Spanish translation and linguistic validation of the North Star Ambulatory Assessment for Duchenne muscular dystrophy functional evaluation

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Introduction. There is a need for reliable and properly validated outcome measures in Duchenne muscular dystrophy, both to monitor functional impairment and to assess the impact of new therapies.

Objective. We aimed to perform a translation of the North Star Ambulatory Assessment scale into Spanish and a linguistic validation of the resulting Spanish version.

Materials and methods. A structured multistage process based on international guidelines was used, with the following steps: translation (preparation, forward translation, reconciliation, back translation, back translation review, clinicians' review), linguistic validation though pilot testing (cognitive interviewing, medical review, review of results and final changes), and finalization (proofreading, final report).

Results. No major difficulties were found during translation steps. Few changes were needed to reconcile forward translations. The linguistic validation process required several meetings to solve comprehension difficulties due to subtle nuances in the meaning of some words. The pilot study was carried out in 10 practitioners from different places in Spain, including both physiotherapists and specialists and registrars in physical medicine and rehabilitation. A total of 6 comments were obtained, including 2 comments on starting positions for items 4-5 (stand on one leg) and item 10 (stand on heels) and 2 comments on scoring instructions for item 3 (stand up from chair) and item 14 (jump).

Conclusion. Our study has resulted in a convenient and reliable instrument for the quantification of functional abilities in boys with Duchenne muscular dystrophy in Spain. Our innovations in methods and our results could be used as a suggested template for the North Star Ambulatory Assessment linguistic validation in other languages.

Key words. Duchenne muscular dystrophy. Functional assessment. Linguistic validation. NSAA. Spanish. Translation.

Introduction

Duchenne muscular dystrophy (DMD) is a rapidly progressing and severe X-linked disorder and results in proximal muscle weakness and increasing disability. It is the most common muscular dystrophy with an incidence of one in about 3,500-5,000 male live births [1]. Early symptoms usually appear between 1 and 3 years of age [2], with mean age of diagnosis being 4.5-5.5 years [3]. DMD usually results in delayed walking; gait abnormalities; difficulty for running, climbing stairs, and rising from the ground; and frequent falls. This is followed by a progressive impairment of muscular strength, with most untreated patients being wheelchair-bound by the age of 8-14 years [2].

There is a need for reliable and properly validated outcome measures in DMD, both to monitor functional impairment progression [4-6] and to assess the potential impact of new therapies [7]. This is particularly so after the recent advent of therapeutic approaches and the approval of disease-modifying drugs for small groups of patients based on controlled clinical trials [8]. Such validated outcome measures are also required for future use in international clinical trials [9,10].

A large number of scales have been developed and published over the years, particularly to assess functional abilities in boys with DMD, with their main limitations being low reliability and sensitivity to change and limited practical application in diverse clinical centres [11]. This led to the development and validation of a functional assessment scale for DMD in ambulant children older than 5 years, the North Star Ambulatory Assessment (NSAA). More recently, the need to extend controlled clinical trials to younger boys with DMD in order to begin proposed therapies at an early stage of the disease when muscle tissue is still largely preserved, was highlighted [12], and the suitability of the NSAA scale use in children younger than 5 years was suggested [13]. Thus, Mercuri et al pub-

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Acknowledgements:
We very gratefully acknowledge the UK North Star Clinical Network, and the Muscular Dystrophy UK (MDUK) who supported the development of the scale. We are thankful to Adnan Manzur and the other developers of the original English version of the NSAA scale for giving us permission to translate it and for their continued support for this project. We are in debt to all the participants volunteering to contribute to the cognitive interviewing.

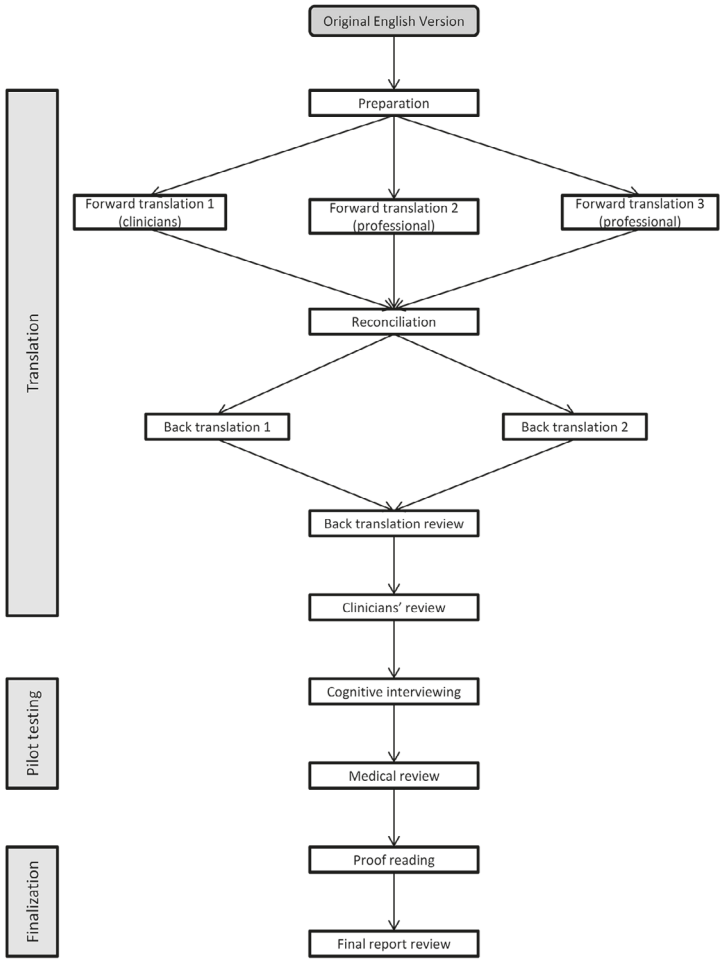
Funding:
Medical writing was supported by PTC Therapeutics with an unrestricted educational grant, and was provided by Content ed Net (Madrid, Spain).

Accepted:
11.01.21.

How to cite this article:
Medina-Cantillo J, Moya-Arcos O, Padrós-Domingo N, Roca-Urraca S, Vigo-Morancho M, Mas S. Spanish translation and linguistic validation of the North Star Ambulatory Assessment for Duchenne muscular dystrophy functional evaluation. Rev Neurol 2021; 72: 337-42. doi: 10.33588/rn.7210.2020604.

Versión en español disponible en neurología.com

Figure. Diagram of the translation and linguistic validation method.



lished a revised version of NSAA scale that can be used also between the ages of 3 and 5 years [12].

Aim

Taking into account the need for assessment instruments that can be used in diverse clinical settings and cross-cultural multinational trials, we aimed to perform a translation of the NSAA scale into Spanish and to carry out a linguistic validation of the resulting Spanish version. We also set out to introduce and assess some refinements in the linguistic validation process for methods intended to allow quantification of functional abilities in clinical settings.

Materials and methods

The NSAA scale was developed through focus groups and workshops in 17 centres in the UK, and its inter- and intra-assessor reliability was confirmed for boys older than 5 years [11]. It is a practical instrument to be used both in speciality centres and community care with limited equipment, and takes about 10 minutes to complete. Moreover, it can be easily and successfully applied in boys with learning/behavioural abnormalities [11].

The revised version of the NSAA scale was used for translation [12]. This version includes the items ordered according to the age at which the actions can be performed and uses a modified scoring scheme based on age-appropriate items [12].

NSAA scale has 17 items ordered according to the age when new abilities are acquired, from standing to hopping on one foot. Items have been selected to reflect activities typically impaired by muscular weakness in DMD. Each item is to be scored on a 3-point scale, and an overall score is obtained by adding the scores for individual items (range 0-34) [12].

Target language was Spanish spoken in Spain. Existing international guidelines for good research practices in translation, cultural adaptation and linguistic validation for outcome measures and multinational trials were followed [14-16], in order to produce a conceptually equivalent and linguistically acceptable instrument. Minor changes in the process were used to adapt it to a scale focused on functional parameters instead of quality of life assessments. A structured multistage process was used, with the following steps: translation (preparation, forward translation, reconciliation, back translation, back translation review, clinicians' review), linguistic validation though pilot testing (cognitive interviewing, medical review, review of results and final changes), and finalization (proofreading, final report) (Figure). Pilot testing was performed in health care workers volunteering to provide their experience when individually using the translated scale; no data from specific patients were used in the process.

Results

Translation

Preparation

The developers of the original English version of NSAA were contacted and their permission was obtained to perform a translation and linguistic

validation of a Spanish version (communication by Adnan Manzur, July 4, 2018). We very gratefully acknowledge the UK North star clinical network, and the Muscular Dystrophy UK (MDUK) who supported the development of the scale. The process was started with the initial version of the scale and subsequently expanded to the later revised version after having been informed about it by the developers. The project director (S.M.), a native Spanish medical doctor and associate professor of medical translation, led the project. She reviewed the scale to clarify the concepts being addressed, and prepared a conceptual definition and references to be provided to professional translators.

Forward translation

Three separate and entirely independent translations into Spanish were performed. All translators were native Spanish speakers and living in Spain. Translation 1 was performed by a group of medical doctors who had clinical experience in DMD. Translations 2 and 3 were performed independently by two highly qualified (master on biomedical translation) professional translators fulfilling the linguistic, cultural and technical standards required. The translators not being medical doctors were informed about the study aims and received explanations on the DMD to allow them keeping the concepts and avoid a word-to-word translation. They were both familiar with medical terms and had worked for several years for international health-related organizations and one of them was a university associate professor of translation. Translators had no contact with each other during the translation process. The three translations were performed uneventfully and were available on time for reconciliation.

Reconciliation

Similarities between the translations were compared and discrepancies were resolved in a meeting of the two professional translators and a Spanish-speaker coordinator who was also proficient in English. A total of 23 translation items required discussion. Six of them were related to the real differences in meaning and required clarification. Seventeen were due to stylistic differences and use of different grammatical constructions or different names for the same concept or object. A consensus was reached for all of them with the contribution by an additional translation consultant. Based on the whole details of the three translations, the discussion and the final consensus, all contributions were reconciled into a single forward translation.

Back translation

As a major measure of quality control to ensure the meaning was preserved, the reconciled Spanish version was used to perform two back translations into British English. Back translations 1 and 2 were performed separately by independent native professional translators who had never seen the original scale and had no previous access to it. Both translators were qualified members of the UK-based Institute of Translation and Interpreting (MITI), a post-experience category achieved after a compulsory entry test and several years of demonstrated experience. Back translations were performed in time and uneventfully.

Back translation review

Both back translations were compared to each other and checked against the original version to ensure conceptual equivalence and address discrepancies and problematic items in order to refine the final forward translation. Apart from a number of stylistic variations and alternatives in grammatical construction, five problematic items with a potential impact on the meaning were found. They all were related to the need to specify the starting position for some of the items in the scale. Slight modifications in Spanish translation wording were applied to avoid such potential confusions.

Clinicians' review

A clinical coordinator (J.M.C.) and a group of 4 clinical consultants (O.M.A., N.P.D., S.R.U., M.V.M.) from the Rehabilitation and Physical Medicine Unit at the Sant Joan de Déu Children's Hospital (Barcelona, Spain) reviewed the Spanish version from the perspective of its clinical use. Comments by clinicians were contributed in two meetings with the translator consultant and led to three wording changes aimed at clarifying the instructions to the children. Thus, a final Spanish version was obtained.

Linguistic validation though pilot testing

Cognitive interviewing

Since NSAA is a scale to be administered by a health care practitioner, no patient or patient's relative was involved in the process, and a sample of 10 potential health care users who had not been involved in the translations project and volunteered to participate was selected (Table). Median (range) age of participants was 27 (20-58) years (mean ± SD: 30.8 ± 12.1 years); most were female (8/10). They were all native Spanish health care profession-

Table. Participants in the cognitive interviewing for the pilot testing.

Age (years)	Sex	Qualification	Centre
58	F	Physiotherapist	Tertiary paediatric hospital
25	F	Physiotherapist	Tertiary paediatric hospital
22	M	Physiotherapist	Tertiary paediatric hospital
30	F	Registrar (Physical Medicine and Rehabilitation)	Tertiary general hospital
29	F	Registrar (Physical Medicine and Rehabilitation)	Tertiary general hospital
47	F	Consultant (Physical Medicine and Rehabilitation)	Tertiary general hospital
26	M	Physiotherapist	Tertiary general hospital
20	F	Final-year physiotherapy student	Tertiary paediatric hospital
23	F	Physiotherapist	Tertiary paediatric hospital
28	F	Registrar (Physical Medicine and Rehabilitation)	Tertiary general hospital

F: female; M: male.

al working in Rehabilitation and Physical Medicine Units at tertiary-level hospitals in different centres in Spain (five in paediatric units in general hospitals and five in children's hospitals). There were five physiotherapists, three Physical Medicine and Rehabilitation registrars, one Physical Medicine and Rehabilitation consultant and one final year physiotherapy student.

All interactions with participants were conducted in the Spanish language only. Standardized interviews were conducted after the participants' review of the instrument and based on their actual experience in the management of young boys for whom the scale was intended to in their clinical activity. The interview included questions aimed at assessing both the instructions to be given to patients and the scoring rules for each item. Two main topics were covered: a) assessment of clarity, intelligibility and comprehension and detection of potential understanding difficulties or misunderstanding; b) assessment of cultural appropriateness and detection of inappropriate words or phrases.

Findings were used to improve translation or reword some items if needed. A total of six comments were obtained, including two comments on starting positions for items 4-5 (stand on one leg) and item 10 (stand on heels) and two comments on scoring instructions for item 3 (stand up from chair) and item 14 (jump). Some slight rewording was applied to solve all such issues. There were also two comments on the overall comprehension and the order of items that only required clarification on the use of the revised scale in younger children. The results from cognitive interviews were used to produce the final Spanish version.

Medical review

The clinical coordinator and the group of clinical specialists reviewed the final version. No changes were needed.

Finalization

Proofreading

Final Spanish version was checked for minor typing, spelling or grammatical errors by a Spanish native professional proofreader in two rounds. Moreover, the translation was checked against the English original version to ensure that no part of the text had been repeated or omitted.

Final report

A final report was written by the project director with contributions by all authors who reviewed and approved its final version.

Discussion

We report the development of a Spanish version for the NSAA scale through a double-checked translation process with several forward- and back-translations and a linguistic validation by means of cognitive interviewing and medical review. International guidelines [14,15] were followed and supplemented with additional steps specific for this kind of instrument, including involvement of clinical experts. The process guaranteed a high-quality translation, and the involvement of expert clinical specialists ensured its medical suitability. This is particularly important because, as stated by the World Health Organization guidelines, translation and linguistic validation is a prerequisite to allow a different version of the same instrument to be 'equally natural and acceptable' and to 'practically perform in the same way' [17]. Our study has resulted in a convenient and reliable instrument for the quantification of functional abilities in boys with DMD in Spain.

Our translation process applied a refinement of the method with the addition of a third forward translation performed by clinical specialists to the

usual two professional translations. This allowed ensuring both conceptual equivalence for the actions to be assessed in each item and a natural language for Spanish children, particularly for the instructions to the patient. Another strength in our study is the involvement of medical doctors with both clinical and translation training and experience in the steering of the process. We recognize, however, this is not easy to achieve in all settings.

No major difficulties were found during translation steps. Few changes were needed to reconcile forward translations, as well as to solve slight discrepancies of back translations when compared to the original version. However multiple independent forward and back translations proved to be necessary to identify small modifications to improve the final version. Most changes consisted in changing one word to make it sound more natural in the Spanish language spoken in Spain.

The linguistic validation process required several meetings to solve comprehension difficulties due to subtle nuances in the meaning of some words or phrases. Although only few modifications were done based on the pilot testing results, such changes highlight the need for this step in a rigorous and accurate linguistic validation of such an instrument. The pilot study was carried out in 10 practitioners from different centres in Spain, including both physiotherapists and specialists and registrars in physical medicine and rehabilitation. The limitation of a short number of participants is recognized. However, we believe they are representative of the practitioners that are likely to use the scale in Spain.

Use of Spanish spoken in Spain is also a limitation and makes our translation version not necessarily valid for other Spanish-speaking countries. NSAA scale includes short instructions to the patient for each item, for which a rather colloquial language is used to allow young children to easily understand them. Such wording has many local variations in Spanish, particularly in Latin America, and should be adapted to specific countries or regions. Nevertheless, our version could be used as a reference material for future adaptation to other Spanish-speaking areas with local differences.

Conclusions

We hope that the Spanish version can be made available to clinicians in Spain and can be useful to improve clinical follow-up of patients in Spain. Together with a previously developed and validated Brazilian Portuguese version [18], it will also allow comparison and aggregation of results from different countries and populations and will help performing multinational clinical trials. We look forward to the publication of other versions in additional languages that will be welcome to enable multinational research. Our innovations in methods and our results could be used as a suggested template for NSAA linguistic validation in other languages and non-English-speaking populations, based on the original English version.

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Traducción al español y validación lingüística de la escala *North Star Ambulatory Assessment* para la evaluación funcional de la distrofia muscular de Duchenne

Introducción. En la distrofia muscular de Duchenne son necesarias medidas de evaluación fiables y validadas para el seguimiento del deterioro funcional y de los efectos de los nuevos tratamientos.

Objetivo. Se realizó una traducción, seguida de validación lingüística, de la escala *North Star Ambulatory Assessment* al español.

Materiales y métodos. Se utilizó un proceso estructurado, de múltiples etapas, basado en las guías internacionales, con los siguientes pasos: traducción (preparación, traducción directa, reconciliación, retrotraducción, revisión de la retrotraducción y revisión por médicos clínicos), validación lingüística mediante una prueba piloto (entrevista cognitiva, revisión médica, revisión de resultados y ajustes finales) y finalización (revisión de pruebas e informe final).

Resultados. No surgieron dificultades importantes durante los pasos de traducción. La reconciliación de las traducciones directas requirió pocos cambios. En la validación lingüística fueron precisas varias reuniones para resolver dificultades de comprensión, en matices sutiles, en el significado de algunas palabras. El estudio piloto se realizó con 10 especialistas clínicos de diferentes lugares de España (fisioterapeutas y especialistas o residentes de medicina física y rehabilitación). Hubo seis comentarios, dos de ellos sobre las posiciones de partida en los ítems 4-5 (de pie sobre un solo pie) y 13 (de pie sobre los talones), y dos sobre las instrucciones de puntuación para los ítems 3 (levantarse de la silla) y 14 (saltar).

Conclusión. Nuestro estudio ha proporcionado un instrumento cómodo y fiable para cuantificar las capacidades funcionales en niños con distrofia muscular de Duchenne en España. Las innovaciones en el método y nuestros resultados podrían usarse como modelo para la validación lingüística en otros idiomas.

Palabras clave. Distrofia muscular de Duchenne. Español. Evaluación funcional. NSAA. Traducción. Validación lingüística.