

RESPIRATORY FUNCTION IN CHILDREN WITH CEREBRAL PALSY

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Abstract

Purpose: to characterize the pulmonary function of children with cerebral palsy and to compare it with children with typical development. **Methods:** the study was with 20 children with typical development and 21 cerebral palsy, both sexes aged between 6-12 years (9.41 ± 1.47), with cognitive ability to perform the spirometry test. Wilcoxon / Mann Whitney test hypothesis compare the values obtained in both groups and intragroup in relation to gender and in the functional classifications of cerebral palsy. **Results:** typical development group showed higher spirometry values in males ($p < 0.05$), whereas for

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children with cerebral palsy there was no difference in relation to sex. In addition, the typical development group demonstrated better values of forced vital capacity, forced expiratory volume in the first second, and peak expiratory flow than the cerebral palsy group ($p < 0.01$). There were significant differences and better spirometric values for children with cerebral palsy less compromised in the comparisons between the functional, topographic levels, and functional gait pattern. **Conclusion:** there is a tendency towards obstructive and restrictive breathing patterns in the children with CP, observable mainly by the decrease in FVC, FEV1, and PEF compared DT. CP children with worse functionality have greater respiratory impairments.

Keywords: Spirometry; Cerebral Palsy; Respiration; Respiratory Muscles; Nervous System Diseases.

Función respiratoria en niños con parálisis cerebral

Resumen

Objetivo: caracterizar la función pulmonar de niños con parálisis cerebral y compararla con niños con desarrollo típico. **Métodos:** 20 niños con desarrollo típico y 21 con parálisis cerebral, de ambos sexos, de 6 a 12 años ($9,41 \pm 1,47$), con capacidad cognitiva para realizar la prueba de espirometría. Test de Wilcoxon/Mann Whitney para la comparación entre los valores obtenidos en los dos grupos e intragrupos en relación al sexo y clasificaciones funcionales de la parálisis cerebral. **Resultados:** El grupo de desarrollo típico mostró mayores valores de espirometría en el sexo masculino ($p < 0,05$), mientras que para los niños parálisis cerebral no hubo diferencia en relación al sexo. Además, el grupo desarrollo típico demostró mejores valores de capacidad vital forzada (FVC), volumen espiratorio forzado en un segundo (FEV1) y flujo espiratorio máximo (PEF) que el grupo parálisis cerebral ($p < 0,01$). Se encontraron diferencias significativas y mejores valores espirométricos para los niños con parálisis cerebral que estaban menos comprometidos en las comparaciones entre los niveles funcionales y topográficos y el patrón de marcha funcional. **Conclusión:** hay una tendencia hacia patrones obstructivos y restrictivos en grupo con parálisis cerebral, observados por los valores más bajos de FVC, FEV1 y PEF en comparación con desarrollo típico. Los niños con parálisis cerebral con peores niveles funcionales tienen mayor compromiso respiratorio.

Palabras llave: Espirometría; Parálisis cerebral; Respiración; músculos respiratorios; Enfermedades del Sistema Nervioso.

Introduction

Children with neuromotor disorders commonly suffer from morphofunctional changes in the respiratory system due to damage to the central nervous system (CNS) and secondary biomechanical changes. This is often seen in children with chronic non-progressive childhood

encephalopathy (CNPCE), commonly called cerebral palsy (CP) [1].

CP is one of the most common causes of physical disabilities in childhood [2], especially with motor impairment, which has a direct impact on development and performance function of the child throughout his life [3]. In this sense,

the child with CP can be classified in relation to the degree of independence of the gait, by the Gross Motor Function Classification System (GMFCS), in 5 motor levels, being level I with independent gait at level V, in which the child is totally dependent, wheelchair-bound and without head control [4].

CP can be classified clinically in relation to topographic distribution in uni or bilateral lesions or in hemiparesis/plegia, diparesis/plegia or quadriparesis/plegia, this being the most usual form [5, 6], tripareisis/plegia [5]. Regarding tonic alteration, there is the spastic, dyskinetic, ataxic, hypotonic or mixed form with spasticity [5, 7] which is characterized by increased tone, the most common [7], and present in about 80% of CP cases [8].

There are also other factors that contribute to respiratory complications, such as gastroesophageal reflux (GER), direct food aspiration, oropharyngeal incoordination, convulsive crises, reduced level of consciousness, abnormal head positions, ineffective cough for sputum [9], and lower level of engagement in physical activity [10]. All of these factors contribute directly or indirectly to the accumulation of secretion in the upper and lower airways, contributing to the occurrence of complications and recurrent respiratory infections, which are considered the most frequent causes of morbidity and mortality in patients with CP [11]. Respiratory infections are considered the major cause of hospitalization in children with CP, representing an additional burden on the public and/or supplementary health system, given the chronic condition of the impairments [12]. The life expectancy of people with CP is lower than that of people without this condition, although it has improved in this issue over time, especially when respiratory interventions are considered [11]. Even with this context, there are still no normative parameters in the literature and/or specific equipment recommended for obtaining pulmonary function tests for children with CP.

Currently, spirometry has been playing a fundamental role in the clinical evaluation and management of respiratory diseases. It is a simple, non-invasive, and useful method for assessing children with lung disorders. The interpretation of spirometric results is largely based on reference values that varied between different ethnic groups and according to the anthropometric data of each individual [13]. And although it is still little used in children with CP, due to the inherent cognitive impairment or attention deficit, the test seems to be effective also in this population [14]. However, the lack of a set of studies that confirm this application limits the recommendation of this test to the population of children with cerebral palsy. Thus, more evidence is needed so that spirometry can be used in this population, with adequate reference parameters. In this sense, the aim of this study was to characterize the pulmonary function of children with CP and to compare it with children with typical development (TD). In addition, compare lung function in relation to sex, topographic, and functional classification in CP children.

Methods

It was a cross-sectional observational study, with a convenience sample, divided into two groups: a group of children with cerebral palsy (CP), and another group with children with typical development (TD).

Children with CP were selected at two Rehabilitation Centers in the city of Curitiba, namely, *Vitória Centro de Reabilitação Neurológica*, and *Happy Kids Centro de Reabilitação Neurológica*. Typical children were selected at the *Centro de Excelência Caixa Jovem Promessa de Ginástica no Paraná*.

The protocol for this study was approved by the Research Ethics Committee of the *Centro Universitário Campos de Andrade*, under opinion No. 1,284,990 and consent by parents or guardians.

Sample

To choose the sample of both groups (CP and TD), the following inclusion factors were adopted: children of both sexes aged between 6-12 years old, who were performing regular physical exercise (TD) or physical therapy 2-3x per week (CP), with cognitive ability to perform the spirometry test.

Interviews were conducted with the heads of both groups to collect information on gestational age at birth, anthropometric data, previous respiratory diseases, medications, and frequency of physical exercise or therapy. Specifically, in children with CP, a motor assessment was performed to classify the child according to the GMFCS levels and topography.

Children who had a previous history of chronic respiratory diseases such as asthma, bronchitis, bronchiolitis, etc., or any acute and/or neurological respiratory exacerbations that interfered with the results obtained at the time of the assessment and the occurrence of associated syndromes were excluded from the study.

At the end of the study, a sample of 20 children with TD and 21 children with CP were obtained, with no sample loss among the selected volunteers.

Respiratory assessment

To check the respiratory function, the Carefusion-Microloop digital bidirectional spirometer, with a resolution of 10 milliliters (ml) per volume and 0.03 l/s per-flow and accuracy of $\pm 3\%$, was used, as recommended from ATS properly calibrated, which was attached to the Samsung brand notebook - expert line and Spirometry PC Software, version 1.03 / 2010.

The interface between the spirometer and the patient was materialized through an individual disposable mouthpiece and it was up to the evaluator to position the nose clip, the correct coupling and the individual's lip seal in the mouthpiece, preventing air leaks and mistakes in the obtained values [15, 16].

All children in the sample received the same instructions, given and carried out in a practical way by the researcher herself, which happened with simple commands, such as filling the lungs with air and then immediately releasing all the air into the straw (mouthpiece) and pulling in the air again as strong as possible. The training took place moments before the evaluation, with an interval of 5 minutes between the training and the evaluation. Through the training it was possible to identify the visual feedback to which the child was most attracted since the equipment provides us with some playful visual feedback options, such as dropping the house of the three little pigs, making soap bubbles, popping the bubble gum or even a dragon dropping fire.

The evaluation took place with the child sitting [17] independently or with the help of the therapist, when considering that the sitting position when compared to the supine position, has benefits such as increasing dynamic lung compliance and decreasing resistance to pulmonary flow, which can be attributed to the increase in functional residual capacity in that posture [18, 19].

With the researcher positioned beside the child, she was instructed to inhale the air until the total lung capacity (TLC), immediately after inspiration, the mouthpiece was placed on the tongue, between the teeth, with the closed lips and the child was instructed to perform a maximum exhalation in the first second and hold it for more than 6 seconds (volume) when it reached its residual volume it could then perform a maximum inspiration (flow), still inside the mouthpiece, as shown in Figure 1.

For each evaluation, the number of attempts contained three acceptable and two reproducible curves. The criteria for acceptance of the curves followed those proposed by the Brazilian Society of Pulmonology and Tisiology. Once the acceptance and reproducibility criteria were met, the highest values for forced vital capacity (FVC), forced expiratory volume in the first second (FEV_1), and peak expiratory flow (PEF) were noted [20].



Figure 1. Protocol for conducting the spirometric test.
Source: own elaboration

Data analysis

Both groups were analyzed for spirometric and anthropometric variables. The group of children with CP was stratified into subgroups for intra-group comparisons, according to the classification of the variables: GMFCS (subgroup I to III vs. subgroup IV and V), functional

level of gait (walkers subgroup vs. non-walkers subgroup), topographic classification (hemiplegia and diplegia subgroup vs. quadriplegia subgroup), frequency of intervention (subgroup 2x vs. subgroup 3x) and prematurity (premature subgroup vs. non-premature subgroup). To verify the distribution of the data, the Shapiro-Wilk test was used, and the Wilcoxon / Mann Whitney test hypothesis test for comparisons between CP and intergroup, adopting $p \leq 0.05$ as significance using the SPSS 25.0 software.

Results

The TD and CP groups had similar mean age, weight, sex distribution, and Tiffenau index between the groups. There was a difference between groups in stature values, and in spirometric variables FVC, $FEV_{1,}$ and PEF (Table 1).

The values with the greatest discrepancy were those of the PEF (l/min), with a difference of 1.65 l/m.

The results of the comparison of the intra-group spirometric variables TD and CP, in relation to gender are shown in Table 2. The results of the intra-group comparison for CP children, regarding the subgroups by GMFCS, functional level of gait, topographic classification, frequency of intervention, and prematurity are shown in Table 3.

Table 1. Anthropometric data and respiratory variables of children with TD and CP, presented by mean \pm standard deviation, Curitiba, Brazil, 2019

	Children with typical development (n=20)	Children with cerebral palsy (n=21)	p value
Sex (Female / Male)	14/6	13/8	0.828
Age (years)	9.45 \pm 1.14	9.38 \pm 2.29	0.761
Weight (kg)	36.20 \pm 10.44	34.51 \pm 8.50	0.725
Height (cm)	141.35 \pm 9.56	133.95 \pm 11.42	0.032*
$FEV_{1,}$ (l)	1.93 \pm 0.47	1.17 \pm 0.57	0.001**
FVC (l)	2.16 \pm 0.53	1.28 \pm 0.66	0.001**
PEF (l/min)	4.38 \pm 0.82	2.73 \pm 1.26	0.000***
Tiffenau index $FEV_{1,}/FVC$ (%)	88.50 \pm 6.56	90.76 \pm 5.98	0.340

Where: Wilcoxon / Mann Whitney test, * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$
Source: own elaboration

Table 2. Comparison of anthropometric and respiratory variables of TD and CP children in relation to sex presented by mean \pm standard deviation, Curitiba, Brazil, 2019.

	Children with typical development		p	Children with cerebral palsy		p
	Female (14)	Male (6)		Female (13)	Male (8)	
Age (years)	9.29 \pm 1.07	9.83 \pm 1.33	0.392	8.77 \pm 2.28	10.38 \pm 2.07	0.123
Weight (kg)	32.50 \pm 9.01	44.85 \pm 8.70	0.021*	34.10 \pm 8.45	35.19 \pm 9.13	0.561
Height (cm)	139.29 \pm 9.12	146.17 \pm 9.58	0.173	131.62 \pm 9.54	137.75 \pm 13.78	0.216
FEV1 (l)	1.74 \pm 0.28	2.39 \pm 0.54	0.006**	1.13 \pm 0.43	1.23 \pm 0.79	0.971
FVC (l)	1.97 \pm 0.32	2.63 \pm 0.66	0.032*	1.22 \pm 0.52	1.40 \pm 0.89	0.971
PEF (l/min)	4.07 \pm 0.68	5.10 \pm 0.68	0.009**	2.66 \pm 1.24	2.86 \pm 1.38	0.800
Tiffenau index FEV1/ FVC (%)	87.21 \pm 7.33	91.50 \pm 2.88	0.173	92.69 \pm 4.66	87.63 \pm 6.84	0.136

Where: Wilcoxon / Mann Whitney test, *p<0.05; **p<0.01
Source: own elaboration

In Table 3 by the intragroup comparison CP, it is possible to notice that when we classify the children by the level of functionality through the GMFCS (I, II and III vs IV and V) and by the functional level of gait, being walkers versus non-walkers, the different spirometric variables were FEV1 and FVC, with lower values for the groups of non-ambulators and GMFCS IV and V.

Although the quadriplegia subgroup was older than the hemiplegia/diplegia subgroup, the FEV1 and FVC variables showed higher values in the hemiplegia/diplegia subgroup. There was no difference in the anthropometric and spirometric values of CP children in relation to the frequency of therapy. Regarding prematurity, although it represents the majority (76%) of CP children, no difference was identified in relation to CP children at term for the anthropometric and spirometric variables analyzed.

Discussion

This study aimed to characterize the pulmonary function of children with CP and compare it with children with typical development (TD). In addition, compare lung function in relation to sex, topographic, and functional classification in CP children. The main finds were TD group showed higher spirometry values in males,

whereas for children with CP, there was no difference in relation to sex. In addition, the TD group demonstrated better values of forced vital capacity (FVC), forced expiratory volume in the first second (FEV1), and peak expiratory flow (PEF) than the CP group.

It is known that the evaluation of respiratory functions in children with respiratory impairment is extremely valuable. However, the absence of reference values for specific populations, makes the identification of data, monitoring the evolution of the intervention, and prognosis more difficult. For this reason, some researchers choose to also evaluate individuals with typical development so that there is a comparison between the control and study groups.

For the comparative effects of the spirometric values between TD and CP, an attempt was made to match the age and sex variables, the same being not possible for the height variable, which was significantly lower for children with CP. This anthropometric difference in relation to children with CP is mentioned in other studies [21, 22] and it is a variable that although it influences lung volumes[23] it was not possible to be controlled in the present study, given the expected growth difference for children with CP, especially from GMFCS II to V [21] when compared to children of the same age group with TD.

Table 3. Intra-group comparison for CP children, regarding the subgroups by GMFCS, functional level of gait, topographic classification, frequency of intervention, and prematurity. Data presented by mean \pm standard deviation, Curitiba, Brazil, 2019.

Children with cerebral palsy	N	Age (years)	Weight (kg)	Height (cm)	FEV1 (l)	FVC (l)	PEF (l/min)	Tiffenau index FEV1/ FVC (%)
Functional level of gait								
walkers	13	8.85 \pm 2.34	34.07 \pm 9.04	133.00 \pm 9.55	1.48 \pm 0.49	1.63 \pm 0.62	2.76 \pm 1.14	91.07 \pm 4.79
Non-walkers	8	10.25 \pm 2.05	35.24 \pm 8.09	135.50 \pm 14.56	0.66 \pm 0.19	0.72 \pm 0.15	2.68 \pm 1.51	90.25 \pm 7.91
p		0.186	0.717	0.689	0.000***	0.000***	0.192	0.689
GMFCS								
I, II e III	11	8.45 \pm 2.34	34.61 \pm 9.55	132.09 \pm 9.67	1.44 \pm 0.53	1.58 \pm 0.66	2.74 \pm 1.24	91.73 \pm 4.90
IV e V	10	10.40 \pm 1.84	34.41 \pm 7.70	136.00 \pm 13.30	0.87 \pm 0.49	0.95 \pm 0.53	2.72 \pm 1.35	89.70 \pm 7.10
p		0.074	1.000	0.376	0.007**	0.005**	0.398	0.377
Topographic classification								
Hemiplegia/Diplegia	15	8.67 \pm 2.29	33.19 \pm 8.85	131.20 \pm 10.43	1.37 \pm 0.54	1.51 \pm 0.66	2.60 \pm 1.14	91.07 \pm 4.43
Quadriplegia	6	11.17 \pm 0.98	37.82 \pm 7.18	140.83 \pm 11.74	0.66 \pm 0.22	0.72 \pm 0.18	3.05 \pm 1.59	90.00 \pm 9.34
p		0.024*	0.185	0.100	0.005**	0.003**	0.938	0.845
Prematurity								
yes	16	9.56 \pm 2.42	33.85 \pm 7.62	134.50 \pm 11.40	1.12 \pm 0.57	1.22 \pm 0.65	2.75 \pm 1.38	90.81 \pm 6.52
no	5	8.80 \pm 1.92	36.64 \pm 11.67	132.20 \pm 12.66	1.31 \pm 0.59	1.47 \pm 0.73	2.65 \pm 0.88	90.60 \pm 4.39
p		0.379	0.741	1.000	0.509	0.509	0.804	0.836
Frequency of intervention								
2x per week	15	9.33 \pm 2.16	35.99 \pm 8.25	135.47 \pm 10.29	1.14 \pm 0.52	1.25 \pm 0.63	2.70 \pm 1.22	90.53 \pm 6.52
3x per week	6	9.50 \pm 2.81	30.83 \pm 8.70	130.17 \pm 14.18	1.24 \pm 0.75	1.36 \pm 0.81	2.83 \pm 1.48	91.33 \pm 4.84
p		0.813	0.185	0.611	0.969	0.969	1.000	0.815

Where: Wilcoxon / Mann Whitney test, *p<0.05; **p<0.01; *p<0.001

Source: own elaboration

In the present study, the significantly higher values of FEV1, FVC and PEF for boys with TD corroborate what is pointed out by Rosa *et al.* [24] and contrasts the studies by França *et al.* [25] and Park *et al.* [26]. Although the influence of males on greater lung capacities is known, it is interesting to note, however, that this difference between genders was not identified for children with CP, as already mentioned by other studies [14, 27].

It was identified that children with CP had lower values of FEV1, FVC and PEF, when compared to those with TD. The results indicating a restrictive pulmonary change in CP resulting from limited movements and not due to dysfunction of the lung parenchyma, as already mentioned by Lee, Cha e Kim [28]. Thus, the decline in the participation of physical activities in children with CP can lead to the development of abnormalities and peripheral muscle dysfunctions due to muscle weakness, increased muscle fatigue and reduced oxidative capacity, [27] justifying the lower values of FEV1, FVC and PEF.

Although brain damage itself is not a direct cause that produces CP breathing problems, children showed lower values, not only in general respiratory function but also in respiratory muscle strength, which is closely related to self-care activities, social adaptability in daily life when compared to typical children, [14] justifying the analysis of the respiratory function of CP children considering their functional levels.

In the present study, both in the grouping of CP children by the functional levels of the GMFCS (I to III vs. IV to V) and by the ability to walk independently (Walkers vs. Non-Walkers) and by the topographic classification (hemiplegia and diplegia vs. quadriplegia) it is observed that less compromised children had significantly higher FVC and FEV1 values. These findings corroborate with previous studies [27, 29-32].

It is known that the function of walking / walking as associated with the preservation or de-

velopment of better respiratory functions in children with CP. Kwon e Kim [29] and Know and Lee [27] evaluated children with CP between GMFCS levels I and III, with hemiplegia and diplegia, and found that children who are unable to walk independently (GMFCS III) would have a low respiratory function and muscle strength due to a decline in lung capacity accompanied by limited functional movement. In addition, there was no significant difference between children categorized at GMFCS levels I and II in terms of respiratory function and muscle strength, due to the fact that their capacity for physical activity is similar. Muammer *et al.* [31] compared children with CP in GMFCS I and II and GMFCS III to V and identified a relationship between the poorest level of GMFCS and lower lung capacity / respiratory muscle weakness, represented by the FVC value. The children evaluated by Muammer *et al.* [31] with GMFCS III had a lower motor function than respiratory functions compared to children with a GMFCS I and II.

In the same direction Pereira *et al.* [32] identified a strong negative correlation between the GMFCS motor classification and the inspiratory pressure of these patients, that is, the higher the GMFCS, the lower the lung function in individuals with brain injuries.

Regarding the functionality of children with CP, it is known that spasticity is present in the vast majority of cases, which would justify the reduction in FVC evidenced by the influence of high muscle tone on the immature breathing pattern, with reduced diaphragmatic action, lack of mobility of the costal grid and spine hindering pulmonary expansion in the anteroposterior and lateral-lateral directions [33].

Another important factor is that the relationships between FEV1, FVC, and FEV1 / FVC are useful parameters for assessing restrictive lung disease, on the other hand, PEF is a measure of the flow produced during exhalation with maximum effort and is useful for assessing lung disease. obstructive. However, among

most of the studies included here, only FEV1 and FVC were listed, with FEV1 / FVC and PEF data omitted. In addition, previous studies have shown that lung function in children with CP has characteristics of obstructive and restrictive lung disease.[34] These findings can lead to the development of generalized microatelectasis and decreased pulmonary distensibility and high mortality rates associated with CP.

It is notable the difficulty in evaluating children with CP due to cognitive impairment [26] and changes in muscle, respiratory, and biomechanical function, make it difficult to perform the spirometry test properly [35]. Clarifying such specific spirometric parameters makes it possible to clarify deficits and respiratory changes, which could guide the conduct and treatment of this population.

The number of the sample can be mentioned as limitations of the present study, due to the need to understand the test. As suggestions for future studies, it is indicated to analyze the number of attempts to perform the test until the three acceptable and reproducible curves are reached, the possibility of a correlation between the increase in heart and respiratory rate in individuals with CP [36].

The strengths of this study were the control in comparing the spirometric variables by sex and age between children with CP and with TD, allowing a reliable comparison of the test variables. The success rate in the assessments was 100%, which can be justified by the use of a computational spirometer that allowed us to use resources such as visual feedback. Another important factor was the comprehensiveness of all levels of the GMFCS (I to V) and the inclusion of quadriplegic patients, a pattern that is rarely found in other studies due to the severe cognitive impairment often associated in the most severe CP cases.

Conclusion

The comparison of respiratory variables between children with cerebral palsy and typical children allowed us to conclude that there is a tendency towards obstructive and restrictive breathing patterns in the group of children with CP, observable mainly by the decrease in FVC, FEV1, and PEF.

It was found that the higher the child's functional level (GMFCS) and the lower the chances of independent walking and functionality, the greater the individual's respiratory impairment. Regarding the topographic classification, it is concluded that the quadriplegics had lower values of FVC and FEV1. In the group of children CP, there were no differences in respiratory variables between the subgroups of the frequency of weekly therapy (2 or 3 times). In the group of typical children, the influence of sex was observed on the spirometric values obtained, which was not observed in children with CP.

This study characterized the spirometric patterns of children with CP. Studies with larger samples are recommended so that normality values can be defined for children with CP, considering their classifications according to the various dimensions analyzed (GMFCS, functional level of gait, topographic classification, frequency of intervention, and prematurity).

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Conflict of Interest

The authors declare that they have no conflict of interest.

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