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ORIGINAL ARTICLE

Motor Function Measure: Validation of a Short Form for Young Children With Neuromuscular Diseases

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Abstract

Objective: To validate a useful version of the Motor Function Measure (MFM) in children with neuromuscular diseases aged <7 years old.

Design: Two prospective cohort studies that documented the MFM completion of children aged between 2 and 7 years old.

Setting: French-speaking rehabilitation departments from France, Belgium, and Switzerland.

Participants: Healthy children (n=194) and children with a neuromuscular disease (n=88).

Interventions: Patients were rated by the MFM either once or twice by trained medical professionals, with a delay between the 2 MFMs ranging between 8 and 30 days.

Main Outcome Measure: Intra- and interrater reliability of the MFM.

Results: The subtests making up the MFM-32, a scale monitoring severity and progression of motor function in patients with a neuromuscular disease in 3 functional domains, were carried out in healthy children aged 2 to 7 years. Twenty items of the MFM-32 were successfully completed by these children and were used to constitute the MFM-20. Principal component analysis of the MFM-20 confirmed the 3 functional domains. Inter- and intrarater reliability of the 3 subscores and total score were high (intraclass correlation coefficient >.90), and discriminant validity was good.

Conclusions: The MFM-20 can be used as an outcome measure for assessment of motor function in young children with neuromuscular disease.

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Neuromuscular diseases include a large number of conditions affecting muscles and the peripheral nervous system. These diseases differ widely in their natural history and clinical features. In terms of motor function, in order to measure the impact of new treatments objectively, it is crucial to know the natural history of each disease, particularly for severe diseases, such as Duchenne muscular dystrophy (DMD) and spinal muscular atrophy (SMA). Ideally, measurement tools should be validated for the age at which the diagnosis is made.

Evaluation of motor function is complementary to measurement of muscle strength.[1,2] Different scales (validated or not) have been developed to quantify motor function in specific pathologies (eg, Hammersmith Motor Ability Score[3] for DMD and Functional Motor Scale[4] for SMA), by region (eg, Brooke et al[5] score for the upper limbs and Vignos et al[6] score for the lower limbs), and by functional limitation (eg, the Egen Klassification scale for non-ambulant DMD).

The Motor Function Measure (MFM)[7] is a tool designed to monitor precisely the severity and progression of motor function in most neuromuscular diseases. It is applicable to all degrees of disease severity, in both ambulant and nonambulant patients. Items are rated on a 4-point Likert scale and are grouped into subscores (determined after factor analysis during the validation study) assessing 3 functional areas: standing position and transfers (the D1 subscore; 13 items), axial and proximal motor function (D2; 12 items), and distal motor function (D3; 7 items). The MFM has been validated in terms of reproducibility, construct validity, and concurrent validity in patients aged between 6 and 60 years old with one of the principal neuromuscular diseases.[7]

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Its sensitivity to change has been also assessed at a 1-year interval: a good correlation between MFM scores and changes reported by patient or investigator was shown.[8,9] French and English versions have been available since 2006, a Spanish version since 2007, and Portuguese, Dutch, Italian, Danish, German, Arab, and Turkish versions are currently available or undergoing translation (available at: http://www.mfm-nmd.org). It is or has been used as an outcome measure in a variety of studies, such as a controlled trial of riluzole versus placebo in SMA (ASIRI study, ClinicalTrials Identifier No.: NCT00774423), the safety and efficacy study of olesoxime (TRO19622) (TROPHOS study, ClinicalTrials Identifier No.: NCT01302600), an observational study of steroids in DMD, the French Pompe disease registry (natural history and evaluation of enzyme replacement therapy), and the Genethon calpainopathy natural history study.

For assessment of a younger patient with a neuromuscular disease, some teams use motor scales specific for 1 condition, such as the Test of Infant Motor Performance[10] or the Hammersmith Functional Motor Scale (HFMS, original or modified/expanded version) in SMA[11-15] or the North Star Ambulatory Assessment in DMD.[16,17] Others tests are used in SMA, but have not been fully validated for such use, such as the Gross Motor Function Measure (validated for cerebral palsy).[18]

For clinical trials involving young children, the standard MFM (MFM-32) is not well suited as an outcome measure: the completion time is long, and achievement of some items is too difficult in terms of children's cognitive and motor development. The scale has not been validated for patients <6 years old, and the number of children between 6 and 7 years old in the initial study was low.

The aim of the present study was therefore to construct and validate a short form of the MFM that is suitable for children <7 years old. The first step was the construction of a reduced scale by testing young healthy children with the standard MFM-32 and removing items inappropriate in terms of cognitive and motor development. The second phase was assessment of this short form in a population of 2- to 7-year-old children with one of the main neuromuscular diseases through a validation process.

Methods

Development of the MFM for young children

In order to identify and remove items from the MFM-32 not suitable for young children in terms of their cognitive and motor development, healthy children were subjected to the MFM-32.

Four French rehabilitation centers from different universities hospitals (Lyon, Montpellier, Nice, and Saint Etienne) took part in this phase. Evaluations were performed by physiotherapists trained and experienced in the MFM within 4 schools (nursery and elementary schools). The criteria for patient inclusion were: aged between 2 and 7 years and no cognitive or neuromuscular disorder. Noninclusion criteria were the following: psychomotor delay, recent surgery, or sensory disability.

Items of the MFM-32 were not considered suitable for use in young children if <80% of subjects obtained the maximum score of 3 points on the item, or if completion of the item could not be graded clearly according to the manual.

List of abbreviations:

ANOVA	analysis of variance
CGI	Clinical Global Impression
CMT	Charcot-Marie-Tooth disease
DMD	Duchenne muscular dystrophy
HFMS	Hammersmith Functional Motor Scale
ICC	intraclass correlation coefficient
MFM	Motor Function Measure
SMA	spinal muscular atrophy
VAS	visual analog scale

Validation of the short version of the MFM

Setting and participants

A total of 13 French-speaking investigating centers (10 in France, 2 in Belgium, and 1 in Switzerland) were involved in the validation study. Five took part in the interrater reliability phase, 7 in the intrarater reliability phase, and 1 in both phases. At least 1 physician and 1 or 2 physiotherapists previously trained and certified for the MFM were appointed to the study at each center. Before the start of the study, an informational meeting was organized in each investigating center in order to explain the protocol and provide the information and material required for optimal MFM completion.

Children had to be between the ages of 2 and 7 at inclusion, have the ability to understand and speak sufficiently to perform the requested tasks, and have pathology from 1 of the following groups, with a suspected (molecular biology ongoing) or confirmed diagnosis: progressive muscular dystrophy (DMD, Becker muscular dystrophy, or other), spinal muscular atrophy, Charcot-Marie-Tooth disease (CMT), congenital muscular dystrophy, congenital myopathy, or myotonic dystrophy. Noninclusion criteria were the following: recent disease or surgery affecting motor function. Patients were included between April 2008 and March 2009.

Structure of the short-version MFM

The 20 items of the short version of the MFM, named the MFM-20, are listed in table 1. Test positions include lying, sitting, and standing. Scoring is on a 4-point Likert scale based on the subject's best abilities without assistance: 0 (does not initiate movements or starting position cannot be maintained); 1 (partially completes the exercise); 2 (completes the exercise with compensation, slowly, or with obvious clumsiness); and 3 (completes the exercise in the standard pattern). The total score ranges from 0 to 60 when summing the 20 items. The total score and subscores are expressed as a percentage of the maximum possible score.

Visits and data collection

Physicians screened patients for trial eligibility on the day of inclusion and submitted patients to a medical interview. Data collected during this interview concerned history of ambulation, previous surgery, and ongoing medications.

A physiotherapist performed an initial evaluation using: (1) Brooke,[5] Vignos,[6] and colleagues grades; (2) 100mm visual analog scales (VASs) of severity of disability (assessed globally and for the motor function domains of standing position and transfers, axial and proximal motor functions, and distal motor function)[19]; (3) Clinical Global Impression (CGI) of motor disability (mild, moderate, severe, or very severe)[20]; and (4) the MFM-20. Concerning the MFM-20 assessment, duration of the scale completion, assessment of subject cooperation (nil, moderate, or optimal), and assessment of patients' fatigue were collected.

Patients participating in the inter- and intrarater reliability study phase underwent a second visit, which took place 8 to 30 days after the first visit. During this visit, they were assessed using the MFM-20 by the same physiotherapist (intrarater reliability study) or by another physiotherapist (interrater reliability study).

Table 1 Thirty-two items of the MFM-32 with starting positions and requested actions

No.	Domain	Starting Position	Exercise Requested	% of Maximum Score Obtained by Healthy Children
1	D2	Supine, head in midline position	Holds the head for 5s in midline position and turns it completely from one side to the other	89.2
2*	D2	Supine	Raises the head and maintains the raised position for 5s	80.0
3	D2	Supine	Flexes the hip and knee >90° by raising the foot from the mat	95.4
4	D3	Supine, leg supported by examiner	From plantar flexion, dorsiflexes the foot to at least 90° in relation to the leg	95.9
5	D2	Supine	Raises the hand and moves it to the opposite shoulder	94.9
6	D1	Supine, lower limbs half-flexed, kneecaps at the zenith, and feet resting on the mat	Raises the pelvis; the lumbar spine, the pelvis and the thighs are aligned and the feet slightly apart	90.8
7	D2	Supine	Turns over into prone and frees both upper limbs from under the body	95.9
8*	D1	Supine	Without upper limb support sits up	31.3
9	D2	Seated on the mat	Without upper limb support, maintains the seated position for 5s and is then capable of maintaining contact between the 2 hands for 5s	96.9
10	D2	Seated on the mat, the tennis ball placed in front of the subject	Without upper limb support, leans forward, touches the ball, and sits back again	97.9
11	D1	Seated on the mat	Stands up without upper limb support	91.3
12	D1	Standing	Without upper limb support, sits down on the chair with the feet slightly apart	96.4
13*	D2	Seated on the chair	Without upper limb support or leaning against the back of the chair, maintains the seated position for 5s, with the head and trunk in midline position	93.3
14	D2	Seated on the chair or wheelchair, head in flexion	Raises the head from the flexed position, the head stays aligned throughout the movement and is maintained raised in midline position for 5s	90.3
15*	D2	Seated on the chair or wheelchair, forearms on the table, but not elbows	Places both hands on top of the head at the same time while the head and trunk remain in midline position	93.3
16*	D2	Seated on the chair or wheelchair, pencil on the table	Without moving the trunk, reaches the pencil with 1 hand, forearm and hand off the table with the elbow in full extension at the end of the movement	76.9
17*	D3	Seated on the chair or wheelchair, 10 coins on the table	Successively picks up and holds 10 coins in 1 hand during the 20-s period	65.6
18	D3	Seated on the chair or wheelchair, 1 finger placed in the center of the fixed compact disk	Goes round the edge of the compact disk with 1 finger without contact of the hand with the table	87.7
19*	D3	Seated on the chair or wheelchair, pencil on the table	Picks up the pencil and draws a continuous series of loops inside the frame and over its full length touching the top and bottom line of the frame	8.7
20*	D3	Seated on the chair or wheelchair, holding the sheet of paper	Tears the sheet of paper folded in 4, beginning from the fold edge	20.0
21	D3	Seated on the chair or wheelchair, tennis ball on the table	Picks up the ball and holding the ball turns the hand over completely	91.3
22	D3	Seated on the chair or wheelchair, 1 finger placed in the center of the diagram	Raises the finger and places it successively on the squares of the diagram without touching the lines	74.2

Table 1 (*continued*)

No.	Domain	Starting Position	Exercise Requested	% of Maximum Score Obtained by Healthy Children
23	D2	Seated on the chair or wheelchair, upper limbs along the trunk	Places the 2 forearms and/or the hands on the table at the same time	98.5
24	D1	Seated on the chair	Stands up without upper limb support and with the feet slightly apart	96.4
25	D1	Standing with upper limb supported	Releases the support and maintains a standing position for 5s with the feet slightly apart, the head, trunk, and limbs in the midline position	96.9
26*	D1	Standing with upper limb support on equipment	Without upper limb support, raises the foot for 10s	32.3
27	D1	Standing	Without support, touches the floor with 1 hand and stands up again	98.5
28*	D1	Standing without support	Takes 10 steps forward on both heels	80.0
29*	D1	Standing without support	Takes 10 steps forward on a line	64.1
30	D1	Standing without support	Runs for 10m	91.3
31*	D1	Standing on 1 foot without support	Hops 10 times in place	36.9
32	D1	Standing without support	Without upper limb support, manages to squat and gets up twice in a row	91.8

NOTE. The percentages of maximum score obtained by healthy children are given.
Items 2, 13, and 15 were not accurately assessed according to the criteria from the reference manual.
* Items not suitable for use with young children.

Statistical methods

Quantitative variables were described as means, SD, and ranges. Categorical variables were reported as frequencies or percentages.

Principal component analysis was performed using Kaiser criterion (eigenvalue >1) as an extraction, followed by a varimax rotation to obtain independent domains.

Inter- and intrarater reliability of each item of the MFM-20 were assessed with Cohen kappa agreement coefficients and that of total scores and subscores (derived from factor analysis) with intraclass correlation coefficients (ICCs) computed with a 1-way random effect analysis of variance (ANOVA) model. These coefficients were interpreted as poor (<0.4), moderate (0.4−0.6), good (0.6−0.8), and excellent (>0.8).[21] A coefficient ≥.40 was considered acceptable for individual items.

The internal consistency of the overall scale and subscales was measured by Cronbach alpha coefficients. A coefficient ≥.70 was considered acceptable.

Convergent validity was assessed by Pearson correlation coefficients between MFM-20 scores and other clinical measures.

Discriminant validity was assessed by comparing scores of the scale according to CGI severity, ambulatory status, and diagnosis. Comparisons were made using ANOVA, followed by *t* tests for pairwise comparisons in case of significance.

P values for significance were set at 5%. Statistical analysis was performed using the software BMDP[a] and SPSS version 18.0.[b]

Ethical consideration

The study was approved by an ethics committee (Comité de Protection des Personnes Lyon Sud-Est II).

Written and oral information on the study's objectives, methodology, duration, and expected benefits were given to children and their parents, and written consent was collected from parents.

Results

Development of the MFM for young children

A total of 194 healthy children aged 2 to 7 years participated in the creation phase. Four infants aged 2 and 3 years old who did not want to carry out some of the 32 items were excluded. The analysis was then carried out with 190 children, 98 boys and 92 girls (mean age ± SD, 4.5±1.2y) (table 2). The average completion time ± SD was 18.4±6.3 minutes for the 32 items (range, 9−41min). There was no significant correlation between completion time and age of the subjects. Cooperation was rated as optimal in 88% of cases and moderate in 12%.

The maximum score was not achieved by 80% of subjects in 10 items (items 8, 16, 17, 19, 20, 22, 26, 28, 29, 31), and scores for 3 items (items 2, 13, 15) could not be assessed accurately according to the criteria given in the reference manual (see table 1). Concerning item 22, which was successfully completed by 74.2% of participants, it was further decided not to remove it, and some adaptations were made in the user manual in order to facilitate the comprehension of the requested task for young children. Twelve items of the MFM-32 were finally removed, leaving an MFM scale of 20 items (see table 1). Their distribution among the 3 subscales derived from the standard MFM-32 was approximately maintained: 8 items in subscale D1 (standing and transfers), 8 items in subscale D2 (axial and proximal motor function), and 4 items in subscale D3 (distal motor function).

Table 2 Characteristics of children participating in the development of the MFM for young healthy children (n=190) and in the MFM-20 validation study (n=88)

	Development of the MFM for Young Children		MFM-20 Validation Study	
Age Range (y)	No. of Patients	B/G	No. of Patients	B/G
2–3	19	11/8	8 (0 IaR, 7 IeR)	4/4
3–4	41	21/20	15 (3 IaR, 8 IeR)	9/6
4–5	72	41/31	23 (3 IaR, 9 IeR)	16/7
5–6	33	12/21	19 (5 IaR, 7 IeR)	16/3
6–7	25	13/12	23 (8 IaR, 5 IeR)	16/7
Total	190	98/92	88 (19 IaR, 36 IeR)	61/27

Abbreviations: B, boy; G, girl; IaR, intrarater reliability study; IeR, interrater reliability study.

Validation of the short version of the MFM

Population studied

A total of 88 children participated in the validation study of the MFM-20, and none were excluded from the statistical analysis. Their mean age ± SD was 4.8±1.3 years (range, 2.0–6.8y) (see table 2). The study consisted of 30.7% girls and 69.3% boys. Boys with DMD (n=29) represented one third of the sample. SMA was the second highest group by number (n=22), followed by CMT (n=13). Other pathologies included congenital myopathy or muscular dystrophy (n=17), Becker muscular dystrophy (n=3), limb-girdle muscular dystrophy (n=3), and Steinert disease (n=1). Nine DMD patients were taking steroids, and 1 SMA patient was receiving valproic acid. Of the participants, 77% had acquired ambulation at a mean age ± SD of 17.8±4.4 months (range, 11–30mo). A few (4%) had lost the ability to walk. According to the physicians' CGI, 40% had mild disability, 28% had moderate disability, 24% had severe disability, and 7% had very severe disability.

Scale completion

It took on average ± SD 26±8.5 minutes (range, 12–50min) to complete the MFM-20 scale. There was no significant correlation between completion time and age of subjects. Cooperation was rated as optimal in 60% of cases and moderate in 38%. For 2 infants (2%), cooperation was low throughout the tests. Cooperation did not relate to age or type of pathology.

Factor analysis and internal consistency

Item loadings obtained by factor analysis were compared for each item with its assigned subscale (D1, D2, or D3) in the original MFM-32 scale (table 3). Only 3 items did not conform to their initial

Table 3 Summary of the construct validity (factor analysis and reliability) of items in the MFM-20

				Reliability (Cohen κ coefficients)	
Subscale (% variance explained) Item No.	D1 (34%)	D2 (24%)	D3 (13%)	Intrarater (n=17)	Interrater (n=34)
1		0.71		0.70	.64
3		0.61		1.00	.46
4			0.61	0.68	.75
5		0.42*	0.61	0.60	.64
6	0.74			0.82	.67
7		0.71		0.80	.55
9		0.83		1.00	.70
10		0.76		1.00	.81
11	0.87			0.71	.90
12	0.87			0.91	.69
14		0.74		1.00	.22
18			0.57	0.56	.59
21		0.59	0.40*	0.85	.51
22			0.64	0.74	.64
23		0.24*	0.75	1.00	.37
24	0.88			0.83	.74
25	0.78			1.00	.77
27	0.87			0.92	.85
30	0.87			1.00	.71
32	0.77			0.70	.94
ICCs	0.99	0.99	0.91		
Cronbach α coefficients	0.96	0.90	0.69		
Mean scores ± SD (%), range	56.4±34.8, 4.2–100.0	89.0±18.8, 12.5–100.0	85.4±17.9, 33.3–100.0		

Abbreviations: D1, standing position and transfers; D2, axial and proximal motor function; D3, distal motor function.
* Factor loading in the subscale in which the item is assigned in the MFM-32 (see text).

subscale: item 5 correlated better with D3 than with D2 (.61 and .42, respectively), item 21 correlated better with D2 than with D3 (.59 and .40, respectively), and item 23 was strongly related to D3 (.75 vs .24 with D2). Nevertheless, it was decided for continuity and clinical reasons to maintain these 3 items in their respective subscales.

Cronbach alpha coefficients calculated with items in their initial subscales (item 5 in D2, item 21 in D3, and item 23 in D2) were above 0.7 for all scores, except D3 (.97, .96, .90, .69 for total score, D1, D2, and D3, respectively).

Convergent validity

We found good correlation of the MFM-20 total score (range, .82−.86) with the severity of disability evaluated by physicians with the CGI and VAS assessing overall disability (fig 1, table 4). D1 (standing position and transfers) was highly correlated with the Vignos grade (−.91) and the corresponding VAS (−.86). D2 (axial and proximal limb motor function) correlated well with the Brooke grade (.74) and the corresponding VAS (−.72). Correlations of the D3 score with other measures were only moderate (−.56 with its corresponding VAS). Moderate correlations were found between MFM scores and age (range, .39−.45).

Discriminant validity

There was a strong inverse correlation between the severity of disability, as assessed by physicians, and the total score ($P<.001$) (see fig 1) and the 3 subscores (not shown). As well, the total score and 3 subscores allowed good discrimination between the 5 principal diagnostic groups (DMD, SMA, CMT, congenital myopathy, and congenital muscular dystrophy; $P<.01$) (fig 2). In this age group, SMA patients (mainly nonambulant children) showed the most impaired scores, whereas those with DMD were less severely affected, as expected at this early stage. Lastly, D1, D2, and total score discriminated well between the 66 ambulant and 22 nonambulant patients ($P<.001$ for all scores) (fig 3).

Reliability

For interrater reliability (n=36) (see table 3), Cohen kappa coefficients were acceptable to excellent (range, .37−.94) for all items except item 14 (.22). For intrarater reliability (n=19) (see table 3), kappa coefficients were acceptable to excellent (.56 to 1.0) for all items. All intra- and interrater agreement coefficients (ICC) were excellent for the total score (.99) and the 3 subscores (range, .91−.99).

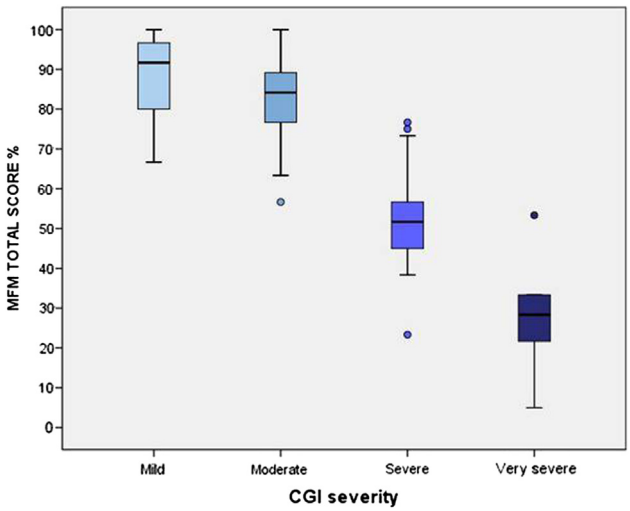

Fig 1 CGI severity versus MFM-20 total score.

Table 4 Pearson correlation coefficients between MFM-20 scores (subscores D1, D2, and D3 and the total score) and physicians' grading of disability (corresponding to each domain and overall disability) on a 100mm VAS, and Vignos[6] and Brooke[5] grades for upper and lower limb deficit, respectively

Scores	VAS	Vignos Grade	Brooke Grade
Subscore D1	−.86	−.91	−.76
Subscore D2	−.72	−.74	−.83
Subscore D3	−.56	−.49	−.65
Total score	−.86	−.89	−.85

Discussion

In order to propose a valid outcome measure to assess the motor capacities in patients with any neuromuscular disease, our group constructed and validated the MFM-32, a generic functional rating scale evaluating severity and progression of motor function in neuromuscular patients from 6 years onward, and applicable whatever the severity of deficiencies, for example both for ambulant and nonambulant patients.[7,8] Its interest in the neuromuscular domain is underlined by its use in several clinical trials (eg, TROPHOS and ASIRI studies) and for the study of the natural history of some diseases.[22,23] However, recent progress made both in terms of research and clinical care of neuromuscular diseases highlights the need for a valid scale to describe the effects of treatments in very young children. If some motor scales are used today for younger children in assessments of a particular disease, no generic motor scale is yet available.[10-18] For its use in the follow-up of patients with a neuromuscular disease, and especially for children older than 6 years old, items of the MFM-32 were designed to be easy and enjoyable, and to need only a minimal level of comprehension. However, one could expect that, because of children's cognitive and motor development, some items would not be suitable for very young children. When applied to 190 healthy children between 2 and 7 years old, although some items could not be correctly performed, particularly those involving fine motor skills and balance, a substantial proportion of items (20 of the 32 items of the MFM-32) was successfully completed (ie, a substantial number of children obtained the score of 3 for these items). These 20 items were collected in a new scale, named the MFM-20, which is a candidate for a generic scale useful for younger children. The psychometric properties of this reduced scale, as assessed in this first phase validation study, are satisfactory.

Items for the MFM-20 were well accepted by neuromuscular patients and their therapists, and no issues of fatigability were reported even among patients with severe SMA. The standard equipment used for the MFM-32 (adult scale) can be used with minor modifications. Time of completion was shorter than for the MFM-32 (average of 26min vs 36min), thus being closer in duration to shorter scales, such as the modified HFMS,[11] which takes 15 to 30 minutes to complete. Factor analysis produced 3 clinical domains close to those obtained for the MFM-32 scale in older patients. Three items (2 in D2 and 1 in D3) could have been reassigned but were maintained for continuity and clinical relevance.

Some lessening of precision might be expected with scale shortening; in particular, one could expect that the most difficult items of the MFM-32 would be removed, making the scale more suitable for assessment of less impacted patients. We analyzed 3378 MFM results from adult or children patients with neuromuscular

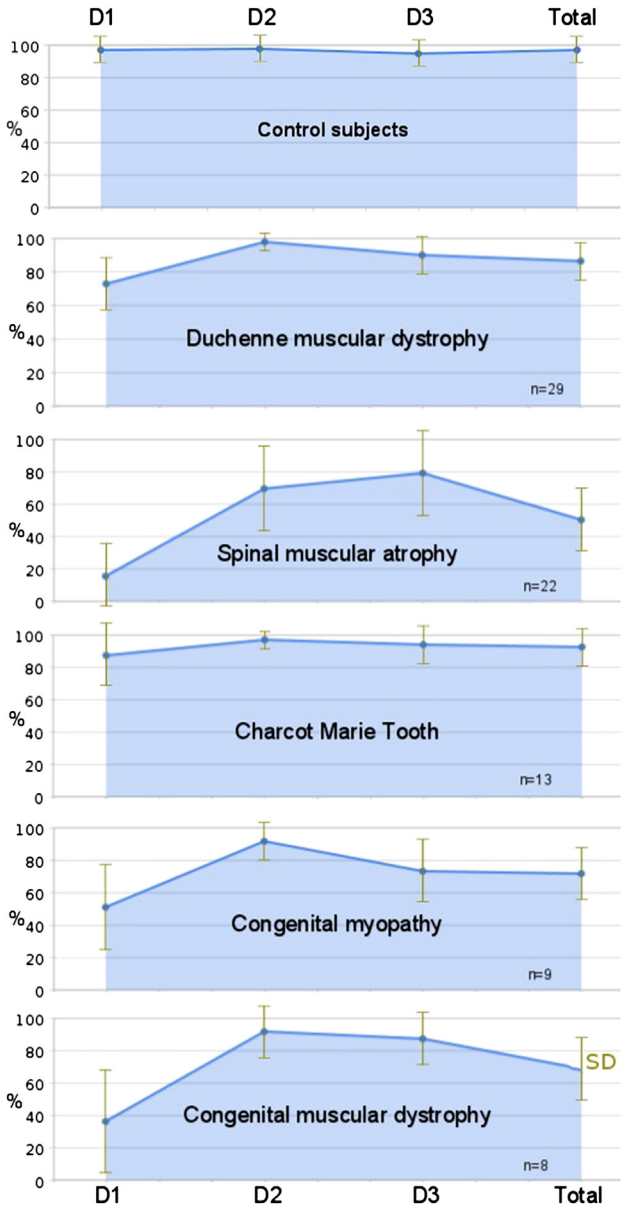

Fig 2 MFM-20 subscores and total scores (expressed as percent of maximal possible score) in controls and in patients with 5 neuromuscular conditions. Abbreviations: D1, standing position and transfers; D2, axial and proximal motor function; D3, distal motor function.

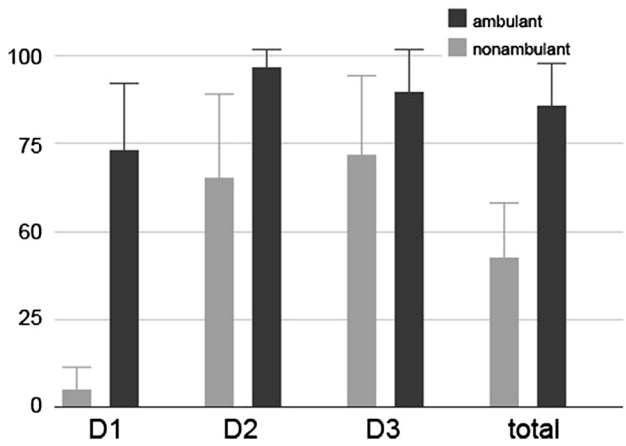

Fig 3 Total and dimensional scores of the MFM-20 expressed as percent of maximal possible score according to ambulant or nonambulant populations: $P<.001$ for all scores. Abbreviations: D1, standing position and transfers; D2, axial and proximal motor function; D3, distal motor function.

disorders collected in an online database (for more information about the database, see www.mfm-nmd.org). We found that some of the most difficult items, determined according to the percentage of patients reaching the highest score on the item, were not removed from each of the 3 functional domains (eg, items 30 and 11 for D1, items 7 and 15 for D2, and item 4 for D3) (data not shown). Moreover, the distribution of items among the 3 functional motor domains remains comparable with that of the MFM-32, with 8 items versus 13 for standing and transfers, 8 versus 12 for axial and proximal motor function, and 4 versus 7 for distal motor function. Our results thus suggest that the MFM-20 scale permits evaluation of motor function of both less and more impacted patients in all 3 functional domains.

Psychometric properties of the reduced scale remain good and close to those of the MFM-32, meeting the criteria set out in the recommendations of the American Psychological Association.[24] Inter- and intrareliabilities were acceptable to excellent, and the total and D1 and D2 scores remain correlated with CGI severity and the Brooke and Vignos grades. Distal motor function (D3) was less well captured than the D1 and D2 domains (Cronbach $\alpha=.69$), but still remains acceptable.

As reported at a Treat-NMD workshop,[2] the MFM can be used in clinical trials in which patients lose ambulation or become significantly weaker over time. The 3 subscales may be treated independently according to clinical or trial focus. As illustrated in figure 2, the subscales characterize the principal motor deficiencies of specific pathologies: standing and transfers in DMD and congenital disorders, along with axial and proximal deficits in SMA. Other more specific scales can be used in conjunction with the MFM, such as Children's Hospital of Philadelphia Infant Test of Neuromuscular Disorders in SMA type I,[25] the six-minute walk test[26] for ambulant patients, or new tools, such as continuous monitoring.[27]

In order to complete the study of the psychometric properties of the MFM-20, a 1-year follow-up study is underway in order to assess its sensitivity to change.

To facilitate the use of the MFM, a user's manual applicable to both the adult and pediatric scales (available in Dutch, English, French, German, Portuguese, and Spanish) is available to download (available at: www.mfm-nmd.org). It provides detailed guidance for the scoring of each item. Nevertheless, practice of the MFM requires rigor. Examiners (physiotherapists, physicians, and other professionals) must be trained and certified, either by attending a day-long training course or through video training, available at the website. Training is essential for the quality of outcome measures, and its importance is emphasized by those who develop other scales.[13]

Study limitations

Although the population included in the validation study reflects the mix of pediatric patients seen in consultation at a rehabilitation department where a predominance of DMD and SMA is observed, a limitation of this validation study includes the relatively low numbers of very young children (2−3y old). Inclusion of a larger

number of patients aged <3 years is difficult, because the diagnosis of some neuromuscular diseases rarely occurs before 3 years of age.

A second limitation of this study is that the item response theory model was not used. Such a model applied to the MFM-20 could lead to a better knowledge of its psychometric properties, further confirming that it can be used as an outcome measure in clinical trials and may solve some of the concerns of the suitability of the items for assessing different neuromuscular diseases in different populations.

Conclusions

The present results suggest that the newly developed MFM-20 is appropriate for use for the follow-up of young patients or in clinical trials. With the availability of this scale, patients can be followed throughout their pediatric course, with a transition from the MFM-20 to the MFM-32 during their seventh year. In our experience, the total score expressed as a percentage provides the best manner to follow progression of the disease across the 2 scales.

Suppliers

a. BMDP Statistical Software, 1964 Westwood Blvd, Ste 202, Los Angeles, CA 90025.
b. SPSS Inc, 233 S Wacker Dr, 11th Fl, Chicago, IL 60606.

Keywords

Disability evaluation; Neuromuscular diseases; Rehabilitation

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