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# Clinimetric Testing of the Comprehensive Cervical Dystonia Rating Scale

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## Abstract

**Objective—**To test the clinimetric properties of the Comprehensive Cervical Dystonia Rating Scale.

**Background—**This is a modular scale with modifications of the Toronto Western Spasmodic Torticollis Rating Scale (composed of three subscales assessing motor severity, disability and pain) now referred to as the revised Toronto Western Spasmodic Torticollis Scale-2.; a newly developed psychiatric screening instrument; and the Cervical Dystonia Impact Profile-58 as a quality of life measure.

**Methods—**Ten dystonia experts rated subjects with cervical dystonia using the comprehensive scale. Clinimetric techniques assessed each module of the scale for reliability, item correlation and factor structure.

**Results—**There were 208 cervical dystonia patients (73% women, age 59±10 years, duration 15±12 years). The internal consistency of the motor severity subscale was acceptable (Cronbach's alpha = 0.57). Item to total correlations showed that elimination of items with low correlations (<0.20) increased alpha to 0.71. Internal consistency estimates for the subscales for disability and pain were 0.88 and 0.95 respectively. The psychiatric screening scale had a Cronbach's alpha of 0.84 and satisfactory item to total correlations. When the subscales of the Toronto Western Spasmodic Torticollis scale -2 were combined with the psychiatric screening scale, Cronbach's alpha was 0.88, and construct validity assessment demonstrated four rational factors: motor, disability, pain and psychiatric disorders. The Cervical Dystonia Impact Profile-58 had an alpha of 0.98 and its construction was validated through a confirmatory factor analysis.

**Conclusions—**The modules of the Comprehensive Cervical Dystonia Rating Scale are internally consistent with a logical factor structure.

### Keywords

Cervical dystonia; focal dystonia; rating scale; Toronto Western Spasmodic Torticollis Rating Scale; Cervical Dystonia Impact Profile-58

## Introduction

Cervical dystonia (CD) is a complex disorder marked by involuntary movements of neck and shoulders, pain, impaired activities of daily living and reduced quality of life. The abnormal movements often combine head turn, tilt, forward or backward flexion, anterior or sagittal shift and shoulder elevation.[1, 2] The involuntary movements are associated with significant disability. In addition, pain occurs in 75% of patients and contributes to a greater degree of disability.[3] CD has also been associated with psychiatric disorders, including depression, anxiety, panic disorders and social phobia.[4–7] Furthermore, several studies have also demonstrated impaired health-related QOL in CD[8–14].

Although there have been many rating scales developed for motor symptoms of CD[15], only 3 of these, the Tsui scale[16], the Cervical Dystonia Severity Scale[17] and the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) have had clinimetric evaluation. None of these scales address the psychiatric symptoms or quality of life. The Tsui rating scale is a 6-item scale that assesses amplitude and duration of involuntary neck movements, shoulder elevation, and head tremor[16]. This scale is designed to assess head and shoulder postures and head tremor but does not take into account the other manifestations of CD. The Cervical Dystonia Severity Scale uses a protractor and wall chart to rate angles of head deviation from neutral in each of three planes[17]. This scale does not evaluate shoulder elevation, tremor or sagittal shift. The Tsui rating scale and CDSS do not address pain, activities of daily living, psychiatric symptoms, or quality of life.

The standard TWSTRS consists of three domains that assess motor severity, pain, and disability[18]. The motor severity subscale consists of 10 items, with variable scaling and weighting. It also includes a disability scale with 6 items, and a pain scale with 3 items. The total score is the sum of each of the subscales. Only the motor domain has undergone evaluation for inter-rater reliability and construct validity, with good to excellent inter-rater reliability[19]. Despite the limited clinimetric studies of the TWSTRS, it has been used extensively in clinical studies of CD and is the scale currently recommended by the Movement Disorder Society task force on dystonia rating scales.[15]

There are no psychiatric rating scales validated for use in CD. While the DSM-V criteria are the gold standard for diagnosis of psychiatric disease, their application requires specific training and is impractical for routine use by most CD providers. There are several self-administered scales that are easy to administer, require no examiner training, and have been assessed for clinimetric properties in primary depression and anxiety. The Beck Depression Inventory[20] is a self-administered scale with 21 components that takes 10 – 15 min to complete. This scale does not emphasize somatic components and therefore avoids the confounding factors of the movement associated with CD.[21] The Hospital Anxiety and Depression Rating Scale is a self-administered scale that consists of 14-item subscales for both depression and anxiety.[22, 23] This scale was specifically developed for use in patients with somatic co-morbidity and has no questions related to the physical signs of depression or anxiety. The Beck Anxiety Index is a self-reported scale[21] designed as a screening tool for anxiety with good positive predictive value for panic disorders.[24] Although these psychiatric rating scales are all well validated in psychiatric practice, they have not been systematically applied to CD.

The effect of CD on quality of life is comparable to that seen in multiple sclerosis, Parkinson's disease, stroke[14] and other chronic diseases.[25] Standard measures of QOL, including generic health-related QOL,[25] EuroQoL, SF-36 and Rosenbergs' self-esteem scale[26] are not consistent in identifying factors predicting reduced quality of life in CD and do not correlate with effective treatment for CD, such as botulinum toxin injections.[12, 27] The Craniocervical Dystonia Questionnaire (CDQ-24), although designed specifically for blepharospasm and CD, has not been extensively used or tested against other scales.[28] The Cervical Dystonia Impact Profile – 58 item (CDIP-58) was developed using a modified Delphi method with Rasch methodology.[29] It is a self-administered scale with 8 subscales

measuring the impact of head and neck symptoms on a variety of quality of life items.[29] The CDIP-58 has been evaluated for reliability and validity in CD[30] and shown to be superior to the SF-36, a widely used but generic QOL measure. The CDIP-58 also demonstrates sensitivity to change following botulinum toxin injections[29, 31] and is the recommended scale for quality of life in CD.[15]

In this study, the original TWSTRS was revised to the TWSTRS-2 to address identified deficiencies, including the variable scaling of items, the lack of an item for head tremor and the weighting of the duration factor by two.[32] The TWSTRS-PSYCH was developed to screen for psychiatric disorders associated with CD. The CDIP-58 with previously established reliability and validity was included in its original form. We combined the TWSTRS-2, TWSTRS-PSYCH and the CDIP-58 to produce the modular Comprehensive Cervical Dystonia Rating Scale or CCDRS. The specific aim of the study was to assess the reliability and construct validity of the CCDRS.

## Methods

The methods for development of the CCDRS have been described in a prior publication.[32] Briefly, the existing TWSTRS motor severity was revised to the TWSTRS-2 motor severity using a modified Delphi method with input from dystonia experts. The TWSTRS–PSYCH was developed using a similar methodology with input from psychiatrists, dystonia experts and patients. The draft TWSTRS-2 included assessments for motor severity (12 items), pain (5 items) and disability (6 items). The TWSTRS-PSYCH included 6 items rated on a 5-point scale from 0 (absent) to 4 (severe) for occurrence over the past month (Figure 2). The maximal score of the TWSTRS-PSYCH was 24. The CDIP-58 includes 58 self-administered questions that define 8 subscales and are transformed into a total score, with a maximal score of 100. The TWSTRS-2, TWSTRS-PYSCH, and CDIP-58 then were combined into the CCDRS and used in the data collection phase of the study along with other demographic and disease-related measures.

Subjects with isolated CD, previously known as primary dystonia, were recruited from 10 sites. Demographic information, including age, gender, and duration of CD were collected. For this study, subjects were videotaped using a standardized protocol during the time that the site investigator rated the subject severity using the TWSTRS-2 motor severity subscale.[32] Subjects were interviewed to complete the TWSTRS-2 disability and pain subscales, as well as the TWSTRS-PSYCH. The subjects completed the self-reported CDIP-58.

There are no accepted formulae for calculating required sample sizes for scale validation studies, particularly factor analytic methods, at given levels of power[33]. Instead, recommended subject-to-item ratios are employed. For the present study, we have a 9.1:1 subject-to-item ratio, which exceeds the recommended 8:1 ratio shown to be adequate for this analysis[34, 35].

Rating scores and video were electronically sent to a central database at Washington University, St. Louis, MO[36]. The video and data were assessed for completeness. Queries

regarding missing data were resolved. Accuracy of the data entry was verified through cross referencing electronic data to paper data collection forms in 10% of cases.

## Statistical approach

Subject demographics and disease-related variables were examined using frequency counts and measures of central tendency and variability, as appropriate. To assess the reliability and validity of the TWSTRS-2 and TWSTRS - PSYCH components of the CCDRS, we employed both Classical Test Theory (CRT) and Item Response Theory (IRT). Classical Test Theory focuses on the relationships of individual items to the entire scale[37] while Item Response Theory focuses on the measurement characteristics of the items in relation to the individual completing the scale[37]. Using Classical Test Theory we examined Cronbach's alpha, a measure of scale reliability, item-to-total correlations, changes in alpha if selected items were removed and distributional skewness, a measure of potential floor or ceiling effects, for the separate subscales of the TWSTRS-2 (motor severity, disability, pain) and TWSTRS-PSYCH modules of the CCDRS. These analyses were conducted using SPSS (Version 21). Additionally, we examined the construct validity through exploratory factor analyses. Because of the ordered categorical level of measurement of the CCDRS we employed an unweighted least squares approach for the factor estimate and a CF-Varimax orthogonal rotation to improve the interpretability of the factors. MPlus (Version 7) was used for these analyses. For the Item Response Theory approach, we used a graded response model analysis with maximum likelihood parameter estimation[38] to examine item discrimination, or the strength of the relationship between the item and the measured domain, and item threshold, or the level of item response to the overall severity of the measured domain. MPlus (version 7) was used for these analyses.

To assess each item's utility in the CCDRS, we identified items with low item-to-total correlations (defined as ≤ 0.3), improvement in Cronbach's alpha if omitted, low factor loading (defined as ≤ 0.4), a skewness outside of the range −1.50 to +1.50 representing possible floor or ceiling effects, non-significant Item Response Theory discrimination scores and thresholds that did not encompass a value of zero. Based on this assessment, each item was considered either as one to keep in the scale or as one to drop or modify. If an item met the criteria for acceptable item-to-total correlation, change in alpha if the item were omitted, appropriate factor loading, skewness and Item Response Theory discrimination and threshold, it was retained. Items not meeting these criteria were deleted.

Because the CDIP-58 module of the CCDRS had already undergone clinimetric examination for reliability and validity, we limited our analysis to assessments of internal consistency (Cronbach's alpha) and confirmatory factor structure (CFA). The CFA was conducted to determine if the 8 factors found in the original publication[29] could be confirmed with the data collected for this study. We evaluated the CFA results based on the Comparative Fit Index (CFI)[39]. To confirm a good fit between the original factor structure and our data, the CFI was required to be 0.90 or greater. Mean and variance adjusted weighted least square (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, with values less or equal than 0.10 indicating an acceptable index. MPlus (version 7) was used for these analyses.

## Results

A total of 208 CD subjects (73% women, mean age 59 years SD± 9.95), onset of CD 44 years (SD ± 12.11) from 10 sites in the United States were included. The mean severity of CD as measured using the TWSTRS 2 total score was (33.24 (SD ± 13.22)), with subscale scores for motor severity of (16.29 (SD ± 5.54)), disability (9.21 (SD ± 5.72)) and pain (7.88 (SD ± 5.56))

### TWSTRS-2 Motor Severity Subscale (Table 1)

Overall Cronbach's alpha for the TWSTRS-2 motor severity subscale was 0.57. Items assessing Rotation, Laterocollis, Shoulder Elevation, Duration, Range of Motion and Time in Midline met criteria for acceptable item-to-total correlation, change in alpha if item omitted, factor loading, skewness, IRT discrimination and IRT threshold. Items assessing anterocollis, retrocollis, lateral shift, sagittal shift, head tremor and effect of a sensory trick failed to meet the criteria for utility in the CCDRS and were deleted from the CCDRS.

### TWSTRS-2 Disability Subscale (Table 1)

Overall Cronbach's alpha for the TWSTRS-2 disability subscale was 0.88. Items assessing Work, Activities of Daily Living, Driving, Reading, Television and Outside of Home Disability met criteria for acceptable item-to-total correlation, change in alpha if item omitted, factor loading, skewness, IRT discrimination and IRT threshold. All items met the criteria for utility in the CCDRS and were retained in the CCDRS.

### TWSTRS-2 Pain Subscale (Table 1)

Overall Cronbach's alpha for the TWSTRS-2 pain subscale was 0.95. Items assessing Pain at its Best, Pain at its Worst, Usual Pain, Pain Duration and Pain Disability met criteria for acceptable item-to-total correlation, change in alpha if item omitted, factor loading, skewness, IRT discrimination and IRT threshold. All items met the criteria for utility in the CCDRS and were retained in the CCDRS. The revised TWSTRS-2 scale is included in figure 1.

### TWSTRS- PSYCH (Table 1)

Overall Cronbach's alpha for the TWSTRS-PSYCH was 0.84. Items assessing Depression, Loss of Interest, Discomfort and Anxiety met criteria for acceptable item-to-total correlation, change in alpha if item omitted, factor loading, skewness, IRT discrimination and IRT threshold and were retained in the CCDRS. Items assessing Panic and Afraid of Going Outside met all criteria except for skewness. The skewed distribution appears to be due to the high percentages of zero scores for Panic (88%) and Afraid of Going Outside (82%). The TWSTRS-PSYCH scale is included in figure 2.

### Combined TWSTRS-2 and TWSTRS-PSYCH

Overall Cronbach's alpha for the combined TWSTRS2 (after removing items assessing anterocollis, retrocollis, lateral shift, sagittal shift, head tremor and effect of a sensory trick) and TWSTRS-PSYCH was 0.88. All items met criteria for acceptable item-to-total

correlation, change in alpha if item omitted, skewness, IRT discrimination and IRT threshold. Factor analysis revealed a satisfactory four-factor solution with items assessing motor severity, disability, pain and psychiatric manifestation loading on separate factors (all factor loadings > 0.40) (Table 2).

### CDIP-58

Overall Cronbach's alpha for the CDIP-58 was 0.98. The CFA of the 8-factor solution of the original CDIP-58 resulted in a CFI of 0.97 with a RMSEA of 0.07 and a model fit chi-square of 48.96 ($p < 0.0005$) using the data from the current study. Thus the pre-specified 8-factor structure was confirmed.

## Discussion

This study demonstrates that the CCDRS assesses distinct components of CD and can be applied as a complete scale or used in a modular format. The current study provides a realistic picture of the clinimetric properties of this scale and each of its modules, and allows the deletion of items that do not demonstrate clinical utility.

The revised motor severity subscale of the TWSTRS-2 demonstrated that certain items (anterocollis, retrocollis, lateral shift, sagittal shift, head tremor and effect of sensory trick) had multiple indicators of poor utility on both CCT and IRT analyses. The reasons for the lack of utility of these items are varied. Anterocollis, retrocollis, lateral shift and sagittal shift ratings had highly skewed distributions, suggesting possible floor-effects. Head tremor and effect of sensory trick had more normal-shaped distributions. However, these items had low item-to-total correlations, and increased the alpha if omitted. Further, the low factor loading of these items indicates that they may not directly contribute to overall CD severity in contrast to the other items, although these may be features of the disorder. Hence, these items were deleted from the rating of motor severity, resulting in a simplified scale that can be used efficiently in a clinical study (Figure 1)

The TWSTRS-2 disability subscale, which was unchanged from the standard TWSTRS, was not revised and had good reliability and content validity. The TWSTRS-2 pain subscale was revised, removing the mathematical manipulations (the multiplication of the usual level of pain by 2 and eliminating the division of the pain scores by 4), and was found to be reliable and valid. The first psychiatric screening tool for CD, TWSTRS-PSYCH (Figure 2), demonstrated good clinimetric properties. The CDIP-58, which has previously been assessed for reliability and validity using a different scale development technique, was found to have acceptable internal consistency and a confirmed factor structure of 8 factors. Inclusion of the CDIP-58 provides a patient reported measure of the impact of CD on quality of life that is distinct from information provided by the other scales in the CCDRS.

Although many rating scales have been developed to evaluate CD, none has been comprehensive[10, 15, 40, 41]. The CCDRS includes measures for motor severity, disability, pain, psychiatric disorders and quality of life measures. Each of these domains may be affected in CD and contribute to overall severity of the condition. The reduction in total items in the TWSTRS-2 motor severity subscale based on these results will allow for easier

use. While the deleted items may be useful as descriptors for CD, these items do not contribute to the overall assessment of CD severity

The results of the factor analysis for the modified TWSTRS-2 and TWSTRS-PSYCH suggest that the scores of the four subcomponents (motor severity, disability, pain and psychiatric concerns) can be used either as independent measures or summarized into a single measure of CD impairment. The previously defined factor structure of the CDIP-58 was confirmed in the present analysis.

The CCDRS provides a tool that allows an assessment of all aspects of CD and can be used in modular format. This study provides the framework for development of rating scales that can be used to assess the varied clinical aspects of focal dystonias involving other body regions. As new therapeutic modalities become available for the treatment of the focal dystonias[42], it is critical that validated outcome measures capture not only the motor features, but also those related to psychological disorders and impact on quality of life.

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## I TWSTRS-2 SEVERITY

### Maximal excursion for items 1-8

The first section of the Severity scale is maximal excursion. This section has rating items for the amplitude of excursion with patients allowing their head and neck to assume the spontaneous abnormal posture, without opposing the movement, during the maneuvers indicated by the videotape examination protocol. The angle of movement is determined for each axis of head movement, shifting of the neck on the shoulders in a forward or backward direction, and shoulder movement.

In scoring each item, it is important to score only for that particular posture. For example, the score for rotation would only include the degree of horizontal deviation separate from the other components of movement observed.

- **For each item, full range is considered the range that a normal person without dystonia can achieve at maximal effort in a particular direction**
- **If a rating lies between two scores, the greater score is marked. There are no 0.5 scores accepted.**

### 1.Rotation (horizontal turn: right or left)

Rotation is defined as the movement of the head along the horizontal axis. The movement of the chin from the midline position to right or left is best seen in the frontal view. In the mid-position, the chin is positioned directly over the sternum, midway between the attachments of the clavicles. Rotation is scored by the greatest degree of deflection from the mid-position.

| | |
|---|---|
| 0 | None |
| 1 | Slight (less than 25% full range) (1 - 22 degrees off midline) |
| 2 | Mild (25 to less than 50% of full range) (23 - 45 degrees off midline) |
| 3 | Moderate (50 to less than 75% of full range), (46 - 67 degrees off midline) |
| 4 | Severe (>75% of full range) (68 - 90 degrees off midline) |

### 2. Laterocollis (tilt right or left, exclude shoulder elevation)

Laterocollis refers to the angle of tilting of the head to the right or left but excludes shoulder elevation. As in rotation, the maximum deviation in a lateral direction is the score to be recorded. A technique for determining head tilt or laterocollis is to draw a line between the eyes or the ears and compare this line to the horizontal plane.

| | |
|---|---|
| 0 | None |
| 1 | Slight (less than 25% full range, 1-22 degrees of tilt) |
| 2 | Mild (25 to less than 50% of full range, 23-45 degrees of tilt ) |
| 3 | Moderate (50 to less than 75% of full range 46-67 degrees of tilt), |
| 4 | Severe (>75% of full range, 67 to 90 degrees of tilt) |

**3. Shoulder elevation/anterior displacement**

This category includes an assessment of the severity of shoulder movement, as well as a duration factor for the shoulder. Shoulder elevation is best evaluated from a frontal or posterior view. Anterior or posterior displacement of the shoulder is best viewed from a lateral or profile view.

| | |
|---|---|
| 0 | Absent |
| 1 | Slight (< 25% full range) intermittent or constant |
| 2 | Mild (greater than 25% but less than 50% of full range) intermittent or constant |
| 3 | Moderate (greater than 50% but less than 75% of full range) intermittent or constant |
| 4 | Severe (greater than 75% of full range) intermittent or constant |

**4. Duration of cervical dystonia during examination**

Duration of cervical dystonia is determined during the course of the entire standard examination session and is an assessment of head deviation in any direction. It consists of two components: (a) the percentage of time during the entire examination that head deviation is present AND (b) the relative intensity of the head deviation during the examination (e.g. when present during the session, the head deviation was most often submaximally or maximally present).

Note that the duration of shoulder movement is not considered in this category, but is rated below in another section

| | |
|---|---|
| 0 | None |
| 1 | Occasional deviation (< 25% of the time), either maximal or submaximal |
| 2 | Intermittent deviation (25 - 50% of the time) either maximal or submaximal<br>or<br>Frequent deviation (50 - 75% of the time), most often submaximal |
| 3 | Frequent deviation (50 - 75% of the time), most often maximal<br>or<br>Constant deviation (> 75% of the time), most often submaximal |
| 4 | Constant deviation (> 75% of the time), most often maximal |

**5. Range of Motion of head and neck**

The range of motion category assesses the ability of the patient to move from the abnormal posture through the midline to the extreme opposite position without the aid of a sensory trick. Range of motion is assessed for each of the three axis of head movement: horizontal rotation, flexion/extension, and lateral tilting. The score for the most severely limited direction of movement is the final range of motion score.

| | |
|---|---|
| 0 | Able to move to extreme opposite position |
| 1 | Able to move head well past midline but not to extreme opposite position |
| 2 | Able to move head barely past midline |
| 3 | Able to move head toward but not past midline |
| 4 | Barely able to move head beyond abnormal posture |

**6. Time holding head in midline**

This item assesses the ability of the patient to hold the head within 10 degrees of the midline, normal head position. Obtaining midline position may be done using verbal direction. Obtaining the midline marks the beginning of the time measure. The ability to remain in midline is obtained twice and the mean duration up to 60 seconds for each attempt is averaged to obtain the score. If the patient cannot reach midline, the score is 4.

| | |
|---|---|
| 0 | > 60 sec |
| 1 | 46 - 60 sec |
| 2 | 31 - 45 sec |
| 3 | 16 - 30 sec |
| 4 | < 15 sec |

**Total TWSTRS-2 Severity Score: Sum of 1-6, Maximal Score 24**

**3. Shoulder elevation/anterior displacement**

This category includes an assessment of the severity of shoulder movement, as well as a duration factor for the shoulder. Shoulder elevation is best evaluated from a frontal or posterior view. Anterior or posterior displacement of the shoulder is best viewed from a lateral or profile view.

| | |
|---|---|
| 0 | Absent |
| 1 | Slight (< 25% full range) intermittent or constant |
| 2 | Mild (greater than 25% but less than 50% of full range) intermittent or constant |
| 3 | Moderate (greater than 50% but less than 75% of full range) intermittent or constant |
| 4 | Severe (greater than 75% of full range) intermittent or constant |

**4. Duration of cervical dystonia during examination**

Duration of cervical dystonia is determined during the course of the entire standard examination session and is an assessment of head deviation in any direction. It consists of two components: (a) the percentage of time during the entire examination that head deviation is present AND (b) the relative intensity of the head deviation during the examination (e.g. when present during the session, the head deviation was most often submaximally or maximally present).

Note that the duration of shoulder movement is not considered in this category, but is rated below in another section

| | |
|---|---|
| 0 | None |
| 1 | Occasional deviation (< 25% of the time), either maximal or submaximal |
| 2 | Intermittent deviation (25 - 50% of the time) either maximal or submaximal<br>or<br>Frequent deviation (50 - 75% of the time), most often submaximal |
| 3 | Frequent deviation (50 - 75% of the time), most often maximal<br>or<br>Constant deviation (> 75% of the time), most often submaximal |
| 4 | Constant deviation (> 75% of the time), most often maximal |

**5. Range of Motion of head and neck**

The range of motion category assesses the ability of the patient to move from the abnormal posture through the midline to the extreme opposite position without the aid of a sensory trick. Range of motion is assessed for each of the three axis of head movement: horizontal rotation, flexion/extension, and lateral tilting. The score for the most severely limited direction of movement is the final range of motion score.

| | |
|---|---|
| 0 | Able to move to extreme opposite position |
| 1 | Able to move head well past midline but not to extreme opposite position |
| 2 | Able to move head barely past midline |
| 3 | Able to move head toward but not past midline |
| 4 | Barely able to move head beyond abnormal posture |

**6. Time holding head in midline**

This item assesses the ability of the patient to hold the head within 10 degrees of the midline, normal head position. Obtaining midline position may be done using verbal direction. Obtaining the midline marks the beginning of the time measure. The ability to remain in midline is obtained twice and the mean duration up to 60 seconds for each attempt is averaged to obtain the score. If the patient cannot reach midline, the score is 4.

| | |
|---|---|
| 0 | > 60 sec |
| 1 | 46 - 60 sec |
| 2 | 31 - 45 sec |
| 3 | 16 - 30 sec |
| 4 | < 15 sec |

**Total TWSTRS-2 Severity Score: Sum of 1-6, Maximal Score 24**

6. **Activities outside the home** (e.g. shopping, walking about, movies, dining and other recreational activities)

0 No difficulty
1 Unlimited activities but bothered by torticollis
2 Unlimited activities but requires simple "tricks" to accomplish
3 Only accomplishes activities when accompanied by others because of torticollis
4 Limited activities outside home; certain activities impossible or given up due to torticollis
5 Rarely if ever engages in activities outside the home

**TOTAL DISABILITY SCORE = Sum 1-6 - Maximum Score = 30**

## III. TWSTRS-2 PAIN SCALE

1. Rate the **severity** of neck pain during the last week on a scale of 0-10 where a score of 1 represents a minimal ache and 10 represents the most excruciating pain imaginable

Best 0-10
Worst 0-10
Usual 0-10

2. Rate the duration of neck pain

0 None
1 Present <10% of the time
2 Present 10% - <25% of the time
3 Present 25% - <50% of the time
4 Present 50% - <75% of the time
5 Present >75% of the time

3. Rate the degree to which pain contributes to **disability**

0 No limitation or interference from pain
1 Pain is quite bothersome but not a source of disability
2 Pain definitely interferes with some tasks but is not a major contributor to disability
3 Pain accounts for some (less than half) but not all disability
4 Pain is a major source of difficulty with activities; separate from this, head pulling is also a source of some (less than half) disability
5 Pain is the major source of disability; without it most impaired activities could be performed quite satisfactorily despite the head pulling

**TOTAL PAIN SCALE SCORE = Sum 1-3 - Maximum Score = 40**

**TOTAL TWSTRS-2 = Sum of severity, disability and pain. Maximal Score: 94**

**Figure 1.**
Toronto Western Spasmodic Torticollis Rating Scale-2 (TWSTRS-2)

1. In the last month has there been a period of time when you were feeling depressed or down?

| | |
|---|---|
| 0 | Absent |
| 1 | Mild: occasional sadness in keeping with the circumstances |
| 2 | Moderate: sad or low but brightens up without difficulty |
| 3 | Marked: Pervasive feelings of sadness or gloominess. The mood is still influenced by external circumstances. |
| 4 | Severe: Continuous or unvarying sadness, misery or despondency |

2. In the last month have you lost interest or pleasure in things you usually enjoyed? (mark according to subjective experience of interest as opposed to actual ability to perform an action)

| | |
|---|---|
| 0 | Normal interest in surroundings and in other people |
| 1 | Mild: reduced ability to enjoy usual interests, activities, hobbies, people, or work but no reduction in initiation of activities |
| 2 | Moderate: moderate loss of interest in activities, hobbies, or work such that it is difficult to initiate activities |
| 3 | Marked: marked loss of interest in surroundings and loss of interest in being with friends and acquaintances with marked reduction in initiation of activities |
| 4 | Severe: continuous and virtually unremitting loss of interest in all activities, including social activities even with the closest friends and relatives; inability to initiate activities |

3. Over the past month has there anything that you have been afraid to do or felt uncomfortable doing in front of other people, like speaking, eating or writing?

| | |
|---|---|
| 0 | Absent |
| 1 | Mild: anxious in some social settings but continues to participate |
| 2 | Moderate: anxious in most social settings + avoidance of some activities involving large groups or being centre of attention (eg raising a toast, asking questions in forum etc) |
| 3 | Marked: pronounced anxiety in most social settings + avoidance of most activities except for 1 or 2 activities |
| 4 | Severe: pronounced anxiety + avoidance of all social settings except in company of closest family/caregivers |

4. In the past month have you been particularly nervous or anxious?

| | |
|---|---|
| 0 | Absent |
| 1 | Mild: worries a little more than necessary about minor matters but with only mild distress |
| 2 | Moderate: intrusive anxious thoughts out of proportion to patient's situation but able to dispel or dismiss them |
| 3 | Marked: continuous worry fluctuating in intensity, distressing thoughts may cease for an hour or two, especially if distracted by an activity requiring attention |
| 4 | Severe: virtually unrelenting dread or anxiety |

5. In the past month have you had a panic attack, when you suddenly felt frightened or suddenly developed a lot of physical symptoms? Common physical symptoms are: palpitations, sweating, trembling or shaking, SOB or smothering, chest pain, nausea, dizzy/faint, paresthesias, chills or hot flushes.

| | |
|---|---|
| 0 | None |
| 1 | Mild: rare episodes (less than monthly) of panic precipitated by specific triggers, |
| 2 | Moderate: at least 2 panic attacks/month + some anticipatory anxiety of recurrence without any avoidance |
| 3 | Marked: at least weekly panic attacks + marked anticipatory anxiety (fear of recurrence) + some avoidance behaviour. |
| 4 | Severe: panic attacks almost daily + pronounced worry of recurrence + significant avoidance |

6. In the past month have you been afraid of going out of the house alone, being in crowds, standing in a line, or traveling on buses or trains?

| | |
|---|---|
| 0 | None |
| 1 | Mild: some discomfort in few specific settings eg lectures, buses, public transport |
| 2 | Moderate: avoids some settings |
| 3 | Marked: avoids most settings |
| 4 | Severe: very rarely if ever leaves home alone |

Total TWSTRS-PSYCH: sum of items 1-6. Maximal Score 24

**Figure 2. TWSTRS-PSYCH**
Circle one number for each question.

**Table 1**

Classical test theory and item response analyses results for items on the TWSTRS-2 Motor, TWSTRS-2 Disability, TWSTRS-2 Pain and TWSTRS-2 Psych components of the Comprehensive Cervical Dystonia Rating Scale.

| Item | Item-Total Correlation | Alpha-if-Item-Removed | Factor Loading | Skewness | IRT Discrimination (p value) | IRT Threshold (Min; Max) | Action |
|---|---|---|---|---|---|---|---|
| TWSTRS-2 Motor | | | | | | | |
| Rotation | 0.314 | No Increase | 0.420 | 0.314 | 1.00 (<0.0005) | −2.75; 2.50 | Keep |
| Laterocollis | 0.466 | No Increase | 0.519 | 0.608 | 1.40 (<0.0005) | −2.21; 6.22 | Keep |
| Anterocollis | 0.180 | Increased | <0.3 | 1.780 | 0.69 (0.06) | 0.68; 3.53 | Drop/Modify |
| Retrocollis | 0.091 | Increased | <0.3 | 1.536 | 0.17 (0.44) | 0.62; 5.31 | Drop/Modify |
| Lateral Shift | −0.068 | Increased | <0.3 | 1.723 | −0.24 (0.19) | 0.71; 5.35 | Drop/Modify |
| Sagittal Shift | 0.136 | Increased | <0.3 | 1.575 | 0.34 (0.05) | 0.45; 5.37 | Drop/Modify |
| Head Tremor | 0.000 | Increased | <0.3 | 0.472 | 0.04 (0.84) | −0.59; 3.91 | Drop |
| Shoulder Elevation | 0.349 | No Increase | 0.481 | 0.542 | 1.11 (<0.0005) | −1.79; 2.88 | Keep |
| Duration | 0.527 | No Increase | 0.626 | −0.648 | 1.58 (<0.0005) | −6.53; 1.06 | Keep |
| Sensory Trick | 0.011 | Increased | <0.3 | 1.107 | 0.09 (0.63) | −1.42; 2.23 | Drop/Modify |
| ROM | 0.373 | No Increase | 0.428 | 0.877 | 1.16 (<0.0005) | −1.81; 3.88 | Keep |
| Time in Midline | 0.472 | No Increase | 0.631 | −0.563 | 1.60 (<0.0005) | −1.59; 0.49 | Keep |
| TWSTRS- 2 Disability | | | | | | | |
| Work | 0.722 | No Increase | 0.704 | 0.483 | 1.00 (<0.0005) | −1.46; 5.50 | Keep |
| ADL | 0.654 | No Increase | 0.604 | 0.774 | 1.66 (<0.0005) | −0.77; 5.45 | Keep |
| Driving | 0.587 | No Increase | 0.542 | 0.294 | 1.40 (<0.0005) | −1.90; 5.13 | Keep |
| Reading | 0.748 | No Increase | 0.805 | 0.183 | 3.32 (<0.0005) | −3.00; 8.12 | Keep |
| Television | 0.734 | No Increase | 0.759 | −0.052 | 3.35 (<0.0005) | −2.92; 6.80 | Keep |
| Outside of Home | 0.728 | No Increase | 0.690 | 0.685 | 2.22 (<0.0005) | −2.00; 5.91 | Keep |
| TWSTRS- 2 Pain | | | | | | | |
| Pain Best | 0.778 | No Increase | 0.752 | 1.13 | 1.000 (<0.0005) | −0.11; 2.92 | Keep |
| Pain Worst | 0.884 | No Increase | 0.848 | −0.210 | 4.853 (<0.0005) | −1.20; 0.96 | Keep |
| Pain Usual | 0.944 | No Increase | 0.900 | 0.312 | 6.169 (<0.0005) | −0.77; 1.87 | Keep |
| Pain Duration | 0.835 | No Increase | 0.778 | −0.169 | 3.564 (<0.0005) | −1.130; 0.55 | Keep |

| Item | Item-Total Correlation | Alpha-if-Item-Removed | Factor Loading | Skewness | IRT Discrimination (p value) | IRT Threshold (Min; Max) | Action |
|---|---|---|---|---|---|---|---|
| Pain Disability | 0.735 | No Increase | 0.669 | 0.472 | 2.320 (<0.0005) | −0.50; 1.74 | Keep |
| TWSTRS- 2 Psych | | | | | | | |
| Depression | 0.657 | No Increase | 0.724 | 0.557 | 1.00 (< 0.0005) | −0.44; 2.38 | Keep |
| Loss of Interests | 0.673 | No Increase | 0.740 | 1.263 | 2.20 (< 0.0005) | 0.22; 2.08 | Keep |
| Discomfort in Public | 0.570 | No Increase | 0.627 | 0.728 | 1.57 (< 0.0005) | −0.17; 2.34 | Keep |
| Anxiety | 0.672 | No Increase | 0.741 | 0.907 | 2.04 (< 0.0005) | −0.22; 2.24 | Keep |
| Panic | 0.559 | No Increase | 0.626 | 3.646 | 2.37 (< 0.0005) | 1.18; 2.57 | Keep |
| Afraid Going Out | 0.576 | No Increase | 0.645 | 2.673 | 2.16 (< 0.0005) | 0.93; 2.85 | Keep |

Note: Item-To-Total = correct item score to total score correlation; Alpha-If-Item-Removed = increase of decrease in Cronbach's alpha if the item is removed from the analysis; Factor Loading = maximum factor loading value; Skewness = a measure of distributional asymmetry; IRT Discrimination = strength of relationship between item and the measured construct; IRT Threshold = level of item response in relation to severity of measured construct, minimum and maximum thresholds displayed.

## Table 2

Factor solution for combined TWSTRS-2 and TWSTRS-PSYCH after deleting TWSTRS-2 Severity items not meeting criteria for inclusion in the Comprehensive Cervical Dystonia Rating Scale.

| Item | Factor | | | |
|---|---|---|---|---|
| | 1 | 2 | 3 | 4 |
| Rotation | | | | .41 |
| Laterocollis | | | | .51 |
| Shoulder elevation | | | | .53 |
| Duration | | | | .62 |
| Range of movement | | | | .42 |
| Time midline | | | | .63 |
| Work disability | | .59 | | |
| ADL disability | | .52 | | |
| Driving disability | | .52 | | |
| Reading disability | | .80 | | |
| Television disability | | .75 | | |
| Outside disability | | .63 | | |
| Pain at best | .76 | | | |
| Pain at worst | .84 | | | |
| Pain at usual | .92 | | | |
| Pain duration | .82 | | | |
| Pain disability | .67 | | | |
| Depression | | | .68 | |
| Loss of interest | | | .70 | |
| Discomfort in public | | | .58 | |
| Anxiety | | | .71 | |
| Panic attack | | | .61 | |
| Fear of outside | | | .62 | |

Note: All factor loadings < 0.40 are not shown in the Table.