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Hamstring length, gross motor function and gait in children and adolescents with cerebral palsy

Comprimento de isquiotibiais, função motora grossa e marcha em crianças e adolescentes com paralisia cerebral

Tamaño de los isquiotibiales, función motora gruesa y marcha en niños y adolescentes con parálisis cerebral

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ABSTRACT | This study aims at assessing the relationship between hamstring length, gross motor function, and gait in children with spastic cerebral palsy (CP). Children and adolescents aged between 6 and 18 years, were classified as levels I, II or III according to the Gross Motor Function Classification System. Participants were assessed using a modified Tardieu Scale to determine hamstring length, evaluating: R1 (first leg extension resistance), R2 (second leg extension resistance) and R2-R1 (difference between R1 and R2) of the left and right lower limbs. The Gross Motor Function Measure (GMFM) was used to evaluate gross motor function and the modified Physicians Rating Scale (PRS) for gait. Twenty-three participants were included in the study and the results showed a moderate correlation between R1, R2, and PRS of the left leg and the GMFM. All other variables exhibited a weak correlation. Hamstring length was weakly to moderately related to gross motor function and gait in children and adolescents with CP.

Keywords | Gait; Cerebral Palsy; Musculoskeletal System; Motor Activity.

RESUMO | O objetivo desse estudo é verificar se há relação entre o comprimento dos isquiotibiais, função motora grossa e marcha em crianças e adolescentes com paralisia cerebral (PC). Os participantes, entre 6 e 18 anos, foram classificados pelo Sistema de Classificação da Função Motora Grossa (GMFCS) nos níveis I, II e III através da escala Tardieu modificada, para avaliar o comprimento dos músculos isquiotibiais, sendo identificados em: R1 (primeira resistência da extensão da perna), R2 (segunda resistência da extensão da perna) e R2-R1 (diferença entre R1 e R2) do membro inferior esquerdo e direito. Para avaliar a função motora grossa, foi utilizado o teste *gross motor function measure* (GMFM-88), e a escala *physicians rating scale* modificada (PRS) foi utilizada para avaliar a marcha. Vinte e três participantes foram incluídos e os resultados evidenciaram correlações moderadas entre R1, R2 e PRS do membro inferior esquerdo e GMFM. As demais variáveis apresentaram uma correlação fraca.

Descritores | Marcha; Paralisia Cerebral; Sistema Musculoesquelético; Atividade Motora.

RESUMEN | El presente estudio tuvo como objetivo verificar si existe una relación entre el tamaño de los isquiotibiales, la función motora gruesa y la marcha en niños y adolescentes con parálisis cerebral (PC). Los participantes con edades entre 6 y 18 años fueron clasificados en el Sistema de Clasificación de la Función Motora Gruesa (GMFCS) en los niveles I, II y III utilizando la escala Tardieu modificada para evaluar el

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tamaño de los músculos isquiotibiales, y que los identificaron en: R1 (primera resistencia de extensión de la pierna), R2 (segunda resistencia de extensión de la pierna) y R2-R1 (diferencia entre R1 y R2) de la extremidad inferior izquierda y derecha. Para evaluar la función motora gruesa, se utilizó la prueba Gross Motor Function Measure (GMFM-88); y para evaluar la marcha, la escala *Physicians Rating Scale* modificada (PRS). Se incluyeron 23 participantes, y los resultados mostraron correlaciones moderadas entre R1, R2 y PRS del miembro inferior izquierdo y GMFM. Las otras variables tuvieron una correlación débil.

Palabras clave | Marcha; Parálisis Cerebral; Sistema Musculoesquelético; Actividad Motora.

INTRODUCTION

Gait changes in children with cerebral palsy (CP), which lead to impaired functionality, are due to changes in motor control characterized by disorders of tone, movement, and posture resulting from non-progressive damage to the developing central nervous system[1,2]. Among the tonic changes exhibited by these children, spasticity is the most common, characterized by high tone, increased tendon reflexes, and resistance to rapid passive movement[1]. The increased tone leads to the installation of movement patterns considered atypical. These patterns predispose to musculoskeletal disorders, secondarily generating biomechanical misalignments that can result in shortening, contractures, and deformities[1,2].

The pathophysiological mechanisms involved in the development of these changes are not yet fully understood, but it is known that in addition to muscle hyperactivity, the reduction in muscle growth is already observed in children with CP early at 15 months of age, which may also contribute to the emergence of muscle contractures[2,3]. These musculoskeletal complications, added to the changes in mobility, balance, and strength, gradually increase the functional limitations of these individuals throughout their development[1,4].

Reduction of the musculoskeletal system flexibility is directly associated with a reduction in the functional capability of children with spastic CP[2]. This decrease in flexibility results in a progressive reduction in hip, knee and ankle joint excursion, a shortening in gait speed and stride length, and an increase in the total support phase time[5]. The reduction in hamstring muscle length is usually observed in children with CP[5], and when this factor is associated with a progressive reduction in active knee extension during gait, a decrease in stride length and a reduction in the peak flexion of the leg during the swing phase, which results in lower walking speed and reduced stability, can be perceived[5,6].

Taking into account these children's gait disorders, this study analyzes the relationship between hamstring muscle length, gait and gross motor function in children with CP at levels I, II and III according to the gross motor function classification system (GMFCS).

METHODOLOGY

This is an observational, descriptive and cross-sectional study. The caregivers confirmed the participation by signing the free and informed consent form, at the child's will. Participants were children and adolescents diagnosed with spastic CP, aged between 6 and 18 years, classified as levels I, II or III of the GMFCS[7-10]. Participants should have the ability to understand simple orders and use anticonvulsants as long as they had not manifested seizures within the three months prior. Children and adolescents with tonic fluctuations who underwent surgical procedures in the musculoskeletal system within 12 months and who underwent botulinum toxin type A or another type of chemical block in the lower limbs up to 6 months before the assessment date were not included in the study.

The sample was selected for convenience, consecutively, and all eligible participants undergoing physical therapy at the two main rehabilitation centers in the city were invited to participate in the study.

Instrumentation

Tardieu scale

The modified Tardieu scale is an instrument used to measure tone and muscle response to static and dynamic stretching[11,12]. With this scale, one can obtain three distinct values, labeled R1, R2, and R3. The R1 value refers to the angle found at the first point of muscular endurance resulting from the hyperactive stretch reflex, which can be observed over a rapid passive range of motion, and thus provides the dynamic muscle length.

The R2 angle determines the static muscle length while performing a passive-slow stretching from resistance point R1. The R3 value corresponds to the range of joint motion calculated by subtracting R1 from R2 (R2-R1). A larger difference between R1 and R2 indicates a large dynamic muscle component, while a small difference may mean a fixed muscle contracture or range of motion (ROM)[13]. This scale relies on respectable intra-examiner and inter-examiner reliability[14,15].

Physicians Rating Scale (PRS)

The modified version of the physicians rating scale (PRS) is an observational analysis instrument[11,12] aimed at assessing the gait of children with CP. The 35 items are split into eight subgroups, with a maximum score of 22 points for each lower limb.

The scale measures the following parameters: (1) knee position at mid support; (2) initial contact of the foot; (3) midfoot foot position; (4) time of foot elevation; (5) support base; (6) midfoot hindfoot; (7) equipment used to assist walking; and (8) changes following injections of botulinum toxin type A[11]. This study used subgroups 1 to 7, obtaining a total of 20 points for each lower limb.

Gross motor function measure-88 (GMFM-88)

GMFM-88 is a standardized instrument used to quantitatively assess gross motor function[16]. This test consists of 88 items, grouped into five dimensions: dimension A (17 items) refers to bedtime and roll activities; dimension B (20 items) is based on sitting posture; dimension C (14 items) refers to crawling and kneeling activities; Dimension D (14 items) comprises the ability to stand up; and dimension E (24 items) is composed of walking, running and jumping activities. Each item score ranges from 0 through 3, where 0 indicates that the child does not start the movement; 1 indicates that the child starts the movement (<10% of the task), 2 applies to children who partially complete the item (10-99%) and 3 to the children who able to complete the task (100%)[16].

Only D and E dimensions were used in this study. GMFM-88 has been validated and translated into Portuguese[17] and is considered reliable for application in Brazilian children with CP[18].

Procedures

The children participating in this study were evaluated at the physical therapy outpatient clinic of the University Hospital of the Universidade Federal de Juiz de Fora (HU-UFJF). Each participant was evaluated a single time by two evaluators trained and experienced in the application of all instruments used, obtaining excellent intra-examiner reliability (test-retest) reliability (ICC>0.90).

Initially, the descriptive data of the children were collected: gender, age, anthropometric characteristics (height and body mass) and clinical picture. The sample was split into three groups: levels I, II and III, according to the GMFCS. The first evaluator measured the R1 and R2 values of the hamstring muscles using a universal goniometer and applied the D and E dimensions of the GMFM-88 test. Afterward, the second evaluator performed the classification according to the GMFCS functional level and the children's gait footage according to the modified PRS.

For the measurement of the hamstring length using the modified Tardieu scale with the universal goniometer, the child remained supine on a stretcher, with the hip and knee of the lower limb to be evaluated in a 90º flexion and the opposite lower limb in thigh extension, supported on the stretcher[11]. The D and E dimensions of the GMFM-88 test were performed in a comfortable environment, with adequate temperature and brightness, without noise and in the presence of the parents, so that the child was able to perform the best in each of the tested items. For the evaluation of the PRS, the children were filmed in the sagittal (five meters distance) and frontal (two meters distance) planes, by two fixed Sony® cameras, adjusted according to the participants' height. The gait was filmed uninterruptedly on a six meters long track, three times the distance predetermined by the scale. Gait analysis, according to the PRS predefined criteria, was later performed by watching the slow-motion footage[12].

Statistical analysis

At first, the Shapiro-Wilk test was used to analyze the normality of the distribution of each variable. For the description of the sample, the following variables were used: age, body mass, height, topographic distribution, lower functional limb, and functional classification according to the GMFCS. The one-way Anova test was used to test differences between the three groups in numerical variables by applying the Bonferroni *post-hoc* test to locate bivariate differences, calculating the statistical power of the test when necessary, and the Chi-square to compare the groups in the descriptive variables.

The nonparametric Spearman test was used to verify the correlation between the dependent variables. The values obtained followed the following interpretation: 0 to 0.25 – poor correlation; 0.25 to 0.50 – weak correlation; 0.50 to 0.75 – moderate correlation; and 0.75 to 1.00 – strong correlation[19]. The significance index of α=0.05 was considered using the Statistical Package for Social Sciences (SPSS®, version 15.0, 2007).

RESULTS

After contacting all potential participants from the two rehabilitation centers that collaborated with this study, 23 children and adolescents with CP agreed to participate. There was no statistically significant difference (P>0.05) between the three groups according to GMFCS regarding age, height and body mass. On the Chi-square test (P<0,05) it can be observed that the GMFCS groups were different concerning the topographic distribution. The descriptive characteristics of the sample and the GMFCS classification are shown in Table 1.

Table 1. Descriptive characteristics of the sample according to the functional group division into GMFCS I, II and III (n=23)

Descriptive variables	GMFCS		
	I (n=11)	II (n=7)	III (n=5)
	Mean (± standard deviation)		
Age (years)	12.55 (±3.93)	11.57 (±3.59)	11.40 (±5.13)
Body mass (kilograms)	40.85 (±12.60)	41.37 (±13.32)	36.89 (±19.92)
Height (meters)	1.50 (±0.17)	1.51 (±0.17)	1.32 (±0.26)
Topographic distribution			
Hemiparesis	10	3	0
Diparesis	0	4	5
Quadriparesis	1	0	0
Most functional lower limb			
Right	4	4	2
Left	7	3	3

GMFCS: gross motor function classification system; topographic distribution and more functional lower limb values represent the number of children and adolescents with cerebral palsy by onset frequency.

The one-way Anova test showed statistically significant differences between the three GMFCS groups concerning the results obtained in the D (P≤0.001) and E (P≤0.001) dimensions of the GMFM-88, in the right lower limb PRS (P=0.010) and left lower limb (P=0.030), with statistical power greater than 0.80. Significant differences were found in the Tardieu scale between groups at left R1 (P=0.009), right R2 (P=0.025), left R2 (P=0.025), and left R3 (P=0.0470) values, with statistical power less than 0.80. Table 2 presents the mean scores, the standard deviation of the variables in each group, Bonferroni *post-hoc* tests to locate the bivariate differences, and the statistical power of the tests employed.

Table 2. Mean scores and standard deviations obtained in the GMFM test on the PRS and Tardieu, scales between the GMFCS I, II and III groups, Bonferroni *post-hoc* test results and the statistical power of the tests employed.

Mean (± standard deviation)						
Variables		GMFCS	I (n=11)	II (n=7)	III (n=5)	Statistical power
GMFM	D		92.77	80.22	42.05	1.000
		I	(±3.77)	(±9.21)	(±8.62)	
		II		0.004*	<0.001*	
					<0.001*	
	E		93.05	72.82	13.86	1.000
		I	(±4.56)	(±14.69)	(±4.71)	
		II		<0.001*	<0.001*	
					<0.001*	
PRS	RLL		15.91	15.57	8.60	0.814
		I	(±4.70)	(±2.64)	(±4.62)	
		II		1.000	0.012*	
					0.029*	
	LLL		17.45	14.00	10.20	0.919
		I	(±3.50)	(±3.61)	(±3.03)	
		II		0.154	0.003*	
					0.223	
R1	R1R		146.18	134.86	126.00	0.561
		I	(±15.55)	(±17.31)	(±9.59)	
		II		0.413	0.068	
					0.989	
	R1L		149.09	127.43	123.60	0.825
		I	(±20.11)	(±10.75)	(±11.61)	
		II		0.036*	0.026*	
					0.922	
R2	R2R		157.45	147.43	141.60	0.697
		I	(±7.95)	(±14.50)	(±8.99)	
		II		0.189	0.034*	
					1.000	
	R2L		156.55	140.00	144.00	0.697
		I	(±13.06)	(±11.75)	(±8.64)	
		II		0.112	0.047*	
					1.000	
R3	R3R		11.27	12.57	15.60	0.120
		I	(±9.39)	(±7.81)	(±3.29)	
		II		1.000	0.990	
					1.000	
	R3L		7.45	15.43	16.40	0.594
		I	(±8.77)	(±5.97)	(±6.23)	
				0.122	0.119	
		II			1.000	

GMFCS: gross motor function classification system; GMFM: gross motor function measure; PRS: Modified physicians rating scale; D: GMFM D dimension; E: GMFM E dimension; RLL: right lower limb; LLL: left lower limb; R1R: first point of resistance to extension of the right leg; R2R: second point of resistance to extension of the right leg; R3R: difference between R2R and R1R; R1L: first point of resistance to left leg extension; R2L: second resistance point to left leg extension; R3L: difference between R2L and R1L. Values represented as means and standard deviation in parentheses;* P<0.05.

Spearman correlation indices were used to test relationships between muscle length (Tardieu scale), gross motor function (GMFM) and gait (PRS). Significant, positive and moderate relationships were found between the left R1 variables, the left lower limb PRS and the GMFM D and E dimensions; and left R2, the left lower limb PRS, and the GMFM D and E dimensions. Positive and weak relationships were observed between the right R1 variables and the GMFM D and E dimensions; between the left R1 variables and the right lower limb PRS; in addition to the left R2 variable and the right lower limb PRS. Finally, negative and weak relationships were found between the right R3 variables and the right lower limb PRS and between the left R3 variables and the PRS of both lower limbs, as well as the GMFM D and E dimensions. Bivariate correlations, including P-value and strength of relationships, can be seen in Table 3.

Table 3. Bivariate correlation between Tardieu scale values, and PRS and GMFM values.

	Correlation (P-value)			
	PRS		GMFM	
Tardieu scale	RLL	LLL	D	E
R1R	0.410 (0.052)	0.317 (0.140)	0.449[1](0.031)*	0.460[1](0.027)*
R1L	0.483[1](0.020)*	0.639[2](0.001)*	0.550[2](0.007)*	0.598[2](0.003)*
R2R	0.334 (0.119)	0.330 (0.124)	0.408 (0.053)	0.486[1](0.019)*
R2L	0.421[1](0.046)*	0.609[2](0.002)*	0.512[2](0.012)*	0.559[2](0.006)*
R3R	-0.424[1](0.044)*	0.062 (0.780)	0.264 (0.224)	0.379 (0.198)
R3L	-0.467[1](0.025)*	-0.483[1](0.019)*	-0.429[1](0.041)*	-0.461[1](0.027)*

PRS: Modified physicians rating scale; RLL: right lower limb; LLL: left lower limb; GMFM: gross motor function measure; D: GMFM D dimension; E: GMFM E dimension; R1R: first point of resistance to extension of the right leg; R2R: second point of resistance to extension of the right leg; R3R: difference between R2R and R1R; R1L: first point of resistance to left leg extension; R2L: second resistance point to left leg extension; R3L: difference between R2L and R1L; * P<0.05; 1: weak correlation; 2: moderate correlation.

DISCUSSION

The results of this study indicate that longer static and dynamic hamstring lengths reflect, moderately to poorly, better gait and gross motor performance in children and adolescents with CP. The strongest correlation was found between the value of the dynamic muscle length of the left lower limb (R1L) and the score obtained in gait analysis with the PRS of the same limb. In general, the values obtained for the dynamic and static muscle lengths in the left lower limb presented a better connection with PRS and GMFM values when compared to the values obtained from the right lower limb. This better correlation may be related to the fact that, in this sample, the left lower limb showed an apparent functional superiority over the right, more compromised limb in most of the evaluated participants. Thus, this reduced functional capability of the right lower limb seems to be due to the differences in dynamic muscle length (R1R) found between GMFCS levels I, II and III groups.

The moderate correlation between static and dynamic left lower limb muscle length with gait and gross motor function agrees with the results obtained by previous studies[6,20]. In contrast, due to the biarticular nature of this muscle group, there is a possibility of alteration in the proportion of shortening of the hamstrings distal and proximal portions. In this situation, the muscle portion near the hip would be elongated, while the region near the knee would be shortened, with both joints remaining flexed[21]. Thus, the Tardieu scale may not have demonstrated the real mechanical deficit of this muscle group, which generated underestimated values to these variables concerning the GMFM.

The fact that a strong correlation has not been found suggests that there is a relationship between other unevaluated components that, jointly, interfere with these children's gait and functionality. The associations between the variables measured in this study have different levels of complexity. While the GMFM gait and activities (D and E dimensions) report on the activity level, the Tardieu scale informs about the characteristics of the body structure and function component. Studies have shown that information about the activity component happens in a context and is directly influenced by environmental factors characteristics[22,23]. The literature has systematically shown that the magnitude of the association between singular structure components and body functions with more complex functions (i.e., gait, mobility, etc.) varies from weak to moderate, reinforcing the results found in this study[22,23]. As a result, it is reasonable to infer that a child with CP needs more than a relatively free active and passive knee joint amplitude to walk. The results reinforce the argument that performing a function in a particular context is a more complex phenomenon than the sum of structural components, such as muscle strength or range of motion.

Therefore, conducts exclusively aimed at stretching to improve motor performance and gait can produce unsatisfactory results. This assumption is consistent with the fact that some children benefit from stretching surgery, while others do not improve or experience functional worsening postoperatively[6]. Thus, an indication for surgery should consider physical and/or social environment, changes in body mass, muscle strength, and other factors that also contribute to functional decline.

Although physiotherapeutic conducts using only muscle stretching being insufficient[24], these results do not discredit their use but highlight their need to be addressed in association with other conducts, such as muscle strengthening techniques, to ensure that ROMs generated are effectively used during functional activities and, consequently, not lost over time. Thus, muscle stretching should not be ruled out, regardless of its effectiveness in improving ROM not being fully understood at the moment[25]. Pin, Dyke, and Chan[25] encourage a shift in focus on muscle range-of-motion stretching to make it more comprehensive in flexibility, with the use of brokering activities such as ballet, Pilates and swimming, in which children with CP actively participate, also benefiting from these activities playful and inclusive characters[25]. These approaches are corroborated by the most recent evidence which points out that CP treatment has better effects on activity and function level when performed globally, with actions such as task-oriented training and home-care programs, instead of treatments aiming only at the body structure, such as multilevel surgery and the application of botulinum toxin type A[26].

A limiting factor of this study was the reduced number of participants, especially in group III, which may have influenced the absence of differences found between levels I and II, and levels II and III of the GMFCS regarding hamstring muscle length, since the statistical power of the tests was less than 0.80. However, all children in the age group studied were contacted and all agreed to participate. Another limitation of this study is that while many participants had gone through puberty and peak growth, others did not, which may have influenced the results achieved by the study.

Future studies should jointly relate muscle length, strength, spasticity, and other structural and functional mechanisms of the muscle with motor function and gait in children with CP, to clarify the relationship between structure and body function, activity, and participation, besides conducting a clinical intervention to improve these children's functionality and gait. This knowledge is considered of paramount importance for professionals dealing with this group of patients, due to the large investment intended to gain and/or maintain muscle length in the treatment of this population.

CONCLUSION

In this study, hamstring length was weakly and moderately related to gross motor function and gait in children and adolescents with CP. Therefore, the presence of muscle shortening, in isolation, does not explain the functional deficits observed in this population's gait, which is probably caused by other components of body structure and function.

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