

Fisioterapia Neurofuncional



ANELICE CALIXTO RUH
(Organizadora)

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APRESENTAÇÃO

A neurologia clínica é um assunto muito pesquisado devido a sua complexidade e suas diferentes manifestações em cada indivíduo. Os sinais e sintomas, reações e consequências variam, tornando-se um desafio para quem diagnostica, trata, para os familiares e para a sociedade.

A fisioterapia está conquistando cada vez mais espaço na realidade da saúde mundial. A prática da profissão baseada em evidências e estratégias científicas levam a credibilidade do tratamento. Para isto torna-se importante estudos científicos com precedentes intervencionistas comprobatórios ou não.

A formação do fisioterapeuta deve ser voltada ao aprendizado de gerir, avaliar, observar, prescrever e tratar, sendo para isso necessário a busca pelo conhecimento em fontes atuais de cada área.

Nas doenças neurológicas, neste caso, devemos estar atentos aos métodos avaliativos, pois mais do que a doença apresentada com suas características gerais, a avaliação minuciosa que deve nortear a assistência.

Nesta coleção de 16 artigos você vai encontrar diversas técnicas avaliativas e de tratamento para doenças neurológicas com alto índice de morbidades, como a Paralisia Cerebral, lesão não progressiva que pode ocorrer no período pré, peri ou pós-natal, que afeta o tônus muscular, a postura e o movimento pode estar acompanhada de diversos outros sinais e sintomas que devem ser bem avaliados para que se possa definir o nível do comprometimento motor delineando o tratamento específico para cada paciente, como equoterapia, realidade virtual, etc.

A lesão medular, incapacidade de grande impacto econômico e social, sendo de extrema importância a utilização diversos métodos avaliativos e terapias diversas, para melhor desempenho motor e qualidade de vida do paciente. Dentre outros assuntos relevantes.

A reabilitação das funções perdidas ou prejudicadas por estas doenças traz um desafio acadêmico e profissional, sendo importante obras como esta que englobam temas relacionados, atualizando a comunidade científica sobre métodos avaliativos, recursos terapêuticos e técnicas, tudo isso visando a recuperação de forma mais proveitosa para o paciente.

Boa Leitura!

Anelice Calixto Ruh

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SELF-PERCEIVED POSTURAL SHIFT IN CERVICAL DYSTONIA PATIENTS

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ABSTRACT: Background: Dystonia is a neurological syndrome characterized by involuntary muscle contractions, leading to repetitive torsion movements and postural alterations. **Aims:** The purpose of this study is to evaluate whether patients with cervical dystonia present alterations in their cervical body shift perception. **Methods:** This study included patients with cervical dystonia with no evidence of degeneration or structural lesion, isolated dystonia, being treated at the botulinic toxin clinic. The exclusion criteria were patients with combined dystonia, dementia, osteoarticular problems or other neurological problem. A control group composed of patient's relatives or companions without dystonia, who fulfill the same inclusion and exclusion criteria, was evaluated.

Every patient was evaluated regarding to his cervical body perception, severity of his dystonia, cognition, and depression. **Results:** Only 11% of patients with dystonia present correct perception of their posture whereas the same was observed in approximately 80% of the control group. **Conclusions:** Patients with cervical dystonia present bad awareness to the cervical shift perception in relation to individuals without dystonia. This fact may be related to physiopathogenic mechanisms of the dystonia. **KEYWORDS:** Dystonias, Cervical Dystonia, Bodily Perception, Botulinic Toxin.

1 | INTRODUCTION:

Cervical dystonia is a form of focal dystonia, although the shoulder can also be involved. It is considered an isolated form when the dystonia is the only motor feature, with the exception of tremor, without evidence of degeneration or structural lesion. The etiology of dystonia is still not fully comprehended. In every case, there is a coactivation of agonists and antagonists that interfere with time, accomplishment and loss of independent movements of articulation (TANABE, MARTIN, DAUER; 2014; AGUIAR, et al, 2004, AGUIAR, FERRAZ, 2000).

It has been reported that patients with dystonia have altered sensory-motor

processing. Recent observations suggest the irregular processing of proprioceptive information from the affected areas may perform a significant role in the origin of the characteristic of symptoms. It is possible that the kinematic deficiency is not limited to the dystonic segments. Due to the processing of information from muscle spindles and the misinterpretation of positional information, it may also be present in asymptomatic regions of the body (OCHUDLO, BRYNIARSKI, OPALA, 2007; SLAWEK, et al, 2007; HILKER, et al 2001).

The altered sensorial function would suffer adaptative or compensatory changes to take information to the sensory-motor system. A possible explanation for this fact is that it represents a secondary phenomenon resulting from alterations in the sensorimotor cortex induced by dystonic movements. These mechanisms could affect the way that the patients perceive their body position (HILKER, et al 2001).

Perception can be defined as the integration of sensorial impressions of information. It represents the capacity of selecting the stimuli that require attention and action to integrate them and with the previous information so they can finally be interpreted. The conscience of the bodily scheme is considered the base of the performance of the entire intentional motor behavior (DAUER et al, 1998).

This study aims to evaluate the cervical bodily perception of patients with cervical dystonia.

2 | MATERIALS AND METHODS

For this study, the inclusion criteria were patients with isolated cervical dystonia who were under treatment at the clinic of dystonias of the Hospital de Clínicas de Porto Alegre (HCPA). Only individuals over 18 years old who agreed and signed the free informed consent form participated in this study. As exclusion criteria, patients who presented combined dystonias, dementia, osteoarticular problems, other neurological diseases and who refused to carry out the evaluations were considered unfit. For the control group, the same criteria were considered, and it was composed of patients' relatives and companions without dystonias.

The Ethics Research Committee of the Hospital de Clínicas de Porto Alegre approved of this project on April 28th, 2014, under the register 140087.

Information of case group patients' age, gender, schooling and clinical history was collected. Patients were controlled as to the treatment with botulinic toxin.

Evaluation of Bodily Perception

The perception of the position/posture of the head was evaluated on a visual analog scale. It was developed based on 11 images with different degrees of head/neck inclination: 3 representing varying degrees of anterocollis, 2 of retrocollis, 3 of torticollis and 3 of laterocollis. The images were shown to the participants of the research when

asked about their cervical bodily perception. They were supposed to point to which model represented their posture, according to the scheme below and the image 1:

➤ Anterocollis

- $A = 0$ (not corresponding to posture)
- $B = 1^\circ - 15^\circ$,
- $C = 16^\circ - 30^\circ$,
- $D = 31^\circ - 65^\circ$,

➤ Retrocollis

- $A = 0$ (not corresponding to posture)
- $B = 1^\circ - 20^\circ$,
- $C = 21^\circ - 50^\circ$,

➤ Torticollis

- $A = 0$ (not corresponding to posture)
- $B = 1^\circ - 15^\circ$,
- $C = 16^\circ - 30^\circ$,
- $D = 31^\circ - 55^\circ$,

➤ Laterocollis

- $A = 0$ (not corresponding to posture)
- $B = 1^\circ - 10^\circ$,
- $C = 11^\circ - 20^\circ$,
- $D = 21^\circ - 40^\circ$.

The posture perception by the individuals was considered correct when there was congruence between the results of self-perception in the visual-analogical scale and goniometry measures, or incorrect or non-congruent when the response of the visual scale differs from real values presented on the goniometry. It was taken into consideration the treatment with botulinic toxin, and the evaluations were carried out at least two months after the last application, reducing the effect of the toxin influence the perception of the patients.

Evaluation of the Mental State and Depressive Symptoms

The participants were evaluated to determine the degree of depression by the Montgomery-Asberg Depression Rating Scale (MADRS). In order to determine their mental state, it was used the Montreal Cognitive Assessment (MoCA) was applied in the case and control groups in this study.

The MADRS values varied as follows: 0 – 6 normal or light symptoms, from 7 – 19 light depression, from 20 – 34 moderate depression and above 34 severe depression

(DRACTU,RIBEIRO, CALIL, 1987).

The MoCA scale considers a score equal or superior to 26 as normal and has the following items: executive visuospatial function, appointment, attention, language, abstraction, deferred evocation and orientation (NASREDDINE et al, 2005).

Evaluation of the Degree of Cervical Dystonia Involvement

- Toronto Western Spasmodic Torticollis Rating Scale

The questionnaire Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) was applied to the case group, aiming to verify the severity of the patients' dystonia. It is composed of three sub-scales: motor severity (interval of 0 to 35), deficiency/activities of daily life (interval of 0 to 30), and pain (interval of 0 to 20) (SALLEM, CARAMELLI, BARBOSA, 2011)..

- Goniometric Measurement

In order to measure the degrees of different postures the patients had, an analysis was carried out through goniometry, which made it possible to obtain the alteration degrees of anterocollis, laterocollis, retrocollis and torticollis the patients really have for further comparison with a visual analog scale.

3 | STATISTICAL ANALYSIS

The data analysis was made by using the software SPSS version 21.0 and the Kappa Pabak calculator available on the website <http://www.singlecaseresearch.org/calculators/pabak-os>.

The quantitative variables were described by average or standard deviation or median and interquartile range. The categorical variables were described by absolute and relative frequencies.

The Student's *t*-test was applied to compare the average between the groups. In the case of asymmetry, the Mann-Whitney test was used. In the comparison of proportions, Pearson's chi-squared test or Fisher's exact test was applied. In the case of significative association, the adjusted residual test was used.

The association between the continuous variables was evaluated by the correlation tests of Pearson (symmetric distribution) or Spearman (asymmetric distribution). The concordance between the scales was evaluated by the Kappa Pabak coefficient, as it regulates the data by the prevalence of closure, along with the interval of 95% of assurance. The level of significance adopted was 5% ($p \leq 0.05$).

4 | RESULTS

Twenty-seven (27) patients with cervical dystonia were evaluated, 18 women (66.7%) and 9 men (33.33%), as well as 27 patients of the control group with the

same distribution. The age average of the case group was of 51.44 years old, from 19 to 79, and the control group had an average of 41.85 years old, from 24 to 67, there was statistically significant association by adjusted residue test as age groups. Table 1 describes the characterization of the sample.

➤ *Analysis of the cervical body shift perception*

In the analysis of bodily perception, only 11% of the patients with cervical dystonia presented a correct perception of the cervical positioning in relation to the goniometric analysis, which was significantly different from the control group whose correct perception was observed in 78% of the people ($p < 0.001$), as shown in Figure 2.

➤ *Factors Associated with cervical bodily perception*

• *Subtype of Cervical Dystonia*

In the studied sample, as to the distribution of the subtype of dystonia, 12 patients with torticollis (44%), 9 patients with laterocollis (33%) and 3 (11%) with retrocollis and anterocollis, respectively, were found in the studied sample. The bodily perception of the people was not related to the subtype of the disease.

• *Severity*

The severity of the evaluated dystonia by the TWTRS scale obtained an overall score with an average of 33.4 DP \pm 13.6, in which the subscale motor severity had an average of 15.5 DP \pm 5.9, deficiency subscale in daily life activity 8.37 DP \pm 5.72 and in the pain scale a score of 4.74 DP \pm 6.30. The bodily perception of the individuals was not related to the severity of the disease.

• *Pain*

When questioned about feeling pain, 21 patients (78%) of the case group and 16 (59%) of the control group reported feeling pain. The intensity of pain in the case group was 6.43 (DP \pm 2.01) and in the control group 5.47 (DP \pm 2.03). The bodily perception of the people was not related to pain.

• *Time of disease*

The age average of the beginning of the illness in the dystonic group was 36.74 years old (DP \pm 17.60), varying from 9 to 72 years old. The average time of the disease was 14.7 years (DP \pm 12.46). The bodily perception of the people was not related to the time of the disease and the beginning age.

• *Botulinic Toxin*

In this sample, 22 (81%) of the patients used to have applications of botulinic toxin. The average the patients had the applications was 26 times (DP \pm 19.33). Twenty (20) patients (91%) reported feeling better after the application. The average of days patients feel the effect of the toxin is 73 days (DP \pm 37.46).^{<0}} The bodily perception of the individuals was not related to the treatment with botulinic toxin.

- Depressive symptoms and Mental State

There was no significant difference between the groups as to the scores of depression ($p=0.151$), despite the case group presents a higher proportion of moderate depression and the control group a higher light degree. In the case group, 96.3% presented cognitive decrease whereas only 29.6% of the control group showed this condition, with significant difference between the groups ($p<0.001$). The bodily perception of the individuals was not related to the depressive symptoms and mental state.

4.1 Concordance between the self-perception and goniometry

Table 3 brings the comparisons carried out between the visual scale (VS) and goniometry, in cases groups.

Patients of the case group do not have a proper perception of the correct positioning of their heads; there was no significant concordance between the visual scale and the goniometry, which corresponds to the real measure of their postures. The control group showed having a real perception, presenting concordance in every gauged position, except in retrocollis positions where two patients were with this posture and did not point it in the VS.

The association between the clinical variables of the patients in the case group with cervical bodily perception was evaluated. However, these data did not show significant association, but demonstrated that patients who are congruent present more time of dystonia and more severity of the disease, do not apply toxin and have a lower level of depression. There was not a significant association between the variables referring to depression and cognition when compared to the perception of the control group.

5 | DISCUSSION

This study demonstrated that patients with cervical dystonia present alteration in the cervical bodily perception in relation to non-dystonic individuals. In this research, the congruence of perception (correct perception) was about 11% in the case group and 78% in the control group. As far we are aware this has been the first study that carried out such approach in patients with cervical dystonia.

The alteration degree in bodily perception in patients with dystonia was not related to the severity, time of disease, treatment with botulinic toxin, depressive symptoms, and cognitive state.

Other essays have demonstrated alterations in the bodily perception of dystonic patients by using different instruments of evaluation. Studies suggest the altered bodily perception in cervical dystonia is connected to the vestibular system, as the patients with cervical dystonia would notice their spatial environment asymmetrically, the torticollis

associates to proprioceptive asymmetries and vestibular inputs, which leads to the mistaken spatial perception and behavior, even if the head deviation changes during the day (ANASTASOPOULOS, et al, 1998; BRONSTEIN, RUDGE, 1986; MÜLLER, GLÄSER, TRÖGER, DENGLER, JOHANNES, MÜNTE, 2005).

In a group of 35 patients with cervical dystonia evaluated as to their vestibular abnormalities either being or not being responsible for their abnormal posture of the head or the result of abnormal interaction of interaction between the motor sensory system, the result was that the primary involvement of the vestibular system in cervical dystonia relates to the transport of sensorial information responsible for the orientation of head and eyes (MÜLLER, GLÄSER, TRÖGER, DENGLER, JOHANNES, MÜNTE, 2005).

In animals vestibular lesions cause posture and positioning of the head problems, and there are reports of vestibular disorders in patients with cervical dystonia, it is not clear, but the vestibular system would be the cause of abnormal head posture or would be acting with another system to cause the change. It is believed that there is an imbalance of tonic muscular activity in both the neck and in the ocular system, diverting the head and eyes in the same direction. The vestibular system is important connections within the brain stem, and a disruption of these connections can lead to imbalance of the muscles that control the positioning of the head (BRONSTEIN, RUDGE, 1986).

In another study evaluating the allocentric and egocentric position of people with cervical dystonia, questioning whether the abnormal posture of the head was associated with spatial reference, it was suggested that patients are not capable of adapting themselves to abnormal proprioceptive entrance over time due to the fluctuation of symptoms being connected or not to the treatment with botulinic toxin, and the vestibular system, visual and proprioceptive are connected to a body position when it must be a reference to other lines or ways (BOVE, et al, 2006).

Analyzing the orientation of body and posture during the locomotion of people with or without cervical dystonia, to verify whether the proprioceptive entrance of the neck is misinterpreted or not by the carrier of dystonia, the authors conclude the system of reference used by dystonic people to control and guide their bodies in space is refractory to a side proprioceptive entrance through the neck, and this is probably connected to the pathogenesis of the disease as an adaptation of the proprioceptive entrance of the muscles involved in the orientation of the head in space (BLACKIE, LEES, 1990).

As to the application of the botulinic toxin, in this study there was not a significant association related to the patients' perception, being of no effect in our population the number of times the applications were made, how many days the patient was without receiving the toxin or even whether they note any difference with the treatment, similar to another study whose results showed there is no significant difference between patients that had never received an application of the toxin or the ones who received

as to how to position their head (BLACKIE, LEES, 1990).

The severity of the dystonia was evaluated through the TWSTRS questionnaire and did not show having significant relation with perception. However, it was possible to identify a tendency: patients who have more time of dystonia and a more severe score in the Toronto test are the ones who have congruence between their perception and the real posture.

In this current study, the depression in the case group had two significant associations: the first one, related to the severity of the disease, and the second one, inversely significant to the time of the disease. These data corroborate the literature, demonstrating that social and emotional factors interfere in posture and have a sensibility under the torticollis. Different levels of depression are found in patients with spasmodic torticollis in relation to the embarrassment caused by postural disfigurement (KUYPER, et al, 2011).

The depression may be related to the patient's global incapability, and in the population with cervical dystonia studied in 2007, the distribution of depressive symptoms seems to be similar to the current study, as the major part of the studied population had indices of light to moderate depression and the minority had severe depression. The results of the referred study corroborate the relation between severity and depression index, demonstrating the higher the depressive symptom, the more severe is the dystonia. However, it was not found any significant association between the depression and the bodily perception (SLAWEK, et al, 2007).

The cognitive deficit etiology is difficult to explain in patients with dystonia. However, it is noted a high variability of age and schooling, which may influence the results of the cognitive tests¹⁸. Such data were also discrepant in our study. The mental state of patients were evaluated in the case and control groups in this study, and although only 3.7% of individuals of the case group and 70.4% of the control group do not have cognitive decline (MoCA \leq 26); these data do not present significant association with bodily perception.

The possible limitations of the study may be the following: the number of participants; only patients with cervical dystonia were evaluated; people from the control group were not evaluated in different head/neck positions; this group is composed of patients' relatives and companions, which may have influenced the scores of depression. Patients of the case group were from only one hospital in the city and receive the same type of treatment, leaving out a chance for other carriers of dystonia, who are treated in other clinics and hospitals, take part of this study.

Based on the observed aspects, it is possible to conclude that to the studied population, the patients' perception with primary cervical dystonia is different from the population without dystonia. As a second important result, this study presented a significant relation of depression with the severity of the disease. We did not obtain answers to the relation of self-perception with depression, cognitive level or use of botulinic toxin. Even so, we still believe the study is valid as it demonstrated through a

simple method of evaluation of the cervical bodily perception the difference between the analyzed groups.

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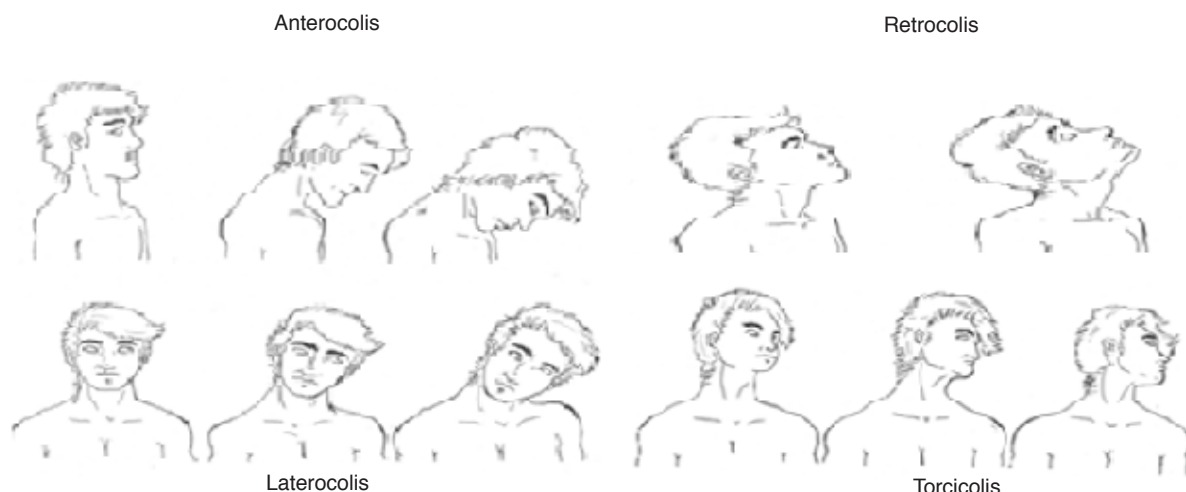


Figure 1 - Representation of the figures of EV (visual scale) subtype of dystonia

Variable	Cases (n=27)	Controls (n=27)	p
Age (years) – average ± DP	51,4 ± 15,9	41,9 ± 14,0	0,023*
Sex – n(%)			1,000***
Men's	9 (33,3)	9 (33,3)	
Female	18 (66,7)		
Age of onset	36,74 ± 17,60		
Disease duration (years) – average ± DP	14,7 ± 12,46		
Subtype – Anterocolis n(%)	3 (11)	18 (66,7)	
Subtype – Retrocolis n(%)	3 (11)		
Subtype – Laterocolis n(%)	9 (33)		
Subtype – Torcicolis n(%)	12 (44)		
Presence of painful symptoms n(%)			0,241***
Yes	21 (77,8)	16 (59,3)	
No	6 (22,2)	11 (40,7)	
Pain frequency (x / week) – average ± DP	4,8 ± 2,2	5,1 ± 1,7	0,588*
Pain scale – average ± DP	6,43 ± 2,01	5,47 ± 2,03	0,168*

Table 1 - Characteristics of the sample of cases and controls

* Test t-student; *** Chi-square test; # Statistically significant association by the testing of waste adjusted to 5% significance.

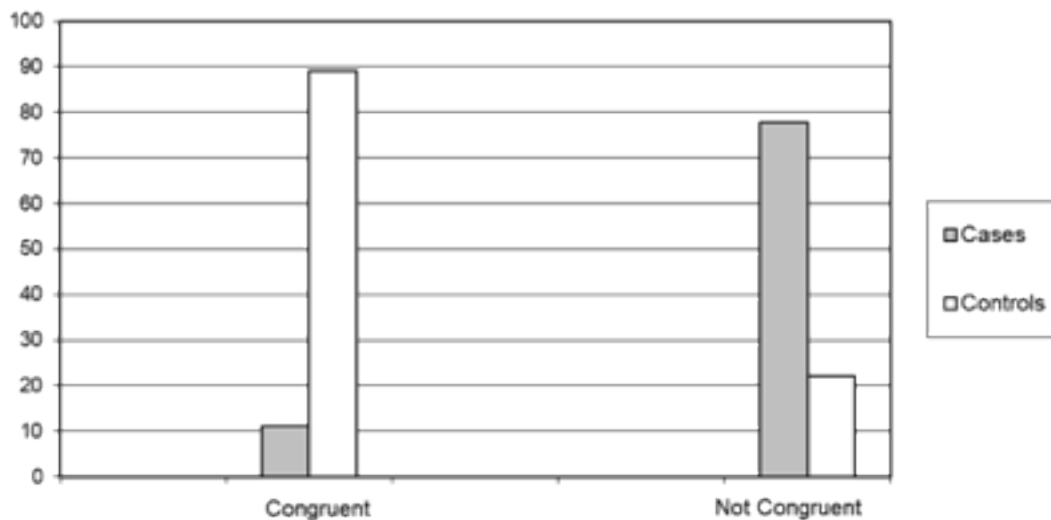


Figure 2 - Perception of cervical position in the group cases and controls

Positions	Kappa Pabak		IC 95%		P	
	Cases	Controls	Cases	Controls	Cases	Controls
Anterocolis	0,50	1,00	0,00 a 1,00	0,71 a 1,00	0,221	<0,001
Retrocolis	-0,20	-0,50	-0,61 a 0,21	-1,00 a 0,15	0,527	0,317
Laterocolis	0,17	0,86	-0,14 a 0,47	0,59 a 1,00	0,480	<0,001
Torcicolis	0,15	0,64	-0,02 a 0,32	0,44 a 0,84	0,204	<0,001

Table 2 - Comparison of the visual analog scale and goniometry in different types of cervical dystonia

ID	Visual Scale Antero	Gonio Antero	Visual Scale Retro	Gonio Retro	Visual Scale Torti	Gonio Torti	Visual Scale Latero	Gonio Latero
1	B	B	B	C	D	B	C	B
2	B	B	A	B	B	B	C	B
3	B	B	B	B	A	B	B	B
4	A	B	A	B	B	B	C	B
5	B	B	B	B	B	B	B	B
6	B	B	B	B	B	B	C	B
7	B	B	B	B	A	B	B	B
8	A	B	B	B	C	B	C	B
9	D	B	A	B	D	C	D	B
10	A	B	A	B	B	C	C	B
11	B	B	B	B	B	B	B	B
12	C	B	A	B	C	C	D	B
13	A	B	A	B	B	C	A	B
14	B	B	A	B	D	C	B	B
15	B	B	A	B	A	B	B	B
16	A	B	B	C	A	C	C	B
17	C	B	D	B	D	B	C	D
18	B	B	A	B	A	B	C	C
19	A	B	B	C	B	B	D	D
20	C	B	B	B	C	B	C	D
21	A	B	B	B	B	B	C	B
22	D	D	C	B	B	B	D	B
23	B	B	B	B	B	B	B	B
24	B	B	A	B	A	B	B	B
25	B	D	A	B	B	B	B	B
26	B	B	B	C	B	C	B	D
27	A	B	A	B	B	C	C	B

Table 3 - Case group comparison as the visual scale (perception) and goniometry

Legend: Consider for anterocolis: A = 0 (not corresponding to posture), B = 1° - 15°, C = 16° - 30°, D = 31° - 65°. For Retrocolis: A = 0 (not corresponding to posture), B = 1° - 20°, C = 21° - 50°. For Torticolis: A = 0 (not corresponding to posture), B = 1° - 15°, C = 16° - 30°, D = 31° - 55°. For Laterocolis: A = 0 (not corresponding to posture), B = 1° - 10°, C = 11° - 20°, D = 21° - 40°.

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