

## FUNCTIONAL LOSS AND POSTURAL CHANGES RESULTED OF SEVERITY AND TIME OF EVOLUTION OF CERVICAL DYSTONIA: AN OBSERVATIONAL STUDY

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**Abstract:** Question. Can the level of severity of cervical dystonia (CD) and the time course of this disease influence daily activities performance and postural pattern? What is the postural pattern of patients with cervical dystonia? Design: Cross-sectional, descriptive and retrospective study. Participants: sixty nine patients who had undergone clinical treatment of cervical dystonia. Interventions: Participants were assessed once, while they came for a medical appointment. Outcomes measures: Clinical data were collected from medical records, level of severity and repercussions of dystonia were assessed by Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS), daily activities were evaluated by Functional Independence Measure (FIM) and postural measures were taken by postural Analysis Software (SAPO®). Results: The longer the disease progresses, the greater the inability to feed, dress, climb stairs, move downstairs and walk ( $p < 0,0001$ ). The higher level of severity and impact of CD, the larger angles between acromions, ears and acromions, among acromions and iliac spines and between ears and iliac spines and less independence was observed ( $p < 0,0001$ ). However, the severity was lower in younger subjects, although the pain and repetitive motion impacts the performance of activities of daily living. Conclusion: The time course and severity of the disease can influence on daily activity and on postural characteristics of these patients. Their postural pattern is characterized by the great discrepancy between the head and the trunk, which could be accentuated by the longer time course of dystonia.

**Keywords:** dystonia, torticollis, posture, physiotherapy, activities of daily living, disability evaluation

## INTRODUCTION

Actually, dystonia is defined as a movement disorder characterized by involuntary sustained or intermittent muscle contraction, which is exacerbated by voluntary movement. Tremor and twisting movement patterns are usually associated with dystonia, which can lead to abnormal voluntary movements and posture. (ALBANESE, et al., 2013)

Cervical dystonia is the most common type of focal dystonia. It involves muscles of the neck, producing sustained contractions that trigger movements in a specific direction (ALBANESE, et al, 2006; PHUKAN, et al., 2011). Besides, typical head movements, head or body tremor and sensorial tricks were often found as clinical signs (VELICKOVIC; BENABOU; BRIN, 2001).

Additionally, due to its characteristics, cervical dystonia has high potential to cause pain. It can be associated with elevation of one shoulder, observed in 54% of cases and scoliosis in 16% of patients with cervical dystonia (DEFAZIO, et al., 2003). Furthermore, due to the large amount of sensory receptors in the head and the neck region, the stability of this region is important for the maintenance of the posture and for daily living activities performance. Then, all that could influence different aspects of life (CAMFIELD, et al., 2002).

However, there are few records of the effects of cervical dystonia on the head and the neck posture and functionality, as well as the correlation between the time of the disease progression, pain, postural disorders and the elements that make up the functionality. Thus the lack of information about those, was a source of motivation for the development of this research.

The research questions were:

1. Can the level of severity of cervical dystonia (CD) and time course of this disease influence daily activities

performance and postural pattern?

2. What is the postural pattern of patients with cervical dystonia?

## **METHOD**

### **DESIGN**

This study was characterized by observational, descriptive and retrospective (HOCHMAN et al., 2005). It investigated the relationship between severity of cervical dystonia, the time course and its consequences. Therefore, level of severity, functionality and posture was evaluated in 69 patients diagnosed with cervical dystonia once, while they came to movement disorder service.

### **PARTICIPANTS, THERAPISTS, CENTRES**

Participants were recruited from movement disorders ambulatory. Patients with clinical diagnosis of cervical dystonia, who were not under botulinum toxin effects in the cervical region and agree voluntarily to participate were included in this study. Among the exclusion criteria were the presence of other concomitant neurological diseases and inability to remain in the orthostatic position for postural evaluation. Out of the 74 patients who participated in the selection, five of them were excluded because they presented other associated neurological diseases (n = 2) and because they could not remain in the orthostatic position for postural evaluation (n = 3).

### **INTERVENTION**

Evaluation procedure was established by a clinical history collected through medical records and interview. Data about functionality was obtained by Functional Independence Measure (FIM) questionnaire, the level of severity and impact was assessed by Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) and the assessment of

static posture of the head and the trunk were held by physical assessment and processed by specific postural software.

## **OUTCOME MEASURES**

### **PRIMARY OUTCOME**

Clinical history was collected through the patient's record, which contained personal data, clinical presentation of the disease and the muscles that receive botulin toxin. In order to supplement the information relating to the history of the disease it was developed an evaluation form containing data such as age of onset of the symptoms, how long ago it was diagnosed, time under treatment, presence of family history of dystonia, evolution of dystonia to other parts of the body, presence of tremor and early retirement caused by dystonia.

### **SECONDARY OUTCOME**

Level of severity of dystonia with cervical involvement was assessed by Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS). This scale is considered the gold standard for checking the level of severity of cervical dystonia and its repercussions. It is recommended by the International Society of dystonia and it is widely used in clinical studies. (ALBANESE, et al., 2013)

Functional assessment is a way of quantifying the ability to perform activities of independently daily living. If the patient is able to perform them or if some kind partial or total aid is required. (RIBERTO, et al., 2004;PARAHYBA; SIMÕES, 2006)

To evaluate static posture of the head and the trunk in coronal plane, photogrammetry was used, to be reliable, reproducible, easy to use, low cost and accessible for clinical practice (IUNES, et al., 2009).

Reference points were selected from the tagging protocol points of Postural Analysis

Software (SAPO®), and were located and marked (right and left). Spherical polystyrene markers

2.5 cm in diameter were used, fixed by means of double-sided tape in those points. Because cervical dystonia provides a clear commitment on the head, the neck and the trunk, it was decided to use those points in order to better characterize these postural effects, extrapolating the subjectivity of observational analysis.

To obtain images of the above reference point, in the coronal plane it was used a digital camera Sony Cyber-shot® 10.1 megapixel, coupled with tripod located 85 centimeters from the floor and three meters away from the point of reference located on the floor, which was at 30 centimeters from the wall. It was the plumb line of 1 meter in length, which was intended to be as a reference point on the Y axis and it was instrumental in the calibration images in the software. Then, distances and angles traced the records of the head and the trunk in the frontal plane were calculated (IUNES, et al., 2005).

## DATA ANALYSIS

Data collected in all evaluation were initially evaluated by means of descriptive statistics. Qualitative variables were expressed as absolute and relative frequencies percentage, while quantitative variables were expressed by mean and standard deviation.

Data of the variables were standardized, and then analyzed using principal component analysis (PCA), after verification of data quality by Kaiser-Meyer-Olkin method (KMO test). The evaluation of the correlation matrix between variables was assessed using Bartlett's test of sphericity and setting the number of principal components were established through the Broken-stick test. With the ACP it was possible to determine the explanatory variables for each variable evaluated, these

being defined a priori according to the time of diagnosis of dystonia. For all tests it was considered  $p \leq 0,05$ .

## INTRA-RATER RELIABILITY INDEX

Intra-rater reliability index (ICC) was calculated for greater accuracy and reliability of data obtained through measurements performed during postural analysis of the head and the trunk. (PORTNEY; WATKINS, 2000). Data were processed by software Bioestat® 5.0, with a 95% confidence interval. Excellent reliability ( $> 0.90$ ) was obtained of distance between tragus measures (0.96), distance between acromions (0.99), DEI (0.99), angles between tragus (0.98), angles between acromions (0.98), angles between iliac spines (0.97) angles between tragus and acromions (0.99), angles between acromions and iliac spines (0.97) DT8-L2 (0.99), angles between C7 and L2 (0.98) and angles between T8 and L2 (0.99). Good reliability (0.75 to 0.89) was observed between tragus and iliac spine angles (0.86).

## RESULTS

### FLOW OF PARTICIPANTS, THERAPISTS, CENTRES THROUGH THE STUDY

Regarding the characteristics of the sample it was observed that the average age of survey participants was  $50 \pm 15$  years, most of them female, corresponding to 60.9% ( $n = 42$ ). 69.6% ( $n$

$= 48$ ) the disease manifested itself in adulthood and it had no identified family history of it by the time of evaluation in 85.5% ( $n = 59$ ). As for the direction of the movement performed, the dystonia torticollis laterocollis was found in 31.9% ( $n = 22$ ), followed by torticollis in 27.5% ( $n = 19$ ) of cases evaluated. Due to the development of the disease, 69.6% ( $n = 48$ ) of people did not

retire early. Therefore, as to the time of disease progression, patients showed median time of diagnosis of  $13 \pm 10$  years, and there are  $6.9 + 4.9$  years of treatment.

**Research question 1** - Can the level of severity of cervical dystonia (CD) and the time course of this disease influence daily activities and postural pattern?

The higher the level of severity and impact of the DC, the larger angles between the head and the trunk and the lower independence to move from bed to chair and to use the toilet.

Functionality, that was assessed by FIM, was observed. A greater impact of cervical dystonia in the item memory, social interaction and difficulty in solving problems was found. Similarly, it was observed that patients who had better memory scores are also more independent, have more sociability interaction, the ability to solve problems and also the ability to feed themselves, to do their personal hygiene, bathing, taking a shower, dressing, using the toilet and using stairs (Figure 1).

Correlation between time of diagnosis and functions of FIM showed that the shorter the time of diagnosis, the more independent the patient becomes for food, to perform personal hygiene, to dress from the waist up to the waist down, to use toilet, to take a shower and to use stairs.

**Research question 2** - What is the postural pattern of patients with cervical dystonia?

A discrepancy in the angles formed between the 7th cervical vertebra and the 8th thoracic vertebra was observed. The same occurred with the angles formed between the 7th cervical vertebra and the 2nd lumbar vertebra, as well as between the 8th thoracic vertebra and the 2nd lumbar vertebra, characterizing great trunk asymmetry (Figure 2).

## DISCUSSION

The most common form of dystonia is the focal one (LE, NILSEN et al., 2006), affecting more women than men at a ratio of 1.5: 1 (MORDIN , et al., 2015). These findings from the literature are similar to the results found in this study, reaffirming the higher prevalence of women with CD.

Regarding the age group of CD, a study with more than one thousand people that aimed to investigate the characteristics of patients with CD, found that the mean age of participants was  $53.2 \pm 11.9$  years. The age group being the most prevalent was found among 45-54 years (32%), 55-64 years (26%) and over 64 years (20%) (COMELLA; BHATIA, 2015; MORDIN , et al., 2015). Thus, the data obtained by this study reveal similarity to the findings mentioned by the authors above. It was observed that the time course of the disease was  $15.5 \pm 5.07$  years (QUEIROZ et al., 2012).

In relation to direction of movement, a study developed with 16 patients with CD, with a mean age of 46.19 years, found that 62.5% of them had laterocollo, 18.75% presented retrocollo and 18.75% torcicolo (USEROS-OLMOS; COLLADOVÁSQUEZ, 2010). In this study, lactalocolus was also more frequent.

According to the severity level, when subjects with CD were evaluated, it was observed that the total score of the TWSTRS was 43.23. The disability subscale was 12.37, the severity score was 19, 79 and in the pain subscale was 9.27. It was also observed that the impairment was greater in the elderly subjects than in the young ones. However, there was no relationship between the gender and the body mass (JOST et al., 2012).

In a study developed to verify the effects of hydrotherapy in 16 patients with CD, it was observed that the total, severity, disability and pain scores of the TWSTRS before the

proposed treatment were  $41.25 \pm 14.56$ ,  $17.50 \pm 5.90$ ,  $13.63 \pm 6.64$  and  $10.13 \pm 5.49$ , respectively. After treatment, the respective scores were  $32.81 \pm 12.66$ ,  $14.19 \pm 5.71$ ,  $11.44 \pm 5.93$  and  $7.81 \pm 5.13$  (USERS-OLMS, COLLADO-VÁSQUEZ, 2010).

The data presented by the aforementioned surveys are related to the values found in the present study, since the respective values were obtained in total score and in the severity, disability and pain dimensions, respectively  $28.5 \pm 11.59$ ,  $8.7 \pm 4$ ,  $10.6 \pm 6.1$  and  $9.8 \pm 5.3$ . These findings are justified by the fact that the research subjects included in this study were already assisted for at least  $6.9 \pm 4.9$  years.

Thus, it is suggested that the time of follow-up and treatment in the outpatient clinic favored the maintenance of these scores, even though at the time of the evaluation the patients did not have BTX effect. Data related to physical activity and physical therapy were inconsistent and for this reason were not included in this study.

Therefore, subjects with the highest scores of severity and impact of the DC presented the highest measurements of the angles between the head and the trunk, as well as

greater dependence to move from a bed to a chair and to use the toilet. Likewise, greater measures of angles of the trunk are related to lower independence for food, to perform personal hygiene, to bathe/take a shower, to dress from the waist up to the waist down, use the toilet, gait and ability to use stairs, when correlated to the variables present in the measure of functional independence.

These indications reinforce the argument about the existence of abnormalities that may interfere in the degree of severity and functionality of subjects affected by DC. The range of

motion of the cervical spine in the frontal plane is up to  $45^\circ$ . Considering that the maximum angle formed between the swallow and the acromion was  $48^\circ$ , this could be enough to cause some type of compensation, which could be represented by greater wear of the joint surfaces, causing pain and functional limitation (Kapandji, 2000). Thus, the results indicate that the greater the discrepancy between the joint angles, the greater the functional impairment and the severity level of the DC.

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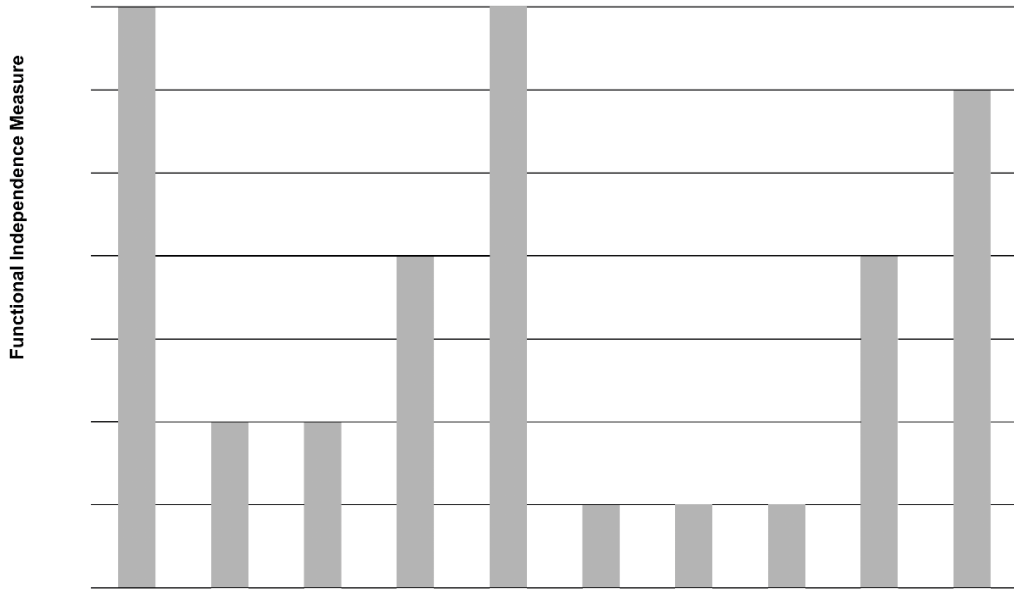
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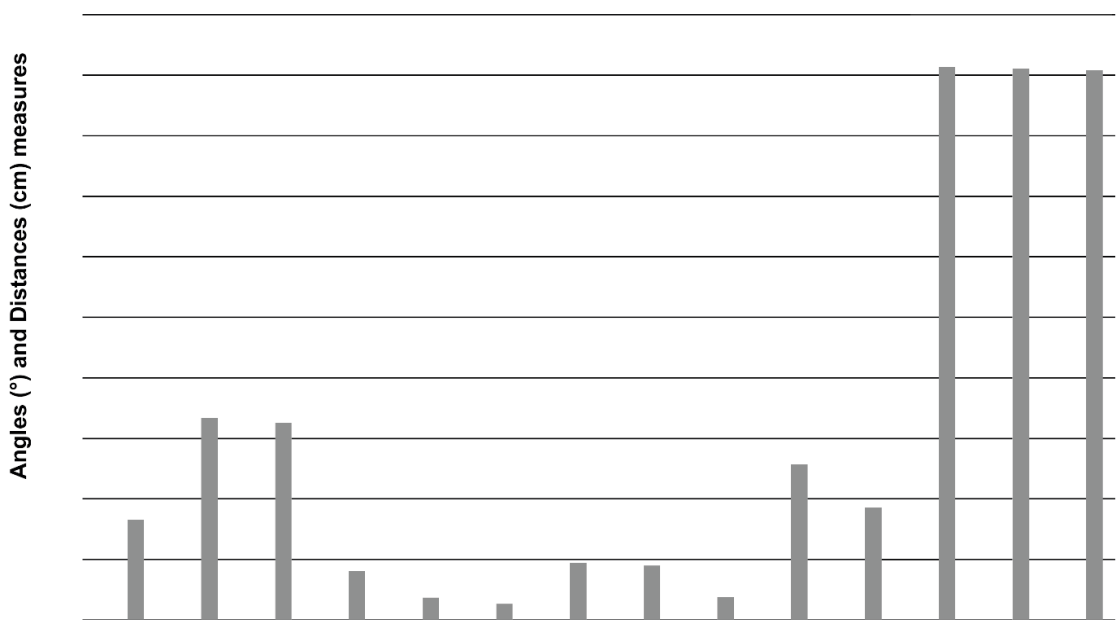
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**Figure 1.** Mean of scores of FIM, that represents the most compromised daily activities by the cervical dystonia.

Daily Activities



**Figure 2.** Postural pattern of cervical dystonia patients.

Postural Pattern (Distances and angles assessed by the head and the trunk)