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ORIGINAL REPORT

DEVELOPMENT AND TESTING PSYCHOMETRIC PROPERTIES OF AN ICF-BASED HEALTH MEASURE: THE NEUROMUSCULAR DISEASE IMPACT PROFILE

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Objectives: **To develop a measure that is based on the International Classification of Functioning, Disability and Health (ICF) and reflects the prevalence and severity of disabilities related to neuromuscular disorders, and to evaluate the psychometric properties of this measure.**

Methods: **A preliminary questionnaire was developed, based on the categories of the ICF Core Set for Neuromuscular Diseases. Next a cross-sectional postal survey was carried out among 702 patients (70% response rate) diagnosed with a neuromuscular disease. Finally, psychometric properties were examined.**

Results: **The preliminary Neuromuscular Disease Impact Profile (NMDIP) consisted of 45 items. Factor analysis showed that the NMDIP comprised domains representing 3 ICF-components: 5 factors in the Body Functions component, 2 factors in the Activities component, and 1 factor in the Participation component. Scales showed moderate to good internal consistency (α = 0.63–0.92) and mean inter-item correlation coefficients (0.38–0.77). Convergent and discriminant validity analysis indicated that the NMDIP measures the impact of neuromuscular disease on physical, mental, and social functioning. The NMDIP discriminates between groups who differ in extent of limitations.**

Conclusion: **The NMDIP is an ICF-based measure that reflects neuromuscular disease-related disabilities. It consists of 36 items divided over 8 scales with satisfactory psychometric properties and 4 single items.**

Key words: neuromuscular disease; health measure; International Classification of Functioning Disability and Health; psychometric properties.

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INTRODUCTION

Neuromuscular diseases (NMD) may be acquired or hereditary. Causes are dysfunction of the anterior horn cell or sensory ganglion cell (neuronopathy), peripheral nerve (neuropathy), neuromuscular junction (myasthenia), or muscle (myopathy) (1). Common symptoms of neuromuscular diseases include muscle weakness, impairment in muscle endurance, involuntary muscle activity (stiffness, myotonia, cramp, and fasciculation), sensory loss, autonomic dysfunction and impairment in control of voluntary movements. Sensations of pain and fatigue are common consequences of these muscle and nerve disturbances (2, 3). These symptoms have a profound impact on daily activities and participation in life situations (4, 5).

In clinical practice and research there is a need for reliable and validated assessment tools as well as outcome measures that cover the broad range of health problems in neuromuscular patients (6).

Over the last 2 decades many health measurement instruments have been developed for use both in clinical practice and in research. As a result, there are generic health-related quality of life (HRQoL) instruments with a broad scope, for example, the Medical Outcome study Short Form Questionnaire (SF-36) (7). An example of a disease-specific HRQoL instrument with a broad scope is the Amyotrophic Lateral Sclerosis Assessment Questionnaire (ALSAQ-40) (8). In addition, there are generic and domain-specific measures to assess limitations in daily living, for example the Groningen Activity Restriction Scale (GARS) (9), or to assess participation in life situations, for example, the Impact on Participation and Autonomy Questionnaire (IPAQ) (10).

The International Classification of Functioning, Disability, and Health (ICF) is a classification developed by the World Health Organization (WHO) and aims at providing a unified and standardized language for describing and classifying health domains and health-related states, and hence providing a common framework for the development of health outcome measures (11, 12).

The ICF comprises 4 key components. The first component, *Body Functions and Structures*, refers to functions of body systems, and to anatomic parts. The second component, *Activities*, refers to "task or action execution by the individual". The third component, *Participation*, refers to "involvement in life situations". The *Environmental Factors* that interact with these 3 components, are described in the fourth component of the ICF (11).

In the model of functioning that underlies the ICF classification system, the components body functions and structures, activities and participation are summarized under the concepts

"functioning" and "disability". These are associated both with health status and with personal and environmental factors. Functioning is an umbrella term encompassing all body functions, activities and participation. Similarly, disability is an umbrella term for impairments in body functions, limitations in activities and restrictions in participation (11). ICF Core Sets have been the first approach to providing ICF-based instruments for clinical practice and research (13). An example of such ICF-based measures is the Multiple Sclerosis Impact Profile (MSIP) (14). This measurement instrument has shown to be a feasible assessment tool in practice and psychometrically sound measures in research. To the best of our knowledge there is no broad ICF-based health measure covering all 600 NMDs. Therefore the aim of this study was to develop an ICF-based measure, the Neuromuscular Disease Impact Profile (NMDIP), with the intention of reflecting the prevalence and severity of a broad range of disabilities related to neuromuscular diseases, using the ICF features such as ICF terminology and ICF qualifiers and to evaluate the psychometric properties of this new measurement instrument.

METHODS

Sample and procedures

A cross-sectional postal survey was conducted among patients diagnosed with a neuromuscular disease and registered at the Department of Neurology of the University Medical Center Groningen, the Netherlands. Criteria for inclusion were: diagnosed with a neuromuscular disease, aged 18 years or older, and able to read and write in Dutch.

In total 1,003 eligible patients were selected from the hospital patient record system with the aim of representing the 4 major neuromuscular disease (NMD) groups defined by Rowland: motor neurone disorders, muscle disorders, junction disorders and, peripheral nerve disorders (15). To prevent any inappropriate sending of the questionnaire, we cross-checked for deceased patients using the national population register.

Patients received information about the study and were invited to participate. Respondents completed the preliminary NMDIP, generic and domain-specific questionnaires, along with some demographic and disease-specific questions. Reminders were sent out after 2 weeks.

Ethical approval was obtained from the local ethics committee. Reference: METc 2009.310.

Preliminary Neuromuscular Disease Impact Profile

The preliminary NMDIP was developed as a disease-specific and ICF-based measure to assess disability among patients with an NMD. We used the 69 ICF categories of the NMD-Core Set (3). These categories are divided over the 4 ICF components. Selected categories were operationalized in order to estimate the patient's objectified opinion (impairment in body functions, limitations in activities or restrictions in participation) of the incidence and severity of a disability, and to estimate the support from relevant environmental factors. Furthermore, ICF terminology for "disabilities" was applied, ICF item labels were used when formulating the subject of the question (e.g. "urination" functions instead of "bladder" functions), and ICF codes (e.g. b280 or p920) were documented for each question (14). Illustrative examples were annexed (using fourth-level ICF-items) to some questions to ensure an adequate response. To record the presence and severity of a problem in functioning, we applied response scales with scoring options specified for each ICF component, based on "qualifiers" proposed by the ICF (14).

The preliminary questionnaire was reviewed by patients, clinicians, nurse specialists, experts on the ICF and methodologists ($n = 24$) for clarity, comprehensiveness, redundancy and patient burden. A modified questionnaire was pre-tested in a random sample of 3 clinicians and 50 patients who were not involved in the first appraisal of the questionnaire. Unclear or ambiguous items and instructions were identified and some modifications of the questionnaire were made.

Finally, the preliminary NMDIP reflects an objectified view of the prevalence and severity of NMD-related disabilities and consists of 45 items representing the 4 ICF components.

Measurement instruments

For evaluating the psychometric properties of the NMDIP, 2 generic and 2 domain-specific measures were used.

The SF-36 is a broad and generic HRQoL measure that consists of 36 items divided over 8 domains (7). For each domain, item scores were transformed to a scale that ranges from 0 (worst health) to 100 (best health). In a previous study among Dutch multiple sclerosis patients the SF-36 domains showed satisfactory levels of internal consistency: Cronbach's alpha ranged between 0.74 and 0.96 (14).

The World Health Organization Quality Of Life (abbreviation version) (WHOQOL-BREF) is a broad and generic measure of global QoL (16), and consists of 26 items divided over 4 domains. For each domain, item scores were transformed to a scale that ranges from 0 (worst health) to 20 (best health). In a previous study among Dutch multiple sclerosis patients the WHOQOL-BREF showed good levels of internal consistency: Cronbach's alpha ranged between 0.80 and 0.81 (14).

The GARS is a domain-specific instrument to measure limitation, and consists of 18 items divided over 2 domains (9). A 4-category response format is used, ranging from 1 (no problem in performing without help) to 4 (impossible to perform). Scores are summed for each subscale. The GARS showed strong levels of internal consistency: Cronbach's alpha ranged from 0.95 to 0.97 in a study in a Dutch sample of multiple sclerosis patients (14).

The IPAQ is a domain-specific questionnaire focusing on person-perceived participation and autonomy (10, 17). The instrument assesses 2 aspects of participation: perceived participation and the perceived problem. In this study we applied the perceived participation part that consists of 24 items divided over 5 domains. Items are scored on a 5-point rating scale with discrete responses, ranging from 1 (very good) to 5 (very poor). Scores are summed for each domain. In a previous study among Dutch multiple sclerosis patients, the IPAQ showed good levels of internal consistency: Cronbach's alpha ranged from 0.86 to 0.94 (14).

Item reduction

Exploratory factor analysis with oblique rotation (Geomin) (20, 23) was used to examine whether the domains measured by the NMDIP represent the 4 ICF components. To improve the content validity the prevalence of each item was examined before entering items in the factor analysis. Items with a low prevalence ($\leq 20\%$) were excluded from further analysis (18, 19). Factor analyses were performed using Mplus 6 software (20). Given the categorical nature of the variables, methods based on polychoric correlations and the robust-weighted least squares estimators (21) were used. Goodness-of-fit of the underlying factorial structure was measured by the root mean squared error of approximation (RMSEA, adequate if below 0.06) and the standardized root mean square residual (SRMR, adequate if below 0.08), the Comparative Fit Index and the Tucker-Lewis Index. For the latter 2 indices, it is recommended that values should be greater than 0.95 (22, 23). Items with factor loadings ≥ 0.40 were selected for scale construction.

Items that could not meet the criteria of scalability were taken into consideration for use as a single indicator.

Missing items

The maximum number of missing items allowed to be replaced by the mean scale score was determined by a sufficient Cronbach's alpha in relation to the number of scale items (19, 24).

Psychometric evaluation

The distribution of scale scores was evaluated by calculating the median, mean and standard deviation and observed score range. Proportion of patients with worst and best possible scores (floor and ceiling effect) were calculated. Proportions ≤20% were considered acceptable (18).

Internal consistency was examined with Cronbach's alpha (25) and the mean inter-item correlation coefficient (MICC) for each scale (26, 27). Alpha was considered sufficient if ≥0.70 (28, 29), and MICC if ≥0.30 (26).

To test whether NMDIP scales measure physical, psychological, social, and environmental domains of functioning, as they purport to measure, convergent and discriminant validity were assessed by examining the extent to which correlation values between NMDIP scales and concurrent measures were consistent with hypotheses (30, 31). Regarding convergent validity, we hypothesized that the NMDIP scales would have a strong correlation (≥0.70) (32) with scales that cover the same domain in concurrent measures. For example, NMDIP scales for physical functions should correlate highly with the SF-36 "Physical Functioning scale". To support discriminant validity, we hypothesized that the NMDIP scales would correlate weakly (<0.40) with scales measuring different domains in NMDIP or concurrent measures. For example, NMDIP scales for physical function would correlate weakly with mental or emotional scales of the SF-36.

Regarding known-groups validity (30, 31), we hypothesized that the NMDIP scales should be able to discriminate between subgroups of respondents known to differ on relevant clinical characteristics. The level of limitations due to a neuromuscular disease was used to create relevant subgroups of respondents. Therefore, the generic question "Extent of limitations" was used. Respondents were asked to answer the question "To what extent are you limited due to a neuromuscular disease?" on an 11-point scale with a score range from 0 (not limited at all) to 10 (severely limited). Next, respondents were divided into 2 groups: those with a "lower extent of limitations" (score 1–4), and those with a "higher extent of limitations" (score 5–10).

Statistical analysis

Patient characteristics were analysed using descriptive statistics. Spearman's correlation coefficient (rho) was used to examine convergent and discriminant validity. Known-groups validity was assessed using the independent Mann-Whitney *U* test.

To estimate the magnitude of the difference in scores between subgroups of respondents, the non-parametric effect size (coefficient *r*) for unrelated samples was calculated for statistically significant group differences (alpha = 0.05) (33). Coefficient *r* is calculated by dividing the z statistic (obtained from the Mann-Whitney *U* test) by the root of the sample size (*n*). To interpret the non-parametric effect sizes using coefficient *r*. Cohen suggested the following thresholds for interpretation: an *r* of <0.10 indicates a trivial effect, an *r* of ≥0.10 to <0.24 a small effect, an *r* of ≥0.24 to <0.37 a moderate effect, and an *r* ≥0.37 a large effect. An *r* ≥0.10 reflects a clinically relevant difference between groups (33, 34). IBM SPSS statistics version 20 was used.

RESULTS

Patient characteristics

In total 702 participants (70% response rate) completed the questionnaires. Demographics and disease-specific characteristics are described in Table I. Mean age was 59 years (SD 16, range 19–92 years), while slightly more than half of the patients were younger than 65 years. Mean number of "years since diagnosis" was 12 (SD 11, range 0–65 years). Approximately 30% of the patients were retired due to a neuromuscular disease. The motor neurone disorder subgroup was a relatively small sample compared with the other neuromuscular disease subgroups according to Rowland's classification (15).

Table I. *Sample characteristics (*n = *702)*

Variable	Total sample
Gender, *n* (%)	
Female	350 (50)
Male	352 (50)
Age, years, mean (SD) [range]	59 (16) [19–92]
Years since diagnosis, mean (SD) [range]	12 (11) [0–65]
Relationship status, *n* (%)	
Relationship (married/partnership)	498 (71)
Single (unmarried/widowed/divorced)	186 (27)
Educational level, *n* (%)	
Primary school/vocational training	235 (33)
Secondary school/vocational training	270 (38)
Higher education/vocational training	161 (23)
University	28 (4)
Employment status (more answers possible), *n* (%)	
Enrolled in a training or study course	36 (5)
Employment (part-time or full-time)	173 (25)
Voluntary work (part-time or full-time)	42 (6)
(Partially) retired due to NMD	213 (30)
Housewife/househusband	171 (24)
Retired due to age	244 (35)
NMD category, *n* (%)	
Motor neurone disorder	43 (6)
Muscle disorder	154 (22)
Junction disorder	234 (33)
Peripheral nerve disorder	271 (39)

NMD: neuromuscular disease; SD: standard deviation.

Non-respondents did not differ from respondents in terms of gender, but were significantly younger (mean 53, SD 19 years) than respondents (mean 59, SD 16 years).

Content validity

Nine of the original 45 items showed a low prevalence (≤20%) and were not entered in the factor analysis. These items were from the component "Activities" ("a350 Conversation", "a360 Using communication devices and techniques", and "a465 Moving around using equipment"), the component "Participation" (p510–p540 items concerning "Personal care", "p360 Communication devices and techniques, "p630 Eating and drinking", "p610 Acquiring a place to live" and "p850 Remunerative employment"), and from the component "Environmental Factors" ("e340 Personal care providers and personal assistants").

Item reduction

The EFA models showed a (very) good fit for the 5-factor model for the Body Functions component, the 2-factor model for the Activities component, and the 1-factor model for the Participation component. Comparative Fit Index and Tucker-Lewis Index values were above 0.95, SRMR values were below 0.08 as recommended. The RMSEA values were below 0.06 for the Body Functions component and Participation component, while the value for the Activities component was slightly higher (0.069), but still acceptable. For the Environmental Factors

Table II. *Factor analysis with Body Functions component categories (*n = *702)*

	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
Muscle Functions (MuF)					
Impairment in…					
b730 Muscle power functions	**0.612**	0.473	0.032	0.015	–0.054
b740 Muscle endurance functions	**0.677**	0.329	–0.003	–0.020	0.195
Movement Functions (MoF)					
Impairment in…					
b760 Control of voluntary movements functions	0.009	**0.777**	–0.100	0.023	–0.038
b765 Involuntary movements functions	–0.315	**0.849**	0.106	–0.022	0.044
b780 Sensation related to muscles and movement functions	0.045	**0.590**	–0.041	0.053	0.198
Swallowing and speech functions (SSF)					
Impairment in…					
b320 Speech functions	0.024	0.144	**0.842**	0.008	–0.013
b5105 Swallowing functions	0.246	–0.035	**0.687**	0.053	0.054
Excretion and reproductive Functions (ERF)					
Impairment in…					
b525 Defecation functions	0.022	–0.030	–0.002	**0.656**	0.168
b620 Urination functions	–0.052	0.028	0.036	**0.785**	–0.032
b640 Sexual functions	0.046	0.281	0.026	**0.426**	0.043
Mental Functions and Pain (MFP)					
Impairment in…					
b134 Sleep functions	–0.028	–0.008	–0.055	0.091	**0.680**
b1300 Fatigue	0.460	0.019	0.062	0.011	**0.593**
b152 Emotional functions	–0.020	–0.014	0.100	–0.148	**0.725**
b160 Thought functions	–0.230	0.040	0.259	0.023	**0.539**
b280 Sensation of pain	0.078	0.267	–0.238	0.040	**0.527**

Bold figures are sufficient factorloadings selected for scale construction.

component no satisfying fit was found. Factor analysis reduced the remaining 36 items of the initial NMDIP to 32 items:

- Five factors within the Body Functions component included 15 items (Table II). Interpretation of item content, using the ICF first- and second-level category labels led to the following scale labels: "Muscle Functions" (MuF), "Movement Functions" (MoF), "Swallowing and Speech Functions" (SSF), "Excretion and Reproductive Functions" (ERF), and "Mental Functions and Pain" (MFP).
- Two factors within the Activities component included 14 items (Table III). These factors were given the labels, "Activities of Moving around"(AMA) and "Self-care and Domestic Activities" (SDA). One factor within the Participation component included 3 items (Table IV). This factor was labelled "Participation in Life Situations" (PLS).

Table III. *Factor analysis with Activities component categories (*n = *702)*

	Factor 1	Factor 2
Activities of Moving around (AMA)		
Limitations in…		
a410 Changing body position	**0.860**	0.005
a415 Maintaining body position	**0.639**	0.270
a420 Transferring oneself	**0.711**	0.251
a450 Walking	**0.952**	–0.035
a470 Using transportation	**0.589**	0.330
a920 Recreation and leisure	**0.411**	0.354
Self-care and Domestic Activities (SDA)		
Limitations in…		
a440 Fine hand use	–0.003	**0.758**
a445 Hand and arm use	–0.102	**0.894**
a510 Washing oneself	0.272	**0.709**
a520 Caring for body parts	–0.007	**0.904**
a530 Toileting	0.335	**0.585**
a540 Dressing	0.200	**0.754**
a630 Preparing meals	0.057	**0.863**
a640 Doing housework	0.398	**0.458**

Bold figures are sufficient factorloadings selected for scale construction.

Finally, the Body Functions component items "Seeing functions" and the Environmental Factor component items "Immediate family", "Social security services", and "Health services" with a sufficient prevalence and clinical relevance were added to the questionnaire as single items.

Scale features are shown in Table V. Internal consistency for 7 NMDIP scales for the total sample was good and moderate for 1 scale. Cronbach's alpha ranged from 0.63 to 0.92 and mean inter-item correlation coefficient ranged from 0.38 to 0.77. Four scales showed a high floor effect.

The NMDIP scales within the NMD groups showed acceptable to good internal consistency. (Table VI). For some scales the Cronbach's alpha was weak, but this was compensated by a sufficient mean inter-item correlation coefficient, except for the "Swallowing and Speech Functions" scale in the peripheral nerve disorder group and the "Excretion and Reproductive Functions" scale in the muscle disorder group.

Table IV. *Factor analysis with Participation components (*n = *702)*

	Factor 1
Participation in Life Situations (PLS)	
Restriction in…	
p460/p470 Mobility	**0.690**
p740-p760 Relationships	**0.719**
p910/p920 Recreation and leisure	**0.903**

Bold figures are sufficient factorloadings selected for scale construction.

Table V. *Scale features of the Neuromuscular Disease Impact Profile (NMDIP) scales (*n = *702)*

	Cases *n*	Items k	Possible score range	Observed score range	Floor effect %	Ceiling effect %	Median	Mean	SD	Alpha	MICC
NMDIP scales											
Muscle Functions	658	2	0–8	0–8	8	3	4	3.4	1.9	0.87	0.77
Movement Functions	594	3	0–12	0–12	18	0	2	2.4	2.1	0.72	0.47
Swallowing and Speech Functions	669	2	0–8	0–6	59	1	0	0.7	1.1	0.69	0.53
Excretion and Reproductive Functions	509	3	0–12	0–12	27	0	1	1.8	3.1	0.63	0.38
Mental Functions and Pain	597	5	0–20	0–16	7	0	4	1.8	1.8	0.80	0.59
Activities of Moving Around	702	6	0–18	0–18	18	2	4	5.1	4.8	0.90	0.62
Self-care and Domestic Activities	701	8	0–24	0–24	28	1	2	4.7	6.0	0.92	0.59
Participation in Life Situations	695	3	0–12	0–12	49	0	1	1.7	2.4	0.72	0.47
NMDIP single item											
Seeing functions	666	1	0–4	0–4			0	0.68	0.88		
Immediate family	697	1	0–2	0–2			0	0.51	0.73		
Social security services	268	1	0–2	0–2			1	0.74	0.77		
Health services	693	1	0–2	0–2			0	0.66	0.75		

MICC: mean inter-item correlation coefficient; SD: standard deviation.

The final version of the NMDIP consists of 36 items divided over 8 scales and 4 single items (see Appendix I).

Psychometric testing

Table VII provides evidence of convergent and discriminant validity for the NMDIP scales, reflecting the impact of a neuromuscular disease on physical, psychological, and social aspects of functioning. The predictions were consistent with the direction, magnitude, and patterns of correlations.

Convergent validity was supported by a strong correlation between the NMDIP "Muscle Functions" scale and the SF-36 "Physical Functioning" scale, and a strong correlation between the NMDIP "Lower Extremity Activities" and "Upper Extremity Activities" scales, and the GARS "Activities of Daily Living" and "Instrumental Activities of Daily Living" scales. The NMDIP "Participation in Life Situations" scale was moderately correlated with the IPAQ participation scales.

Discriminant validity was supported by weak correlation values found, for example, between the NMDIP "Mental Functions and Pain" scale and the SF-36 "Physical Functioning" scale, and the GARS "Activities of Daily Living" and "Instrumental Activities of Daily Living" scales. Similarly, the NMDIP "Muscle Functions" scale correlated weakly with the SF-36 "Mental Health and "Role Emotional" scales, and the NMDIP "Participation in Life Situations" scale correlated weakly with the SF-36 "Mental Health" scale.

Evidence of known-groups validity was obtained for all NMDIP scales by statistically significant group differences and clinically relevant effect sizes (Table VIII). Patients classified as having a higher extent of limitation had statistically significant higher scores on all NMDIP scales compared with those classified as having a lower extent of limitation. Effect sizes were moderate for 2 scales, and large for 6 scales.

DISCUSSION

The objective of this study was to develop a psychometrically sound ICF-based measure for estimating the prevalence and severity of a broad range of disabilities related to neuromuscular diseases using ICF features such as ICF terminology and ICF qualifiers.

The results provide evidence to support the validity and reliability of the final version of the Neuromuscular Disease Impact Profile (NMDIP) as an instrument to measure the prevalence and severity of a broad spectrum of consequences of a neuromuscular disease including disabilities in Body Func-

Table VI. *Internal consistency (Cronbach's alpha) and the mean inter-item correlation coefficient (MICC) of the Neuromuscular Disease Impact Profile (NMDIP) scales per neuromuscular diseases group*

	Motor neurone disorders (*n* = 43)		Muscle disorders (*n* = 154)		Junction disorders (*n* = 234)		Peripheral disorders (*n* = 271)	
	Alpha	MICC	Alpha	MICC	Alpha	MICC	Alpha	MICC
Muscle functions	0.86	0.77	0.79	0.67	0.87	0.78	0.86	0.76
Movement functions	0.72	0.48	0.72	0.48	0.67	0.42	0.67	0.41
Swallowing and speech functions	0.82	0.72	0.74	0.59	0.63	0.47	0.42	0.27
Excretion and reproductive functions	0.71	0.47	0.54	0.30	0.63	0.36	0.68	0.45
Mental functions and pain	0.69	0.34	0.77	0.40	0.73	0.36	0.72	0.33
Activities of moving around	0.90	0.62	0.90	0.60	0.85	0.50	0.90	0.59
Self-care and domestic activities	0.95	0.69	0.93	0.63	0.83	0.43	0.88	0.47
Participation in life situations	0.80	0.57	0.74	0.49	0.67	0.40	0.67	0.41

Table VII. *Results of convergent and divergent validity analyses of the Neuromuscular Disease Impact Profile (NMDIP) scales (*n = *702)*

	Body functions					Activities		Participation	
NMDIP	MuF	MoF	SSF	ERF	MFP	AMA	SDA	PLS	Alpha
NMDIP									
Muscle Functions (MuF)	1								0.87
Movement Functions (MoF)	0.57	1							0.72
Swallowing and Speech Functions (SSF)	0.26	0.23	1						0.69
Excretion and Reproductive Functions (ERF)	0.35	0.43	0.31	1					0.63
Mental Functions and Pain (MFP)	*0.49*	*0.59*	*0.34*	*0.48*	1				0.80
Activities of Moving around (AMA)	**0.72**	0.58	0.23	0.41	0.48	1			0.90
Self-care and Domestic Activities (SDA)	**0.63**	0.52	0.31	0.45	0.51	**0.79**	1		0.92
Participation in Life Situations (PLS)	0.54	0.46	0.26	0.38	0.53	0.68	0.61	1	0.72
SF-36									
Physical functioning	**–0.71**	–0.52	–0.23	–0.40	–0.47	**–0.85**	**–0.77**	–0.61	0.94
Role physical	–0.43	–0.42	–0.26	–0.37	–0.52	–0.43	–0.45	–0.40	0.88
Bodily pain	–0.38	–0.51	–0.17	–0.32	–0.65	–0.42	–0.40	–0.38	0.91
General health	–0.49	–0.49	–0.32	–0.44	–0.60	–0.49	–0.48	–0.46	0.78
Mental health	*–0.21*	–0.33	–0.18	–0.18	**–0.53**	*–0.21*	*–0.25*	*–0.32*	0.83
Role emotional	*–0.18*	–0.30	–0.20	–0.21	–0.39	*–0.23*	*–0.26*	–0.25	0.87
Social functioning	*–0.44*	–0.46	–0.29	–0.40	–0.63	–0.46	–0.46	**–0.51**	0.77
Vitality	–0.41	–0.42	–0.34	–0.33	–0.68	–0.35	–0.39	–0.39	0.81
WHOQOL-BREF									
Physical health and autonomy	–0.29	–0.30	–0.22	–0.29	–0.49	–0.29	–0.29	–0.33	0.84
Psychological health	*–0.25*	*–0.33*	*–0.25*	*–0.23*	*–0.43*	*–0.29*	*–0.30*	–0.35	0.80
Social relation	*–0.23*	*–0.22*	*–0.14*	*–0.40*	*–0.37*	*–0.26*	*–0.25*	**–0.33**	0.60
Environment	*–0.34*	*–0.39*	*–0.24*	*–0.27*	*–0.51*	*–0.41*	*–0.39*	–0.46	0.82
GARS									
Activities of daily living	0.64	0.54	0.26	0.45	0.44	**0.81**	**0.81**	*0.62*	0.95
Instrumental activities of daily living	0.67	0.49	0.30	0.44	0.49	**0.78**	**0.84**	*0.62*	0.93
IPAQ									
Autonomy indoors	0.54	0.52	0.25	0.43	0.50	0.66	0.65	**0.56**	0.94
Family role	0.53	0.52	0.30	0.41	0.59	0.61	0.61	**0.53**	0.92
Autonomy outdoors	0.57	0.50	0.34	0.48	0.63	0.68	0.66	**0.66**	0.84
Social relations	0.34	0.35	0.32	0.41	0.53	0.41	0.45	**0.48**	0.85

NMDIP: *n* = 484–658; SF-36: *n* = 654–657; WHOQOL-BREF: *n* = 628–649; GARS: *n* = 655–658; IPAQ: *n* = 654–657.
Bold correlations = expected convergent correlations. Italic correlations = expected discriminant correlations.
MuF: Muscle functions; MoF: Movement functions; SSF: Swallowing and Speech functions; ERF: Excretion and Reproductive functions; MFP: Mental functions and Pain; AMA: "Activities of Moving around"; SDA: "Self-care and Domestic Activities"; PLS: Participation in life situations; GARS: Groningen Activity Restriction Scale; IPAQ: Impact on Participation and Autonomy Questionnaire.

tions, Activities and Participation, and lack of support from Environmental Factors. The NMDIP can be used as a clinical and research instrument for the assessment of the impact of a neuromuscular disease.

The original 45 items in the preliminary NMDIP could be reduced to 36 items: 32 items covering 8 domains representing 3 ICF-components, and 4 clinically relevant items (1 Body Functions item and 3 Environmental Factors items), which were applied as single items in the questionnaire (see final version in Appendix I).

Although the NMDIP used the same items as the initial MSIP (14), results of the factor analysis showed some differences compared with the final MSIP scales. For example, the MSIP "Muscle and Movement functions" 4-item scale is represented in the NMDIP in 2 separate and recognizable scales "Muscle functions" with 2 items and "Movement functions", also with 2

Table VIII. *Results of known-groups validity analyses of the Neuromuscular Disease Impact Profile scales (*n = *702)*

	Low (1–4) vs high (5–10) Extent of limitations				
	n	Low Mean Rank	High Mean Rank	*p*-value (Z-statistic)	Effect size
Muscle functions (MuF)	640	197.84	390.25	0.000 (–12.973)	0.51
Movement functions (MoF)	577	201.52	342.52	0.000 (–9.994)	0.42
Swallowing and speech functions (SSF)	651	273.86	359.14	0.000 (–6.445)	0.25
Excretion and reproductive functions (ERF)	495	198.34	279.74	0.000 (–6.374)	0.29
Mental functions and pain (MFP)	581	201.45	346.38	0.000 (–10.170)	0.42
Activities of moving around (AMA)	683	210.67	422.72	0.000 (–13.704)	0.52
Self-care and domestic activities (SDA)	682	227.58	411.25	0.000 (–11.956)	0.46
Participation in life situations (PLS)	677	239.81	400.46	0.000 (–11.110)	0.43

items. Furthermore, the 3-item "Mental functions" MSIP scale appeared in the NMDIP as a 5-item version: new scale items were Fatigue and Pain. This can be explained by the fact that pain and fatigue are the direct result of (using) weakened muscles, which is a common symptom in neuromuscular diseases. Unlike neuromuscular diseases fatigue in multiple sclerosis is most likely related to the process of inflammation, while pain originates from spasticity and/or neuropathy. Furthermore, scale construction also identified a construct that was not present in the MSIP: "Swallowing and Speech Functions". This can be explained by the fact that some myopathies and the myasthenia's tend to affect bulbar musculature. Finally, analysis showed no consistent factor for the Environmental Factors items.

Reliability of the NMDIP scales for the total sample was sufficient for 2 scales and good for 6 scales. The internal consistency of the scales per separate NMD group was generally sufficient, except for the "Swallowing and Speech Functions" scale in the peripheral nerve disorder group and the "Excretion and Reproductive Functions" scale in the muscle disorder group. Some caution is advised in the interpretation of the results of these scales. Convergent and discriminant validity analysis indicated that the NMDIP measures the impact on physical, mental, and social functioning for people with a neuromuscular disease.

The correlation between the NMDIP "Participation in Life Situations" scale and the SF-36 "Physical Functioning" scale was unexpectedly higher. It is likely that the activity-related participation items in the NMDIP scale are responsible for this moderate correlation.

Known-groups validity was supported for the 8 NMDIP scales. Scales discriminated sufficiently between groups of patients with a neuromuscular disease that differed in extent of limitations.

An important strength of this study is the large and broad group of participating patients with a neuromuscular disease, and the sound conceptual basis used in developing the NMDIP (3, 35).

A possible limitation of this study is the small sample size of the motor neurone disorder group, compared with the sample size of the 3 other NMD groups. However, in our opinion the disabilities in this group are sufficiently represented in the NMDIP because the basis of the NMDIP, the NMD ICF-Core set, covers all items of the disease-specific Amyotrophic Lateral Sclerosis Assessment Questionnaire-40 (8). Another limitation could be the high floor effect of some scales that might affect the reliability of these scales (36). However, these floor effects match the course of the slowly progressive nature of most NMDs. This means that some disabilities appear years after onset, such as speech and swallowing functions or upper extremity activities.

Further research should focus on psychometric evaluation concerning stability and sensitivity to change of the NMDIP scales, and validation across other populations of neuromuscular disease patients in other cultures. It would also be interesting to examine the differences in prevalence and severity of disabilities between the 4 major NMD groups, as defined by Rowland (15). Finally, it would be interesting to investigate the impact of the broad range of NMDIP-related disabilities on HRQoL of patients with neuromuscular diseases.

We considered the possibility of undertaking group invariance testing; however, the sub-groups are relatively small and will affect the test of Differential Item Functioning. We therefore suggest further examination of the factor structure in a new sufficient sample.

In clinical practice, especially in multidisciplinary rehabilitation teams, the NMDIP may contribute to better understanding the patients' health problems when used as an assessment tool. Although positive results were found in the feasibility studies with the preliminary NMDIP and the MSIP, it is advisable to combine this application with research; for example, in order to investigate the effects on the healthcare plan when using the NMDIP.

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REFERENCES

1. Phillips M, Flemming N, Tsintzas K. An exploratory study of physical activity and perceived barriers to exercise in ambulant people with neuromuscular disease compared with unaffected controls. Clin Rehabil 2009; 23: 746–755.
2. Marchettini P, Lacerenza M, Mauri E, Marangoni C. Painful peripheral neuropathies. Curr Neuropharmacol 2006; 4: 175–181.
3. Bos I, Stallinga HA, Middel B, Kuks JBM, Wynia K. Validation of the ICF core set for neuromuscular diseases. Eur J Phys Rehabil Med 2013; 49: 179–187.
4. Graham C, Rose M, Grunfeld E, Kyle S, Weinman J. A systematic review of quality of life in adults with muscle disease. J Neurol 2011; 258: 1581–1592.
5. Peric' S, Rakocevic-Stojanovic V, Stevic Z, Basta I, Pavlovic S, Vujanac V, et al. Health-related quality of life in patients with myotonic dystrophy type 1 and amyotrophic lateral sclerosis. Acta Neurol Belg 2010; 110: 71–77.
6. Smith PC, Papanicolas I. Health system performance comparison: an agenda for policy, information and research. Copenhagen, Denmark: WHO, Regional Office for Europe; 2012.
7. Ware JE Jr, Sherbourne CD. The MOS 36-item short-form health survey (SF-36). I. Conceptual framework and item selection. Med Care 1992; 30: 473–483.
8. Jenkinson C, Fitzpatrick R, Brennan C, Swash M. Evidence for the validity and reliability of the ALS assessment questionnaire: the ALSAQ-40. Amyotroph Lateral Scler Other Motor Neuron Disord 1999; 1: 33–40.
9. Kempen GI, Miedema I, Ormel J, Molenaar W. The assessment of disability with the Groningen Activity Restriction Scale. Con-

ceptual framework and psychometric properties. Soc Sci Med 1996; 43: 1601–1610.
10. Cardol M, de Haan RJ, van den Bos GA, de Jong BA, de Groot IJ. The development of a handicap assessment questionnaire: the Impact on Participation and Autonomy (IPA). Clin Rehabil 1999; 13: 411–419.
11. World Health Organization (WHO). International Classification of Functioning, Disability and Health (ICF). Geneva: World Health Organization; 2001.
12. Stucki G, Ustun TB, Melvin J. Applying the ICF for the acute hospital and early post-acute rehabilitation facilities. Disabil Rehabil 2005; 27: 349–352.
13. Stucki G, Cieza A, Rauch A, Hoogland-Eriks I, Brach M, editors. Case studies: translating interventions into real-life gains – a rehab cycle approach. CH-6207 Nottwil, Switzerland: Swiss Paraplegic Research; 2007.
14. Wynia K, Middel B, van Dijk JP, De Ruiter H, De Keyser J, Reijneveld SA. The Multiple Sclerosis Impact Profile (MSIP). Development and testing psychometric properties of an ICF-based health measure. Disabil Rehabil 2008; 30: 261–274.
15. Rowland LP, McLeod JG. Classification of neuromuscular disorders. J Neurol Sci 1994; 124: 109–130.
16. Kuyken W, Orley J, Herrman H, Schofield H, Power M. The World Health Organization Quality of Life assessment (WHOQOL): Development and general psychometric properties. Social Sci Med 1998; 46: 1569–1585.
17. Cardol M, de Haan RJ, de Jong BA, van den Bos GA, de G I. Psychometric properties of the Impact on Participation and Autonomy Questionnaire. Arch Phys Med Rehabil 2001; 82: 210–216.
18. Hobart JC, Riazi A, Lamping DL, Fitzpatrick R, Thompson AJ. Improving the evaluation of therapeutic interventions in multiple sclerosis: development of a patient-based measure of outcome. Health Technol Assess 2004; 8: iii, 1–iii, 48.
19. Van Sonderen E. [How to handle missing data in particular scale items]. Verpleegkunde, Nederlands-Vlaam wetenschappelijk tijdschrift voor verpleegkundigen 2000; 15: 104–111 (in Dutch).
20. Muthén L, Muthén B. Mplus user's guide (1998–2010). 6th edn. Los Angeles, CA: Muthén & Muthén; 2010.
21. Flora DB, Curran PJ. An empirical evaluation of alternative methods of estimation for confirmatory factor analysis with ordinal data. Psychol Methods 2004; 9: 466–491.
22. Hu L, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: conventional criteria versus new alternatives. Structural Equation Modeling 1999; 6: 1–55.
23. Muthen B. Mplus Technical appendices (1998–2004). Los Angeles, CA: Muthen & Muthen; 2004.
24. Van der Heijden GJMG, Donders ART, Stijnen T, Moons KGM. Imputation of missing values is superior to complete case analysis and the missing-indicator method in multivariable diagnostic research: a clinical example. J Clin Epidemiol 2006; 59: 1102–1109.
25. Clark LA, Watson D. Constructing validity: basic issues in objective scale development. Psychol Assess 1995; 7: 309–319.
26. Eisen M, Ware JE, Donald CA, Brook RH. Measuring components of children's health status. Med Care 1979; 17: 902–921.
27. Piedmont RH, Hyland ME. Inter-item correlation frequency distribution analysis: a method for evaluating scale dimensionality. Educ Psychol Meast 1993; 53: 369–378.
28. Nunnally JC. Bernstein IH, editor. Psychometric theory. Third edn. New York: Mcgraw-Hill, Inc.; 1994.
29. Cortina J. What is coefficient alpha? An examination of theory and applications. J Appl Psychol 1993; 78: 98.
30. Polit DF, Beck CT. editor. Nursing research: principles and methods. Philadelphia, New York, Baltimore: Lippincott; 2004.
31. Streiner DL, Norman GR. Health measurement scales, a practical guide to their development and use. Fourth edition. Oxford: Oxford University Press; 2008.
32. Cohen J. Statistical power analysis. New York: Academic Press; 1988.
33. Andersen M, Johnson U, Lindwall M, Ivarsson A. To adjust or not adjust: nonparametric effect sizes, confidence intervals, and real-world meaning. Psychol Sport Exerc 2013; 14: 97–102.
34. Cohen J. Statistical power analysis for the behavioural sciences. 2nd edn. Hillsdale, NJ: Lawrence Erlbaum Associates; 1988.
35. Wynia K, Middel B, van Dijk JP, De Ruiter H, Lok W, De Keyser JH, et al. Broadening the scope on health problems among the chronically neurologically ill with the International Classification of Functioning (ICF). Disabil Rehabil 2006; 28: 1445–1454.
36. Terwee CB, Bot SD, de Boer MR, van der Windt DA, Knol DL, Dekker J, et al. Quality criteria were proposed for measurement properties of health status questionnaires. J Clin Epidemiol 2007; 60: 34–42.

APPENDIX I. Neuromuscular Disease Impact Profile (NMDIP). (Final 36-item version)

NMDIP		Body functioning questions
Scale		Response options 0 = no, not at all 1 = yes, I have a slight impairment 2 = yes, I have a moderate impairment 3 = yes, I have a severe impairment 4 = yes, I have a complete impairment
MuF	b1	Do you face loss of your muscle power functions? (b730)
MuF	b2	Do you face loss of muscle endurance functions? (b740)
MoF	b3	Do you face loss of control of voluntary movements? (b760)
MoF	b4	Do you face involuntary movements? (e.g. tremors or tics) (b765)
MoF	b5	Do you face muscle stiffness or muscle spasm? (b7800/b7801)
SSF	b6	Do you face impairment in your speech functions? (b320)
SSF	b7	Do you face impairment in your swallowing functions? (b5105)
ERF	b8	Do you face impairment in your defecation functions? (e.g. changes in frequency, constipation, incontinence) (b525)
ERF	b9	Do you face impairment in your urination functions? (e.g. frequency of urination, incontinence, difficulties with urination) (b620)
ERF	b10	Do you face limitations in sexual functions? (b640)

APPENDIX I. Contd.

NMDIP		Body functioning questions
MFP	b11	Do you face impairment in your sleep functions? (e.g. onset of sleep, the maintenance of sleep or the quality of sleep) (b134)
MFP	b12	Do you experience fatigue? (b1300/b455)
MFP	b13	Do you face changes in your emotional functions? (e.g. fear, depression, happiness) (b152)
MFP	b14	Do you face changes in your thought functions? (e.g. the ability to think logically, the ability to memorize, the ability to concentrate) (b160)
MFP	b15	Do you experience sensation pain? (b280)
single	b16	Do you face impairment in your seeing functions? (With eyeglasses on or item lenses in) (b210)
NMDIP		Activities questions
Scale		Response options 0=No 1=Yes, but assistance devices and/or adaptations are not necessary 2=Yes, and assistance devices and/or adaptations are necessary 3=Yes, and assistance devices and/or adaptations and another person's help are necessary
AMA	a1	Do you face limitations in changing your body position? (e.g. moving from lying down to standing up or from standing to sitting) (a410)
AMA	a2	Do you face limitations in maintaining your body position? (e.g. maintaining kneeling, standing, and sitting postures) (a415)
AMA	a3	Do you face limitations in transferring yourself? (e.g. moving from a chair into bed; from a wheelchair into a car) (a420)
AMA	a4	Do you face limitations in walking? (a450)
AMA	a5	Do you face limitations in using transportation? (a470)
AMA	a6	Do you face limitations in activities you would like to undertake for recreation and leisure? (a920)
SDA	a7	Do you face limitations in your fine hand use? (e.g. picking up small objects; manipulating a keyboard) (a440)
SDA	a8	Do you face limitations in your arm(s) and hand(s) use? (e.g. pulling or pushing objects; turning or twisting knobs or handles; reaching for kitchen cupboard) (a445)
SDA	a9	Do you face limitations in washing yourself? (a510)
SDA	a10	Do you face limitations in caring for body parts? (e.g. brushing teeth, clipping your nails, combing your hair, shaving) (a520)
SDA	a11	Do you face limitations in toileting? (a530)
SDA	a12	Do you face limitations in dressing yourself? (a540)
SDA	a13	Do you face limitations in preparing meals? (a630)
SDA	a14	Do you face limitations in doing housework? (a640)
NMDIP		Participation questions
Scale		Response options 0=no 1=Yes, as a consequence I have some trouble with …. 2=Yes, as a consequence I have trouble with.. 3=Yes, as a consequence I have a lot of trouble with … 4=Yes, as a consequence …. is (nearly) impossible
PLS	p1	Are there obstacles in your environment that complicate your participation in community, recreation, and leisure? (e.g. accessibility of clubs or associations) (p910/p920)
PLS	p2	Are there obstacles in your environment that complicate the maintenance of your relationships with your closest family, friends, or relatives? (e.g. the travel distance, the attitude of others) (p740–p760)
PLS	p3	Are there obstacles in your environment that complicate your mobility inside or outside your home? (e.g. thresholds; curbs; absence of elevators) (p460/470)
NMDIP		Environmental factors questions
Scale		Response options 0=Yes, very supportive; 1=Yes, somewhat supportive; 2=No, not supportive
Single Item	e1	Is your relationship with your immediate family supportive for you? (e.g. partner, children, parents, brothers, sisters) (e310)
Single Item	e2	Are the social security services supportive for you? (e.g. income support) (e570)
Single Item	e3	Are the health services supportive for you? (e.g. medical and nursing care) (e580)

MuF: Muscle functions; MoF: Movement functions; SSF: Swallowing and Speech functions; ERF: Excretion and Reproductive functions; MFP: Mental functions and Pain; AMA: Activities of moving around; SDA: Self-care and domestic activities; PLS: Participation in life situations.