

Independent Spanish Validation of the Unified Dyskinesia Rating Scale

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Abstract: The Unified Dyskinesia Rating Scale (UDysRS) assesses the severity and disability caused by dyskinesias in Parkinson's disease (PD). As part of the UDysRS development plan, the International Parkinson and Movement Disorder Society (MDS) established guidelines for official non-English translations. We present here the formal process for completing this program and the data on the first officially approved non-English version of the UDysRS (Spanish). The UDysRS translation program involves four steps: translation and back-translation; cognitive pretesting to ensure that raters and patients understand the scale and are comfortable with its content; field testing of the finalized version; and analysis of the factor structure of the tested version against the original English-language version. To be designated an official MDS translation, the confirmatory factor analysis comparative fit index (CFI) had to be ≥ 0.90 . The Spanish UDysRS was tested in 253 native-Spanish-speaking patients with PD. For all four parts of the UDysRS, the CFI, was ≥ 0.94 . Exploratory factor analyses of the Spanish version revealed a very clear factor structure, with three factors related to ON dyskinesia, OFF dystonia, and patient perceptions of the functional effect of dyskinesias. The Spanish version of the UDysRS successfully followed the MDS Translation Program protocol, reached the criterion to be designated as an official translation, and is now available on the MDS website for use.

In Parkinson's disease (PD), one of the most troubling clinical treatment issues is drug-induced dyskinesia.^{1,2} At present, there is no biomarker or other gold-standard index to determine the severity of drug-induced dyskinesias in PD, and severity assessment relies on clinically based rating scales. In this regard, the Unified Dyskinesia Rating Scale (UDysRS) has been developed to provide a comprehensive rating tool of dyskinesia in PD.³ The UDysRS has four parts: (1) Historical Disability (patient perceptions) of ON-Dyskinesia impact (maximum, 44 points); (2) Historical Disability (patient perceptions) of OFF-Dystonia impact (maximum, 16 points); (3) Objective Impairment (Dyskinesia severity, anatomical distribution over seven body regions, and type (choreic or dystonic) based on four activities observed or video recorded (28 points); and (4) Objective Disability based on Part 3 activities (maximum, 16 points). As a result, the UDysRS has been used in several studies because it

has demonstrated acceptable levels of internal consistency, inter- and intrarater reliability, temporal stability, and detects treatment response.³⁻⁶ The UDysRS includes a teaching program that includes a DVD-based training program with instructions, and a Web-based certification exercise also available.⁴ Because dyskinesia therapies involve multicenter studies, having a scale that is validated in multiple non-English languages is pivotal to international efforts to treat dyskinesia. To obtain equivalent, locally validated non-English versions of the UDysRS, a translation program protocol has been established by the International Parkinson and Movement Disorder Society (MDS) under the direction of the rating scale committee (www.movementdisorders.org). The aim of this study was to organize and perform an independent validation of the UDysRS Spanish version following the methodology of the English-language validation process.³

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Methods

Design

This study was an international, multicenter, observational, cross-sectional assessment.

Procedure

Phases in the development of the Spanish version of the UDysRS followed the prescribed protocol established by the MDS for official translations of the UDysRS: (1) translation, back-translation, comparison with the original, and amendments of the scale text. All wording was selected to meet the criterion of being at a seventh-grade reading level. Documents were reviewed by the administrative team and reviewers independent of the Spanish organizers and a draft-translated scale was developed based on feedback; (2) cognitive pretesting of a small sample of raters and PD patients, to establish task difficulty for examiner and respondent, as well as respondent interest, attention span, discomfort, and comprehension.⁷ Once cognitive pretesting was taken into account, the final translation was obtained: (3) field testing in a large sample of native Spanish speakers; and (4) statistical analyses including validity testing and factor analyses.

Patients and Ethical Aspects

This study included consecutive patients, at any age and disease stage, with PD as per the UK Parkinson's Disease Society Brain Bank Criteria.⁸ The program for validation of the UDysRS was approved by the ethics committee of the Complejo Hospitalario Asistencial Universitario of Burgos, Spain. Patients gave their signed consent to participate after receiving the pertinent information. Sampling was monitored to ensure an adequate representation of all levels of dyskinesia severity.

Raters

Neurologists from the Spanish Movement Disorder Society were invited by e-mail to participate in this project. Furthermore, the project was announced during the Movement Disorders Meeting in the Spanish Neurological Society Annual Meeting of 2010. To qualify for inclusion as a rater, each investigator successfully completed the MDS UDysRS training program.⁴

Data Analysis

In addition to descriptive statistics, the following attributes were tested as the primary analyses of the Spanish data: (1) Cronbach's alpha index⁹ assessed the internal consistency of the translated scale. This index provides the lowest estimate of reliability that can be expected for an instrument and is based on the average correlation between two halves of the scale across all possible split halves. We also examined the corrected item-

to-total correlation as well as the change in Cronbach's alpha if an individual item was removed; (2) factor analyses of the Spanish data included an exploratory factor analysis (EFA) and a confirmatory factor analysis (CFA) to examine the underlying factor structure for the Spanish-language scale. Question 1 (time of ON dyskinesia) and question 12 (time of OFF dystonia) were considered as descriptive indices, rather than measures, of impairment or disability and were omitted from the EFA and CFA. Mplus (version 6.11) was used to do the EFAs as the variables are categorical.¹⁰ We used an unweighted least squares approach to factor estimation that minimizes the sum of squared differences between observed and estimated correlation matrices not counting diagonal elements. To assist in interpretation of the factors, we used an orthogonal CF-VARIMAX rotation that constrains the factors to be uncorrelated. We used a Scree plot to choose the number of factors retained for UDysRS. The subjective Scree test¹¹ is a scatter plot of eigenvalues plotted against their ranks with respect to magnitude to extract as many factors as there are eigenvalues that fall before the last large drop (i.e., an "elbow" shape) in the plot. Once the factors were chosen, an item was retained in a factor if the factor loading for the item was 0.40 or greater. As a secondary analysis, we conducted CFA¹² for the Spanish version of UDysRS. We evaluated the CFA results based on the comparative fit index (CFI). The CFI of UDysRS was required to be 0.90 or greater. Mean and variance adjusted weighted least square estimator was used to confirm model fit. We also used the root mean square error of approximation (RMSEA) to check goodness of fit. It is a population-based index that relies on the noncentral chi-square (χ^2) distribution, which is the distribution of the fitting function when the fit of the model is not perfect.

The sample size for the translation study was based on the need for 7 to 10 subjects per item of the questionnaire in order to perform the tasks needed to validate the instrument.¹¹ Because there are 26 items on the UDysRS, a sample of at least 250 was required. The investigators obtained human subjects' approval before data collection. Deidentified data (without patient names or medical record numbers) were transferred to the analytic team by a secure website.

Results

Translation/Back-Translation Phase

The local translation/back-translation program included two independent teams (translators and back-translators) that operated under the Spanish coordinator (E.C.). For each team, two movement disorder neurologists fluent in English and Spanish were included. Subsequently, the translation and back translation was submitted and approved by the administrative team (C.G.G., G.T.S., B.C.T., and N.R.L.).

Cognitive Testing

Two rounds of cognitive testing were conducted, during which the data were collected. Six raters and 24 patients were involved

in round 1, and three raters and 10 patients were involved in round 2 of the cognitive pretesting phase. Cognitive pretesting identified issues with words directly translated from English (“twisting and jerking”) used to describe dyskinesia. Additionally, some of the language was thought to be too complex and lengthy. Recommendations were made to remove the words “dyskinesia” and “dystonia” anywhere in the scale where the patient has to respond to these words, using instead a clear definition. We recommended the use of lay language for ON dyskinesia and OFF dystonia as well as the shortening of sentences as needed for a seventh-grade reading level. The second round of cognitive testing identified primarily issues unique to specific patients, except that 2 patients did not identify the word “spasm” with dystonia, and a recommendation was made to investigate other wording possibilities.

Final Field-Testing Phase

Of 30 neurologists expressing initial interest, 22 (73.3%) participated in 19 different centers throughout Spain. All raters successfully completed UDysRS training through the MDS Web-based training program (C.G.G., director).⁴ Centers provided between 5 and 29 cases to the study cohort.

Baseline Characteristics

Demographic characteristics of the Spanish patients are shown in Table 1. The Spanish data set included 253 native Spanish-speaking patients with dyskinesia who were examined using the UDysRS. Figure 1 gives the distributions of answers to each question.

Cronbach’s Alpha Index and Correlation Analysis

The overall raw and standardized Cronbach’s coefficient alpha were 0.94 and 0.95, respectively, indicating that the Spanish UDysRS was reliable. Table 2 shows the correlation matrix for

TABLE 1 Demographics and PD history

Patients		All Patients	Nonzero ON-Dyskinesia Time
Total patients	N	253	208
Male	N	122	97
	%	48.2	46.6
Age	Mean	69.2	69.4
	SD	10.5	10.6
Race (white)	N	252	207
	%	99.6	99.5
Ethnicity (Hispanic)	N	251	206
	%	99.2	99.0
Years of dyskinesia	Mean	4.9	5.6
	SD	4.6	4.5
Years of PD	Mean	12.5	13.4
	SD	6.8	6.7
Years of education	Mean	10.4	10.6
	SD	5.8	5.8

SD, standard deviation.

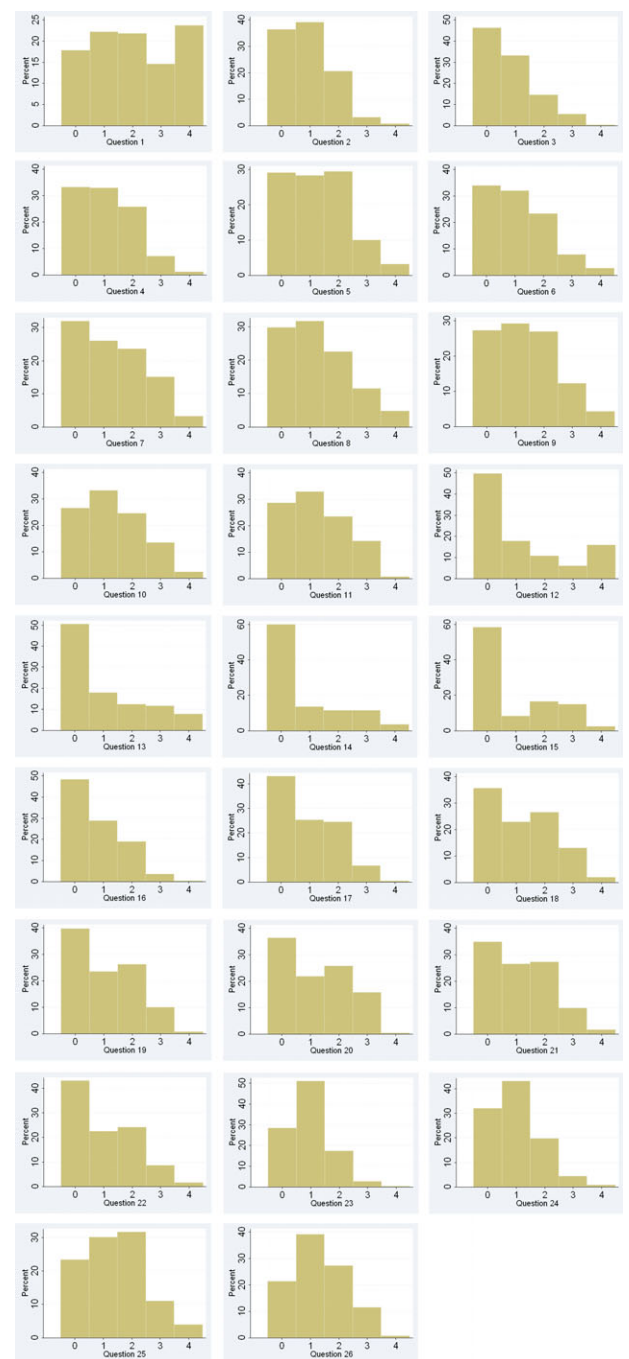


Figure 1 First histogram is all patients showing distribution of time with dyskinesia. In remaining histograms, 0 indicates those with 0 ON-dyskinesia time and 1 indicates those with ON-dyskinesia time greater than 0.

each question. Examination of the correlation of individual items to the total score revealed lower correlations for items 12 to 15.

EFA and CFA

Mplus performs list-wise deletion of cases with any missing data. That is, any case with one or more missing data points is

TABLE 2 Item-to-total correlations and Cronbach's alpha with deleted items

Cronbach Coefficient Alpha with Deleted Variable*				
Deleted Variables	Raw Variables		Standardized Variables	
	Correlation**	Alpha	Correlation	Alpha
Q1	0.671	0.946	0.683	0.951
Q2	0.648	0.946	0.660	0.951
Q3	0.668	0.946	0.681	0.951
Q4	0.767	0.945	0.776	0.950
Q5	0.760	0.944	0.764	0.950
Q6	0.781	0.944	0.787	0.950
Q7	0.761	0.944	0.771	0.950
Q8	0.736	0.945	0.739	0.950
Q9	0.737	0.945	0.743	0.950
Q10	0.777	0.944	0.778	0.950
Q11	0.705	0.945	0.699	0.950
Q12	0.320	0.951	0.294	0.955
Q13	0.381	0.949	0.348	0.954
Q14	0.353	0.949	0.320	0.954
Q15	0.302	0.950	0.269	0.955
Q16	0.581	0.946	0.600	0.951
Q17	0.657	0.946	0.670	0.951
Q18	0.690	0.945	0.706	0.950
Q19	0.667	0.946	0.676	0.951
Q20	0.659	0.946	0.674	0.951
Q21	0.636	0.946	0.645	0.951
Q22	0.602	0.946	0.608	0.951
Q23	0.656	0.946	0.672	0.951
Q24	0.735	0.945	0.748	0.950
Q25	0.737	0.945	0.749	0.950
Q26	0.737	0.945	0.740	0.950

Cronbach coefficient alpha is a measure of squared correlation between observed scores and true scores.

*Shows what the Cronbach coefficient alpha would be if that variable were deleted.

**The correlation between an individual item and the sum of the remaining items.

omitted entirely from analyses. Thus, the sample size in factor analysis is 247. Table 3 displays the factor structure for UDysRS and associated factor loading. The Scree plot is displayed in Figure 2. Based on these findings, we selected a three-factor solution: factor 1 covering questions 2 to 11 (ON dyskinesia); factor 2 covering questions 16 to 26 (patient perceptions); and factor 3 covering questions 13 to 15 (OFF dystonia). The CFI for the Spanish-language factor structure was 0.97, exceeding our prespecified criterion of 0.90 or greater, and the RMSEA of 0.083 (247 patients).

Discussion

The Spanish version of the UDysRS fulfills the criteria to be designated as an MDS-approved official translation (http://www.movementdisorders.org/publications/rating_scales). The total Spanish UDysRS scale demonstrated excellent internal consistency, suggesting that the scale provides a reliable measure of dyskinesia. In addition, the scale demonstrates a very clear factor structure, with three factors related to ON dyskinesia, OFF dystonia, and patient perceptions of the functional effect of dyskinesia. This three-factor structure was confirmed in the CFA. The original English version was clinimetrically evaluated

TABLE 3 Exploratory factor analysis without items "time ON dyskinesia" and "time OFF dystonia"

Factors	Items	N = 247		
Factor 1	Speech	0.626	0.421	0.088
	Chewing/swallowing	0.649	0.461	0.023
	Eating tasks	0.769	0.433	0.073
	Dressing	0.852	0.328	0.113
	Hygiene	0.809	0.42	0.064
	Hand writing	0.782	0.412	0.1
	Doing hobbies/activities	0.739	0.406	0.121
	Walking/balance	0.748	0.397	0.082
	Public/social	0.704	0.455	0.151
	Exciting situations	0.711	0.287	0.199
Factor 2	Dystonia effects on activities (not pain)	0.154	0.004	0.891
	Effect of pain from dystonia	0.121	0.046	0.979
	Dystonia pain severity	0.079	0.081	0.925
Factor 3	Face	0.298	0.731	-0.02
	Neck	0.331	0.755	0.024
	Right hand/arm/shoulder	0.417	0.694	0.034
	Left hand/arm/shoulder	0.408	0.658	0.077
	Trunk	0.352	0.753	-0.042
	Right foot/leg/hip	0.345	0.662	0.054
	Left foot/leg/hip	0.285	0.669	0.076
	Communication	0.298	0.793	0.06
	Drinking	0.427	0.781	0.018
	Dressing	0.401	0.696	0.083
Ambulation (walking)	0.373	0.653	0.168	

Values in bold indicate significant factor loading.

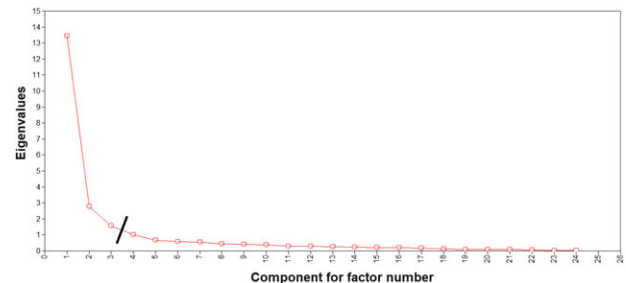


Figure 2 Questions "Time Spent with ON Dyskinesia" and "Time Spent with OFF Dystonia" were not considered in the Scree plots because they were not included in the factor analysis.

to establish internal consistency and inter-rater reliability, but the small sample size of the English version precluded a comprehensive analysis of factor structure.³ Because of this limitation, we could not compare the resultant structure from the present study with that of the original English version. Therefore, the Spanish version of the UDysRS stands as the first large-scale clinimetric analysis of this instrument.

The MDS has organized a world-wide program to provide official and clinimetrically validated versions of the MDS-UPDRS and the UDysRS. Several official non-English versions of the MDS-UPDRS have been developed, but the Spanish UDysRS is the first completed program for this scale. As of 1 June 2013, the overall translation program includes teams

organized to provide UDysRS versions including 16 other languages (French, German, Italian, Turkish, Greek, Portuguese, Dutch, Estonian, Polish, Chinese, Japanese, Korean, Hebrew, Russian, Slovak, and Hungarian). Enrollment of other language teams can be initiated at www.movementdisorders.org.

The present study was planned to develop a cross-cultural adaptation of the UDysRS in order to provide the official Spanish version of this scale, equivalent to the original version, and hereafter available for use in Spanish-speaking settings internationally. According to a scientific requirement (replication of outcomes), independent validation of health measures is necessary to confirm or reject the findings obtained by the developers of the instrument. This version is owned by the MDS, and for clinical trials or multinational research efforts, scale access rights can be arranged through the MDS. After the analysis was completed, the administrative team suggested that the accuracy of the UDysRS time-based questions on percent of the day with ON dyskinesia and OFF dystonia could be enhanced with the addition of clarifying statements that remind raters and patients to remain focused on the time spent with the two forms of dyskinesia over the past week. With their advice, for the English, Spanish, and all subsequent translated UDysRS scales, we have added three clarifying statements to ensure harmonization of the time-based questions with the patient/caregiver questionnaire and interview items: In the initial instructions to the full scale, we alert the rater to review the patient questionnaire after completion to ensure that, if item scores indicate the presence of dyskinesia or dystonia over the past week, the time-based items also reflect their occurrence (rating 1, 2, 3, or 4, but not 0); at the end of each questionnaire section (ON dyskinesia and OFF dystonia), the same alert is inserted.

A potential limitation is that the translation is only validated in a cohort of European Spanish-speaking patients, and cultural differences in Latin America and other regions of the world where Spanish is spoken could provide different clinimetric profiles. In contrast to our program anchored only in Spain, the MDS-UPDRS validation study included North and South American centers.¹³ The design of the present study did not include data for analyses of test-retest reliability or concurrent validity, important aspects of scale validation. Likewise, we acknowledge potential sample selection bias, because all subjects were patients under the care of neurologists at a specialty center. However, selection bias is a minor issue because neurologists at specialty centers are the most likely group to be using this scale for their research. Within Spain, we consciously chose a wide representation of patients from across the country because Spain remains regional in many cultural perspectives. The cross-cultural adaptation of the scale and the confirmation that the UDysRS Spanish version is structurally equivalent to the original English measure is relevant for future transnational studies enrolling patients from Spanish-speaking countries.

Author Roles

(1) Research Project: A. Conception, B. Organization, C. Execution; (2) Statistical Analysis: A. Design, B. Execution,

C. Review and Critique; (3) Manuscript: A. Writing of the First Draft, B. Review and Critique.

E.C.: 1C, 3B

C.G.G.: 1A, 1B, 1C, 2A, 2C, 3A, 3B

G.T.S.: 1A, 1B, 1C, 2A, 2C, 3B

B.C.T.: 2C, 3B

L.W.: 2B, 2C, 3A, 3B

S.L.: 2C, 3B

N.R.L.: 2A, 2B, 3B

Disclosures

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Appendix

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