Effect of incentive spirometer exercise on pulmonary functions in children with spastic cerebral palsy

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Background Spastic cerebral palsy (CP) patients have lower pulmonary functions than normal healthy individuals as they usually have decreased chest wall mobility, deviation of optimal chest wall structure, and weak respiratory muscles.

Purpose The aim was to study the effect of incentive spirometer exercise (ISE) on spirometry pulmonary function in children with spastic CP.

Materials and methods Fifty spastic CP patients were randomly divided into two groups: the study group consisted of 30 patients and the control group consisted of 20 patients. Both groups were following and doing physiotherapy in the National Institute of Neuromotor System, the study group added incentive spirometer exercise to their physiotherapy program. We assessed forced expiratory volume at first second (FEV₁%), the forced vital capacity (FVC %), FEV₁/ FVC ratio, and maximal mid-expiratory flow before and after 4 weeks of exercise and lastly after another 4 weeks of exercise.

Introduction

Cerebral palsy (CP) is the most common physical disability in children [1]. Children with CP have a higher incidence of respiratory dysfunction than healthy children. They usually have recurrent chest infections, bronchiectasis, atelectasis, sleep apnea, and chronic obstructive lung disease [2]. They have high risk of morbidity and mortality due to excessive drooling and frequent aspiration that result in chest infections [3,4].

Children with spastic CP have decreased chest wall mobility, weak respiratory muscles, and deviation of optimal chest wall structure [5], resulting in lower pulmonary function than healthy children [6].

The use of incentive spirometer exercise (ISE) has been found to increase or at least to maintain inhaled lung volume, improve expectoration of lung secretions, and to decrease the frequency of chest infections [7]. It was also found to improve pulmonary function according to recent studies [8]. ISE is easy. The patient is asked to seal his lips well around the mouthpiece and then to take a slow deep breath and is motivated to achieve a preset volume by visual feedback promoting opening up of the possibly collapsed lung areas and mobilization of lung secretions [9].

Some studies have shown the effect of ISE on improving pulmonary functions and blood gases in chronic obstructive lung disease and ankylosing spondylitis **Results** The authors found significant improvements in $FEV_1\%$, FVC %, and maximal mid-expiratory flow in the study group, but not in the control group.

Conclusion The authors support the use of ISE for improving pulmonary functions in children with spastic CP. *Egypt J Bronchol* 2019 13:716–721 © 2020 Egyptian Journal of Bronchology

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patients [10]. However, the available data regarding its effect on pulmonary function in children with CP and the duration needed to gain this effect is still lacking.

Materials and methods

A prospective and randomized case-control study was conducted in the National Institute of Neuromotor System in 2019. Among patients attending the outpatient clinic, 50 patients were chosen for the study. Inclusion criteria: spastic CP patients between the age 6 and 18 years with reasonable cognitive functions and a reasonable IQ to understand the test and who are cooperative enough to allow measurement of their pulmonary function. Exclusion criteria: patients with other psychiatric and/or neurological disorders than CP, patients with medical conditions that can affect respiratory functions such as cardiac disease or respiratory disease, patients who are taking medications that has an effect on pulmonary functions like bronchodilators or beta-blockers.

The 50 patients enrolled in the study were randomly assigned into a study group that consisted of 30 CP patients and the control group that consisted of 20 CP patients. All patients were given Growth Motor Function

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Incentive spirometer device, Londonio,12 (Milano) Italy.

Classification System (GMFCS) scores [11]. The study group patients were treated using a flow-oriented incentive spirometer (Londonio, Milano, Italy) (Fig. 1).

The device has three colored balls that can be raised in their chamber once the inspiratory flow is generated by the cases through the mouthpiece. The balls are lifted according to the inspiratory flow and they remain suspended by the sustained inspiratory flow that serves as visible feedback. The instructions on how to use the ISE that were given to the study group children were as follows: the incentive spirometer should be held upright, then the patient was asked to seal his lips well around the mouthpiece and then take a slow and deep breath. The patient was motivated to achieve a preset volume by visual feedback. The patient should hold his breath for 2-3s at full inspiration. Expiration should be calm and slow. After each set of 10 breaths the patient was asked to cough so that mucus is cleared from the lungs [9]. The cases were instructed to do the exercise 10 times per session for two sessions per day for 4 weeks before doing the pulmonary function test and then continued the exercise for another 4 weeks.

Both groups were doing traditional physiotherapy. Both groups had pulmonary function testing before the study, after 4 weeks of the exercise, and after another 4 weeks of the exercise.

The Local Ethics Committee, Pediatric Department, Ain Shams University approved the study. A verbal and written consent was obtained from the caregiver of each child.

Statistical analysis

Data analysis was done using the SPSS program version 23 (IBM, Armonk, New York, USA).

Quantitative data were presented as mean and SD while qualitative data were presented as count and percentage. Student's *t*-test was used to compare quantitative data between two groups and χ^2 -test was used to compare qualitative data.

Repeated measures analysis of variance test was used to compare quantitative data at different time points for the same group.

Pearson's correlation test was used to measure the correlation between different quantitative data.

A P value less than 0.05 was considered statistically significant.

Results

Only 50 CP patients were enrolled in the study, 30 patients as the study group and 20 patients as the control group because of the difficulty in collecting cooperative patients with a reasonable IQ to understand the test. The mean age of the study group was 10.63 ± 2.44 years, while the mean age of the control group was 11.75 ± 2.10 years. Both study and

	Mean±SD		Tests of significance	P value	
	Cases (N=30)	Controls (N=30)			
Age (years)	10.63±2.44	11.75±2.10	<i>t</i> =1.673	0.101	
Weight (kg)	32.63±8.95	36.35±8.55	<i>t</i> =1.464	0.150	
Height (cm)	130.53±16.82	142.15±9.00	<i>t</i> =3.164	0.003	
GMFCS	2.83±0.70	2.85±0.67	<i>t</i> =0.084	0.933	
Sex [n (%)]					
Male	20 (66.7)	14 (70)	$\chi^2 = 0.06$	0.80	
Female	10 (33.3)	6 (30)			

GMFCS, Gross Motor Function Classification system scores.

	Cases (N=30)		Controls (N=20)		Student's t test	P value
	Mean	SD	Mean	SD		
FVC (%)	56.17	18.43	54.55	13.45	0.337	0.783
FEV ₁ (%)	60.17	18.45	58.15	13.15	0.422	0.675
FEV ₁ /FVC	107.70	8.60	106.95	6.14	0.336	0.783
MMEF (%)	67.40	26.46	65.25	16.82	0.322	00.749

FEV₁, percentage of the predicted forced expiratory volume in the first second; FVC, percentage of predicted forced vital capacity; MMEF, percentage of the predicted maximal mid-expiratory flow.

	Cases	Cases (N=30)		(<i>N</i> =20)	Student's t test	P value
	Mean	SD	Mean	SD		
FVC %	67.70	18.13	56.55	13.48	2.348	0.023
FEV ₁ %	71.63	18.56	60.40	13.66	2.318	0.025
Ratio	106.00	12.73	106.95	4.65	0.373	0.711
MMEF %	69.70	23.63	65.10	16.71	0.753	0.455

FEV₁, percentage of the predicted forced expiratory volume in the first second; FVC, percentage of the predicted forced vital capacity; MMEF, percentage of the predicted maximal mid-expiratory flow.

control groups were at a mean GMFCS of between 2.83 and 2.85, respectively. The two groups were not significantly different in age or body weight, but there was a significant difference in height. In the study group, 66.7% were men while in the control group 70% were men (Table 1). The pulmonary function parameters and GMFCS scores at baseline were not significantly different in the two groups (Tables 1, 2). The improvement in forced expiratory volume at first second (FEV₁) and forced vital capacity (FVC) was significantly higher in the study group than in the control group after 1 month of the exercise and also after 2 months (Tables 3 and 4, Figs 2, 3). There was significant negative correlation between the level of GMFCS and FVC and FEV₁ preintervention, after 1 month of exercise, and after 2 months of the exercise in both groups but more in the study group (Tables 5, 6).

Discussion

Morbidity and mortality associated with CP is mainly related to respiratory compromise, particularly in low functioning CP. Therefore, interventions for enhancing respiratory function should be a part of the management of CP patients [12]. Pulmonary function in children with CP is known to have both restrictive and obstructive characteristics of lung disease according to previous studies, and thus, the percentage of the predicted FVC, FEV₁, FEV₁/ FVC, and maximal mid-expiratory flow (MMEF) were assessed as outcome measures in the present study [13].

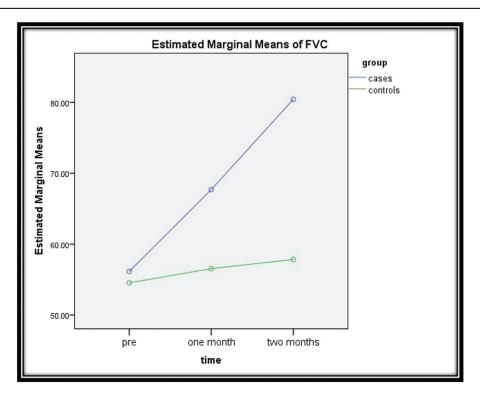
ISE is extremely used in chest physiotherapy. The patient is encouraged to perform a deep slow inspiration through visual feedback, which allows the stretching and opening of the collapsed airways. ISE is considered helpful as it is simple, inexpensive, and safe as it has no known side effects. One more advantage is that it requires no supervision once the child is trained on how to use it. Patient compliance is promoted by achieving the visual target so it encourages the patient to do his best [14].

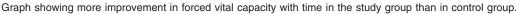
Table 4 Comparison between cases and controls, pulmonary function data

	Mean	SD	F*	P value
FVC pre (%)	56.17 ^a	18.43	64.32	< 0.001
FVC after 1 month (%)	67.70 ^b	18.13		
FVC after 2 months (%)	80.43 ^c	14.27		
FEV ₁ pre (%)	60.17 ^a	18.45	62.99	< 0.001
FEV ₁ after 1 month (%)	71.63 ^b	18.56		
FEV ₁ after 2 months (%)	85.43 ^c	12.38		
FEV ₁ /FVC ratio pre	107.70	8.60	0.33	0.72
FEV ₁ /FVC ratio after 1 month	106.00	12.73		
FEV ₁ /FVC ratio after 2	106.87	13.04		
MMEF pre (%)	67.40 ^a	26.46	4.26	0.02
MMEF after 1 month (%)	69.70	23.63		
MMEF after 2 months (%)	73.13 ^b	26.74		

FEV₁, percentage of the predicted forced expiratory volume in the first second; FVC, percentage of the predicted forced vital capacity; MMEF, percentage of the predicted maximal mid-expiratory flow. *Repeated measure analysis of variance test (a,b,c) post-hoc test.

Figure 2





The aim of the work was to study the ISE effect on spirometry pulmonary function in children with spastic CP.

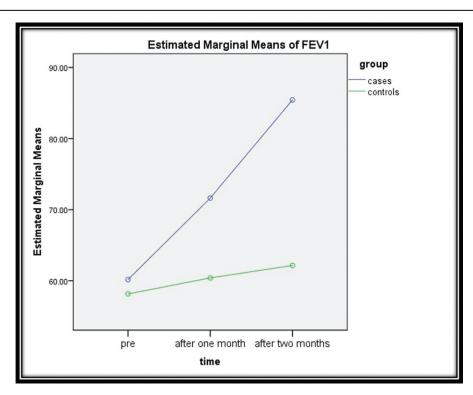
In the current study, 66.7% of the studied cases were men. This male predominance agrees with the study by Choi *et al.* [15] in which 60% of the studied cases were men. Also in the study by Lee and Kim and El-Refaey and colleagues there was male predominance [16,17].

The positive effect of exercise programs on vital capacity was reported in previous studies. But, the

effect of exercise programs on children with CP has been reported only in few studies [8].

A randomized, controlled study was done by Lee *et al.* [8] on feedback respiratory training (FRT) which consists of performance of repetitive and continuous maximal inspiration and expiration training. This study showed a significant rise in FVC and FEV₁, and not in peak expiratory flow, but this study was conducted only on nine children. So, the improvement in peak expiratory flow after feedback respiratory training did not reach a statistically significant level.





Graph showing more improvement in forced expiratory volume at first second with time in the study group than in control group.

 Table 5 Correlation between pulmonary function tests and

 Growth Motor Function Classification System among cases

Cases	GI	MFCS
	r ^a	P value
FVC % pre	-0.28	0.13
FEV ₁ % pre	-0.23	0.23
FEV ₁ /FVC ratio pre	0.35	0.06
MMEF % pre	0.004	0.98
FVC % after 1 month	-0.49	0.01
FEV ₁ % after 1 month	-0.43	0.02
FEV ₁ /FVC ratio after 1 month	0.17	0.36
MMEF % after 1 month	0.03	0.88
FVC % after 2 months	-0.64	< 0.001
FEV ₁ % after 2 months	-0.51	0.004
FEV ₁ /FVC ratio after 2 months	0.35	0.06
MMEF % after 2 months	-0.04	0.83

FEV₁, percentage of the predicted forced expiratory volume in the first second; FVC, percentage of the predicted forced vital capacity; GMFCS, Growth Motor Function Classification System scores; MMEF, percentage of the predicted maximal mid-expiratory flow. ^aPearson's correlation coefficient.

Kim *et al.* [18] concluded that FRT resulted in larger chest expansion of stroke patients, who are neurologically similar to CP patients.

Sartori *et al.* [19] used FRT in a group of cystic fibrosis patients; although they used a Spiro Tiger FRT device and they used different measures of ventilation, their results support the results of our study, as also ISE training gives visual feedback and pulmonary function

 Table 6 Correlation between pulmonary function tests and

 Growth Motor Function Classification System among controls

Controls		GMFCS
	r ^a	P value
FVC % pre	-0.83	<0.001
FEV ₁ % pre	-0.86	< 0.001
FEV ₁ /FVC ratio pre	0.34	0.14
MMEF % pre	-0.33	0.16
FVC % after 1 month	-0.84	< 0.001
FEV ₁ % after 1 month	-0.87	< 0.001
FEV ₁ /FVC ratio after 1 month	0.15	0.53
MMEF % after 1 month	-0.35	0.14
FVC % after 2 months	-0.82	< 0.001
FEV ₁ % after two months	-0.84	< 0.001
FEV ₁ /FVC ratio after 2 months	0.28	0.23
MMEF % after 2 months	-0.31	0.19

FEV₁, percentage of the predicted forced expiratory volume in the first second; FVC, percentage of the predicted forced vital capacity; GMFCS, Growth Motor Function Classification System scores; MMEF, percentage of the predicted maximal mid-expiratory flow. ^aPearson's correlation coefficient.

testing includes both tests for ventilation and tests for respiratory muscle function.

In another randomized, controlled trial by Choi and colleagues, ISE training combined with comprehensive rehabilitation therapy in spastic CP children resulted in significant improvements in pulmonary function in the study group only while the control group, which received only comprehensive rehabilitation therapy showed no significant improvement [16].

Another study done by Kwon and Lee [6] reported that GMFCS level III children had a better increase in FVC after FRT than GMFCS level I or II children. They suggested that the absence of significant improvement in GMFCS level I or II children after RFT is because they may have already reached the upper level of their respiratory function capacity. But in our study, the children with lower GMFCS had better improvement in FEV₁ and FVC. We suggest that these children have better IQ allowing them to perform the exercise more efficiently leading to better improvement in pulmonary function.

According to several other studies, FRT can improve respiratory function, according to exercise capacity, dyspnea perception, quality of life, and especially motivation of children [8,16]. However, its effect on CP children was studied only in few studies. Lee et al. [8] evaluated the efficacy of using Spiro Tiger which is a FRT device on the ventilator functions, whereas Choi et al. [16] evaluated the efficacy of using incentive spirometry (IS) on the ventilatory functions and the gross motor function measure in CP children. Both of them included children with spastic CP with different topography classification not focusing on spastic diplegia, which is the most common type of spastic CP. Also El-Refaey et al [16] investigated the efficacy of FRT using IS on respiratory muscle strength, upper to lower chest wall ratio, gross motor function, and pediatric quality of life inventory of spastic diplegic CP children and they found significant improvements of respiratory muscle strength and upper to lower chest wall ratio in the study group with nonsignificant changes in the control group.

Conclusion

This prospective, randomized case-control study demonstrated significant positive effect of ISE on parameters of pulmonary function in spastic CP children. Wider use of ISE may be beneficial for children with CP and other children who are at risk of pulmonary dysfunction as it is inexpensive, simple, and safe for children to use.

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Conflicts of interest

There are no conflicts of interest.

References

- 1 Stavsky M, Mor O, Mastrolia SA, Greenbaum S, Than NG, Erez O. Cerebralpalsy-trends in epidemiology and recent development in prenatal mechanisms of disease, treatment, and prevention. Front Pediatr 2017; 5:21.
- 2 Proesmans M. Respiratory illness in children with disability: a serious problem? Breathe (Sheff) 2016; 12:e97–e103.
- 3 Arvedson JC, Clin EJ. Feeding children with cerebral palsy and swallowing difficulties. *Nutr* 2013; 67:S9–S12.
- 4 Parkes J, Hill N, Platt MJ, Donnelly C. Oromotor dysfunction and communication impairments in children with cerebral palsy: a register study. *Dev Med Child Neurol* 2010; 52:1113–1119.
- 5 Wang HY, Chen CC, Hsiao SF. Relationships between respiratory muscle strength and daily living function in children with cerebral palsy. *Res Dev Disabil* 2012; 33:1176–1182.
- 6 Kwon YH, Lee HY. Differences of respiratory function according to level of the gross motor function classification system in children with cerebral palsy. J Phys Ther Sci 2014; 26:389–391.
- 7 Pathmanathan N, Beaumont N, Gratrix A. Respiratory physiotherapy in the critical care unit.Continuing Education in Anaesthesia. *Critical Care Pain J* 2015; 15:20–25.
- 8 Lee HY, Cha YJ, Kim K. The effect of feedback respiratory training on pulmonary function of children with cerebral palsy: a randomized controlled preliminary report. *Clin Rehabil* 2014; 28:965–971.
- 9 Kotecha SJ, Watkins WJ, Paranjothy S, Dunstan FD, Henderson AJ, Kotecha S. Effect of late preterm birth on longitudinal lung spirometry in school age children and adolescents. *Thorax* 2012; 67:54–61.
- 10 Scherer TA, Spengler CM, Owassapian D, Imhof E, Boutellier U. Respiratory muscle endurance training in chronic obstructive pulmonary disease: impact on exercise capacity, dyspnea, and quality of life. Am J Respir Crit Care Med 2000; 162:1709–1714.
- 11 Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy. *Develop Med Child Neurol* 2007; 109:8–14.
- 12 Duruflé-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; 57:24–37.
- 13 Kwon YH, Lee HY. Differences in respiratory pressure and pulmonary function among children with spastic diplegic and hemiplegic cerebral palsy in comparison with normal controls. J Phys Ther Sci 2015; 27:401–403.
- 14 So MW, Heo HM, Koo BS, Kim YG, Lee CK, Yoo B. Efficacy of incentive spirometer exercise on pulmonary functions of patients with ankylosing spondylitis stabilized by tumor necrosis factor inhibitor therapy. J Rheumatol 2012; 39:1854–1858.
- 15 Choi JY, Rha DW, Park ES. Change in pulmonary function after incentive spirometer exercise in children with spastic cerebral palsy: a randomized controlled study. *Yonsei Med J* 2016; 57:769–775.
- 16 EI-Refaey B, Abd-EI Maksoudb G, Ali O. Efficacy of feedback respiratory training on respiratory musclestrength and quality of life in children with spastic cerebralpalsy: Randomized controlled trial. *Bull Faculty Phys Ther* 2017; 22:46–52.
- 17 Lee HY, Kim K. Can walking ability enhance the effectiveness of breathing exercise in children with spastic cerebral palsy? J Phys Ther Sci 2014; 26:539–542.
- 18 Kim K, Fell DW, Lee JH. Feedback respiratory training to enhance chestexpansion and pulmonary function in chronic stroke: a double-blind, randomized controlled study. J Phys Ther Sci 2011; 23:75–79.
- 19 Sartori R, Barbi E, Poli F, Ronfani L, Marchetti F, Amaddeo A, et al. Respiratory training with a specific device in cystic fibrosis: aprospective study. J Cyst Fibros 2008; 7:313–319.