ORIGINAL ARTICLE



Validation of the Czech Version of the Dysphagia in Multiple Sclerosis Questionnaire (DYMUS)

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Abstract

Dysphagia is a common symptom of neurological disease, including multiple sclerosis (MS). The DYsphagia in MUltiple Sclerosis (DYMUS) questionnaire was developed as a screening tool for swallowing problems. The purpose of the present study was to validate the Czech version of the DYMUS questionnaire. We validated the questionnaire on a sample of 435 patients with MS and 135 healthy controls (HC) chosen by accidental sampling from larger, long-term studies conducted by the Prague MS Center. For the purposes of this study, we used both electronic (primary method of distribution) and paperbased (backup) versions of the questionnaire. The internal consistency of the whole scale was satisfactory (Cronbach's α =0.833). The DYMUS mean score in HC was 0.215 (standard deviation [SD]=0.776). Normative data suggested a cutoff value for dysphagia between 1 and 2 points. Principal component analysis (PCA) showed a two-factor structure of the adapted scale. However, the structure did not completely correspond to the originally proposed dimensions of dysphagia between HC and patients MS (U=25,546, p < 0.001) and by a positive correlation with the EDSS (Kendall's tau-b=0.169, p < 0.001) and other patient-reported outcomes. The Czech version of the DYMUS questionnaire is a valid and reliable tool for evaluating swallowing impairment in Czech-speaking patients with MS. Moreover, the questionnaire can be administered electronically, with a paper-based backup.

Keywords Dysphagia · DYMUS · Multiple sclerosis · Validity · Reliability

Introduction

Dysphagia (swallowing difficulties) is a common symptom of neurological diseases, including multiple sclerosis (MS). Swallowing is a semi-autonomic motor action that requires coordination of the respiratory, oropharyngeal and gastrointestinal tract muscles [1]. Dysphagia is prevalent in at least one third of all patients with MS [1–3]. The prevalence varies from 31% of people, based on subjective questionnaires, to 81% of people diagnosed with the use of instrumental evaluation [3, 4]. Relying on self-reports of dysphagia may be problematic, because silent aspiration cannot be realised by patients themselves, and in some patients, there can be a lack of realistic insight due to cognitive deficit [1, 3, 4].

Even in patients with mild symptoms of MS (an Expanded Disability Status Scale [EDSS] score \leq 2), dysphagia has been reported in 17%–20% of cases [2, 5]. The presence of dysphagia seems to correlate with the level of disability due to MS, reaching up to 65% in patients with greater neurological disability [2, 6]. Dysphagia can be one of the most life-threatening symptoms of MS [7–9]. Therefore, early identification of dysphagia can help reduce the risk of potential complications. Routine screening of dysphagia requires the use of a reliable and validated assessment tool.

There are relatively few validated, self-report questionnaires for dysphagia available to people with neurodegenerative diseases [10], of which the DYsphagia in MUltiple

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Sclerosis (DYMUS) questionnaire is the most used patientreported outcome for MS. It has been validated in several languages including English, French, Italian, Greek, Persian, Portuguese and Turkish [11–21]. The DYMUS questionnaire was developed as a screening tool of swallowing problems to identify those patients who should be referred to special assessment and rehabilitation intervention [11, 13]. The DYMUS questionnaire has shown that the presence of dysphagia occurs three times more often than when relying on a patient's self-report of dysphagia [15, 16]. The DYMUS questionnaire correlates adequately with other dysphagia questionnaires or other disease burden measures such as the EDSS [14, 16–19].

With only 10 items, the DYMUS questionnaire is a brief 1-min self-evaluation tool [2]. The original questionnaire reported a two-dimensional structure (dysphagia for solids and dysphagia for liquids) and solid internal consistency, with Cronbach's α equal to 0.877 [13]. The high internal consistency has been replicated across subsequent validation studies, with Cronbach's α or Kuder–Richardson 20 scores between 0.72 and 0.91 [11, 12, 14–19, 21].

Despite the short length of the DYMUS questionnaire, its factor structure and the functioning of some of its items have faced an ongoing discussion [11, 12, 16–19, 21]. Based on principal component analysis (PCA), two shortened versions of the questionnaire have been proposed. Printza et al. [17] skipped items Q2, Q8, Q9 and Q10 and proposed a 6-item version, the so-called Mod-DYMUS. On the other hand, Alali et al. [12] dropped items Q3, Q5, Q8, Q9 and Q10 to create a shortened 5-item version of the scale. The last question (Q10), asking about loss of weight, is the most controversial, with the majority of studies showing its poor functioning within the scale [12, 16–19, 21].

Building on the previous findings, the purpose of the present study was to validate the Czech version of the DYMUS questionnaire, primarily using an electronic version of the questionnaire, with a paper-based backup.

Methods

The Questionnaire

The original DYMUS 10-item questionnaire [13] was adapted from English to the Czech language using the adapted Wild et al. [22] guidelines of good practice. The author of the original DYMUS questionnaire gave us consent for the translation and adaptation. Two forward translations (by an independent translator and by a research team member) were carried out and merged after discussion (language validity). The appropriateness and clarity of the phrases used in the forward translation was discussed with a speech therapist experienced in clinical assessment of patients with neurodegenerative diseases (content validity). The backward English translation was provided by an independent professional translator and reviewed in discussion with the research team. The pre-final version was pilot tested with five MS Center employees and eight patients with more severe MS (EDSS \geq 5). The pilot-testing led to a minor revision of item Q10. The final Czech version of the DYMUS questionnaire is provided in Appendix 1.

The DYMUS questionnaire evaluates the ability to swallow solids (7 items) and liquids (3 items). The items are coded as 0=No or 1=Yes, with a total score ranging from 0 to 10. At least one point on the DYMUS scale indicates dysphagia, whereas a score of 3 or more indicates severe dysphagia [13]. For the purposes of this study, we used both a classical paper-based version and an electronically distributed version of the questionnaire.

Sample

We validated the questionnaire on a sample of 435 patients with MS and 135 healthy controls (HC) chosen by accidental sampling from larger long-term prospective observational studies conducted in the MS Center of the General University Hospital in Prague, Czech Republic (GA UK 154,218 study, AZV grant NV18-04–00,168). Ninety patients with MS were reassessed after 1 year.

Accidental sampling from the larger, long-term observational studies was based specifically on time-frame availability (i.e., examination planned between October 2018 and February 2020). The DYMUS questionnaire was not administered to the whole sample of the original long-term observational studies because of unforeseen COVID-19 prevention measures, which caused early discontinuation of the planned monitoring by the DYMUS questionnaire. In total, 515 patients with MS and 147 HC from the original studies, available during the given time-frame, were asked to complete the DYMUS questionnaire. The complete answers from 435 patients with MS and 135 HC thus represent a response rate of 84.5% and 91.8%, respectively.

The original, long-term observational studies consisted of 638 patients with MS selected by judgmental sampling [23] to proportionally represent all major MS forms as seen in the Prague MS Center population, and 211 HC selected by judgmental sampling following the age distribution of the originally studied MS population.

The inclusion criteria for patients with MS were as follows: clinically isolated syndrome or clinically definite MS confirmed by magnetic resonance imaging (MRI) and cerebrospinal fluid examination [24], native Czech speaker, participation in a brain MRI volumetric assessment programme and aged \geq 18 years. The exclusion criteria were signs and symptoms suggestive of a disease other than MS and a serious psychiatric disorder. For HC, the inclusion criteria were as follows: Czech native or Czech near-native speakers and aged 18–65 years. The exclusion criteria were presence of neurological or psychiatric disease, head trauma history, presence of other diseases which could affect cognitive functioning, addiction to alcohol or drugs and active participation in a neuropsychological assessment one year prior to the study. The presence of dysphagia was not an exclusion criterion for HC because validation of the DYMUS questionnaire was not the main purpose of the GA UK 154,218 study. Both studies were approved by the ethical committee of the General University Hospital in Prague, Czech Republic.

Data Collection

All data were collected at the MS Center of the General University Hospital in Prague, Czech Republic. The DYMUS questionnaire was distributed as a part of a questionnaire battery (e.g., together with the Fatigue Severity Scale [FSS], the Beck Depression Inventory-II [BDI-II] and the State-Trait Anxiety Inventory [STAI-X1, STAI-X2]) used in the above-mentioned studies.

Patients with MS completed the questionnaire during their planned routine visit to the MS Center (as part of a long-term prospective observational study with annual MRI monitoring of the brain and spinal cord and a comprehensive clinical assessment, consisting of neurological and neuropsychological examinations). HC completed the questionnaire during their planned study visit to the MS Center. The study protocol for patients with MS and HC was identical, except for the neurological examination, which was conducted only in patients with MS.

Study data were collected and managed using Research Electronic Data Capture (REDCap) tools hosted at the Department of Neurology of the General University Hospital in Prague and the First Faculty of Medicine of Charles University in Prague. The electronic version of the DYMUS questionnaire was distributed via the REDCap Survey Environment [25, 26].

The electronic version of the questionnaire was the primary method of distribution. Participants were allowed to fill in the paper-based version of the questionnaire in these cases: (1) patients with MS motorically unable to deal with the electronic version, (2) participants who actively asked for the paper-based version (e.g., older participants unfamiliar with, or afraid of, computer-based technology), (3) participants who failed to use the electronic version or (4) technical issues preventing use of the electronic version (e.g., inability to establish an internet connection).

Most of the participants (88.9%) completed the electronic version of the questionnaire. The electronic version was mostly completed on site using 7" Lenovo Table 4 tablets (76.6%). Of note, 11.4% of participants used a smartphone or desktop

computer, either on-site or at home. The paper-based version was distributed solely on-site and in almost all cases among patients with MS.

Statistical Analysis

Validation of the questionnaire followed standard procedures of validity and reliability assessment. First, the content and construct validity were assessed through item analysis using Cronbach's α measure of internal consistency and through PCA, using varimax rotation to identify items belonging to the dimensions. Additionally, we assessed comparability of the novel electronic and the classical, paper-based scale by calculating Cronbach's α measure of internal consistency for both scales and by conducting binomial logistic regression, controlling for collinearity, to test the similarity between both versions of the questionnaires. Versions of the questionnaire were included in the regression model as a dependent variable and selected variables (DYMUS-10 total score, the EDSS, years of education and age) were included as covariates.

Reliability was assessed through the above-mentioned measures of internal consistency. Although we did not assess test-retest reliability, we tested long-term stability of the results in 90 patients with MS after 1 year using Kendall's tau-b correlation coefficient.

We assessed criterion validity by comparing patients with MS and HC total scores on the DYMUS questionnaire using the Mann–Whitney U test and by assessing correlation (Kendall's tau-b) between DYMUS total scores and other disease burden markers such as the EDSS, BDI-II, STAI X1, STAI X2 and FSS. We based this approach to criterion validity on previous findings that dysphagia is more frequent in the MS population and that patients with MS and a higher EDSS score and greater disease burden face dysphagia more often [2, 6, 14, 17, 18]. We selected this approach because we did not have access to any other Czech standardised measure of dysphagia.

We performed all analyses using the statistical software jamovi 1.0 [27]. We used parametric or non-parametric tests according to the distribution of the variables. We assessed normality of the distribution by visual inspection of the histograms, inspection of the Q-Q plots and the Shapiro–Wilk test. We tested differences in demographic characteristics between the groups using a chi-squared test or a t-test based on the variable type.

Results

Participants

There were 435 patients with MS (68.5% female) assessed by the DYMUS questionnaire. The mean age of the patients was 40.9 years (range 19–65), with a median EDSS score of 2 (range 0–7). There were 135 HC (71.9% female) assessed by the questionnaire. The mean age was 41.5 years (range 19–65). See Table 1 for details.

Content and Construct Validity

Item Analysis

The scale items' general mean was 0.0495, with a standard deviation (SD) of 0.137, a minimum (Min) of 0 and a maximum (Max) of 1. Cronbach's α of the whole scale was 0.833. All items correlated positively with the rest of the scale (Table 2).

Paper Versus Electronic Version

The discrete item analysis of the electronic and paper-based questionnaire is comparable to the overall item statistics. The items' general mean (electronic scale) was 0.0454 (Sd=0.126, Min=0, Max=1). Cronbach's α of the whole electronic scale was 0.810. All items correlated positively with the rest of the scale. The items' general mean (paper-based scale) was 0.0825 (Sd=0.201, Min=0, Max=1). Cronbach's α of the whole paper-based scale was 0.902. All items correlated positively with the rest of the scale. Cronbach's α of both versions would be improved if item Q10 were dropped.

Binomial logistic regression with the version of the questionnaire as a dependent variable and selected variables (DYMUS-10 total score, the EDSS, years of education and age) as covariates predicted the version of the questionnaire used ($\chi^2(4) = 21.47$, p < 0.001, Nagelkerke's R²=0.094). However, from all covariates included in the model, only age appeared to be a meaningful predictor of the administered version (Table 3).

Dimensionality

The number of initial eigenvalues above 1, the proportion of explained variability and the Cattell scree plot suggested the presence of two or three latent dimensions in PCA.

 Table 1
 Demographic

 characteristic of participants

 Table 2
 DYMUS Component loadings based on parallel analysis

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Item	Componen	Uniqueness	
	1	2	
DYMUS 01	0.695	0.325	0.411
DYMUS 02		0.776	0.353
DYMUS 03	0.647		0.546
DYMUS 04	0.626	0.467	0.390
DYMUS 05	0.303	0.639	0.500
DYMUS 06		0.855	0.269
DYMUS 07	0.808		0.300
DYMUS 08	0.751		0.436
DYMUS 09	0.596	0.333	0.534
DYMUS 10			0.926

Note: ,varimax ' rotation was used, loadings below 0.3 hidden

Parallel analysis suggested two components, and analysis based on the number of initial eigenvalues above 1 suggested three components. The oscillation between two and three factors is caused by item Q10. In the component loadings based on parallel analysis (Supplementary Table 1), item Q10 did not show a loading above 0.3 in either of the two components. PCA also showed other differences from the originally proposed dimensional structure: few items sufficiently explained both factors and at the same time did not correspond completely to the original factor loadings (items Q1, Q4, Q5, Q9). Component 1 explained 30.3% of the total variance and component 2 explained 23.1% of the total variance. Cumulatively, both components explained 53.3% of the variance.

Long-Term Stability of Results After 1 Year of Follow-Up

Ninety patients with MS completed the DYMUS questionnaire again after 1 year of follow-up (the mean number of days between test–retest was 378 [Sd=82]). At baseline, 76.7% of retested participants had a minimal DYMUS total score of 0; after 1 year, this was true for 78.9% of the retested sample. Moreover, 72.2% of retested participants had a score of 0 at both timepoints, and 6.7% of retested participants showed a total score of 0 at only one of the two

MS patients	HC	<i>p</i> -value	Effect size
435	135		
$40.9(\pm 8.97)$	41.5 (±12.2)	0.542^{a}	Cohen's $d = 0.0601$
$15.3 (\pm 3.25)$	$17.2 (\pm 3.02)$	< 0.001 ^a	Cohen's $d = 0.5915$
298/137	97/38	0.462 ^b	Cramer's $V = 0.0308$
2 (0 – 7)	N/A		
$12.4 (\pm 7.66)$	N/A		
	$43540.9 (\pm 8.97)15.3 (\pm 3.25)298/1372 (0 - 7)$	435 135 40.9 (±8.97) 41.5 (±12.2) 15.3 (±3.25) 17.2 (±3.02) 298/137 97/38 2 (0 - 7) N/A	435 135 $40.9 (\pm 8.97)$ $41.5 (\pm 12.2)$ 0.542^{a} $15.3 (\pm 3.25)$ $17.2 (\pm 3.02)$ $< 0.001^{a}$ $298/137$ $97/38$ 0.462^{b} $2 (0 - 7)$ N/A

^aIndependent samples t-test, ${}^{b}\chi^{2}$ test

Table 3Item AnalysisDYMUS-10

Item	Frequency of "Yes" among MS patients	Frequency of "Yes" among HC	Item-Rest Cor- relation	Cronbach's α if Item Dropped
DYMUS 01	23 (5.3%)	3 (2.2%)	0.626	0.808
DYMUS 02	24 (5.5%)	0 (0%)	0.540	0.816
DYMUS 03	36 (8.3%)	5 (3.7%)	0.524	0.818
DYMUS 04	29 (6.7%)	6 (4.4%)	0.665	0.802
DYMUS 05	14 (3.2%)	2 (1.5%)	0.512	0.820
DYMUS 06	24 (5.5%)	3 (2.2%)	0.445	0.825
DYMUS 07	29 (6.7%)	3 (2.2%)	0.679	0.801
DYMUS 08	20 (4.6%)	2 (1.5%)	0.475	0.822
DYMUS 09	29 (6.7%)	4 (3%)	0.576	0.812
DYMUS 10	25 (5.7%)	1 (0.7%)	0.208	0.846

timepoints. Overall, there was long-term stability of scores after 1 year (Kendall's tau-b = 0.788, p < 0.001).

Criterion Validity

Patients with MS Versus HC

Differences in the frequency of 'Yes' answers to single items (Table 2) suggested that dysphagia is more frequent in the MS population. In total, 10.4% of HC (N=14) and 23.2% of patients with MS (N=101) answered 'Yes' to at least one question. The mean difference between patients with MS and HC in the DYMUS total scores tested with the Mann–Whitney U test confirmed this difference (U=25,546, p < 0.001, Cohen's d=-0.269).

The EDSS and Other Disease Burden Measures Versus the DYMUS Questionnaire

There was a weak, positive correlation between the DYMUS total score and the EDSS (Kendall's tau-b=0.169, p < 0.001) and a small positive correlation with other patient-reported outcomes (Table 4). There was no correlation between the DYMUS total score and disease duration (Kendall's tau-b=0.035, p=0.357).

Czech DYMUS-9 Questionnaire

Our data suggested removing item Q10 from the original DYMUS questionnaire. Therefore, we have reported normative data for both the Czech DYMUS-9 and DYMUS-10 questionnaires (Table 5).

We applied the standard cut-off value of the 5th percentile as the clinical cut-off point for the general population. The best cut-off value for symptoms of dysphagia based on this criterion for both the DYMUS-10 and DYMUS-9 questionnaires lies between total scores of 1 and 2. To confirm that, we were able to contact four out of the six HC who scored above 1 on the DYMUS questionnaire via telephone. All four confirmed that they had some symptoms of dysphagia at the time they completed the questionnaire (Table 6).

Discussion

The DYMUS questionnaire can provide clinicians with information about the risk of dysphagia in patients with MS. Our study brings novel data about dysphagia in a sample of Czech patients with MS and provides a validated Czech version of the DYMUS questionnaire, supplemented by basic normative data.

Our study provides a basis for the evaluation of dysphagia by an electronic version of the DYMUS questionnaire,

Table 4Predictors of theVersion of the DYMUS-10 inMS patients (HC excluded)

Predictor	Estimate	SE	Z	р	Odds ratio
Intercept	3.9909	1.1508	3.468	< 0.001	54.106
Czech DYMUS-10 total score	-0.0508	0.0870	- 0.584	0.559	0.950
EDSS	- 0.0934	0.1270	- 0.736	0.462	0.911
Years of Education	0.0630	0.0516	1.221	0.222	1.065
Age	- 0.0636	0.0183	-3.469	< 0.001	0.938

Note: Estimates represent the log odds of "Electronic version of Czech DYMUS-10" vs. "Paper-based version of Czech DYMUS-10 "

Table 5 Correlations between DYMUS-10 and other selected measures

	Kendall's tau-b	p-value
EDSS	0.169	< 0.001
Disease duration	0.035	0.357
BDI-II	0.212	< 0.001
STAI X1	0.201	< 0.001
STAI X2	0.204	< 0.001
FSS	0.224	< 0.001

EDSS-Expanded Disability Status Scale, BDI-Beck Depression Inventory-II), STAI X1, STAI X2-State Traite Anxiety Inventory, FSS-Fatigue Severity Scale

with a possibility of a paper-based backup, if necessary. In our study, the paper-based backup was requested especially by older participants who are not digital natives.

Although we did not directly compare the electronic and paper-based DYMUS, there were no significant differences between the versions. In our sample, the DYMUS questionnaire results were not related to the type of version administered. Furthermore, both versions achieved similar item and scale statistics.

Based on the PCA, we saw a similar two-component questionnaire structure (dysphagia for solids and liquids) as in the original English version of the questionnaire [13]. However, a few items did not completely correspond to the original factor loadings as proposed in the English version. This corresponds to other studies using PCA [12, 17] and suggests that the discriminative validity of the DYMUS questionnaire for dysphagia to liquids or solids needs further research attention. Moreover, our results support previous findings [12, 16–19, 21 showing poor functioning of

Table 6 Normative data of Czech DYMUS-9 and Czech DYMUS-10 Normative Data

	MS patients	HC
N	435	135
Czech DYMUS-10		
Mean (SD)	0.582 (±1.5)	$0.215 (\pm 0.776)$
Median (Min – Max)	0 (0 – 10)	0 (0 – 5)
Count (%) of people with symptoms of dysphagia (DYMUS-10 \geq 2)	50 (11.5%)	6 (1.4%)
Percentiles (P _i) for DYMUS-10 Total Scores		
0 (DYMUS-10 Total Score)	$100^{\text{th}} P_i$	$100^{\text{th}} P_i$
1	$23^{\rm rd} P_i$	$10^{\text{th}} P_i$
2	$11^{\text{th}} P_i$	$4^{\text{th}} P_i$
3	$7^{\text{th}} P_i$	$3^{\rm rd} P_i$
4	$5^{\text{th}} P_i$	$1^{\text{st}} P_i$
5	$3^{\rm rd} P_i$	$< 1^{\text{st}} P_i$
6		
7	$2^{nd} P_i$	
8	$1^{st} P_i$	
9	$< 1^{st} P_i$	
10 (DYMUS-10 Total Score)		
Czech DYMUS-9		
Mean (SD)	$0.524(\pm 1.43)$	$0.207 (\pm 0.754)$
Median (Min – Max)	0 (0 – 9)	0 (0 – 5)
Count (%) of people with symptoms of dysphagia (DYMUS- $9 \ge 2$)	48 (11%)	6 (1.4%)
Percentiles (P _i) for DYMUS-9 Total Scores		
0 (DYMUS-9 Total Score)	$100^{\text{th}} P_i$	$100^{\text{th}} P_i$
1	$20^{\text{th}} P_i$	$10^{\text{th}} P_i$
2	$11^{\text{th}} P_i$	$4^{\text{th}} P_i$
3	$7^{\text{th}} P_i$	$2^{nd} P_i$
4	$4^{\text{th}} P_i$	$1^{st} P_i$
5	$3^{\rm rd} P_i$	$< 1^{\text{st}} P_i$
6		
7	$2^{nd} P_i$	
8	$1^{\text{st}} P_i$	
9 (DYMUS-9 Total Score)	< 1 st P_i	

item Q10 (loss of weight); therefore, we have proposed a shorter 9-item version of the questionnaire.

Concerning item Q10, some authors have argued that the item should be preserved because a loss of weight is one of the most endangering symptoms of dysphagia [28]. However, others have pointed out the item's low specificity because a loss of weight can be caused by many other medical conditions, including malnutrition due to MS [12]. After pilot-testing, we tried to reword the item to exclude a voluntary loss of weight from the question (e.g., weight-reduction diet). As our results showed, this attempt did not improve the item's functioning. Considering the original purpose of the DYMUS questionnaire as a screening tool before performing detailed dysphagia instrumental assessment [1, 29–31], we argue that item Q10 with its low specificity is rather problematic and should be dropped. It would be an essential question only in a population with already instrumentally confirmed dysphagia.

Similarly to other study [18], our normative data suggested the cut-off value for symptoms of dysphagia to lie between total scores of 1 and 2 for both the original 10-item and the shorter 9-item versions of the questionnaire. Using the newly proposed criteria, the prevalence of dysphagia in our sample of patients with MS was around 11%. Even with the originally recommended one positive answer criterion [13], only 23% of people with MS in our Czech validation study showed a risk of experiencing dysphagia. This contrasts with other validation studies which have described a prevalence of dysphagia between 31 and 58% [16, 18, 19, 32]. This difference might be explained by a lower EDSS (the median EDSS score was 2.0) in our sample, which is further supported by previous studies in which the incidence of dysphagia in people with mild disability was similar to our current findings – that is, 17%–21% [2, 5].

Although we did not assess test-retest reliability, we tested long-term stability of the results in patients with MS after 1 year. Kendall's tau-b of 0.788 is very satisfactory, especially considering the longer 1-year retest period, possible fluctuations in the results due to MS and the rather conservative correlational measures applied. Our data on long-term stability are comparable to classical test-retest measures presented in other validation studies [12, 14, 17–19].

We also proved the criterion validity by comparing the results of patients with MS and HC, and by a small positive correlation between the DYMUS score and disease severity measures such as the EDSS, and the level of anxiety, depression and subjective perceived fatigue. Similarly to dysphagia, these measures worsen in people with greater disease burden [33]. The EDSS has proved to be correlated positively with DYMUS scores across multiple studies [14, 16–19, 28]. However, in contrast to previous findings [2, 14, 28], we found no correlation between the DYMUS score and

disease duration. This is not that surprising given the fact that disease duration does not necessarily provide information about disease severity, and it is in line with other authors with similar results [17, 18, 32, 34]. A larger sample that is more representative of people with more severe MS could generate stronger correlations with other disease severity measures, also including disease duration.

Two Variants of the Czech DYMUS Questionnaire

While preparing this manuscript, validation of a second Czech version of the DYMUS questionnaire was published [28]. This is a rather unique situation given the fact that both teams received the DYMUS author's permission to translate the questionnaire. Both studies showed strengths of the Czech DYMUS translation. There was satisfactory internal consistency comparable with other language versions [11, 12, 14–19, 21] for both Czech versions. The version of Kolčava et al. [28] was validated on a smaller sample of patients with MS and HC; however, the authors provided data on excellent test-retest reliability (assessed telephonically after 5 days; Spearman's r = 0.94, p < 0.001). They also confirmed the translation's criterion validity through correlations with the EDSS, disease duration and the 100 ml water swallowing test. On the other hand, Kolčava et al. [28] did not assess dimensionality and did not provide detailed normative data.

There are three minor content discrepancies between the Czech translations worthy of discussion. Specifically, they are in items Q4 (Kolčava et al. [28] does not follow the original English version so precisely and asks about the 'feeling of' food stuck in your throat), Q7 (our version does not mention literally 'solid food' as the English original does) and Q10 (based on pilot-testing, we modified Q10 by asking for 'unintentional' loss of weight, to eliminate an affirmative answer by people on voluntary weight-reduction diets). The discrepancies between the versions are rather small, yet it would be beneficial to propose a merged consensual Czech version in the future.

Limitations

There are some limitations to this study. First, we did not validate the DYMUS questionnaire with objective instrumental tests such as a videofluoroscopic swallowing study (VFSS), fibreoptic endoscopic evaluation of swallowing (FEES) or EMG activity. Second, the sample in our study was not random. We distributed the DYMUS questionnaire as a part of the neuropsychological assessment in larger observational prospective studies. Even though our MS sample chosen by judgemental sampling represents all major MS disease subtypes, patients with a low burden and shorter disease duration are overrepresented in our sample. Third, because of COVID-19 preventive measures, we decided to discontinue questionnaire distribution early. We believe it did not cause any bias because patients were assigned over time independently of their demographic or disease characteristics. Fourth, this validation study was performed in a single centre; however, the main results can be applied to other Czech-speaking MS centres. Fifth, in patients with MS the administration was part of their routine visit to the MS Center, a factor that could negatively affect their motivation, concentration or time possibilities. This could influence the different response rate in patients with MS and HC. If there was a lack of time, patients were asked to complete the questionnaire remotely. However, the size of this sample did not allow us to control for differences between on-site and remote distribution.

Conclusion

The results of this study showed that the Czech version of the DYMUS questionnaire is a valid and reliable tool for evaluating swallowing impairment in Czech-speaking patients with MS. The DYMUS questionnaire is a fast screening tool for regular clinical monitoring, supported by our novel normative data. We have also shown that the questionnaire can be administered electronically, bringing new possibilities of swift evaluation or remote distribution. This simple DYMUS questionnaire can be easily used by health care professionals to identify patients who should be referred to speech therapists for more specific instrumental assessments and for some therapeutic programmes.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s00455-022-10530-5.

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References

- Prosiegel M, Schelling A, Wagner-Sonntag E. Dysphagia and multiple sclerosis. Int MS J. 2004;11(1):22–31.
- Solaro C, Rezzani C, Trabucco E, Amato MP, Zipoli V, Portaccio E, Giannini M, Patti F, D'Amico E, Frau J, Lorefice L, Bonavita S, Della Corte M, Grasso MG, Finamore L, Ghezzi A, Annovazzi P, Rottoli M, Gasperini C, Restivo D, Maimone D, Rossi P, Stromillo ML, Bergamaschi R. Prevalence of patient-reported dysphagia in multiple sclerosis patients: an Italian multicenter study (using

the DYMUS questionnaire). J Neurol Sci. 2013;331(1–2):94–7. https://doi.org/10.1016/j.jns.2013.05.020.

- Hartelius L, Svensson P. Speech and swallowing symptoms associated with Parkinson's disease and multiple sclerosis: a survey. Folia Phoniatr Logop. 1994;46(1):9–17. https://doi.org/ 10.1159/000266286.
- Guan XL, Wang H, Huang HS, Meng L. Prevalence of dysphagia in multiple sclerosis: a systematic review and meta-analysis. Neurol Sci. 2015;36(5):671–81. https://doi.org/10.1007/ s10072-015-2067-7.
- Abraham SS, Yun PT. Laryngopharyngeal dysmotility in multiple sclerosis. Dysphagia. 2002;17(1):69–74. https://doi.org/10. 1007/s00455-001-0103-7.
- Abraham S, Scheinberg LC, Smith CR, LaRocca NG. Neurologic impairment and disability status in outpatients with multiple sclerosis reporting dysphagia symptomatology. Neurorehabil Neural Repair. 1997;11(1):7–13. https://doi.org/10.1177/ 154596839701100102.
- Slawta JN, Wilcox AR, McCubbin JA, Nalle DJ, Fox SD, Anderson G. Health behaviors, body composition, and coronary heart disease risk in women with multiple sclerosis. Arch Phys Med Rehabil. 2003;84(12):1823–30. https://doi.org/10.1016/ s0003-9993(03)00466-0.
- Sumelahti ML, Hakama M, Elovaara I, Pukkala E. Causes of death among patients with multiple sclerosis. Mult Scler. 2010;16(12):1437–42. https://doi.org/10.1177/1352458510 379244.
- Pasquinelli S, Solaro C. Nutritional assessment and malnutrition in multiple sclerosis. Neurol Sci. 2008;29(4):S367-369. https:// doi.org/10.1007/s10072-008-1046-7.
- Keage M, Delatycki M, Corben L, Vogel A. A systematic review of self-reported swallowing assessments in progressive neurological disorders. Dysphagia. 2015;30(1):27–46. https://doi.org/ 10.1007/s00455-014-9579-9.
- Bergamaschi R, Rezzani C, Minguzzi S, Amato MP, Patti F, Marrosu MG, Bonavita S, Grasso MG, Ghezzi A, Rottoli M, Gasperini C, Restivo D, Maimone D, Rossi P, Stromillo ML, Montomoli C, Solaro C, Group D. Validation of the DYMUS questionnaire for the assessment of dysphagia in multiple sclerosis. Funct Neurol. 2009;24(3):159–62.
- Alali D, Ballard K, Vucic S, Bogaardt H. Dysphagia in multiple sclerosis: evaluation and validation of the DYMUS Questionnaire. Dysphagia. 2018;33(3):273–81. https://doi.org/10.1007/ s00455-017-9864-5.
- Bergamaschi R, Crivelli P, Rezzani C, Patti F, Solaro C, Rossi P, Restivo D, Maimone D, Romani A, Bastianello S, Tavazzi E, D'Amico E, Montomoli C, Cosi V. The DYMUS questionnaire for the assessment of dysphagia in multiple sclerosis. J Neurol Sci. 2008;269(1–2):49–53. https://doi.org/10.1016/j.jns.2007. 12.021.
- Jafari Z, Shahbodaghi MR, Maroufizadeh S, Naser Moghadasi A. Validation of the Persian version of dysphagia in multiple sclerosis questionnaire for the assessment of dysphagia in multiple sclerosis. Iran J Neurol. 2018;17(3):99–104. https://doi.org/10. 18502/ijnl.v17i3.365.
- Asadollahpour F, Mehri A, Khatoonabadi A, Mohammadzaheri F, Ebadi A. Validation of the Persian version of the dysphagia in multiple sclerosis questionnaire for the assessment of dysphagia in multiple sclerosis. J Res Med Sci. 2019;24:52. https://doi.org/ 10.4103/jrms.JRMS_248_18.
- Sales DS, Alvarenga RM, Vasconcelos CC, Silva RG, Thuler LC. Translation, cross-cultural adaptation and validation of the Portuguese version of the DYMUS questionnaire for the assessment of dysphagia in multiple sclerosis. Springerplus. 2013;2:332. https:// doi.org/10.1186/2193-1801-2-332.

- Printza A, Boziki M, Bakirtzis C, Nikolaidis I, Kalaitzi M, Triaridis S, Grigoriadis N. The modified DYMUS questionnaire is a reliable, valid and easy-to-use tool in the assessment of dysphagia in multiple sclerosis. Eur J Neurol. 2020;27(7):1231–7. https:// doi.org/10.1111/ene.14219.
- Printza A, Kalaitzi M, Bakirtzis C, Nikolaidis I, Proios H, Grigoriadis N. Reliability and validity of the DYMUS questionnaire for the assessment of dysphagia in multiple sclerosis (Greek version) and proposed modification. Mult Scler Relat Disord. 2018;23:62– 8. https://doi.org/10.1016/j.msard.2018.05.004.
- Tenekeci EG, Kara B, Cetiz A, Demirkaya S, Demir N, Acikel C. Validity and reliability of the Turkish version of the questionnaire for the assessment of dysphagia in multiple sclerosis. Noro Psikiyatr Ars. 2018;55(3):243–7. https://doi.org/10.5152/npa.2016. 18105.
- Villain M, Heinzlef O. DYMUS questionnaire for dysphagia in multiple sclerosis: French validation and clinical considerations. In: Multiple Sclerosis Journal: ECTRIMS 2013, 2013. Sage.
- Sadeghi Z, Afshar M, Ebadi A, Baghban K, Qureshi ZS. Swallowing disorder in multiple sclerosis: modified version of the screening tool. J Rehabil. 2020;21(2):236–55. https://doi.org/10.32598/ rj.21.2.3036.1.
- 22. Wild D, Grove A, Martin M, Eremenco S, McElroy S, Verjee-Lorenz A, Erikson P. Principles of Good Practice for the Translation and Cultural Adaptation Process for Patient-Reported Outcomes (PRO) Measures: Report of the ISPOR Task Force for Translation and Cultural Adaptation. Value in Health. 2005;8(2):94–104. https://doi.org/10.1111/j.1524-4733.2005.04054.x.
- Timmerman ME, Voncken L, Albers CJ. A tutorial on regressionbased norming of psychological tests with GAMLSS. Psychol Methods. 2020. https://doi.org/10.1037/met0000348.
- Thompson AJ, Banwell BL, Barkhof F, Carroll WM, Coetzee T, Comi G, Correale J, Fazekas F, Filippi M, Freedman MS, Fujihara K, Galetta SL, Hartung HP, Kappos L, Lublin FD, Marrie RA, Miller AE, Miller DH, Montalban X, Mowry EM, Sorensen PS, Tintoré M, Traboulsee AL, Trojano M, Uitdehaag BMJ, Vukusic S, Waubant E, Weinshenker BG, Reingold SC, Cohen JA. Diagnosis of multiple sclerosis: 2017 revisions of the McDonald criteria. The Lancet Neurology. 2018;17(2):162–73. https://doi.org/ 10.1016/s1474-4422(17)30470-2.
- Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, McLeod L, Delacqua G, Delacqua F, Kirby J, Duda SN, RED-Cap Consortium. The REDCap consortium: building an international community of software platform partners. J Biomed Inform. 2019;95: 103208. https://doi.org/10.1016/j.jbi.2019.103208.
- 26. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)–a metadata-driven

methodology and workflow process for providing translational research informatics support. J Biomed Inform. 2009;42(2):377–81. https://doi.org/10.1016/j.jbi.2008.08.010.

- 27. The jamovi project (2019) jamovi. 1.0 edn., Sydney, Australia
- Kolčava J, Lasotová N, Vlčková E, Štourač P, Bednařík J. Validation of DYsphagia in MUltiple Sclerosis questionnaire – Czech version of DYMUS. Česká a slovenská neurologie a neurochirurgie. 2020;83–116(3):285–90. https://doi.org/10.14735/amcsn n2020285.
- Alali D, Ballard K, Bogaardt H. Treatment effects for dysphagia in adults with multiple sclerosis: a systematic review. Dysphagia. 2016;31(5):610–8. https://doi.org/10.1007/s00455-016-9738-2.
- Restivo DA, Casabona A, Centonze D, Marchese-Ragona R, Maimone D, Pavone A. Pharyngeal electrical stimulation for dysphagia associated with multiple sclerosis: a pilot study. Brain Stimul. 2013;6(3):418–23. https://doi.org/10.1016/j.brs.2012.09.001.
- Lee SY, Seo HG, Paik NJ. Botulinum toxin injection for dysphagia: a blinded retrospective videofluoroscopic swallowing study analysis. Am J Phys Med Rehabil. 2009;88(6):491–4. https://doi. org/10.1097/PHM.0b013e3181a0dc47.
- 32. Pajouh SD, Moradi N, Shaterzadeh Yazdi MJ, Latifi SM, Mehravar M, Majdinasab N, Olapour AR, Soltani M, Khanchezar F. Diagnostic evaluation of dysphagia in multiple sclerosis patients using a Persian version of DYMUS questionnaire. Mult Scler Relat Disord. 2017;17:240–3. https://doi.org/10.1016/j.msard. 2017.08.012.
- Chwastiak L, Ehde DM, Gibbons LE, Sullivan M, Bowen JD, Kraft GH. Depressive symptoms and severity of illness in multiple sclerosis: epidemiologic study of a large community sample. Am J Psychiatry. 2002;159(11):1862–8. https://doi.org/10.1176/appi. ajp.159.11.1862.
- Printza A, Triaridis S, Kalaitzi M, Nikolaidis I, Bakirtzis C, Constantinidis J, Grigoriadis N. Dysphagia prevalence, attitudes, and related quality of life in patients with multiple sclerosis. Dysphagia. 2020;35(4):677–84. https://doi.org/10.1007/ s00455-019-10075-0.

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