Efficacy of feedback respiratory training on respiratory muscle strength and quality of life in children with spastic cerebral palsy: Randomized controlled trial

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Background/aim
Respiratory muscle weakness and a low upper to lower chest diameter ratio are common respiratory dysfunction manifestations in children with cerebral palsy (CP), which negatively affect their quality of life. This study was conducted to investigate the effect of incentive spirometry (IS) training on these manifestations in those children.

Materials and methods
Totally, 30 children with spastic diplegic CP, aged between 6 and 12 years, were randomly assigned into two groups: the study group and the control group. Both groups received a traditional physical therapy program for 60 min. The study group received also IS training twice per session (15 min each). Children in both groups received 20 training sessions (five times/week for 4 weeks).

Outcomes
Respiratory muscle strength, ratio of upper to lower chest wall, gross motor function, and health-related quality of life were assessed before and after training.

Results
After training, significant improvements of maximal inspiratory pressure, maximal expiratory pressure, and ratio of upper to lower chest wall were obtained in the study group (P=0.002, 0.002, 0.005, respectively), whereas nonsignificant changes were obtained in the control group (P=0.719, 1, 0.284, respectively). In addition, a significant difference was observed between both groups in maximal inspiratory pressure, maximal expiratory pressure, but in favor of study group (P=0.001, 0.001, respectively), whereas there was a nonsignificant difference in the ratio of upper to lower chest wall, gross motor function, and health-related quality of life (P=0.279, 0.527, 0.876, respectively).

Conclusion
Feedback respiratory training improves respiratory muscle strength in spastic CP children.

Keywords:
cerebral palsy, incentive spirometry, quality of life, respiratory muscle strength

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Introduction

Cerebral palsy (CP) is one of the most common lifelong developmental disabilities. It is a considerable diagnostic and therapeutic challenge to the healthcare provider with degree of involvement ranging from mild with minimal disability to severe associated with many comorbid conditions. It causes substantial hardship to the patients and their families [1]. CP occurs because of an injury to the developing central nervous system, which may occur in the uterus, during delivery, or during the first 2 years of life. The manifestations depend on the magnitude, extent, and location of the insult that causes irreversible injury to the brain, brain stem, or spinal cord [2].

Respiratory factors are the principal causes of death in CP [3]. Although CP itself does not directly cause airway or parenchymal lung dysfunction, the consequences of neuromuscular impairment may lead to lung impairment and low cardiopulmonary performance [4,5]. Therefore, some of the children with CP suffer from respiratory problems, such as sleep apnea, recurrent pneumonia, widespread microatelectasis, bronchiectasis, and decreased lung

distensibility, which impairs the motor development and performance of their daily living activities [6–9]. Pulmonary function is thus an important matter in life preservation of children with CP. Previous studies have reported an association in children with CP between respiratory problems and reduced chest wall mobility, deviation of optimal chest wall structure, and insufficient respiratory muscle strength [4,9,10].

An intercostal muscle weakness was observed in patients with spinal muscular atrophy leading to a paradoxical ventilator pattern [11]. A chronic failure to expand the upper chest wall and lungs in these children was revealed by kinematic analysis of breathing [12]. The shallow respiration observed in severe CP may lead to an inability to expand the upper chest wall, which may affect upper chest development. This can lead to a lower ratio of upper chest to lower chest diameter in severe CP children compared with those normally developed [10]. Early beginning of pulmonary rehabilitation may result in maintenance and improvement of respiratory muscle strength, chest expansion, and it can also be suitable therapy in comprehensive management of CP children [13].

Several studies have reported that feedback respiratory training can improve respiratory function, endurance for exercise capacity, dyspnea perception, quality of life, and especially motivating for children [13–18]. However, the effect of feedback respiratory training in CP children has rarely been reported. Lee *et al.* [13] evaluated the efficacy of using feedback respiratory training device known as the SpiroTiger on the ventilatory functions, whereas Choi *et al.* [18] evaluated the efficacy of using incentive spirometry (IS) on the ventilatory functions and the gross motor function measure (GMFM) 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. To the best of our knowledge, no study to determine whether IS training can have an effect on the respiratory muscle strength, ratio of upper to lower chest wall, and pediatric quality of life inventory of children with spastic diplegic CP has been reported. In addition, many devices can be used in feedback respiratory training, but IS is easy to use and also an inexpensive device. Therefore, the current study was designed to evaluate the efficacy of IS training on these variables among spastic diplegic CP children.

Materials and methods

Trial design

This study is a parallel-group, active-control, randomized clinical trial. Totally, 30 children who suffered from spastic diplegic CP were randomly assigned into two groups (the study group and the control group). The study group received a traditional physical therapy exercise program in addition to IS training, and the control group received the same traditional physical therapy exercise program only. Children in both groups received traditional physical therapy program for 60 min, five times per week for a period of 4 weeks. Moreover, children in the study group received IS training twice per session (15 min each) before the traditional physical therapy program and after a 30 min rest period following it. Evaluative procedures were conducted before and after the treatment period.

Participants

This study was conducted at the outpatient clinic of Faculty of Physical Therapy, Cairo University and Resala Charitable Organization, Ministry of Social Collaboration, where the study candidates were recruited. Totally, thirty spastic diplegic children with CP from both sexes, with ages ranging from 6 to 12 years, participated in this study. The inclusion criteria were moderate spasticity that ranged from grade 2 to 3 according to the Modified Ashworth Scale, levels III and IV on the Gross Motor Function Classification System [19], ability to sit even with support, and sufficient cognition to allow them to follow simple verbal commands and instructions during tests and training. The exclusion criteria were severe cognitive impairment, chest diseases, chest or cardiac surgery, fixed deformity of affected extremities or spine, significant perceptual disorders, and visual and auditory defects.

After completion of the initial assessments, the children who matched the inclusion criteria were randomly assigned to the study group (n=15) or the control group (n=15). For randomization, a computer-generated sequence was used. For allocation concealment, sealed opaque envelopes were used. They were prepared in advance and marked inside with A or B, indicating the study group or the control group. An independent therapist, not involved in the research, labeled the training with A and B. Therefore, both the parents of the participating children and the assessors were blinded to the group separation.

The aim and nature of the study were explained for each candidate and/or parent before starting the study.

An informed written assent was obtained from parents/surrogates before enrollment. This study was approved by the Research Ethical Committee of the Faculty of Physical Therapy, Cairo University.

Evaluative procedures

Study candidates were initially evaluated by thorough medical history and physical examination. Eligible participants were further evaluated by the following.

Anthropometrics

Body height (cm) was measured with the patient standing barefoot. Body weight (kg) was measured with individuals in light clothing and was established to the nearest 0.1 kg. The body weight and height were measured using weight and height scale (health scale 70, made in China) with assistance from the therapist or the parents.

Respiratory muscle strength

Children of both groups underwent respiratory muscle strength measurement using respiratory pressure meter (The MicroRPM; CareFusion, Basingstoke, UK). The maximal inspiratory pressure (MIP) and maximal expiratory pressure (MEP) are used as measures of respiratory muscle strength [20].

MIP is the lowest pressure developed during a forceful inspiration against an occluded airway. It is recorded as a negative number. MEP is the highest pressure that can be developed during forceful expiration against an occluded airway. It is recorded as a positive number [20].

The tests were explained clearly to each child. It was important to ensure that the patient's posture is correct – i.e. they were seated upright. MIP test was performed by instructing the child to insert the mouthpiece into the mouth, ensuring that the flange is positioned over the gums and inside the lips, whereas the 'bite blocks' were between the teeth. The individual then exhaled to residual volume, and then performed a forced inhalation against the MicroRPM with as much effort as possible for as long as possible (1–3 s).

MEP test was performed by instructing the child to inspire to total lung capacity, and then perform a forced exhalation against the MicroRPM with as much effort as possible for as long as possible (1–3 s). Both tests were repeated three times to ascertain a best value [20].

Ratio of the upper to lower chest wall

Measuring the ratio of the upper to lower chest wall was used as an easy assessment of chronic hypoventilation of the upper chest in those patients [10]. Supine anteroposterior chest radiographs were taken for all children. Using the anteroposterior view of the chest radiograph, horizontal lines were drawn from the inner margin of the rib on one side to the other side, perpendicular to the line connecting the spinous processes. The longest line of the second rib (*D*apex) and ninth rib (*D*base) was measured and then the percentage ratio of the upper to lower chest wall was calculated as follows: *D*apex/*D*base×100(%) [10].

Gross motor function

The GMFM-88 test was used to assess the changes in gross motor function, as it is useful for measuring the effect of intervention programs. The GMFM test incorporates 88 items grouped in five dimensions: (A) lying and rolling; (B) sitting; (C) crawling and kneeling; (D) standing; and (E) walking, running, and jumping. Each item of the test is scored on a four-point scale, and the percentage score is calculated for each dimension. The total score is obtained by calculating the mean of the five dimension scores [21]. In the present study, the five dimensions (A, B, C, D, E) were used to evaluate the gross motor function for all children who participated in the study. The test was applied according to instructions in its user's manual [22] by the same examiner. Finally, the total score was calculated and taken for analysis in this study.

Health-related quality of life

Pediatric Quality of Life Inventory (PedsQL) is a questionnaire that assesses health-related quality of life (HRQOL) in pediatric patients with chronic health conditions. The PedsQL is a brief, standardized, generic assessment instrument. The PedsQL is based on a modular approach to measure HRQOL and consists of a 15-item core measure of global HRQOL and eight supplemental modules assessing specific symptom or treatment domains. It contains questions regarding child's physical, emotional, social, and school functioning. Parents/child self-administered the PedsQL after introductory instructions from the administrator. On the PedsQL Generic Core Scales, for ease of interpretability, items were reverse-scored and linearly transformed to a 0–100 scale, so that higher scores indicate better HRQOL. To reverse score, transform the 0–4 scale items to 0–100 as follows: 0=100, 1=75, 2=50, 3=25, and 4=0 [23]. Finally, the scale score for each dimension and total scale score were calculated.

Intervention

Traditional physical therapy program

The selected therapeutic exercise program included inhibition of abnormal movement pattern and

facilitation of normal movement patterns based on neurodevelopmental technique; training of equilibrium, righting, and protective reactions from sitting on a ball, roll, and also from standing position; and weight-bearing exercises for upper extremities from sitting and quadruped positions; stretching exercises to restore flexibility of tight muscles in the extremities (ankle planter flexors, knee and hip flexors, hip adductors, flexors of fingers, wrist and elbow, forearm pronators, and shoulder adductors and internal rotators). In addition, all children in both groups received the following exercises to improve gross motor function (sitting and standing) with the therapist guidance and assistance for the children to perform each exercise correctly:

(1) Sitting on floor; roll; chair with help if it was needed.
(2) Standing against a support surface (wall).
(3) Standing holding onto support (stand bar or sticks) with manual help at knee joints.
(4) Standing with manual support at both knee joints than at one knee joint.
(5) Rising reactions (from supine and prone to sitting position, and from sitting to standing position).

Incentive spirometer training

Children in the study group received IS training using flow-centered incentive spirometer (Triflow II, Respirogram; India). It is made from plastic material, contains three balls, connected to a tube, and a mouthpiece. When the individual takes deep inspiration, the balls rise. The ball shows the created flow. The procedure of using IS was as follows:

(1) Each child was asked to sit and relax quietly for a few minutes and pay attention to their present breathing. Then, the patient held the flow-based IS by one hand and the tube and mouthpiece by the other hand if he could or with therapist assistance if he could not.
(2) Each child was asked to take three to four slow and easy breaths, and then the child was asked to place the IS in his/her mouth and slowly and maximally inhale through the spirometer to raise the ball in the cylinder as high as he/she can, and then hold the inspiration for at least 2–3 s before exhaling normally outside the mouthpiece [24]. These steps were repeated for a total of five times, and then he/she was instructed to stop and rest for 60 s. This sequence was repeated for 30 min, 15 min before the conduction of traditional physical therapy exercise program and for 15 min after it.

Sample-size determination

On the basis of a pilot study, the primary clinical outcome of the current study was MIP that determined to obtain a power of 0.8 with α level of 0.05 with an effect size of 1.22; total sample-size estimation would be 12 participants per group using G*Power 3.1 software (Institut für Experimentelle Psychologie: Heinrich-Heine-Universität niversitätsstraße, Düsseldorf, Germany), and to account for dropout rates, the sample size was increased to 15 per group.

Data analysis

Data analysis was performed with statistical package for the social sciences version 18.0 (SPSS; SPSS Inc., Chicago, Illinois, USA). Numerical data were explored for normality by checking the distribution of data, calculating the mean, median, and SD values, using the tests of normality (Kolmogorov–Smirnov and Shapiro–Wilk tests), drawing box plot, and histogram. Respiratory muscle strength variables (MIP, MEP) and ratio of the upper to lower chest wall revealed nonparametric distribution.

The demographic data were expressed as the mean (SD) [i.e. age, weight, height, and modified Ashworth scale (MAS)]. Dependent variables (i.e. MIP, MEP, ratio, GMFM, and PedsQL) were expressed as median (interquartile range). Parametric statistical analysis was used for age, weight, height, and spasticity (MAS) using unpaired *t*-test. Nonparametric statistical analysis was used for respiratory muscle strength variables (MIP, MEP), ratio of the upper to lower chest wall, gross motor function (GMFM), and PedsQL including the Wilcoxon-signed rank test and the Mann–Whitney *U*-test for within-group and between-group comparison, respectively. The chi-square test was used for gender. A Bonferroni correction was conducted to account for increased inflation of type I error. A *P*-value of less than 0.01 was taken as significant.

Results

Overall, 26 children (12 in the study group and 14 in the control group) completed the duration of the treatment for 4 weeks, but unfortunately four could not be followed up because of illnesses or withdrawal during trial. The analysis of baseline values between the two groups, as shown in Table 1, revealed that there were no statistically significant differences with respect to age (P=0.660), weight (P=0.872), height (P=0.786), MAS (P=0.514), MIP (P=0.148), MEP (P=0.68), ratio (P=0.303), GMFM(P=0.667), and PedsQL (P=0.918).

Respiratory muscle strength

Within-group comparison between the pretreatment and post-treatment values of MIP and MEP revealed a significant improvement in the study group (P=0.002 in both parameters) and nonsignificant improvement in the control group (P=0.719 and >0.999, respectively). Between-group comparison of MIP and MEP values before treatment revealed a nonsignificant difference (P=0.148 and 0.68, respectively), whereas post-treatment comparison found a statistically significant difference in favor of the study group (P=0.001 in both parameters) (Tables 1 and 2).

Ratio of upper to lower chest wall

Within-group comparison of the pretreatment and post-treatment values of the ratio of upper to lower chest wall (%) revealed a significant improvement in the study group (P=0.005), whereas a nonsignificant improvement was observed in the control group (P=0.284). Between-group comparison of the ratio (%) values revealed a nonsignificant difference in the pretreatment and also in the post-treatment results (P=0.0.303 and 0.279, respectively), as recorded in Tables 1 and 2.

Gross motor function

The analysis of the pretreatment and post-treatment values of GMFM within each group showed nonsignificant improvement in both the study and control groups (P=0.109 and 0.18, respectively). Comparing GMFM values before and after treatment between the groups revealed a nonsignificant difference in both the pretreatment and post-treatment results (P=0.667 and 0.527, respectively), as represented in Tables 1 and 2.

Health-related quality of life

Within-group comparison of the pretreatment and post-treatment values of PedsQL score revealed a nonsignificant improvement in the study group (P=0.168) and in the control group (P=0.581). Between-group comparison of PedsQL score values revealed a nonsignificant difference in the pretreatment and also in the post-treatment results (P=0.0.918 and 0.876, respectively), as shown in Tables 1 and 2.

Table 1 Demographic and baseline clinical characteristics of children in both groups

Characteristics	Study group (n=12)	Control group (n=14)	P-value
Age (years)	9.56 (1.45)	9.83 (1.62)	0.660
Weight (kg)	31.88 (7.01)	32.34 (9.81)	0.872
Height (cm)	136.0 (13.52)	134.61 (12.37)	0.786
Sex (male/female)	(9/3)	(8/6)	0.589
MAS	2.45 (0.52)	2.53 (0.51)	0.514
GMFCS [n (%)]			
Level III	7 (58.33)	7 (50)	
Level IV	5 (41.67)	7 (50)	
MIP (cmH_2O)	44 (13)	37 (37)	0.148
MEP (cmH_2O)	39 (32.8)	52 (37)	0.68
Ratio (%)	63.2 (4.1)	63.9 (8.3)	0.303
GMFM	56.8 (4.07)	56.5 (4.06)	0.667
PedsQL	71.6 (31.7)	67.4 (22.8)	0.918

Values of age, weight, height, and MAS are expressed as mean (SD); sex distribution and GMFCS are expressed as frequency; and MIP, MEP, Ratio, GMFM, and PedsQL as median (interquartile range). GMFCS, Gross Motor Functional Classification System; GMFM, gross motor function measure; MAS, modified Ashworth scale; MEP, maximal expiratory pressure; MIP, maximal inspiratory pressure; PedsQL, pediatric quality of life; Ratio, ratio of the upper to lower chest wall.

Table 2 Statistical analysis of maximal inspiratory pressure, maximal expiratory pressure, ratio (%), gross motor function measure, and pediatric quality of life scores within each group and post-treatment comparison between groups

Parameters	Study group (n=12)			Control group (n=14)			Between-group P-value
	Pre	Post	P-value	Pre	Post	P-value	
MIP (cmH_2O)	44 (13)	59 (21.8)	0.002*	37 (37)	32 (29.3)	0.719	0.001*
MEP (cmH_2O)	39 (32.8)	62 (12.4)	0.002*	52 (37)	49 (28.3)	1	0.001*
Ratio (%)	63.2 (4.1)	66.4 (5.9)	0.005*	63.9 (8.3)	64.2 (7.4)	0.284	0.279
GMFM	56.8 (4.07)	57 (3.2)	0.109	56.5 (4.06)	56.8 (4.1)	0.18	0.527
PedsQL	71.6 (31.7)	65.2 (22)	0.168	67.4 (22.8)	70.7 (22.5)	0.581	0.876

Values are expressed as median (interquartile range). Between-group P-value, post-treatment comparison between groups; GMFM, gross motor function measure; MEP, maximal expiratory pressure; MIP, maximal inspiratory pressure; PedsQL, pediatric quality of life; P-value, before versus after treatment within each group; Ratio, ratio of the upper to lower chest wall. *Significant.

Discussion

The main purpose of this study was to investigate the efficacy of feedback respiratory training using IS on respiratory muscle strength, ratio of upper to lower chest wall, gross motor function, and pediatric quality of life inventory of spastic diplegic children with CP. According to the current results, significant improvements of respiratory muscle strength and ratio of upper to lower chest wall were observed in the study group with nonsignificant changes in the control group, indicating an augmented therapeutic effect of IS training.

The significant improvement in these variables in the study group could be attributed to respiratory muscle training. It improves inspiratory muscle strength and lung expansion and raises production of surfactant, which results in decreasing surface tension, enhancing lung compliance, better aeration of the alveoli, and decreasing the work of breathing [14,25,26]. IS allows slow maximal inspiration. During inspiration, there is elevation of balls, which encourages the patients, through a visual feedback, to perform slow and deep inspirations to their best. This pattern determines the increase of inspiratory volumes, increase of transpulmonary pressure, stretching and opening of collapsed airways, improving the performance of inspiratory muscles, and thus re-establishing the pattern of pulmonary expansion [27,28].

The current study findings are supported by the findings of previous studies. In a randomized controlled trial by Choi *et al.* [18], IS training combined with comprehensive rehabilitation therapy in spastic CP children resulted in significant improvements in pulmonary function in the experimental group, but not in the control group, which received comprehensive rehabilitation therapy only. Although they did not measure the current study variables, they suggested that the use of IS training benefits inspiratory and expiratory muscle strengthening, which supports our present results. Lee *et al.* [13] used feedback respiratory training using a device known as the SpiroTiger (Idiag AG, Volketswil, Switzerland), which trains both inspiration and expiration. Training using this device resulted in significant improvement in forced vital capacity and forced expiratory volume at 1 s with nonsignificant changes in other measured pulmonary function variables probably because of the small sample size. Although they concluded an augmented therapeutic influence of feedback respiratory training but not a comprehensive training method for improvement of pulmonary function of CP children, it has worth it, as a pilot trial supports the use of respiratory muscles training by using visual and auditory stimulations.

In addition, Lee and Kim [29]compared differences in respiratory pressures and the effect of biofeedback respiratory training in accordance with walking ability in CP children using the SpiroTiger for respiratory training. Respiratory pressures had significantly improved following training, which supports current study results.

Kim *et al.* [17] concluded that feedback respiratory training resulted in larger chest expansion of stroke patients, who are neurologically similar to CP patients.

Sartori *et al.* [16] used feedback respiratory training in a group of patients with cystic fibrosis; although they used SpiroTiger feedback respiratory training device and they used different measures of ventilation, their results support the results of the present study, as also IS training gives visual feedback and pulmonary function testing includes both tests for ventilation and tests for respiratory muscle function.Our study focused on spastic diplegic children with CP, as it was indicated that those children have weaker respiratory muscle, poorer waist expansion, and less pulmonary function values compared with spastic hemiplegic CP [30].

Regarding gross motor function and HRQOL, no significant changes were observed in both groups probably because of the short duration of the intervention (4 weeks).

It was reported that there is a positive correlation between respiratory muscle strength and daily living in domains of self-care and social function in CP children. It was suggested that this should be taken into consideration in rehabilitation programs aiming to improve abilities of daily living in those children [9].

In a pilot study by Litchke *et al.* [31], concurrent respiratory flow resistance training enhanced some aspects of HRQOL (vitality and bodily pain) in wheelchair rugby athletes. Their results differ from current results because a concurrent flow resistance training exercises both inspiratory and expiratory muscles simultaneously and also their intervention period was longer (9 weeks).

The current study has some limitations: the small sample size and short duration of the training period. The long-term effects after stopping IS training was not measured; therefore, further research into the various effects, especially the long-term research of IS training on children with CP, should be carried out. Comparison with other feedback respiratory training methods may also be needed, taking into consideration the economic cost of each.

Conclusion

IS training is an effective intervention that can be added to the traditional physical therapy exercise program for the improvement of pulmonary function of spastic CP children.

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Conflicts of interest

There are no conflicts of interest.

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