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Research Article

Screening for Scoliosis in Children and Adolescents with Cerebral Palsy: A Pilot Study

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Abstract

Introduction: Cerebral palsy (CP) is a non-gradual brain disease, caused by damage to the brain, resulting in permanent motor impairment. Vertebral deformity in a patient with CP has a multifactorial cause, muscle weakness, spasticity and inefficient muscle control. The aim of this study was to perform a screening of neuromuscular scoliosis in children and adolescents with cerebral palsy.

Methods: A prospective, observational and cross-sectional study, where children with CP and healthy aged 3 to 15 years were evaluated. The trunk rotation angle (ATR) of children with cerebral palsy was measured using the Scoliometer app on cervical, thoracic and lumbar spine regions. The Early Clinical Assessment of Balance (ECAB) was also applied to assess trunk balance.

Results: Thirteen children diagnosed with cerebral palsy were evaluated. Four children were able to move independently (functional group) and nine children were functionally incapacitate (non-functional group). The ATR of functional CP children were 5.25 ± 2.0 (cervical), 10.25 ± 2.1 (thoracic), 4.75 ± 2.1 (lumbar) and 10.25 ± 2.1 (highest ATR). The ATR of non-functional CP children were 5.88 ± 3.4 (cervical), 14.33 ± 9.9 (thoracic), 14.89 ± 10.3 (lumbar) and 16.13 ± 10.5 (highest ATR). The trunk balance were 38.63 ± 3.6 (functional group) and 18.11 ± 12.2 (non-functional group). Healthy children presents ATR = 13.08 ± 8.4 (highest ATR).

Conclusions: All children with CP had ATR > 7°. Non-functional group have a significant higher lumbar angle of trunk rotation ($p = 0.01$) and worse trunk balance than functional children with CP ($p = 0.0009$). Children with CP had a significant higher ATR than healthy children ($p < 0.0001$).

Keywords: Scoliosis; Cerebral Palsy; Chronic; Trunk; Spine; Diagnosis

Abbreviations

CP: Cerebral Palsy; ATR: Angle of Trunk Rotation; GMFCS: Gross Motor Function Classification System; ECAB: Early Clinical Assessment of Balance; SOSORT: International Society on Scoliosis Orthopaedic and Rehabilitation Treatment

Introduction

Chronic encephalopathy of childhood or cerebral palsy (CP) was first described in 1843 by William John Little, an English orthopedist. The expression cerebral palsy appeared in 1897 and includes several conditions that compromise the immature central nervous system, and the motor disorder is the most evident manifestation [1]. It results from brain injury in the maturation phase, resulting in motor dysfunction such as movement, postural and tonus disorders [2].

Risk factors for CP are those that negatively influence by mother health, such as the condition and nutrition of the baby, delivery, hypoxic or traumatic events in the perinatal period, prematurity and low birth weight. In addition to regional malformations with motor deficits, defects in migration and embryogenesis are common etiologies of cerebral palsy and may occur in children who do not have a history of gestational or perinatal risk [3]. In countries such as Brazil, which has a notable regional diversity and inequalities, it is possible that the prevalence in population groups is higher [4]. In Brazil, studies reveal that for every 1,000 children that are born, seven are carriers of CP, with the emergence of approximately 30,000 to 40,000 new cases per year [5].

The motor disorder presents in abnormal patterns of posture and movements, associated with abnormal postural tone. The lesion that affects the brain does not progress but interferes with the child's motor development [6]. CP can also be classified according to changes in the child's movements and also based on the anatomical location and topography of the lesion symptom. CP can thus be divided into four groups: spastic, ataxic, dyskinetic/dystonic or choreoathetotic and mixed [7].

Among tonic alterations, spasticity is the most common, corresponding to 75% of cases [8]. The spastic type can be divided into: diplegia, quadriplegia, hemiplegia and double hemiplegia. As spasticity predominates in some muscles, the appearance of deformities in this type of cerebral palsy is common [9].

Scoliosis is a three-dimensional morphological deformation of the spine that is characterized by the lateral inclination of the vertebrae in the frontal plane, rotation in the axial plane and their posteroflexion in the sagittal plane [10]. According to the Scoliosis Research Society – SRS), the lateral curvature of the spine with a Cobb angle greater than 10° is called scoliosis, measured on an anteroposterior radiograph of the patient in orthostatism [11].

The risk of scoliosis is 1% for GMFCS level I at 10 years old and 5% at 20 years, but 30% for GMFCS V at 10 years and 80% at 20 years [12]. Risk factors for progressive neuromuscular scoliosis include scoliosis of 40° or more, thoracolumbar curves, and total body involvement [13]. Scoliosis progression does not stop at skeletal maturity at curves of 40° or more. In children with CP-associated scoliosis, internal rotation and reduced range of flexion on the high side of the hip can result in problems with sitting [14].

The abnormal curvature is a consequence of the gravity effect on the spine, posterior spinal and abdominal muscles is weak during the accelerated growth period, the degree of deformity is related to neuromuscular involvement and locomotion capacity [15]. Neuromuscular scoliosis develops secondary to muscle imbalance, congenital disorders, degenerative diseases or syndromes, where the pace of its progression is conditioned to the children growth [11].

Spasticity, muscle weakness and incomplete muscle control found in CP cases contribute to impaired trunk control and development of spinal deformities. Severe scoliosis can cause additional motor dysfunction, problems with sitting and transferring, compromised cardiopulmonary function, pain and reduced quality of life [16].

The International Society on Scoliosis Orthopaedic and Rehabilitation Treatment (SOSORT) recommends school-based screening programs for early detection, identification of adolescents at risk of developing scoliosis and those with curves that require conservative treatment [17]. Therefore, the primary aim of this work is to perform the screening of neuromuscular scoliosis in children and adolescents with cerebral palsy. The secondary objectives of this study were: to compare the curves according to the locomotion skill, to correlate the curve degree with trunk balance, and to compare the curvatures between children with and without cerebral palsy.

Materials and Methods

This study was an observational, prospective cross-sectional study approved by the Institutional Review Board under protocol CAAE: 38195320.0.0000.5020. The volunteers were only evaluated after signing the informed consent statement by those responsible.

The study sample consisted of subjects aged 3 to 15 years with a confirmed CP diagnosis. The inclusion criteria for the study were: having a confirmed diagnosis of CP, being under 18 years of age, and being a resident on Coari, a town in a remote area of Amazonia and/or surrounding riverside communities. Will be excluded from the study volunteers who have congenital diseases that can affect the musculoskeletal system such as primary myopathies or muscular dystrophies, have undergone previous surgery on the spine, inability to collaborate in the evaluation tests, if their responsible does not authorize the evaluation of the volunteer, presence of COVID-19 signs and symptoms in the last 15 days. For comparison purposes, healthy children of the same age were recruited. The inclusion criteria were being residents of the same region and aged between 3 and 15 years old. Children with any congenital disease and/or congenital physical deformity or resulting from trauma were excluded.

To spinal evaluation, a digital inclinometer will be used through the smartphone app Scoliometer version 2.4 (Figure 1). The assessment follows the protocol described by Penha., *et al*.18. Volunteers able to maintain themselves in an upright position, they will be instructed to flex their trunk anteriorly, keeping their feet approximately 15 cm apart, knees extended, shoulders relaxed, elbows extended, and palms of the hands touching and positioned in front of the knees. The volunteer performed a trunk flexion to verify the greatest angle of rotation of the trunk of the cervical, thoracic and lumbar spine. Volunteers unable to maintain himself in an upright posture, he will be positioned seated on a platform, with his feet on the ground, and will perform the same procedure as for flexing the trunk.

Figure 1: Evaluation of the Angle of Trunk Rotation (ATR) of a child with cerebral palsy.

The inclinometer will be positioned on the vertebrae spinous process of the thoracic, cervical and lumbar spine, and the highest Angle of Trunk Rotation (ATR) value found will be noted on the evaluation form. To facilitate, the spinous processes of the seventh cervical, eighth thoracic, and first lumbar vertebrae were marked with self-adhesive labels. The measurement was performed three times with the volunteer returning to the upright position between each test, the highest value observed being considered. When the ATR is greater than or equal to 7° in any of the regions (thoracic, thoracolumbar and lumbar), the volunteer will be indicated of a possible scoliotic curve.

The trunk balance of these patients was evaluated using the Early Clinical Assessment of Balance (ECAB) version 2. The ECAB is an instrument that quantifies posture stability and can be used to assess children and adolescents with CP at all levels of the Gross Motor Function Classification System (GMFCS). It aims to assess head and trunk control during static and dynamic activities. The first part assesses postural control of the head and trunk through seven items, the second part assesses postural control sitting and standing, with a score of 64 points. The lower the score, the worse the subject trunk balance.

GraphPad Prism v8.0 software (GraphPad Software, SanDiego, CA, USA) was used for tabulation, graphing and analysis of statistical data. To verify the distribution and normality of the quantitative data, the Komogorov-Smirnov test was used. For mean comparison analysis, the unpaired t test was used. To verify the association between ATR and trunk balance, Spearman's correlation was used.

Results

Participated in this study (n = 13) individuals with cerebral palsy (9 children and 4 adolescents) with a mean age of 7.7 years, ranging from 3 to 15 years old. The CP volunteers were divided into two groups according to ability to move independently (Functional Group) and inability to move (Non-Functional Group).

Table 1 presents the functional and non-functional characteristics of the participants with their respective ATR with cervical, thoracic, lumbar and mean measurements. Cervical ATR was the lowest angle rotation in comparison to other spinal regions and was similar on both groups. Thoracic ATR was the higher angle on

	Functional Group	Non-Functional Group		
n	4	9		
	Mean/SD	Mean/SD	Mean difference	P value
Age	7 ± 4.7	8.8 ± 5.7	---	---
Cervical ATR	5.25 ± 2.0	5.889 ± 3.4	0,6389 ± 1,534	0.68
Thoracic ATR	10.25 ± 2.1	14.33 ± 9.9	4,083 ± 3,449	0.26
Lumbar ATR	4.75 ± 2.1	14.89 ± 10.3	10,14 ± 3,608	0.01*
Average ATR	6.75 ± 0.8	10.4 ± 6.6	3,69 ± 2,38	0.16
Highest ATR	10.25 ± 2.1	16.13 ± 10.5	5,875 ± 3,860	0.16
Trunk Balance	38.63 ± 3.6	18.11 ± 12.2	-20,51 ± 4,477	0.0009*

Table 1: Functional and non-functional characterization of individuals with cerebral palsy ATR (Angle of Trunk Rotation). *: there was a statistically significant difference for $p < 0.05$.

functional group, while the greater value was the lumbar ATR on non-functional group.

It is noted that the non-functional group had a greater ATR at all levels of the spine and a worse result for trunk balance. There was no significant difference between groups for cervical and thoracic ATR. However, there was a statistically significant difference on lumbar ART, as in the functional group the ART mean = 4.75, while in the non-functional group mean = 14.89 (p = 0.01). There was also no difference between the groups when comparing the mean spinal ATR (the average between cervical, thoracic and lumbar values) or the highest ATR value across the entire spine. The trunk balance of the non-functional group was statistically lower than the functional group.

The results presented in figure 2 refer to the association of ATR and trunk balance. It was observed that greater trunk balance was associated with a decrease in the angle of rotation. However, the correlation values were considered moderate to low (Cervical: R = -0.07; Thoracic: R = -0.3; Lumbar: R = -0.4).

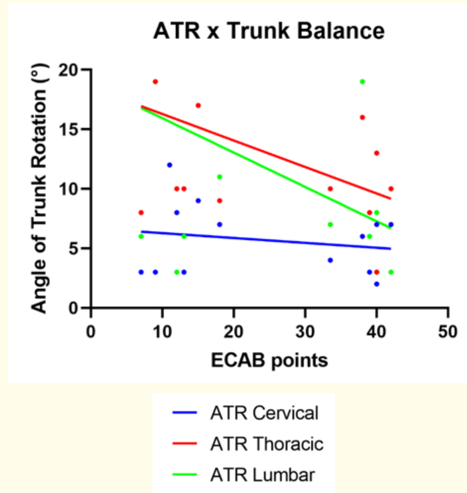

Figure 2: Graph of dispersion showed a relationship between the angle of trunk rotation and trunk balance level measured by ECAB.

Regarding scoliosis screening, when the angle of rotation is greater than 7º, the patient is indicated to undergo an X-ray to identify possible scoliotic deviation. In view of this, all patients (n = 13) presented some rotation angle greater than 7º, which may indicate a possible deviation.

For the purpose of comparing healthy subjects with CP, 34 healthy children with a mean age of 9.8 ± 1.1 years were recruited. Children with CP had a statistically higher ATR (13.08 ± 8.4) than healthy children (1.21 ± 0.2) ($p < 0.0001$).

Discussion

The results show that individuals in the non-functional group (who are dependent for locomotion) had a greater degree of trunk rotation, therefore a greater risk for scoliosis, and also not having trunk balance. The functional group (who move independently) showed more trunk balance and a lower level of trunk rotation, presenting a lower risk for scoliosis. The spine level whose rotation showed the higher differences was in the lumbar region.

According to Majnemer., *et al*. [19], individuals with cerebral palsy achieve postural control and balance as a result of the connection between several factors such as: injury in the Central Nervous System maturation, neuromuscular and somatosensory system impairments and other possible signs and symptoms. In addition, it is normal for these people to have changes in stability, posture and balance, which are essential requirements for the development of locomotor skills, as well as for the effective performance of activities of daily living [20]. According to Bax., *et al*. [21], this loss of stability leads to body imbalance and can cause the individual to lose part of their functionality.

According to Cunha., *et al.* [22], there was a significant moderate to high correlation between postural alignment and motor function on the sitting dimension in children with CP (R = 0.748; p = 0.01). These findings suggest that how much better a postural alignment, better the motor function. Therefore, in these cases, a greater trunk balance is expected.

According to our findings, there was a difference on the averages of the lumbar ATR between in functional and non-functional children. This has become a new and relevant finding, since the occurrence of changes in the lumbar curvatures is more uncommon in relation to the thoracic or thoracolumbar. Prudente., *et al.* [23] reports that this theme is still little studied, however extremely important, as there are increased motor and cognitive disorders that lead to more dependent the child becomes. Mancini., *et al*. [24] demonstrate that CP children with a more severe motor level present lower performance in all areas of functional impairment.

Musculoskeletal changes that involve muscle weakness, shortening, contracture and atrophy, little pelvic movement with fixations in pelvic anteroversion or retroversion, slowing motoneuron recruitment, modulation problems on muscle recruitment, as well as loss of control of selective movements, dexterity and co-contraction, are common in children with CP, and this makes postural control sitting and standing is worse than in healthy children, in addition to causing delay in the start of movement [25-27].

The risk of developing scoliosis in individuals with CP increases with advancing age and at high levels in the GMFCS. Persson-Bunke., *et al*. [28] observed that the GMFCS level was the only significant factor in the Cobb angle increase, showing that children at GMFCS IV and V have a 50% chance of acquiring moderate or severe scoliosis at 18 years, with the risk of deformity progression. These GMFCS levels correspond to our classification of the non-functional group (unable to move independently) and it was in this group that we observed higher ATR values.

Our study has some limitations that should be mentioned. Only the ATR measurement was performed, and the ideal was also to use a radiograph to measure a possible scoliosis. However, future data will be collected and presented with panoramic radiographs. Another limitation of our study was the relatively low number of cases. Due to the low number, it was not possible to stratify the sample according to the GMFCS, so we used the division between functional and non-functional. As this is a study in a town whose HDI is very low (0.586), many patients live in rural and riverside areas and have difficult access to participate in the study. However, the use of mobile solutions has allowed access to remote areas by health professionals. Therefore, this pilot study showed that it is possible to expand the column evaluation of this population with CP.

Conclusion

Our study concluded that children with cerebral palsy who are unable to independently locomotion have a higher lumbar angle of trunk rotation and worse trunk balance than functional children with CP. All children with CP present ART > 7°, that supposing a deviation in the curvature of the spine. There was a low to moderate negative correlation between trunk balance and thoracic and lumbar ATR, respectively. Children with CP present a significant higher ATR value than healthy children.

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Conflict of Interest

All authors declare no conflict of interest.

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