Vestibular symptoms in children with spastic cerebral palsy

Sintomas vestibulares em crianças com paralisia cerebral espástica

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Abstract

Introduction: In children with spastic cerebral palsy (CP), the factors associated with the development of symptoms of vestibular system dysfunction are not yet fully understood. **Objective:** To determine the association between clinical symptoms of vestibular system disorders and the severity of gross motor function impairment, characteristics of the brain lesion and age at the beginning of physiotherapy in a sample of children with spastic CP. **Methods:** A cross-sectional study was conducted involving 82 children with spastic CP. Assessments were performed of gross motor function and characteristics of the brain lesion, along with a vestibulo-oculomotor clinical examination. **Results:** The severity of gross motor function impairment, location of the brain lesion (motor cortex) and age at which the child began physiotherapy (more than 2 years of age) were associated with symptoms of vestibular system disorders, specifically with abnormalities of the vertical and horizontal vestibulo-ocular reflexes and the presence of central nystagmus. **Conclusion:** The severity of the impairment of gross motor functions, brain lesion (motor cortex) and the age at which the child began physiotherapy are associated with symptoms of vestibular system disorders.

Keywords: Cerebral palsy. Vestibular system. Brain injury. Motor skills. Physiotherapy.

Resumo

Introdução: *Em crianças com paralisia cerebral (PC) espástica, os fatores associados com o desenvolvimento de sintomas de disfunções do sistema vestibular ainda não estão totalmente esclarecidos.* ***Objetivo:*** *Determinar a associação entre sintomas clínicos de distúrbios do sistema vestibular e a gravidade do comprometimento da função motora grossa, características da lesão cerebral e idade no início da fisioterapia em uma amostra de crianças com PC espástica.* ***Métodos:*** *Um estudo transversal foi conduzido envolvendo 82 crianças com PC espástica. Foram realizadas avaliações da função motora grossa e características da lesão cerebral, juntamente a um exame clínico vestíbulo-oculomotor.* ***Resultados:*** *A gravidade do comprometimento da função motora grossa, a localização da lesão cerebral (córtex motor) e a idade em que a criança iniciou a fisioterapia (mais de 2 anos de idade) foram associadas a sintomas de distúrbios do sistema vestibular, especificamente com anormalidades dos reflexos vestíbulo-oculares verticais e horizontais e com a presença de nistagmo central.* ***Conclusão:*** *A gravidade do comprometimento das funções motoras grossas, a lesão cerebral (córtex motor) e a idade em que a criança iniciou a fisioterapia estão associados aos sintomas de distúrbios do sistema vestibular.*

Palavras-chave: *Paralisia cerebral. Sistema vestibular. Lesão cerebral. Habilidades motoras. Fisioterapia.*

Introduction

Inadequate balance control is related to the brain lesion in cerebral palsy (CP), which affects essential areas of movement control, determining sensory-motor abnormalities.[1–3] Impaired bodily balance can be seen in the early development of affected children and has repercussions in the form of substantial limitations in the performance of motor activities and participation.[4]

Besides impairments directly related to the neurological lesion, such as spasticity, restrictions to active movements, including those of the head and trunk, which exert a direct impact on the development of the vestibulo-ocular and vestibulo-spinal reflexes, tend to aggravate the impaired bodily balance.[5] Thus, although children with CP do not have a greater likelihood of having structural lesions of the vestibular system due mainly to the intact peripheral vestibular function, symptoms of disorders of the processing of vestibular information are often found in this population.[6] Such disorders can exert a negative impact on the rehabilitation process,[7] further compromising motor performance, and even the implementation process of augmentative and alternative communication, which is often performed through ocular movements that control communication programs.

Although the literature describes a considerable frequency of symptoms of vestibular disorders in children with CP, evidence on factors related to the development of such disorders is limited and generally restricted to children with walking ability categorized on levels I to III of the Gross Motor Function Classification System (GMFCS).[6] The understanding of factors related to vestibular symptoms can contribute to a better understanding of the clinical condition and the selection of more effective rehabilitation strategies.[7] Therefore, the aim of the present study was to investigate associations between clinical symptoms of vestibular system disorders and the severity of gross motor function impairment, characteristics of the brain lesion and age at the beginning of physical rehabilitation in a sample of children with spastic CP. The hypothesis of the study was that the presence of vestibular symptoms would be associated with the severity of gross motor function impairment as well as the anatomical and etiological characteristics of the brain lesion.

Methods

A cross-sectional study was conducted with children recruited from the Pediatric Neurostimulation Center (CENEPE REAB), in São Paulo, Brazil. The study was approved by the ethics committee of Faculdade de Ciências Médicas da Santa Casa de São Paulo, under the number 80601924.2.0000.5479. The legal guardians and participants signed an informed consent form and no financial compensation was provided.

Participants

From October 2024 to January 2025, the children were screened based on the eligibility criteria. The following were the inclusion criteria: 1) diagnosis of spastic CP confirmed through a clinical examination; 2) magnetic resonance exams demonstrating a lesion in the pyramidal system with no impairment of the extrapyramidal system

or cerebellum; 3) age between 6 and 12 years; 4) agreement from a legal guardian by signing a statement of informed consent; and 5) acceptance from the participant through a term of assent. The exclusion criteria were: 1) children who had neurological or neuromuscular diseases or syndromes along with CP; 2) those who had been submitted to orthopedic or neurological surgeries in the 12 months prior to the assessment procedures; 3) those who had orthopedic deformities with an indication for surgery; and 4) those with a degree of cooperation incompatible with the adequate performance of the proposed activities.

Outcome measures

All assessment procedures were performed in an environment reserved to the development of the study, at CENEPE REAB, with individual scheduling according to the availability of the participant's guardian.

The participants were assessed on two non-consecutive days. On the first day, clinical history was collected, neuroimaging exams were analyzed, and gross motor function was evaluated. On the second day, the vestibular system was examined. All assessment procedures were conducted by three neurofunctional physiotherapists who were adequately trained to perform each stage of the evaluation.

Each participant was assessed by a single physiotherapist; however, the vestibular examination was recorded on video to ensure increased methodological rigor. The vestibular findings described by the evaluator were subsequently reanalyzed by a physiotherapist with 20 years of experience in vestibular assessment and rehabilitation, with the aim of increasing the reliability of the results.

Gross motor function

Gross motor function was classified according to the five levels proposed by the GMFCS.[8] Moreover, the reference curves for the gross motor function measure were used, which consider motor function measured by the Gross Motor Function Measure-66 (GMFM-66), considering age and GMFCS level. The reference curves enable the analysis of gross motor function in percentiles (3rd, 5th, 10th, 25th, 50th, 75th, 90th, 95th and 97th). For the purposes of analysis, the results were categorized considering gross motor function equal to or above the 50th percentile or below the 50th percentile.[9]

Clinical history

Medical reports and patient records were consulted to collect data on the etiology of the brain lesion and the identification of the time of the lesion (prenatal, perinatal or postnatal). The guardians were asked about the time in which the physiotherapeutic intervention was initiated, which, for the purposes of analysis, was categorized as up to 2 or after 2 years of age.

Location of brain lesion

The structural magnetic resonance exams performed for the confirmation of the diagnosis of spastic CP were analyzed by a specialist to categorize the location of the brain lesion: cortical lesion – lesions involving the primary motor cortex that could extend to the underlying white matter; and subcortical lesion – deeper lesions of the internal capsule (excluding the cerebral cortex, brainstem and cerebellum).[10]

Vestibulo-oculomotor examination

The examination was performed with the children in the sitting position. Those on GMFCS levels IV and V were adequately position with trunk support. For cases in which the child was unable to perform head movement voluntarily, the movements were performed by the examiner. The oculomotor examination consisted of the assessment of the ocular range of motion (capable or not to follow a target), inspection of the presence or absence of nystagmus (spontaneous, semi-spontaneous and after head shaking) and categorization of nystagmus (characteristics of peripheral or central nystagmus). During the examination, toys were used as targets to ensure the children's attention. Ambient lighting was controlled to maintain pupil stability. Each variable was assessed only once to minimize participant discomfort.

Initially, the presence of lateral head tilt, spontaneous eye movements, and ocular alignment were observed. Ocular inspection was performed through direct observation by the evaluator to assess spontaneous nystagmus, using a fixed target positioned one meter from the participant. Subsequently, the evaluator instructed the child to follow a moving target in front of them, which was moved by drawing the letter "H" in the air, to evaluate the amplitude and integrity of the oculomotor system.[11]

In another stage of the oculomotor assessment, the presence or absence of nystagmus was observed

following passive head movements. With the child properly seated and starting from a neutral head position while fixating on a target in front, the evaluator passively moved the head slowly to the sides (15 repetitions). Eye movements were observed after the completion of each head movement. Nystagmus was categorized as present or absent, and its probable origin was noted as peripheral or central. Monoplanar nystagmus, or nystagmus without a torsional component that was not inhibited by visual fixation mediated by the reticular formation, was classified as of central origin.[11]

The assessment was concluded by documenting signs and symptoms of discomfort during head movements and position changes from supine to sitting. Discomfort was identified through observation and/or self-report by the child, considering facial expressions, eye closure during movement, reports of vertigo or dizziness, imbalance or sensations of body instability, and nausea.

Horizontal and vertical vestibulo-ocular reflex (VOR)

The ability to stabilize the vision during head movements was assessed with the participants in the sitting position. Those classified between GMFCS levels I and III remained sitting without support. Those on levels IV and V were adequately positioned in the sitting position with support in the trunk region. A fixed target was positioned at a distance of approximately one meter. After confirmation that the child was able to see the target, the participant was instructed or assisted, if necessary, to move the head horizontally, during the assessment of horizontal VOR and verticall, during the assessment of vertical VOR at a range of 35 degrees, with a velocity of 2 Hz or 120 bpm, measured with a metronome (Figure 1). The assessment was solely clinical, without the use of equipment. VOR gain was considered altered when the child exhibited difficulties in stabilizing their gaze during head movements, which could be accompanied by reports of discomfort such as vertigo, nausea, or body imbalance. An alteration in VOR gain indicates a dysfunction in the automatic response of the vestibular system that controls eye movements in response to head movements, leading to instability of the image on the retina during such movements. To confirm the alteration, three tests were performed for both horizontal and vertical VOR.[11]

Statistical analysis

Descriptive analysis of the sample and variables of interest was performed using measures of central tendency and dispersion for quantitative variables and absolute and relative frequency for categorical variables. We conducted chi-square (X^2) analyses to determine whether the clinical characteristics, characteristics of the brain lesion and gross motor function impairment were associated with symptoms of vestibular processing disorders. The strength of the associations between these variables was analyzed using Cramer's V for chi-square analyses. A p-value < 0.05 was indicative of a statistically significant result.

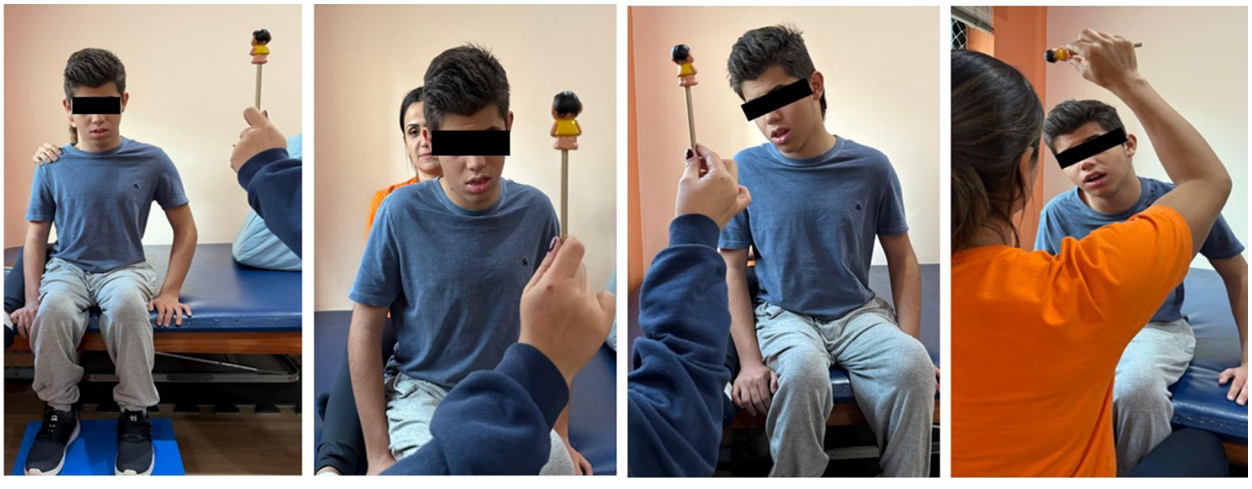

Figure 1 - Demonstration of the assessment procedures performed in the vestibular system examination.

Results

The sample was composed of 82 children with spastic CP (43 girls and 39 boys), with a mean age of 6.2 ± 3.8 years. Table 1 displays the clinical characteristics of the participants.

Table 1 - Clinical characteristics and characteristics of brain lesion in sample studied

Clinical characteristics	n (%)
Spastic cerebral palsy	
Hemiparesis	16 (19.5)
Paraparesis	28 (34.1)
Quadriparesis	38 (46.3)
Gross Motor Function Classification System	
GMFCS I	8 (9.7)
GMFCS II	10 (12.1)
GMFCS III	13 (15.8)
GMFCS IV	29 (35.3)
GMFCS V	22 (26.8)
Gross motor function curve	
≥ 50th percentile	28 (34.1)
< 50th percentile	54 (65.8)
Location of brain lesion	
Cortical	61 (25.6)
Subcortical	21 (25.6)
Etiology of brain lesion	
Hypoxic-ischemic	60 (73.1)
Brain hemorrhage	19 (23.1)
Possible congenital malformation	1 (1.2)
Not defined	2 (2.4)
Time of brain lesion	
Prenatal	1 (1.2)
Perinatal	52 (63.4)
Postnatal	27 (32.9)
Not defined	2 (2.4)
Beginning of physical rehabilitation	
Before 2 years of age	60 (73.1)
After 2 years of age	22 (26.8)

Considering the levels of gross motor function, the statistical analysis demonstrated a significant association between the GMFCS level and both abnormal horizontal and vertical VOR and nystagmus of a central origin after head shaking, with a moderate correlation between these variables. The results demonstrated that greater impairment of gross motor function (levels IV and V) was related to a more frequent occurrence of these symptoms of disorders in the processing of vestibular stimuli (Table 2). Figure 2 illustrates the categorization of nystagmus according to the GMFCS level. There was no change in the type of nystagmus observed during the clinical-functional examination of the vestibular system.

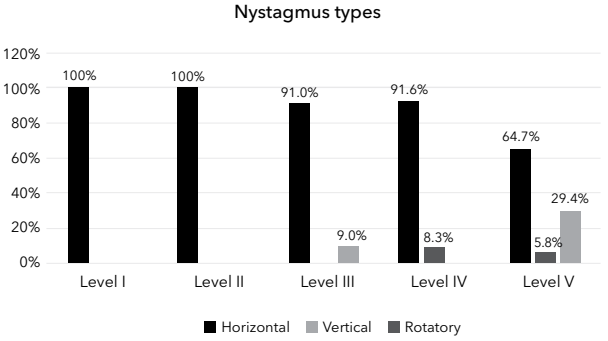

Figure 2 - Types of nystagmus presented by the study population, considering the levels of gross motor function.

The association between vestibular symptoms and gross motor function was also analyzed considering the percentiles obtained by the sample on the gross motor function reference curves. The statistical analysis demonstrated significant associations between the gross motor functions of the participants and abnormal horizontal VOR horizontal ($X^2 = 7.6$, $p = 0.006$, Cramer's V = 0.30), abnormal vertical VOR ($X^2 = 5.4$, $p = 0.019$, Cramer's V = 0.25), the observation of nystagmus after head shaking ($X^2 = 3.9$, $p = 0.046$, V = 0.22) and nystagmus of a central origin ($X^2 = 5.6$, $p = 0.022$, Cramer's V = 0.25), demonstrating an association between these symptoms and poor performance with regards to gross motor functions (Figure 3A).

The location of the lesion (cortical or subcortical) was also significantly associated with abnormal horizontal VOR ($X^2 = 44.1$, $p < 0.001$, Cramer's V = 0.73), abnormal vertical VOR ($X^2 = 32.0$, $p < 0.001$, Cramer's V = 0.62) and the observation of nystagmus of a central origin ($X^2 = 6.3$, $p = 0.012$, Cramer's V = 0.27). These symptoms were associated with cortical lesions (Figure 3B).

The beginning of physiotherapeutic treatment after two years of age was associated with abnormal horizontal VOR (X^2 = 7.8, p = 0.005, Cramer's V = 0.31) and abnormal vertical VOR (X^2 = 6.1, p = 0.013, Cramer's V = 0.27), but with a weak correlation. This variable was not associated with any of the other variables of interest (Figure 3C). Likewise, the other variables analyzed, such as the topography of motor impairment (hemiparesis, paraparesis or quadriparesis), etiology of the lesion and time of the lesion (prenatal, perinatal or postnatal), were not associated with the symptoms of vestibular disorders studied.

Table 2 - Frequency of symptoms of dysfunctions in the processing of information of the vestibular system according to level of gross motor function

Dysfunctions	GMFCS I n = 8	GMFCS II n = 10	GMFCS III n = 13	GMFCS IV n = 29	GMFCS V n = 22	Total n = 82
Abnormal horizontal vestibulo-ocular reflex	3 (37.5)	5 (50.0)	9 (69.2)	26 (89.6)	22 (100)	n = 65 (79.2%) x^2 = 22.1 p < 0.001 Cramer's V = 0.52
Abnormal vertical vestibulo-ocular reflex	4 (50.0)	6 (60.0)	10 (76.9)	26 (89.6)	22 (100)	n = 68 (82.9%) x^2 = 15.6 p = 0.004 Cramer's V = 0.43
Spontaneous nystagmus	1 (12.5)	3 (30.0)	8 (61.5)	17 (85.6)	11 (50.0)	n = 40 (48.7%) x^2 = 7.6 p = 0.107 Cramer's V = 0.30
Nystagmus after head shaking	2 (12.5)	5 (50.0)	11 (84.6)	24 (82.7)	17 (77.2)	n = 59 (71.9%) x^2 = 14.1 p = 0.007 Cramer's V = 0.41
Nystagmus of central origin	2 (25.0)	3 (30.0)	11 (84.6)	24 (82.7)	16 (72.7)	n = 56 (68.2%) x^2 = 19.2 p = 0.001 Cramer's V = 0.48
Nystagmus of peripheral origin	1 (12.5)	2 (20.0)	0 (0.0)	0 (0.0)	1 (4.5)	n = 4 (4.8%) x^2 = 4.6 p = 0.336 Cramer's V = 0.23
Head movement*	0 (0.0)	1 (10.0)	6 (46.1)	10 (34.4)	11 (50.0)	n = 28 (34.1%) x^2 = 10.3 p = 0.040 Cramer's V = 0.25
Postural changes*	0 (0.0)	1 (10.0)	1 (7.6)	2 (6.8)	3 (13.6)	n = 7 (8.5%) x^2 = 1.6 p = 0.805 Cramer's V = 0.14

Note: Data presented as n (%). *Signs and symptoms of discomfort. GMFCS = Gross Motor Function Classification System.

Discussion

The present study investigated associations between symptoms of vestibular system disorders and the severity of gross motor function impairment, characteristics of the brain lesion and age at the beginning of physical rehabilitation in a sample of children with spastic CP.

The importance of investigations of this type is related to the possibility of determining children with risk factors for the development of vestibular disorder and then implementing therapeutic strategies capable of preventing or diminishing the development of such disorders, which tend to aggravate the impairment of postural control.

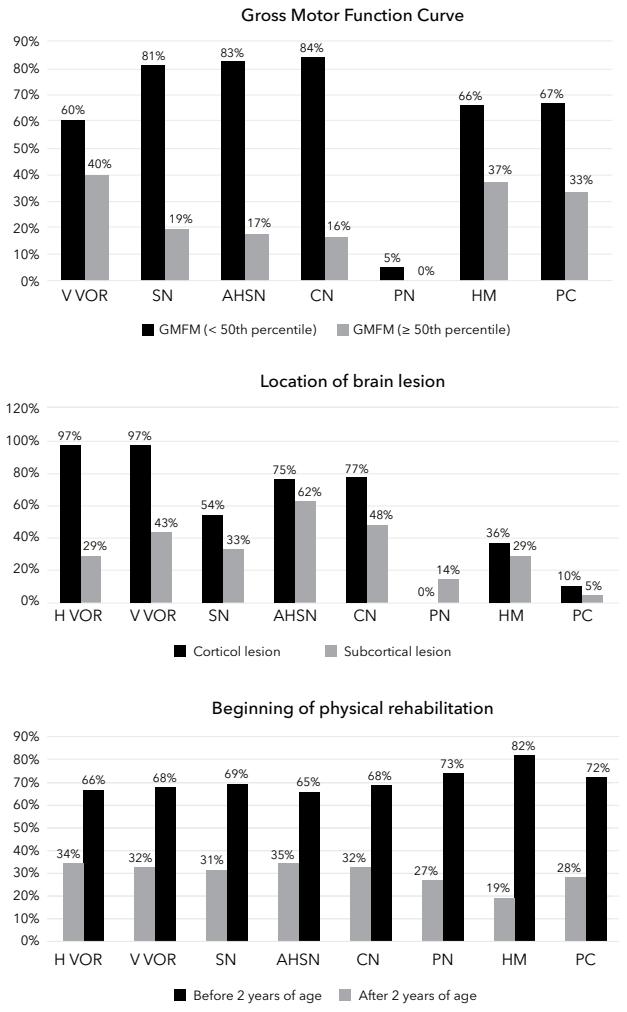

Figure 3 - Frequency of vestibular dysfunction symptoms according to gross motor function, location of brain lesion (cortical or subcortical), and age at which the child began physical rehabilitation.

Note: GMFCS = Gross Motor Function Classification System; H VOR = horizontal vertical vestibulo-ocular reflex; V VOR = vertical vestibulo-ocular reflex; SN = spontaneous nystagmus; AHSN = after head shaking nystagmus; CN = central nystagmus; PN = peripheral nystagmus; HM = head movements; PC = postural changes.

As described, evidence demonstrates that the prevalence of vestibular system disorders is high among children with CP and vestibular hypofunction can exert a negative impact on the management of bodily balance.[6,12] However, detailed knowledge about the characteristics and impact of vestibular dysfunctions in CP, especially considering the different levels of gross motor function, still requires further investigation. In a systematic review[13] aimed at establishing the prevalence of these dysfunctions in children with neurological impairments, authors identified only one high-quality study on the subject.[13]

This study reported a prevalence of 48.4% for saccular dysfunction, assessed through the Cervical Vestibular Evoked Myogenic Potential Test, in children aged 7 to 12 years with spastic CP, classified as levels I and II of the GMFCS.[14] Conversely, in the present study, the prevalence of vestibular dysfunction symptoms varied according to the severity of the clinical presentation, showing a progressive increase with the degree of motor impairment: Level I (50.0%), Level II (60.0%), Level III (76.9%), Level IV (89.6%), and Level V (100%).

In a general context, the studies cited were conducted primarily with children whose gross motor function was classified on levels I to III of the GMFCS,[6,12] which refers to children with the ability to walk independently, limiting the analysis of the impact of motor impairment severity on the frequency of symptoms of vestibular disorders. One example is the recent study conducted by Conti et al.,[15] which involved 13 children with CP, classified as GMFCS levels I to III. The study considered a wide range of clinical presentations of neurological lesions, including ataxia (n = 3), spasticity (n = 6), and other categories (n = 7). The authors presented the results of a cross-sectional study that aimed to evaluate the feasibility of assessing the function of the angular VOR using the Video Head Impulse Test (vHIT). They found that this assessment was feasible for the studied population, which had a mean age of 7.76 ± 2.65 years. Additionally, they observed that six of the 13 children had at least one dysfunctional horizontal semicircular canal, showing a strong correlation with gross motor function, as all dysfunctions were identified in children with ataxia and categorized as GMFCS level III.[15]

The sample of the present study involved all five GMFCS levels, but primarily with more severely impaired children (GMFCS IV and V). Thus, the data analysis enabled broadening knowledge on the subject, as we identified an association between the severity of motor impairment and possible vestibular disorders, such as abnormalities of the vestibulo-ocular reflex.

Children with spastic PC on levels IV and V of the GMFCS have impaired motor control, including poor control of the head and trunk and mobility restricted to a wheelchair.[8] If we consider that the vestibular system is structurally developed in the gestational period, but is a

system dependent on head movements for its functional development,[16,17] we may infer that the restrictions to mobility faced by children with CP result in restrictions with regards to stimuli offered to the vestibular system.

The restriction of stimuli offered to the vestibular system at an essential age for the development of its functioning on the central nervous system level may explain the findings of the present study. In the sample of children studied, the abnormalities found in the vestibulo-ocular reflex and the pattern of nystagmus of a central origin were associated with the GMFCS level and lower than expected performance in terms of gross motor functions. These findings demonstrate that children with greater impairment regarding the ability to execute gross motor functions are more likely to develop symptoms of vestibular system disorders.

However, as demonstrated in previous studies, children on levels I to III also have an abnormal vestibulo-ocular reflex,[6,12] often progressively increasing according to the severity of gross motor function impairment. We believe that this may be related to the delay in the motor steps of development and independent gait.

Another aspect that should be highlighted is the greater frequency of an abnormal vertical vestibulo-ocular reflex compared to the horizontal reflex. Among the postural abnormalities found in children with spastic CP, trunk flexion associated with flexion of the head is a postural pattern often found in this population.[18,19] Thus, voluntary control of the movement of head extension compromised by sequelae of the brain lesion is aggravated by the biomechanical limitations imposed by the postural pattern, reducing vertical stimuli offered to the vestibular system.[19]

Such statements should be considered possible explanations for the abnormalities observed, but it is not possible to establish a cause-and-effect relationship. However, the results of the present study offer a basis for the development of a longitudinal study in which the assessment of the vestibular system should be included early in the follow-up of children with spastic CP. Studies of this type could contribute to broadening knowledge on the development of vestibular system disorders in this population and enable the early identification of symptoms as well as the implementation of effective therapeutic strategies for treatment.

Another relevant aspect was the association between an abnormal vestibulo-ocular reflex and age at the beginning of physical rehabilitation. Beginning rehabilitation after two years of age was associated with a greater frequency of symptoms of vestibular system disorders. We believe that this result underscores the importance of early rehabilitation. Even with a reserved prognosis regarding the acquisition of gross motor functions, children placed into physical rehabilitation early experience movements and postures that contribute to the functional development of the vestibular system. Considering the substantial impact of this system on motor control, we believe that interventions focused on the stimulation of this system could contribute so that children with spastic CP reach their maximum potential in terms of postural control and overall functioning, with a possible reduction in the discomfort and insecurity with regards to movement found in some cases.

Cortical brain lesions were also associated with an abnormal vestibulo-ocular reflex. Evidence demonstrates a more reserved motor prognosis in cases of cortical compared to subcortical lesions.[20] In such cases of spastic CP, the brain lesion affects the pyramidal system, involving the primary motor cortex, with a variable extension of the lesion depending on the etiology and time in which the brain was exposed to the lesional condition. Considering the proximity of the primary motor cortex and vestibular cortex, future studies should analyze the incidence of lesions in the vestibular cortical area in cases of spastic CP to determine a possible relationship between the lesion of this brain restrictor and neurofunctional abnormalities in this population.

The present study has limitations that should be considered, such as the number of participants according to the GMFCS levels. As spastic CP is one of the most prevalent health conditions at rehabilitation services throughout the world, the number of participants was too limited to offer a basis for definitive conclusions. Another limitation is related to the vestibular system assessment methods employed in the study. There are currently high-technology instruments that enable precise assessment of the vestibular system, such as vHIT. However, many instruments are not easily adapted for use on children.

Conclusion

Based on the analysis of the present results in a sample of children spastic CP, the severity of the impairment of gross motor functions (GMFCS levels IV and V),

the topography of the brain lesion (motor cortex) and age at which the child began physical rehabilitation (older than 2 years of age) are associated with symptoms of vestibular system disorders, specifically abnormal vertical and horizontal vestibulo-ocular reflexes and the presence of central nystagmus.

Authors' contributions

AQ, CSO, VMPU, and LACG contributed significantly to the conceptualization; AQ, CSO, ALSS, and LACG, to the methodology; AQ, MOA, and VMPU, to the investigation; ALSS and LACG, to the formal analysis; AQ, CSS, MOA, ALSS, and LACG, to writing the original draft. CSO and LACG were responsible for the project administration, and ALSS for its supervision. All authors read and approved the final manuscript version.

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