “*Involvement in a life situation.*” Participation is conceptually viewed as functioning at the societal level, and a problem is called a participation restriction <sup>[25]</sup>. The construct of participation would appear to fit well with the construct of occupational performance captured by the CMOP-E. The ICF can classify aspects related to the person as well as the health care professionals’ point of view, thus both the ICF as well as the CMOP-E allow for an objective as well as a subjective description of the findings. In the ICF terminology the value of culture is an environmental factor e.g. the attitude of society towards employment.

The CMOP-E identifies three major constructs related to human occupation: person, occupation and environment <sup>[26]</sup>. The CMOP-E examines the fit in the interaction of these three constructs. It portrays human occupation as the result of the dynamic interaction of person, environment, and occupation, whereas occupational engagement is individual to each person. The CMOP-E provides a listing of necessary variables to enable occupation such as paid work, but does not give insight into how to combine these ingredients to enable occupation. The CMOP-E captures the occupational perspective of occupational therapy and positions it as an internationally important occupational model, whereas the Person-Environment-Occupation (PEO) Model developed by Law et al. in 1996 <sup>[22]</sup> provides more explicit information on the degree of fit between person, environment and occupation that is to say, the actual occupational performance, see figure 3.

Both models, the ICF and the CMOP-E, point to the importance of the interaction of the person and the environment to human functioning in context at one moment

in time. Aspects related to employment of Bernadette’s case can be explained in the CMOP-E. As Bernadette’s case underpins, physical dysfunction influences her performance and engagement in three areas of occupation: self- maintenance, productivity and leisure. In solving employment issues the three areas have to be considered in relation to each other to address Bernadette’s occupational challenges related to the area ‘work’.

The CMOP-E can be used to understand the source of those challenges, and how the strengths, resources, conditions and opportunities of enablement affect them. However, given the strength of the congruence between the ICF and the CMOP-E in identifying factors related to employment for persons with NMD, the ICF has been chosen in this dissertation because it supports interdisciplinary understanding of health and contains contextual factors related to employment participation<sup>[27]</sup>.

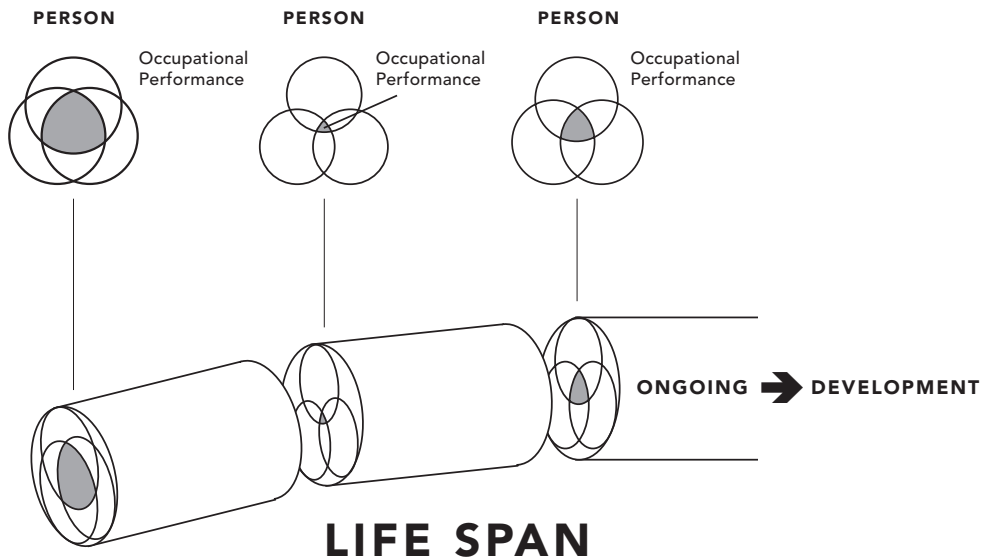


Figure 3. The PEO-Fit<sup>[22]</sup>

### The case study explained in terms of the ICF

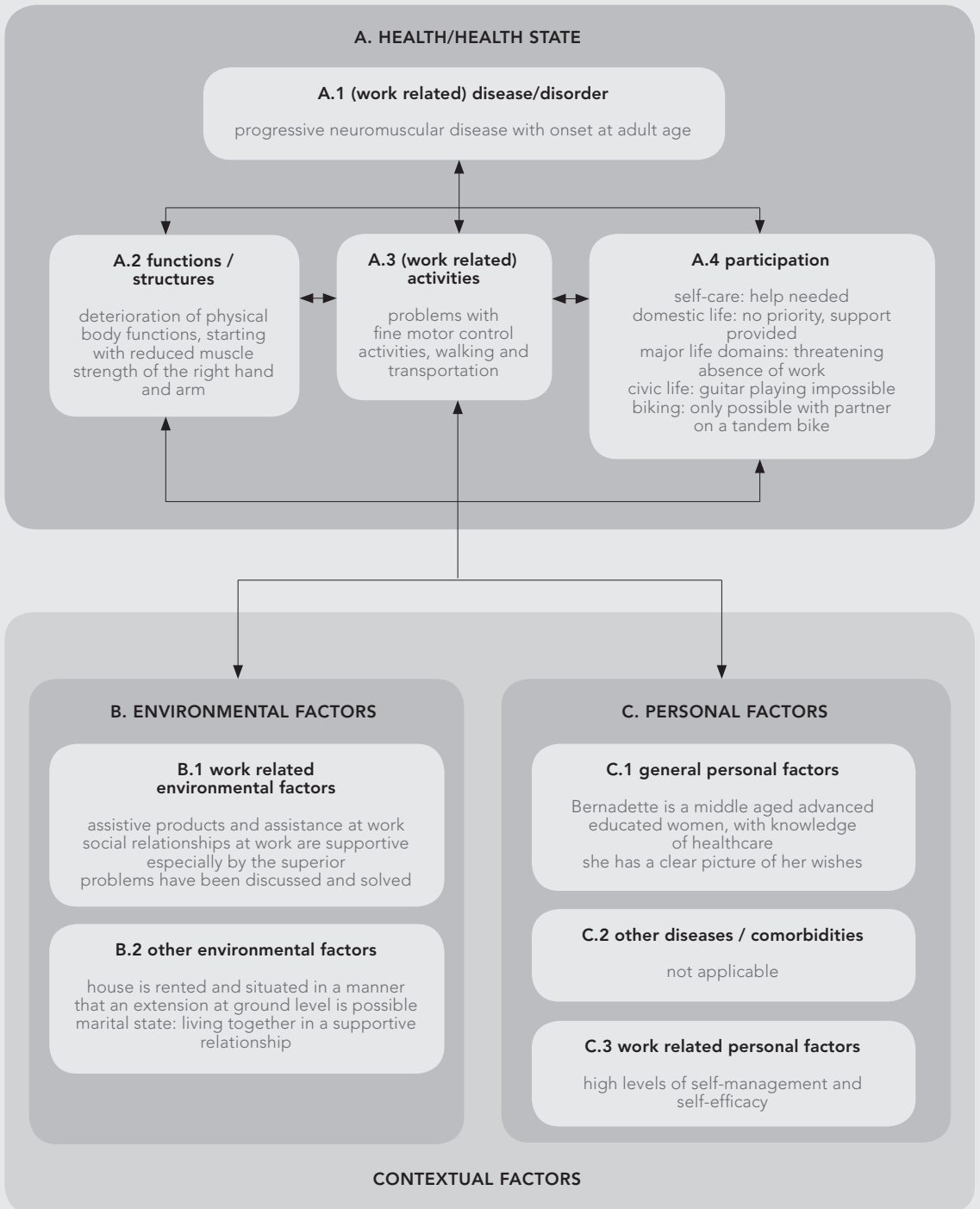
To organize factors influencing employment participation for persons with three types of NMD an expanded version of the ICF-scheme has been used (figure 4). Heerkens et al.<sup>[27]</sup> developed an expanded ICF model by subdividing environmental

and personal factors influencing the health of employees. Environmental factors were subdivided into work related environmental factors and other environmental factors. Personal factors were subdivided into general personal factors, other diseases and co-morbidities and work related personal factors<sup>[27]</sup>. The diagram depicts the multiple factors affecting Bernadette's employment status.

### **Occupational Therapy', a possible bridge between 'Occupation' and 'Health'**

Occupational therapy, in Dutch 'ergotherapie', is derived from the Greek word 'εργον' or 'ergo'. Ergo means: work, meaningful activity, active involvement, physical and mental activity and performance. From its very beginning, occupational therapy has been concerned with 'occupation' and its role in 'health and well-being'. Meyer, one of the first occupational therapists, stated as early as 1922 that *"There is a fundamental relationship between health, work and activities"*<sup>[28]</sup>. This statement is in line with the 1948 definition of health of the World Health Organization (WHO): *"Health is a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity"*<sup>[29]</sup>.

Circumstances, other than health conditions can be limiting factors in occupational performance and engagement, as can be seen by the way health was defined by the WHO in 1986: *"Enable focuses on equity and justice in opportunities, and resources to enable all people to achieve their fullest potential through a supportive environment, access to information, and opportunities for making health choices"*<sup>[30]</sup>. The most recent effort to define health has been formulated as follows: *"The ability to adapt and self-manage"*<sup>[31]</sup>. The basis of occupational therapy formulated in 2007 is *"The importance of having an occupation for human health, well-being and justice"*<sup>[32]</sup>. The definition of health focuses on empowerment instead of just removing symptoms with treatment with medication. This definition refers to the perceived quality of life and sense of well-being just as the definition of occupational therapy does. The term occupation refers to all manner of human doing, i.e., self-care, leisure, and productivity including paid work<sup>[32]</sup>. The focus on daily occupations orients occupational therapists to engage clients in the determinants of health which include paid employment<sup>[26]</sup>. These definitions seem to focus on the same thing, namely to empower a person. Occupational therapists' competencies include skills analyzing the consequences of diagnosis specific hindrances for people, not only in their daily home lives but also in the work place, and the impact of the injury on their productivity<sup>[33]</sup>. It can be expected that occupational therapists can provide support in job retention for persons with chronic diseases. *"The Canadian Association of Occupational Therapy (CAOT) believes that engagement in meaningful occupations, including those in paid work environments, are an important determinant of health and as such views return-to-work (RTW) as a valued occupational therapy service area"*<sup>[34, 35]</sup>.



**Figure 4.** Expansion of the ICF-scheme of the World Health Organization <sup>[24]</sup>, resulting in a version of the scheme as published by Heerkens et al. <sup>[27]</sup>.

### The Dutch healthcare system concerning employment participation

A challenge to client-centred practice is the bio-medical context of health services<sup>[36]</sup>. The unease for the occupational therapist willing and able to provide support in work maintenance occurs when their practice management is narrowed to self-care, leisure, and unpaid work with minimal attention to paid work<sup>[37]</sup>. For occupational therapists it is not their background or knowledge that has been a hindrance to intervention in clients' employment issues, but their position in the health care system, i.e., the different laws and separate forms of funding applicable to health care and employment. This is in contrast to other countries such as Germany, Finland and the USA where medical care for sick workers and their reintegration after absenteeism form a much more integrated package<sup>[38]</sup>. This is illustrated by the fact that the functions of general practitioner and occupational health physician can be combined in one and the same person. In these countries, laws and funding for sickness and absenteeism are interrelated<sup>[39]</sup>.

In this respect it is interesting to look at the report of Ybema who investigated the effect of the Dutch law launched in 2001 with the aim to make both employer and employee responsible for rapid reintegration (Dutch law pertaining to reintegration from sick leave / Dutch Gatekeeping Improvement Act [in Dutch: Wet Verbetering Poortwachter])<sup>[40]</sup>. The authors investigated the effect of the new law (wvvp) on return to work (RTW) and concluded that RTW is expedited when the relationship with the employer is good<sup>[41]</sup>. In the case of Bernadette, her relationship with her employer was also fine, and that may be the reason that she had not been on sick leave. The employer was willing to support her and provided assistive products and personnel to support her with her work.

Apparently, intentions to support job retention for clients at risk differ among health care professionals and governmental representatives in the Netherlands. The insurance physician evaluates the possibilities for job retention with governmental regulations and legislation in mind and judges whether someone with a disability is still able to work. If someone is restricted in the performance of his current job, due to his health condition, and, according to the insurance physician, the client's capacities match another job, the person will be asked to apply for that other job in the same organization or somewhere else when that job is not available in his own organization. If the person is not successful in finding a job, he has to rely on general social benefits (Unemployment benefit act [in Dutch: Werkloosheidswet, 1986]), not on disability benefits [Work and Income according to Labour Capacity Act (in Dutch: Wet Werk en Inkomen naar Arbeidsvermogen, 2005] because he has no work disability. In contrast, professionals working in health care, who are focused on well-being for the individual client (a client-centred approach), the governmental insurance policy is to seek the best solutions from economic and societal perspective (a system approach).

The content of referrals by physicians to occupational therapists working in health care is generally broadly formulated and minimally related to the client's occupation or 'gainful employment'. It is the professional responsibility to initiate first contact, set the stage, assess and evaluate occupational performance, and agree on treatment goals in concord with the client <sup>[26]</sup>. However, under current regulations (in the Netherlands) if the occupational therapist is working in a health care setting, he is restricted from intervening in employment issues. It is Anema's view that occupational health care should be brought into mainstream health care with work participation as a shared treatment goal <sup>[42]</sup>. This will be a great opportunity for occupational therapists to embrace work productivity as a main focus of their efforts in health care settings.

### **Neuromuscular diseases and consequences for employment participation**

#### *FSHD and employment*

Only one study has been found that describes the current occupational situation, vocational disability and past career and gives an indication of the job perspectives and preferable vocational training of a large group of persons with FSHD <sup>[3]</sup>. Wevers et al. <sup>[3]</sup> found that 64% of the persons with FSHD required substantial mental effort to retain the job and 38% found their job physically strenuous.

#### *HMSN and employment*

Persons with HMSN experience significant fatigue and pain influencing quality of life <sup>[43]</sup>. Overall physical deterioration of HMSN varies greatly among families from minimal impact (except for walking difficulties, stumbling and falls, which can be remediated with special shoes) to very disabling. Hence, it can be expected that the impact of HMSN on employment status is quite varied.

#### *MD and employment*

The only previous study regarding employment of individuals with MD was a French Canadian study <sup>[44]</sup>. The results of this study clearly indicate that clients with MD exhibit a socio-economic profile associated with disadvantaged milieu: high unemployment, low income and limited education. The wage-earning history of persons with MD is closely linked to their self-evaluation of their working capabilities. After the age of forty, a large proportion of these persons is unable to maintain paid employment. It is therefore expected that MD might have a huge impact on employment participation <sup>[44]</sup>.

### **Objectives and research questions of this dissertation**

Related to the aim of this dissertation the following research questions were formulated:

- How many working age persons with NMD are employed? Are there differences in employment status between persons with FSHD, HMSN, and MD?
- Which health related factors, environmental factors and personal factors can be identified that are associated with employment status of persons with FSHD, HMSN, and MD?
- To what extent do persons with NMD receive referrals to occupational therapists, physiotherapists and speech therapists working in a multidisciplinary team in a hospital setting for interventions to support them in employment?
- How do persons with an NMD perceive their own employment participation?

### **Dissertation outline**

This dissertation covers scientific perspectives and client perspectives related to employment status of persons with NMD in order to contribute to the improvement of employment prospects for people with NMD. By exploring factors influencing employment of persons with NMD, lessons can be learned that may be beneficial to health professionals and occupational health professionals so as to enable persons with NMD to retain their jobs and for persons with NMD to enhance their self-management strategies.

*Chapter 2* contains the methods and results of a systematic review conducted to summarize scientific evidence about factors that influence employment status for persons with NMD.

In *Chapter 3* the results of a secondary analysis of the data on the influence of fatigue in daily functioning are presented <sup>[45]</sup>, to gain more insight into factors related to maintaining employment for a large group of 591 gainfully employed persons with NMD. Disease related personal factors, fatigue severity, and functional health status factors related to employment of a large NMD group and subgroups of patients with FSHD, HMSN, and MD have been determined. Also differences in employment status among the three subgroups of patients have been investigated.

In *Chapter 4* recommendations about employment issues formulated by allied health professionals in a clinical health setting have been explored. This exploration was based on the premise that occupational therapists working in their professional roles could have played a role in job retention of persons with a disability. By exploring recommendations about employment as part of the advice given in the clinical setting by specialized occupational therapists, physical therapists and speech therapists after one-off consultations, it was possible to determine whether employment problems were found and to what extent patients with NMDs received referrals for interventions regarding employment.



In *Chapter 5* the results of a qualitative study are presented to approach the client perspective. A deeper understanding of the clients' point of view on the meaning of paid employment is gained based on the results of open in-depth interviews with 16 employees with NMD. Respondents were asked how they perceive employment in order to find key elements which help them to stay employed while the disease deteriorates physical and sometimes mental functioning. Findings might be helpful to create awareness about the impact of a physical condition on job retention for health care professionals and occupational professionals and to better support and empower persons with NMD to retain their jobs.

*Chapter 6* contains the general discussion addressing the main findings and clinical implications of the studies for the improvement of employment prospects for people with NMD and more effective guidance by physicians, health care professionals and occupational professionals involved.

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## **CHAPTER II**

### **Classification of Employment Factors according to the International Classification of Functioning, Disability and Health in Patients with Neuromuscular Diseases: A Systematic Review**

Published as:

Minis MA, Heerkens Y, Engels J, Oostendorp R, van Engelen B.

Classification of Employment Factors according to the International Classification of Functioning, Disability and Health in Patients with Neuromuscular Diseases:

A Systematic Review

*Disability and Rehabilitation* 2009;31(26):2150-63.



## Abstract

*Purpose:* A systematic evaluation of the literature to identify health and contextual factors associated with employment in patients with Neuromuscular Diseases (NMD) and to perform a best evidence synthesis, taking into account the design of studies, methodological quality, and the statistical significance of findings.

*Method:* Publications were retrieved by a computerised search in medical and psychological databases. Two reviewers assessed titles and abstracts first and assessed the quality of the remaining full text publications independently as well. Of the residual publications health and contextual factors associated with employment in NMD patients were extracted. The factors found were included in a recently developed expanded International Classification of Functioning, Disability and Health scheme.

*Results:* 662 titles and abstracts were screened. The main reason to exclude a title and/or abstract was the absence of the study population selected: Facioscapulo-humeral Muscular Dystrophy (FSHD), Hereditary Motor and Sensory Neuropathy (HMSN) & Myotonic Dystrophy (MD). Of the remaining 20 full-text publications, eight publications fulfilled the inclusion criteria: two repeated survey designs and six cross-sectional studies. Factor extraction resulted in 94 factors related to employment. Ten factors in five publications were indicative for an association with employment status: Disease related factors HMSN, MD & NMD in general), factors related to functions (physical functions, muscle power functions), general personal factors (age, gender and education), work related personal factors (type of occupation, and expressed interest in employment by NMD patients).

*Conclusion:* In the best evidence synthesis ten factors were indicative for an association with employment status in NMD patients in five publications with good to excellent methodological quality.

## Introduction

The impact of a chronic illness can cause problems in participation, including limitations in the ability to work, which can negatively influence economic and social status<sup>[1]</sup>. Patients with a disability value their work in the same positive way as healthy people<sup>[2-5]</sup>. Together with quality of life (QOL), return to work (RTW) is one of the principal endpoints in rehabilitation<sup>[6]</sup>. In order to support people with a chronic disease to maintain a job or RTW, it is important to detect which factors influence the ability to work, especially factors that are within the scope of allied health care interventions such as vocational therapy. Scientific evidence of factors related to health state, work and other environmental and personal factors is needed to improve care and services by allied health professionals and organizations involved in the (re-) integration process.



To investigate factors that influence employment status in chronically ill patients, we selected patient groups with chronic, slow progressive, hereditary neuromuscular diseases (NMD). This choice is based on three reasons. Firstly, because NMD patients state that participation in the work force is an important factor for their quality of life<sup>[7]</sup>, secondly, because the onset of NMD mainly occurs during working ages (18-65 years) and thirdly, because the main feature of these three NMDs is muscle weakness<sup>[8-11]</sup>. In general NMD patients strive to stay employed without disclosing their illness or progression of their illness. NMD patients will not easily admit to disease related problems when this creates a risk to their employment<sup>[7]</sup>.

The factors found will be included in a recently developed scheme expanding on the International Classification of Functioning, Disability and Health (ICF)<sup>[12,13]</sup>. Within this scheme a distinction can be made between state of health factors (functions, structures, activities and participation), environmental factors (work related environmental factors and other environmental factors) and personal factors (general personal factors, other diseases/comorbidities and work related personal factors).

Until now, no systematic review has been reported on the presence of health and contextual factors in relationship to employment in NMD patients. The objective of our review is to identify state of health, personal and environmental factors associated with employment of NMD patients. Identified factors will be categorized according to the scheme developed by Heerkens et al.<sup>[13]</sup>.

## Methods

### *Search strategy*

Publications were retrieved by an extensive computerised search, using the following resources: PUBMED (1966 until May 2006), MEDLINE (1966 until April 2006), CINAHL (1982 until April 2006), PSYCHINFO (1972 until May 2006), ERIC database (1966 until April 2006), WEB OF SCIENCE (1996 until April 2006), the Cochrane Controlled Trials Register (1996-2006), EMBASE (1996 until 2006), PICARTA and the Dutch library of medical and rehabilitation literature (Dutch Institute of Allied Health Care) (NPI) (1960 until April 2006). In Pubmed the search terms of the neuromuscular diseases Facioscapulo-humeral Muscular Dystrophy (FSHD), Hereditary Motor and Sensory Neuropathy (HMSN) and Myotonic Dystrophy (MD) were entered as MeSH terms and free text words, combined with several MeSH terms and free text words related to employment. In Medline, also a search was done with only free text words and the same text words were used to search the other databases. For full description of the computerized search strategy in PUBMED, using MeSH terms and the adapted search applicable to the other databases see appendix 1. An experienced medical librarian advised on the search strategy.

*Selection criteria*

The procedure for inclusion of the studies was based on the recommendations described by Van Tulder et al <sup>[14]</sup> and consisted of two phases: first screening of titles and abstracts and, secondly, screening of selected full text publications. Application of inclusion and exclusion criteria were pilot tested by two reviewers (MK & YH) and adjusted until consensus was reached.

The first phase screening of titles and abstracts was independently performed by two reviewers (MK & YH). The inclusion criteria were: design, age, language, population (FSHD, HMSN & MD), and subject (work). The following designs were included: Randomized Clinical Trials (RCTs), Controlled Clinical Trials (CCTs), and Other Designs (ODs) such as observational studies (cross sectional, cohort, case-control studies, repeated survey design and longitudinal study studies (prospective and retrospective studies). Inclusion was also limited by age. The age range was set between 18 and 65 years old (working population). Only English or Dutch languages were accepted and included for the full text articles. Study populations included the following types of NMD: FSHD or HMSN or MD or Hereditary Neuromuscular Disease or Neuromuscular Disorder/Disease, combined with one of the subjects: vocation or occupation or work or absenteeism or (un)employment or sick leave. An abstract was also included for further review when (some of) the inclusion criteria were not clearly specified. However, this was only done, when, in case of any doubt, a third reviewer (JE) was consulted and agreed affirmatively to the abstract.

Exclusion criteria for titles and abstracts were all other Neuromuscular Diseases/ Disorders, that are classified as NMD with a different aetiology. Examples are diseases or disorders of neuromuscular nature caused by circumstances at work e.g. complaints of arm, neck, and shoulder (CANS), Carpal Tunnel Syndrome, Entrapment Neuropathies, aids, acquired NMD or other neurological chronic diseases. Narrative reviews, updates of systematic reviews, qualitative research such as case reports, congress abstracts, books and commentary on other papers, letters to the editor, economical evaluations and guidelines were excluded.

In the second selection phase the remaining full text publications were independently assessed by two reviewers (MK & YH). In this phase, the inclusion criteria were combined with research outcomes or interventions related to employment. Full text papers were included when all inclusion criteria were met and none of the exclusion criteria were applicable. Consensus was reached by discussion. When consensus was not reached a third reviewer (JE) decided.

*Methodological quality*

Criteria lists developed by the Cochrane review committee were used for RCTs and CCTs <sup>[15]</sup>. Lists developed by Van der Windt et al <sup>[16]</sup> and Van Tulder et al <sup>[14]</sup> were adapted by the first author using Checkoway's <sup>[17]</sup> research methods for ODs, see appendix 3. These adapted criteria were used to identify the methodological quality of the ODs. Separate lists were designed for repeated survey and cross-sectional studies. Each

criterion was graded as ‘+ = present’ or ‘- = not present’. If insufficient information was provided in the studies, the criterion was assessed as ‘? = not described or insufficiently described’. Excellent quality studies were defined as scores of  $\geq 10$  ‘+’ (of a possible 13) for repeated survey studies and  $\geq 7$  ‘+’ (of a possible 9) for cross-sectional studies. Studies were considered ‘good’ with a score between 6-9 for repeated survey and 4-6 for cross-sectional studies. Studies with scores below 6 and 4 respectively were considered ‘low quality’. In case of any disagreement between the two reviewers (MK & YH), a consensus method was used to discuss and solve the disagreement. When the disagreement persisted a third reviewer (JE) decided.

### *Best Evidence Synthesis*

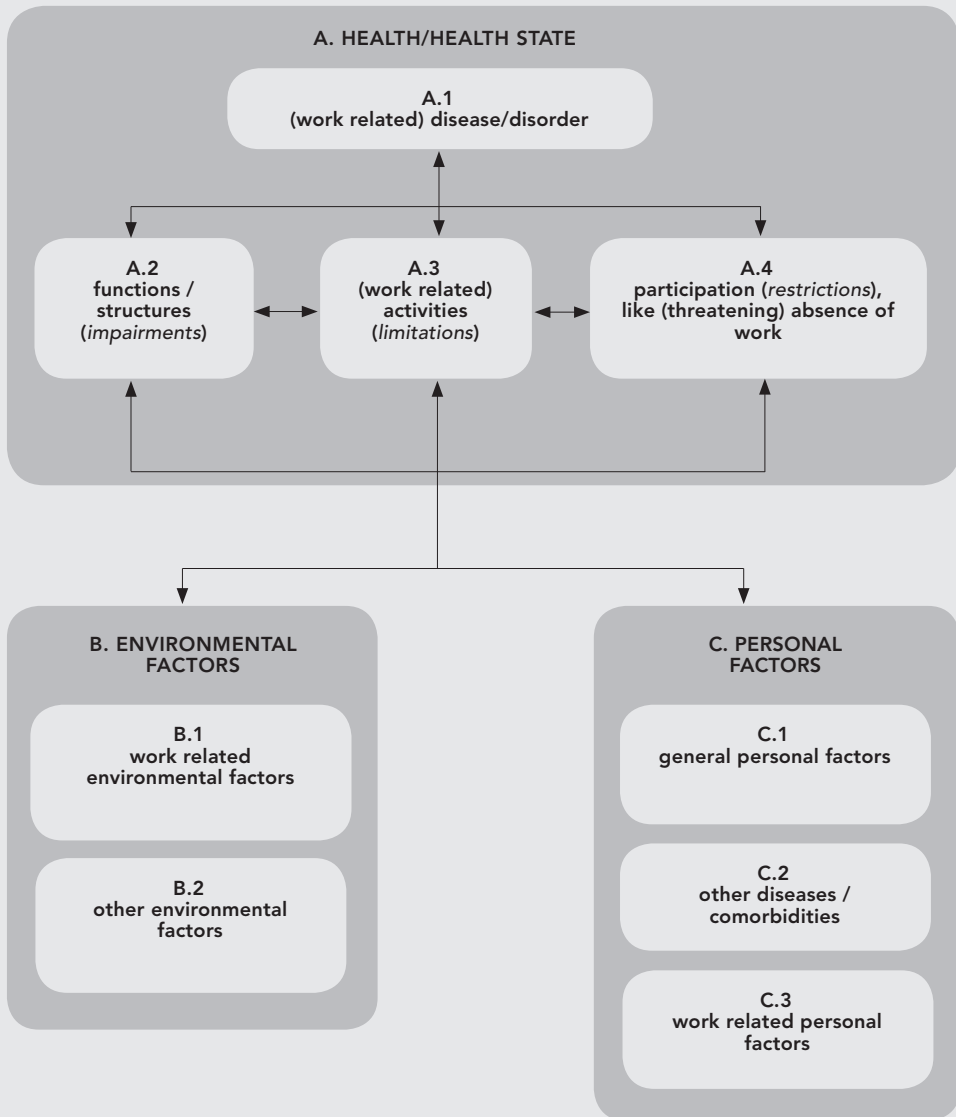
The best evidence synthesis for quantitative studies based upon the methods of Van Tulder et al <sup>[14]</sup> was used for the purpose of this review (table 1).

**Table 1.** Best-evidence synthesis for quantitative research

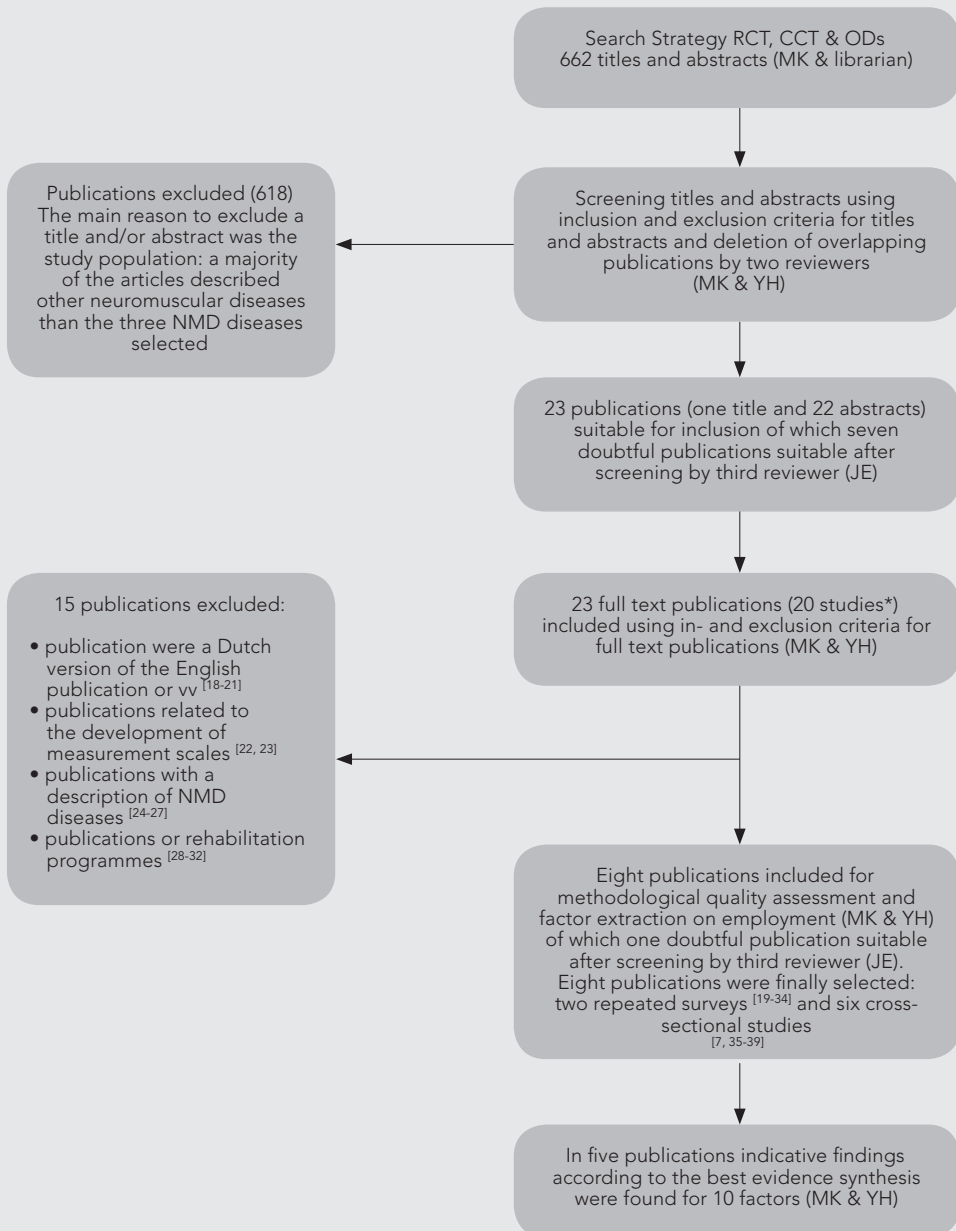
Strong evidence:	provided by consistent, statistically significant findings in outcome measures in at least two high quality RCTs*
Moderate evidence:	provided by consistent, statistically significant findings in outcome measures in at least one high quality RCT and at least one low quality RCT or high quality CCT*.
Limited evidence:	provided by statistically significant findings in outcome measures in at least one high quality RCT* or: provided by consistent, statistically significant findings in outcome measures in at least two high quality CCTs* (in the absence of high quality RCTs)
Indicative findings:	provided by statistically significant findings in outcome and/or process measures in at least one high quality CCT or low quality RCT (in the absence of high quality RCTs) or provided by consistent, statistically significant findings in outcome and/or process measures in at least two ODs with sufficient quality (in absence of RCTs and CCTs*)
No or insufficient evidence:	in cases of results of eligible studies do not meet the criteria for one of the above stated levels of evidence or in case of conflicting (statistically significant positive and statistically significant negative) results among RCTs and CCTs or in case of no eligible studies

\* If the less than 50% of the total number of eligible studies within the same category of methodological quality and study design (RCTs, CCTs or ODs) found no evidence we categorize this as no evidence.

RCT, randomized clinical trial; CCT, controlled clinical trial; OD: other design



**Figure 1.** Expansion of the ICF-scheme of the World Health Organization (WHO) <sup>[12]</sup>, resulting in a simplified version of the scheme as published in 2004 by Heerkens et al <sup>[13]</sup>



**Figure 2.** Flowchart of the review process and factor extraction  
\*Publications describing the same study were considered as one study

In case of a lack of RCTs and CCTs, a best evidence synthesis analysis was performed by attributing various levels of evidence to factors contributing to employment status of NMD patients, taking into account the design of the studies, the methodological quality, and the statistical significance of findings. Resulting ODS with good or excellent quality and statistically significant findings were selected. Indicative findings are provided by consistent, significant findings in outcome and/or process measures in at least two ODS.

Factor extraction using the International Classification of Functioning, Disability and Health (ICF). Two reviewers (YH & MK) independently performed an extraction of factors influencing employment of NMD patients on the remaining full text publications. The identified factors that can be associated with participation in the work force are included in an expansion of the ICF-scheme, designed by Heerkens et al.<sup>[13]</sup>. The scheme can be useful to describe health problems of persons related to work and to identify the points of intervention by different professionals. The schematic representation adds a further subdivision of environmental and personal factors to the ICF-scheme. The codes of the ICF were used to categorize the main factors: Health State (A), Environmental factors (B) and Personal factors (C). Environmental factors (B) were subdivided in work related environmental factors and other environmental factors. Personal factors (C) were subdivided into general personal factors, other diseases and comorbidities and work related personal factors. This expanded scheme of the ICF was used with the expectation that the categorization would be helpful for ordering health and contextual factors found. When codes were not known for the factor present, MK & YH assigned the factor to a category (figure 1).

## Results

### *Selection of studies*

The flow chart of the review process is shown in figure 2.

Our search strategy resulted in 662 citations: PUBMED (115), MEDLINE (232), CINAHL (105), PSYCHINFO (11), ERICDatabase (2), WEB OF SCIENCE (27), Cochrane Controlled Trials Register (54) and EMBASE (104). The search in the Dutch database PICARTA and the Dutch Institute of Allied Health Care resulted in four and eight more studies respectively. The 662 titles and abstracts were screened for inclusion or exclusion by two reviewers (MK & YH). Twenty three publications (1 title and 22 abstracts) were selected by the two reviewers. The main reason to exclude a title and/or abstract was the study population: a majority of the articles described other neuromuscular diseases than the three NMD diseases selected. Based on the exclusion criteria for full text articles fifteen studies were excluded for the following reasons: four publications described the same study population, because the publications were a Dutch version of the English publication or vice versa<sup>[18-21]</sup>, or two publications related to the

**Table 2.** Methodological quality assessment of other designs of publication related to employment and NMD according to the adapted version of Van Tulder et al<sup>[14]</sup> and Van der Windt et al. <sup>[16]</sup>

<b>Criteria of methodological quality repeated survey design</b>																			
Author(s)	Criterion <sup>a</sup>	1	2	3	4	5	6	7	8	9	10	11	12	13	Internal validity Criteria 5, 6, 7, 9	Descriptive Criteria 1, 2, 8, 10, 11	Statistical Criteria 3, 4, 12, 13	Quality	
Andries et al	2000	+	+	+	?	+	+	-	+	+	+	+	+	-	3	5	2	excellent	10
	2005a	+	+	+	-	+	+	+	+	+	+	+	+	-	4	5	2	excellent	11
<b>Criteria of methodological quality cross-sectional design</b>																			
Author(s)	Criterion <sup>a</sup>	1	2	3	4	5	6	7	8	9	10	11	12	13	Internal validity Criteria 6	Descriptive Criteria 1, 2, 6, 8	Statistical Criteria 3, 4, 12, 13	Quality	
Andries et al	1997	+	+	+	+	+	+	+	+	+	+	+	+	+	1	4	4	excellent	9
	2005 <sup>b</sup>	+	+	+	+	+	+	+	+	+	+	+	+	-	1	4	3	excellent	8
Fowler et al	1997	+	+	+	-	+	+	+	+	-	-	+	+	-	1	3	2	good	6
Natterlünd & Ahlström	1999 <sup>a</sup>	+	+	+	+	+	+	+	+	-	-	+	+	-	1	3	3	excellent	7
Ville et al	1992	+	+	+	+	+	+	+	+	+	+	+	+	-	1	4	3	excellent	8
Wevers et al	1993	+	+	+	+	+	-	+	+	+	+	+	-	-	0	4	2	good	6

**Legend:**

Criterion<sup>a</sup> A repeated survey design was considered to be of excellent quality if the methodological quality score was 10 or more. Maximum score = 13.

Criterion<sup>b</sup> A cross-sectional study was considered to be of excellent quality if the methodological quality score was 7 or more. Maximum score = 9.

### Methodological criteria list for repeated survey studies/cross-sectional studies

- + = positive,
- = negative,
- ? = described or insufficiently described,
- NA = Not Applicable

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#### Objective of the study

- 1/1 A specific, clearly stated objective is described.

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#### Study population

- 2/2 Description of inclusion or exclusion criteria is adequate.
- 3/3 The main features of the study population (cohort definition) are described (sampling frame and distribution of the population)
- 4/4 Positive if the main features of the study population (cohort definition) are described (sampling frame and distribution of the population by age and sex)
- 5/Na was the composition of the population of each of the surveys similar with respect to relevant responder characteristics / exposures to employment factors
- 6/6 The number of non-responders is described and acceptable. Participation rate at baseline is at least 80% or if participation rate is 60-80% and non-response is not selective (data presented)
- 7/Na the number of non-responders is described and acceptable: the response at main moment of follow up is at least 60-80% or if non-response is not selective (data presented)

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#### Exposure measurements: factors associated with employment

- 8/8 Method for measuring factors related to employment
- 9/Na repeated measure methods involve similar health and exposure methods

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#### Measurements

- 10/10 The employment measures are relevant and defined adequately
- 11/Na data were collected for at least one year or at least every three months

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#### Analysis and data presentation

- 12/12 Statistical analyses are adequately chosen in relationship to the research question and data presented
- 13/13 Statistical analyses are sufficiently powered.



**Table 3.** Study characteristics of eight selected publications on employment and neuromuscular disease (NMD) patients

First Author (reference)	Design	Analysis	Type of NMD	Number of participants	Male (%)	Employed (%)	Source population selected from:	Instruments	Follow up duration
Andries 1997	Cross-sectional	t-test, Chi-square, Logistic regression analysis (stepwise LR)	MD HMSN SMA MG	1072	47%	Phase 1 41%	Seven university clinics	Descriptive data on four NMD disorders Disorder related characteristics (Former) job and career related characteristics Present job characteristics RAND-36	NA
Andries 2000	Repeated survey	Chi-square t-test%, mean, Linear regression	MD MG SMA HMSN	1994 987 1999 822	46%	Phase 2 38,6%	NMD members of the VSN, the Netherlands	Work and Handicap Questionnaire for employees Questionnaire for direct chief Questionnaire for employers	5 yrs
Boström 2005 <sup>a</sup>	Repeated survey	ANOVA Paired t-tests	MD	T0 58 T1 44	Not reported	T0 = 48% T1 = 25%	Comprehensive prevalence study in the county Örebro, Sweden	Self report Activities of Daily Living (ADL) SIP	10 yrs
Boström 2005 <sup>b</sup>	Cross-sectional	Kruskal-Wallis Mann-Whitney	MD	46	42%	39.1%	A comprehensive prevalence study in the county Örebro, Sweden	Demographic variables Subjective estimation of Quality of life SCoL	NA
Fowler 1997	Cross-sectional	ANOVA Chi-square	LGD FSHD BMD SMA HMSN MD	27 16 9 17 43 42 Total 154	Not reported	40%	Ambulatory patients from a regional teaching hospital California DR counsellors	Patient Occupational Survey Questionnaires for DR counsellors to determine their experience with NMD's	NA

Questionnaire to determine physician experience with NMD's

Regional physicians  
Reviews of DR caseload files

MMPI  
CPI

Objective evaluations of impairment and disability

Natterlünd 1999 <sup>b</sup>	Cross-sectional	Unpaired t-test, ANOVA	MD Post-polio syndrome	33 46 Total 119	50%	44,5 %	A comprehensive prevalence study in the county Örebro, Sweden	Personal characteristics Five occupational forms and their basic activities	NA
Ville 1992	Cross-sectional	Comparison of means, correlations and descriptive multivariate statistics	FSHD and MD	68	47%	83.8 %	Through contact of a specialist in neuromuscular diseases	Manual testing of 38 different muscles Brook Functional Scale Questionnaire on the social construction of identity: Personal characteristics Population characteristics	NA
Wevers 1993	Cross-sectional		FSHD	138	Not reported	44.2 %	Seven university clinics, the Netherlands	Patient/worker Questionnaire	

ANOVA = Analyses of Variance, BMD = Becker's Muscular Dystrophy, CPI = California Psychological Inventory, DR = Department of Rehabilitation, HMSN = Hereditary Motor and Sensory Neuropathy, LGD = Limb-Girdle Dystrophy, MD = Myotonic Dystrophy, MG = Myasthenia Gravis, MMPI = Minnesota Multiphasic Personality Inventory, NMD = Neuromuscular Diseases, SIP = Sickness Impact Profile, SMA = Spinal Muscular Atrophy, SQol = Subjective estimation of Quality of life, VSN = Vereniging Spierziekten Nederland.

development of measurement scales <sup>[22, 23]</sup> or nine publications with a description of NMD diseases <sup>[24-27]</sup> or rehabilitation programmes <sup>[28-32]</sup>. Eight publications were finally selected: two repeated surveys <sup>[33, 34]</sup> and six cross-sectional studies <sup>[7, 35-39]</sup>.

#### *Methodological quality of the studies*

The methodological quality of the studies included is presented in table 2.

The two repeated survey studies had an adequate description of inclusion/exclusion criteria, fulfilled the internal validity criteria, had descriptive as well as statistical criteria and described the repeated survey design duration (table 2). The two studies met the criteria for 'excellent' quality <sup>[33, 34]</sup>. The six cross-sectional studies had an adequate description of inclusion/exclusion criteria, fulfilled the internal validity criterion, had descriptive as well as statistical criteria and were rated as good (6 of 9) <sup>[35, 36]</sup> or excellent ( $\geq 7$  of 9) <sup>[7, 37-39]</sup>. Fowler did not describe the study population adequately <sup>[35]</sup> and both Fowler and Nätterlund did not present relevant employment measures <sup>[35, 26]</sup>. Adequate statistics were used for parametric and non-parametric data analyses in seven of the eight articles. Wevers et al <sup>[36]</sup> presented only a description of the study population and percentages of findings. The statistical criterion 'sufficiently powered' was only present in one paper <sup>[7]</sup>.

#### *Study characteristics*

The characteristics of the studies included are presented in table 3.

Types of NMD in the studies varied. Seven included MD, three included FSHD, HMSN and Spinal Muscular Atrophy (SMA), and two included Myasthenia Gravis (MG). Limb Girdle Dystrophy (LGD) and Becker's Muscular Dystrophy (BMD) were present in one of the studies. The number of participants and the variety of types of NMD in the studies varied from large numbers <sup>[7, 33]</sup> and one type of disease <sup>[30, 35, 38]</sup> to relatively small numbers <sup>[21, 31, 34, 35, 37, 39]</sup> and many types of NMD combined <sup>[36]</sup>. Five studies reported percentages of gender for the total group of included subjects. Percentages of males varied from 42% to 50% <sup>[7, 31, 33, 37, 39]</sup>. Source populations were recruited from university clinics <sup>[7, 33, 37]</sup>, a patient association for people with NMD <sup>[7, 33]</sup>, from rural areas <sup>[25, 34, 37]</sup> or through contact of a hospital or neurologist <sup>[35, 39]</sup>. For the two repeated survey studies follow up duration ranged from five to ten years <sup>[33, 37]</sup>. Intervention strategies were not performed. The studies focused on different instruments used in NMD research and different aspects of employment. These aspects included mental or physical impairments (e.g. fatigue) and activities <sup>[11, 19, 34]</sup>, participation in work <sup>[33, 35]</sup>, health care <sup>[11]</sup> and referral patterns <sup>[35]</sup>. Fowler focuses especially on mental impairments using the Minnesota Multiphasic Personality Inventory (MMPI) and the Wechsler Adult Intelligence Scale Revised (WAIS) showing that some intellectual functions influence employment status. For the combined NMD group mental impairments, measured with the MMPI, and psychosocial adjustment, measured with the California Psychological Inventory (CPI), were found to be significant factors in relation to employment <sup>[35]</sup>. All studies described factors related to employment. For details of instruments and outcome of all studies, see table 3.

*Factor extraction*

The results of the factor extraction related to employment were included in the scheme of Heerkens<sup>[13]</sup> presented in appendix 2.

For ten of the 94 factors found, the results are scored as indicative findings. These indicative findings are categorised as disease related factors (type of NMD: HMSN and MD and other types of NMD), factors related to functions (physical functions, muscle power functions), general personal factors (age, gender and education) and work related personal factors (type of occupation, and expressed interest in employment by the NMD patient). No evidence was found for the other 84 factors. 77 of these factors were present in only one study; of these factors, 44 had a significant relation with employment; 32 were not related to employment and in one study the factor was significantly related to employment in MD patients, but not in HMSN patients<sup>[33]</sup>. The remaining seven factors were present in two studies: six factors were significantly related to employment in one of the two studies, but not in the other study and only one factor was not related to employment in both studies.

*Best evidence synthesis*

Considering the eight studies with good or excellent methodological quality for ODs, ten factors in five publications were indicative for employment of NMD patients (table 4).

## Discussion

Two repeated surveys and six cross-sectional studies of 20 selected publications met the criteria for factor extraction. These eight studies showed good to excellent methodological quality. By critically reviewing the full text of these eight studies we identified 94 factors; for ten of these factors there were indicative findings with respect to their relation to employment. These ten factors were found in five publications. Three of ten factors of the two cohort studies concerned type of disease: HMSN, MD and NMD in general. Remarkably, in previous studies influence of disease factors on employment were not found e.g. in studies about chronic diseases such as chronic obstructive pulmonary disease (COPD)<sup>[40]</sup> rheumatoid arthritis (RA), asthma and ischemic heart disease (IHD)<sup>[41]</sup>. Impaired body function such as decreased physical function and muscle power however were found to be indicative factors to employment, both in our review and in other studies<sup>[41]</sup>. Indicative personal factors in NMD patient groups were younger age, being male and higher education all of which increased the chance of employment. The labour force participation in RA patients was found to be only slightly lower than the general population<sup>[42]</sup> so we might conclude that these three personal factors are related to employment for every human being. Furthermore work related personal factors such as type of occupation (heavy manual work versus non manual work) and expressed interest in employment by the NMD

patient seemed to be of consequences on employment. In the review of Detaille et al<sup>[41]</sup>, of the category personal factors also age, gender and education were found as common personal factors predicting employment. Type of occupation was also found to be a prognostic factor in RA, Astma and IHD patients as well<sup>[41]</sup>.

Of the residual 84 factors 44 factors had a relationship to employment in NMD patients however these factors were each present in only one of the studies. Evidence for work related environmental factors and other environmental factors were not found. In order to reach the lowest level of evidence according to the evidence synthesis a factor has to be present in at least two OD studies to be indicative. According to the best evidence synthesis uncontrolled studies or ODs can only contribute to indicative of the best evidence synthesis.

In our view, building a best evidence synthesis solely on RCTs or CCTs, under values other types of study designs because outcomes of prospective outcome research is valuable when subsets of a defined population have to be identified<sup>[43]</sup>. Those outcomes cannot be reached with RCTs or CCTs. Cohorts are defined populations, which are followed in an attempt to determine distinguishing subgroup characteristics. Repeated survey research is an observation of a population for a sufficient number of persons over a sufficient number of years to generate incidence or mortality rates subsequent to the selection of the study group. A longitudinal study is one in which two or more measures or observations of a comparable form are made of the same individuals or entities over a period of at least one year<sup>[44]</sup> and therefore more applicable to observe change over time in the disposition of (work related) factors than RCTs or CCTs.

Recently a network of methodologists, researchers and journal editors was established and they developed a position statement for reporting observational research: Strengthening the Reporting of Observational studies in Epidemiology (STROBE) statement<sup>[43]</sup>. The STROBE statement is an acknowledgement of the value of observational research. Unfortunately, the recommendations for reporting observational research of the STROBE initiative cannot be used as criteria to select methodological quality of publications. To select OD studies of sufficient methodological quality an applicable set of criteria had to be found. Two lists were found<sup>[14,16]</sup>. These lists were designed for specific diagnostic groups and had to be redesigned for purpose of screening publications in our review. We adapted the version of van Tulder and van der Windt using the description of the design of a cross-sectional study of Chekoway to finalize the criteria<sup>[14,16,17]</sup>.

Different focuses of the studies and instruments used in NMD research related to employment status made it hard to compare the studies. Although not much research is available, the 44 factors related to employment we found in at least one publication might be very important in spite of the fact that no indicative evidence was found through the best evidence synthesis. Many factors are known from clinical experiences by the first, fourth and fifth author and from the perspective of patients. Factors such as solving work related problems, institute work adaptations, work accommodations,

workload, and job coaching/counselling are helpful to sustaining employment or helping to reintegrate NMD patients in the work force. From clinical experience and findings in one of the publications, it is also obvious that rehabilitation should focus on finding a match between physical and mental demands of a job on the one hand and work capacities of disadvantaged persons on the other hand. Therefore we suggest to create an adapted version of the Work and Handicap Questionnaire (WHQ) of Andries et al<sup>[33]</sup>. Combined with the indicative findings, the factors that were only present in one article, and the factors derived from clinical experience and patient perspective can be used in this adapted version of the WHQ. This questionnaire, or a core set of factors relevant for employment, can be used to investigate in a prospective cohort study for all kind of study populations. Outcomes will then also become internationally comparable. This will provide better scientific evidence to improve care and services by allied health professionals and organizations involved in the (re-)integration process. The value of vocational training for chronically ill employees to help them (re)enter the labour market, was already recommended more than a decade ago by Wevers<sup>[36]</sup> and in a recently published review from 1988-2004 by Varekamp et al<sup>[1]</sup>. Cup et al<sup>[45]</sup> found an underuse of referrals to OT and Speech therapy in NMD patients.

The codes of the ICF were used to categorize the main factors related to employment in the expanded ICF scheme developed by Heerkens et al<sup>[33]</sup>. All factors found could be accommodated by the scheme indicating that the expanded ICF scheme indeed works well to fill the gap between the terminology used by professionals in health care, and the terminology used by professionals in occupational medicine (table 2)<sup>[33]</sup>. Factors influencing employment such as general personal factors, work related personal factors, terms of employment and task content are not present in the ICF. Classification and coding within ICF terms was therefore not possible for these factors. MK & YH made a decision on how to classify these factors, realizing that other choices might be possible.

## Recommendations for future research

In order to support people with a chronic disease in maintaining a job or return to work, it is important to detect facilitators and barriers influencing the ability to work, especially factors that are within the scope of influence by allied health care interventions such as vocational therapy. We have used a way of investigating factors related to employment in NMD patients by searching for evidence according to the best evidence synthesis for quantitative studies. In this way our results become comparable with other reviews. Qualitative studies, such as case studies, case reports, clinical guidelines and narrative reviews might also be valuable to find relevant factors as well. A qualitative review could therefore be a profound next step in finding factors related to employment in NMD patients. Furthermore we propose the creation of an adapted version of the Work and Handicap Questionnaire (WHQ) of Andries et al<sup>[19]</sup>.

This will provide better scientific evidence to improve care and services by allied health professionals and organizations involved in the (re-)integration process.

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CLASSIFICATION OF EMPLOYMENT FACTORS

Appendix 1. Literature search

Databases were searched as described below:

	<b>MESH terms and free text words combined with 'OR'</b>	<b>'AND'</b>	<b>MESH terms and free text words combined with 'OR'</b>
PUBMED	Hereditary Motor and Sensory Neuropathy Myotonic Dystrophy Facioscapulo-humeral Muscular Dystrophy Facioscapulo-humeral neuromuscular disorder* neuromuscular disease*		work employment unemployment occupation absenteeism sick leave vocation
	<b>MESH terms and free text words combined with 'OR'</b>	<b>'AND'</b>	<b>MESH terms and free text words combined with 'OR'</b>
MEDLINE CINAHL PSYCHINFO ERIC database WEB OF SCIENCE COCHRANE Controlled Trials Register EMBASE	Neuromuscular dis* Myotonic dis* Myotonic Dystroph* Hereditary Motor Sensory Neuropath* Fascioscapulo-humeral*		Vocation* Occupation* Work* Absenteeism* Employ* Unemploy* Sick Leave*
	<b>Dutch free text words combined with 'OR'</b>	<b>'AND'</b>	<b>Dutch free text words combined with 'OR'</b>
PICARTA Doc.online	Neuromusculaire ziekten Neuromusculaire aandoeningen spierziekten		arbeid werk baan ziekteverzuim

**Appendix 2.** Factors of health, environmental factors and personal factors associated with employment in Neuromuscular Disease (NMD) patients. Health related items and environmental factors are coded according to ICD-10 and ICF if applicable

ICF Code	Main factors	Sub factors	Author	Association with employment present	Evidence synthesis
<b>A</b>	<b>HEALTH / HEALTH STATE</b>				
<b>A.1</b>	<b>(Work related) disease/disorder</b>				
	Difference between types of NMD	MD (ICD-10/G71.1)	Fowler, Böstrom <sup>a</sup>	no, yes	No evidence
	Type of NMD	HMSN (ICD-10/G60.0) FSHD (ICD-10/G71.0) Other type of NMD	Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> Fowler Fowler, Andries <sup>a</sup> , Andries <sup>b</sup>	yes, no, yes/no yes, no, yes/no yes yes, no (MG)/ yes (SMA), no no, no	Indicative findings Indicative findings No evidence Indicative findings
	Disease duration		Fowler, Böstrom <sup>b</sup>		No evidence
<b>A.2</b>	<b>Functions (incl. mental functions) &amp; structures (impairments)</b>				
	Higher number of complaints	Full Scale IQ *	Andries <sup>b</sup>	yes	No evidence
	Intellectual functions (ICF/b117)	Verbal IQ *	Fowler	yes	No evidence
		Performance IQ *	Fowler	yes	No evidence
		Anxious *	Fowler	no	No evidence
	Temperament and personality functions (ICF/b126)	Self-controlled *	Fowler	no	No evidence
		Sociable *	Fowler	no	No evidence
		Dominance *	Fowler	no	No evidence
		Capacity for status *	Fowler	yes	No evidence
		Social presence *	Fowler	no	No evidence
		Self-acceptance *	Fowler	yes	No evidence
		Responsibility *	Fowler	no	No evidence
		Tolerance *	Fowler	no	No evidence
		Communality *	Fowler	yes	No evidence
		Achievement via conformance *	Fowler	yes	No evidence
		Good impression *	Fowler	no	No evidence
		Achievement via independence *	Fowler	no	No evidence
		Intellectual efficiency *	Fowler	no	No evidence
		Psychological mindedness *	Fowler	no	No evidence
		Flexibility *	Fowler	no	No evidence
		Ego strength *	Fowler	no	No evidence

(continued)

Other mental functions	Attention functions (ICF/b140) Memory functions (ICF/b144) Higher-level cognitive functions (ICF/b164)	Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup> Fowler, Andries <sup>b</sup> Andries <sup>b</sup>	yes yes yes yes, yes yes	No evidence No evidence No evidence Indicative findings No evidence
Physical functions	Fatigue			
General physical endurance (ICF/b4550)	Articulation functions (ICF/b320)	Andries <sup>a</sup>	yes	No evidence
Voice and speech functions (ICF/b3)	Sensation of pain (ICF/b280)	Andries <sup>a</sup>	yes (MD)/no (HMSN)	No evidence
Pain and sensory functions (ICF/b2)	Seeing functions (ICF/b210) Hearing functions (ICF/b230) Muscle power functions (ICF/b730)	Andries <sup>a</sup> Andries <sup>a</sup> Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> , Ville	yes yes no, no, yes, yes	No evidence No evidence Indicative findings
Neuromusculoskeletal and movement related functions (ICF/b7)				
<b>A.3 Activities (limitations)</b>				
Mobility (ICF/d4)	Changing and maintaining body position (ICF/d410-d429) Carrying, moving and handling objects (ICF/d430-d449) Walking and moving (ICF/d450-d469)	Fowler, Andries <sup>a</sup> Fowler, Andries <sup>a</sup>	no, yes no, yes	No evidence No evidence
Communication (ICF/d3)				
Self care and domestic life (ICF/d5-d6)		Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup>	yes yes yes	No evidence No evidence No evidence
Severity of physical disability		Fowler Ville	yes	No evidence
Extent of physical disability		Fowler	yes	No evidence
Impact of disability on performance		Andries <sup>b</sup>	yes	No evidence
Total amount of limitations			yes	No evidence
<b>A.4 Participation (restrictions)</b>				
Remunerative employment (ICF/d850)	Periods of unemployment Length of time unemployed	Andries <sup>a</sup> Andries <sup>a</sup> Ville	no yes yes	No evidence No evidence No evidence
Perceived handicap	Time spent at social activities	Andries <sup>b</sup>	no	No evidence
Socializing (ICF/d9205)				

(continued)

ICF Code	Main factors	Sub factors	Author	Association with employment present	Evidence synthesis
<b>B</b>	<b>ENVIRONMENTAL FACTORS</b>				
<b>B.1</b>	<b>Work related environmental factors</b>				
	General characteristics of organization	Legislation Knowledge of employer Size of the company	Andries <sup>b</sup> Andries <sup>b</sup>	no no	No evidence No evidence
	Terms of employment	Work adaptations needed (unspecified) Work adaptations realized (unspecified) Working in shifts Working hours Working part-time Adapted working hours realized Chances for promotion	Andries <sup>a</sup> , Andries <sup>b</sup> Andries <sup>a</sup> , Andries <sup>b</sup> Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>b</sup> Andries <sup>a</sup>	yes, no yes, no no yes no yes	No evidence No evidence No evidence No evidence No evidence No evidence No evidence
	Social relationship at work	Consideration of superior (ICF/e330) Remarks of superior (ICF/e330) Consideration of colleagues (ICF/e330) Remarks of colleagues (ICF/e325) Taken seriously by colleagues (ICF/e325) General health perception of the employer	Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup> Fowler	no no no no no no	No evidence No evidence No evidence No evidence No evidence No evidence
	Task content	Tasks Physical demands Mental demands Autonomy Work adaptation	Andries <sup>a</sup> , Andries <sup>b</sup> Andries <sup>a</sup> Andries <sup>b</sup> Andries <sup>a</sup>	no, yes no yes no	No evidence No evidence No evidence No evidence
	Working conditions	Adaptation in task content, realized Adaptation in pace, realized Adaptation of tools, realized (ICF/e325) Adaptation of furniture, realized (ICF/e325)	Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup> Andries <sup>a</sup>	yes yes yes yes	No evidence No evidence No evidence No evidence
<b>B.2</b>	<b>Other environmental factors</b>				
	Home environment/social support/etc.	Negative experience social support (ICF/e3)	Andries <sup>b</sup>	yes	No evidence
	Health systems (ICF/e5801)	Referral patterns to department of rehabilitation % of acceptance in department of rehabilitation	Fowler Fowler	yes yes	No evidence No evidence

(continued)

<b>C. PERSONAL FACTORS</b>				
<b>C.1</b>	<b>General personal factors</b>			
	Age		yes, yes, yes/no, no	Indicative findings
	Gender		no, yes, no/yes	Indicative findings
	Education		yes, yes, yes/no	Indicative findings
	Marital status		no	No evidence
		Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> , Böstrom <sup>b</sup> Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> Fowler, Andries <sup>a</sup> , Andries <sup>b</sup> Böstrom <sup>b</sup>		
<b>C.2</b>	<b>Other diseases/co morbidities *</b>			
	Hypochondriasis * (ICD-10/F45.2)	Fowler	yes	No evidence
	Depression * (ICD-10/F32)	Fowler	yes	No evidence
	Hysteria * (ICD-10/F44)	Fowler	yes	No evidence
	Schizophrenia * (ICD-10/F20)	Fowler	yes	No evidence
	Psychopathic deviation * (ICD-10/F84.5)	Fowler	yes	No evidence
	Paranoia * (ICD-10/F60.0)	Fowler	no	No evidence
	Psychastenia * (ICD-10/F48)	Fowler	yes	No evidence
	Mania * (ICD-10/F30)	Fowler	no	No evidence
<b>C.3</b>	<b>Work related personal factors</b>			
	Type of occupation			
	Quality of work of employee (judgement employer)	Fowler, Andries <sup>b</sup> Andries <sup>b</sup>	yes, yes yes (HMSN)	Indicative findings No evidence
	Productivity of employee (judgement employer)	Andries <sup>b</sup>	yes	No evidence
	Flexibility of employee (judgement employer)	Andries <sup>b</sup>	yes (HMSN)	No evidence
	Expressed interest in employment by the NMD patient	Fowler, Andries <sup>b</sup>	yes, yes	Indicative findings
		Heavy labour/blue colour/commercial		

The evidence synthesis is based on the best evidence synthesis of Table 1.  
 Yes: Indicates the factor is present; in relation to employment.  
 No: indicates the factor is present, however not in relation to employment.  
 Yes/no = one factor is reported in one article as well with, as without significance,  
 All factors with\* = all types of NMD combined.  
 Andries<sup>a</sup> = Andries 2000, Andries<sup>b</sup> = Andries 1997;  
 Böstrom<sup>a</sup> = Böstrom 2005<sup>a</sup>, Böstrom<sup>b</sup> = Böstrom 2005<sup>b</sup>.  
 FSHD = Facioscapulo-humeral Dystrophy, HMSN = Hereditary Motor and Sensory Neuropathies,  
 ICD-10 = Classification of Diseases, ICF = International Classification of Functioning, Disability and Health,  
 IQ = Intelligence coefficient, MD = Myotonic Dystrophy, MG = Gravis, NMD = Neuromuscular Disease,  
 SMA = Spinal Muscular Atrophy.

**Appendix 3.** Example of an adapted Methodology Checklist

**Methodological criteria list for repeated survey studies**

**First author** : .....  
**Reviewer** : .....  
**Title** : .....  
**Source** : .....

+ / - / ?                      + = positive, - = negative, ? = not described or insufficiently described

*Objective of the study*

X 1		Positive if a specific, clearly stated objective is described.
-----	--	--

*Study population*

X 2		Positive if description of inclusion or exclusion criteria is adequate
-----	--	--

V 3		Positive if the main features of the study population (cohort definition) are described (sampling frame and distribution of the population).	
		Sampling frame T0 =	Sampling frame T1 =

V 4		Positive if the main features of the study population (cohort definition) are described (sampling frame and distribution of the population by age and sex).	
		Age: Males:                      Females:	Age: Males:                      Females:

O 5		Was the composition of the population of each of the surveys similar with respect to relevant responder characteristics / exposures to employment factors.
-----	--	--

O 6		The number of non-responders is described and acceptable: Positive if the participation rate at baseline is at least 80% or if participation rate is 60-80% and non-response is not selective (data presented).                      .....%
-----	--	--

O 7		The number of non-responders is described and acceptable: Positive if the response at main moment of follow up is at least 60-80% or if non-response is not selective (data presented).                      .....%
-----	--	--

*Exposure Measurements: Factors associated with employment*

X 8		Method for measuring factors related to employment: <input type="radio"/> direct measurement (+) <input type="radio"/> observation (+) <input type="radio"/> self-reported (-) <input type="radio"/> questionnaire (+)
-----	--	--

O 9		Repeated measure methods involve similar health and exposure methods
-----	--	--

*Measurements*

X 10		Positive if the employment measures are relevant and defined adequately
------	--	---

X 11		Positive if data were collected for at least one year or at least every three month
------	--	---

(continued)

CLASSIFICATION OF EMPLOYMENT FACTORS

*Analysis and Data Presentation*

V 12		<p>The statistical analyses are adequately chosen in relationship to the research question and data presented:          Positive if the appropriate statistical model is used for continuous data (uni or multivariate model).          Student t-test or ANOVA          Linear regression analyses is used to determine the most important variables.          Positive if measures of association are presented for dichotomous outcomes:          Prevalence ODD's ratio (POR)          Prevalence ratio (PR)          Prevalence difference (PD) including confidence interval, p-value, and numbers in the analysis.          Logistic regression analyses is used to determine the most important variables.</p>	
V 13		<p>The statistical analyses are sufficiently powered:          e.g. Positive if the number of cases in the multivariate analysis is at least ten times the number of independent variables in the analysis (final model).</p>	# =

<b>Quality Score</b>	<b>X = descriptive (5)</b>	<b>V = statistics (4)</b>	<b>O = internal validity (4)</b>	<b>Quality</b>
Total '+':				
Total '-':				
Total '?':				





## **CHAPTER III**

### **Employment Status of Patients with Neuromuscular Diseases in relation to Personal factors, Fatigue and Health Status: A Secondary Analysis**

Published as

Minis MA, Kalkman JS, Akkermans RP, Engels JA, Huijbregts PA, Bleijenberg G, Oostendorp RA, van Engelen BG. Employment status of patients with neuromuscular diseases in relation to personal factors, fatigue and health status: a secondary analysis *J Rehabil Med* 2012;42(1):60-5.



## Abstract

*Objective:* To determine the number of employed people in a group of patients with neuromuscular diseases and in 3 separate subgroups (facioscapulo-humeral muscular dystrophy, hereditary motor and sensory neuropathy, and myotonic dystrophy) to investigate any differences in employment status between the patient groups, and to identify factors related to employment status.

*Design:* Cross-sectional study.

*Patients:* A total of 591 patients with neuromuscular diseases participated in the study, 138 with facioscapulo-humeral muscular dystrophy, 135 with hereditary motor and sensory neuropathy, and 318 with myotonic dystrophy.

*Methods:* Self-report questionnaires, the Checklist Individual Strength (CIS) and the Short Form-36 (SF-36).

*Results:* Of the patients with neuromuscular diseases in the study, 56.7% were employed. Younger age, being male, and higher education contributed significantly to employment status of the neuromuscular diseases group and the hereditary motor and sensory neuropathy and myotonic dystrophy subgroups. Significant between-group differences for employed vs not employed subjects were present in the total neuromuscular diseases group on all subscales of the CIS and SF-36. Factors related to employment status differed for the 3 neuromuscular diseases subgroups.

*Conclusion:* More than half of the patients with neuromuscular diseases were employed. Patients with facioscapulo-humeral muscular dystrophy and patients with hereditary motor and sensory neuropathy were more often employed than patients with myotonic dystrophy. Between-group analyses for differences in baseline factors revealed 11 significant factors related to employment. Multivariate logistic analyses revealed 6 factors contributing to employment for the group of patients with neuromuscular diseases.

## Introduction

Employment is recognized worldwide as important to all people, including those with disabilities and chronic diseases. People in gainful employment, again including those with chronic diseases, enjoy greater health, wellbeing, and sense of fulfillment in their lives [1-8]. Healthcare professionals, politicians, and patient organizations all recognize the value of empowering people with a disability by way of gainful employment.

Three moderately prevalent progressive neuromuscular diseases (NMD) were chosen for this study based on their onset at working age with a progressive impact on performing daily activities that is also likely to affect gainful employment [9,10]. Facioscapulo-humeral muscular dystrophy (FSHD) is a genetic muscular dystrophy. Hereditary motor and sensory neuropathy type I (HMSN) is a genetic peripheral nerve disease. Adult-onset myotonic dystrophy (MD) is a genetic multi-organ disorder also

affecting cognitive function. A systematic review revealed 10 factors associated with employment status: disorder-related factors, factors related to functions, general personal factors, and work-related personal factors <sup>[11]</sup>. This study is a secondary analysis of data from the study by Kalkman et al. <sup>[10]</sup>. We selected from this study those measurement instruments that were likely to impact on employment status.

This analysis seeks to answer the following research questions: (i) How many NMD patients of working age are gainfully employed and what are their characteristics? (ii) Are there differences in employment status among the 3 subgroups of patients with FSHD, HMSN, and MD? (iii) Which disease related, personal, fatigue severity, and functional health status factors, are related to employment of the NMD patient group and the 3 patient subgroups?

## Methods

### *Subjects*

In their cross-sectional study Kalkman et al. <sup>[10]</sup> contacted patients with a definitive medical diagnosis of NMD using patient databases from the Neuromuscular Centre Nijmegen and the Dutch Neuromuscular Patient Association (Vereniging Spierziekten Nederland). With a response rate of 72%, these researchers were able to recruit 598 patients with FSHD, HMSN and MD aged 18–68 years. The study was approved by the local ethics committee and all subjects signed an informed consent. The sample was described in terms of general personal factors, participation in volunteer work, fatigue, functional health status, and employment status. For this secondary analysis examining employment status, one patient was excluded for being over the retirement age of 65 years, and 6 patients were excluded due to incomplete or missing data with regard to employment status. Thus, 591 patients were included in this secondary analysis: 138 patients with FSHD, 135 with HMSN, and 318 with MD.

### *Measurement instruments*

For this analysis those data from measurement instruments used in the primary study <sup>[10]</sup> that were likely to be associated with employment status were selected. These included data from a patient questionnaire, the Checklist Individual Strength (CIS), and 4 subscales of the Short Form-36 (SF-36) functional health status measure.

### *Patient questionnaire*

For the original study, employment was defined as the state of being engaged in an activity or service for salary (gainful employment). In the questionnaire used by Kalkman et al. <sup>[10]</sup> 5 categories of employment status were defined: 0–8, 9–16, 17–24, 25–32 or > 32 h/week. Although this cut-off value does not correspond to the Dutch governmental recognition of economic importance of working  $\geq 12$  h/week <sup>[12]</sup>, for this secondary analysis the data available on employment status were dichotomized into

either employed (working  $\geq 9$  h/week) or not employed (working 0–8 h/week). Marital status was dichotomized as single or domestic union/married. Participation in volunteer work was dichotomized into performing volunteer work or not, because no data on the number of hours of voluntary work was included in the primary study. Data collected on highest educational level attained consisted of 3 categories: lower educated (primary school), intermediate educated (secondary school), and higher educated (post-secondary level).

#### *Checklist Individual Strength*

The CIS is a multidimensional instrument with 4 subscales (fatigue severity, concentration problems, reduced motivation, and reduced activity) to measure the level and impact of fatigue in patients with chronic diseases and in healthy people<sup>[13–15]</sup>. Each subscale item can be scored on a 7-point Likert scale. A higher composite subscale score indicates higher levels of fatigue, higher levels of concentration problems, lower motivation, and lower levels of activity<sup>[13]</sup>. Although in the literature the fatigue scale has been dichotomized, with a score  $\geq 35$  used to identify severe fatigue<sup>[10,14]</sup>, all subscale scores were analyzed here as ratio-level data. The total CIS score has demonstrated good reliability and validity<sup>[16,17]</sup>.

#### *Functional health status*

The Medical Outcomes Study Short Form General Health Survey (SF-36) uses 36 questions to measure 8 aspects of functional health status<sup>[18]</sup>. Item scores are added and transformed to a 100-point scale to produce subscale scores. Higher scores indicate better health status.

For this secondary analysis 4 subscales were used that were thought to be relevant to employment status: physical functioning (10 items), social functioning (2 items), vitality (4 items), and bodily pain (2 items). The SF-36 has been shown to have adequate reliability and validity, both for total and subscale scores<sup>[18]</sup>.

#### *Data analysis*

Descriptive statistics are presented separately for all variables of employed and not employed NMD patients. These descriptive data were analysed for between-group differences with univariate analyses (ANOVA) for numeric and continuous variables and  $\chi^2$  analyses for ordinal variables both for the total NMD group (table 1) and for the 3 subgroups (table 2). We regarded  $p < 0.05$  as statistically significant. Further statistical analyses were performed for the total NMD group and for the 3 subgroups with employment status as the dependent variable. Stepwise forward multivariate logistic regression analyses using SPSS software (Version 15.0 for Windows®) were performed to examine the contribution of the variables to employment status for the total NMD group and for the 3 patient subgroups, allowing us to present odds ratios and calculate variance (table 3). In the stepwise procedure we used  $p < 0.15$  for inclusion and  $p > 0.05$  for exclusion of variables in the model.

## Results

Data on personal factors, level and impact of fatigue, and functional health status for the total NMD group of employed, and not employed NMD patients are presented in table 1. The results for the 3 subgroups are presented in table 2. In table 3 the contribution of different factors to employment status is presented for the total NMD group and the subgroups.

**Table 1.** Personal factors, level of fatigue, and health status of the total with patient group neuromuscular diseases (NMD) employed and not employed (n=591)

Variable	Employed†	Not employed†	p-value*
Total, % (n)	56.7 (335)	43.3 (256)	
Age mean (SD)	40.4 (9.96)	46.1 (9.52)	< 0.001
Gender % (n)			
Men	52.8 (177)	35.2 (90)	< 0.001
Women	47.2 (158)	64.8 (166)	
Marital Status, % (n)			
Single	31 (104)	27.1 (69)	0.291
Living together/married	69 (231)	72.9 (187)	
Education level, % (n)			
Lower	18.8 (63)	40.8(104)	< 0.001
Intermediate	50.7 (170)	47.8 (122)	
Higher	30.4 (102)	11.4 (29)	
Volunteer, % (n)			
Yes	23.5 (78)	19.6 (50)	0.269
No	76.5 (254)	80.4 (205)	
CIS mean (SD)			
- CIS-fatigue (8-56)	36.5 (12.17)	41.7 (11.44)	< 0.001
- CIS-concentration (5-35)	13.4 (7.72)	17.3 (9.04)	< 0.001
- CIS-motivation (4-28)	13.2 (6.35)	16.5 (6.76)	< 0.001
- CIS-activity (3-21)	9.8 (5.32)	13.9 (5.57)	< 0.001
SF-36 Health Status‡ mean (SD)			
- Physical functioning (0-100)	52.5 (27.63)	43.8 (29.08)	< 0.001
- Social functioning (0-100)	72.4 (22.65)	66.8 (25.74)	0.006
- Vitality (0-100)	49.0 (19.22)	45.2 (19.82)	0.017
- Bodily pain (0-100)	74.0 (23.73)	69.9 (26.02)	0.046

Differences in proportions tested with Chi-squares; group means tested by ANOVA's

\* p-value <0.05 was regarded as statistically significant. † Employment defined as working >9 h/week; ‡ Not employed defined as working < 9 h/week

‡ SF-36. A higher value indicates a better health status on a scale of 0-100

CIS: Checklist Individual Strength; SF-36: 36-item Short Form Health survey;

SD: standard deviation

**Table 2.** Personal factors, level of fatigue and health status of patients with facioscapulo-humeral muscular dystrophy (FSHD), hereditary motor and sensory neuropathy (HMSN) and myotonic dystrophy (MD) employed and not employed

Variable	FSHD n = 138			HMSN n = 135			MD n = 318		
	Employed†	Not employed†	p-value*	Employed†	Not employed†	p-value*	Employed†	Not employed†	p-value*
Total, % (n)	70.3 (97)	29.7 (41)	< 0.001	63.7 (86)	36.3 (49)	0.001	47.8 (152)	52.2 (166)	0.432
Total, 95, % CI	39.98-44.03	44.61-50.46		38.27-42.70	42.61-48.58		37.84-40.95	44.55-47.43	
Mean age (SD)	42 (10.05)	47.5 (9.26)	0.003	40.5 (10.33)	45.6 (10.38)	0.007	39.4 (9.61)	45.9 (9.35)	< 0.000
Gender, % (n)									
Men	55.7% (54)	31.7 (13)	0.010	43 (37)	32.7 (16)	0.235	56.6 (86)	36.7 (61)	< 0.000
Women	44.3 % (43)	68.3 (28)		57 (49)	67.3 (33)		43.4 (66)	63.3 (105)	
Marital Status, % (n)									
Single	24.7 % (24)	26.8 (11)	0.797	32.6 (28)	30.6 (15)	0.815	34.2 (52)	26.1 (43)	
0.114									
Living together/married	75.3 % (73)	73.2 (30)		67.4 (58)	69.4 (34)		65.8 (100)	73.9 (122)	
Education level, % (n)									
Lower	14.4 (14)	31.7 (13)	0.005	11.6 (10)	18.4 (9)	0.294	25.7 (39)	49.7 (82)	< 0.000
Intermediate	48.5 (47)	56.1 (23)		50.0 (43)	55.1 (27)		52.6 (80)	43.6 (72)	
Higher	34.1 (36)	12.2 (5)		38.4 (33)	26.5 (13)		21.7 (33)	6.7 (11)	
Volunteer work % (n)									
Yes	33.0 (32)	26.8 (11)	0.475	26.7 (23)	31.3 (15)	0.579	15.4 (23)	14.5 (24)	0.808
No	67.0 (65)	73.2 (30)		73.3 (63)	68.8 (33)		84.6 (126)	85.5 (142)	
CIS mean (SD)									
CIS-fatigue (8-56)	34.8 (12.10)	41.4 (10.90)	0.003	37.0 (12.33)	36.8 (13.08)	0.929	37.3 (12.10)	43.2 (10.70)	< 0.000
CIS-concentration (5-35)	11.6 (6.99)	14.1 (10.29)	0.105	13.0 (8.28)	15.2 (9.27)	0.169	14.7 (7.65)	18.6 (8.36)	< 0.000
CIS-motivation (4-28)	11.8 (5.50)	13.0 (6.54)	0.257	11.9 (5.84)	12.7 (6.45)	0.459	14.9 (6.75)	18.4 (6.07)	< 0.000
CIS-activity (3-21)	8.9 (4.77)	13.0 (5.80)	< 0.00	1 9.1 (5.58)	9.9 (5.35)	0.450	10.8 (5.37)	15.3 (4.96)	< 0.000
SF-36 Health Status ± mean (SD)									
Physical functioning (0-100)	52.2 (30.67)	29.0 (26.41)	< 0.001	53.8 (24.67)	53.9 (28.53)	0.974	52.0 (27.33)	44.6 (28.58)	0.019
Social functioning (0-100)	74.1 (22.55)	67.9 (25.47)	0.164	68.7 (24.61)	66.2 (23.73)	0.568	73.3 (21.46)	66.7 (26.49)	0.015

Differences in proportions tested with 2, group means tested by analysis of variance (ANOVA).  
 \* p-value <0.05 was regarded as statistically significant. † Employment defined as working >9 h/week; Not employed defined as working < 9 h/week.  
 ‡ SF-36. A higher value indicates a better health status on a scale of 0-100; CI: confidence interval; CIS: Checklist Individual Strength; SF-36: 36-item Short Form Health survey; SD: standard deviation.



*Between-group analysis*

*Employment status.* In the total NMD group, 56.7% of patients were gainfully employed. Employment status differed significantly from this percentage for the total group for both the FSHD (70.3% employed;  $p < 0.001$ ) and the HMSN (63.7%;  $p = 0.001$ ) subgroups, but not the MD subgroup (47.8%;  $p = 0.432$ ) (table 1 and 2).

*Gender and age.* Between-group analysis showed significant differences with regard to gender and age: NMD patients who were employed were younger and more often male than the not employed patients (table 1). In all 3 subgroups, younger patients were employed to a significantly greater degree than older patients. For the FSHD and MD subgroups, the correlation between being male and being gainfully employed was also significant (table 2).

*Participation in volunteer work.* The employed NMD patients did not perform significantly more volunteer work than not employed patients (table 1 and 2).

*Marital status.* No significant differences were found for marital status for the total NMD group or for the 3 subgroups between employed and not employed patients (table 1 and 2).

*Educational level.* Significant between-group differences were noted in the level of education for the total NMD patient group. Employed NMD patients were, on average, significantly higher educated than not employed patients. Of the employed NMD patients, 30.4% had higher, 50.7% intermediate, and 18.8% lower education. For the not employed patients these percentages were 11.4%, 47.8%, and 40.8%, respectively (table 1). Of the employed patients, 81.1% had higher or intermediate educational background versus 59.2% in the non-employed group (table 1). Significant between-group differences with regard to educational level were also noted in the FSHD and MD-subgroups (table 2).

*Checklist Individual Strength.* Significant between-group differences for the 4 subscales of the CIS were present in the total MD group. Employed NMD patients reported significantly less fatigue, fewer concentration problems, better motivation, and higher levels of activity than the not employed NMD group (see table 1). Of note was that both employed and not employed NMD patients reported severe fatigue (cut-off  $\geq 35$  on the CIS fatigue subscale) (table 1). Despite this CIS fatigue score (mean 36.5, SD 12.1) a high percentage of NMD patients were still gainfully employed.

Subgroup analysis also showed significant between-group differences in the FSHD and MD subgroups with regard to fatigue severity level. Employed FSHD and MD patients reported significantly less fatigue compared with not employed patients. The fatigue score for the employed FSHD group (mean 34.8, SD 12.1) was below the cut-off for severe fatigue (table 2). The employed FSHD and MD patient group also reported significantly higher levels of activity. The employed MD patient group reported significantly fewer concentration problems and better motivation (table 2). No significant between-group differences were present for the HMSN subgroups for the 4 CIS subscales (table 2).

**Table 3.** Logistic regression analyses to predict employment status in the total neuromuscular diseases group and for the 3 patient groups: facio-oscipulo-humeral muscular dystrophy (FSHD), hereditary motor and sensory neuropathy (HMSN) and myotonic dystrophy (MD)

		Dependent variable employment							
		NMD n = 579 (256/335)*		FSHD n = 138 (41/97)*		HMSN n = 135 (49/86)*		MD n = 318 (166/152)*	
Independent variables	OR (95% CI)	p-value	OR (95% CI)	p-value	OR (95% CI)	p-value	OR (95% CI)	p-value	
Age	0.94 (0.92-0.96)	< 0.001			0.94 (0.89-0.98)	0.001	0.97 (0.89-0.96)	< 0.001	
Gender (male)	0.33 (0.22-0.49)	< 0.001					0.28 (0.161-0.508)	< 0.001	
Marital status									
Education									
Lower	1	< 0.001	1	0.015			1	< 0.001	
Intermediate	1.9 (1.21-3.01)	< 0.001	1.2 (0.41-3.66)	0.008			2.2 (1.25-4.13)	< 0.001	
Higher	5.6 (3.12-10.37)	0.005	6.6 (1.64-26.69)	0.708			9.9 (3.93-25.19)	0.007	
Volunteer									
Volunteer work									
CIS									
CIS- Fatigue									
CIS- Concentration	0.97 (0.95-0.99)	0.026			0.95 (0.91-0.99)	0.023			
CIS- Motivation									
CIS- Activity	0.89 (0.86-0.93)	< 0.001			0.86 (0.79-0.94)	0.001			
SF-36 Health Status									
Physical functioning	1.072 (> 1.000-1.149)	0.050			1.233 (1.047-1.452)	0.012			
Social functioning									
Vitality									
Bodily pain									
Nagelkerke R <sup>2</sup>	0.372		0.397		1.26 (1.077-1.490)	0.004	0.424		

\* Not employed (0-8 h)/Employed (9-40 h).

R<sup>2</sup> is the percentage of variance of the dependent variable score, explained by the independent variables together.

Relationship between patient factors with the dependent variable employment (p < 0.05).

Only significant odds ratios with accompanying 95% confidence interval.

CIS: Checklist Individual Strength;

SF-36: 36-item Short Form Health Survey

*Functional health status.* For the total NMD group between-group analysis showed significant differences in health status. The employed NMD patient group had a significantly higher score on the SF-36 subscales physical functioning, social functioning, vitality, and bodily pain compared with the not employed patient group, indicating a better health status (table 1).

Subgroup analysis showed more varied between-group differences. The employed FSHD group reported significantly better physical functioning, more vitality, and less bodily pain than the not employed FSHD group. The employed HMSN group reported significantly less bodily pain. The employed MD group reported significantly better physical and social functioning (table 2).

#### *Multivariate analysis*

*Total neuromuscular diseases group.* All of the factors that were found to contribute significantly to employment status from the multivariate logistic regression for the total NMD group are shown in table 3, presented as odds ratios (OR). Factors that contributed significantly to employment status for the total NMD patient group were age, gender, and educational level, CIS concentration score, CIS activity score, and SF-36 physical functioning score. Together these factors explained 37.2% of variance with regard to employment status. For the total NMD group the odds of being employed decreased 6% for each year with increasing age, controlling for other variables in the model. The odds for a male being employed were 3 times higher than for a woman, and higher educated NMD patients had 5.6 higher odds of being employed than lower educated NMD patients (table 3). The odds of being employed increased 3% with each higher level of CIS concentration (expressed in a lower score OR 0.97) and the odds of being employed increased 11% with each higher level of CIS activity (expressed in a lower score OR 0.89). The odds of being employed increased 0.72% with each unit increase of the score on the subscale SF-36 physical functioning (table 3).

*Subgroups.* Similar analyses were performed to predict employment status for all 3 subgroups. For the FSHD group, male patients with a higher education, higher CIS activity score, and better SF-36 physical functioning score were more likely to be employed: 39.7% of the variance was explained by these factors (table 3). For the HMSN group age, higher levels of concentration and less pain contributed significantly to employment status: 20.6% of the variance was explained by these factors (table 3). For the patients with MD, younger males with higher education levels and higher CIS activity score were most likely to be employed: These factors explained 42.4% of the variance in employment status (table 3). Of note was that educational level was an important explanatory variable for employment in both the FSHD and MD subgroups (OR 6.6–9.95).

## Discussion

In contrast to earlier published reports <sup>[9,19]</sup>, this study indicates that a high percentage of patients with NMD are gainfully employed, particularly in the FSHD and HMSN subgroups. A lower employment percentage was confirmed only for the MD subgroup in this study. In addition, it was found that employed NMD patients are younger, more often male, and higher educated than the not employed NMD patients. Four factors identified in a systematic review of the literature on factors associated with employment status in patients with NMD <sup>[11]</sup> were consistent with the findings of this study: NMD type, age, gender, and educational level. This impact of age is also in agreement with the findings by Andries <sup>[20]</sup>, who reported a more rapid decrease in employment status in patients with NMD increasing age.

In our study the HMSN and patient with MD subgroups reported a significant relationship between age and employment status. Furthermore, being male was a relevant factor for the FSHD and MD subgroups. On average, the employed MD patients had attained a lower educational level than the other 2 subgroups. We hypothesize that the fact that MD is a multi-organ disorder that also affects cognitive function might explain this lower educational level and, related to the nature of this pathology, lower employment level compared with the FSHD and HMSN subgroups <sup>[21]</sup>. Fowler et al. <sup>[9]</sup> also noted a correlation between the type of NMD and educational level. They stated that patients with higher education were more frequently working in clerical positions. Clerical jobs are less physically strenuous than heavy manual jobs, and higher education thereby provides a plausible explanation for employment status.

We expected to find a lower educational level in the MD subgroup, based on the nature of the pathology. Lower educational levels were expected to result in a higher percentage of these patients working in more strenuous jobs, leading to earlier disability pension and a lower employment rate <sup>[10,19]</sup>. In this study the percentage of higher educated patients with MD was found to be low. It was notable that the chance of being employed was almost 10 times better for the patients with MD higher education compared with those with lower education. The clinical implication of this finding is that special attention appears warranted to explain to patients with MD at an early age the importance of educational level and to encourage them to study, if their condition allows it, as education has been found to have a strong correlation with future opportunities in the labor market.

The variance explaining employment status for the contributing factors identified was relatively high for the FSHD (39.7%) and the MD (42.4%), but more limited for the HMSN subgroup (20.6%). Of all the factors included in the regression analyses, none were relevant for all 3 subgroups. Education was shown to be the most important variable in the FSHD and MD subgroup in explaining association with employment status. Age was an important variable for the HMSN and MD subgroups and gender for the FSHD and MD subgroups. Severe fatigue and pain are major complaints in patients with NMD <sup>[10, 22–28]</sup> and we expected that fatigue and pain would influence employment

status in the NMD patient groups. The variability with regard to fatigue severity for type of NMD was large. However, despite high levels of fatigue severity, the levels of employment were also relatively high. This seems to indicate that fatigue severity may not be the reason why the patients in this study were not participating in the workforce. There was a significant between-group difference with regard to reported pain in the total NMD group and in the FSHD and HMSN subgroups. However, less pain was a contributing factor only with regard to employment for the HMSN subgroup. In the regression analyses the CIS fatigue and motivation subscale scores did not contribute to the calculated variance in employment status in any of the 3 subgroups. This study indicates that fatigue severity and motivation do not affect participation in the workforce for patients with MD.

A significant between-group difference was found between the 4 SF-36 subscales: physical functioning, social functioning, vitality, and bodily pain. Subscale scores were higher in those patients who were employed, indicating better health status. Regression analysis detected the subscale factor SF-36 physical functioning as a variable that could partly explain the variance of employment status in the total NMD group and in the FSHD subgroup. We suggest that if factors could be found that influence employment in patients with NMD, it might be helpful in the development of treatment strategies for (allied) health professionals to assist patients with NMD to return to work and/or to stay employed.

Although 11 factors that influenced gainful employment status were found in the sample of patients with NMD studied, 6 of which were also found to be relevant using multivariate logistic analyses, a maximum of only 42.4% of the variance was explained, which leaves 57.6% unexplained with the factors in the present model. Due to the limitations with regard to data selected, additional relevant predictive factors influencing employment were probably not taken into account.

Environmental factors are, for example, not encountered in this study and might influence employment status in NMD patients, as was found in other cross-sectional studies [4, 9, 12, 29–31]. Other studies also recommend that determinants such as adequate referral patterns, goal setting, motivation, expectation, job seeking, work maintenance, work demands, terms of employment (type of job, amount of hours worked, ability to regulate working hours) should be addressed [9, 12, 32, 33]. Social support, facilitation, and compensation for barriers to employment when disabled and chronically ill patients such as NMD patients attempt to return to work also need to be addressed in future studies [21, 33, 34]. This stresses the importance of specific qualitative research to identify further modifiable variables related to employment in order to enhance labour participation of the patient group in our study.

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## **CHAPTER IV**

### **Exploring employment in consultation reports of patients with Neuromuscular Diseases**

Published as

Minis MA, Cup EH, Heerkens YF, Engels JA, van Engelen BG, Oostendorp RA.  
Exploring Employment in Consultation Reports of Patients with Neuromuscular  
Diseases

*Arch Phys Med Rehab* 2012;93(12):2276-80.



## Abstract

*Objectives:* To explore consultation reports for patient and employment characteristics and recommendations on employment regarding patients with neuromuscular diseases (NMDs).

*Design:* Retrospective study of multidisciplinary reports.

*Setting:* An outpatient neuromuscular clinic at a university hospital.

*Participants:* Reports (N=102) of patients with NMDs.

*Interventions:* Based on one-off consultations by occupational therapists, physical therapists, and speech therapists and a multidisciplinary meeting, recommendations were developed regarding therapy content and volume in primary care or rehabilitation settings.

*Main Outcome Measures:* A checklist has been developed to examine employment characteristics. A general questionnaire has been used including demographic variables and data on employment.

*Results:* Of the 102 reports available, 86 were included for analysis. Sixty-nine reports contained information on employment. Thirty-seven patients (43%) with NMD were employed, most in white-collar or moderately strenuous jobs. Of the 37 employed patients, 28 (76%) worked using adaptations. Thirty-two (87%) had employment problems; of these, 15 (40%) needed improvement in 1 or more environmental factors. Twenty patients (54%) needed advice regarding participation in employment, of whom 19 were referred to primary care or rehabilitation settings for treatment to enhance employment participation.

*Conclusions:* Eighty percent of the included consultation reports contained information on employment. Less than half the patients with NMD were employed, most in office-related jobs, using some kind of adaptations. Nineteen of 20 patients who agreed to recommendations regarding therapy were adequately referred by occupational therapists and physical therapists for treatment of employment problems.

## Introduction

Employment plays a major role in the lives of people. This is not only the case for healthy people but also for people with a chronic progressive disease, such as a neuromuscular disease (NMD) <sup>[1, 2]</sup>. In a systematic review <sup>[3]</sup> of factors influencing employment of patients with NMD, few studies were found in which patients with a slowly progressive NMD were subject of investigation. In addition to factors affecting employment of healthy people, such as age, sex, and education, the type of NMD also influenced possibilities for employment <sup>[4]</sup>. In a study of 591 patients with NMD, employment status varied from 47.8% for patients with myotonic dystrophy, to 63.7% for patients with hereditary motor and sensory neuropathy type I, to 70.3%

for patients with facioscapulo-humeral muscular dystrophy <sup>[4]</sup>. Early identification of the aforementioned factors is important in facilitating continued engagement in employment activities for patients with a progressive disease such as NMD. Because of the slow progressive deterioration, patients with NMD need regular support (e.g., by a job coach or case manager) to maintain employment participation <sup>[4]</sup>. Employment was defined as “the state of being engaged in an activity or service in exchange for wages or salary” <sup>[5]</sup>. Based on their professional domain, occupational therapists, physical therapists, and speech therapists, supporting chronically ill patients in their daily performance may play a role in preventing job loss <sup>[6]</sup>. Occupational therapists, physical therapists, and speech therapists are in a position to identify participation problems related to employment <sup>[6]</sup>. If needed, these therapists can also refer patients to occupational health or vocational rehabilitation services <sup>[6]</sup>.

Generally, patients with NMDs visit neurologists for medical diagnoses and treatment, not primarily for participation problems regarding employment. After a visit to the neurologist, a group of patients with NMD were consecutively referred to a specialized team of occupational therapists, physical therapists, and speech therapists for a one-off consultation <sup>[7]</sup>. Based on the results of these consultations and a multidisciplinary meeting in which a neurologist, a rehabilitation physician, and the therapists participated, treatment recommendations were formulated specifying content (including frequency) by occupational therapy (OT), physical therapy (PT), and speech therapy (ST) in primary care or rehabilitation settings <sup>[7]</sup>. There is a lack of knowledge regarding the contribution of occupational therapists, physical therapists, and speech therapists in optimizing employment participation of patients with NMDs. The aim of this study was to explore the consultation reports for patient characteristics and employment characteristics, as well as frequency and content of recommendations on employment, of patients with NMDs.

## Methods

### *Design and Setting*

A retrospective analysis was performed using multidisciplinary reports of 102 consecutive patients with NMD visiting the Neuromuscular Centre Nijmegen from December 2002 until June 2003. Inclusion criteria were: Probable or definite NMD according to the medical records; age 18 years or older; and sufficient command of the Dutch language <sup>[7]</sup>. Reports of patients with a fast progressive type of NMD, such as amyotrophic lateral sclerosis <sup>[8]</sup> and of patients older than 64 years were excluded from analyses.

The multidisciplinary reports consisted of 5 sections: a semi-structured general section containing diagnosis, demographic data, marital status, data on education, leisure activities, work status, and presence of personal equipment or adaptations; 3 specific sections with findings of OT, PT, and ST consultations; and a fifth section

with general conclusions and recommendations for treatment (including volume) based on the one-off consultations and multidisciplinary meetings.

#### *General Questionnaire*

A general questionnaire including demographic variables (age, sex, marital status, education) and information related to the disease, such as medical diagnosis and duration of complaints, was used. Specific items of the general questionnaire related to paid employment were as follows: employment status (yes/no), source of income (job income, allowance, both job income and allowance, or no personal income), weekly job hours (8-12h, 13-20h, 21-40h), monthly income (€ 1000, € 1000-€ 1500, € 1500-€ 2000, >€ 2000, does not know or wants to keep it private), weekly hours of sick leave (4-12h, 13-20h, 21-40h), percentage working disability (8%-50%, 51%-80%, 81%-100%), and patient status currently under review for disability percentage assessment <sup>[7,9]</sup>.

#### *Procedure*

For analysis of employment problems, the multidisciplinary reports were made anonymous and numbered. For analysis of the reports, a checklist was developed (Appendix 1). The first researcher (M.A.M.) analyzed all reports, and 2 other raters (Y.F.H. and R.A.O.) each rated half of the reports. Meetings were used to reach consensus for detailed exploration of employment problems. The procedure consisted of 6 steps:

- Step 1: Selection of reports eligible for analysis.
- Step 2: Exploration of the presence of information on employment in the different sections of the reports.
- Step 3: Division of the reports into 2 groups: “not employed” patients and “employed” patients. Regarding patients who were not employed, reasons for unemployment were recorded. The reports on employed patients were checked in more detail; that is, they were examined to identify employment problems and type of job (office-related white-collar jobs, moderately strenuous jobs for patients with NMD such as nursing and teaching, and labor-intensive blue-collar jobs) <sup>[10]</sup>. If patients worked under preconditions of the presence of adaptations, this was also registered.
- Step 4: Examination of the reports pertaining to work-related environmental factors that needed improvement; that is, terms of employment, task content, working conditions, and social relationships at work <sup>[11]</sup>.
- Step 5: Examination of whether therapists made recommendations regarding the employment for patients with NMD. Patients’ indications that recommendations were not yet necessary or their decisions to postpone them were also registered.
- Step 6: Examination of the reports regarding recommendations for further treatment, patients’ consent regarding these recommendations, and how many patients were referred accordingly.

The Medical Ethics Committee of the Radboud University Nijmegen Medical Centre in the Netherlands approved the research project in accordance with the Helsinki Declaration <sup>[12]</sup>.

### *Statistical analysis*

Percentage agreement regarding the presence of information on employment in the reports between M.A.M., Y.F.H., and R.A.O. was calculated. For the general questionnaire of the employed and not employed groups, descriptive data were presented. These descriptive data were tested for between-group differences regarding age, sex, marital status, education level, medical diagnosis, or duration of complaints with an unpaired t test for the continuous variable 'age' and chi-square analyses for the nominal variables.

A p-value of < .05 was considered statistically significant. If data were missing, patients were excluded from analyses. The variables with empty cells or only applicable for the employed were excluded from the table and described in the text. All statistical analyses were performed with the statistical package SPSS for Windows (Version 17.0).

## **Results**

Of the 102 multidisciplinary reports of consecutively referred patients with NMDs, 86 were eligible for analysis. Sixteen reports were not eligible for analysis because of age ( $\geq 64$ y) or the progressive type of NMD such as amyotrophic lateral sclerosis (fig 1, step 1). Two consensus meetings between M.A.M., Y.F.H., and R.A.O. were necessary to reach 100% consensus about the presence of information on employment in 1 or more sections of the multidisciplinary reports (see figure 1).

The presence of work-related information was found in the general section of 69 multidisciplinary reports (80%), in 1 or more specific sections (OT section, 41%; PT section, 23%; ST section, 7%), or in the conclusion (8%) (fig 1, step 2). The reports eligible for analyses (n=86) showed that 37 patients (43%) were employed and wished to stay employed, and 49 (57%) were unemployed (fig 1, step 3).

The demographic data of the 86 patients included showed that the employed patients with NMD (n=37) were significantly younger, predominantly men, and more highly educated. No significant differences between the employed group and the unemployed group were found for marital status, medical diagnosis, and duration of complaints (table 1). Working hours and level of income and percentage disability for work differed significantly between the 2 groups. More than half of the employed patients worked 21 to 40 hours a week. Eight employed patients with NMD reported weekly hours of sick leave, and 3 patients were under review for disability percentage assessment.

**Table 1.** Demographic Characteristics of Employed and Not employed Patients With Neuromuscular Diseases (n=86)

	Employed	Not-employed	p-value*
Patients	37 (43)	49 (57)	
Age (y)	41.1 ± 11.4	48.8 ± 10.8	.002
Sex			
Men	27 (73)	19 (18.8)	.002
Marital status			
Living with others	26 (70.3)	36 (73.5)	.743
Education level			
Lower	11 (29.7)	31 (66)	
Intermediate	17 (45.9)	4 (8.5)	
Higher	9 (24.3)	12 (25.5)	<.001
Medical diagnoses			
Muscular diseases (FSHD & MD)	26 (70.3)	35 (71.4)	.907
Duration of complaints (n; %)> 1 y	34 (91.9)	46 (93.4)	.720

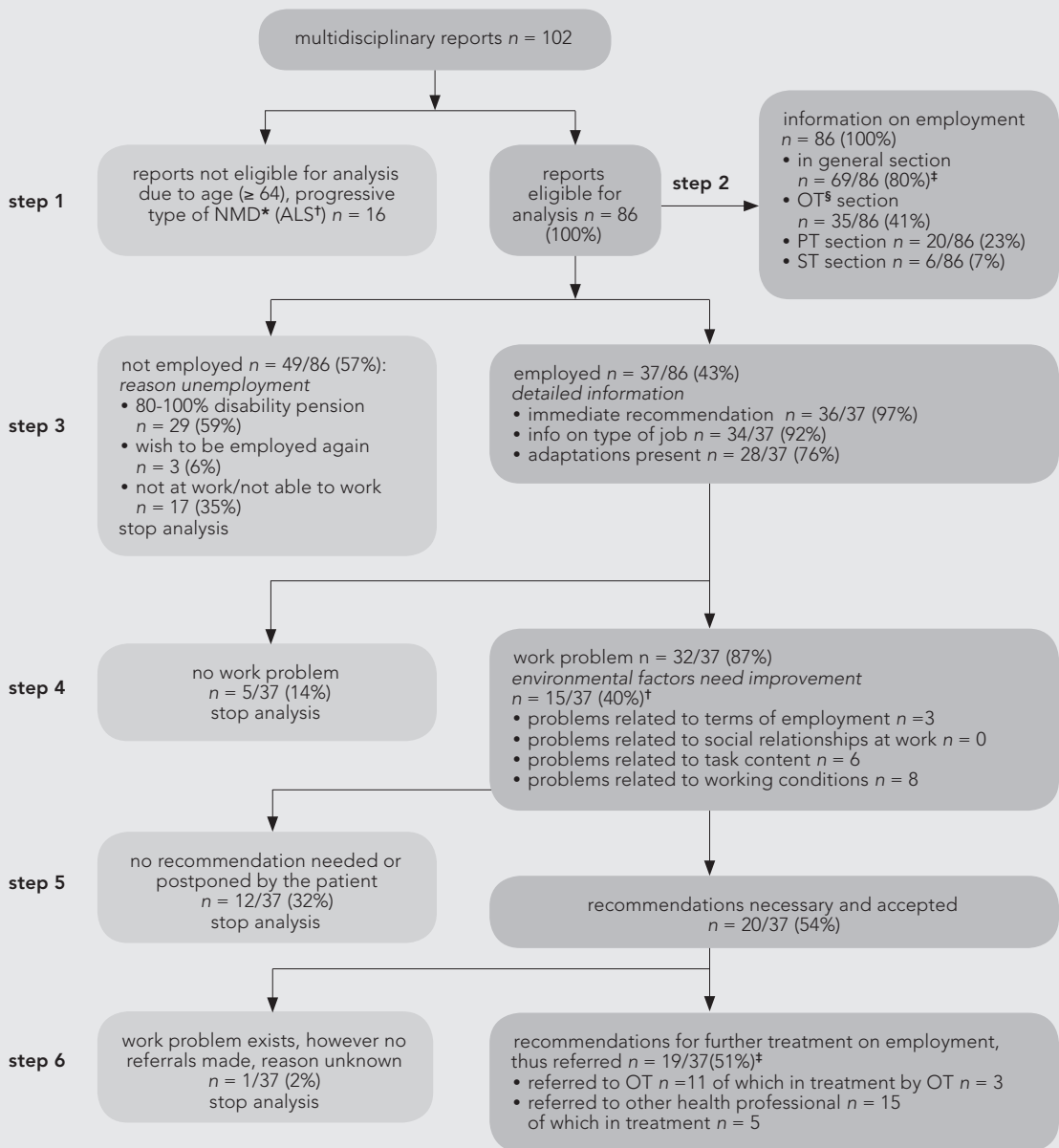
**NOTE.** Values are n (%), mean ± SD, or as otherwise indicated. Differences in proportions tested with  $\chi^2$  test; group means tested by unpaired t-test. Abbreviations: FSHD, facioscapulo-humeral muscular dystrophy; MD myotonic dystrophy. \* $p \leq .05$ .

The unemployed group (n=49) comprised 29 patients (59%) on disability pension, 3 patients (6%) who wished to return to work, and 17 (35%) for whom no information was present in the reports for obvious reasons. They either were unable to work because of their illness, were not employed because of their age (>62y), and/or carried out their role as a housewife (see fig 1, step 3). As a result of her illness, 1 of the 3 patients who wished to return to work was changing her profession, pursuing a university education, and hoped to find a job. The second patient did not like having to rely on social benefits and still wanted to work. A reintegration agency was involved in helping him find a new job. The third patient was unemployed and was considering returning to work in a white-collar job.

Thirty-six of the 37 employed patients received work-related recommendations during the one-off consultations. Eighteen patients were employed in office-related jobs, 13 in moderately strenuous jobs, and 3 in labor-intensive jobs; for 3 the type of job was not specified. For 28 employed patients (76%), 1 or more adaptations at work were identified. In 32 (87%) of the 37 reports of the employed patients, work problems were present despite existing adaptations at work (fig 1, step 4). In 15 (40.5%) of these 37 reports, patients identified specific environmental problems; however, problems with social relationships at work were not mentioned (see fig 1, step 4). Twelve (32%) of the 37 patients with a work problem indicated no need for referral or indicated



CHAPTER IV



**Figure 1.** Flow chart of analysis of integrated multidisciplinary reports on presence of employment information, type of job, realization and need for advice and adaptations at work. *Step 1:* Selection of reports eligible for analysis. *Step 2:* Presence of information on employment in different sections. *Step 3:* Division of the reports into 2 groups: employed and not employed. Employed screened for immediate recommendation, type of job, and adaptations present. *Step 4:* Presence of employment problems and type of adaptation problems. *Step 5:* Recommendations necessary and appreciated. *Step 6:* Referred for further treatment on employment.

\*NMD, neuromuscular disease. †ALS, amyotrophic lateral sclerosis. ‡Information can be present in 1 or more sections. §OT, occupational therapist; PT, physiotherapist; ST, speech therapist.

having solved their employment problems (fig 1, step 5). For 20 (54%) of the 37 patients, recommendations were made and consented. To further address these employment problems, 19 (51%) of the 37 patients were referred to an occupational therapist or other health professional (see fig 1, step 5). These 19 patients received specific recommendations 34 times in 1 or more sections of the multidisciplinary reports. Eight of these patients already in treatment by an occupational therapist (n=3) or other health professional (n=5) were additionally referred for their employment problems (fig 1, step 6). For 1 patient who had a work problem, no referral was made. The reason for that omission is not known.

## Discussion

Our main finding was that all 86 multidisciplinary reports contained information on employment. The percentage of patients with NMD at work (43%) was less than in a previous study<sup>[4]</sup> of patients with different NMDs, where on average 61% were employed. Previous findings that younger patients, men, and more highly educated patients with NMD are more likely to be employed are confirmed in the current study<sup>[4]</sup>.

Employment status and working disability were adequately addressed by occupational therapists and physical therapists, indicating that they are more appropriately qualified than speech therapists to identify problems of patients with NMD at risk for working disability. This was also the case for patients with rheumatoid arthritis<sup>[6]</sup>. The general section, for which the information was gathered by the occupational therapist, and the OT section produced most of the information on employment, followed by the PT section and the ST section. This was probably because of the primary focus of the OT professional. Work is recognized as an important domain of OT, whereas the physical therapist is mainly focused on motor functioning and the speech therapist on speech and communication skills<sup>[9]</sup>. These findings indicate that participation restrictions in work are systematically integrated into one-off consultations, most often by occupational therapists. The fact that information on employment is effectively collected is no guarantee that recommendations are implemented. A follow-up study<sup>[13]</sup> of implementation of therapy recommendations showed variations in frequency and volume based on treatment settings. Further research is needed to monitor whether recommendations for referrals toward a primary care or rehabilitation setting aimed at enhancement of employment participation for patients with NMD were implemented effectively. More than 80% of the employed patients were working in white-collar or moderately strenuous jobs. In the literature, often a distinction is made only between white-collar and blue-collar jobs<sup>[10]</sup>. In our opinion, some jobs cannot be categorized as purely white-collar or blue-collar jobs. There are jobs such as nursing and teaching that might involve heavy lifting, require holding the arms up in elevated positions for longer periods, or require fine motor control but that cannot be categorized as blue collar. Hence, we added the category

“relatively strenuous jobs” to determine how many patients with NMD were able to work in moderately physically demanding jobs. Indeed, a relatively large group did have a moderately physically demanding job in spite of the severe type of NMD. Less strenuous jobs were also positively related to employment in a group of 591 patients with NMD <sup>[4]</sup>.

The presence and need for adaptations at work were mentioned in most reports on employed patients with NMD. Adaptations were obviously often a precondition for being able to work. During the one-off consultations, immediate recommendations were given to almost all patients. In nearly all reports, 1 or more work problems were recorded. Recommendations for further treatment given by occupational therapists, physical therapists, and speech therapists were accepted by more than half of the patients with NMD experiencing employment problems, indicating that a one-off consultation is fairly effective in efforts to maintain a job in case of a deteriorating NMD. The most prevalent adjustments included adaptations of working conditions, terms of employment, and task content, as seen in other studies <sup>[1,14]</sup> on employment and patients with NMD. Andries et al <sup>[1]</sup> also found the presence or need for adaptations and adjustment of working hours, physical demands, and autonomy at work as a precondition for maintaining employment for patients with NMD. Because only 1 study <sup>[3]</sup> was found with these associations, the scientific level of evidence for these factors in our review was low. Our study revealed the same factors associated with employment, lending support to previous findings. Generally, it cannot be expected that all employment problems can be addressed in a one-off consultation; however, we found it surprising that none of the reports described the influence of social relationships and support at work. This factor deserves more attention because it is 1 of the 4 main work-related environmental factors that influence employment participation <sup>[15,17]</sup>. All employed patients wished to keep their jobs. An important prerequisite for keeping a job or returning to work is the person’s interest in employment <sup>[1]</sup>. Explicit interest in employment has been found to be a positive indicator for employment status <sup>[3]</sup>. The general questionnaire used in this study included items on employment not addressed by the therapists, such as level of income and weekly hours employed. A low level of income can be an indicator of employment problems. The general questionnaire also contained 3 items to identify employment problems: percentage disability for work, hours of sick leave, and employment under review. For identification of employment facts and problems, the general questionnaire can be used as a checklist to pay attention to the various aspects during a one-off consultation.

#### *Study Limitations*

This study was limited to only 1 setting, the outpatient clinic of a university hospital. A secondary analysis was performed with data originally collected to answer a different question - that is, to investigate referral patterns to occupational therapists, physical therapists, and speech therapists. These data were not collected to give

information and recommendations about employment status and the need for referral regarding employment for patients with NMD<sup>[7]</sup>. Data purposefully collected to investigate the adequacy of one-off consultation for assessment of employment problems could have provided more rigorous and replicable data. Our intention was, however, to determine whether problems related to employment are given sufficient attention during a generic one-off consultation as a starting point for intervention.

The presence of information on employment was presented descriptively. Because of the small sample of 37 reports and even smaller samples of type of job, frequency of work problems, type of adaptations at work, and work-related recommendations, the estimated percentages have a margin of error that can rise to a maximum of 16%.

### *Recommendations*

Based on our findings, we recommend the continuation of one-off consultations in the current manner, with emphasis on 4 questions addressing employment: (1) Does the patient have a job? (2) If the patient is not employed, does the patient want a job? (3) Are there concerns about employment? and (4) Does the patient need support by a health professional to address employment problems? Follow-up research is also recommended to investigate whether referral to allied health professionals leads to enhanced employment participation for patients with NMD.

Qualitative research is recommended to (1) provide more insight into the meaning of work for patients with NMD; (2) obtain more insight into the influence of social relationships and support at work; and (3) gain in-depth information on the changing ability of employed patients with NMD and their needs to stay at work. Until now, few qualitative studies have been performed to learn more about employment experiences of adults with NMD<sup>[18-20]</sup>.

## **Conclusions**

Eighty percent of the consultation reports contained information on employment. Less than half of the patients with NMD were employed, most in office-related or moderately strenuous jobs, using some kind of adaptation. Of the patients employed, a small proportion indicated they had not experienced employment problems, and a third claimed no need for recommendations or postponed the referral suggested. The remaining patients with employment problems, except 1, were adequately referred by occupational therapists and physical therapists for treatment of employment problems.

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## Appendix 1. Checklist multi-disciplinary advices of allied health professionals

### Instruction

Each advice has to be selected based upon paid work.

Use the complete advice to find the applicable items.

Select the items solely on what has been written, without interpretation.

Job adaptations must be interpreted as changes in terms of employment, task content, and work conditions. Only one answer can be selected, unless otherwise indicated.

### A. Number

#### B. File number

### C. Employment status

1. Paid employment (including clients employed and actually (partly) at sick leave or working at a therapeutic basis) go to E
2. Not employed
9. Missing

### D. Intention to (re-)integrate in paid employment

1. Following a course or education to (re)start work
  2. Would like to be employed// aiming at a job
  3. No intention to be employed (again)
  4. Hesitate about (re-)integration in paid employment
  9. Missing
- Go to I

### E. Type of job

1. Regular clerical / white collar
2. Regular job blue collar
3. Sheltered work environment
9. Missing

### F. Adaptation of work

1. No adaptation needed
2. Adaptations needed go to G
3. Adaptations realized go to H
9. Missing

### G. Type of adaptation needed (more answers possible)

1. Working hours (shifts, less/more hours)
  2. Task content (e.g. less physical or mental demanding, pace, more autonomy)
  3. Working conditions, adapted physical circumstances (e.g. tools, furniture)
  4. Other
  9. Missing
- Go to I

### H. Type of adaptation realized (more answers possible)

1. Working hours (shifts, less/more hours)
2. Task content (e.g. less physical or mental demanding, pace, more autonomy)
3. Working conditions, adapted physical circumstances (e.g. tools, furniture)
4. Other
9. Missing

### I. Weekly job hours

1. > 32 hours
2. 20-31 hours
3. 12-19 hours
4. < 12 hours
9. Missing

**J. Source of income**

1. Job income (salary, (early) retirement, business revenues.
  2. Allowance
  3. Both job income and allowance
  4. No personal income (e.g. income is generated by partner)
  9. Missing
- K. Presence of work problems**
1. Work problems expressed by the client YES
  2. Work problems expressed by the client NO
  9. Missing

**L. Problem Experience**

With regard to occupation, to what extent does the illness or disability cause problems in client's work according to the health professional?

1. No problems
2. Minor problems
3. Severe problems
9. Missing

**M. Impact on participation and autonomy (IPA) observed by a health professional (answer all questions)**

In the context of illness or disability

- A. the chances of doing the work or occupation he want to do are...
  - B. the chances of doing my work the way he want to do are...
  - C. the contacts with his colleagues are...
  - D. the chances of achieving or maintaining the job he want, are...
  - E. the chances of getting a different job with the same or different employer, are...
  - F. the chances of getting the training or education he wants are...
1. very good
  2. good
  3. fair
  4. poor
  5. very poor
  9. missing

**N. Action**

1. Work problems exist, no advice needed stop analyses
2. Work problems exist, advice needed, not wanted stop analyses
3. Work problems exist, advice needed and accepted
4. Work problems existed, problem solved/situation stable stop analyses
9. Missing stop analyses

**O. Diagnostic phase**

1. Received work advice by OT during assessment
2. Did not receive work advice by OT during assessment
9. Missing

**P. Intervention phase (more answers possible)**

1. Actually in treatment by OT
2. Actually in treatment by other allied health professional, rehab, social worker, occupational physician, pain ward
3. Work advice needed, client is referred to OT.
4. Work advice needed client is referred to other allied health professional, rehab, social worker, occup. physician, pain ward
5. Work advice needed in near future
6. Work related rehab or OT in the past
9. Missing





## **CHAPTER V**

### **How Persons with a Neuromuscular Disease perceive Employment Participation: A Qualitative Study**

Published as

Minis MA, Satink A, Kinébanian A, Engels JA, Heerkens YF, van Engelen BG, Nijhuis-van der Sanden MW. How persons with a Neuromuscular Disease perceive employment participation: A Qualitative Study. *J Occup Rehab* 2013; May, 5. [Epub ahead of print].



## Abstract

*Introduction:* A qualitative study was carried out to understand how people with a slow progressive adult type neuromuscular disease (NMD) perceive employment participation.

*Methods:* 16 paid employed persons with NMD were interviewed in open, in-depth interviews. Data were analyzed using the constant comparison method.

*Results:* Four themes were identified in the analyses: (1) Experiences regarding the meaning of work; (2) Solving problems oneself; (3) Reaching a turning point; and (4) Taking into account environmental aspects. Persons with NMD highlighted benefits of staying at work as well as the tension they felt how to shape decisions to handle progressive physical hindrances in job retention. This study shows how participants at work with NMD were challenged to keep up appearances at work and at home, the tension felt around when and if to disclose, the effect of their condition on colleagues and work reorganisation challenges. Participants experienced that disclosure did not always make things better. With increasing disability participants' focus shifted from the importance of assistive products towards considerate colleague, in particular superior's willingness in supporting job retention.

*Conclusions:* Implications for health professionals might include awareness of the significant impact of changes in physical condition on employment. Timely communication and if appropriate referral to a health or occupational professional may empower employees with NMD to handle employment issues at an appropriate way for themselves. Assistive products and a supportive superior might enhance employment participation.

## Introduction

The importance and meaning of paid employment has been studied over decades. Gainful employment is seen as a human right inclusive of people with a disability <sup>[1,2]</sup>. Persons with slow progressive adult types of Neuromuscular Disease (NMD) experience few tangible hindrances because of their disease during their upbringing or education, however decreasing muscle strength, increasing functional problems and in some cases cognitive decline become apparent during adulthood <sup>[3]</sup>. As a result persons with NMD start employment in good health with a seemingly normal career potential. Although persons with a hereditary NMD might be aware of the impact of possible future physical deteriorations on activities of daily life, individual experiences differ as result of the clinical heterogeneity of NMD which varies even within families <sup>[4-6]</sup>.

According to a Dutch report on Public Health Forecasting, the employment rate of chronically ill persons (35%) is in sharp contrast with that of healthy persons (65%) <sup>[7]</sup>. The employment rate did not differ from the normal population for persons with

Facioscapulo-humeral Dystrophy (FSHD) (64%) and Hereditary Motor and Sensory Neuropathy (HMSN) (70%). In a group of 591 persons with an adult type of NMD 57% were found to be employed between the age of 35 to 65<sup>[7,8]</sup>. Thus, persons with NMD seemed less disadvantaged in this respect compared to chronically ill people in general. These percentages were more promising than in former Dutch studies in which an average of 40-50% of persons with NMD was employed<sup>[9,10]</sup>. Andries et al, investigated employment rates for persons with NMD and found that labour participation tends to decrease around the age of 34 years onwards until middle-age<sup>[11]</sup>. From the age of 34 the majority of employees with NMD require assistive products<sup>[12]</sup> in order to remain employed<sup>[11,13]</sup>. Employment rates for men were better than for women. A 1995 study found that at age 54 years, an average of 60% of the men with NMD were employed compared to 90% of the overall male population; for females the percentage was 34% after the age of 34 compared to 54% of the overall female population<sup>[11]</sup>. Although comparison of studies is difficult due to different time frames and sample populations, a tendency towards a drop in employment rates seemed related to the diagnosis and in line with the deterioration process taking place in persons with NMD<sup>[10,11]</sup>.

A systematic review of employment factors in patients with NMD revealed a number of factors that seemed to influence the employment rate in persons with the three types of NMD. In addition to general factors, such as age, gender, and education, factors such as physical function, muscle strength, fatigue, type of NMD, interest in employment and type of job influenced the employment rate in persons with NMD<sup>[14]</sup>. However, according to a best evidence synthesis for quantitative research<sup>[15]</sup>, these studies were not based on rigorously measured evidence<sup>[13]</sup>. Although employed and not-employed persons with NMD reported severe fatigue (cut-off  $\geq 35$  on the CIS fatigue subscale)<sup>[8,16]</sup> the employed group reported significantly less fatigue than their not-employed peers. Fatigue as an influencing symptom in employment participation is also found in persons with other slow progressive neurological diseases, such as Multiple Sclerosis (MS), Parkinson's disease and for persons who have experienced a Cerebral Vascular Accident<sup>[17]</sup>. Persons with Myotonic Dystrophy (MD) were not only severely fatigued, but also had the least social participation, the most psychosocial problems and the poorest psychosocial well-being compared to persons with other types of NMD<sup>[18]</sup>. These social consequences are likely related to cognitive decline, a typical characteristic for this multi-system disorder. Besides a huge impact on daily life, this disease-related characteristic likely impacts their ability to work<sup>[8]</sup> although health care providers may not always have recognized this. For example, recently it was reported by couples, of which one had been diagnosed with MD, that health care providers lacked awareness of the consequences of MD for daily life<sup>[19]</sup>. Early identification of employment issues for persons with a disability is thus recommended so that timely support can be provided during the early stages of decline and before employees encounter the stress of deteriorating abilities<sup>[20,21]</sup>.

Although the above-mentioned studies provide some information about relevant factors influencing employment participation, the experiences of persons with NMD,

i.e. the client perspective, regarding job retention is lacking. This client perspective can provide knowledge about the impact of the disease on employment capacity and employment performance and factors which can enable continuation of their participation in the work force. Information from employed persons with NMD in particular can give insight into the meaning of work for life fulfilment, individual capacities and environmental requirements for enhancement of employment participation.

The objective of this study was to explore how persons with NMD still at work perceive the impact of the slow process of NMD related deterioration on employment participation and the strategies used in maintaining their jobs.

## Methodology

### *Design*

In 2010 and 2011 a qualitative study was performed. Purposeful sampling was used to include currently employed adult persons in the age range of 18 to 65 years representing three different neuromuscular genetically well defined, homogeneous, and large client populations with relatively common neuromuscular disorders: (1) FSHD<sup>[4]</sup>, (2) HMSN<sup>[5]</sup>, and (3) MD<sup>[6]</sup> and who experienced hindrances in employment participation. Included persons confirmed that employment participation and performance were influenced by their disease symptoms and that they were employed at least 12 hours a week conforming to the definition of a working population defined by the Dutch government<sup>[22]</sup>. The intention was to include persons from different age groups, levels of education, types of jobs and stages of the disease in order to look at problems concerning work from a variety of perspectives. Recruitment was carried out using four methods; notices on the website of the Dutch Federation of Muscular Diseases<sup>[23]</sup>; in the periodical of the same organization; from the database of persons with NMD previously admitted to a neuromuscular center of a university hospital in the Netherlands; and via participants.

### *Data collection and participants*

Data collection consisted of a single, open, in-depth interview (16 in total) at a location selected by the participant: 12 opted for an interview at home, two preferred to be interviewed at the university hospital and two invited the researcher to their work place. Some informal talk about the hospitality at their home or work place and their willingness to participate preceded the interview. Furthermore, the participants' understanding of the purpose and format of the interview was ascertained, and additional explanation was provided when necessary.

The first researcher (MM) began the session by showing interest in the interviewee's job and asking a few factual questions about education and training in order to understand the context of his/her work. The main question was "What does

work mean to you?” Interview topics focused on an ordinary day at work, experiences, satisfaction, importance of working for remuneration and engagement with the current job. The researcher encouraged elaboration on topics such as the impact of the disease on the ability to work, the requirements or preconditions enabling job retention, and the possibilities for either job rotation or applying for another job within the company or with another employer.

Each interview proceeded in an informal conversational style, lasting forty-five minutes to one hour. Based on the researcher’s written memos on the impressions and reflections of one interview and first analysis, the interview guide was adapted in the consecutive interviews with probing questions if new themes showed up. This was done to deepen and expand the topics. For example: “What is your own contribution to keeping your job?”; “What is your relationship with your colleagues and your superior?”; “How does the state of the economy influence your career opportunities?”. After the interview with the 12th participant, saturation was reached, since the interviews with four additional persons did not produce any new themes.

The researcher checked eligibility, explained the objective of the interview to potential participants in a telephone conversation and sent written information about the study. All candidates confirmed participation in the study, and when contacted a week later, the researcher made an appointment for the interview. Participants were explicitly informed that participation was entirely voluntary and that withdrawing from the study was possible at any time. Written informed consent was obtained. Permission to audiotape the interview as well as reassurance of confidentiality was confirmed during the visit. Following the interview, participants were invited to reflect on their experiences and indicate potential need for formal after care but none of the participants needed this.

The Medical Ethics Committee of the Radboud University Nijmegen Medical Centre in the Netherlands CMO nr 30076, approved the research project in accordance with the Helsinki Declaration<sup>[24]</sup>.

Data were gathered from 16 currently employed persons with three different types of NMD (5 women and 11 men). All participants resided in the Netherlands, were Caucasian, and aged 24 to 62 years. Seven persons contacted the researcher in response to the announcement on the aforementioned website. Seven persons from the database of persons with NMD previously admitted to a neuromuscular center of a university hospital in the Netherlands were approached by telephone and two persons were asked by other participants to enroll in the study. One participant lived with his parents, ten lived with a spouse or partner and five lived alone. Five persons were diagnosed with MD (type 1 or type 2), five participants with FSHD and six participants with HMSN. Severity of symptoms, educational level, socio-economic background and type of job varied. Fourteen participants were employed and two participants were entrepreneurs. Working experience varied between two and 40 years. All participants were breadwinners, of whom five were joint breadwinners. Five participants worked full-time, the others worked part-time or were partially work disabled and received

both a salary and disability pension. All participants used assistive products or had been facilitated, for example, working fewer hours or at a slower pace (table 1). The order in the table was based on the type of diagnosis.

### *Data Analysis*

The analysis was inspired by a hermeneutic approach involving a constant comparison method. Atlas.ti© computer software version 6 was used to assist in organizing the data. After completing five interviews, in-depth analysis started following each additional interview. The researcher transcribed the audiotapes verbatim. The analysis of the data consisted of the following steps <sup>[25]</sup>:

- 1) Familiarization with the interviews, and understanding the experiences on the job by multiple readings of the transcripts, the production of summaries and reading the reflective memos and field notes.
- 2) Assignment of in vivo codes related to the research questions to meaningful units so as to stay close to the participants' original phrasing.
- 3) Categorization of meaningful units, reorganization and interpretation, resulting in the formulation of themes and subthemes.
- 4) Interpretation of relations between themes and subthemes. Use of other sources, including literature, field notes and personal experiences, leading to new insights and better understanding of the meaning of work for participants with NMD.

The first researcher (MM) executed all these steps. During the analysis, a number of conceptual maps of the emerging themes were constructed and discussed by a small peer review group and experts in qualitative methodology (TS and AK). Text extracts that supported each theme were discussed for consensus on several occasions and over time with discussion. To capture the thoughts and insights, the first researcher wrote minutes and memos to assist and documented the development of the themes and subthemes. The findings were discussed five times within the whole research team which reached consensus on the themes and subthemes in a final meeting. Whenever necessary for the purpose of anonymity, the participant's wording was paraphrased (omitting dates and names) <sup>[19]</sup>.

### *Interview Findings*

Four themes were identified in the analyses: (1) *Experiences regarding the meaning of work*: "Work is belonging"; (2) *Solving problems oneself*: "I am not going to tell"; (3) *Reaching a turning point*: "I have to be realistic"; and (4) *Taking into account environmental aspects*: "I can understand both sides" (table 2). Each of these themes and related subthemes are described in detail with quotes to illustrate the themes.



CHAPTER V

**Table 1.** Demographic profile of participants with facioscapula-humeral muscular dystrophy (FSHD), Hereditary Motor and Sensory Neuropathy (HMSN) and Myotonic Dystrophy (MD). Neuromuscular diseases (NMD)

<b>Participant</b>	<b>Gender</b>	<b>Age</b>	<b>NMD type and other conditions</b>	<b>Severity of symptoms</b>	<b>Marital state</b>
1	Female	43	FSHD	Moderate	Married
2	Male	30	FSHD	Moderate	Co-habiting
3	Male	62	FSHD	Severe	Married
4	Male	53	FSHD	Moderate	Married
5	Male	45	FSHD	Moderate	Married
6	Male	24	HMSN	Little	Living with parents
7	Male	49	HMSN & hernia	Severe	Living alone
8	Male	28	HMSN & hip dysplasia	Moderate	Married
9	Female	41	HMSN	Moderate	Married Former head nurse
10	Female	42	HMSN	Moderate	Living alone
11	Male	42	HMSN	Severe	Married
12	Male	46	MD	Little	Married
13	Male	43	MD	Severe	Living alone
14	Male	51	MD	Severe	Living alone
15	Female	46	MD	Moderate	Living alone
16	Female	44	MD	Moderate	Married

## HOW PERSONS WITH NMD PERCEIVE EMPLOYMENT PARTICIPATION

Education level	Profession	Assistive products and adjustments at the work place or at home
Intermediate	Residential counselor	Reduced working hours; Wheeled office chair
Advanced	Builder website applications	Reduced pace at work
Advanced	IT consultant	Wheeled office chair; Computer facilities
Lower	Shovel operator	Adapted shovel Support from employer and colleagues
Advanced	Teacher primary school	Wheeled office chair. Printer, scanner, copy machine in the classroom. Partner taking over home duties
Lower	Truck driver	Adaptation in the truck present and needed
Intermediate	Office job at the municipality	Adapted car to commute Powered office chair Voice controlled computer Support from employer and colleagues
Intermediate	Arborist	Extra pair of protection shoes Quad and quad transportation system
Advanced	IT consultant	Changed job with same employer: Reduced working hours and -pace at work Wheeled office chair Support from employer and colleagues
Advanced	Psychiatrist	Reduced working hours Assistance by secretary; No shifts Support from employer and colleagues
Intermediate	Entrepreneur Accountancy	Able to manage own pace at work Office at ground level Adapted computer mouse
Intermediate	Entrepreneur Director	Able to manage own pace at work
Intermediate	Buyer and manager of products at a food company	Reduced working hours Different job with same employer Office at ground level
Advanced	Accountant at major corporation	Home care & Adapted car to commute
Intermediate	Former assistant accountancy Now musician	Changed type of job Reduced working hours & reduced pace
Lower	Homecare	Reduced pace at work

## **Experiences Regarding the Meaning of Work: “Work is Belonging”**

The first theme dealt with the way participants with NMD at work experienced the meaning of work. Most participants indicated that work was an important activity throughout their lives, irrespective of the phase of the disease or severity of symptoms. Reasons why work was meaningful and what aspects were meaningful varied among the participants, but most of them felt that work connected them to the community. The following quote underlined the significance of work for participants with NMD.

*Work is 80% of my life fulfillment, yes (P14).*

However, paid work seemed to become a burden when working with their disease related limitations was felt to be a necessity, i.e. when participants experienced that they had no other choice than staying at work.

### **Working for Personal Benefits**

Aspects that participants mentioned to express the advantageousness of a paid job and what made work meaningful to them were the pleasure, status and self-esteem derived from work. Some participants stated that work justified their personal value, others who were single and experienced severe symptoms had chosen to put all their energy into their work at the cost of other social contacts and emphasized that work was important since work seemed to be their only remaining connection to society. Participant 4, diagnosed with FSHD, had the same view, and although he had a family and hobbies at home he expressed that he valued his paid job because filling time with hobbies that were difficult to perform due to physical deterioration seemed a poor alternative for his paid work.

*At work I talk with my colleagues, there are stories to tell, I am busy. I like that... having colleagues, taking part in society, ... I am glad that I work ... Indeed, I can do a lot of chores at home, once I did, but this is getting increasingly difficult too (P4).*

### **Working for Economic and Social Benefits**

Analysis showed that the value of employment was evident in the economic and social benefits that participants experienced from working. Some participants mentioned

**Table 2.** Themes and subthemes identified through analysis of interviews with 16 employed participants with a Neuromuscular Disease

**Experiences regarding the meaning of work: “Work is belonging”**

- Working for personal benefit
- Working for economic and social benefits
- Working as a burden

**Solving problems oneself: “I am not going to tell”**

- Disguising limitations thus postponing disclosure at work
- Contemplating job demands and other roles in life

**Reaching a turning point: “I have to be realistic”**

- Struggling to let go
- Accepting change: an emotional process
- Using strategies to keep one’s job in the long run

**Taking into account environmental aspects: “I can understand both sides”**

- Valuing assistive products
- Considering colleagues’ positions
- Considering employer’s position
- Interfering macro-economic influences from society

that receiving a salary enabled them to pay for their children’s education, health care or hired assistance for physically demanding domestic activities. Obligations connected to personal roles in life positively influenced participants’ meaning of being gainfully employed. Participants who were breadwinners (men and women) indicated that for them this role was linked to a sense of full participation in society. It felt good to be able to provide the necessary income for oneself and the family.

*I have many choices and a lot of freedom, due to the fact that I work. I do the work I can do, with which I earn money that allows me to hire people for physically demanding jobs at home (P10).*

*For me it is important to be useful for society, not just having to rely on support by society (P11).*

**Working as a Burden**

Although participants preferred to work instead of having to rely on social benefits, analysis showed that having to work for economic reasons was experienced as a burden, especially when participants experienced symptoms such as fatigue, which affected their employment performance.

*As head of the family and being responsible for my children, I must make sure that there is food on the table... I also have responsibilities. I have my own house. I need to pay my mortgage every month (P5).*

*... and besides, it is currently a financial necessity. I must work now. I'm not a breadwinner, but my son wants to become an interior decorator. That training costs a lot of money. I want to give my child the opportunity to follow his preferred education (P1).*

### **Solving Problems Oneself: "I Am Not Going to Tell"**

Analysis showed that participants held on to the job and subsequently searched for solutions themselves to relieve their workload. Strategies for solving problems differed among participants and depended on the type of job, their upbringing, and stages of deterioration. Most participants stated that their first reaction to physical signs of deterioration, such as reduced muscle strength or increased fatigue, was to avoid going on sick leave as it was against their values and norms. To them a reliable employee was someone who works hard, without complaints or periods of sick leave. The first reaction after diagnosis was to pretend that nothing was the matter. One participant stated that she feared losing her job from the moment she was diagnosed in 2003, so that she did not talk about her problems at work as long as she was able to hide the symptoms.

*Colleagues pass by, now and then. What's up? Are you able to keep up? Yes, I'm fine. I carry on, even in bad periods. As long as it's not getting worse, I am not going to tell (P1).*

### **Disguising Limitations thus Postponing Disclosure at Work**

Physicians' unfamiliarity with early symptoms delayed the initiation of support in daily functioning or appropriate interventions on behalf of some participants. Moreover, after having been diagnosed, many participants tended to deny their condition and the challenges within their environment. Postponing disclosure at work could last for years as long as symptoms and consequences for employment participation could be hidden even when symptoms started hindering occupational performance.

*The GP claimed that referral to a neurologist was unnecessary. After my insisting I was referred and then the diagnosis was confirmed (P1).*

*For years it troubled me, yes there was something wrong, but I didn't know what it was. When I was diagnosed in '87, I just kept going on and work went reasonably well. (P4)*

*I denied my illness for 15 years. That is true for many persons with MD, indeed of 3000 people in the Netherlands with MD; only 900 are members of the association (P14)*

*...and I've always thought as long as I do not have it, I have nothing; I mean so long as I did not notice anything (P15).*

Examples of ways participants disguise their limitations, thus avoiding sick leave varied from voluntary reduction of working hours to asking for parental leave, even when this meant a lower income. Seeking a less strenuous job within the organization without mentioning the reason was found to be another solution. Other reasons mentioned by participants for avoiding disclosure were the fear of being fired and repercussions by the organization.

*At work I started doing things that fitted my physical condition... In 2002 I [nurse] applied for an office job... yes, at a certain moment in time, it was in 2007, I decided to ask for a period of parental leave. I wanted to work so badly, but was not able to continue anymore (P9).*

*As long as it does not really get worse, then I am not going to tell. If it gets worse, yes, I must tell, but as long... Yes, he [employer] might as well fire me, when I am on sick leave. Indeed, I have just completed one year in this organization (P16).*

### **Contemplating Job Demands and Other Roles in Life**

Analysis indicated that participants became aware that the choice to continue to work had consequences for their private lives and family members. If one of the partners was diagnosed with NMD, couples shared the struggle. Indeed most participants with a partner did not start rebalancing job demands, but began by giving up responsibilities and duties in the private domain, in their roles as a parent or a spouse. Concerns about income, or the possible need to change roles within the family due to the disease could be emotional. For one couple, roles were changed: the woman quit her job because the combination of working and caring for her family without the support of her partner with NMD was no longer an option for her. He wanted to work fulltime and did not have enough energy to support his wife in domestic roles as he used to. The resulting dilemma was that because of his partial disability the current decisions might cause unexpected financial problems and need to be altered.

*Two years ago my wife quit her job. At some point she could no longer sustain a busy family of four children. I work fulltime and therefore my energy is low and she had to do more at home. The balance we have now might have to be adapted again in the near future. Of course that makes me sad sometimes and this is where I start to have my doubts. Where will this lead? (P5).*

Analysis showed that some participants made use of legislation and regulations in order to rebalance roles among partners. For example, after years of proving himself in paid jobs, one participant purposely chose to make temporary use of a disability pension in favor of his wife who wanted to work. Unfortunately in this latter situation the tension over how to shape decisions at home became apparent when she lost her job. In another case, the participants' partner became the health care provider for her partner and got paid by health insurance, with the advantage of providing the care in a timely fashion.

*Yes my wife worked in the field of health care. When we heard about the new legislation that provides financial compensation instead of home care, several years ago, we took the opportunity and now my wife is being paid for the care she provides to me. She has resigned her job and has started her own day-care business from home. So she is always at home for work, to take care of our and other people's children and she can help me any moment of the day when I work in my office next door (P11).*

Consequences of keeping up appearances at work might have repercussions at home. To save energy a participant sacrificed familial obligations to fulfill work obligations, were not able to do anything at home after work or as another participant explained the intensity of social contacts was lowered.

*My wife knows the other me, completely exhausted, physically and mentally burnt out, and now completely worn out in my chair (P12).*

*At the moment I'm not present [for music practice in the evening]; my friends know that today my energy level is low. Then I grab my moment of rest (P5).*

## **Reaching a Turning Point: “I Have to be Realistic”**

From the moment that symptoms became obvious and participants were no longer able to work in the same way in spite of measures they had taken, a turning point was reached. Some participants had reached this turning point long ago, for others it was a fresh reality. The most common reason to admit to themselves that continuation of

work as before was no longer possible, was that symptoms disrupted job productivity. The participant that reached this stage of openness during the interview became emotional, however still referred to this turning point in the impersonal ‘you’ form, as though it was not yet his own concern, while some other participants at a later stage of the disease or close to retirement seemed to be beyond that stage of grief and showed an active coping attitude towards changes and lived day-to-day.

*Of course that makes you feel miserable sometimes and you think where does this end. At what point, is it going to be a real problem... Erm..., okay you're not present for a day, ... it still feels very difficult. Then you wonder, is it worth continuing to work full time, if besides work, you have no energy for your family, hobbies or your social network (P5).*

### **Struggling to Let Go**

The participants’ story showed a changing attitude about their ability to continue work or to change jobs, however it was clear that this change had an emotional effect with fear for the future, grief and feelings of insecurity. This changing attitude was caused by symptoms that started to impact employment participation as a result of severe physical hindrances and the inability to combine all roles in life. Many quotes began with the phrase: “*I had to be realistic...*” or “*I came to realize*” indicating being forced by their physical conditions to admit that going on in the old way was no longer an option. A changed attitude was seen in participants openly engaging in dialogue with colleagues about the disorder and the work-related consequences of their physical condition.

*...then I came to realize, I must have been a problem for my colleagues, by not asking for help (P9).*

Participants covertly indicated that sooner or later performing the way they were accustomed to was no longer possible, which made them struggle. In the process of letting go and changing, rational considerations alternated with emotional reactions. For several participants the struggle was prolonged, probably since no experts were consulted and they could not foresee the benefits of openness. Participants related that their fear of losing connection to society kept them from giving in to symptoms and accepting change. Several participants blamed others, or mistrusted superiors who threatened job retention. Due to uncertainty these participants expressed openly fear of loneliness, knowing that being fired was inevitable.

*When I applied for the job in 2006 I didn't report that I had a muscle disease. I had a bit of fear that I wouldn't be hired... and... erm... now [2010], I think that there*



*is a chance of losing my job. Don't think you're indispensable when you are not functioning. Now I still can manage those two days, but I think ultimately the disease will hinder me so much, that maybe a whole day will be too much anyway. Now I can do it, but whether it still works in five years I do not know (P1).*

At an advanced stage of the disease, analysis revealed that changing jobs or finding another employer was no longer an option and it was obvious that work might even become impossible. The story of the participants showed a change in attitude to their own ability to continue work. One participant anticipated that the career would end earlier than under normal circumstances and that changing jobs in the future would be difficult.

*Being mobile is the most important thing. I'm neither mobile nor presentable. Then one must be realistic, with my handicap I will never ever get another job anymore (P14).*

*... but perhaps it will be more difficult to find work again somewhere else, when you carry your condition with you in your luggage. That is something that is going round in your head (P7).*

### **Using Strategies to Keep One's Job in the Long Run**

In order to maintain long-term employment, strategies were chosen from the beginning of the process of deterioration. Looking back into their past career, some participants emphasized that having been able to build goodwill turned out to be helpful in negotiating current job retention, although it was recognized that goodwill and personal strategies alone were not adequate to ensure employment participation.

*Someone said to me: Your boss does not want to lose you, he wants to keep you. Indeed, I contributed a lot when I used to work fulltime. I don't know how to say it, but with my 12 hours, I am still worthwhile for the company (P9).*

After being diagnosed many years ago and anticipating future symptoms, some participants developed strategies such as doing work they knew that would make them indispensable, to guarantee job retention later on.

*Your position partly depends on your indispensability. In fact, the better your position at work, doing work that needs to be done, the better it is of course (P8).*

## **Taking into account environmental aspects: “I can understand both sides”**

However it should be noted that ultimately personal strategies for keeping one’s job were no guarantee of success. Many aspects influenced the chances of job retention.

### **Valuing Assistive Products**

All participants at all stages of the disease required adapted work environments to be able to work or to continue working when physical limitations hindered employment performance. Analysis revealed that participants who used assistive products and were provided with adapted work conditions did not experience these measures negatively. As a matter of fact these measures enabled them to continue their work and this was experienced as positive. The type of measures that had been taken at home and at work that facilitated ongoing employment varied greatly among participants, depending on both the physical abilities of participants and job demands.

*Now, they [homecare] come simply every day at 7:30 AM, so that I am at work on time of course (P14).*

*I have a battery powered office chair so that I don’t have to walk so much and voice recognition on the computer. I chat to the computer and the computer does what I say (P7).*

*I am now at home on Wednesdays. The other days I work from 8:00 to about 15:30 pm. The load is nicely distributed over the week (P10).*

One participant was explicit that one should not take these measures for granted and was aware of the need to be sufficiently productive, to make the firm’s investments in these measures worthwhile.

*If you want to have that kind of equipment, I think it is important to realize that this must be paid for. That’s why you have to make money for your boss and provide quality at work. Yes, I think that way. It is important to have good relationship with your manager, and do your job so well that they like to keep you (P8).*

### Considering Colleagues' Positions

In addition to the importance of assistive products and facilities, analysis provided evidence on the importance of good work relationships. With increasing disability, relationships at work became crucial. Compassionate colleagues and, above all, good relationships with superiors willing to support participants' job retention became increasingly important as dependency due to physical deterioration progressed. Absence of this consideration negatively influenced employment status.

Having sympathetic colleagues was very much appreciated. Work enjoyment was boosted by colleagues' understanding and empathy. The importance of social contacts at work was emphasized especially by singles, for those who put work first and foremost at the cost of their private contacts and those who experienced more severe physical dysfunction at a later stage of the disease. Because they put all their energy into work, the work place provided the majority of social relationships, which in turn made work even more meaningful.

*Yes, and colleagues, I have very nice colleagues, who understand my situation too. They help with everything, where they can help, and they are very considerate. During holidays my colleagues carry my bags, assist me from the room to the dining room, ... erm, from hotel to bus. They help me get on the bus, when we get to the ski-hill then they lift me onto the ski lift, and once I am in the lift... then we can go... this makes me very emotional (P7).*

Participants realized they had been unaware of how their condition had impacted colleagues. After disclosing their work problems, participants found that their colleagues were relieved to have this in the open. Several participants then came to realize that trying to hide symptoms was a burden for their colleagues. It turned out that openness made it easier for their colleagues to provide support.

*It seemed to be a relief for my colleagues. My colleagues took over my evening and weekend shifts immediately when I announced that I would like to quit these obligations in a few months (P10).*

However openness about the disease did not guarantee empathy from colleagues. Some participants experienced disappointment that after having shared the grief of their progressive illness, misunderstandings concerning their inability to perform certain tasks remained or support from the superior was lacking. One participant was unpleasantly surprised by the negative attitude of his employer, in particular since his superior had experienced periods of severe fatigue himself recently. Several participants highlighted this emotional dimension in the process of change.

*I am very open and I find it no problem to talk about my things; however it sometimes makes me tired, that I have to explain my disability over and over again. For instance why is it not obvious, that I won't be able to be present, if the meeting is on another floor? (P5).*

*He [superior] wants me to do this work anyway, even though he knows what it's like to be tired because he has experienced the same in recent months. He was very tired, could not work as much. This is why I was disappointed by him (P2).*

### **Considering Employer's Position**

Participants considered things from their employers' perspective including the challenges for the employer involved in supporting participants' employment retention in spite of potential or existing physical limitations. The policy of the organization and the way this policy was carried out by the superior also determined whether participants managed to keep their jobs. Overall the superior's attitude was experienced as particularly important. Participants working in firms with a policy mandating accommodations to facilitate employment for persons with reduced capacities had confidence in the future and dared to be open with their superior about the problems in employment performance that emerged.

*Yes, yes it is a great employer here, who will never say you must go, because we cannot do anything for you anymore. That never happens here. That gives me a safe feeling. I have job security here (P4).*

When companies were reorganized and new superiors were in charge, the former arrangements could easily fall apart. Lack of understanding and negative actions by the company caused decreased pleasure in work. Absence of a sense of security restrained these participants from daring to speak openly about their problems.

*A colleague who had heart problems was advised by the occupational physician to work less. Then she was offered another position, less suitable, supposedly there was no other option. Eventually she decided to leave. Then they [employer] have you where they want to have you, because they want to get rid of you... (P1).*

Although analysis revealed that for some participants their condition was taken into account by superiors, others were quite dissatisfied about the lack of understanding by their superiors. The way participants judged their employers' attitudes regarding the presence or absence of employment support depended on participants' own situation

and disease severity. One young participant who had vague symptoms and two others with a severe condition were indignant about not getting or keeping their favourite job. One of them, although annoyed, and still able to hide her symptoms, understood the choice of her potential employer to not to award her the job after having heard the possible diagnosis.

*As soon as my superior realised my condition, I could imagine that he would be afraid that something would soon go wrong, that I would be forced to work less and that the company would have to pay for my absence because I have a contract (P1).*

*The day after he [the future employer] phoned back. I immediately explained what was going on and that the diagnosis was not clear yet. Then he announced that he was not going to offer me a contract (P6).*

*I think, he [employer] thought I would be totally disabled very quickly. And this was what he really feared. It was a small company that did not want to take that risk, just financially. And somehow, I can still understand it too. If I was an employer and I had 10 people and I wanted someone there and I would have to choose between someone who is ill and someone who is healthy, I would know what to choose (P15).*

For participants with severe physical hindrances the impossibility of changing jobs was probably a good reason to fight to keep their current job. During years of living and working with NMD, one participant became an expert in legislation and regulations for persons with a disability. He convinced his employer of his rights to work with his handicap, and overruled the employer's point of view that this employee was a potential danger to the firm with regard to accountability in case of an accident. This participant had firm opinions about employment participation and expectations for unconditional job retention and employers' responsibility in supporting this.

*I said to my employer that I do not qualify for the disability law. My skills are in my head and those capabilities are not affected by my illness. For motor skill problems, there are other solutions."... "That he [the employer] should be positive towards you, by saying: "You are disabled, okay, but we want to do everything to keep you at work so we will try to create opportunities" (P14).*

The two entrepreneurs named the advantages of being able to make one's own choices, set one's own pace and working times fitting their capabilities. These entrepreneurs felt blessed by their independence and their ability to regulate working hours and to take other measures themselves.

*...as an entrepreneur I am able to manage my work. I can judge for myself what is really important (P11).*

### **Interfering Macro-Economic Influences from Society**

Last but not least, analysis revealed that participants were fully aware of being in a more vulnerable position than healthy colleagues in the event of an imminent reorganisation. The participants expressed that the risk of job loss due to economic crises or reorganization seemed to be larger for them than for healthy persons in their organization. From the participants' point of view reorganization could be improperly used to dismiss someone. Some participants with severe physical hindrances discussed with their employers ways to keep their jobs. Although some participants had been successful in surviving several reorganizations in the past the threat of dismissal remained. When companies were reorganized and other superiors were in charge, the former arrangements were at risk.

*We need to reorganize, and this is the result. Except me, everyone is healthy. All personnel who lost their jobs because of the reorganization were offered another position and continue to work, except me. That is very strange. I worked hard for the firm and now they lay me off (P13).*

*In that sense, it is important that someone has a job where his boss is also nearby... But our company was getting bigger and bigger... . And since May last year ... erm... this empathetic boss has gone. I have seen no boss until March... and actually the first time I talked to my new boss in June, he called me to say that I was on the list of persons whose work went to India. So it is very important to have your boss close by so that in addition to your work at your work place, your physical sight of the work, perhaps your psyche in relation to the work can be assessed. In the past that's always been perfect. These days arrangements can be easily discarded (P3).*

The willingness to work was found throughout participants' lives, irrespective of disease severity. When macro-economic circumstances affect employment, participants tended to be in a disadvantaged position compared to healthy colleagues.

## **Discussion**

This study explored the meaning of work for 16 employed persons with three slow progressive types of NMD and the experiences, challenges and strategies regarding

job retention. Also the stages participants encountered in the process of physical deterioration which impacted employment participation were identified. Although the employment rate of persons with NMD was relatively high compared to persons with other chronic diseases <sup>[9, 11, 22]</sup>, the findings of this study may be useful in helping professionals to empower persons with NMD to maintain employed.

#### *Various Experiences Underpinning the Meaning of Work*

Participants experienced work as an important activity throughout their lives irrespective of disease severity. At first glance, the reasons that work was meaningful to them did not differ from those of healthy people, namely deriving economic benefits from work, developing talent, self-realisation (self-esteem, social contacts, pleasure), and contributing to society <sup>[26]</sup>.

The first theme, *Experiences regarding the meaning of work: "Work is belonging"* highlighted the meaning of work and was recognized as an overall theme throughout participants' lives. The way participants elaborated on the meaning of work in spite of the accompanying challenges emphasized the importance of work to them, a phenomenon recognized in another study on the meaning of work in the lives of people with disabilities <sup>[27]</sup>. Participants with a severe condition highlighted the meaning of paid work and willingness to work even more euphorically, probably because for them work was the main link to society. Apparently participants valued the advantages of having a job as opposed to receiving wage replacement benefits as provided in the Netherlands <sup>[28]</sup>. However, it is a fact that they were aware that from an economic point of view this was a more beneficial choice.

The three subsequent themes: (2) *Solving problems oneself: "I am not going to tell"*; (3) *Reaching a turning point: "I have to be realistic"*; and (4) *Taking into account environmental aspects: "I can understand both sides"*, reflected participants' shift from an internal towards an external focus by changing strategies to maintain their position at their jobs while adapting to physical decline. Comparison with the process of self-management tasks identified in persons with a chronic disease after being diagnosed (focusing on illness needs, activating resources, and living with a chronic disease) became apparent <sup>[29]</sup>. Participants' goals of compensating for physical deterioration varied with the phase of the disease. Initially participants struggled and did not always recognize the significance of vague symptoms. In families with hereditary records it could be expected that persons with these symptoms were, to some extent, prepared for future challenges. However, there was no indication that this knowledge was used by the participants to choose an education or profession that could accommodate their subsequent physical deterioration.

#### *The Moral Pressure at Work to 'Keep Up Appearances'*

As soon as participants were diagnosed, their first reaction was to solve problems themselves, which was identified as the second theme. Comparison with the first stage of the self-management process, namely focusing on illness needs, evolved <sup>[29]</sup>.

Individual problem solving, disguising limitations and postponing disclosure might be a reflection of the tensions participants experienced about when and if to disclose their condition and might coincide with a stage of denial<sup>[30]</sup>. Disclosure was apparently experienced as 'irreversible' and therefore avoided. Reasons for the participants' reluctance to complain and their decision to postpone disclosure was apparently related to fear of being put in an unfavorable position, fear of being dismissed and consequent financial instability. Risk of financial instability and loss of self-worth might have been reasons for delaying disclosure which was also found in persons with slow progressive neurological diseases such as early-onset MS<sup>[31]</sup> and for persons with a chronic disease in regards to job retention<sup>[32, 33]</sup>. The struggle with decisions regarding whether to disclose and discuss disabilities at work is seen in other studies<sup>[34]</sup>. This dilemma and the accompanying fear of potentially negative consequences are actively debated on websites of client associations<sup>[35]</sup> and uncertainty seemingly caused pressure to keep up appearances at work.

*How Keeping Up at Work and at Home Shapes Decisions about Remaining at Work*  
 Problem solving or coping included pondering and negotiating first within personal relationships and later on in work relationships with the aim to maintain their job was strongly developed by participants of this study. Developing coping strategies as a way of adjusting to a chronic disease and activating resources was also an important finding in other studies<sup>[29, 36, 37]</sup>. They did not always recognize the impact of their condition and problem solving strategy on colleagues. Based on colleagues' reactions after disclosure it became apparent that it might have been difficult for colleagues to notice participants' work problems. However, these colleagues did not dare to start talking about it with them. It can be assumed that participants' ignorance of symptoms and their reluctance to complain about their problems at work, possibly leading to less production, might make it difficult for colleagues, line managers (LM), human resource managers (HRMs), occupational physicians (OPs), physicians or health care workers to provide adequate assistance<sup>[38]</sup>. Only after disclosure some participants realized that it might have been wiser to communicate work issues at an earlier stage, instead of continuing working to the detriment of the family as some participants did. Haafkens et al. found that according to LMs and HRMs, employees must accept their own share of responsibility for job retention<sup>[38]</sup>.

This dilemma was apparently not experienced equally among participants. Some participants communicated openly with colleagues and superiors, and successfully bargained about necessary changes aimed at continuation of the job. In these cases communication and sharing the grief of future physical, personal and societal consequences seemed to be instrumental in their relief and resulted initially in compassion by some colleagues and superiors. However in other cases open communication failed when colleagues and superiors seemed to have forgotten participants' needs and workload was increased again and again. This means that there were differences in environmental conditions which influence the vulnerability



of the persons with NMD. This was confirmed by data of Jans et al.<sup>[39]</sup> They discussed the complexity of decision making regarding whether, when, and how to discuss disability, which depended on work place culture and personal choices<sup>[39]</sup>. Our findings that insufficient support from colleagues or LMs were felt as threatening are understandable. In some studies about work and the chronically ill it was found that these factors were positively associated with withdrawal from the labour force<sup>[37]</sup>.

When open communication at work was not effective, participants expressed being disillusioned or even angry. Thus communication and disclosure did not always make things better.

When physical deterioration could no longer be concealed, most participants reached a turning point, identified as theme three, in which the struggle of letting go and accepting change became apparent. Reaching a turning point was true for every participant although at different points of time or phases of disease and this seemed to be a moment to re-evaluate life<sup>[29]</sup>. Although wanting to be realistic, participants struggled with the process of letting go, changing and balancing private life and work needs. Some participants kept bargaining till the bitter end and seemed not to reach a turning point, seeking for normalcy at work<sup>[29]</sup> carrying out work tasks and responsibilities as long as possible.

#### *Environmental Aspects Influencing Job Retention*

Irrespective of the time line described in the findings, a variety of environmental aspects influencing job retention for participants were identified. First of all, the provision of assistive products, timely work adaptations, work adjustments and accommodation through adapted work conditions may enhance participants' employability. In other studies LMs, HRMs and OTs confirmed these preconditions for job retention of the chronically ill.<sup>[38, 40-42]</sup> As Andries et al, stated "Where disabilities in daily life and job demands coincide, work handicaps may be the result. Work handicaps can be removed or their effect mitigated by work adjustments"<sup>[40]</sup>. Secondly, previous and current relationships at work with colleagues as well as superiors and the policy of the company were valued by participants. In the study by Haafkens et al, good manager/employee cooperation and knowledge transfer within the company was identified as an important cluster by LMs and in the same study HRMs perceived organizational policy and culture as the most important mechanism<sup>[38]</sup>. Superiors' willingness to support participants depended on their ability to manage company policies. However these policies could be overruled by reorganizations and macro-economic circumstances<sup>[43]</sup>. These issues might also influence opportunities for job retention or possibilities for alternate employment for participants. According to some participants these circumstances were covertly used for termination and they experienced being in a disadvantaged position compared to healthy colleagues with regard to job retention.

### *The Chain of Delay*

The relative rarity and variety in the hereditary types of NMD<sup>[23]</sup> might explain the delay in physicians' recognition of early symptoms. This, along with the participants' lack of awareness of the seriousness of first symptoms, might create the first delay in receiving proper support related to job retention. The apparent delay in diagnosis and the subsequent delay in disclosure of work related problems by persons with NMD deserve attention. A survey of the opinion of disabled people's organization in the UK about factors hindering return to work or remaining at work revealed, amongst other factors, delayed consultation and a lack of appreciation of work place issues. Employers were seen as unresponsive to the needs of workers, with negative attitudes to disability<sup>[44]</sup>.

The first challenge might therefore be augmenting general practitioners' (GP) alertness to early symptoms, and timely referral to a neurologist for a proper diagnosis. The diagnosing physician's awareness of the implications for employment and initiation of a timely referral of persons diagnosed with NMD to allied health care professionals (HCPs) may be important in this respect<sup>[45]</sup>. If the recently diagnosed person is still employed, but not on sick leave and thus not under the care of an OP, a hospital based OT might be the appointed professional to assist in possible employment dilemmas<sup>[33, 42]</sup>.

At a later stage of the disease, when participants themselves became aware of possible negative consequences of not facing their condition they became aware that they had chosen only short term solutions e.g. decreasing hours, which worked negatively in the long term. It might be worthwhile to have peer discussions about work problems and successful and less successful strategies. Timely information from peers for persons with NMD who are struggling with the issue of disclosure, might help them to identify their chances of success. These findings are in concord with another study on employment of people with a slow progressive disability that revealed that struggling alone and hiding the diagnosis did not always seem to produce the desired results<sup>[37]</sup>. In another study, disclosing an illness was the strongest predictor of receiving work adjustments and social support for persons with chronic illnesses<sup>[46]</sup>. These studies underline the potential benefits of early disclosure but there is also a risk of not getting support.

### *Strengths and Limitations of the Study*

Persons with one of three types of NMD, varied demographics (age, gender and education) and diverse jobs were interviewed. Data were saturated with a variety of positive and negative experiences. To minimize bias by misunderstanding, analysis was conducted on Dutch transcripts of the interviews<sup>[19]</sup>. To guarantee meaningful translation, only the selected quotations used in this article were translated into English in a side-by-side cooperative procedure with the first researcher and a professional translator. The researcher explained to the translator the intended meaning and its context in Dutch. The translated quotes and subtle meaning

differences were discussed by Skype<sup>®</sup> sessions with a native speaker, knowledgeable about the content and type of research<sup>[47]</sup>.

All interviews were conducted by the same researcher, while the process of the data collection and analysis of the content of the interviews were conducted with 2nd and 3rd researchers. It was anticipated that the discussion of the findings within a diverse research team would prevent a narrow or biased view of the data, given that only the researcher read all of the transcripts in their entirety<sup>[25]</sup>.

We choose to interview persons with NMD who were employed at least 12 hours a week. Even participants with relatively severe physical hindrances in our study viewed work as meaningful, likely due to the people sampled. It is possible that persons with NMD who were no longer employed would have had different experiences.

The validity of this study may be enhanced by observations on the job<sup>[25]</sup> and interviewing employers about their experiences in supporting employees in job retention<sup>[48]</sup>. However, we did not choose these methods so as to guarantee that participants provided their work narrative under confidential circumstances. Neither was it the intention to interfere in employer-employee relationships. Interviewing employers about company policies in general would not have provided information regarding management of NMD related problems. It is unlikely that an individual manager would experience this rare condition on a regular basis<sup>[48]</sup>.

Although at the start of the interviews the main question was related to their current work, participants tended to narrate their story from the past to the present with a glimpse into the future. Instead of using an open in-depth interview we could have chosen to use a regressive narrative as a starting point to detect a more detailed course of deterioration or decline along with participants' employment experiences<sup>[49]</sup>.

#### *Implications for Professionals and Policymakers*

These findings might be useful for professionals and policymakers. Of note was that participants' narratives gave insight into the complexity of the process of working with NMD. It was apparent that participants still at work tended not to complain, and delayed disclosure at work until symptoms became tangible. Participants who gained insight into the complexity of maintaining employment with NMD concluded that they would advise others to communicate their work problems earlier. Therefore, empowerment of employees by professionals to help them not to postpone, but to communicate employment issues at an early stage seem to be important. Indeed the importance of timely identification of employment issues in the working life of persons with a chronic disease to minimize the stress of deteriorating abilities was emphasized in other studies<sup>[50]</sup>. The diagnosing physician could play a role in supporting persons with chronic diseases by identifying actual or expected employment issues and providing information on the benefits of referral to a multidisciplinary team or occupational health services<sup>[51]</sup>. Physicians could be provided with brief checklists to assist them to explore the possible impact of the disease on work and to look for early symptoms of deterioration. These checklists have been

developed earlier <sup>[13, 43, 52]</sup>. Johnson et al (2004) emphasized the importance of proper timing of decisions before recommending action impacting employment. Health care providers' awareness of the complexity of the process and of the issues related to employment were found to be important determinants <sup>[53]</sup>.

Our study revealed that supportive social relationships at work, especially a considerate superior were perceived as key in job retention. Based upon the findings we might conclude that the main reason that employers support employees in job retention might be the 'goodwill' built up in the past and a good personal employee-employer relationship.

A study of the effectiveness of the Dutch Law (Wet Verbetering Poortwachter) <sup>[54]</sup> implemented in 2001 obliging companies to develop a return to work plan in case of sick leave and imposing penalties to force both employers and employees to develop an adequate (re-)integration plan resulted in the same findings, namely that in spite of this law the main reason for employers to support employees with a disability was employers' personal involvement, not the sanctioning law <sup>[55]</sup>.

#### *Future Research*

This research was limited to the experiences of employees with NMD still at work regarding their strategies pertaining to job retention. Further research including employees with NMD after job loss, and health professionals' views on employment issues for employees with NMD is necessary to better understand possible problems and determining factors in job retention.

Research on the effect of educating physicians and allied health professionals in early recognition of the impact of chronic diseases on employment participation, as well as cohort studies on the long term effect of immediate one-off consultation by an interdisciplinary team to support persons with NMD at risk for unemployment after diagnosis is recommended.

Seeking the perspectives about work by persons with NMD who have already left work could also be a valuable addition to this research.

## **Conclusion**

Paid employed persons with a slow progressive adult type of NMD who are confronted with decreasing physical abilities in their most productive years valued employment as an important aspect of their lives. The meaning of work as well as the tension of decisions accompanying employment throughout the lives of participants was identified in four themes: (1) *Experiences regarding the meaning of work*: "Work is belonging"; (2) *Solving problems oneself*: "I am not going to tell"; (3) *Reaching a turning point*: "I have to be realistic"; and (4) *Taking into account environmental aspects*: "I can understand both sides". Paid employed persons with NMD might have a strong willingness to work, irrespective of disease severity, likely due to the persons

sampled (11). Despite tensions, participants expressed why they choose to stay at work by elaborating on social, economic and emotional benefits they experience from being employed. This study also showed that participants were challenged to keep up at work versus home, and how this shaped decisions about remaining at work. Participants' deployed coping strategies focusing on keeping their job by hiding symptoms and postponing disclosure. Participants experienced a moral pressure at work to 'keep up appearances', and tensions with regards to when and if to disclose, since disclosure didn't always make things better. Often the impact of their condition and behaviour on colleagues became apparent to them only after disclosure, when communication became more open. It is then that they perceived assistive products, communication of work problems, work accommodations, considerate colleagues and above all good relationships with superiors willing to support their job retention as increasingly important along with greater dependence due to physical deterioration. Support from superiors was valued most highly, since the participants perceived their role as most important in the final decision making regarding participants' job retention. Apparently some participants felt also threatened by work reorganization. Indeed macro-economic circumstances and reorganization could overrule employers' intentions and governmental regulations that are meant to support people with a disability in job retention. In that case participants with an NMD felt disadvantaged compared to healthy colleagues. Implications for health professionals might include awareness of the significant impact of a slowly decreasing physical condition on employment. Timely communication and appropriate referral of persons with a work problem to a health or occupational professional is recommended in order to inform and empower employees with NMD in job retention.

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# **CHAPTER VI**

## **General discussion**



## Introduction

In this last Chapter the main findings of the studies in this dissertation are discussed and recommendations for clinical practice are given.

The case in *Chapter 1* concerning a woman with an adult onset neuromuscular disease (NMD) represents the real life problems which persons with a slow deteriorating NMD encounter. The case starts with her uncertainty about the future when the first clinical features become manifest up and until severe hindrances start to impact daily functioning, including paid work.

This dissertation focuses on persons with three relatively common adult types of NMD and describes the confrontation with disease symptoms in the middle of a person's career. These types are: facioscapulo-humeral muscular dystrophy (FSHD), a myogenic disorder; hereditary muscular and sensory neuropathy (HMSN), a neurogenic disorder; and adult onset myotonic dystrophy (MD), a multisystem disorder.

The first two quantitative studies give information about factors influencing work employment status from epidemiologic and scientific perspectives (Chapter 2 and 3). In *Chapter 4*, the professional perspective was studied by investigating to what extent persons with NMD receive referrals to and subsequent recommendations from occupational therapists, physical therapists and speech therapists, categorized as health care professionals (HCPs), in a hospital setting to support them with their employment problems. Lastly, the client perspective was investigated by performing a qualitative study in which employed clients with NMD were interviewed (Chapter 5).

The dissertation as a whole focuses on the discovery of factors related to sustainability of employment of people with the NMD types investigated. Based on our findings clinical implications and possibilities for future research are presented.

### Employment status of persons with NMD: what did we learn?

A relatively high employment rate was found among persons with FSHD and HMSN compared to persons with MD (Chapter 3) or persons with other chronic diseases<sup>[1]</sup>. They seem to be able to retain employment more successfully than persons with MD, a multisystem disorder that also affects cognitive functions<sup>[2]</sup>. In general, persons with HMSN and FSHD who experience slow progressive physical deterioration do not tend to take sick leave and adapt gradually to physical changes during their working life. This adaptation process takes place in concert with interaction with the family and work environment. Therefore it is not surprising that sustainability of employment is mainly influenced by personal factors as well as by factors in the family and work environment. However, as could be expected, employees with NMD are at an increased risk of reduced performance at work and work disability (Chapter 3). Although the employment rate of employees with NMD (Chapter 3) is somewhat lower than for

healthy persons<sup>[3]</sup>, many persons with NMD are able to cope with NMD at work and maintain their employment for a relatively long time. An important factor here is that these people already have a steady job before symptoms become visible and/or bothersome.

This dissertation adds information to the existing scarce literature about participation in paid work of persons with NMD which can be used to support them to remain employed during the slow progressive process of physical and sometimes cognitive deterioration (Chapter 2). Most studies on return to work (RTW) and occupational rehabilitation are focused on employees returning to normal job responsibilities after a period of absence<sup>[4]</sup> and not on maintaining employment despite a chronic disease. There is growing interest in work place challenges and limitations posed by chronic medical conditions and whether these are effectively identified and addressed, in order to prevent long-term absence and work disability<sup>[5]</sup>. This shift of focus from 'return to work' to 'staying at work' requires other ways of thinking about monitoring, risk factor identification, health care practices, employer policies and interventions. This is especially important in NMD where the slow deterioration of physical conditions means that decision-making should not only focus on the current situation but also on the anticipated future. From the in-depth interviews we learned that most people with NMD seem not to take future challenges into account (Chapter 5).

To categorize factors contributing to employment status as found in the four studies performed, the expanded version of the International Classification of Functioning, Disability and Health (ICF)<sup>[6]</sup> was used (*first column* table 1). For each study, as outlined in columns 2-5, the main findings are ordered in conformance with the ICF categories and for each of these findings the level of evidence is described.

As can be seen in this table, Chapter 3-5 contribute to the level of evidence of some factors found in Chapter 2: the Systematic Review (SR). The *second column* of table 1 shows the ten factors that were indicative for employment status as found in the SR (Chapter 2). However, only limited evidence for this could be found (Chapter 2, appendix 1 of the SR). Eight of the 84 potential factors in the studies included in the SR were also found to be significant factors or relevant (sub)themes in at least one of the other three studies. For this reason these factors are presented (in italics) in the second column. All factors, barriers as well as facilitators, were significantly related to employment status or were clinically relevant, and could be accommodated by the expanded ICF-scheme<sup>[6]</sup> indicating that the scheme works well to fill the gap between terminologies used by researchers, HCPs, professionals in the work environment and employed persons with NMD.

An overall view of table 1 shows that factors significantly related to employment status in persons with NMD were better general physical and mental function (including more muscle power, better concentration and lower levels of fatigue and less pain) and higher levels of activity. Predictive factors for employment status varied among the groups, i.e. better concentration and less pain for the HMSN group, better

physical functioning for the FSHD group were found, and for both the FSHD and MD group, higher activity levels. Strong predictive personal factors were younger age, being male and high levels of education. Work related personal factors included motivation to work and more active coping strategies. Advantageous environmental factors were the presence of adaptations and good relationships at work especially with the superior. Our subsequent studies revealed a shift from disease related factors and physical/mental functions and activities/participation in the first two quantitative studies to work related environmental factors and work related personal factors in studies 3 and 4. A better understanding of the consequences of NMDs on sustainability of work can be obtained by considering more than disease specific categories<sup>[7]</sup>, in particular, environmental and personal factors<sup>[6, 8]</sup>.

The main factors are discussed below and compared with results from other studies to highlight their significance for clinical practice. It is important to determine which of the factors found in this research can be influenced by an intervention to support employees with an NMD to keep their jobs.

## A. Health

### A.1. Work impacting disease

The presence of NMD negatively influences employment status. Persons with an NMD experience deteriorating stages in the course of their lives to which they gradually adapt, typically with a significant decline in their thirties. It was remarkable that persons with NMD first adapt silently to physical deterioration and only communicated problems at work when problems could no longer be concealed. Pro-activity, e.g. a strategic choice in type of work that could be performed later on despite severe physical hindrances, was rarely found (Chapter 5). Thus at the time of diagnosis it might be helpful to offer information to enable affected persons to plan their career more effectively.

### A.2. Functions

The factors categorized as physical and mental functions - such as fatigue, pain and decreased concentration that can interfere with employment participation - were predominantly present in the first two quantitative studies.

*Fatigue severity* differed significantly between employed and not employed employees with NMD and the separate groups FSHD and MD. Fatigue severity was not a predictive factor for work participation in persons in this study, however it is noteworthy that all groups in the study reported high levels of fatigue above the cut-off point of >35 'severe fatigue' on the subscale CIS-fatigue, as defined by Vercoulen et al.<sup>[9]</sup>. Some participants in the qualitative study also mentioned fatigue explicitly in terms of being exhausted after work or used other words to indicate fatigue such as low energy levels, or '*contemplating job demands and other roles in life*' in order to describe

**Table 1.** An overview of factors associated with employment status of persons with NMD, extracted from all studies included in this dissertation

	<b>Classification of employment factors</b>	<b>Employment status of patients with neuromuscular diseases</b>
<b>A. HEALTH</b>		
<b>A.1 (Work related) disease/disorder</b>		
<b>Presence of NMD</b>	Having an NMD is negatively associated with employment status	
<b>Type of NMD</b>	HMSN is negatively associated with employment status MD is negatively associated with employment status	
<b>A.2 Functions</b>		
<b>General physical functioning</b>	Physical functions are associated with employment status	
<b>Muscle power functions</b>	Muscle power functions are associated with employment status	
<b>Fatigue</b>		Employed people with NMDs, and the separate FSHD and MD groups are <b>significantly</b> less fatigued than the same not-employed groups
<b>Mental functions</b>		Higher levels of concentration <b>significantly</b> contribute to employment status of persons with NMDs and the separate MD group
		Higher levels of concentration are a <b>predicting</b> factor for employment status of the HMSN group
<b>Pain and sensory functions</b>		Less bodily pain contributed <b>significantly</b> to employment status of persons with NMD and the FSHD and HMSN group
		Less bodily pain is a <b>predicting</b> factor for employment status of persons with HMSN
<b>A.3 Activities / A.4 Participation</b>		
<b>Activities and participation</b>		Higher levels of activity for persons with NMD, FSHD and MD contribute <b>significantly</b> to employment status
		Better physical functioning for persons with MND in general and the separate groups FSHD and MD contributed <b>significantly</b> to employment status
		Better physical functioning <b>predicts</b> employment status for persons with MND in general and the separate groups FSHD
		Better Social functioning contributed <b>significantly</b> to employment for persons with NMD

Exploring consultation reports	How persons with NMD perceive employment participation.	Level of evidence*
		+
		+
		+
		+
		+
		++
		++
		+++
		++
		+++
		++
		+
		++
		+



	Classification of employment factors	Employment status of patients with neuromuscular diseases
<b>B. ENVIRONMENTAL FACTORS</b>		
<b>B.1 Work related environmental factors</b>		
<b>Determinants at macro level</b>		
<b>Terms of employment</b>	<i>Adapted working hours enhance sustainability of work</i>	
<b>Social relationships at work</b>	<i>Consideration of superior and colleagues enhance sustainability of work</i>	
<b>Task contents</b>	<i>Adaptations in task content and in pace enhance sustainability of work</i>	
<b>Working conditions</b>	<i>Adaptations of tools and furniture, enhance sustainability of work</i>	
<b>C. PERSONAL FACTORS</b>		
<b>C. 1 General personal factors</b>		
<b>Age</b>	Younger clients with NMD have a higher employment rate	Younger persons with NMD, FSHD and HMSN have higher employment rates
<b>Gender</b>	Male clients with NMD have a higher employment rate	Male employees with NMD, FSHD and MD have higher employment rates
<b>Education</b>	Clients with NMD with higher education have a higher employment rate	Higher educated persons with NMD, FSHD and MD have higher employment rates
<b>C.2 Other diseases/co-morbidities</b>		
<b>C.3 Work related personal factors</b>		
<b>Type of occupation</b>	Type of occupation influences employment rate in persons with NMD	
<b>Motivation to work/ drive to work</b>	Persons with NMD with an expressed interest in employment have a higher employment rate	Better motivation for persons with NMD and MD <b>significantly</b> influences employed rates
<b>Coping strategies</b>	<i>Flexibility of the employee</i>	
<b>Employability</b>		Better vitality <b>significantly</b> influences employment rates for persons with MND and FSHD

\* Highest level of evidence presented: +++ Factor predicting employment status (based on regression analyses); ++ Factor significant different between employed and not employed people with NMD; + Indication for a relationship with employment status; +/- findings of the qualitative study.>

Exploring consultation reports	How persons with NMD perceive employment participation.	Level of evidence*
	Taking into account environmental aspects	+/-
	Interfering macro-economic circumstances	+/-
Terms of employment need improvement		+/-
	Considering colleagues' positions Considering employer's position	+/-
Task content needs improvement		+/-
Work conditions: assistive products were present in 76% or needed improvement in 40% of the cases.	Appreciating assistive products	+/-
Employed persons with NMD were younger		++
More men than women with NMD were employed		++
The level of education was higher for the employed persons with NMD		++
Type of job		++
<b>Experiences regarding the meaning of work:</b> Working for personal benefit Working to be a member of society Working for economic and social benefits Contemplating job demands and other roles in life Experiencing work as a burden		++
<b>Solving problems oneself</b> Disguising limitations thus postponing disclosure at work <b>Reaching a turning point</b> Struggling to let go Accepting change an emotional process		+/-
Using strategies to keep one's job in the long run		++

> ICF: International Classification of Functioning, Disability and Health; NMD: neuromuscular diseases; HMSN: hereditary muscular and sensory neuropathy; MD: myotonic dystrophy; FSHD = facioscapulo-humeral muscular dystrophy

the influence of fatigue on participation. Fatigue severity is thus a clinically relevant finding. Fatigue can lead to '*presenteeism*', showing up at work too tired to effectively carry out the job. Among other physical factors fatigue was also found to be the most common reason for loss of employment at work in previous qualitative studies on multiple sclerosis, which is another type of slow progressive neurological disease<sup>[10,11]</sup>.

*Pain* was found to be a factor that can interfere with employment participation in persons with FSHD and MD (Chapter 2). A recent study in which the impact of bio-psycho-social factors on chronic *pain* in persons with FSHD and MD was investigated<sup>[12]</sup> confirms that pain is associated with these two types of NMD. When looking at the employment rate of persons with FSHD, they tend to work despite pain.

*Mental functions, such as concentration*, differed among the three types of NMD investigated. Poorer concentration is most often present in persons with MD indicating that special attention needs to be given to cognitive decline in persons with MD, in particular because persons with MD tend not to ask for support due to their lack of initiative<sup>[13]</sup>.

The HMSN group reported *less fatigue, less pain* and higher levels of *concentration* which might explain their relatively high employment rate (Chapter 3)<sup>[14]</sup>. Although disease symptoms vary among the types of NMD and differ in progression and severity, persons with NMD increasingly have to deal with the consequences of the disease, such as loss of muscle strength, reduced concentration, fatigue, and/or pain. Adapting to these changes can be done by conceding the difficulties to oneself and asking for support from the environment. Biological, social and psychological strategies determine ultimate capabilities and adaptability of people with NMD. The complexity and individual differences are large and therefore interventions need to be tailored and fine-tuned for each individual.

#### A.3. Activities & A.4. Participation

One subtheme of the qualitative study: '*Contemplating job demands and other roles in life*' revealed the impact on relatives of participants who were at risk of job loss as was also found by Varekamp and Van Dijk<sup>[15]</sup>. They reported that finding a balance between work and life at home, i.e. work-home interference, posed a problem for 90% of their participants with a chronic disease and this was a severe problem for more than half of them<sup>[15]</sup>. The theme revealed the difficult choices participants encounter in keeping up appearances at work at the expense of family relationships or leisure activities, before coming to the decision to disclose their condition. This theme also showed that having to make choices between duties at home and at work caused a struggle. When lack of energy causes employment problems and the person with NMD chooses not to disclose this problem at work, family duties are sacrificed in favour of work duties. *Social support at home*, as such, was not identified as a theme but was expressed as an issue among partners to continue work, not always taking into account future prospects. We found that decisions made by participants or by couples could hugely impact their families in positive and negative ways. The desire to work was strong

and choices were made at the expense of the family and consequences were not always foreseen. One spouse had quit her job to enable her husband with NMD to stay employed, a decision that would probably have to be reconsidered in the near future due to the progression of the disease. A better understanding of the prognosis could have contributed to the decision-making process and to insight into the consequences.

### **B.1. Environmental factors**

In order to know which factors are clinically relevant it was important to consider which employment participation themes emerged from the recommendations of HCPs and persons with NMDs themselves (Chapter 4 and 5). It was helpful that these two studies emphasized personal and environmental factors at micro, meso and macro level.

#### *B.1.1. Work related environmental factors at micro-level*

The majority of persons with NMD experiencing physical hindrances at work are able to continue work when the required accommodations and assistive products at work are available (Chapter 4 and 5). The environmental factors related to work found in the literature are the so called '4 A's' in Dutch, covering prerequisites for job retention: 1) Arbeidsvoorwaarden [Terms of employment: such as *adapted working hours*], 2) Arbeidsinhoud [Task content: such as *adaptations in task content and pace*], 3) Arbeidsomstandigheden [Working Conditions: such as *adaptations of tools and furniture*], and 4) Arbeidsverhoudingen [Personal relationships at work: such as *consideration and support of superior and colleagues*]. Physical deterioration hindering employment participation can be compensated at micro- and meso-level<sup>[6]</sup>. The fact that persons with FSHD and HMSN mainly have difficulties with their physical condition (which can be compensated by adapted *terms of employment*, such as adapted *working hours* and permission to work at a slower pace, and changing *task content*, such as job rotation or retraining for a new position), might explain the relatively large percentage of clients with HMSN and FSHD who are employed. It can be expected that there are also negative consequences of taking such measures for job retention. For example when terms of employment are adapted, and the person with NMD is allowed to work fewer hours, this will have consequences for the level of income and opportunities for promotion. Paying more attention to *improving working conditions* and providing adjustments and assistive products offer opportunities to reduce the negative consequences of having a chronic disease with physical limitations<sup>[6]</sup>.

### *B.1.2. Work related environmental factors at meso-level*

*Personal relationships at work*, in particular the relationship with superiors, were an important theme for the participants in the qualitative study. Having been able to build goodwill and personal relationships previously at work was perceived as the most important prerequisite later on for getting support from superiors to stay at work (Chapter 5). Although personal relationships at work were not a significant factor in the SR and findings in qualitative studies cannot be generalized, the clinical relevance seems clear. Evidence for the importance of good personal relationships at work to stay employed or to return to work were found in many other studies in which factors or determinants related to disability and work were investigated<sup>[17-19]</sup>. MacEachen et al.<sup>[20]</sup> stated that goodwill and trust are overarching conditions that are central to successful RTW. The factor social support at work was reported earlier as a key precursor in the RTW process of injured employees<sup>[21, 22]</sup>. The implication for employers might be that they have to create a safe environment for employees with a health condition so that these employees can openly communicate their work problems, and find solutions without fear of being dismissed<sup>[11]</sup>. Competent workers, including those with a disability, have a better chance of access to jobs and the work environment, as well as to accommodations necessary to assure equal participation in society<sup>[23-25]</sup>. The willingness to give support to competent employees with a disability can however be overruled by the impact of macro-economic circumstances, such as an economic crisis, which was also an important theme in the qualitative study (Chapter 5), when participants explained their fear of losing their job. Participants experienced being in a disadvantaged position compared to healthy colleagues with regard to job retention in circumstances of reorganization. It is true that people with a disability, but also older people, not only feel disadvantaged compared to healthy and younger people, but are actually in a less favourable position in spite of anti-discrimination acts and governmental legislation<sup>[26, 27]</sup>.

From a healthcare perspective, it might be important for diagnosing neurologist to identify patients' risk factors early, taking work into account, and refer them promptly to rehabilitation facilities in or outside their hospital in order to provide support to overcome fear and hesitation to disclose. The designated professional, e.g. the occupational therapist might be able to assess employment issues and inform the person at risk how to get support to stay employed when the impact of the disease gets worse as found in chapter 3. Practical solutions can be provided and paid for by the Dutch government to support sustainable employment for persons with a chronic disease<sup>[28]</sup>. However, on the job the law dictates responsibility for job retention for both the employer as well as the employee<sup>[29]</sup>. HRM managers and line managers collaborate with the employee to make job retention possible. Furthermore, occupational physicians assist employers to follow this process carefully, in order to avoid sanctions dictated by the law. As an expert in the field of work and health, the role of the occupational physician is to guide the employee in case of an illness or disability and objectively decide whether measures can be taken for the employee to

stay employed or to recommend a disability pension<sup>[30]</sup>. This gate keeping role may be in conflict with the willingness of people with NMD to remain employed and the intentions of health physicians, i.e. neurologist or rehabilitation physician as well as HCPS, such as occupational therapists or physiotherapists, to empower persons with a disability to stay at work. With their professional background occupational therapists are the members of the rehabilitation team that can play a role in assisting employers and human resource personnel in determining ‘reasonable prerequisites for job retention’<sup>[23]</sup>. Occupational therapists should therefore better utilize their full range of skills when involved in vocational rehabilitation, work hardening programs, or occupational settings<sup>[23, 31-33]</sup>.

### *B.1.3. Work related environmental factors at macro-level*

The impact of work related environmental factors on employment, such as HRM-policy, legislation and macro-economic circumstances have a tremendous impact on employment opportunities for people with a chronic disease or disability. The impact varies within companies, countries and over time<sup>[26, 34]</sup>.

Several initiatives in the Netherlands and elsewhere indicate that supporting sustainability of work for people with a disability is an important societal issue. One of these initiatives is the *International Forum on Disability Management* (IFDM), a worldwide network of researchers initiated in 1999 with the aim of supporting employers in their employees’ job retention. Many countries and companies are connected to this network and IFDM congresses have been held where strategies, interventions, policies and legislation have been shared and discussed<sup>[35]</sup>.

In the same year of the development of DM, a Dutch initiative: the *Early Intervention Rehabilitation Care Foundation* (EIRCF) was established to support people with threatening and/or existing limitations with various forms of vocational rehabilitation (VR)<sup>[36]</sup>. VR refers to a multi-professional approach that is provided to individuals of working age with work-related impairments, limitations, or restrictions. The primary aim of VR is to optimize work participation<sup>[37]</sup>.

Currently 14 accredited partners of EIRCF are legally recognized rehabilitation centers affiliated to rehabilitation. The EIRCF partners have developed a unique method in the field of VR and reintegration. They are specialized in offering various high quality rehabilitation products and provide services that enable people to remain in the labor force. With 17 locations across the country they have nationwide coverage. The EIRCF provides outpatient rehabilitation care that is assured for every Dutch citizen through basic health insurance policy. VR requires various disciplines, e.g. kinesiologists, occupational therapists, physiotherapists, psychologists, social workers, that work closely together in an interdisciplinary team in collaboration with a rehabilitation physician. The first large effect studies on the benefit of VR have been performed on large client groups such as persons with low back pain<sup>[38]</sup>, but not yet on persons with specific diseases such as NMD.

Governmental legislation is intended to help persons at risk to take part in the world of work. It should ensure that it is safe for individuals to openly communicate about issues at work. The European Foundation for the improvement of living and working conditions has developed employment guidance services for persons with a disability<sup>[39]</sup>. These macro-economic initiatives are valuable for VR programmes, health professionals and occupational health professionals who support individuals with a chronic disease or disability in job retention.

Another, more recent initiative is the WDPI, *Work disability participation and integration*, a scientific conference for research on preventing work disability and promoting safe and sustained return to work across a range of conditions, formed in 2011. It was organized to provide an opportunity for interdisciplinary and international exchange of ideas on preventing work disability<sup>[40]</sup>.

Improved knowledge and mutual understanding of each other's field (occupational care and health care), however, could maximize sustainability of employment for persons with a disability. Initiatives to increase knowledge exchange on topics concerning employment and health are evident in research<sup>[36, 41, 42]</sup>, but also in the development of a large number of post academic courses on this subject in the Netherlands<sup>[43, 44]</sup>.

### C. Personal factors

#### C.1. General personal factors

The three quantitative studies revealed similar demographic factors related to employment status: persons with a younger age, being male, and having a higher education showed higher employment rates.

The factors *age and gender* cannot be influenced by an intervention. However, the relevance of the influence of age is in line with the progression of the disease. That age influences employment was also found in other studies on chronic diseases<sup>[45, 46]</sup>. It is possible that in the future gender differences in employment status between men and women will disappear, not only in the healthy population but also for people with a chronic disease<sup>[11]</sup>.

The *level of education* in the second study was higher for persons with HMSN and FSHD than for persons with MD (Chapter 3). To sustain employability for persons with MD it might be necessary to strive for work environments that fit both their intellectual capacities as well as their physical capacities. The lower employment rate of persons with MD might be explained by complicating factors related to the symptoms of this multisystem disorder, such as lack of initiative, subsequent reduced performance and not asking for help, which puts persons with MD in a disadvantaged position<sup>[13]</sup>. Decrease of cognitive performance plays a role in job loss as was found in other studies of persons with slow progressive neurological diseases<sup>[11, 47]</sup>. The persons included in the studies were persons with an adult onset disease which had given

them the opportunity to have a normal childhood and education. Level of education and employment are strongly related<sup>[26]</sup>. Well-educated persons with NMD are apparently able to start a regular career and are better able to compensate for physical hindrances with work accommodations such as assistive devices and adaptations at the work place. Less educated persons working in blue collar jobs are in general more vulnerable to job loss, especially if physical functions deteriorate. The impact on employment of having a chronic disease in combination with a low *educational level* was also found in a study by Holland<sup>[27]</sup>, who investigated how macro-level contexts and policies affect the employment chances of disabled people.

### C.3. *Work related personal factors*

A person with a slow progressive chronic disease at work is confronted with a variety of personal dilemmas, not only to cope with the disease and with the slow progressive deterioration of functions itself, but also how to cope with the environment<sup>[8]</sup> such as expectations of relatives, colleagues and superiors at work, and professionals in occupational care and health care.

In Chapter 2, 3 and 5 the work related personal factor ‘motivation to work or drive to work’ emerged, stated as *expressed interest in employment, better motivation* and by participants’ arguments regarding *the advantages and disadvantages of having a paid job*. The participants were selected because they were employed, and thus in spite of some disadvantages related to disease symptoms such as fatigue, they formed a motivated, but selective group of persons with NMD. Their willingness to work was persistent for a variety of reasons. If the study had included unemployed persons, arguments against employment, like not having the pressure to work, might have been heard. Some participants revealed social and economic reasons for having to work. Others expressed awareness of the risk of being marginalized and social exclusion. The fear of social exclusion is often seen in persons with a chronic disease and might also play a role in the motivation to stay at work, although sometimes this results in presenteeism, instead of taking sick leave, consulting an occupational physician at work<sup>[5]</sup>.

Having a job was as rewarding for participants, in both financial as well as psychological sense, as it is for other people, but frequently participants seemed to apply inefficient coping strategies to sustain employment as they ignored the implications of their disease for themselves as well for other people involved (Chapter 5). Communicating hindrances was postponed due to fear of discrimination resulting in loss of promotion or being fired, based on experiences of colleagues at the work place. Their narrative described subsequent coping strategies progressing from hiding symptoms to acceptance such as communicating problems and coping with progressive hindrances by asking for material and personal support.



### **The potential role of professionals in the support of employed persons with NMD**

When work performance is threatened, persons with a chronic disease have to deal with the world of work and the world of health care, i.e. with occupational professionals and HCPS, but also with company policies, legislation, regulations, social security, and macro-economic circumstances. The type and impact of the different stakeholders and environmental circumstances vary over time along with the progression of the adult onset NMD; from keeping the job to RTW after sick leave and finally to being deemed (partially) disabled for work<sup>[48]</sup>. Thus, when studying employment issues of persons with a chronic disease, a variety of perspectives needs to be considered. This dissertation focuses on how to keep a job. During the slow deteriorating process, when a person has vague complaints which impact daily life, and is not yet or newly diagnosed, the diagnosing physicians should be aware of unspoken work issues. The rehabilitation physician or occupational therapist might be the professionals in the hospital setting to identify labor problems early and to inform the person what to do in case of future hindrances impacting their daily life and work.

Our exploration of recommendations of HCPS in a hospital setting revealed an encouraging attention to work related issues (Chapter 4). Caution about the generalization of our findings is necessary. We studied recommendations related to adequate referral to HCPS in primary care and rehabilitation settings in only one hospital setting specialized in NMD. This might not be a realistic representation of the regular care provided with other client groups, in other hospitals or other settings<sup>[23, 49]</sup>.

As diagnosing physicians are sometimes unfamiliar with possible interventions or facilities, referrals not always take place. Neither general practitioners' nor physicians' first concern is their patients' work. It seems that the amount of support given to employees with NMD varies greatly depending on the setting where professionals work and on the individual professional. Either referral does not take place or does not address work issues. Another barrier affecting timely and proper interventions might be that communication between health care providers and the employer is prohibited without permission from the client. These barriers are not supportive in striving for sustainability of work for a person with NMD.

Rehabilitation physicians and occupational therapists in the hospital setting might be able to bridge the gap between health care and occupational settings when persons with NMD who have been recently diagnosed, but are not yet on sick leave, however, experience some hindrances in occupational performance. When the person diagnosed with NMD is still working, and the employer, HR manager or occupational physician is not yet aware of problems, support from these professionals in the hospital might be helpful in shared decision-making regarding measures to be taken for job retention<sup>[50]</sup>.

Cognitive decline and the lack of initiative, symptoms especially present in persons with MD, could also be a cause of suboptimal treatment regarding work issues. For

these persons offering a consultation to a rehabilitation physician or occupational therapist in association with the neurologist's consult might be a good solution.

At a later stage of NMD physical ailments can hinder job continuation, especially if these result in dangerous situations at the work place. At this stage, vocational rehabilitation programs might be helpful to sustain work for people with NMD <sup>[36]</sup>.

### **Experiences of persons with a neuromuscular disease at work**

The participants of the qualitative study revealed a narrative of their working life. Based upon their experiences several recommendations can be formulated. An overall complicated aspect was the response to early symptoms by participants themselves and the timing of this response. Not recognizing symptoms, ignorance of a probable inherited disease and postponing seeking a diagnosis might create delays. Hesitation to disclose problems as well as lack of understanding about consequences for the future further delayed timely communication. Client centred practice might lead to persons feeling very much heard and understood, however, that might not always lead to suitable interventions <sup>[51]</sup>. It is the professionals' ethical obligation to offer adequate and timely information about possible interventions to empower well informed shared decision making and not to wait for the proper questions to be asked by the client <sup>[52]</sup>.

Although measures taken can fail or be thwarted by reorganizations or a change of superior, that should not be a reason not to take precautions and ask for accommodations and support at work (Chapter 5).

### **Methodological considerations**

The combination of quantitative studies and qualitative studies combined the perspective of science, health professionals, and clients. In addition the qualitative studies provided a platform to draw from the experiences of workers with NMD. Triangulation is a method used by researchers to check and establish validity in their studies by analysing a research question from multiple perspectives <sup>[52]</sup>. Methodological triangulation involves the use of multiple qualitative and/or quantitative methods to study the subject. For example, our results from the systematic review, secondary analysis, methodological exploration of the actual state of health care and semi-structured interviews can be compared to determine if similar results are being found. If the conclusions from each of the methods are the same, then validity is established <sup>[52]</sup>. Although we aimed for triangulation, (methodological) saturation was not reached due to a lack of studies on the subject in general.

What we found was a shift from work related disease factors such as type of NMD, physical functions and activities in the quantitative studies to aspects related to work related environmental and work related personal factors in the qualitative studies.

Although several environmental and personal factors did not emerge in the results of the SR, because they were only present in one or two studies with low methodological quality, four work related environmental factors that corresponded to factors found in the qualitative studies were added (in italics) to the 2nd column of figure 1. The level of evidence might increase if the findings of our subsequent studies are taken into account in an updated version of the SR.

As the field of SRs reviews has been dominated by the development and application of meta-analyses of controlled trials to synthesize evidence about the effectiveness of health and social interventions, SRs traditionally include only quantitative studies.

As no RCTs or CCTs were found in our SR, we compromised by including other types of research provided that the studies were quantitative. By trying to fit the primary research into the traditional method of performing an SR, our findings resulted in a small quantity of studies and limited evidence for the factors found to be related to employment status of persons with NMD (Chapter 2). Over the past 10 years, other methods for reviewing have been developed<sup>[54, 55]</sup> advising that the review method chosen should reflect the approaches, assumptions and methodological challenges of the primary research<sup>[55]</sup>. To meet international standards for SR, our initial review excluded some possibly relevant research such as narrative reviews and other qualitative research such as case reports and clinical guidelines. If we had used a mixed-method SR and include qualitative studies, we might have found additional factors related to employment for people with NMD. However the level of evidence would not have increased with these qualitative findings as they are not generalizable.

The ICF scheme combines the health status (disease/disorder and problems in functioning) of an employee with NMD and contextual factors (environmental and personal factors). For the research question of our study, the ICF-scheme, when expanded with concepts of the Van Dijk model (including work-related environmental factors), seemed to give us a good grip on the categorization and positioning of factors<sup>[6]</sup>. But there is more to the subject than can be conveyed by merely summarizing all of the relevant factors. It would be interesting to know whether the factors we have found influence each other, and if so, what kind of relationship they have and how that relationship is to be interpreted<sup>[56]</sup>. For instance: what is the relationship between motivation to work and work as a burden, or postponing disclosure and contemplating job demands and other roles in life? What is the influence of access to assistive technology for communication on securing one's job? Do persisting problems of fatigue have an impact on one's occupational choice?

The advantage of performing a secondary analysis of data from previous studies not yet processed is that people with health conditions do not have to be bothered by unnecessary interviews or questionnaires. However by not being able to choose primary and secondary outcome measures, a secondary analysis has a higher risk of missing information related to the object of interest of the researcher. A targeted questionnaire provides more specific information on the subject of the research. For instance the definition of employment status in the study of Kalkman et al.<sup>[57]</sup> differed

from the definition set by the Dutch government, i.e. being employed 12 hours a week, versus eight hours<sup>[3]</sup>. This makes comparison with other studies of employment status and factors related to employment more difficult. The secondary analysis did not reveal factors such as adequacy of referral patterns, goal setting after referral, motivation, expectations, efforts to get a job, job retention, job content, working conditions, job type, number of hours worked, possibility of adjusting working hours, control options, social support at work, relationships at work, compensation options and barriers at work, but only provided employment status in relation to personal factors, fatigue, health status and two environmental factors.

With regard to the predictive value of fatigue it should be kept in mind that cut-scores used to identify the significance of fatigue impose an artificial dichotomy in what is an essentially continuous distribution of knowledge, skill or ability<sup>[58]</sup>. When using a cut-off point, one may lose important information<sup>[59]</sup>. For this reason fatigue should be treated as a continuous variable whenever possible.

The factors related to employment (Chapter 3) were derived from a cross-sectional study. To find a relation between higher activity levels and better health and thus higher employment rates or vice versa, a prospective longitudinal study is needed. In performing these prospective studies the Canadian Occupational Performance Measure (COPM) can be used to determine and prioritize the importance of work problems related to other activities in daily life in order to get an overview of personal and environmental barriers and facilitate clients' and occupational performance wishes<sup>[60]</sup>. The Worker Role Interview (WRI)<sup>[61]</sup>, the Work and Disability Questionnaire (WHQ)<sup>[62]</sup>, the Work Role Functioning Questionnaire (WRFQ)<sup>[63]</sup> or the Work Ability Index (WRA)<sup>[64]</sup> can be used to assess the consequences of NMD on job retention.

## Recommendations for practice

### *Recommendations for persons with NMD*

Easily accessible information needs to be developed and implemented for all stakeholders involved. First of all, clients need systems to support them in finding information about accessing support in job retention. Better understanding of the prognosis might contribute to the decision-making process, and to insight into the consequences of NMD for employment.

The website of the Dutch association for muscle diseases<sup>[64]</sup> could serve as an independent repository of information. For the members a guidebook on the website might provide information on which professionals could be consulted for what problem. Peer support also can be very effective in supporting job retention<sup>[65]</sup>.

For people with NMD it is important to fully consider the private and societal implications of their choices before communicating their challenges with relevant stakeholders in order to be supported in work retention. Employees with NMD are recommended to communicate problems at work with a general practitioner or

neurologist in a timely manner in order to initiate referral to professionals in health care or early vocational rehabilitation.

To sustain employability for persons with MD and those suffering from cognitive decline, it is necessary to strive for work environments that fit both their intellectual capacity as well as their physical load capacity. To compensate for the lack of initiative, which is a feature of MD, offering a consultation to a rehabilitation physician or occupational therapist in association with the neurologist's consult on a regular basis might be a good solution.

*Recommendations for physicians and HCPs working in health care settings*

In a hospital setting or outpatient unit a timely, appropriate diagnosis seems to be a key factor.

Implications for diagnosing neurologist and rehabilitation physicians in a hospital include awareness of the significant impact of a slow progressive physical deterioration on current and future employment and the importance of paid work for persons with a disability. The diagnosing neurologist needs to identify work problems as part of the medical assessment. It is recommended to use checklists for a first screening of people with a chronic disease to ensure identification of the employee at risk and their prompt referral to rehabilitation physicians or HCPs specialized in occupational issues, like the occupational therapist<sup>[66, 67]</sup>. Tailored and timely referral of recently diagnosed persons with NMD for early and thorough analysis of job concerns, providing information, formulating a plan that enables them to keep their job, and consultation with professionals who have employment-related expertise are found to be important as well. Occupational therapists could benefit from guidelines that not only focus on their professional skills<sup>[68]</sup>, but also on the organization of vocational rehabilitation and on the roles of each professional involved in employment retention.

When the client has not yet disclosed problems at work, the diagnosing neurologist, working in a hospital setting can start to discuss perspectives with the client, as they are able to consult the rehabilitation physician, the occupational therapist, psychologist and other members of the team, which is important especially at a stage of the disease when physical and cognitive deterioration are not yet related to a diagnosis. The rehabilitation physician and the occupational therapist are appropriate health professionals from the multidisciplinary team to enhance the cooperation between the world of health and the world of work. In collaboration with the team, recommendations can be formulated to outline future measures that have to be taken in order to enhance sustainability of work for the person with NMD at risk of future job loss. These might include for example, choices concerning appropriate accommodations, assistive products and adaptations at home, at work, and for commuting to and from work and if that does not cover the needs, referral to vocational rehabilitation.

It is recommended to be aware of the risk of severe fatigue in persons with NMD and to provide them with support in balancing home and work demands as well as

balancing demands within the workplace so that they maintain their competence to work. Occupational therapists coach persons struggling with balancing daily activities<sup>[68]</sup>.

Self-management programmes for employees with a chronic disease so as to empower persons at risk for work-disability to rebalance work demands and actively discuss their condition at work are also recommended<sup>[69-71]</sup>. The results of two randomized controlled trials indicated that participants were more empowered to cope at work and had fewer physical complaints after training related to their disease<sup>[69,71]</sup>. It is recommended to develop a self-management program for employees with a neurological disorder, to teach them to manage their participation in work. By informing persons with NMD how to deal with future challenges they are empowered not only to discuss employment problems but also to take appropriate measures.

*Recommendations to strengthen collaboration between professionals in health care and professionals working in or for occupational settings*

Although there are administrative and financial barriers, it is recommended to strengthen the collaboration between professionals working in health care and occupational settings in support of employment performance and full participation of clients with a disability. In order to enhance sustainability of work, communication between professionals in health care and the work sectors must be facilitated. For the benefit of the employee at risk of job loss it is recommended that professionals working in health care collaborate with stakeholders at the workplace taking into account each other's professional responsibility. Expertise may vary between occupational and health settings and even within the same profession working in different settings. As long as these professionals know and make use of each other's expertise, there should be no problem. Physicians as well as HCPS can develop specialisms within their field of practice beneficial to the client who then will receive the most appropriate cure and care.

The separation of the two worlds of work and health care has advantages, e.g. independent opinion and recommendations of occupational physicians about the impact of symptoms on the ability to work<sup>[41]</sup>. This responsibility has been laid down in one hand to avoid confusion for the person who is ill. When more professionals give recommendations, the person with a disability will likely listen to the advisor who gives the most appropriate answer in the view of that person<sup>[48]</sup>.

Increase of mutual understanding and knowledge between the world of health care and the world of work by joint continuing training is needed and recommended. Courses aimed at increasing mutual understanding about the impact of governmental legislation on job retention are widely available<sup>[43, 44]</sup>.

We embrace the initiative of the association of neuromuscular diseases that has chosen for the theme 'Work and NMD' at their annual conference this year<sup>[64]</sup>. These conferences have been a yearly event attended by neurologists, rehabilitation physicians, HCPS and persons with NMDs.

Fortunately attention for work is on the increase in programs offered by interdisciplinary teams in hospitals as well as VR that have been strongly developed in the Netherlands in the recent decade. It is recommended to inform general practitioners, health care physicians and occupational physicians about the advantages of referring their clients to these programs.

*Recommendations at the macro-economic level*

Many programs have been developed and are meant to empower disabled people to find work in the open competitive employment market and give disabled people choice and control over the support they receive<sup>[65]</sup>. These types of interventions vary largely within settings and among countries<sup>[39]</sup>.

Implementation of already available inter-professional guidelines to facilitate timely and adequate referral and support in job retention of people with a disability is recommended.

**Recommendations for future research**

*Replenish the systematic review*

New quantitative and qualitative studies now provide additional information about the focus of this study and would justify a fresh systematic review, and since qualitative studies revealed new factors to consider, preferably a mixed method systematic review should be performed<sup>[55]</sup>.

*Enhance triangulation by incorporating more stakeholders in employment related research*

This dissertation covered sustainability of work for persons with NMD from a variety of perspectives: a scientific perspective, the perspective of professionals in health care and the perspective of clients. The perspective of vocational rehabilitation programmes and social security initiatives related to the world of work were beyond the scope of this study and are future topics that have to be addressed.

It is important to support relatives who are confronted with the consequences of the slow deteriorating process on job retention. This was found in a recent study in which members of the Dutch association for neuromuscular diseases were asked to define important research subjects. They prioritized partner support in rehabilitation research<sup>[72]</sup>. Studies of the impact of interventions by occupational therapists providing just-in-time information for relatives revealed significantly better coping strategies for dealing with the progression of their partner's disease<sup>[73]</sup>.

*Enhance evidence for VR interventions*

Documents and positions statements, case studies, descriptive reports and book chapters on possible contributions of occupational therapy services in job retention

for people with a disability are wide spread<sup>[21, 23, 74, 75]</sup>. Interventions like jobsite analysis, during which the client is actually observed by an occupational therapist, while performing job related tasks is a process which can help determine barriers and problems in employment participation. This information can subsequently be utilized to formulate solutions to adapt, create and modify tasks, work stations and equipment, in order to maximize occupational performance and safety of disabled people<sup>[21, 76]</sup>. Evidence about the effect of these occupational therapy interventions is still rare<sup>[76]</sup>. Recommendations to enhance the value of occupational therapy in this area of practice have been formulated by several authors<sup>[75, 77]</sup>. Based on current powerful VR programmes, we recommend strengthening the value of interdisciplinary approaches in different client groups.

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## **SUMMARY**

In this dissertation the impact of NMD on sustainability of employment has been explored. To reveal factors that affect employment status, we focused on people with one of three relatively common types of NMD: 1) a muscle dystrophy: facioscapulo-humeral muscular dystrophy (FSHD); 2) a motor nerve root disorder and peripheral neuropathy that not only affects motor but also sensory nerves: hereditary muscular sensory neuropathy (HMSN); and 3) an adult onset multisystem disorder: myotonic dystrophy (MD). These types of NMD have in common that symptoms do not become manifest until adulthood, thus during the active working life. Given that the costs and cure of health care for people with a neuromuscular disease (NMD) are relatively high due to the early onset and progression, it is surprising that problems in the domain of employment have received little attention in the literature.

Reason to start this study was the clinical experience of occupational therapists working in a hospital that no or insufficient attention was paid to the consequences of and conditions for maintaining employment of people with NMD.

In *Chapter 1*, the general introduction, a case study illustrates employment problems and the necessary strategies for work retention in a person with NMD. Using the International Classification of Functioning, Disability and Health (ICF), the relationships between NMD related impairments, limitations in activities and participation restrictions are shown, taking into account the personal and environmental (work related) factors which influence daily functioning. At the micro level (individual level), the client perspective was shown, and at the meso level (organizational level), the health setting in which health care professionals work as well as the work environment of the client. At the macro level (societal level) legislative and funding barriers to achieve optimal health care and occupational functioning are identified. The introduction explains the importance of paid work for people with NMD, how this is part of their health and which problems they encounter if they want to continue to work. A recent international effort to define health has been formulated as follows: *'The ability to adapt and to self manage'*. The fundamental principle of occupational therapy, as formulated in 2007, is *'The importance of having an occupation for human health, well-being and justice'*. It is explained how the paradigm of occupational therapy fits with this important social domain of health. In conclusion, it means that the occupational therapist is well qualified to play a role in facilitating optimum employment for employees with NMD. The objective of this introduction is to enlarge awareness about and understanding of the right to work for persons with NMD.

*Chapter 2*. A systematic review (SR) was performed to find factors associated with employment of people with NMD – in this case FSHD, HMSN and MD. The results of the SR confirmed our assumption that research on employment problems of people with these three common types of NMD is scarce. No randomized clinical trials or controlled clinical trials were found. Therefore, all other design (OD) studies [cross-sectional, cohort, case-control studies, repeated survey design and longitudinal studies (prospective and retrospective studies)] were included and reviewed.

Due to a lack of high quality studies, the best evidence synthesis for quantitative research revealed no strong, moderate or even limited evidence for determinants in employment status. Ultimately eight studies were identified, revealing 10 indicative factors impacting employment. These factors were related to state of health (type of NMD, especially HMSN and the category 'other types of NMD'), factors related to functions (physical functions and muscle strength), general personal factors (age, gender and education), and work related personal factors (type of occupation, and expressed interest in employment). Although we found 84 other, probably relevant clinical factors, these could not be validated because they were only present in one or two cross-sectional studies. All 10 indicative factors which related to employment status were categorized using an adapted ICF-scheme.

In *Chapter 3* the results of a secondary analysis of data from a large cohort study on fatigue, 591 persons with the same three types of NMD, were presented. The analysis was conducted to investigate whether indicative factors found in the review study were confirmed and whether there were additional factors associated with employment status. Employed and not employed persons with NMD were compared on the following independent variables: personal factors, the Checklist individual Strength (CIS), and the Short Form Health Survey (SF-36) with employment as the dependent variable. There were significant differences between the employed and not employed group with NMD, all of which were found on both the CIS and the SF-36. Logistic regression analyses revealed that a maximum of 37.2% of the employment rate for the whole group was explained by six factors: younger age, being male, higher educational level, greater ability to concentrate, higher activity levels and better physical condition. In the different diagnostic groups factors that predict employment status varied: being younger, better concentration and less pain for the HMSN group (20.6%), being male, higher educated, higher activity levels and better physical functioning for the FSHD group (39.7%), and for the MD group younger age, being male, higher level of education and higher activity levels (42.4%).

Our hypothesis was that fatigue would affect employment status. All persons with NMD reported severe fatigue independent of employment status. The total group of employed persons with NMD and the two separate groups with FSHD and MD reported significantly less fatigue than the unemployed persons with NMD. However, the logistic regression analyses revealed that fatigue was not a predictive factor for employment. In total four factors were found comparable with those in the review study: health condition (type of NMD) and three personal factors (age, gender, and education) and two work-related personal factors (motivation and vitality) enhancing evidence for the relationship of these factors with employment status.

In the third study (*Chapter 4*), daily practice of health care professionals in the hospital setting was reviewed. The availability of 102 systematically written multidisciplinary reports of one-off consultation of clients with NMD in a hospital provided a unique opportunity to explore to what extent recommendations on employment were given directly by occupational therapists, physiotherapists and

speech therapists. Furthermore, we investigated how many clients these therapists subsequently refer to their colleagues in primary care, rehabilitation, or occupational health settings for follow up treatment of employment issues. Of the 102 files, 86 met the inclusion criteria (presence of report in the file, age group between 18 and 64 and slow progressive type of NMD). Eighty per cent of the reports contained information about work status. Occupational therapists wrote the general section of the reports as well as their own section and reported 80% of the work related issues as part of activities of daily life. In spite of the fact that 76% of the patients used assistive products at work or received immediate advice during the first consultation (97%), a large percentage (87%) still reported work-related problems for which 54% asked for support related to task content, terms of employment, and/or working conditions. Except for one person, all were subsequently referred for further investigation and intervention to solve their employment issues. Exploration of the reports also revealed significant differences in age, gender and educational level between employed and not employed people with NMD. We concluded that these findings supported the relevance of the factors for employment status of people with NMD found in the first two studies. Future research on the follow up of employment recommendations by colleagues in primary care, rehabilitation centers, and in occupational health settings is needed to explore the effectiveness of the recommended interventions.

In *Chapter 5* the client perspective, i.e. how people with NMD perceive employment participation, was examined to obtain a better understanding of client perspectives on employment *in the process of deteriorating ability*. Open in-depth interviews were held with 16 people with FSHD, HMSN and MD. This study indicated that people with a slow progressive adult type of NMD pinpoint paid employment as a very important aspect of life.

Four themes were identified in the analyses: the first theme (1) 'Experiences regarding the meaning of work', dealt with the way participants with NMD perceived their situation in the work environment. Most participants indicated that work was an important activity, irrespective of the phase of the disease or the severity of symptoms. Conditions under which work was meaningful and which aspects were meaningful varied. Participants who had no choice but to continue working with physical hindrances experienced employment as a burden. The other three themes were connected to the stage of deterioration: participants described how they changed strategies to maintain their position while adapting to their physical decline. Participants went from an internal focus to an external focus: (2) Solving problems oneself; (3) Reaching a turning point; and (4) Taking into account environmental aspects. Participants had a strongly developed self-management strategy of postponing disclosure of their disease or resulting problems on the job; they did not communicate hindrances until symptoms became apparent to their colleagues at work. Over-reliance on family might be the result of this strategy. *Only when physical hindrances obviously impacted employment performance, did participants negotiate with their superior to overcome or compensate for these issues in order to be able to stay at work.* With



increasing disability participants' focus shifted from the need for assistive products, *adapted pace and working hours* towards the importance of colleagues who took their condition into account. Support from superiors was valued most highly, because of their role as final decision makers in job retention.

In the general discussion, *Chapter 6*, an overview of the factors found in this dissertation are presented in the ICF scheme and discussed: factors related to health, the factors related to functions factors at the level of activity and participation as well as environmental and personal factors.

When work performance is at risk, persons with NMD have to deal with both the world of work as well as health care, i.e. with occupational professionals such as occupational physicians and human resource managers, with health care professionals, as well as company policies, legislation, regulations, insurance, and macro-economic circumstances. Thus, when studying employment issues of persons with a chronic disease, a variety of perspectives have to be taken into account. This dissertation approached employment issues from a health care perspective. The research questions were approached at three levels: scientific, health practice and the client viewpoint. The ICF scheme worked well to fill the gap between terminologies used by health care professionals, occupational professionals and employed persons with NMD.

#### *Recommendations for practice*

Persons with NMD need to be informed about their options in a timely manner to make shared treatment decisions concerning their job retention. Easily accessible information and websites might be helpful to them. Preferably the content of such a site and other sources of information need to be developed in cooperation with people with NMD who dealt with employment issues in their lives. For people with NMD it might be important to learn to communicate their job challenges with relevant stakeholders in order to be supported in work retention, if they wish to continue working. Recently, self-management programmes have been developed to empower persons at risk for work disability. Such programmes can be adapted for persons with NMD.

Guidelines for all stakeholders involved need to be checked on work-related issues and implemented. It is important to create awareness among diagnosing physicians and health care professionals about the impact of NMD on participation especially on work sustainability. Neurologist needs to identify work problems as part of the medical assessment, for which existing checklists can be used. Tailored and timely referral of person recently diagnosed with NMD to an occupational therapist in the hospital where the persons has been diagnosed early and thorough analysis of job concerns, provision of information, formulation of a plan to enable job retention and consultation with professionals who have employment-related expertise is recommended, in particular when the person involved has not yet revealed his problems at work. If the situation is more complex, referral to a rehabilitation physician with an interdisciplinary team is recommended. They can provide

information and, in concert with the person involved, formulate and implement plans to maintain the job. For the benefit of persons whose sustainability of employment is under pressure as a result of their progressive disease good cooperation between health care and occupational health care is required. Rehabilitation physicians and occupational therapists working in a hospital setting are well suited to bridge the gap between health care professionals and occupational professionals in supporting sustainability of employment of employees with a work disability, especially at a stage when problems are not yet visible or disclosed at work. Therefore their role in this should be clearly identified and expanded from current practices.

Several initiatives in the Netherlands and abroad support people with restrictions in maintaining their job is a socially important theme. One of these initiatives is the 'International Forum on Disability Management' (IFDM), a global network of researchers engaged in employment of disabled people, established in 1999. The IFDM focuses on the role of employers in job retention of employees. In the same year, the 'Early intervention vocational rehabilitation care' program was established in the Netherlands to assist people with an impending or existing work disability and their employers in job retention. The company can directly refer an employee with an impending labor problem to this vocational rehabilitation program. Nowadays, in the Netherlands 14 rehabilitation centers have affiliated labor rehabilitation centers where people can be referred to. Another more recent initiative, founded in 2011, is the so called '*Work Disability Participation and Integration*' (WDPI) meeting which, like the IFDM, intends to exchange knowledge among scientists with regard to the prevention of occupational disability from a health perspective. The exchange of scientific knowledge and expertise through these conferences and in-service training, as currently offered by organizations such as '*Gezond in Bedrijf*', is essential to improve existing boundaries between health and occupational health.

The initiative of the Dutch Association of Neuromuscular Diseases (VSN) to choose labor and NMA as a theme for the annual conference in 2013 for people with NMA, neurologists, rehabilitation doctors and allied health professionals, we warmly applaud.

#### *Future research*

There may be newer and more applicable ways to perform a systematic review related to this subject. It would be helpful to add to the SR the studies contained in this dissertation using a mixed method model and including qualitative studies.

A longitudinal study is recommended to determine causal relationships between the factors found in this dissertation and sustainability of work in employees with NMD.

Outcome studies are needed to prove the effectiveness of the since 2004 established ability of occupational physicians to directly refer to allied health professionals and interdisciplinary vocational rehabilitation is recommended. The effect of vocational rehabilitation programmes are scarce and mainly investigated

in large groups of people with low back pain. To determine the effectiveness of these programs for job retention for people with slowly progressive diseases such as NMD is also recommended.

Participation of patients in research is expected to show a shift of the research perspective of disease-related factors on the implications of chronic illness to everyday life, including the maintenance of a paid job.



**NEDERLANDSE SAMENVATTING**  
**(SUMMARY IN DUTCH)**

In deze dissertatie is onderzocht welke impact neuromusculaire aandoeningen (NMA) hebben op deelname aan het arbeidsproces. De factoren die van invloed zijn op het hebben en houden van een betaalde baan zijn onderzocht bij mensen met drie typen NMA die relatief veel voorkomen: 1) facioscapulo-humerale spierdystrofie (FSHD), een (erfelijke) spierziekte die in het begin vooral tot uiting komt in de spieren van het gezicht, het schouderblad en de bovenarmen 2) hereditaire motorische sensorische neuropathie (HMSN), een erfelijke aandoening van de zenuwen die signalen doorgeven van de (gevoels)zintuigen naar de hersenen en van de zenuwen die van de hersenen naar de spieren gaan en 3) myotone dystrofie (MD), een aandoening die tot veranderingen in meerdere organen leidt. MD wordt gekenmerkt door het vertraagd ontspannen van aangespannen spieren, en een langzaam toenemende spierzwakte. Ook organen kunnen klachten geven en er kan sprake zijn van futloosheid en een verhoogde behoefte aan slaap. De drie bestudeerde typen NMA hebben gemeen dat de symptomen zich veelal openbaren op volwassen leeftijd, dus tijdens het werkzame leven.

De kosten voor de gezondheidszorg en de maatschappelijk kosten van mensen met NMA zijn relatief hoog vanwege het vroege ontstaan voor of tijdens de levensfase waarin de arbeidsparticipatie start en de progressiviteit van deze aandoeningen. Het is opmerkelijk dat de arbeidsparticipatie van mensen met NMA tot op heden weinig aandacht heeft gekregen in wetenschappelijk onderzoek. De reden om dit onderzoek te starten was de klinische ervaring van ergotherapeuten werkzaam in een ziekenhuis, dat er geen of onvoldoende aandacht werd besteed aan de gevolgen van en voorwaarden voor het behoud van arbeidsparticipatie van mensen met NMA.

In de algemene inleiding (*hoofdstuk 1*) illustreert een casus de problemen die een persoon met NMA kan tegenkomen in de werksituatie en de strategieën die door deze persoon worden gebruikt om haar baan te behouden. Met behulp van een uitgebreide versie van het schema van de 'International Classification of Functioning, Disability and Health' (ICF), worden de relaties getoond tussen NMA aanverwante stoornissen, (beperkingen in) activiteiten, participatie(problemen) en de persoonlijke en omgevingsfactoren die het dagelijks functioneren beïnvloeden. Op microniveau (individueel niveau), wordt het klantperspectief getoond, en op mesoniveau (organisatieniveau) de werkomgeving van de cliënt en de gezondheidsetting waarin de paramedicus werkzaam is. Op macroniveau (maatschappelijk niveau) wordt uitgelegd welke barrières aanwezig zijn in de wetgeving en de invloed van de gescheiden financiële regelgeving tussen de gezondheidszorg en de bedrijfsgezondheidszorg.

In de inleiding wordt toegelicht wat voor personen met NMA het belang van betaald werk is, hoe dit een onderdeel vormt van gezondheid en met welke problemen zij te maken krijgen als zij aan het werk willen blijven. De meest recente internationale poging om gezondheid te definiëren, heeft geresulteerd in de volgende concept formulering: *'The ability to adapt and to self manage'*. De feitelijke grondslag van ergotherapie, geformuleerd in 2007, luidt: *'The importance of having an occupation for human health, well-being and justice'*. De wijze waarop het paradigma van

ergotherapie past bij het belangrijke maatschappelijke domein 'gezondheid' wordt in dit hoofdstuk toegelicht. Op basis van de uitgangspunten van de ergotherapie wordt geconcludeerd dat ergotherapeuten in staat zijn een rol te vervullen om de arbeidsparticipatie van mensen met NMA te vergemakkelijken.

In *hoofdstuk 2* staan de opzet en de resultaten van een systematische review (SR). De SR is uitgevoerd om factoren te vinden die samenhangen met de arbeidsparticipatie van mensen met FSHD, HMSN en MD. De resultaten van de SR bevestigden dat onderzoek naar problemen met arbeidsparticipatie bij mensen met deze drie veel voorkomende vormen van NMA schaars zijn. Er zijn geen gerandomiseerde klinische trials of gecontroleerde klinische studies gevonden. Daarom zijn alle andere typen kwantitatieve studies (cross-sectionele studies, cohortstudies, case-control studies, en longitudinale studies (prospectieve en retrospectieve studies)) opgenomen in de SR. Wegens het gebrek aan studies met een goed onderzoeksdesign, leverde de 'Best Evidence Synthesis' voor kwantitatief onderzoek in totaal tien factoren op uit acht studies, waarvoor indicaties zijn gevonden dat ze gerelateerd zijn aan arbeidsparticipatie van mensen met NMA. Het betrof factoren gerelateerd aan de gezondheidssituatie: (het type NMD, vooral HMSN en de categorie overige typen NMD, lichamelijke gesteldheid en spierkracht), algemene persoonlijke factoren (leeftijd, geslacht en opleiding) en werk gerelateerde persoonlijke factoren (de fysieke eisen die aan het werk worden gesteld en de getoonde interesse in werk). Naast de tien 'indicatieve' factoren werden ook 84 'waarschijnlijk klinisch relevante' factoren gevonden. Deze konden niet worden gevalideerd omdat ze slechts benoemd werden in één of twee cross-sectionele studies. Alle tien 'indicatieve' factoren in relatie tot de arbeidspositie zijn ingedeeld met behulp van een uitgebreide versie van het ICF-schema.

In *hoofdstuk 3* staan de resultaten van een secundaire analyse van gegevens van een grote cohortstudie naar vermoeidheid waaraan 591 personen met FSHD, HMSN en MD deelnamen. De analyse werd uitgevoerd om te onderzoeken of de indicatieve factoren, die gevonden waren in de SR, konden worden bevestigd en of er bijkomende factoren zouden worden gevonden die van invloed zijn op de arbeidsparticipatie van mensen met NMA. Werkende en niet-werkende mensen met NMA werden vergeleken op de volgende onafhankelijke variabelen: persoonlijke factoren, de Checklist Individuele Sterkte (CIS) en de Short Form Health Survey (SF-36). Werkgelegenheid was de afhankelijke variabele. Zowel de CIS en de SF-36, alsook de afzonderlijke sub-schalen van deze instrumenten verschilden significant tussen de werkende en niet werkende groepen met NMD. Uit regressie analyse bleek dat de arbeidsparticipatie voor het hele cohort NMA voor maximaal 37,2 % werd voorspeld door de volgende zes factoren: jongere leeftijd, behorend tot het mannelijke geslacht, hoger opleidingsniveau, betere concentratie, hoger activiteiten niveau en een betere lichamelijke conditie. Voorspellende factoren voor de arbeidspositie verschilden tussen de diagnose groepen. Van bovengenoemde factoren waren dat voor mensen met HMSN leeftijd, concentratie en pijn (20.6%) voor mensen met FSHD, geslacht,

opleidingsniveau, activiteitsniveau en fysiek functioneren (39.7%) en voor mensen met MD leeftijd, geslacht, opleidingsniveau en activiteitsniveau (42.4%).

Onze hypothese was dat ervaren vermoeidheid ook de arbeidsparticipatie zou beïnvloeden. Bij alle diagnosegroepen was er inderdaad sprake van ernstige vermoeidheid, maar dat had geen verband met het al dan niet hebben van een betaalde baan. Wel rapporteerde het gehele cohort werkende mensen met NMA en de twee afzonderlijke groepen met FSHD en MD significant minder vermoeidheid dan de niet werkende mensen met NMA. Uit de logistische regressie analyse bleek echter dat vermoeidheid geen voorspellende waarde had voor het al dan niet participeren in arbeid.

De uitkomsten uit deze secundaire analyse verhogen het niveau van bewijs voor de relatie met arbeidsparticipatie van mensen met NMD van vier factoren die ook in de SR gevonden zijn (de gezondheidssituatie (type NMD) en drie persoonlijke factoren (leeftijd, geslacht en opleidingsniveau).

In de derde studie (*hoofdstuk 4*), is onderzocht in hoeverre er aandacht wordt gegeven aan arbeidsparticipatie van mensen met NMA in de dagelijkse paramedische praktijk op de polikliniek van een universitair ziekenhuis. De beschikbaarheid van 102 systematisch opgebouwde multidisciplinaire patiëntdossiers van cliënten met NMA bood een unieke gelegenheid om te onderzoeken in hoeverre er na een eenmalig consult in het ziekenhuis door ergotherapeuten, fysiotherapeuten en logopedisten aan collega's in de eerstelijns gezondheidszorg en de revalidatie of aan de bedrijfsgezondheidszorg aanbevelingen worden gedaan voor een follow-up behandeling bij arbeidsproblemen van mensen met een NMA. Van de 102 dossiers voldeden 86 aan de inclusiecriteria (aanwezigheid van het volledige verslag in het dossier, leeftijd tussen 18 en 64 en een langzaam progressieve vorm van NMA). Tachtig procent van de 86 dossiers bevatte informatie over de arbeidsstatus. De ergotherapeuten schreven het inleidende deel van de verslagen en het ergotherapeutisch verslag. De fysiotherapeut en logopedist rapporteerden ieder hun eigen professioneel advies. De ergotherapeuten rapporteerden daarmee 80% van de werk gerelateerde zaken, als onderdeel van de activiteiten van het dagelijks leven. Tijdens het eerste consult bleek dat 76% van de cliënten met een betaalde baan gebruik maakte van aanpassingen en/of voorzieningen. Tijdens datzelfde consult kreeg 79% adviezen voor de arbeidssituatie, waarvan 87% aangaf werk gerelateerde problemen te ervaren. Een deel van deze 87%, namelijk 54% vroeg direct aanvullend advies voor ondersteuning bij de taakhoud, arbeidsvoorwaarden, alsook de arbeidsomstandigheden. Op één persoon na werden deze mensen doorverwezen voor verder onderzoek en behandeling om hun arbeidsproblemen op te lossen.

Uit onderzoek van de verslagen bleek dat mensen die betaald werk verrichten significant verschilden met niet werkenden. Dat wil zeggen dat zij jonger waren, vaker van het mannelijk geslacht en hoger opgeleid. Hieruit kan de conclusie worden getrokken dat het niveau van bewijskracht van de factoren leeftijd, geslacht en opleidingsniveau voor de arbeidspositie van mensen met NMA, zoals gevonden in de eerste twee studies opnieuw wordt verhoogd.



Toekomstig onderzoek moet uitwijzen in hoeverre de aanbevelingen voor het behoud van de werkgelegenheid wordt opgevolgd door mensen met NMA, en door collega's in de eerstelijns gezondheidszorg, revalidatiecentra en bedrijfsgezondheidszorg waar de cliënten naar zijn verwezen. Dit om de effectiviteit van een dergelijke benadering te toetsen.

Om een beter begrip van het cliëntperspectief op arbeidsparticipatie te krijgen werd in de vierde studie (*hoofdstuk 5*), het cliëntperspectief onderzocht. Hoe ervaren en waarderen mensen met een op volwassen leeftijd openbarende langzaam progressief type NMA zelf hun arbeidsparticipatie gedurende het proces van verslechtering van lichamelijke mogelijkheden. Er zijn diepte interviews afgenomen met open vragen bij 16 mensen met FSHD, HMSN en MD met een betaalde baan. De deelnemers gaven aan arbeidsparticipatie als een zeer belangrijk aspect van het leven te ervaren. Er werden vier thema's geïdentificeerd in de analyses: het eerste thema (1) 'Ervaringen met betrekking tot de betekenis van het werk', ging over de manier waarop deelnemers met NMA hun situatie in de werkomgeving zien. De meeste deelnemers gaven aan dat werk een belangrijke activiteit voor hen was, ongeacht de fase van de ziekte of de ernst van de symptomen. Enkele deelnemers gaven aan dat arbeid van belang was omdat zij zich verantwoordelijk voelden voor het gezinsinkomen. De omstandigheden waaronder het werk zinvol was en in welke mate aspecten betekenisvol waren varieerden.

Deelnemers die bijvoorbeeld om economische redenen geen andere keuze hadden dan te blijven werken met fysieke belemmeringen ervoeren het werk meer als een last.

De andere drie thema's zijn te plaatsen in een tijdslijn waarmee beschreven wordt hoe tijdens het proces van fysieke verslechtering mensen omgaan met toenemende problemen op het werk. Zij veranderden hun strategieën om de arbeidspositie te behouden waarbij zij zich aanpasten aan hun lichamelijke achteruitgang. De deelnemers gingen van een intern focus naar een externe focus: (2) het zelf oplossen van problemen, (3) het bereiken van een keerpunt, en (4) het rekening houden met omgevingsfactoren. De deelnemers hadden een sterk ontwikkelde zelfmanagement strategie gericht op het behoud van hun baan door als eerste zo lang mogelijk het moment uit te stellen waarop zij op het werk bekend maakten dat zij NMA hadden en met welke problemen dat gepaard ging. Pas als de symptomen niet meer te verbergen waren voor collega's op het werk werd openheid gegeven over de ziekte. Pas als fysieke problemen obstakels werden en zij de arbeidsparticipatie onmiskenbaar beïnvloedden, onderhandelden de deelnemers met hun leidinggevende om deze problemen te overwinnen of te compenseren om op die manier toch aan het werk te kunnen blijven. Naarmate de functioneringsproblemen verder toenamen, verschoof de focus van de deelnemers van de behoefte aan aanpassingen en voorzieningen, zoals een aangepast werktempo en aangepaste werktijden, naar het belang van collega's die rekening hielden met hun situatie. Steun van leidinggevendenden werd het meest belangrijk gevonden vanwege hun rol als uiteindelijke beslissers bij het behoud van de baan. De algemene discussie in *hoofdstuk 6* betreft een overzicht van de factoren die samenhangen met arbeidsparticipatie zoals deze zijn gevonden in de studies. In het

ICF-schema zijn de gezondheidsfactoren, de functie gerelateerde factoren, factoren op activiteiten en participatieniveau als ook omgevings- en persoonlijke factoren uit de verschillende studies van deze dissertatie geordend.

Wanneer het vermogen om te werken onder druk staat door de aandoening, hebben personen met een NMA te maken met de gezondheidszorg en de bedrijfsgezondheidszorg, maar ook met het bedrijfsbeleid, wet- en regelgeving, verzekeringen en macro-economische omstandigheden. Bij het bestuderen van vraagstukken over baanbehoud dient er met deze verscheidenheid aan perspectieven rekening te worden gehouden. Deze dissertatie beperkt zich tot arbeidsparticipatie van mensen met NMA vanuit gezondheidszorgperspectief waarbij zowel het wetenschappelijke, het professionele als ook het klantperspectief in ogenschouw is genomen.

#### *Aanbevelingen voor de praktijk*

Op basis van deze dissertatie zijn aanbevelingen geformuleerd voor mensen met een NMA en voor neurologen en paramedici werkzaam in de gezondheidszorg.

Personen met NMA moeten tijdig en goed geïnformeerd zijn om in samenspraak met deskundigen beslissingen te nemen ten gunste van hun baanbehoud. Het is voor hen belangrijk om problemen bij de beroepsuitoefening te bespreken met relevante belanghebbenden om er voor te zorgen dat ze worden ondersteund bij het behoud van werk, als dit belangrijk is voor hen. Gemakkelijk toegankelijke en laagdrempelige informatie in folders of op de website van <http://www.spierziekten.nl> zijn daarbij belangrijk om hen te informeren over en te ondersteunen bij procedures, wetgeving, rechten en plichten. De inhoud van informatiefolders en website dient bij voorkeur ontwikkeld te worden in samenspraak met mensen met NMA. Er zijn recent ook zelfmanagement programma's ontwikkeld om mensen die gevaar lopen arbeidsgehandicapt te worden, te ondersteunen. Deze zouden in een aangepaste vorm ook kunnen worden aangeboden aan werknemers met NMA.

Het is eveneens van belang om bewustwording te creëren bij artsen en paramedici om hen inzicht te geven in de impact die NMA heeft op het behoud van werk. De neuroloog dient problemen ten aanzien van werk mee te nemen als onderdeel van de probleeminventarisatie tijdens de diagnostische fase van het medisch onderzoek, zeker als de persoon met NMA zijn problemen op het werk nog niet kenbaar heeft gemaakt. De neuroloog kan voor de eerste check bestaande topiclijsten gebruiken. Voor vroegtijdige en grondige analyse van de werkproblemen is op maat gesneden en tijdige doorverwijzing van recent gediagnosticeerde mensen met NMA naar een ergotherapeut of indien de situatie complex is, een revalidatiearts met een interdisciplinair revalidatieteam van groot belang. Zij kunnen informatie verstrekken en samen met de persoon plannen formuleren en uitvoeren om het werk te behouden. Ten behoeve van de personen wier arbeidsparticipatie ten gevolge van hun progressieve ziekte onder druk staat is goede samenwerking nodig tussen de gezondheidszorg en de bedrijfsgezondheidszorg. Goed overleg tussen

zorgprofessionals en professionals werkzaam bij een bedrijfsgezondheidsdienst is daarbij essentieel. Revalidatieartsen en ergotherapeuten die werkzaam zijn in het ziekenhuis hebben de noodzakelijke kennis en kunde om de kloof tussen professionals in de gezondheidszorg en bedrijfsgezondheidszorg te overbruggen in de ondersteuning van de duurzaamheid van het werk van werknemers met een arbeidshandicap, in het bijzonder in een stadium waarin de arbeidsproblemen nog niet zichtbaar zijn of bekend zijn gemaakt op het werk.

Diverse initiatieven in Nederland en in het buitenland onderschrijven dat het ondersteunen van mensen met een arbeidshandicap bij baanbehoud een maatschappelijk belangrijk thema is. Een van deze initiatieven is het in 1999 opgerichte '*International Forum on Disability Management*' (IDFM), een wereldwijd netwerk van onderzoekers die zich bezig houden met arbeidsparticipatie van arbeidsgehandicapten. Het IDFM richt zich op de rol van werkgevers bij baanbehoud van werknemers. In datzelfde jaar is in Nederland het '*Vroege interventie arbeidsrevalidatiezorg*' programma opgericht om mensen met een dreigende of bestaande arbeidshandicap en hun werkgevers te begeleiden bij baanbehoud. De bedrijfsarts kan een medewerker met een dreigend arbeidsprobleem direct verwijzen naar de arbeidsrevalidatie. Er bestaan inmiddels in Nederland 14 aan revalidatiecentra gelieerde arbeidsrevalidatiecentra waar mensen met beginnende arbeidsproblemen naar kunnen worden verwezen om hen te ondersteunen bij behoud van hun werk.

Een ander meer recent initiatief is het in 2011 opgerichte '*Work Disability Participation and Integration*' (WDPI) congres dat eveneens de intentie heeft om wetenschappelijke kennis uit te wisselen met betrekking tot het voorkomen van arbeidshandicaps vanuit gezondheidszorgperspectief. Wederzijdse uitwisseling en deskundigheidsbevordering door middel van deze congressen en bij- en nascholing, zoals dat nu wordt aangeboden door organisaties zoals '*Gezond in Bedrijf*', is essentieel om bestaande grenzen tussen gezondheidszorg en de bedrijfsgezondheidszorg te verbeteren.

Het initiatief van de Vereniging Spierziekten Nederland (VSN) om voor het jaarcongres 2013 voor mensen met NMA, neurologen, paramedici en revalidatieartsen het thema arbeid en NMA te kiezen, juichen wij van harte toe.

#### *Toekomstig onderzoek*

Recente publicaties geven aan dat er nieuwere en meer toegesneden manieren zijn om een systematische review uit te voeren, bijvoorbeeld een gemengde onderzoeksmethode (a mixed method review), waarbij zowel kwantitatieve als kwalitatieve studies worden onderzocht. Het zou bijvoorbeeld zinnig kunnen zijn om de in dit proefschrift uitgevoerde SR aan te vullen, niet alleen met recenter kwantitatief onderzoek maar ook met kwalitatieve studies.

Om vast te stellen of er een causale relatie is tussen de in dit proefschrift gevonden factoren en arbeidsparticipatie wordt het uitvoeren van een longitudinale studie aanbevolen.

In richtlijnen, statements, artikelen en handboeken staat dat paramedische zorg en arbeidsrevalidatie effectiever zijn geworden door de directe verwijstructuur van bedrijfsartsen naar paramedische en interdisciplinaire arbeidsrevalidatieprogramma's, zoals dat sinds 2004 bij de Nederlandse wet is geregeld. De bewijskracht voor de effectiviteit van deze interventies is echter laag en vooral onderzocht bij grote groepen mensen met lage rugklachten. Er zijn ook studies nodig om de effectiviteit van programma's voor baanbehoud aan te tonen bij mensen met een langzaam progressieve aandoening zoals NMA. Participatie van patiënten in onderzoek zal naar verwachting een verschuiving laten zien van het onderzoeksperspectief van ziekte gerelateerde factoren naar de consequenties die chronische aandoening hebben op het dagelijks bestaan, waaronder het behoud van een betaalde baan.

# **DANKWOORD (ACKNOWLEDGEMENT)**

Velen hebben een belangrijke bijdrage geleverd aan dit proefschrift.

Allereerst dank ik met veel respect de 16 mensen met een neuromusculaire aandoening voor hun openhartigheid om met mij de situaties te delen waarin zij moeten dealen met toenemende beperkingen in de wereld die werken heet. Jullie verhalen hebben mijn klinische ervaring bevestigd dat mensen met een ‘echte’ beperking graag willen werken i.t.t. wat de politiek ons soms wil doen geloven. Ik ben de Vereniging Spierziekten Nederland dank verschuldigd voor de hulp bij het werven van deelnemers aan het kwalitatieve onderzoek door het plaatsen van een advertentie op de website.

Dit proefschrift zou er nooit zijn geweest zonder mijn promotoren prof. dr. Ria Nijhuis-van der Sanden, prof. dr. Baziël van Engelen, copromotoren dr. Josephine Engels, dr. Yvonne Heerkens en voormalig promotor prof. dr. Rob Oostendorp.

Beste Rob, centraal voor mij in je begeleiding staat wel het woord geduld. Je hebt mij met zorg en respect voor mijn klinische achtergrond, die maar al te vaak het wetenschappelijk denken overruledde, gecoacht in het wetenschappelijk denken. Je bent heel betrouwbaar en zorgvuldig in het geven van een reactie op mails. Je feedback was altijd opbouwend en bemoedigend. Knap hoor want je moet wel eens momenten van wanhoop hebben gehad bij het lezen van mijn conceptteksten. Je gastvrijheid thuis op de vertrouwde maandagmorgen met een koekje bij de koffie van Ank heb ik zeer gewaardeerd. Jammer dat we het karwei niet samen hebben kunnen afmaken. Je hebt het stokje met enige weemoed overgedragen aan je opvolger Ria. Daarmee draagt mijn proefschrift gelukkig wel bij aan de output van de door jou geïnitieerde leerstoel paramedische wetenschappen.

Beste Ria, we hebben een langjarig gezamenlijk professioneel verleden waarbij wij als collega's in het Radboudumc onze afdelingen ergotherapie en kinderfysiotherapie goed op de kaart hebben gezet. We deelden met de andere leidinggevenden gebundeld in Staf Zorg de ambitie om de paramedische disciplines en zorg disciplines te academiseren. Wie had toen gedacht dat we elkaar op deze wijze weer tegen zouden komen. Je volgde Rob voortvarend op en hoewel ik opzag tegen de verandering van begeleiding is dat heel goed gegaan. Je gaf mij mijn vertrouwen terug na een dieptepunt waardoor ik nu toch de mijlpaal heb bereikt waar ik zo hard aan heb gewerkt. Dank voor dat vertrouwen.

Beste Baziël, je weet dat twee opmerkingen in mijn geheugen gegrift blijven. Jouw constatering dat het zo moeilijk is om gevestigde HBO-ers met veel praktijkervaring, te begeleiden in de wetenschap. Je golden question: ‘So what?’ zette mij steeds aan het denken. Dan moest ik de transfer maken van mijn vooringenomen klinische bevindingen en opinie naar een wetenschappelijke redenering. Je merkte dat het raakvlak met de klinische praktijk bij het kwalitatieve onderzoek voor mij een fijne afsluiting was van mijn proeve van bekwaamheid.

Beste Josephine, jij was degene die mij in 2003 de vraag stelde of ik de ambitie had te promoveren, waar ik volmondig ja tegen heb gezegd. Ik had toen geen idee wat dat voor de kwaliteit van mijn leven zou gaan betekenen. Je gedrevenheid werkte

aanstekelijk. Het was voor jou niet gemakkelijk om in te schatten hoe je zo'n ervaren zelfstandig opererende vrouw als ik ben zou moeten begeleiden. Er bleek veel geduld nodig te zijn om iemand met zoveel klinische ervaring, totaal andere competenties bij te brengen.

Beste Yvonne, je bent de vrouw van classificaties, systematiek en schema's. Je hebt mij goed geholpen zaken in kaders te plaatsen. Ook ben je expert in het i-dotten en t-crossen. Dat moedigde mij aan om ook de hoogste mate van zorgvuldigheid te betrachten.

De aandacht en feedback van jullie, copromotoren heb ik zeer gewaardeerd. Ook de ruimte die mij gegund werd om andere begeleiders te kiezen voor specifieke onderwerpen.

Het onderzoek is financieel mogelijk gemaakt met subsidie van het 'HBO Mobiliteitsfonds' en de Hogeschool van Arnhem en Nijmegen (HAN University of Applied Sciences) te Nijmegen. Dat ik gebruik kon maken van deze subsidies dank ik aan de directie van de Faculteit Gezondheid, Gedrag en Maatschappij, in de personen van Bart van Bergen, Walter van Gisbergen en hun rechtsopvolger Frank Stöteler en de directie van het Instituut Paramedische studies in het bijzonder Theo Joosten en Menno Pistorius en hun voorgangers. Dank ook voor de morele en aanvullende financiële ondersteuning en het vertrouwen dat jullie in mij hebben gesteld.

Ook mijn opvolgers, opleidingsmanagers en collega docenten ergotherapie die de opleiding ergotherapie aan de HAN in Nijmegen hebben helpen bouwen, inrichten en bestendigen zeg ik dank. Zij zijn mij ook in moeilijke tijden privé tot steun geweest. Enkele namen wil ik op deze plaats noemen. Erik Entken, Ank Eijkelkamp, Bernadette ter Heine, Marleen Kaijen, Henk van Loon, Ineke Stijnen, Merel van Uden, Alex de Veld, Marlies Verkade en Anke Visser. Op de HAN dank ik ook dr. Wietske Kuijer-Siebelink, die mij leerde promotiesuccessen te vieren.

Leden van de kenniskring Arbeid & Gezondheid in wisselende samenstelling. Ik heb geprobeerd om met intervisie tot uitwisseling te komen en onze leermomenten te delen. Ik hoop dat ik de HAN verder kan helpen het wetenschappelijk klimaat verder uit te bouwen voor toekomstige onderzoekers ten behoeve van onze studenten.

Professor dr. Gijs Bleijenberg en dr. Joke Kalkman veel dank voor het beschikbaar stellen van de grote dataset, die de basis is geweest voor het tweede artikel. Van jullie zeer kritische begeleiding heb ik veel geleerd. Vooral het zeer kort en krachtig uitdrukken van de resultaten was een leerzaam proces.

Lorian Kennedy my best friend and colleague, master in occupational therapy, thank you for your valuable suggestions concerning the English texts of all articles and the final manuscript. Dr. Peter Huijbregts, helaas plotseling overleden dank voor de correcties van artikel 2.

Ton Satink mijn grote inspirator t.a.v. de waarde van kwalitatief onderzoek voor ons boeiende vak ergotherapie. Wij hebben heel wat rolwisselingen meegemaakt. Sinds je aanstelling bij de HAN was ik eerst je leidinggevende. Ik moedigde je aan om ons te helpen de opleiding ergotherapie uit te bouwen. Op het moment dat de tijd

daar rijp voor was, is het je gelukt je eigen promotietraject uit te zetten. Veel succes met het vervolg van je innovatieve en interessante onderzoek over zelfmanagement van mensen die een CVA hebben doorgemaakt. In je rol van begeleider van de kwalitatieve studie was je als deskundige mijn meerdere, maar we wisten de rollen goed te scheiden en het heeft de samenwerking op andere fronten versterkt. Je bent een kanjer op het gebied van kwalitatief onderzoek en je feedback was altijd opbouwend en verrijkend. Ik zal je waar je dat nodig acht ook bijstaan in de totstandkoming van jouw proefschrift.

Astrid Kinébanian, ik ben er trots op dat ik jou in mijn dankwoord kan opnemen. Zo graag wilde ik van je wijsheid leren en wat vond ik het fijn een ergotherapie collega te hebben die ook het vakjargon en de passie voor het vak met mij deelt. Van jou had ik vaak enkele kritische tips nodig om verder te kunnen. Een super docent! Ik zal de opgedane kennis verder uitbreiden en doorgeven aan de volgende generaties ergotherapeuten.

Reinier Akkermans en Hans Elvers dank voor jullie lessen statistiek, en dank aan Angelina van Zadelhoff voor de ondersteuning bij het plannen van afspraken met mijn begeleidingscommissie.

Mijn zus Sieta Koet-Minis en nicht Eveline Stoffer dank ik voor het uittypen van de interviews.

Ergotherapie collega's uit de regio, jullie hebben mijn ontwikkeling van dichtbij meegemaakt en zijn vol vuur meegegroeid in mijn ambities en de academisering van het mooie vak ergotherapie. Omdat ik als Mater Familias het risico loop mensen te vergeten wil ik op deze plaats slechts enkele collega's met namen noemen. Nelleke Nederkoorn, Madeleine Corstens, Margot van Melick en Margina Draaisma met wie we de afdeling ergotherapie van het UMCN een eerste gezicht hebben gegeven. Mabel van Esch en Marjolein Thijssen die een uitdaging zijn aangegaan mee te gaan naar de HAN en het onderwijs.

Dr. Edith Cup, Margo van Hartingsveldt-Bakker, Lucelle van de Ven-Stevens, Ria van Gelder † en dr. Pauline Aarts, de laatste twee van de St. Maartenskliniek. Allen zijn we de uitdaging aangegaan een wetenschappelijke studie te gaan volgen en te (gaan) promoveren.

Dank aan dr. Maud Graff die ik in de negentiger jaren heb aangezocht en heb gestimuleerd om in het Radboudumc te komen werken en mee te helpen om de academisering van het vak ergotherapie vorm te geven. Dat heb je subliem gedaan met een cum laude afgeronde promotie naar de effectiviteit van ergotherapie voor mensen met beginnende dementie en hun partners. We hebben veel bereikt, en van alle publicaties en andere output op congressen in boekvorm blijf ik genieten als mooie oogst van onze gezamenlijke inspanningen.

Het Netwerk Ergotherapie Onderzoekers Nijmegen (NEON) dank voor de interessante uitwisseling op wetenschapsgebied. Het NEON is een mooie opstap naar een lectoraat of misschien ooit een leerstoel ergotherapie? De tijd zal het leren. Aan ons, de kenniskring ergotherapie, zal het niet liggen. Dank aan dr. Esther Steultjens, o.m. lid van het NEON die in de laatste periode mijn mentor is geweest.



Fons Wierink who contacted me with my colleagues at the University of Alberta, Edmonton, who became role models in my professional life. From the first meeting in Edmonton, I realized the importance of research in occupational therapy. Thanks colleagues of the University of Alberta for giving me an award for best foreign student. You encouraged me to continue my PhD after graduation, prof. dr. Helen Madill my student coach and second author of my first scientific article, prof. dr. Jean Wessels, my thesis coach, prof. dr. Lili Liu, inspiring teacher, with whom I published my second scientific article and last but not least prof. Sharon Brintnell international contact person of the University of Alberta.

Wat we als promotieteam met elkaar hebben gedeeld zijn vele ingrijpende ‘life events’ die in meer of mindere mate invloed hebben gehad op de voortgang van mijn promotietraject. Toch hebben we steeds de draad weer opgepakt. Voor mij was dat het wegvallen van mijn man en grote supporter Harry van Kuyk. Ik had Harry graag de omslag van het proefschrift laten ontwerpen en heb dit symbolisch gedaan door een ontwerp dat hij heeft gemaakt voor het Nederlands Gezelschap voor Handtherapie, waar ik de auteursrechten van heb geërfd, te gebruiken op de voorzijde van mijn dissertatie. De handen krijgen voor deze gelegenheid een andere betekenis. De twee handen symboliseren de samenwerking tussen mensen die ondersteuning nodig hebben bij het behoud van een baan enerzijds en de gezondheidszorg professionals, bedrijfsgezondheidszorg professionals, wetgevers en werkgevers anderzijds die er alleen in goede samenwerking voor kunnen zorgen dat mensen met een handicap hun baan kunnen behouden.

Met Harry aan mijn zijde met zijn bijzonder ontwapenende, controversiële leefstijl leefde ik een totaal andere wereld naast het werk. Dit zorgde voor balans in mijn leven. Ik ben blij dat ik na dit proefschrift weer tijd heb om hem verder te vereeuwigen in een boek en website over zijn leven en werk. Dit project geef ik samen gestalte met dr. Joost de Wal, kunsthistoricus en zijn broer Frederik de Wal, typograaf, e.e.a. met ondersteuning van het bestuur van de Harry van Kuyk Stichting. Frits Deys, bestuurder en typograaf bedankt voor de prachtige website <http://www.harryvankuyk.nl>, waarop iedereen het vervolg van de ontwikkelingen van het boek kan volgen.

Lieve vrienden en familie, allemaal bedankt voor de belangstelling en steun, in het bijzonder mijn moeder, mijn beste vriendin, met wie ik alle ‘life events’ en ‘hobby’s’ kan delen. Jullie, mijn ouders, hebben indertijd een veer moeten laten om mijn dure particuliere opleiding ergotherapie te betalen van een toen nog niet erkend beroep. Daarmee zijn jullie in positieve zin medeverantwoordelijk voor de ontwikkeling van het beroep ergotherapie in Nederland. Papa heeft zijn avontuurlijke dochter altijd intensief gevolgd en zou niet verbaasd zijn geweest, maar ook trots, dat ik deze uitdaging, het bewijzen van een proeve van wetenschappelijke bekwaamheid ben aangegaan in mijn carrière.

Gelukkig heeft mijn nieuwe levenspartner Ruud Bronsveld mij de zin in het leven en in mijn werk weer terug gegeven. Jij vindt mij een bijzondere vrouw. Je mooiste

statement is: 'Alles komt goed'. Je hebt mij gesteund als ik zat te werken in vakanties, weekenden en vrije avonden. Je hielp mij de Engelse teksten te verbeteren, zorgde voor het huishouden en voor afleiding. Na deze tour de force is mijn vaste voornemen vrije tijd ook echt als vrije tijd in te gaan vullen en met jou nog vele leuke dingen te ondernemen.

Marie-Antoinette

# **CURRICULUM VITAE**

Marie-Antoinette Hubertine Minis werd geboren op 27 april 1955 in Nijmegen. In 1972 haalde zij haar MMS diploma aan het Geert Groote College (nu: Etty Hillesum Lyceum) te Deventer en zij studeerde een jaar aan de opleiding tuinbouw Rollecate N21, (nu: 'Nieuw Rollecate') te Deventer. Aansluitend ging zij van 1973-1976 studeren aan de Revalidatie Academie Hoensbroeck (nu: Zuyd Hogeschool te Heerlen), opleiding Ergotherapie.

Zij werkte dertien jaar als ergotherapeut in de St Maartenskliniek, een ziekenhuis gespecialiseerd in Reumatologie, Orthopedie en Revalidatie. In 1980-1981 ging zij ruim een half jaar voor een expeditie naar Afrika samen met haar echtgenoot grafisch kunstenaar Harry van Kuyk, die in 2008 is overleden.

In de St Maartenskliniek specialiseerde zij zich in reuma hand revalidatie en ze verzorgde jarenlang post HBO cursussen op het gebied van handrevalidatie en het vervaardigen van handorthesen in Amsterdam en in 1983 initieerde zij de landelijke werkgroep Ergotherapie en Reumatologie (WER) (nu onderdeel van de Nederlandse Health Professionals in de Reumatologie [NHPR], divisie van de Nederlandse Vereniging voor Reumatologie). In 1990 was zij medeoprichter en eerste bestuurder van het Nederlands Gezelschap voor Handtherapie (NGHT) voor welk gezelschap zij tien jaar lang de eindredactie van het Nederlands Tijdschrift voor Handtherapie verzorgde. Sinds 2002 is zij erelid van het Gezelschap.

In 1990 maakte ze de overstap naar het St Radboud ziekenhuis (nu Radboud universitair medisch centrum) waar zij hoofd werd van de afdeling Ergotherapie. In 1992 slaagde zij voor de Hoger Management opleiding voor non-profit organisaties aan het VDO van de Hogeschool van Nijmegen (nu: Hogeschool van Arnhem en Nijmegen [HAN]). Zij heeft aan de wieg gestaan van de academisering van de paramedische beroepen.

In 1998 rondde zij haar Master of Science Degree in Occupational Therapy aan de University van Alberta, Edmonton, Canada af. Haar eindschrift ging over de responsiviteit van de Sequential Occupational Dexterity Assessment (SODA).

In opdracht van de directie van de Faculteit Gezondheid, Gedrag en Maatschappij (GGM) van de HAN heeft zij in 1993 en 1998 een adviesaanvraag geschreven voor een Opleiding Ergotherapie in Nijmegen. De toekenning vanuit het Ministerie van Onderwijs en Wetenschappen volgde in 2000. In 2002 startte de Bachelor Opleiding Ergotherapie waaraan zij van 2002-2008 leiding gaf. Sinds 2003 is zij lid van de Kenniskring van het lectoraat Arbeid & Gezondheid van de HAN. In 2005 behaalde zij het diploma didactiek in het hoger onderwijs.

Thans is zij hoofddocent bij het Instituut Paramedische Studies van de Faculteit GGM en HAN Master Programma's (HMP), opleiding Neurorevalidatie. Daarnaast is zij als wetenschappelijk onderzoeker verbonden aan het Radboudumc en aan de HAN.

Zij is lid van het in 2009 opgerichte Netwerk Ergotherapie Onderzoekers Nijmegen ([www.neonijmegen.nl](http://www.neonijmegen.nl)). Naast deze werkzaamheden werkte zij vanaf 2004 aan dit proefschrift.

Het Medisch Comité Nederland-Vietnam heeft haar onlangs gevraagd een opleiding ergotherapie op te richten in Hanoi, Vietnam.

In haar vrije tijd hielp zij haar echtgenoot Harry van Kuyk met het voorbereiden en inrichten van tentoonstellingen, de opmaak van zijn uitnodigingen, affiches en privéuitgaven. Verder wijdt zij zich aan zilver- en goudsmeedwerk en vervulde zij een aantal maatschappelijke functies onder meer voorzitter van de plaatselijke afdeling van de VVD, president van de regionale serviceclub Soroptimisten. Momenteel is zij het bestuurslid van de Harry van Kuyk Stichting.

## **PUBLICATIONS MISCELLANEOUS**

Naast de publicaties in deze dissertatie is zij (mede)auteur van drieëntwintig nationale en internationale publicaties, waarvan zestien in peer reviewed tijdschriften.

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