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ORIGINAL ARTICLE

English Cross-Cultural Translation and Validation of the Neuromuscular Score: A System for Motor Function Classification in Patients With Neuromuscular Diseases

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Abstract

Objective: To develop and validate an English version of the Neuromuscular (NM)-Score, a classification for patients with NM diseases in each of the 3 motor function domains: D1, standing and transfers; D2, axial and proximal motor function; and D3, distal motor function.

Design: Validation survey.

Setting: Patients seen at a medical research center between June and September 2013.

Participants: Consecutive patients (N=42) aged 5 to 19 years with a confirmed or suspected diagnosis of congenital muscular dystrophy.

Interventions: Not applicable.

Main Outcome Measures: An English version of the NM-Score was developed by a 9-person expert panel that assessed its content validity and semantic equivalence. Its concurrent validity was tested against criterion standards (Brooke Scale, Motor Function Measure [MFM], activity limitations for patients with upper and/or lower limb impairments [ACTIVLIM], Jebsen Test, and myometry measurements). Informant agreement between patient/caregiver (P/C)-reported and medical doctor (MD)-reported NM scores was measured by weighted kappa.

Results: Significant correlation coefficients were found between NM scores and criterion standards. The highest correlations were found between NM-score D1 and MFM score D1 ($\rho = -.944$, $P < .0001$), ACTIVLIM ($\rho = -.895$, $P < .0001$), and hip abduction strength by myometry ($\rho = -.811$, $P < .0001$). Informant agreement between P/C-reported and MD-reported NM scores was high for D1 ($\kappa = .801$; 95% confidence interval [CI], .701−.914) but moderate for D2 ($\kappa = .592$; 95% CI, .412−.773) and D3 ($\kappa = .485$; 95% CI, .290−.680). Correlation coefficients between the NM scores and the criterion standards did not significantly differ between P/C-reported and MD-reported NM scores.

Conclusions: Patients and physicians completed the English NM-Score easily and accurately. The English version is a reliable and valid instrument that can be used in clinical practice and research to describe the functional abilities of patients with NM diseases.

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According to Rossi et al,[1] neuromuscular diseases (NMDs) encompass "all disorders caused by an abnormality of any component of the lower motor neuron system: anterior horn cell, peripheral nerve, neuromuscular junction, or muscle." In fact, whatever the cause, most NMDs involve impairment and most frequently a progressive decline of motor function.

There is an increasing interest in developing valid outcome measures to assess motor function and describe the effects of interventions especially in multinational trials[2-4]; thus, several functional quantitative scales have been developed. Some are disease-specific (eg, Hammersmith Motor Ability Score,[5] Modified Hammersmith Functional Motor Scale,[6] expanded version of the Hammersmith Functional Motor Scale,[4] North Star Ambulatory Assessment[7]); others, such as the Motor Function Measure (MFM),[8] are designed for all NMDs and applicable regardless of the patient's ambulant status. These quantitative scales measure motor functional capabilities under controlled conditions and are administered by trained physical therapists within 20 to 30 minutes. Nevertheless, several scales lack the sufficient sensitivity to change that would enable detecting the effects of interventions on motor function in clinical trials.

The work by Palisano's team[9] (ie, the Gross Motor Function Classification System [GMFCS] in cerebral palsy) emphasizes clinical severity scales/classifications that describe a patient's functional status. These classification systems are helpful in clinical research to stratify patients of equal abilities, thereby reducing sample variability. Previous functional rating scales developed specifically for individuals with dystrophinopathies did not consider all components of motor function; Brooke[10,11] and Vignos[12] and colleagues grades describe only the upper body function and the ambulatory capacities, respectively.

To address the need for a standardized method to classify the motor functions of patients with NMDs, the Neuromuscular (NM)-Score was first developed and validated in French.[13] Based on a statement by the World Health Organization about the potential influence of personal and environmental factors on the way patients with NMDs manifest their mobility capacities,[9] the NM-Score focuses on motor function performance under typical current circumstances, not under controlled test conditions.

To extend the applicability of the NM-Score to English-speaking countries, the NM-Score was translated and cross-culturally adapted and is here tested for concurrent validity against other established outcome measurements (MFM,[8] ACTIVLIM [activity limitations for patients with upper and/or lower limb impairments],[14] Brooke Scale,[10,11] Jebsen-Taylor Hand Function Test,[15] and selected myometry measurements) in a cohort of patients with congenital muscular dystrophy (CMD). Because this instrument can be equally used by both patients and parents (the most familiar with the patient's performance in daily living), informant agreement between patient-reported and clinician-reported clinical severity scores was also assessed.

List of abbreviations:

ACTIVLIM	**activity limitations for patients with upper and/or lower limb impairments**
CI	**confidence interval**
CMD	**congenital muscular dystrophy**
GMFCS	**Gross Motor Function Classification System**
LAMA2-RD	**laminin alpha 2–related disease**
MD	**medical doctor**
MFM	**Motor Function Measure**
NIH	**National Institutes of Health**
NM	**neuromuscular**
NMD	**neuromuscular disease**
P/C	**patient/caregiver**

Methods

This study is part of a National Institutes of Health (NIH) protocol entitled "Clinical and Molecular Manifestations of Neuromuscular and Neurogenetic Disorders of Childhood" (Clinical Trial Registration No.: NCT01568658) and has obtained the ethical approval from the NIH Institutional Review Board. Participant consents were obtained according to the Declaration of Helsinki.

NM-Score classification

The NM-Score is a questionnaire-based clinical severity score first developed in French to assess the functional ability of patients with NMDs in 3 domains: standing position and transfers (D1), axial and proximal motor function (D2), and distal motor function (D3).

A rigorous methodology of development and validation of the NM-Score has demonstrated its content validity through a Delphi process that involved 23 experts.[13] Its reproducibility was considered good. For D1, D2, and D3, the levels of agreement between raters were 82.9%, 78.1%, and 75.6%, and the kappa coefficients were .77, .69, and .64, respectively. The disagreements were minimal, a maximum of 1 point whatever the domain. There were significant correlations between the NM scores and the MFM scores: −.881 for D1, −.702 for D2, and −.495 for D3. In addition, the correlations were strong between the NM score D2 and Brooke Scale (r=.642), and between the NM score D1 and Vignos Scale (r=.862).

Translation and adaptation of NM-Score into English

Guillemin et al[16] proposed a set of standardized guidelines for cross-cultural adaptation of health-related quality-of-life measures based on previous research in psychology, sociology, and published methodological frameworks. These guidelines include recommendations for obtaining semantic, idiomatic, experiential, and conceptual equivalence in translation by using back-translation techniques and committee reviews.

In accordance with these guidelines as described in figure 1, two independent French to English translations were completed and compared, thereby leading to a reference version. The cultural sensitivity of the questionnaire phraseology was carefully considered because of the difference in the approach to disability between the French and the American culture. Then, a back-translation of the reference version was completed and compared with the initial French version by a committee of NMD experts (physical therapists, physicians, nurse practitioners) to identify any misinterpretations and finalize the NM-Score English version (appendix 1).

Validation study of English NM-Score

As per the NIH protocol, all the participants were diagnosed with CMD (clinical, muscle biopsy ± genetic testing): collagen 6–related disease, laminin alpha 2–related disease (LAMA2-RD), or