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Validation of the Finnish Version of the Unified Dyskinesia Rating Scale

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Abstract
Introduction: The Unified Dyskinesia Rating Scale (UDysRS) was developed to provide a comprehensive rating tool of dyskinesia in Parkinson's disease (PD). Because dyskinesia therapy trials involve multicenter studies, having a scale that is validated in multiple non-English languages is pivotal to international efforts to treat dyskinesia. The aim of the present study was to organize and perform an independent validation of the UDysRS Finnish version. ***Methods:*** The UDysRS was translated into Finnish and then back-translated into English using 2 independent teams. Cognitive pretesting was conducted on the Finnish version and required modifications to the structure or wording of the translation. The final Finnish version was administered to 250 PD patients whose native language is Finnish. The data were analyzed to assess the confirmatory factor structure to the Spanish UDysRS (the reference standard). Secondary analyses included an exploratory factor analysis (EFA), independent of the reference standard. ***Results:*** The comparative fit index (CFI), in comparison with the reference standard factor structure, was 0.963 for Finnish. In the EFA, where variability from sample to sample is expected, isolated item differences of factor structure were found between the Finnish and Reference Standard versions of the UDysRS. These subtle differences may relate to differences in sample composition or variations in disease status. ***Conclusion:*** The overall factor structure of the Finnish version was consistent with that of the reference standard, and it can be designated as the official version of the UDysRS for Finnish speaking populations.

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Introduction

Dyskinesia is a frequent complication of levodopa-treated Parkinson's disease (PD). Although the prevalence of disabling dyskinesia has decreased over recent years due to more cautious use of levodopa and increased use of device-aided therapies [1], dyskinesia remains a major source of discomfort that ultimately affects a majority of PD patients [2]. The Unified Dyskinesia Rating Scale (UDysRS), originally developed in English language, provides a comprehensive rating tool of dyskinesia in PD [2]. The UDysRS consists of 4 sections: (1) historical disability of on dyskinesias (patient perception), (2) historical disability of off-dystonia (patient perception), (3) objective impairment (severity, anatomical distribution, and type), and (4) objective disability based on 4 activities. Internal consistency, inter- and intrarater reliability, and temporal stability of the UDysRS have proven to be acceptable [3, 4].

Because dyskinesia therapy trials involve multicenter designs, having a scale that is validated in multiple non-English languages is pivotal to international efforts to evaluate and treat dyskinesia. To provide locally validated equivalent non-English versions of the UDysRS, a translation program was established by the International Parkinson and Movement Disorder Society (MDS) under the direction of the MDS Rating Scales Translation Committee (www.movementdisorders.org). The aim of the present study was to organize and perform an independent validation of the UDysRS Finnish version. Finnish is a Uralic/Finnic language with a total user population of 5.83 million [5].

Materials and Methods

The UDysRS was translated into Finnish and then back-translated into English using 2 independent teams. Cognitive pretesting was conducted on the Finnish version, and required modifications to the structure or wording of the translation were completed. The final Finnish version was administered to a large cohort of PD patients whose native language is Finnish irrespective of race or ethnicity (e.g., Finnish vs. Sámi or other groups). To validate the UDysRS in full range of dyskinesias, investigators were instructed to recruit all eligible dyskinetic PD patients with mild, moderate, and extreme dyskinesia severity. The study was approved by the Turku University Hospital Ethics Committee, and patients provided written informed consent before participation.

To conduct the cognitive pretesting and data entry of the large cohort, a cognitive pretesting packet and a data entry guide were translated into Finnish. The data from the large Finnish cohort were analyzed to assess the confirmatory factor structure [6] to the Spanish UDysRS (the reference standard) [7]. Secondary analyses included an exploratory factor analysis (EFA), independent of the reference standard.

For cognitive pretesting, PD participants were recruited from the neurology outpatient clinic of the Turku University Hospital, Finland. For the large-scale validation, PD patients were recruited from outpatients and inpatients of university hospitals in Turku, Helsinki, and Oulu and from regional hospitals in Joensuu and Turku city. Both the cognitive pretesting and the validation were performed between 8 a.m. and 5 p.m. after the patients had taken their normal antiparkinsonian medications.

Cognitive Pretesting

Cognitive pretesting is a qualitative approach to assessing task difficulty for examiners and respondents, and respondent interest, attention span, comfort, and comprehension [8]. Before testing, we carried out a cognitive pretesting procedure to further investigate several items on the scale and specifically (1) items that were identified as different between the back-translated Finnish to English version and the original English version and (2) items that had been flagged as raising concerns in cognitive pretesting of the English version. Items in cognitive pretesting were instructions to raters and instructions to patients; time spent with dyskinesia; chewing and swallowing; exciting or emotional settings; effects of spasms or cramps separate from pain on activities; objective impairment ratings; and objective disability ratings. After each item, examiners and patients were instructed to evaluate the wording and usefulness of the questionnaire item using a numeric scale of 1–6 (e.g., how easy or difficult was it for you to understand this question? very difficult = 1, very easy = 6). Based on the results of the initial cognitive pretesting, other round(s) of translation and back-translation and cognitive pretesting could be required. Once cognitive pretesting was taken into account, the final translation was obtained.

Factor Analysis

For conducting the factor analysis of the UDysRS, we omitted question 1 (time of on dyskinesia) and question 12 (time of off dystonia) and considered these items as descriptive indices, rather than measures of impairment or disability. To maximize accuracy of these time indices, we added 3 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.

M-plus, version 7.4 was used to conduct the confirmatory factor analysis (CFA) and EFA as the variables are categorical [9]. We used the weighted least squares (WLSMVs) approach for factor estimation that minimizes the weighted sum of squared differences between observed and estimated correlation matrices. To assist in interpretation of the factors, we used an orthogonal VARIMAX rotation that constrains the factors to be uncorrelated.

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

Table 1. Demographics

	Total patients, *n*	Male		Age, years		Years of PD		Years of dyskinesia	
		n	%	mean	SD	mean	SD	mean	SD
Reference standard	253	122	48.2	69.2	10.5	12.5	6.8	4.9	4.6
Finnish	250	120	48	65.7	9.1	11.9	4.9	4	3.4

PD, Parkinson's disease.

Primary Analysis

As the primary analysis, we conducted a CFA, comparing the Finnish data to those from the reference standard UDysRS data. We determined if the factor structure for the reference standard UDysRS could be confirmed in the data collected using the Finnish translation. This was the primary question of interest. We evaluated the CFA results based on the comparative fit index (CFI). To confirm a good fit between the Finnish and reference standard UDysRS, CFI 0.90 or greater was required. Mean and variance adjusted WLSMV estimator was used to confirm model fit. We also used the root-mean-square error of approximation (RMSEA) to check the goodness of fit (threshold value: < 0.09). It is a population-based index that relies on the noncentral χ^2 distribution, which is the distribution of the fitting function when the fit of the model is not perfect.

Secondary Analysis

As a secondary analysis, we conducted an EFA to explore the underlying factor structure for the Finnish language translation, without constrain of a prespecified factor structure, using a WLSMV approach. We used a scree plot to choose the number of factors retained for the UDysRS. The subjective scree test [10] is a scatterplot 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 [10]. Once the factors were chosen, an item was retained in a factor if the factor loading for the item was 0.40 or greater. To assist interpretation of the factors, an orthogonal CF-VARIMAX rotation was used, which set the factors to be uncorrelated.

Results

Baseline Characteristics

The demographic characteristics of the Finnish patients are shown in Table 1. The Finnish data set included 250 native Finnish-speaking levodopa-treated patients with dyskinesia who were examined using the UDysRS between October 2017 and August 2020. see online suppl. Table 1; for all online suppl. material, see www.karger.com/doi/10.1159/000517369 gives the severity distributions of answers to each question.

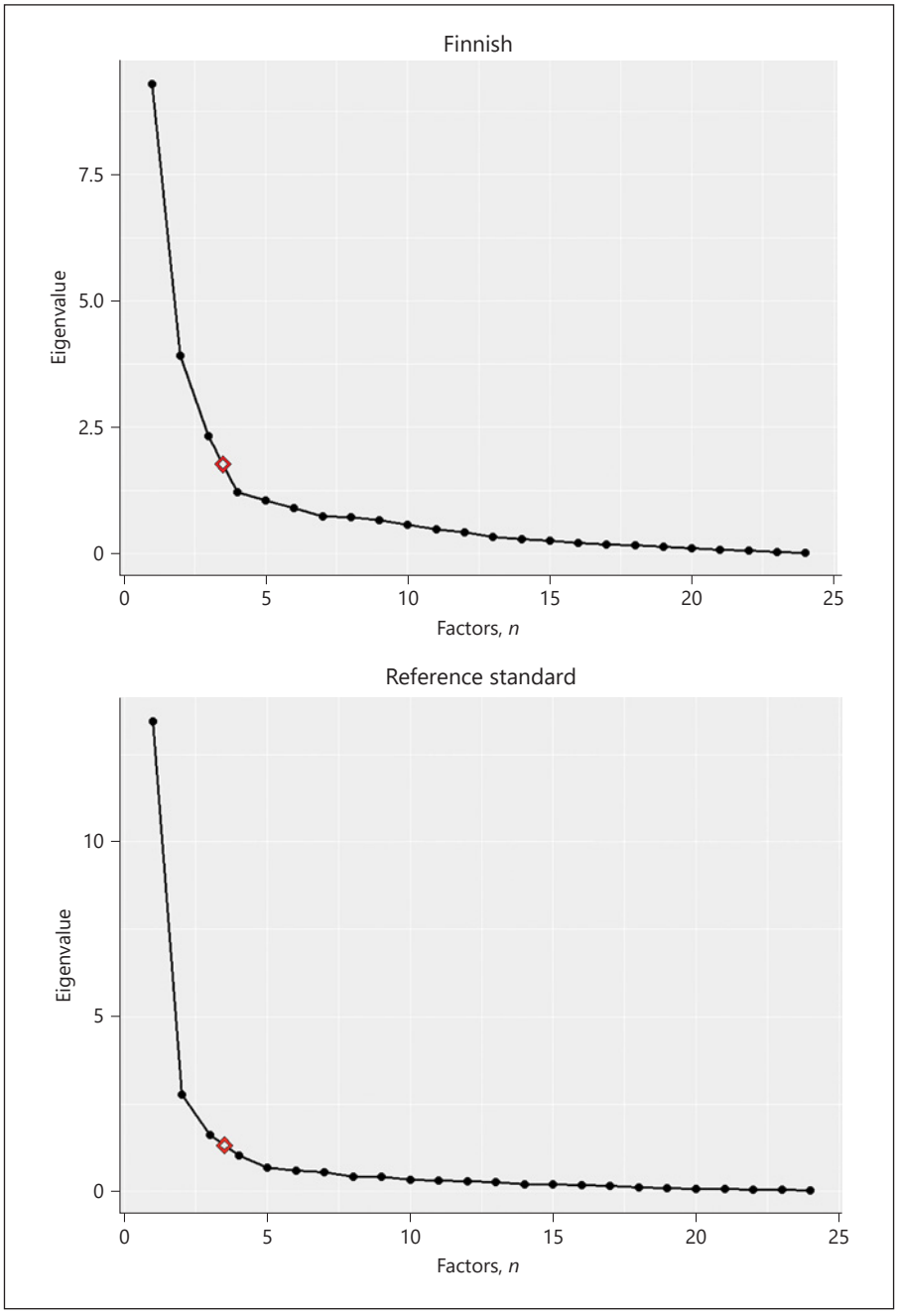
Table 2. Exploratory Factor Analysis (CFI = 0.953, RMSEA = 0.091)

	Reference standard (*N* = 246)	Finnish (*N* = 250)
Factor 1		
Speech	0.698	0.696
Chewing/swallowing	0.749	0.804
Eating tasks	0.800	0.895
Dressing	0.861	0.887
Hygiene	0.825	0.721
Handwriting	0.780	0.846
Doing hobbies/activities	0.728	0.689
Walking/balance	0.731	0.805
Public/social	0.686	0.712
Exciting situations	0.718	NA
Factor 2		
Face	0.717	0.574
Neck	0.752	0.862
Right hand/arm/shoulder	0.701	0.809
Left hand/arm/shoulder	0.663	0.727
Trunk	0.769	0.747
Right foot/leg/hip	0.711	0.731
Left foot/leg/hip	0.741	0.792
Communication	0.775	0.858
Drinking	0.755	0.871
Dressing (objective)	0.739	0.738
Ambulation	0.729	NA
Dystonia pain severity	NA	0.620
Factor 3		
Dystonia effects on activities	0.883	0.954
Effect of pain from dystonia	0.971	0.896
Dystonia pain severity	0.945	NA
Exciting situations	NA	0.596

EFA, exploratory factor analysis; CFI, comparative fit index; RMSEA, root-mean-square error of approximation.

Cognitive Pretesting

A total of 10 patients with PD and 3 examiners were interviewed using the type of structured interview format typical for cognitive pretesting. On the first round of cog-



Color version available online

Fig. 1. Scree plots used to determine the number of factors to retain for EFA. EFA, exploratory factor analysis.

nitive pretesting, the only issue identified as problematic was the font size for the patient-reported sections of the scale. The font size was increased, and the scale was approved as the official working document of the Finnish UDysRS for testing in a larger group of patients with PD.

Primary Analysis: CFA

Mplus performs list-wise deletion of cases with any missing data; that is, any case with 1 or more missing data points is omitted entirely from analyses. The original sample size was 251; 1 case was deleted due to missing data, and the final sample size in factor analysis was 250. The CFI, in comparison with the reference standard factor structure, was 0.963 for Finnish (RMSEA = 0.077, n = 250; reference standard CFI = 0.973, RMSEA = 0.083, n = 246). Because the CFI was greater than our prespecified CFI criterion, we concluded that the prespecified reference standard factor structure was confirmed in the Finnish data set.

Secondary Analysis: EFA

Table 2 shows the results of the EFA for all patients of the reference standard and Finnish UDysRS without the items for "time spent with on dyskinesia" and "time spent with off dystonia." The scree plots are given in Figure 1. From the scree plot, we extracted 3 factors. The factor structure of Finnish UDysRS is quite consistent with that of reference standard UDysRS.

Discussion

The Finnish language is a Uralic language and one of two official languages spoken in Finland. Its linguistic origins are obscure and unrelated to many other European national languages. In this study, we recruited only participants who are native Finnish speakers. Even though Swedish is also an officially recognized language in Finland, we did not include subjects whose primary language was Swedish and whose secondary language was Finnish.

Although the early assumptions of the genetic homogeneity of the isolated Finnish population have now proven to be false [11], Finns may be particularly valuable to the study of treatment interventions that target specific mechanistic genes. There is also literature which points to a specific pharmacogenomic profile of the Finnish population as compared to other North European populations [12, 13]. Further, certain neurological disorders, such as cervical dystonia, have a relatively high prevalence in Finland [14], whereas others, such as Huntington's disease, seem to be rare compared to other countries [15]. Although the prevalence of PD in Finland appears to be similar compared to other European countries [16] and it seems to be particularly high in eastern and rural parts of the country [17, 18], there is no knowledge of possible differences in the prevalence of levodopa-induced dyskinesias in Finland. As PD research moves increasingly to deep phenotyping and gene-based therapies, it is particularly important that neurologists in Finland will be equipped with rating scales applicable to this population. The value of this validated scale not only pertains to participation of Finnish teams in multicenter dyskinesia trials but specifically to the aforementioned targeted genetic studies that may emerge in the future.

The overall factor structure of the Finnish UDysRS version was consistent with that of the reference standard based on the CFIs for all models of the UDysRS in the CFA (CFI = 0.98). Therefore, this version can be designated as the official Finnish version of the UDysRS. In the EFA, where variability from sample to sample is expected, identified isolated item differences of the factor structure were found between the Finnish and reference standard versions of the UDysRS. Also in previous UDysRS validations from other languages, such as Hebrew [19], Turkish [20], Hungarian [21], Slovak [22], and Italian [23], similar minor differences have been observed in relation to Spanish, the reference language. These subtle differences may relate to differences in sample composition or variations in disease status.

Conclusions

In terms of future efforts, similar validation processes for Finnish versions of the MDS-Unified Parkinson's Disease Rating Scale (MDS-UPDRS) and MDS Non-Motor Rating Scale (MDS-NMS) will benefit PD research of the Finnish population.

Statement of Ethics

The study was approved by the Ethics Committee of Turku University Hospital district (decision number ETMK 51/2017) and was conducted according to the principles of the Declaration of Helsinki. Written informed consent was obtained from all PD participants included in the study.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

V.K.: data acquisition, data analysis and interpretation, drafting of the original manuscript draft, and funding acquisition; F.S., M.K., J.K., A.B., J.S., J.Jo., J.Jä., J.E.-R., M.H.M., K.A., and E.P.: data acquisition and revising the manuscript; G.S., P.M.-M., C.G., J.L., and S.L.: design of the study, data analysis and interpretation, and revising the manuscript.

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