Craniocervical Dystonia Questionnaire (CDQ-24): Validation and Cross-Cultural Adaptation in Serbian Patients

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ABSTRACT

The purpose of this study was to investigate the validation of the translated and culturally adapted CDQ-24 questionnaire on a group of Serbian patients. The study was comprised of 100 consecutive patients with idiopathic cervical dystonia (CD) and blepharospasm (BSP) who were evaluated at the Institute of Neurology, Clinical Centre of Serbia in Belgrade between March and June 2007. The linguistic validation of CDQ-24 involved 3 steps, according to an internationally accepted methodology. Most of the patients with CD and BSP accepted the CDQ-24 questionnaire. The internal consistency reliability ranged from 0.81 to 0.97. The mean total score of the CDQ-24 was 35.6±23.5. Patients with BSP had better HRQoL scores in the Pain subscale (p=0.025) compared with CD patients. However, patients with CD had better HRQoL sores in the Activities of Daily Living subscale (p=0.028) compared with BSP patients. Statistically significant positive correlations were registered between the Dystonia Movement Scale score and almost all CDQ-24 scales. The Serbian version of CDQ-24 should be recommended for HRQoL evaluation among patients with CD and BSP as an important outcome measure.

Key words: *dystonia, quality of life, questionnaire, CDQ-24, validation, treatment*

Introduction

Dystonia is involuntary movement characterized by sustained muscle contractions usually producing twisting and repetitive movements or abnormal postures[1]. In a cross-sectional study of primary dystonia in 8 European countries, the prevalence was 15.2/100 000 with focal dystonia having the highest value of 11.7/100 000[2]. According to the Serbian epidemiologic study the prevalence of late onset focal dystonia in Belgrade (capital of Serbia) was 15.4/100 000[3].

All types of primary dystonia are frequently associated with depression and social disabilities, emphasizing its impact on the impairment of health-related quality of life (HR-QoL)[4]. Although the SF-36 is the most widely used generic measure of health status and has been already used in dystonia[4–6], there is a need for development of the disease-specific instruments for measuring specific problems in patients with different types of dystonia, particularly for assessment of their functional health and treatment effects[7]. The Craniocervical Dystonia Questionnaire (CDQ-24) was developed at Innsbruck University (Austria) as a disease-specific instrument to measure the quality of life of patients with craniocervical dystonia[7].

A large number of patients with different types of dystonia from all parts of Serbia are treated and followed at both inpatient and outpatients settings of the Department of Movement Disorders, the Institute of Neurology, Clinical Center of Serbia, in Belgrade, which is a primary referral national center for diagnosis and treatment of these disorders in Serbia. Thus, we had opportunity for studing the wide range of problems related to different aspects of dystonia[8–12]. However, keeping in mind all limitations of HR-QoL generic measures, as well as the large number of our patients, we decided to make possible usage of CDQ-24, the first patient-based rating scale both for cranial and cervical dystonia.

The objective of this study was to determine the validation of the translated and culturally adapted CDQ-24 questionnaire on a group of 100 Serbian patients.

Patients and Methods

The study was comprised of 100 consecutive patients with idiopathic cervical dystonia (CD) and blepharospasm (BSP) who presented for botulinum toxin type A treatment in the outpatient setting of the Movement Disorders Department, the Institute of Neurology, Clinical Centre of Serbia, Belgrade, between March and June, 2007. We did not include patients with neurologic abnormalities in addition to dystonia, a robust response to levodopa (dopa-responsive dystonia), a history of exposure to dopamine receptor blocking agents within 6 months before the onset of dystonia, severe head trauma with a loss of consciousness, and features suggesting various types of secondary dystonia. The standard supplementary investigations included magnetic resonance imaging (MRI) and/or computed tomography (CT) of the brain and cervical region, electroencephalography, extensive laboratory analyses (including serum levels of ceruloplasmin, urine copper levels, thyroid function tests, and a blood smear for accanthocytosis).

The study was approved by the Ethical Committee of the Clinical Center of Serbia. All participants were included after giving the informed consent.

In this study, HRQoL was measured by the disease-specific instrument, CDQ-24 which includes 24 questions divided in 5 domains: Stigma (9 items), Emotional Wellbeing (5 items), Pain (3 items), Activities of Daily Living (7 items), and Social/Family Life (4 items). Each item consists of 5 statements representing increasing severity of impairment, scored from 0 to 4. In order to obtain comparable scores for individual subscales, the raw subscores (sum of the individual item scores) were linearly transformed to a 0–100 scale, where a score of 0 indicates the best and a score of 100 the worst possible quality of life[7].

We performed translation and cultural adaptation of the original version of the CDQ-24[7], with the aim to produce a translated version in Serbian which is conceptually equivalent to the original version, as well as being clear and easy to understand. The linguistic validation involved three steps: a) translation of the original CDQ-24 into Serbian (forward translation); b) the questions of the first Serbian version of CDQ-24 were translated back into English (backward translation) and c) patient testing. Two professional translators, with the assistance of quality of life experts and clinicians performed both forward and backward translations. They discussed controversial items to produce a final version of the CDQ-24 which would be most appropriate for the cultural milieu of Serbia and acceptable for testing on patients with CD and BSP. In the 3rd step, we performed patient testing with the aim to test the translated questionnaire on patients to determine whether it was acceptable, whether it was understood in the way it was supposed to, and whether the language used was simple and appropriate. In order to verify acceptability and interpretation of the translated items, the questionnaire was tested on 5 patients, 3 with CD and 2 with BSP. In this phase the CDQ-24 was performed through face-to-face interviews during which the physician-investigator inquired whether the patient had any difficulty in understanding the questionnaire and checked the patient's interpretation of all items. The results of these tests were discussed in the same group of experts, which led to the final Serbian version of the CDQ-24.

The CDQ-24 was filled in by patients in the presence of a physician who assisted the patient, if necessary. A neurologist collected the main clinical data and assessed disability using the Tsui score[13] for CD, the Blepharospasm Rating Scale[14] for BSP and the Dystonia Movement Scale[15] for both.

The internal reliabilities of CDQ-24 (Serbian version) were assessed by Cronbach's alpha coefficient, with a range from 0–1, the latter indicating perfect reliability.

Clinical validity was quantified by the comparison of CDQ-24 subscales scores and the values of the Score for Spasmodic Torticollis[13] for CD, the Blepharospasm Rating

TABLE 1
DEMOGRAPHIC AND CLINICAL CHARACTERISTICS OF PATIENTS WITH CERVICAL DYSTONIA AND BLEPHAROSPASM

Characteristic	Cervical dystonia	Blepharospasm
Number	60	40
Gender		
Male	26 (43.3%)	13 (32.5%)
Female	34 (56.7%)	27 (67.5%)
Current age[a] (years)	51.0±12.1 (26–76)	59.1±11.1 (26–74)
Education (years)		
Primary (1–8)	11 (18.3%)	12 (30.0%)
Secondary (9–12)	36 (60.0%)	15 (37.5%)
University (13+)	13 (21.7%)	13 (32.5%)
Occupation		
Housewife	2 (3.3%)	8 (20.0%)
Manual worker	26 (43.3%)	11 (27.5%)
Clerk	19 (31.7%)	8 (20.0%)
Professional	13 (21.7%)	13 (32.5%)
Current employment status		
Employed	26 (43.3%)	10 (25.0%)
Retired	15 (25.0%)	21 (52.5%)
Unemployed	19 (31.7%)	9 (32.5%)
Marital status		
Married/cohabiting	44 (73.3%)	29 (72.5%)
Single (never married)	9 (15.0%)	2 (5.0%)
Divorced/widowed	7 (11.7%)	9 (22.5%)
Disease duration[a] (years)	8.4±6.4 (1–24)	7.6±7.0 (1–30)
Severity of dystonia [a]		
Dystonia Movement Scale	3.3±3.5 (0–18)	4.3±4.4 (0–20)
Score for Spasmodic Torticollis	5.9±3.3 (0–19)	–
Blepharospasm Rating Scale	–	9.2±3.9 (4–21)
De novo patients[b]	11 (18.3%)	4 (10.0%)

[a] $\overline{X}$±SD (range), [b] No previous botulinum toxin treatment

TABLE 2
DESCRIPTIVE STATISTICS FOR THE CDQ-24 (SERBIAN VERSION)

CDQ-24 subscale	Cervical dystonia $\overline{X}\pm SD$	Blepharospasm $\overline{X}\pm SD$	p	Total sample $\overline{X}\pm SD$
Stigma	40.2±29.3	37.8±32.9	0.707	39.3±30.7
Emotional well-being	37.8±29.6	37.5±32.1	0.958	37.7±30.5
Pain	30.1±24.7	18.9±22.9	0.025	25.6±24.5
Activities of daily living	38.9±20.7	49.3±25.3	0.028	43.1±23.1
Social/family life	32.1±24.9	32.2±28.6	0.995	32.1±26.3
Total score	35.8±22.3	35.1±25.5	0.884	35.6±23.5

Scale[14] for BSP, and the Dystonia Movement Scale[15] for both. Pearson's correlation coefficient (r) was used to investigate the relationship between the scores mentioned above and the main clinical and demographic variables.

Results

One hundred consecutive patients with CD (60) and BSP (40) were included in CDQ-24 (Serbian version) validation. Selected demographic and clinical characteristics of our patients with CD and BSP are presented in Table 1.

Most of the patients with CD and BSP accepted the CDQ-24 questionnaire well. The majority of the patients (97%) estimated the questions as clear and understandable. None of the items were interpreted as unpleasant and embarrassing. The mean time to complete the questionnaire was 11 minutes (range, 4–21 minutes).

Descriptive statistics of the CDQ-24 subscales are shown in Table 2, both for the total sample, and for CD and BSP separately. The mean total score of the CDQ-24 was 35.6±23.5 (range 1.7–96.7). Patients with BSP had a better HRQoL score (lower score) in the Pain subscale (p=0.025) compared with the CD patients. However, patients with CD had a better HRQoL score (lower score) in the Activities of Daily Living subscale (p=0.028) compared with patients with BSP.

Internal consistency reliability ranged from 0.81 on the Activities of Daily Living subscale to 0.97 on the Stigma, the Emotional Wellbeing and the Overall Quality of Life subscales (Table 3).

There was a weak statistically non-significant correlation between the scores of the CDQ-24 subscales and the Score for Spasmodic Torticollis, except for the Activities of Daily Living subscale (r=0.381, p<0.01). A statistically significant positive correlation was also found for the Overall Quality of Life score and the Score for Spasmodic Torticollis (r=0.255, p<0.05) (Table 4).

In BSP patients, a statistically significant positive correlation was found between the Blepharospasm Rating Scale and all of the CDQ-24 subscales, except for the Pain subscale (Table 5).

According to the findings shown in Table 6, a statistically significant positive correlation was registered between the Dystonia Movement Scale score and all CDQ--24 scales, except for the Emotional Wellbeing subscale in CD patients, and the Pain subscale in BSP patients.

TABLE 3
INTERNAL CONSISTENCY RELIABILITY FOR THE CDQ-24

CDQ-24 subscale	Number of items	Cronbach's Alpha (Serbian version)	Cronbach's Alpha (Original version)
Stigma	6	0.97	0.89
Emotional well-being	5	0.97	0.88
Pain	3	0.84	0.79
Activities of daily living	6	0.81	0.77
Social/family life	4	0.90	0.85
Total score	24	0.97	0.94

TABLE 4
CLINICAL VALIDITY: CORRELATION OF CDQ-24 (SERBIAN VERSION) WITH SCORE FOR SPASMODIC TORTICOLLIS IN PATIENTS WITH CERVICAL DYSTONIA

CDQ-24 subscale	Pearson Correlation
Stigma	0.201
Emotional well-being	0.103
Pain	0.245
Activities of daily living	0.381**
Social/family life	0.226
Total score	0.255*

*p<0.05, **p<0.01

TABLE 5
CLINICAL VALIDITY: CORRELATION OF CDQ-24 (SERBIAN VERSION) WITH BLEPHAROSPASM RATING SCALE IN PATIENTS WITH BLEPHAROSPASM

CDQ-24 subscale	Pearson Correlation
Stigma	0.428**
Emotional well-being	0.460**
Pain	0.165
Activities of daily living	0.535**
Social/family life	0.525**
Total score	0.480**

*p<0.05, **p<0.01

TABLE 6
CLINICAL VALIDITY: CORRELATION OF CDQ-24
(SERBIAN VERSION) WITH DYSTONIA MOVEMENT SCALE

CDQ-24 subscale	Pearson Correlation (Cervical dystonia)	Pearson Correlation (Blepharospasm)
Stigma	0.290*	0.480**
Emotional well-being	0.219	0.483**
Pain	0.394**	0.22
Activities of daily living	0.445**	0.520**
Social/family life	0.400**	0.541**
Total score	0.393**	0.510**

*$p < 0.05$, **$p < 0.01$

Discussion and Conclusion

The Medical Outcome Study 36-item Short Form Health Survey (SF-36) is the most widely used generic measure of health status and has been already used in dystonia[5,6]. A recent study revealed that HRQoL in CD patients is mainly predicted by factors such as self-esteem, self-deprecation, social participation, social support, stigma, anxiety and depression, all of which are not addressed by a generic HRQoL instrument[4,7,16].

The CDQ-24 was created as a disease-specific quality of life instrument that addressed the special perceptions and concerns of CD and BSP patients, and can be used to evaluate the impact of the disease on areas not covered by generic measures[7]. The CDQ-24 has revealed good reliability properties according to generally accepted criteria in quality of life assessment[17]. It does mean that the scales of CDQ-24 are appropriately measured.

In our study, the mean total CDQ-24 score was 35.6± 23.5, which is identical to the one obtained in the original study[7]. In both studies, BSP patients scored statistically significantly better HRQoL in the Pain subscale compared with CD patients. We found that CD patients showed a statistically significantly better HRQoL score in the Activities of Daily Living subscale, compared to BSP patients. The differences are also observed in the Social/family life subscale, i.e., Austrian patients with both CD and BSP scored better in this domain compared to Serbian patients, implicating a lack of psychological support and social functioning of our patients.

In this investigation, statistically significant positive correlations were registered between the Dystonia Movement Scale score and all CDQ-24 scales, except for the Emotional Wellbeing subscale in patients with CD, and the Pain subscale in patients with BSP, implicating acceptable clinical validity. Similarly to the original study[7], CDQ-24 subscores showed a weak correlation with the Score for Spasmodic Torticollis in CD patients. We did not find a reasonable explanation for absence of these correlations in the extant literature. In agreement with these findings, it has been taken into consideration that the Score for Spasmodic Torticollis is based on objective neurologic examination, while the quality of life scores represent subjective patients' perception. In other words, some patients with severe CD might ignore their difficulties caused by the disease and show better scores in the quality of life assessment.

In conclusion, the Serbian version of CDQ-24 should be recommended for HRQoL evaluation among patients with both CD and BSP, as an important outcome measure.

Acknowledgements

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UPITNIK KRANIOCERVIKALNE DISTONIJE (CDQ-24): VALJANOST I KULTURALNA PRILAGODBA ZA SRPSKE PACIJENTE

S A Ž E T A K

Cilj ove studije je istražiti valjanost prevedenog i kulturalno prilagođenog upitnika kraniocervikalne distonije (CDQ-24) na grupi srpskih pacijenata. Studija je sadržavala 100 pacijenata s idiopatskom cervikalnom distonijom (CD) i blefarospazmom (BSP) koji su bili ispitani na Institutu za neurologiju, Sveučilišnog kliničkog centra Srbije u Beogradu, u periodu od ožujka do lipnja 2007. godine. Lingvistička valjanost CDQ-24 uključivala je 3 koraka, prema međunarodno prihvaćenoj metodologiji. Većina pacijenata s CD i BSP prihvatila je upitnik. Interna dosljednost pouzdanosti rangirala je od 0,81 do 0,97. Aritmetička sredina sveukupnog rezultata je 35,6±23,5. Pacijenti s BSP imali su bolje rezultate kvalitete života povezane sa zdravljem (HRQoL) na podskali boli (p=0,025) u odnosu na pacijente sa CD. Međutim, pacijenti sa CD imali su bolje HRQoL rezultate na podskali aktivnosti svakodnevnog života (p=0,028) u odnosu na pacijente s BSP. Statistički zančajna pozitivna korelacija registrirana je između rezultata skale distonijskog micanja i gotovo svih CDQ-24 rezultata. Srpska inačica CDQ-24 trebala bi biti preporučena za evaluaciju HRQoL među pacijentima sa CD i BSP kao važna ishodišna mjera.