

# A new method for the evaluation of cervical dystonia

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### ABSTRACT

**Objectives:** The study aimed to develop a measurement method that yielded objective data for the clinical assessment of cervical dystonia using a Kinect camera system.

**Patients and methods:** This double-blind, parallel-group method development study included 22 patients with cervical dystonia (3 males, 19 females; mean age: 47 years; range, 34 to 60 years) and 20 healthy individuals (13 females, 7 males; mean age: 32 years; age range, 22 to 65). Using cameras and a computer software, the head-neck postures of 40 healthy participants were recorded in the virtual environment. Using the device, 22 patients with cervical dystonia were examined both at rest and while moving with different parts of the body. Two different experts evaluated and scored the cases using the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) and the Tsui scale.

**Results:** A three-way comparison revealed interclass correlations between the coefficients of 0.799 (79.9%) and 0.784 (78.4%) for at rest and with movement, respectively. The two-way comparison of the experts revealed correlation coefficients of 0.717 (71.7%) and 0.692 (69.2%) for at rest and with movement, respectively. A three-way comparison of the device and Expert 1 and Expert 2 TWSTRS scores revealed interrater agreement values of 0.6 and 0.8 (good) and 0.6 and 0.8 (good) while at rest and with movement, respectively. A three-way comparison of the device and Expert 1 and Expert 2 Tsui scores revealed interrater agreement values of 0.6 and 0.8 (good) and 0.6 and 0.8 (good) while at rest and with movement, respectively. A three-way comparison of the device and Expert 1 and Expert 2 Tsui scores revealed interrater agreement values of 0.6 and 0.8 (good) and 0.4 and 0.6 (moderate) while at rest and with movement, respectively.

**Conclusion:** The newly developed system was a sensitive tool for use in the kinematic evaluation of patients with cervical dystonia and could prove beneficial in diagnosis and treatment follow-up.

Keywords: Cervical dystonia, kinect camera, Toronto Western Spasmodic, Torticollis Rating Scale, Tsui.

Cervical dystonia is a hyperkinetic movement disorder that causes abnormal posture in the head and neck. It is characterized by tonic or clonic contractions and is the third most common movement disorder in adults, with an estimated prevalence of 16.4 in 100,000 individuals, affecting 3 million people worldwide.<sup>[1]</sup>

Various evaluation scales have been developed for dystonia. As the first evaluation scale, the Fahn-Marsden scale was proposed by Burke et al.<sup>[2]</sup> in 1985. The Tsui scale was developed by Tsui et al.<sup>[3]</sup> in 1985 as a relatively shorter evaluation scale. The scale evaluates the posture, the amplitude, and duration of interval head movements and the presence of shoulder elevation and head tremor.

Poewe et al.<sup>[4]</sup> modified the Tsui scale to improve its sensitivity to the postural deviance of the head. The interrater correlation demonstrated that the scale yielded replicable results.<sup>[5]</sup> However, some studies identified significant discrepancies between the scores and the evaluations of the therapeutic responses of the patients.<sup>[4,6]</sup>

In 1990, Consky et al.<sup>[7]</sup> developed the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS), a composite scale incorporating

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different characteristics of cervical dystonia. The scale evaluates the severity of cervical dystonia and includes two subscales for disability and pain. It is the most used scale for cervical dystonia. However, the routine clinical use of the scale is complicated despite its clinical value. The weaknesses of the TWSTRS include the unclear definition of the midline for evaluating the range of motion, the lack of a separate scoring category for the evaluation of dystonic tremor, and the specification of duration for the effects of sensory tricks. The Movement Disorders Society recommended the use of the Tsui scale and TWSTRS for the evaluation of cervical dystonia.<sup>[8]</sup>

The postures and movements of individuals or objects can be analyzed using optical depth data. The cameras that are used for this purpose include time of flight sensor systems that send one or two rays of artificial light (laser or LED [light-emitting diode]) to the surface of an object and record the time elapsed during the return of the same ray of light. The sensors thus calculate the difference between the distances and detect the direction and speed of the movement in the case of a moving object (Figure 1).

Kinect cameras discern human body parts using depth data and define the skeletal structure by dividing the human body into 20 joint locations. They contain depth sensors and RGB (red-green-blue) holographic light sensors. The cameras can identify faces and bodies using the depth data obtained using infrared light. Through their structural light sensors, Kinect cameras can generate images with higher resolution and are cheaper than technologies providing similar measurement results. Kinect cameras can record the depth, position, and movement of objects. The camera can take measurements at 0.4 to 4.5 m. The Kinect version 2.0 divides the human body into 25 joint locations, and the new version allows muscle simulation and the tracking of the face and hands.<sup>[9]</sup>

The study aimed to develop a measurement method that yielded objective data for the clinical assessment of cervical dystonia using a Kinect camera system. The hypotheses of the study were that *(i)* the dystonia-induced postural changes could be mathematically calculated in the computer environment using optic systems and appropriate computer software, and *(ii)* the method would yield objective data for the evaluation of cervical dystonia.

#### PATIENTS AND METHODS

This double-blind, parallel-group method development study was conducted with healthy individuals and patients with primary cervical dystonia who were admitted to the Movement Disorders Clinic of the Neurology Department, Ege University Medical Faculty, between December 2018 and January 2020. The study's inclusion criterion was being aged between 20 and 60 years. For the cervical dystonia group, additional criteria



Figure 1. Kinect camera sensors.

included a diagnosis of primary cervical dystonia, the absence of any other condition that might cause abnormalities in cervical posture or shape, and being actively followed at the Ege University Faculty of Medicine, Department of Neurology. For the healthy group, eligibility was contingent upon the absence of any diseases or conditions that could lead to abnormalities in the head or neck's shape or posture. The exclusion criteria included the presence of systemic disease, the acquired or congenital orthopedic problems of the neck, the presence of diplopia, and the presence of acute or chronic vestibular pathology. The final sample of the study consisted of 22 patients (3 males, 19 females; mean age: 47 years; range, 34 to 60 years) with cervical dystonia and 20 healthy individuals (13 females, 7 males; mean age: 32 years; age range, 22 to 65). The study was approved by the Ege University Faculty of Medicine Clinical Research Ethics Committee (date: 20.02.2018, no: 18-8/28). Written informed consent was obtained from all participants. The



**Figure 2.** The head-neck posture simulation of Kinect camera software. Using a Kinect version 2.0 camera and custom software, participants' head and neck postures were transferred to a virtual environment through angle measurements. Three 10-sec recordings at the same position ensured replicability, with data analyzed using the software.

study was conducted in accordance with the principles of the Declaration of Helsinki.

Initial measurements were performed using a dummy with a 360° rotatable head, a manual protractor (Figure 4), and the Kinect camera system (Xbox 360<sup>™</sup>; Microsoft Corp., Washington, USA) for the optimization of the measurement method and the specially developed software. The position of the dummy's head was adjusted to 10°, 20°, and 30° in the right-left direction and 10° and 20° in the anterior-posterior direction. Measurements were performed using the Kinect camera system, which revealed that the anterior and posterior deviations could not be measured using the system. The right-left 10°, 20°, and 30° error margins in torsion and lateral position was calculated to be ±2°. The mean margin of error for the clinical use of the Kinect system was calculated to be  $\pm 5^{\circ}$  using the data collected from the 20 healthy participants. The limitations of the system and program were identified, and the system and program were revised accordingly by computer engineers. After the revisions, measurements were repeated with 20 different healthy participants, and the mean margin of error of the Kinect system was calculated to be ±3°. Afterward, measurements were performed with the 20 patients with cervical dystonia, with simultaneous video recordings captured from four directions.



**Figure 3.** The second measurement of the healthy participants.



**Figure 4. (a)** A dummy with a 360° rotatable head. **(b)** Manuel protractor. A manual protractor used in neurology is a simple, handheld tool used to quantitatively measure joint angles, range of motion, or alignment.

The video recordings of the cervical dystonia patients were evaluated by two experts (Expert 1 and Expert 2) who were blind to each other's evaluations. Moreover, the experts were asked to indicate exact numerical values for their estimations of the rotation angle. The data collected from the device and the experts were compared. The protocol for the m'easurements with the Kinect camera was as follows: (i) a 1-m distance was set between the camera and the participant; (ii) the height of the camera was adjusted to the glabellum of the participant; (iii) the participant was recorded while sitting and facing the camera; (iv) the healthy participants were recorded while at rest, and the head-neck postures were simultaneously assessed using a manual protractor; (v) the cervical dystonia patients were recorded both at rest and while moving (opening and closing their hands); (vi) the 20-sec video recordings of the left-right and anterior-posterior profiles of the cervical dystonia patients were made so that the shots displayed the shoulders of the patients as well. The first 10 sec of the videos were recorded while the patients were at rest, while the last 10 sec were recorded with the patients moving (opening and closing their hands). The video recordings were evaluated by Experts 1 and 2.

The TWSTRS can be applied in approximately 20 to 40 min. The scale includes three subscales comprising physical findings (severity subscale), disability subscale, and pain subscale. The scale includes a videotape protocol that allows the standard evaluation of all patients. The rotation and laterocollis subscales of the TWSTRS were employed in the study. The items of the TWSTRS-Severity scale are as follows: maximal

excursion (rotation, tilt, anterocollis-retrocollis, lateral shift, sagittal shift); duration factor; effect of sensory tricks; shoulder elevation/anterior displacement; range of motion (without sensory tricks); time (the duration up to 60 sec for which the patients can maintain their heads in the neutral position [±10°] without the use of sensory tricks). The maximum total score is 35 (Table 1, and 2). Previous studies demonstrated TWSTRS to have acceptable internal consistency and interrater agreement. The highest interrater agreement was obtained for rotation, anterocollis, and retrocollis, while the results on lateral shift yielded the lowest interrater agreement.<sup>[10]</sup> The TWSTRS scale was strongly correlated with the Tsui scale scores.<sup>[8]</sup>

The Tsui scale can be applied in 5 min. The scale is employed in routine clinical controls and scientific studies. The results are rated from 0 to 25, with increasing scores indicating greater clinical severity. It is a short and practical scale for the evaluation of the amplitude and duration of the dystonic movement in spasmodic torticollis. The interrater reliability of the scale was previously established as 86%.[3] The subscale A of the Tsui scale was employed in this study. The subscale was scored by the experts in three components (where A represents the amplitude of the dystonic movement): (i) A-1 (rotation score), (ii) A-2 (laterocollis scoring), and (iii) A-3 (anterocollis/retrocollis scoring). In all three components, the following grading was used: 0= absent, 1= mild (1° to 15°), 2= moderate (15° to 30°), and 3= severe (>30°) (Tables 1, 2).

## Statistical analysis

Data were analyzed using IBM SPSS version 21.0 (IBM Corp., Armonk, NY, USA) and Stata

TABLE 1       Sample measurements by Expert 1										
	TWSTRS						mTsui			
	0	1	2	3	4	0	1	2	3	
	0	01-22	23-45	46- 67	68-90	0	<15	15-30	>30	
01R		Х					Х			
01M		Х					Х			
02R		Х					Х			
02M							Х			
03R	Х	Х				Х				
03M		Х					Х			
05R		Х								
05M										
06R	Х					Х				
06M	Х					Х				
07R	Х					Х				
07M	Х					Х				
08R	Х					Х				
08M		Х					Х			
09R		Х					Х			
09M		Х						Х		

TWSTRS: Toronto Western Spasmodic Torticollis Rating Scale; mTsui: Modifiye Tsui Scale; R: At rest; M: Diffusion, with facilitation; 0: None; X: Present; YY: Yes Yes (sagittal).

TABLE 2       Sample measurements by Expert 2										
		TWSTRS					mTsui			
	0	1	2	3	4	0	1	2	3	
	0	01-22	23-45	46-67	68-90	0	<15	15-30	>30	
01R		Х					Х			
01M		Х					Х			
02R			Х					Х		
02M			Х					Х		
03R	Х					Х				
03M		Х					Х			
05R		Х					Х			
05M		Х					Х			
06R		Х					Х			
06M		Х					Х			
07R		Х					Х			
07M		Х					Х			
08R		Х					Х			
08M		Х					Х			
09R			Х					Х		
09M			х					х		

TWSTRS: Toronto Western Spasmodic Torticollis Rating Scale; mTsui: Modifiye Tsui Scale; R: At rest; M: Diffusion, with facilitation; 0: None; X: Present; YY: Yes Yes (sagittal).

version 13.0 software (StataCorp LLC, College Station, TX, USA). The descriptive statistics of the data were given in mean angles, minimum and maximum values, and frequencies. The conformity of the sample to normality was checked using the Kolmogorov-Smirnov test. The t-test and paired samples correlation tests were used to compare the mean values of the groups. The data of the device, Expert 1, and Expert 2 were compared using the intraclass correlation method. The interrater reliability for ordinal data was calculated using the percent agreement, Cohen's or Conger's kappa, and Gwet's AC2 (weighted) coefficients, first between pairs of raters and then among all three raters. All coefficients were presented with a 95% confidence interval. Due to the issues in the use of the kappa coefficient, the Gwet's AC2 was preferred for yielding more consistent and reliable results.<sup>[1]</sup> However, considering the demand of the guideline for reporting multiple coefficients of agreement, two other coefficients were also presented. The coefficients were interpreted using the Gwet's probabilistic method while adhering to the Landis and Koch's scale. The results were interpreted by referring to the benchmark scale.

## RESULTS

The rotation and lateral position of the necks of 20 healthy individuals were measured at the angles of 10°, 20°, and 30° to calculate the mean margin of error for the clinical use of the Kinect system, which yielded a margin of error of  $\pm 5^{\circ}$ . Thus, the limitations of the system and program were identified, and the system and program were revised accordingly by computer engineers.

The first measurement yielded a margin of error of  $\pm 5^{\circ}$  at 10° to 20° in the right-left direction during rotation and in the laterocollis position, while the margin of error increased at the angles equal to or above 30°. Additionally, the participants struggled to hold their heads straight for more than 10 sec. Furthermore, the brightness of the environment affected the measurements. Fluorescent light harmed the measurement quality, and the best measurement quality was obtained with daylight. The computer software was updated correspondingly, and the filming method was optimized. Thus, the measurements were done under daylight, and the filming duration was adjusted to 10 sec.

After the revision of the system, the second measurement was carried out with 20 healthy

individuals. The rotation and lateral position of the neck at 10° to 20° in the right-left direction were evaluated. Consecutive 10-sec recordings were done in three repetitions in each position and at each angle. Thus, 480 recordings were obtained and examined. The second measurement revealed a maximum margin of error of  $\pm 3^{\circ}$  for the rotation and lateral position of the neck at 10° to 20° in the right-left direction. This deviation was deemed acceptable.

Afterward, using the Kinect system, measurements were performed on 35 healthy individuals with cervical dystonia. The measurements of seven patients were excluded from the final evaluation due to technical difficulties, leading to a final number of 22 for the evaluation of rotation (Table 3).

Three consecutive 10-sec recordings were done while the patients were at rest or during movement (opening and closing their hands). A reduction in the dystonic posture was observed in two patients. The coevaluation of the cases revealed mean rotation angles of  $13.7\pm6.4$  and  $15.2\pm5.8$  while at rest and during movement, respectively. The difference between the mean values was determined to be not statistically significant (p=0.158).

The three-way comparison of the rotation angles measured by the device, Expert 1, and Expert 2 revealed interclass correlation coefficients of 0.799 (79.9%) and 0.784 (78.4%) while at rest and during movement, respectively.

The agreements between the device and Expert 1 were 0.896 (89.6%) and 0.856 (85.6%) while at rest and during movement, respectively. The agreements between the device and Expert 2 were 0.478 (47.8%) and 0.499 (49.9%) while at rest and during movement, respectively. The two-way comparison of the experts revealed correlation coefficients of 0.71 (71%) and 0.69 (69.2) while at rest and during movement, respectively (Table 4).

The three-way comparison of the device and the TWSTRS data of Experts 1 and 2 while the patients were at rest revealed agreement coefficients of 0.6 and 0.8, which was a good level of agreement according to the Landis-Koch benchmark. The two-way comparisons of the data revealed that the agreements between the device and Expert 1, the device and Expert 2, and Expert 1 and Expert 2 were 0.6 and 0.8 (good), 0.6 and 0.8 (good), and 0.4 and 0.8 (moderategood), respectively. The three-way comparison of

			TABLE 3			
	The demograph	ic characteris	stics of the 22	2 patients wit	h cervical dy	stonia
Case	Age	Sex	Rot-R	Rot-M	Lat-R	Lat-M
1	35	Female	Left	Left	None	None
2	57	Male	Left	Left	None	None
3	41	Female	None	Right	Left	Left
5	60	Female	Left	Left	None	None
8	51	Female	None	Right	None	None
9	52	Female	Left	Left	None	None
11	46	Female	Right	Right	None	None
12	55	Female	Right	Right	None	None
13	47	Female	Right	Right	None	Right
14	44	Female	Right	Right	None	None
16	43	Female	Left	Left	None	None
17	45	Female	None	Left	Right	Right
24	40	Female	Left	Left	None	None
25	56	Female	Right	Right	None	None
26	34	Female	Right	Right	None	None
28	48	Female	Right	Right	None	None
29	53	Female	Right	Right	None	None
30	36	Female	Left	Left	None	None
32	37	Male	Left	Left	None	None
33	53	Female	Left	Left	None	None
34	43	Female	Right	Right	None	None
35	58	Male	Right	Right	Right	Right

R: At rest; M: Diffusion, with facilitation.

the device and the Tsui scores of Experts 1 and 2 while at rest revealed a good level of interrater reliability, with values of 0.6 and 0.8.

The three-way comparison of the device and the TWSTRS data of Experts 1 and 2 while the patients were moving revealed agreement coefficients of 0.6 and 0.8 (good). The two-way

TABLE 4The comparison of the rotation coefficients measuredwhile at rest and during movement						
	Mean±SD					
Device-Rotation angle-resting	13.782±6.4315					
Expert 1-Rotation angle-resting	13.727±8.5422					
Expert 2-Rotation angle-resting	17.591±6.9327					
Device-Rotation angle-movement	15.282±5.8783					
Expert 1-Rotation angle-movement	15.409±7.9861					
Expert 2-Rotation angle-movement	18.182±6.4190					

SD: Standard deviation

comparisons of the data revealed that the agreements between the device and Expert 1, the device and Expert 2, and Expert 1 and Expert 2 were 0.6 and 0.8 (good), 0.2 and 0.4 (sufficient), and 0.6 and 0.8 (good), respectively. The three-way comparison of the device and the Tsui scores of Experts 1 and 2 while moving revealed a good level of interrater reliability with values of 0.4 and 0.6. The two-way comparisons revealed that the agreements between the device and Expert 1, the device and Expert 2, and Expert 1 and Expert 2 were 0.2 and 0.6 (sufficient-moderate), 0.4 (moderate), and 0.4 and 0.6 (moderate), respectively.

#### DISCUSSION

Numerous scales have been developed for the assessment of the severity of cervical dystonia. In an evidence-based critique, the TWSTRS and Cervical Dystonia Impact Scale-58 were proposed as the appropriate scales for the evaluation of cervical dystonia.<sup>[7]</sup> Both scales successfully passed the first clinimetric test, but none could provide the all-around evaluation of the motor and nonmotor clinical findings specific to cervical dystonia. These scales evaluate the severity of cervical dystonia and impairment of daily life. Electrophysiological studies are used as an objective diagnostic method in the evaluation of dystonia, but it is difficult for patients to tolerate because it is an invasive method.<sup>[11]</sup>

Reliable and valid assessment scales are needed in studies of the outcomes of the treatment of patients with cervical dystonia. The patients are visually scored by experts, leading to subjective results and reduced reliability and replicability. Moreover, the scales should be applied by trained and experienced clinicians in movement disorders. The results of this study revealed a better agreement between the device and Expert 1, indicating differences between raters and demonstrating the subjectivity of rater-dependent data. The use of both the TWSTRS and Tsui scales requires educated and experienced raters in movement disorders. In this study, the agreement between the device and the more experienced expert was evidently better. The use of the Kinect system does not require specific training, and the system yields reliable and replicable results through the measurements of the patients in the sitting position.

The agreement between the device and experts and between the two experts were lower when the patients were moving compared to the agreement rates at rest. The rotation angle while moving can be altered by the overflow phenomenon, which is the activation of dystonia during the voluntary movement of a nonprimary area. Furthermore, the assessment of dystonia with movement is more complicated than that of dystonia at rest.

A review of the literature revealed a variety of systems that analyze movement in cervical dystonia cases. In their study, Boccagni et al.<sup>[12]</sup> employed motion sensors that utilize electromagnetic fields to kinematically evaluate cervical dystonia. During the measurements, four sensors were placed on the coronal, axial, and sagittal planes of 15 cervical dystonia patients and 10 healthy participants. Measurements were done while at rest and with voluntary movement, head range motion was calculated, and head and neck movements were examined in detail. The head postures at rest were determined to be more impaired in cervical dystonia patients compared to those of the normal group in all planes. The analysis of voluntary motion revealed a reduced range of voluntary excursion, while the time elapsed until the motion was completed increased in cervical dystonia cases, and the impairment was clearer when the patients moved their heads against the dystonic side.

In their study, Barr et al.[13] examined the impairment of balance, gait, and stepping reaction in cervical dystonia with 10 cervical dystonia cases and 10 healthy controls. Walking speed, step length, step time, and cervical movement range were measured using a computed walkway, head-mounted goniometer, and special apparatuses. Compared to those in the control group, walking speed, balance, and stepping reaction were determined to be significantly lower in patients with cervical dystonia. However, stepping time was longer in patients with cervical dystonia. In Barr et al.'s study, various materials were mounted on the participants, and the practitioner had to receive comprehensive training before the measurement. In our study, these prerequisites were eliminated, and the need for physical contact with the participants was rendered unnecessary. The newly developed optic system in this study distinguishes itself from other metric systems in the literature by eliminating the need for the mounting of sensors and physical contact with patients.

In the present study, an easily applicable measurement method that yields objective data at rest and during movement was developed. The method may be used in patients with cervical dystonia with torticollis (10° to 20°) and showed good agreement with the TWSTRS and medium-good agreement with the TWSTRS and medium-good agreement with the Tsui scale. The system was shown to be a sensitive method for the kinematic evaluation of patients with cervical dystonia. The findings indicated the Kinect system as a beneficial tool in the diagnosis and treatment of patients with cervical dystonia. However, it should be improved to obtain more detailed data.

This study had some limitations. In the study, 10° to 20° torticollis and laterocollis were measured, while anterocollis and retrocollis could not be evaluated. Moreover, the system could only be optimized at 10° to 20°. Thus, the cases with rotation angles >25° and cases with complex dystonic patterns were excluded from the study. The improvement of the software system in this regard necessitates the use of additional materials,

such as a second Kinect camera. This study was a preliminary examination of a new measurement system. Therefore, the reliability and validity of the data yielded by the device can be improved and optimized with the help of computer engineers. The study is a pilot study of method development and clinical practicability. In the future, carrying out greater numbers of multicenter studies could prove beneficial. The system should be optimized to eliminate the limitations of this study. The limited number of cases is another drawback of the study. Multicenter studies with a greater number of participants are needed to support our findings.

In conclusion, the medical applications of advanced motion analysis systems have become topics of discussion. However, the number of studies on the issue is still limited. The evaluation of the scales that are used in the assessment of dystonia is dependent on the visual interpretations of clinicians, which yields subjective results. The results revealed that the system yielded objective and replicable data. The new method was deemed suitable for individual use in the assessment and follow-up of patients. Moreover, the method can be used for the evaluation and development of the motor disability subscales for the treatment and follow-up of patients. However, the system should be improved by adopting a multidisciplinary approach that includes computer and software engineers to obtain more detailed data.

**Data Sharing Statement:** The data that support the findings of this study are available from the corresponding author upon reasonable request.

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