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RESEARCH PRIORITIES OF PATIENTS WITH NEUROMUSCULAR DISEASE.

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ABSTRACT

Purpose This paper describes a study in which patients with neuromuscular disease (NMD) were engaged to list top-priorities for scientific research in order to complement the researchers' agenda.

Method A dialogic model for research agenda setting was used. Interviews, focus groups and expert meetings with patients were held to identify research topics. Research topics were prioritized via a questionnaire. Agendas were integrated in a dialogue meeting with professionals and patients.

Results The research agenda of NMD patients is divided in four research domains, with a total of 24 research topics. These domains include 1) health; 2) quality of life; 3) quality of care and support, and 4) basic issues. Among the research domains highest priority was given to research on health, followed by research on quality of life.

Conclusions Both patients and professionals agreed a proper balance needs to be found between fundamental research and research on symptomatic treatment and quality of life. They concluded that more attention is required for research on the effective treatment of symptoms, quality of life and implementation of knowledge about NMD in regular care.

5.1 Introduction

Popular notions like 'shared decision making' and 'self management' indicate a growing acknowledgement of the value of patients' perspectives in treatment decisions. Patients' perspectives are also considered relevant for enhancing the quality of care and support (Hartley, Goodwin, & Goldbart, 2011; Scherer & Lane, 1997; Wain, Kneebone, & Billings, 2008; Wicks & Frost, 2008). In line with this trend we see a growing sense of urgency to involve patients in medical research and setting research agendas (Entwistle, Renfrew, Yearley, Forrester, & Lamont, 1998; Tallon, Chard, & Dieppe, 2000).

Funding agencies increasingly demand the inclusion of patient reported outcome measures in clinical trials, like the Food and Drug Administration in the United States and the European Medicines Agency. This is also encouraged in the TREAT-NMD network which aims at "bringing together leading specialists, patient groups and industry representatives to ensure preparedness for the trials and therapies of the future" (http://www.treat-nmd.eu/).

It is expected that patient involvement in research will ultimately make practice and policy more relevant to consumers' needs, leading to outcomes that include greater patient satisfaction, improvement in treatment adherence and better acceptance of research findings. Given these benefits patients are therefore more frequently consulted about the topics they prioritize for scientific research. Research agendas from patient perspectives are, for example, developed for kidney disease (Schipper & Abma, 2011; Tong et al., 2008), spinal cord injuries (Abma, 2005), asthma and COPD (Caron-Flinterman, Broerse, Teerling, & Bunders, 2005), burns (Abma & Broerse, 2010), cancer (Wright, Corner, Hopkinson, & Foster, 2006), ulcerative colitis (Welfare, Colligan, Molyneux, Pearson, & Barton, 2006) and people with an intellectual disability (Nierse & Abma, 2011).

In the field of neuromuscular diseases (NMD) patient organizations have since long taken an active role with regard to scientific research (Boon & Broekgaarden, 2010; Rabeharisoa, 2003, 2006). A sociological study described a successful engagement of patient associations in research, which was initiated by the French MD Organisation, and resulted in a partnership model in which the patient organisation is an active player in decision making processes on research (Rabeharisoa, 2003, 2006). The Dutch Patient Association for NMD ("Vereniging Spierziekten Nederland", VSN) also has taken an active role to develop research networks in

The Netherlands (Boon & Broekgaarden, 2010; Streng, 2003). The VSN also keeps track and reports on scientific developments relevant for their members and has put issues like reimbursement of expensive drugs on the policy agenda (Boon & Broekgaarden, 2010). In these initiatives patients are not just consulted on an ac hoc basis, but structural relationships have been developed with researchers.

Despite this active engagement with scientific research no studies have been found that specifically focus on patients' involvement in research agenda setting for NMD.This article aims to describe research priorities listed by NMD patients. It is based on a study commissioned by the Netherlands Organisation for Health Research and Development (ZonMw) and was carried out by a team consisting of two researchers with a background in social sciences, a staff member of the Patient Association for NMD (VSN) and a neurologist who was the head of the Dutch Neuromuscular Research Centre (ISNO) during the study period. All team members were not engaged in clinical care of any of the NMD patients who participated in this study. The project started in April 2006 and lasted till January 2008. The project aimed to integrate the research agendas of both patients and researchers/clinicians.

5.2 Methods

5.2.1 Design

A dialogic model for research agenda setting was used integrating interviewing, focus groups, expert meetings and a digital questionnaire (Abma & Broerse, 2010). Interviews were used to capture illness experiences and their impact on the lives of patients (Britten, 1995). Focus groups were organized to broaden and deepen the interview data, and to work towards topics for research (Balch & Mertens, 1999; Kitzinger, 1995). Expert meetings were organized with active members of the patient association (NMD patients and representatives) to discuss the patients' agenda and to complement and validate the research topics. The questionnaire among patients aimed at prioritizing the research topics, and was used to reach a larger group of patients than can be achieved by only using qualitative methods. A dialogue meeting with active members of the patient association and clinicians/researchers, among them a rehabilitation physician / professor, was organized to deliberate about these topics in order to achieve a shared agenda for research.

The mixed methods design follows a sequential path as the qualitative data form the input for the quantitative survey, followed by a qualitative interpretation of the findings (Curry, Nembhard, & Bradley, 2009; Greene, 2007). See figure 1 for a chronological overview of the research activities.

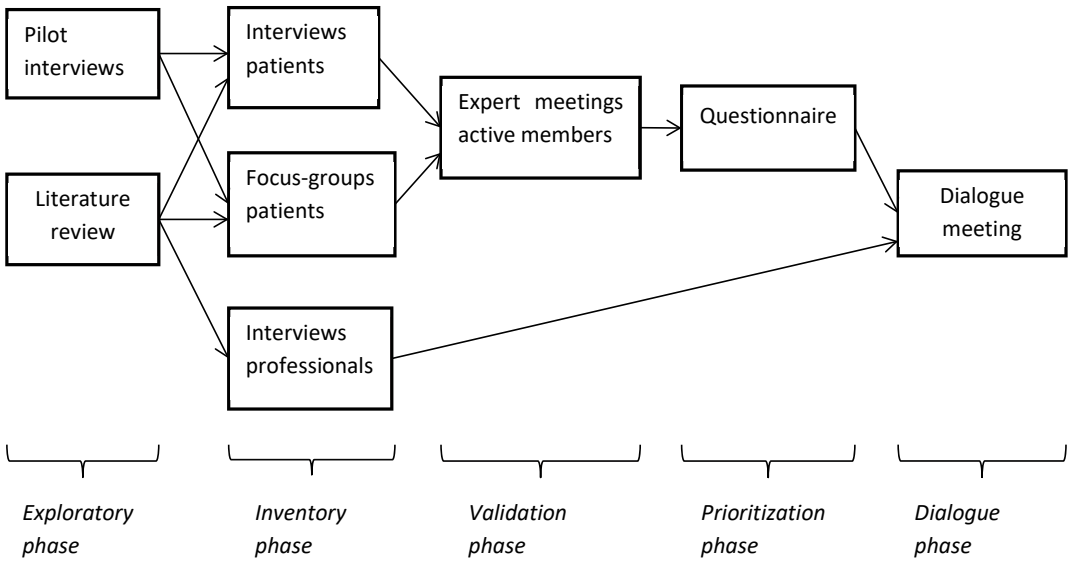

Figure 5.1. Flow chart of the phases and subsequent research activities during the research process. In the exploratory phase a literature study and pilot interviews were conducted. In the inventory phase interviews and focus groups with patients with NMD, and interviews with researchers/clinicians were held. In the validation phase expert meetings with active members (patients and representatives) of the patient association were organised. In the prioritization phase a questionnaire was set out among patients who were asked to give their top three priorities for research. In the dialogue phase a dialogue meeting was organised with patients and representatives and researchers/clinicians. The arrows indicate that the research process followed a iterative design, meaning that data that resulted from one phase in the process were used as input for the following phase.

5.2.2 Participant selection

NMD patients were eligible to participate if they were able to speak Dutch, 16–80 years of age and able to give informed consent. Given the diversity of NMD it was decided to focus on a

representative set of non-curable NMD conditions, because this is by far the greatest group within NMD and because it was expected that here the greatest benefits within research could be realized. Selection criteria used to include patients with diverse characteristics were degree of progressiveness, hereditariness, severity of symptoms, age of onset and rarity of the disease. After intense discussions the following diagnoses were included: Amyotrophic Lateral Sclerosis (ALS), Spinal Muscular Atrophy (SMA), Post-Polio Syndrome (PPS), Hereditary Motor and Sensory Neuropathies (HMSN), Duchenne Muscular Dystrophy (DMD), and Myotonic Dystrophy (MD).

Participants were approached by the VSN and recruited by the research team. Participants were intentionally sampled for the interviews to achieve a good division of participants over the diagnostic groups (Kuper, Lingard, & Levinson, 2008). Participants were asked to tell about their whole illness history from the (process of) diagnosis till their current situation. We also interviewed the parents of a son with DMD.

The interviews thus were directed at the stories of individual patients, whereas subsequently the focus groups aimed at exchanging experiences and generating shared understandings about possible topics for research (Britten, 1995; Kitzinger, 1995). Participants were selected based on their presumed ability and willingness to interact with other patients.

One focus group was held with youngsters with one of the selected diagnoses (MD, PPS and HMSN). Since ALS patients could not join focus-groups due to their condition, additional interviews were held with them. Focus group participants were offered reimbursement for their time and transport expenses.

Also, researchers, physicians and paramedics were interviewed about research on NMD. The latter data are not included in this article as it focuses on the patients' agenda.

5.2.3 Setting

Patients could choose the location of the interview and most interviews were held at home. The focus groups and expert meetings were held in neutral locations to encourage openness during discussion.

5.2.4 DATA COLLECTION

A topic list was developed to structure the interviews with patients, which was based on the International Classification of Functioning, Disability and Health (ICF) framework, a literature review, ego-documents and discussion among the research team. It was adjusted after two pilot-interviews. The 11 interviews with patients (table 1) were completed by duos composed of an academic researcher and VSN staff member. The interviews lasted 1,5 to 3 hours.

From the analysis of the interviews with patients, we identified several research areas that were further explored in three focus groups (N = 26). The focus groups were led by a social scientific researcher and VSN staff member. About eight to nine participants were in each focus group. The facilitators were experienced in how to conduct focus groups and how to use strategies to elicit and equalize participation. A structured and well suited protocol was developed and used for each focus group. To validate the themes an expert meeting with 11 active members of the patient association (both patients and representatives, selected on their knowledge of stories of other patients; good division over the six diagnostic groups) was organized.

Table 5.1 Overview of patient participants.

	Interview (N=8)	**Focus-group (N=3)**	**Expert meeting (N=2)**	**Total**
ALS	3	-	2[1]	5
DMD	1[2]	2	2[3]	5
HMSN	1	6	2	9
MD	1	8[4]	2	11
PPS	1	5	2	8
SMA	1	5	1	7

[1] Partners of a patient with ALS.
[2] Parents of a son with DMD.
[3] A parent of a son with DMD.
[4] Of which two partners of patients with MD.

Then a questionnaire was used to prioritize the research topics of patients. Important issues derived from the interviews, focus groups and expert meeting were translated to research domains and topics. These domains and topics formed the backbone of the questionnaire, which was piloted and discussed with the expert group of patients and representatives and subsequently amended to make it assessable for NMD patients. The questionnaire consisted of four parts. One part concerned relevant characteristics of the respondents. The second part focused on prioritizing of four research domains. In the third part research topics were prioritized per domain. The fourth part of the questionnaire concerned evaluative questions and invited the respondents to make remarks. The patient organization distributed 246 digital questionnaires to their members. One week after distribution a reminder was sent. The outcomes were discussed among the research team and among the expert group.

In the last stage a dialogue was organized between eight patient representatives from the VSN (one father of a child with DMD; one partner of a person with ALS; six patients with other NMD) and two researchers (neurology and MD in rehabilitation medicine, who were both not involved in clinical care for the participants) to integrate both agendas. This dialogue meeting was facilitated by the two social scientific researchers.

5.2.5 Data analysis

The interviews and focus groups were digitally audio-recorded after consent and transcribed in full. The transcripts were read and coded by two team members (CN, TA) independently. They compared and discussed their individual coding choices afterwards. Any disagreements were resolved by discussion. Through a process of careful analysis and comparisons we inductively developed the research themes. To check the validity of the analysis each respondent and focus group participant received an account with the question if they recognized the analysis (member check) (Kuper et al., 2008). All patients agreed and some extended their account. Triangulation of methods added to the breath of the study (Greene, 2007). The results of the digital questionnaires were scored by a VSN staff member using Excel.

5.3 RESULTS

The research agenda is divided in four domains, with a total of 24 research topics from the patients' perspectives. These domains include: research on health; quality of life; quality of care and basic issues. In the domain quality of life a distinction was made between social aspects and societal aspects. The research domains and research topics are visualized in figure 2.

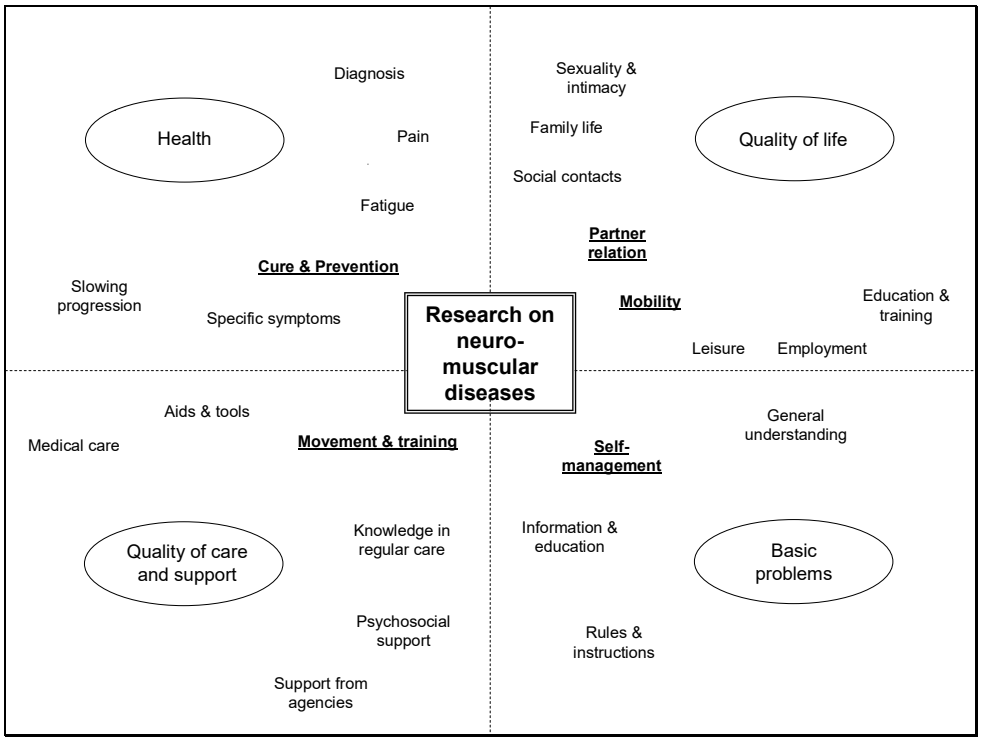

Figure 5.2 Research topics from the perspectives of patients with neuromuscular disease. These topics were collected during the interviews, focus groups and expert meetings with patients. The research topics are divided in four domains: Health; Quality of life; Quality of care and support; Basic problems. Per domain one topic (underlined) was prioritized in the questionnaire among patients.

The main research domains also have been prioritized. The response rate of the questionnaire was: 246/171 (70 %). Table 2 gives an overview. The grey areas indicate that research on health was most often prioritized as number 1. Priority 2 was most often given to research on

quality of life, including social and societal aspects, and research on quality of care. Basic issues did not gain priority. Basic issues refer to more general bottlenecks patients experience in life including the lack of self-management, information and education and bureaucracy. Below the research domains and topics will be described.

Table 5.2 Priorities among research domains as scored by patients with NMD.

Research domains	priority 1	priority 2	priority 3
Health	83%	6%	6%
Quality of life, social aspects	7%	30%	17%
Quality of life, societal aspects	4%	28%	26%
Quality of care	3%	30%	33%
Basic issues	3%	6%	19%

5.3.1 Health

NMD patients regard their health as a central problem on its own. At the same time, patients indicate that their health problems lie at the core of problems that relate to several other domains in their lives. Research on the cure, slowing down progression and prevention of the occurrence of symptoms was initially prioritized within this domain. In the dialogue meeting with professionals the priority given to cure and prevention was placed in perspective, and more emphasis was placed on research on symptomatic treatment. Patients realized that a cure will only become available in the long run for most NMD, and that in the meantime the majority of NMD patient suffers from various symptoms such as pain and fatigue on a day-to-day basis. Patients and professionals therefore agreed that a proper balance is needed between fundamental research and research on symptomatic treatment.

Cure & prevention

Most patients having the diagnosis for quite some time realize that their disease has no cure and will be a lasting influence in their lives. Some patients say they have reconciled with this situation, whereas others place their hopes on a curative treatment.

"The wish for a cure is important, but we have to realize that this is basic research. … Apart from that you need to have some money for applied research. How a NMD can be cured can cost millions, and that only for a start."

Research should focus on finding the cause of the disease, the prevention of the occurrence of symptoms, finding a cure for the disease and how specific physical complaints can be cured.

Slowing progression

Sometimes patients indicate that they want to live day-by-day, and do not dare to look at the future. However, many say that they are distressed about the future and its insecurities, and that the progression of the disease is what limits them the most: what will be the next thing that they will no longer have the capacity for, and when will it happen? It is the recurring confrontation and adaptation to new limitations which causes distress.

"I suffer most from the progression. I feel that I have to keep on making sacrifices all the time."

Research should focus on what patients can do themselves and finding effective treatments to slow down the progression of the disease. Also research into making more accurate and patient specific predictions on the development of the progression is considered desirable.

Pain, fatigue and disease specific complaints

Pain is a common complaint. Not all NMD patients experienced pain chronically or frequently, but they do report on problems in dealing with pain complaints. Fatigue is also a frequent feature and includes having limited energy during the day. Fatigue is surrounded with ignorance and negative comments by the environment. Patients easily become overburdened when they do not succeed in dividing their energy or in prioritizing activities during the day. In addition patients experience various disease specific complaints like cardiac diseases, respiratory problems, sleep disturbances, scoliosis and intestinal problems. Research should concentrate on effective treatment and prevention of pain, fatigue and disease specific complaints.

5.3.2 QUALITY OF LIFE; SOCIAL AND SOCIETAL ASPECTS

Many problems NMD patients experience are related to daily life. Aspects of daily life gain a different meaning due to the illness, therefore the quality of life is under pressure. The quality of life domain consists of a broad array of topics, which can be further distinguished between social topics (partner relation, sexuality, social contacts, family life) and societal topics (mobility, leisure, employment, education). In the dialogue with professionals it was acknowledged that research on the factors that influence quality of life is extremely important.

Partner relation and sexuality

NMD patients may experience problems in their partner relation. These are partly related to fatigue and increasing dependence.

> *"Someone within my disease (Myotonic Dystrophy) sleeps, for instance, the whole evening after some time. What do you then still have in common as a couple? The equality is fading away."*

Feelings of guilt may arise, because of the reliance on the partner for help. Also, the relationship changes and may become less equal if the partner acts as informal caregiver. ALS patients mention bereavement as a complicated aspect in the partner relation. Problems to have children may put pressure on the relation, as well as sexual limitations. Research should focus on ways to support of patients their partners to maintain quality in their relationship.

Social contacts

Building and maintaining friendships can be a problem for people with NMD.

> *"My boys have hundred thousand contacts, they chat and use msn, but they hardly see anybody. I don't know if they miss it. But a problem can arise if you grow older and do develop a need for human contact."*

The lack of contacts may relate to mobility limitations, fatigue and limited energy as well as to problems to socialize with others and lack of empathy. Research should focus on finding ways to prevent social isolation and to create conditions to participate more equally in society.

Family life

If a family member has NMD this can have its impact on the whole family. Especially in the case of a progressive disease. Family members are coping differently with the disease, and this may function well but can also lead to tensions in the communication and interaction.

> *"Everyone in the family experiences it in a different manner, and at various moments. At one moment someone can be very down, while you just see it positively. Than you can support each other, that is actually very special."*

A hereditary disease can put pressure on the whole family, including feelings of guilt. Daily care is often carried out by the parents or partner and may cause overburdening (physical and emotional). In the case of ALS too many professional caregivers may negatively affect the family life. Research should focus on how to prevent overburdening, and achieving an optimal balance between informal and professional care

Mobility and leisure

Many NMD patients are limited in their mobility, and dependent on aids, other persons or institutions. Walking and cycling is often no longer possible. Driving a car is often complicated due to limited financial compensation for adjustments. Public transport is limited. Limitations have consequences for social contacts and leisure activities.

> *"There is just less possible with a wheelchair. I would like to do more […] I can do many leisure activities, but travelling is impossible."*

Research should concentrate on ways to improve mobility inside and outside the home, effective technical aids to become more mobile, and how best to improve accessibility of public transport and leisure activities.

Employment and education & training

Work and education are important aspects in the lives of NMD patients. It helps to regain a feeling of independence. Patients feel, however, that agencies do not support them to find a job or internship.

"I said to the agency that I am finished with school and look for a job, but they thought that was weird."

Some experience troubles dealing with colleagues and disbelief, especially if the disease is less visible. Patients who have been ill since their youth indicate they have been bullied at school. Research should focus on ways to support people to find appropriate work or internships, or to make effective adjustments in work activities, and finally finding ways to prevent disbeliefs and ignorance among employers, colleagues and at schools.

5.3.3 Quality of care and support

Many NMD patients expect help and support from caregivers and providers for the reduction of their complaints and improvement of their quality of life. Within this research domain patients gave initially priority to movement and training. In the dialogue with professionals also emphasis was placed on implementation research as the lack of knowledge about NMD in regular care became more clear.

Movement & training

Movement and physical training are considered as important means to build one's condition, to stop excessive deterioration and to counter fatigue. Problems relate to the appropriateness of certain training schedules, finding a balance between building ones condition and preventing over burdening.

"You get tired. On the one hand you have to stay moving, but on the other hand you risk fatigue. It is hard to find a proper balance."

Finally, NMD patients doubt how they can integrate training into their lives. Research should focus on the best ways to tailor training to the unique person with NMD, whether training can improve one's condition and a better balance between movement and overburdening.

Medical care and knowledge in regular care

Patients have to deal with many care providers and the coordination and continuity of care can be problematic. After the rehabilitation phase patients experience a lack of support.

"You go to a rehabilitation doctor, and are being thrown into a trajectory for six weeks, you visit a psychologist, a physiotherapist, a vocational therapist. But you are suddenly finished and then you are just left along all by yourself."

Patients experience shortcomings in hospital care if this care it not adjusted to their disease. They mention the lack of knowledge about the side effects of anaesthetics and about the NMD specific side-effects of regular medicines and combinations of medicines. Regular treatments or examinations can be burdening and painful. General practitioners have limited knowledge of NMD.

Research should focus on finding ways to increase knowledge among professionals about NMD-specific side-effects and combinations of medicines, appropriate narcotics and physical examinations for NMD patients. Furthermore, research should concentrate on improving the coordination and the continuity of care.

Aids & tools and support of agencies

Most NMD patients are dependent on technical aids in their daily lives. They are in need of aids to make them more independent and more mobile. Youngsters expect much from technological innovations and ICT.

"I would like to have a robot who can take over care, so that you are no longer dependent of somebody. A robonurse."

NMD patients find it difficult to apply for support. They may wait too long, and institutions not always take the progression into account. Approvals may come (too) late.

"An example is the special chair. You only get such chair if you cannot stand up anymore, while such chair is meant to prevent fatigue."

Research should focus on how best to assist NMD patients in their trajectory to apply for support from agencies, to improve the accessibility of aids and on developing new, attractive aids to help patients to function more independently.

Psychosocial support

Progression of NMD requires patients to adjust their lives all the time. Finding a balance can be difficult. Patients may also experience psychosocial needs to deal with their partner relation, family, social contacts and in the case of ALS with end-of-life. Patients express a need for psychosocial support of themselves, but also indicate support is required for their partner and family. Research should focus on how best to support people to cope with the disease.

5.3.4 Basic issues

Finally there are some basic bottlenecks many respondents mentioned and recognized. These bottlenecks have an influence on all domains of life. Self management was given priority in this domain.

Self management and information & education

NMD patients are for many things dependent on others. This is not at all easy, and many long for more independence and more control over their lives. They like to become the managers of their own lives. It helps if they gain honest information that anticipates on the future. Although there is many information, many patients feel uninformed. They do not know where to get practical and medical information. Research should focus how patient can regain their autonomy and become more in control over their lives, and how available information on NMD can be better disseminated to patients and others involved.

General understanding

It is hard for NMD patients to participate in society due to the inaccessibility of buildings, mobility problems and disbeliefs about limitations and fatigue. Sometimes patients feel mistreated. Research should focus on how society can be influenced to be more welcoming and enabling for NMD patients.

Rules and instructions

Patients with NMD experience many problems with the bureaucracy and procedures for technical aids, housing adjustments, social welfare, driver licenses etc. Rules are not transparent, communication unclear and people receive contradictory advises. Research

should focus on how greater transparency and consistency of rules and instructions can be facilitated.

5.4 Discussion

In this process, consisting of different phases we showed how patients with NMD were able to compose an agenda for research. This and other studies show that patients can actually formulate relevant questions for research if one starts with their life world experiences and creates a platform for deliberation to develop their voice (Abma, 2006; Abma, Nierse, & Widdershoven, 2009; Nierse & Abma, 2011; Nierse, Schipper, van Zadelhoff, van de Griendt, & Abma, 2012). Our study led to the identification of 24 research topics from the perspective of NMD patients covering four research domains.

In a dialogue between patients and representatives from the expert group and researchers and clinicians we further developed a scientific research agenda based on the experiences and needs of NMD patients. Although professionals have, historically, denied the validity of the knowledge of patients because of its perceived lack of objectivity, verifiability, universality or rationality (Caron-Flinterman, Broerse, & Bunders, 2005; Entwistle et al., 1998; Epstein, 1995), lately in the field of NMD research priorities have shifted in the direction of symptoms such as fatigue (Erdmann, Lindeman, Cats, & Van Den Berg, 2010; Kalkman, Zwarts, Schillings, van Engelen, & Bleijenberg, 2008; Minis et al., 2010). In the dialogue meeting a mutual learning process occurred and professionals acknowledged diverse areas for research that mattered for patients. The exchange of perspectives resulted in a shared agenda for research.

One of the limitations of this study relates to the fact that we only consulted members of the VSN. We have involved members who were not active in the patient association by consulting them in the interviews, in the focus groups, and by sending them the survey for prioritization of topics. For example, patients with ALS could not attend focus groups or expert meetings, due to the progressive nature of their diagnosis. We therefore decided to do some extra interviews, in order to have their voices heard in the research agenda.

The participants of the expert meetings and the dialogue meeting were active members of the patient association. These members may be the more vocal, assertive and critical patients.

The inclusion of "quieter", non-active members as well as active members provided a balanced selection of NMD patients.

In future research, selection bias can be avoided by not only approaching members of a patient association, but also recruiting patients via other canals, like centres for rehabilitation or placing ads in general patient magazines.

Another limitation is that we used a digital questionnaire to ask patients to prioritize research topics. The VSN regularly uses this method to reach their members, because it is considered a less intensive method for people with NMD to respond, than via a paper questionnaire. However, it excluded patients who did not have internet access, or who were uncomfortable with digital technologies. It could be expected that typically elderly are underrepresented in a digital questionnaire, but in this study it was not the case, since a considerable amount of the respondents who returned the questionnaire were in the age group 61 years and older. Contrary to what is sometimes assumed the implementation of the patient agenda does not mean that current research projects should be stopped or diminished. NMD patients acknowledge the importance of biomedical and health research. They rank research on cure and prevention on the occurrence of symptoms very high. Patients and representatives in the expert group explained that many patients want to go back to normalcy. The disease has such an impact on their lives that people long for a life without the disease. The progression requires continuous adaptation and cure is seen as the ultimate solution even if this is not available within life time. The heredity of the disease is a strong motivator: patients hope research will be of help to their families in the future. However, patients and professionals indicate a balance is needed between fundamental research and research on preventing and treating symptoms (e.g. pain, fatigue and other disease specific symptoms) as well as research on slowing down the progression.

Research on the factors that influence the quality of life of patients with NMD is beginning to gain more attention (Cup, 2011; De Groot, Post, Heuvelen, Van den Berg, & Lindeman, 2007), and the agenda presented here might be seen as a legitimization of such research. Within this area NMD patients prioritize research on the partner relation and social contacts as well as research on mobility. Research in those areas is emerging and contribute to the relevance of medical research. Within the Quality of care research domain NMD patients prioritized

research on movement and physical training. Although research in this area has been started (Erdmann et al., 2010; Voet, Bleijenberg, Padberg, van Engelen, & Geurts, 2010), more emphasis is needed on finding the proper way to incorporate training in daily life. Another theme that is gaining more attention is the importance of implementation of existing knowledge to enhance the quality of care in regular settings.

The patient research agenda does not compete but rather complements the research already conducted on NMD. One may even state that the patient-driven agenda on NMD is validating the professional conceptualization and the wide scope on rehabilitation captured in the ICF model. The broad array of topics, arising from experiential knowledge concerning their bodies as well as what it means to live with NMD, requires research in new, emerging areas like societal participation (Cardol et al., 2002; Hammel et al., 2008; Schipper, Visser-Meily, Hendrikx, & Abma, 2011), of care services (Hartley et al., 2011; Wain et al., 2008), and of new technologies (Scherer & Lane, 1997). The patient-driven agenda also points to the need for more interdisciplinary research. To deal with the interrelated topics in the domains of quality of life, quality of care and basic problems researchers in NMD – neurologists and rehabilitation physicians - and social scientific researchers increasingly work together and expand their collaborations. Jointly they can work on the research priorities mentioned by patients in order to unite with patients' perspectives. Structural involvement of patients in NMD research helps to stay tuned to patients' needs, and reinforces their social and political participation in decision making processes concerning research.

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DECLARATION OF INTEREST

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