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Diaphragmatic excursion and pulmonary function in children with cerebral palsy – comparative study

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Tintu Thomas[1], Amitesh Narayan[1], Gopala Krishna Alaparthi[2], Shreekanth D. Karnad[1]

[1] Department of Physiotherapy, Kasturba Medical College, Mangalore, Manipal Academy of Higher Education, Manipal, India

[2] Department of Health Professions, Faculty Health and Education, Manchester Metropolitan University, Manchester, UK

Abstract

Introduction. Cerebral palsy (CP) is a common paediatric disorder occurring in about 2 to 2.5 per 1000 live births. Studies have reported that children with cerebral palsy have a high incidence of respiratory problems, with limited evidence regarding diaphragmatic impairment. Thus, the study aims to analyse and compare the diaphragmatic excursion and pulmonary function in children with CP compared to typically developing children.

Methods. The sample size was calculated as 38, divided into groups of typically developed children (19) and spastic CP children (16). An experienced radiologist assessed the diaphragmatic excursion. The respiratory parameters were assessed using a portable Pulmonary Function Test machine.

Results. A positive significance was seen in the pulmonary function test values ($p = 0.00$) and diaphragmatic excursion ($p = 0.00$), showing better values among the typically developed children than the CP group.

Conclusions. The study concludes that children with CP have a reduction in diaphragm excursion and poorer pulmonary values than typically developing children, making them prone to respiratory diseases, thus requiring early intervention.

Key words: cerebral palsy, diaphragm excursion, respiratory function tests

Introduction

Cerebral palsy (CP) is a common paediatric disorder occurring in about 2 to 2.5 per 1000 live births [1]. Many studies have reported that children with cerebral palsy have a high incidence of respiratory problems, i.e., recurrent pneumonia, atelectasis, bronchiectasis, and restrictive lung disease, which may increase morbidity and mortality [2–4]. For breathing, the children with CP rely on abdominal muscles more than chest wall muscles, due to which they develop poor breathing pattern coordination. This may lead to restriction in chest wall movements, impacting the chest muscles' strength and affecting the capacity to take large breaths [5, 6]. It is worth noting that children with CP fail to adjust their lung volume and depend on the intercostal and oblique muscles as a compensatory strategy against poor respiratory biomechanics [7].

Respiratory functions are assessed by Pulmonary Function Tests (FVC, FEV1, FEV1/FVC & PEF) [8]. Diaphragm and abdominal muscles are essential for inspiration, and their weakness causes respiratory morbidity and recurrent lower respiratory tract infections in patients with neurological diseases, especially in children with CP [3, 6]. Weakness of the abdominal muscles influences the stability of the central tendon of the diaphragm, which may restrict lateral chest wall expansion [9]. Diaphragmatic function can be assessed by ultrasonogram by analysing diaphragmatic motion, i.e., the descent and ascent of the diaphragm during inspiration and expiration [10]. Among many impairments observed in CP, respiratory system impairments are less explored. It is known that children with CP exhibit poor performance in spirometry evaluation, which is related to their innate poor postural alignment, causing them to develop malalignment such as scoliosis, thereby leading to poor chest mobility [11].

Though there is considerable evidence of a reduction in pulmonary function and morphological and mechanical changes to the chest wall kinematics in children with CP, there is a lack of retrievable studies on diaphragmatic function abnormalities in children with CP. Therefore, this study aims to analyse the diaphragmatic excursion and pulmonary function in children diagnosed with cerebral palsy and to compare these values obtained among the CP children with age and sex-matched typically developed children. The obtained data further strengthens the evidence regarding the need for early intervention among children diagnosed with cerebral palsy.

Subjects and methods

This was a cross-sectional analytical study conducted at Kasturba Medical College Hospital, Attavara, Mangalore, following approval from the Institutional Ethics Committee. The duration of this study was one year (February 2014 – February 2015). The study population was children diagnosed with cerebral palsy by a paediatrician or a paediatric neurologist and referred to the neurosensory development clinic KMC Mangalore for evaluation and treatment. The sample size was calculated using the following formula:

$$N = \frac{2(Z_\alpha + Z_\beta)^2\,\sigma^2}{C^2}$$

where, $\alpha = 1.96$, $\beta = 1.28$, $C = 0.719$, $\sigma^2 = 9$, power of the study: 90%, Confidence Interval (CI): 95%.

Using the purposive sampling method, the sample size of (n) 38 was obtained and further divided into two groups – one with children diagnosed with CP and the other with typically developing children (Figure 1). Children in the age group of 6 to 15 years of either sex and children diagnosed with

Correspondence address: Amitesh Narayan, Department of Physiotherapy, Kasturba Medical College, Mangalore, Manipal Academy of Higher Education, Manipal, Karnataka, India, e-mail: amitesh.narayan@manipal.edu; https://orcid.org/0000-0001-7587-8414

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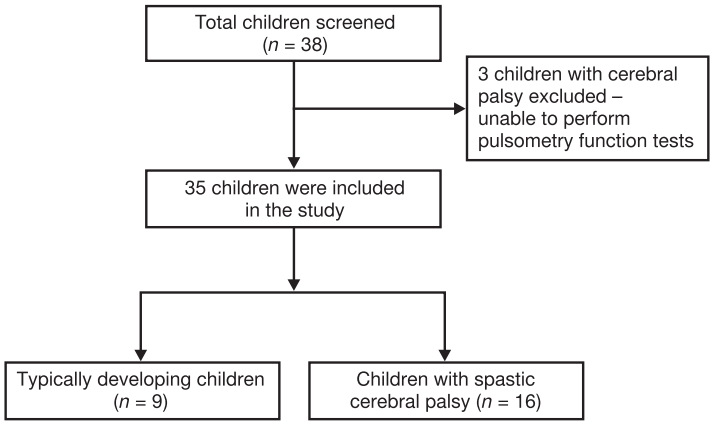

Figure 1. Flow chart showing screening and recruitment of children for the study

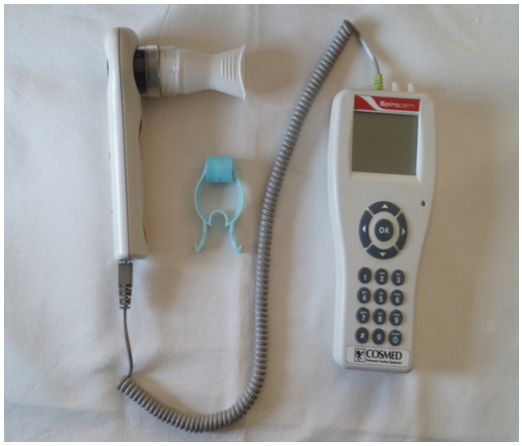

Figure 2. Pulmonary function test machine

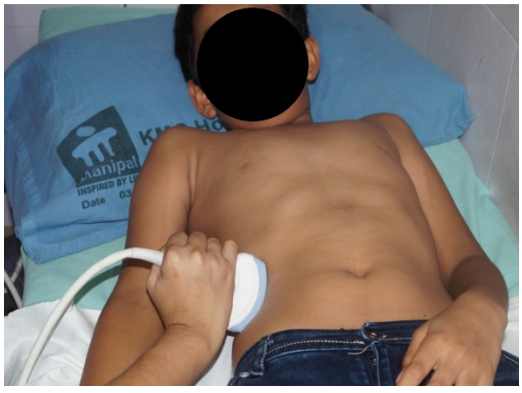

Figure 3. Diaphragmatic excursion testing procedure

spastic diplegic cerebral palsy by the paediatrician were included. Gross Motor Function Classification System Level I & II with Modified MMSE score of > 24, were included. Age and sex-matched typically developing children recruited from the community.

At screening, children suffering from upper or lower respiratory tract infections/inflammations, children diagnosed to have any pulmonary, cardiovascular, metabolic, or other systemic conditions that can alter the diaphragmatic function or interfere with the performance of test procedures, highly irritable children, who may not be able to co-operate with the test procedure, who were on anti-epileptics and anti-spastic drugs and children with congenital diaphragmatic hernia were excluded.

Study procedure

The study protocol was presented to the scientific committee and the institutional ethics committee. First, children diagnosed with cerebral palsy were referred to the neurosensory development clinic and screened for the inclusion criteria. Those who met the inclusion criteria were recruited for the study after obtaining informed assent from the parent/guardian. Age- and sex-matched typically developing children were recruited from the community as controls by notifications through newspaper advertisements, newsletters, or by direct contact, calling for voluntary participation.

Data collection procedure

Pulmonary Function Test Procedure: The measurement was carried out by a qualified physiotherapist trained in employing the PFT device. All children were measured in a sitting position on a chair with the head and trunk maintained erect, and the hip and knee joints flexed at 90°. In the test, each child was instructed to inhale a breath and then blow the entire volume through the mouthpiece of a spirometer as deeply and rapidly as possible. Before the actual measurement, the procedure was explained and demonstrated to each subject until successful performance was achieved. All respiratory function measurements were performed at least three times, and the best of the three trials was considered. Respiratory function tests, including forced vital capacity (FVC), forced expiratory volume at one second (FEV1), ratio of forced expiratory volume at one second to forced vital capacity (FEV1)/(FVC), and peak expiratory flow (PEF) measurement were taken by the same examiner using a spirometer (Spiropalm®) throughout the entire experiment in both the typically developing children and children with cerebral palsy (Figure 2) [12].

Procedure for assessing diaphragmatic excursion

An experienced radiologist assessed the diaphragmatic excursion using a *GE Voluson 730E* ultrasound machine in B-Mode. The probe was placed between the midclavicular and anterior axillary lines in the sub-costal area and directed medially, cranially, and dorsally so that the ultrasound beams reached the posterior third of the right hemidiaphragm perpendicularly. Diaphragm excursion during inspiration was measured as the maximum caudal descent of the midpoint of the posterior third of the diaphragm between end-expiration and end-inspiration. The average of 3 consecutive readings were obtained by the same investigator (Figure 3) [9].

Statistical analysis

The statistical data were coded and entered into the Statistical Package for the Social Sciences (SPSS®) version 16. The results were expressed as a summary measure (mean and standard deviation) and presented in an appropriate table. The diaphragmatic excursion and pulmonary function were compared using an unpaired *t*-test.

Ethical approval

The research related to human use has complied with all the relevant national regulations and institutional policies, has followed the tenets of the Declaration of Helsinki, and has been approved by the institutional scientific committee, and institutional ethics committee, KMC Mangalore, Manipal Academy of Higher Education (approval No.: IEC KMC MLR 11-13/229.

Informed consent

A duly signed written informed consent form was obtained from the parents as the study included children with an average age below 10.

Results

Thirty-eight children (both typically developing and children with cerebral palsy) were screened, and 35 children (TD-19, CP-16) were selected for the study. The mean age of the typically developing (TD) children was (9.78 ± 2.56), and the children with CP was (9.75 ± 3.01). Based on the Gross Motor Function Classification System (GMFCS) for CP, 68.7% (*n* = 11) were at level I and 31.25% (*n* = 5) at level II (Table 1 and Figure 1).

Table 1. Demographic characteristics of children represented as percentage, mean and standard deviation

Variables		TD (mean ± *SD*)	CP (mean ± *SD*)
Sex	Male	13 (37.1%)	11 (31.4%)
	Female	6 (17.1%)	5 (14.3%)
Age (years)		9.7 ± 2.5	9.7 ± 3
Height (cm)		134.10 ± 9.2	128.25 ± 10.2
Weight (kg)		30.37 ± 5.7	27.62 ± 7.6
GMFCS	I	–	11 (68.7%)
	II	–	5 (31.2%)

TD – typically developed, CP – cerebral palsy,
GMFCS – Gross Motor Function Classification System

The pulmonary function tests analysis suggests that typically developing children demonstrated better pulmonary functions capacities, i.e., FVC (1.83 ± 0.46), FEV1 (1.68 ± 0.44), PEF (2.89 ± 0.96) compared to children with CP, i.e., FVC (1.14 ± 0.32), FEV1 (1.05 ± 0.38) and PEF (1.62 ± 0.57). There is statistical significance in all variables: FVC, FV1, PEF ($p \leq 0.00$) except FEV1/FVC ($p \leq 0.09$) (Table 2). The result indicates that diaphragmatic excursion was found to be more in typically developing children 4.142 ± 0.656 (R) and 4.168 ± 0.0619 (L) compared to children with CP 3.075 ± 0.407(R) and 3.250 ± 0.495 (L) ($p \leq 0.00$) (Table 2).

Table 2. Descriptive analysis of Pulmonary Function Tests and diaphragmatic excursion in typically developing children and children with cerebral palsy

Variables	TD (*n* = 19) (mean ± *SD*)	CP (*n* = 16) (mean ± *SD*)	Mean diff.	*p*-value
Diaphragmatic excursion				
DE (R)	4.14 ± 0.65	3.07 ± 0.40	1.06	0.00*
DE (L)	4.16 ± 0.61	3.25 ± 0.49	0.91	0.00*
Pulmonary Function Tests				
FVC	1.83 ± 0.46	1.14 ± 0.32	0.69	0.00*
FEV1	1.68 ± 0.44	1.05 ± 0.38	0.62	0.00*
FEV1/FVC	91.15 ± 3.35	86.81 ± 9.28	4.34	0.09
PEF	2.89 ± 0.96	1.62 ± 0.57	1.26	0.00*

* $p < 0.05$, significant, DE – diaphragmatic excursion,
FVC – forced vital capacity, FEV1 – forced expiratory volume in 1 second, PEF – peak expiratory flow, TD – typically developed, CP – cerebral palsy

Discussion

This study compares diaphragmatic excursion and pulmonary function through pulmonary function tests (FVC, FEV1, FEV1/FVC & PEF) between children with spastic cerebral palsy and typically developing children within the mean age of (9.75 ± 3.010) and (9.79 ± 2.566), respectively. Results of the study indicate a reduced excursion of the hemidiaphragm in children with CP and that they performed significantly poorer in all pulmonary function outcomes.

There is an abundance of existing literature emphasising the susceptibility of children with CP to developing respiratory ailments. Biomechanical alterations to chest wall mobility and physiological variations in respiratory functions have been attributed to the elevated prevalence of reduced excursion of the hemidiaphragm [2, 3, 5, 13]. In the current study, the physiological outcomes of the pulmonary function and kinematics of the diaphragm in a volitional effort have been studied concurrently. It has been established that, as speculated, reduced diaphragmatic mobility coexists with poor pulmonary function in children with CP.

A child with spastic cerebral palsy develops changes in the ribcage and thoracic spine structure due to a lack of muscular action. The ribcage either adapts a barrel shape with excessive and sustained superficial muscle activity or is flattened with distal rib flaring because many muscle groups lack activity, which may cause shallow respiration and facilitates an immobile ribcage during inhalation and exhalation. In both situations, the ribcage is elevated, and the thoracic spine develops a more kyphotic posture, limiting the anterior part of chest expansion [13].

Children with spastic cerebral palsy have weak anti-gravitational potential in an upright position, which, together with the ribs' insufficient concave position, reduces the chest mechanics in the sagittal and lateral planes. Insufficient tonus of the internal intercostal muscles, oblique abdominal muscles and the diaphragm fibres (sternal part) lower the sternum at inspiration. This, in turn, together with insufficient chest decompression, flattens the breathing, increases the breathing rate, reduces the ventilatory capacity, and leads to abnormal chest wall development [14].

In our study, variables of pulmonary function (FVC, FEV1 & PEF) were found to be reduced in children with spastic cerebral palsy than typically developing children, and these differences were significant. Our findings are comparable to a study done on 42 children (14 spastic diplegic, 14 hemiplegic CP, and 14 normal children), where it was found that children with CP had significantly weaker respiratory function as compared to normally developing children, and among the children with CP, the spastic diplegia children showed more profound impairment compared to those with hemiplegia. Pulmonary function was assessed using the variables of FVC, FEV1, FEV1/FVC and PEF. The observed impairment in pulmonary function was attributed to reduced exercise tolerance stemming from limited physical activity and immobility of the chest wall imposed by reduced neuro-muscular control and musculoskeletal impairments in CP.

Poor postural control, spasticity, impaired activation, and weakness of the muscles of respiration have been identified as predisposing factors to impairment of respiratory function in children with CP [2, 5, 13]. In our study, three subjects who fulfilled the inclusion criteria and were recruited for testing could not be tested for the variable of FEV1, as they failed to elicit a valid response. It was found that they could not sustain an ample enough expiratory effort for the prerequisite 6 seconds, and their expiratory effort stopped well short of the

required time. It is well understood that sudden or sustained volitional activity, performed especially with effort, can lead to unwanted tonic fluctuations and associated reactions in children with CP [15]. The invalid effort of the three children while testing for FEV1 could be attributed to these unwanted reflexes and reactive motor activities.

Limitations

One limitation of the study is difficulty in generalisation due to the small sample size. Also, a few parameters, such as TV and TLC, were not considered in the study. Future studies need to establish the associations and relationship between coexisting physiological and morphological variables of respiratory function in children with CP. The variations in pulmonary function and diaphragm mechanics across the various topographical classifications of CP, the relationship between functional impairment as measured by GMFCS, and the changes in respiratory dynamics need deeper analysis and understanding.

Conclusions

Therapeutic efforts in managing respiratory disorders in children with CP should not be limited only to maintaining bronchial hygiene and airway clearance. A deeper understanding of the predisposing factors to respiratory ailments and their extrapulmonary causes, such as maladaptive postural /trunk control and global changes to neuro-motor control, are required. Hence, interventions designed to address these multitudes of issues will lead to superior and sustainable rehabilitative outcomes.

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Disclosure statement

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Conflict of interest

The authors state no conflict of interest.

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