

Comparison of Pulmonary Function in Children with Cerebral Palsy and Normally Developed Children

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ABSTRACT

Introduction: Cerebral Palsy is one of the most common neurological disorders in children. It is characterised by varying degrees of disability affecting multiple domains. Respiratory complications, especially in severely disabled cerebral palsy patients are a major cause of morbidity and mortality. However, respiratory deficiencies in children with cerebral palsy is often overlooked and rarely assessed in the clinical setting.

Aim: To assess the pulmonary function in children with cerebral palsy and compare the same with normally developed children.

Materials and Methods: Thirty seven patients diagnosed with spastic cerebral palsy in the age range of 5-18 years and 37 age matched normally developed controls were enrolled. Assessment of pulmonary function in terms of Forced Vital

Capacity (FVC), Forced Expiratory Volume in 1st second, FEV₁/FVC, Peak Expiratory Flow (PEF), Forced expiratory flow over 25-75% of FVC (FEF25-75), Inspiratory Reserve Volume (IRV), Expiratory Reserve Volume (ERV), Tidal Volume (TV) and Vital Capacity (VC) was done for each subject. Statistical analysis was done using SPSS version 21.0 and a p-value of <0.05 was considered as statistically significant.

Results: Most of the children with cerebral palsy were found to have lower FVC, FEV₁, PEF25-75, IRV, ERV, TV and VC in comparison to normally developed children indicative of predominantly restrictive pulmonary pathology.

Conclusion: Routine pulmonary function testing in cerebral palsy children may reveal compromised pulmonary function. This will enable early therapeutic intervention for management of respiratory dysfunction in these children.

Keywords: Neurological disorder, Paediatrics, Respiratory dysfunction

INTRODUCTION

Cerebral Palsy is one of the most common neurological disorders encountered in paediatric population affecting 2-3 per 1000 live births [1,2]. It is due to non-progressive disturbances in the developing foetal or infant brain which results in a set of permanent developmental disorders of movement and posture. Although motor disorders are predominant, they are often accompanied by disturbances of sensation, perception, cognition, communication and behaviour. Epilepsy and secondary musculoskeletal problems are also frequently encountered leading to varying degree of disability [3,4].

Owing to this wide spectrum of abnormalities, cerebral palsy is rendered one of the most common causes of childhood disability. Restricted mobility and the afore-mentioned abnormalities in sensation, perception, cognition, communication, behaviour and other neurological conditions like seizures make cerebral palsy patients vulnerable to respiratory complications. Respiratory compromise may further be augmented by associated upper airway obstruction due to inadequate nasopharyngeal motor function, obstructive sleep apnoea syndrome or poor nutrition [5-7]. Respiratory complications, especially in severely disabled cerebral palsy patients are reported to be a major cause of morbidity and mortality and are a matter of great concern [8-14].

However, despite presence of evidence, not much attention is directed towards evaluation of respiratory deficiencies in children with cerebral palsy. This study has been designed to address this issue with an aim to compare pulmonary functions in children with cerebral palsy and normally developed children.

MATERIALS AND METHODS

This cross-sectional comparative study was conducted in Department of Physical Medicine and Rehabilitation of a tertiary care hospital during the period from October 2013 to February 2015.

Ethical approval was taken before initiating the study (S.No. IEC/VMMC/SJH/Thesis/Oct-13/152).

Thirty seven patients with spastic cerebral palsy diagnosed on the basis of history and clinical findings, within the age range of 5-18 years of either gender (Group 1), were included in the study. Thirty seven age matched normally developed controls (Group 2) of both genders were enrolled from the relatives of the children with cerebral palsy and children attending Physical Medicine and Rehabilitation OPD for other problems. Written informed consent was taken from parents/guardians of all participants. The sample size was calculated based on a study by Kwon YH et al., comparing respiratory functions of CP children with normally developed children [15].

Children with mental retardation, apparent chest deformity and Gross Motor Function Classification System (GMFCS) 4, 5 were excluded from study. Subjects were thoroughly examined clinically after obtaining detailed medical history from participants/parents using the study proforma.

Demographic characteristics recorded were age, sex, residence, and socio-economic status. Assessment of pulmonary function was done by using computerised Medisoft spiro air spirometer. Clear instruction regarding the procedure was given to all the participants. They were first accustomed to the apparatus and the procedure, following which a minimum of three readings were taken for each parameter. The assessment parameters recorded were FVC, FEV₁, FEV₁/FVC, PEF, FEF25-75, IRV, ERV, TV and VC.

STATISTICAL ANALYSIS

Data collected were entered into Microsoft Excel spread sheet and analysed using SPSS version 21.0 software. Categorical data was presented in number and percentage and continuous variables were presented as mean±SD. Normality of data was tested by Kolmogorov-Smirnov test. Quantitative variables were compared using Unpaired t-test/Mann-Whitney test and qualitative variables

were compared using Chi-square test and Fisher's-exact test. A p-value <0.05 was considered as statistically significant.

RESULTS

Seventy-four children, 37 with spastic cerebral palsy (Group 1) and 37 normally developed children (Group 2) were enrolled in study. The two groups were comparable with respect to demographic characteristics [Table/Fig-1]. In group 1, 18 (49%) children were spastic hemiplegic and 19 (51%) were spastic diplegic. Among the spastic hemiplegic children, 12 were right and 6 were left sided hemiplegic.

Baseline characteristics	Group 1 (n=37)	Group 2 (n=37)	p-value
Mean age in years	11.24±3.43	12.16±2.72	0.234
Mean height in centimetre	139±17.31	143.11±13.14	0.207
Mean weight in kilogram	32.95±10.95	36.16±10.78	0.355
Gender distribution	Male=27 Female=10	Male=29 Female=8	0.588

[Table/Fig-1]: Demographic characteristics of the children.

[Table/Fig-2,3] shows the observed values of different parameters used for assessment of pulmonary function. Significant difference between the two groups was found in FVC. All the children in group 1 had decreased FVC while in group 2 only one child had so. Two children in group 1 were found to have normal FEV₁ while in group 2, seven children had decreased FEV₁. This difference in FEV₁ between the two groups is statistically significant. While comparing the ratio of FEV₁ and FVC significant difference was observed between the two groups. Decreased PEF was found in 24 group 1 children and 9 in group 2 children had normal, while, FEF over middle of FVC was found to be decreased in 28 children in group 1 and 22 in group 2. The mean IRV and ERV of group 1 child was found to be significantly lower than that of group of 2 children. Tidal volume of group 1 children ranged from 0.15 to 0.78 litres while in group 2 children the same was from 0.35 to 0.81. While comparing the mean tidal volumes, the difference between the two groups was found to be significant. Only two children in group 1 had normal vital capacity. 35 children in group 1 were found to have decreased vital capacity, while only three children in group 2 showed so.

Parameters	As compared to predictive value	Group 1 (n=37)	Group 2 (n=37)	p-value
FVC	Decreased	37	1	<0.001
	Normal	0	34	
	Increased	0	2	
FEV ₁	Decreased	35	7	<0.001
	Normal	2	28	
	Increased	0	2	
FVC/FEV ₁	Normal	11	27	<0.001
	Increased	26	10	
PEF	Decreased	24	2	<0.001
	Normal	12	9	
	Increased	1	20	
FEF 25-75	Decreased	28	22	0.007
	Normal	6	9	
	Increased	3	6	
VC	Decreased	35	3	<0.001
	Normal	2	30	
	Increased	0	4	

[Table/Fig-2]: The flow rates and vital capacity of the children.

Parameters	Group 1 (n=37)	Group 2 (n=37)	p-value
Mean IRV (litre)	0.61±0.4	0.96±0.44	<0.0001
Mean ERV (litre)	0.53±0.31	0.94±0.53	<0.0001
Mean TV (litre)	0.46±0.15	0.53±0.1	0.037

[Table/Fig-3]: Different lung volumes of the children.

Thus, observed results indicate significantly compromised respiratory function in spastic CP children as compared to children with normal development.

The observed pulmonary function of the children in the group 1 were further analysed for type of dysfunction. Out of 37 children included in the group 1, 28 (75.68%) children were found to have restrictive type of lung dysfunction, 8 (21.7%) showed obstructive type of lung dysfunction and 1 (2.7%) child showed non-conclusive parameters.

DISCUSSION

Cerebral palsy is a common cause of physical disability in children. Respiratory infection and diminished pulmonary function rank as one of the chief causes of morbidity and mortality [16]. In this study we assessed and compared pulmonary function among children with spastic cerebral palsy with normally developed children. Results of the study revealed compromised pulmonary function in the cerebral palsy group. The difference in FVC, FEV₁ and FEV₁/FVC ratio between the patient group and control was statistically significant. The peak and mid expiratory flow rates, vital capacity, tidal volume, expiratory and inspiratory reserve volumes were also found to be decreased in cerebral palsy group. However, out of these flow rates and static lung volumes, tidal volume failed to show statistically significant difference between the two groups. These findings are in accordance with previous studies, which reported significantly poorer pulmonary function in children with cerebral palsy as compared to normally developed cerebral palsy children [15-17].

The results of the pulmonary function tests are indicative of restrictive dysfunction in these children, in spite of the fact that cerebral palsy children with obvious structural deformities of the chest were excluded from the study. The observed difference between the two groups can be attributed to a decrease in physical activity and chest mobility caused by motor disability in these children. Ersoz M et al., reported that children with cerebral palsy had restricted chest mobility which mainly resulted from muscle weakness, spasticity, and impaired neuro-motor control rather than involvement of costo-vertebral joint movement [18]. Muscle weakness may further be affected by decreased physical activity. It is possible that low level of physical activity and trunk control is accompanied by a decrease of respiratory function, which leads to deterioration of lung expansion by restriction of chest mobility. Restrictive lung diseases mainly result from impairment of the elastic properties of the lungs and chest wall and are characterised by reduced lung volumes and capacities. Deformities of spine and thorax are often encountered [19-21]. Also, individuals with neuromotor impairment rely on abdominal muscles for respiration showing poorly coordinated breathing [22,23]. Gradually chest movements become restricted and muscles are weakened. Eventually, the ability to take deep breath, generate expiratory force, and cough effectively is impaired [24]. These children are therefore more prone to develop respiratory infections. Although a matter of serious concern, respiratory problems in cerebral palsy has not received due attention in the clinical setting and pulmonary function test are not done routinely. Only a few studies comparing pulmonary function in cerebral palsy children with normal children have been reported in the literature. But, most studies have reported impaired pulmonary function in cerebral palsy children mostly of the restricted variety. The present study highlights the fact that simple measurement of pulmonary function can help in administration of timely rehabilitative/preventive measures that can greatly reduce morbidity and mortality in these children. Prevention of mucus stasis is of utmost importance and should be the principal focus of respiratory management for children with cerebral palsy. Exercise programs that encourage deep breathing, increase airflow rates, and stimulate cough may enhance secretion clearance. Application of pulmonary rehabilitation is important for children with cerebral palsy who have pulmonary dysfunction in order to reduce morbidity and to improve quality of life.

The findings of this study provide rationale for early clinical assessment, prevention and rehabilitative intervention for management and treatment of diverse pulmonary diseases in children with cerebral palsy.

LIMITATION

The major limitation of this study is difficulty in extrapolation due to small sample size and difficulty in performing PFT in non-cooperative children. In addition, we did not consider a variety of clinical factors related to pulmonary function in terms of oxygen saturation, chest wall excursion, etc. So, further study of overall pulmonary function including these factors will be needed.

CONCLUSION

Routine pulmonary function testing in cerebral palsy children will provide insight into aberration in respiratory function and help in introducing timely rehabilitative measures and is an invaluable tool for assessment of pulmonary function. In total, 75.68% of the cerebral palsy children showed restrictive type of pulmonary dysfunction. Significantly, lower pulmonary function in children with cerebral palsy compared to normal children. Early clinical assessment and therapeutic intervention for respiratory function should be carefully considered for children with cerebral palsy.

REFERENCES

- [1] Odding E, Roebroek ME, Stam HJ. The epidemiology of cerebral palsy: Incidence, impairments and risk factors. *Disability and Rehabilitation*. 2006;28(4):183-91.
- [2] Krageloh-Mann I, Cans C. Cerebral palsy update. *Brain Dev*. 2009;31:537-44.
- [3] Cooper MS, Mackay MT, Fahey M, Reddihough D, Reid SM, Williams K, et al. Seizures in children with cerebral palsy and white matter injury. *Pediatrics*. 2017;139(3):e20162975.
- [4] Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A Report: The definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol*. 2007;49(s109):8-14.
- [5] Wilkinson DJ, Baikie G, Berkowitz RG, Reddihough DS. Awake upper airway obstruction in children with spastic quadriplegic cerebral palsy. *J Paediatr Child Health*. 2006;42(1-2):44-48.
- [6] Strauss D, Brooks J, Rosenbloom L, Shavelle R. Life expectancy in cerebral palsy: an update. *Dev Med Child Neurol*. 2008;50(7):487-93.
- [7] Seddon PC, Khan Y. Respiratory problems in children with neurological impairment. *Arch Dis Child*. 2003;88(1):75-78.
- [8] Meehan E, Reid SM, Williams K, Freed GL, Sewell JR, Vidmar S, et al. Hospital admissions in children with cerebral palsy: A data linkage study. *Dev Med Child Neurol*. 2017;59:512-19.
- [9] Blackmore AM, Bear N, Blair E, Langdon K, Moshovis L, Steer K, et al. Predicting respiratory hospital admissions in young people with cerebral palsy. *Arch Dis Child*. 2018;103:1119-24.
- [10] Evans PM, Alberman E. Certified cause of death in children and young adults with cerebral palsy. *Arch Dis Child*. 1991;66(3):325-29.
- [11] Maudsley G, Hutton JL, Pharoah PO. Cause of death in cerebral palsy: A descriptive study. *Arch Dis Child*. 1999;81(5):390-94.
- [12] Hutton JL, Colver AF, Mackie PC. Effect of severity of disability on survival in north east England cerebral palsy cohort. *Arch Dis Child*. 2000;83(6):468-74.
- [13] Reddihough DS, Baikie G, Walstab JE. Cerebral palsy in Victoria, Australia: mortality and causes of death. *J Paediatr Child Health*. 2001;37(2):183-86.
- [14] Durufflé-Tapin A, Colin A, Nicolas B, Lebreton C, Dauvergne F, Gallien P. Analysis of the medical causes of death in cerebral palsy. *Ann Phys Rehabil Med*. 2014;57(1):24-37.
- [15] Kwon YH, Lee HY. Differences of respiratory function in children with spastic diplegic and hemiplegic cerebral palsy, compared with normally developed children. *J Pediatr Rehabil Med*. 2013;6(2):113-17.
- [16] Evans PM, Evans SJW, Alberman E. Cerebral palsies: why we must plan for survival. *Arch Dis Child*. 1990;65(12):1329-33.
- [17] Wang HY, Chen CC, Hsiao SF. Relationships between respiratory muscle strength and daily living function in children with cerebral palsy. *Res Dev Disabil*. 2012;33(4):1176-82.
- [18] Ersoz M, Selcuk B, Gunduz R, Kurtaran A, Akyuz M. Decreased chest mobility in children with spastic cerebral palsy. *Turk J Pediatr*. 2006;48(4):344-50.
- [19] Yazici M, Şenaran H. Cerebral palsy and spinal deformities. *Acta Orthop Traumatol Turc*. 2009;43(2):149-55.
- [20] Häggglund G, Pettersson K, Czuba T, Persson-Bunke M, Rodby-Bousquet E. Incidence of scoliosis in cerebral palsy. *Acta Orthopaedica*. 2018;89(4):443-47.
- [21] Koop SE. Scoliosis in cerebral palsy. *Dev Med Child Neurol*. 2009;51(s4):92-98.
- [22] Dias RC, Miller F, Dabney K, Lipton GE. Revision spine surgery in children with cerebral palsy. *J Spinal Disord*. 1997;10(2):132-44.
- [23] Benditt JO. Management of pulmonary complications in neuromuscular disease. *Phys Med Rehabil Clin N Am*. 1998;9(1):167-85.
- [24] McCool FD, Leith DE. Pathophysiology of cough. *Clin Chest Med*. 1987;8(2):189-95.

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