



# The Effect of Inspiratory Muscle Training on Respiratory Pressure, Pulmonary Function and Walking Ability in Preschool Children with Cerebral Palsy

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## Abstract

The aim of the present study was to evaluate the effect of Inspiratory Muscle Training (IMT) on the respiratory function and the walking ability of preschool children with Cerebral Palsy (CP). A sample of seven children with spastic CP, aged 4.5 to 7 yrs., participated in a control condition (6 weeks) and in an experimental condition (6 weeks afterwards). During the experimental period, the children performed Inspiratory Muscle Training (IMT), at 40% of their Maximum Inspiratory Pressure (MIP) for 20 mins, 2 times per week. Measured variables were: Maximum Inspiratory Pressure (MIP), Maximum Expiratory Pressure (MEP), and Forced Expiratory Volume in the First Second (FEV<sub>1</sub>), Forced Vital Capacity (FVC) and Six-Minute Walk Test (6-MWT). The statistical analyses with t-tests for dependent samples showed a significant effect of the experimental condition at MIP (p=0.016) and MEP (p=0.018), while ANOVA - repeated measurements and the within subjects' contrasts were significant for FEV<sub>1</sub> (p=0.013), FVC (p=0.005) and 6-MWT (p=0.004). Overall, the present study supported the effectiveness of IMT program on respiratory function and functional capacity in preschool children with CP. Despite the limited sample size and the absence of a control condition, the IMT may be considered with caution in the rehabilitation context of children with CP.

**Keywords:** Cerebral palsy; Inspiratory muscle training; Respiratory function; MIP

## Introduction

Cerebral Palsy (CP) is the common name used for a group of disorders related to mobility and postural development due to a non-progressive disturbance in immature brain during fetal period or childhood [1]. CP causes physical dysfunction such as sensor motor and respiratory dysfunction [2,3]. Motor disturbances of CP individuals induce paralysis or weakness of respiratory muscles [4,5] that lead to chest wall deformation and decreased mobility as well, restriction of physical activity, delayed development of the cardiopulmonary system and limitations in functional capacity [6,7].

Inadequate chest wall expansion, due to insufficient respiratory muscle strength, lead to ineffective alveolar ventilation, poor airway clearance, shortness of breath and to increased risk of developing atelectasis, pneumonia or chronic respiratory failure [8,9]. In children with CP respiratory problems are always related with decreased chest wall mobility [6,10] and often result in hospital admission or deaths [11].

Respiratory muscle strength has shown a significant association with indicators of health fitness (heart and respiratory rates) and daily functional ability in children with CP [5]. The evaluation of respiratory muscle strength should be an integral part of a holistic assessment and intervention of body function for children with CP [5]. Limitations in the functional ability of this population are due to muscle spasticity, joint range limitation and insufficient motor control [12,13].

To our knowledge, Inspiratory Muscle Training (IMT) was used in many populations and showed improvement in:

- a. Cardiorespiratory function [14]

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- b. Pulmonary function [15-20]
- c. Respiratory muscles strength [15-23]
- d. Endurance of the inspiratory muscles [21]
- e. Functional capacity (lifting, pushing, pulling, reaching and handling, climbing and balance, hand strength and dexterity and basic sitting, standing and walking) [20]
- f. Feeling of fatigue [22,19] and
- g. Dyspnea [19].

However, there are limited data for the effectiveness of IMT on pulmonary function and functional capacity in CP children. No relevant information is available for the pre-school age. According to the above, the purpose of this study was to evaluate the effectiveness of an IMT program on respiratory muscle strength, pulmonary function and functional capacity in pre-school children with CP.

## Materials and Methods

The study was approved by the local ethics committee of the School of Physical Education and Sport Science, of the National and Kapodistrian University of Athens, in accordance with the Helsinki declaration. All parents of children provided written informed consent before participation.

### Study design

1. The present study was a clinical trial and consisted of two phases. In the first phase the validity and reliability of the Six-Minute Walk Test (6-MWT), on a sample of Greek pre-school children with CP [24] was examined. More specifically, the research team assessed the: Criterion validity

- 2. Construct validity
- 3. Test-retest reliability and
- 4. Inter-rater reliability.

In the second phase, the design consisted of 3 repeated measures to evaluate individuals before and after participation in an intervention program incorporating elements of IMT. The protocol included screening and enrolment (Visit 0), baseline assessment within 2 weeks of screening (Visit 1), a second assessment after six-week period before the start of the intervention program (Visit 2), and a post assessment, immediately after the end of the intervention (Visit 3).

### Participants

Sample size was estimated using Power Analysis based on the results of Lee and Kim [25]. Based on the initial ( $M=36.9 \pm 12.3$ ) and final ( $M=42.8 \pm 12.9$ ) Maximum Inspiratory Pressure (MIP) measurements, with a power of 0.80, a significance level at 0.05, for three (3) repeated measurements, it was estimated that the minimum sample size of three (3) individuals was necessary to detect significant findings.

In the first phase, 5 children with spastic CP ( $M=5.02 \pm 0.35$ ) and 5 children ( $M=5.78 \pm 0.72$ ) with typical development were recruited. In the second phase, the recruited sample consisted of 7 children aged from 4.1 to 6.8 years old ( $M=5.81 \pm 1$ ) with spastic CP. Participants with CP were selected from a respective rehabilitation institution (ELEPAP) for CP children in Athens.

Only children with spastic CP were recruited, according to the

following inclusive criteria:

- 1) Children with spastic CP diagnosed by a paediatric neurologist from their brain MRI image,
- 2) No history of psychiatric or neurological disorders except CP,
- 3) Belonged to levels I, II, and III of the Gross Motor Function Classification System (GMFCS) and
- 4) Not participating in any organized physical activity programs during the study, outside the rehabilitation center [5,25,26-29].

The exclusion criteria were:

- 1) Any uncontrolled, clinically significant medical condition, such as coexistent cardiac disease or respiratory disease,
- 2) Children with cognitive impairment who were unable to comply with the protocol-required procedure,
- 3) Children who were taking medications that may affect respiratory function and
- 4) Participating in therapeutic approaches such as orthopaedic surgeries or botox injection over the last 6 months prior to the study [5,25-29].

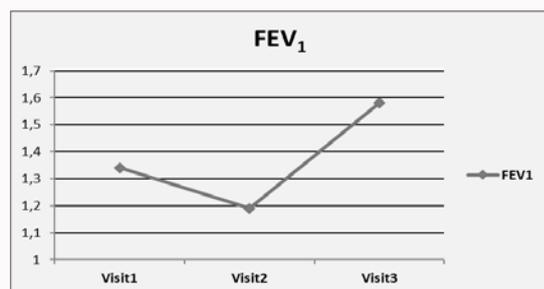
### Intervention

During the first phase, 5 children with CP and 5 children without CP performed the 1-MWT test and the 6-MWT, within a period of one week. The goal was to establish the concurrent validity of the 6-MWT and decide whether it was an appropriate walking ability assessment to use for the purposes of the study [27,30,31]. Furthermore, the construct validity was examined between children with and without CP. The 6-MWT was assessed for children with CP after 4 weeks, to check the test-retest reliability. The assessment of children with CP was conducted by two physiotherapists in order to evaluate the inter-rater reliability.

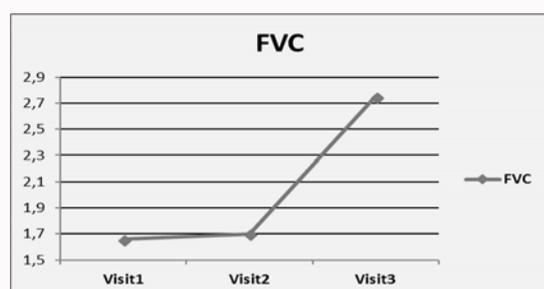
During the second phase, the 7 children with CP were assessed at baseline (Visit 1) and six weeks afterwards (Visit 2). Accordingly, they received twelve respiratory training sessions, performed twice a week, for a period of six (6) weeks. Each training session lasted twenty minutes. At the end of the 6-week intervention the children were finally re-assessed (Visit 3).

The protocol for respiratory training was planned using the respiratory training device known as Threshold-loading IMT device (Respironics, Cedar Grove, NJ, USA), which is used for training breathing muscles and for improvement of pulmonary function. The equipment has a spring-loaded valve so that inspiratory resistance can be adjusted. The IMT has been found to be reliable and reproducible for loading inspiratory muscles. Breathing exercises were performed with children seated comfortably with their head and trunk straight, in order to provide respiratory re-education and awareness. During the sessions, Threshold IMT was used in 10 series of 30s each, separated by rest periods of 30s, in order to develop muscle strength.

During the exercises, children were taking the mouthpiece into their mouth and placed a nose clip on their nose, ensuring that breathing occurred exclusively through the device. After a quiet expiration, they were encouraged to close their lips tightly around the mouthpiece and then instructed to inhale slowly and deeply as they



**Figure 1:** Average force expiratory volume at 1 second (FEV<sub>1</sub>) for the participants 6 weeks before intervention (Visit 1), the week just before start the intervention (Visit 2) and after intervention (Visit 3).



**Figure 2:** Average force vital capacity (FVC) for the participants 6 weeks before intervention (Visit 1), the week just before start the intervention (Visit 2) and after intervention (Visit 3).

could. The breathing frequency during exercise was chosen as the low frequency of 8 to 12 breaths/minute in order to prevent fatigue or dizziness. When symptoms of fatigue or hyperventilation were observed, training was stopped, and the participants were allowed to rest.

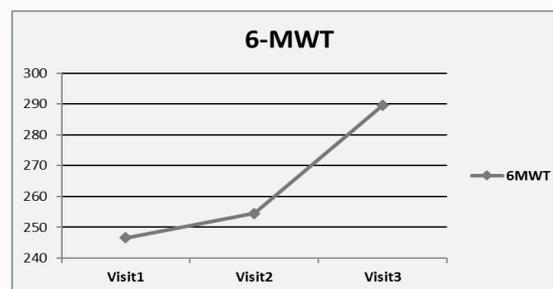
In the first session, children started training at a resistance of 20% of their MIP, in order to familiarize themselves to the IMT process. The resistance was then increased to 40% of MIP, which was well tolerated. Pressure threshold load of MIP for practice was calculated one week prior to the intervention. All training sessions were performed at 13:00 pm, under the supervision of a paediatric physical therapist, with verbal encouragement to motivate the participants to complete each session. Besides the intervention program, all children received conventional physical and occupational therapy, 2 or 3 times per week, which focused mainly on gross motor, fine motor tasks and functional activities.

### Assessment procedure

Upon enrolment, demographics and other clinical information was collected by the Primary Investigator (PI). The assessments were conducted by the PI and another paediatric physical therapist with 5 years of clinical and research experience with CP children.

### Outcome measures

**Six-minute walk test (6-MWT):** The test was performed in the corridor; between 2 lines set 15 m apart [32], according to standard recommendations, always at noon (13 pm and 14 pm) and under the supervision of the primary researcher and another physiotherapist. Participants were instructed to walk as far as possible in 6 mins. They were not allowed to run. Children received standardized vigorous verbal encouragement, and every 30 seconds were advised of the distance covered and the remaining time. Distance was calculated



**Figure 3:** Average 6-minute walking test (6-MWT) for the participants 6 weeks before intervention (Visit 1), the week just before start the intervention (Visit 2) and after intervention (Visit 3).

to the nearest meter using markings on the track. During testing children wore their own comfortable clothing, shoes and splints (as appropriate) and used their walking aid/s as appropriate.

**One-minute walk test:** The 1-MWT was conducted in the same environment with the 6-MWT, under the same conditions and included standardized instruction. Participants were asked to complete a 1-min walk using the same procedure as with the 6-MWT. The same assessors carried out both the 6-MWT and 1-MWT.

**Measurement of respiratory pressure:** For respiratory muscle strength, the Maximal Inspiratory (MIP) and Maximal Expiratory (MEP) Pressures constituted the combined respiratory strength assessment for diaphragm, abdominal, intercostal, and accessory respiratory muscles [26]. The recorded pressure during MIP (negative pressure, in cm H<sub>2</sub>O) or MEP (positive pressure, in cm H<sub>2</sub>O) testing represented the strength of inspiratory and expiratory muscles, respectively. During the assessment, the highest inspiratory and expiratory pressure against a closed mouthpiece attached to a respiratory pressure meter was recorded.

**Pulmonary function tests (PFT):** The PFT was administered during the resting period and at least one hour after termination of the physical and occupational therapy sessions. The PFT was performed before and after the 6-week IMT period by the same investigators. The PFT was performed using a portable spirometer (Micro Spirometer MS01; Micro Medical Ltd., Kent, UK) assessing Forced Expiratory Volume at one second (FEV<sub>1</sub>) and Forced Vital Capacity (FVC). The children were seated on a chair with the head and trunk straight and the hip and knee joints flexed to 90°, with use of external supporting pad. The children were instructed to inhale as deeply as possible and to blow their entire lung volume through the spirometer. This process was repeated at least three times, and the highest value was recorded [33-36].

### Statistical analysis

All data was analysed using the SPSS (Version 23.0). Descriptive statistics were used to summarize all demographic variables and subject characteristics.

During the first phase the concurrent validity of the 6-MWT was evaluated using the Pearson correlation coefficient [37]. Construct validity was evaluated by the difference between groups (children with and without CP) using Independent samples t-test [37]. Test-retest reliability was assessed through the Intraclass Correlation Coefficient (ICC) [37]. Inter-rater reliability was tested using Interclass correlation Coefficient (ICC) [37].

During the second phase, data were examined for normality via

**Table 1:** Demographic information of the two groups.

	Children with cerebral palsy (n1=5)	Children with typical development (n2=5)
Age (years)	5.02 (0.4)	5.9 (0.7)
Gender (M/F)	04-Jan	03-Feb
Height (cm)	108.8 (5.4)	113.6 (3.4)
Weight (kg)	17.9 (2.1)	20.6 (1.8)

Values are expressed as frequencies or means  $\pm$  SD.

**Table 2:** Participant's characteristics at enrolment.

Participant	Characteristics				
	Age (years/months)	Gender (boys/girls)	GMFCS level	Height (cm)	Weight (kg)
1	5.8	Female	III	106	24.3
2	6.5	Male	I	113	19.7
3	5	Male	I	115	19.1
4	6.8	Male	III	107	13.8
5	6.8	Male	I	115	23.2
6	4.1	Male	III	104	16
7	5.7	Male	I	111	19.1

GMFCS indicates Gross Motor Function Classification System.

the Shapiro-Wilk and Kolmogorov-Smirnov tests [37]. The main analysis was planned with repeated measures ANOVA or the non-parametric equivalent (Friedman's test) for 6-MWT, FEV<sub>1</sub> and FVC variables. The within subjects' contrasts were used as post hoc comparisons. The dependent t-test or the non-parametric equivalent (Wilcoxon test) was planned to compare MIP and MEP variables for the values recorded before and after the intervention. The alpha level for statistical significance was set at 0.05. [37].

## Results

### First phase-validity and reliability of 6-MWT

Five children with spastic CP and five children without CP were enrolled in the first phase. Demographic information for the children with and without CP is shown in Table 1. No statistical differences between the two groups were found in terms of age, gender, height and weight ( $p < 0.05$ ). The descriptive statistics revealed some deviation from normality for the group of children with CP. This was probably due to the fact that one of the CP children had extreme values ( $> +3$  SD) in the 1-MWT and 6-MWT. This particular child was classified in GMFCS level III. However, it was decided to retain the respective scores in the subsequent statistical analysis, since the CP population is considered as highly heterogeneous [38]. Respectively, no deviation from normality was evident for the group of children without CP.

Accordingly, the inter-correlation between the 1-MWT and 6-MWT was highly significant ( $r=0.99$ ,  $p < 0.000$ ). The independent samples t-test revealed statistically significant differences between children with and without CP ( $p < 0.007$ ). Finally, the test-retest reliability ( $ICC=0.995$ ,  $p < 0.000$ ) and the inter-rater reliability ( $ICC=0.999$ ,  $p < 0.000$ ) of the 6-MWT were high.

### Second phase-main study

Seven children with spastic CP were enrolled in the second phase of the study. Demographic information of our sample is presented in Table 2. The descriptive statistical analysis revealed that the assumption of normality was met in all outcome measures except FVC. The FVC appeared to deviate from normality probably due to the fact that one of seven children showed extreme values

in the respective measurement according to the Shapiro Wilk test. However, it was decided to retain the respective data in the statistical analysis because it did not affect the normality of the remaining variables. The normality of the outcome measures was examined and confirmed using the Shapiro Wilk test. The Shapiro-Wilk Test is more appropriate for small sample sizes instead of Kolmogorov-Smirnov Test [37].

Table 3 shows the means and standard deviations of the outcome variables, at the three-time points assessed, for the CP children who participated in the IMT and also test-retest reliabilities of the repeated assessments during the second phase.

Accordingly, the repeated measures Analysis of Variance (ANOVA) was used for the 6-MWT, FEV<sub>1</sub> and FVC measures. The ANOVA was perceived appropriate, despite the small sample size, because the assumption of normality was met (Mauchly's test). Separate t-tests for dependent samples were used to examine the differences between the repeated MIP and MEP assessments.

**MIP:** The t-test for dependent samples revealed significant difference with respect to the MIP ( $t = -3.325$ ,  $p = 0.016$ ).

**MEP:** The t-test for dependent samples revealed significant difference with respect to the MEP ( $t = -3.215$ ,  $p = 0.018$ ).

**a. FEV<sub>1</sub>:** Repeated measures ANOVA revealed significant difference between the 3 assessments ( $F=6.33$ ,  $p=0.013$ ,  $\eta^2=0.51$ ). The within subjects' cNo significant differences between the 1<sup>st</sup> and 2<sup>nd</sup> assessment ( $F=4.55$ ,  $p=0.077$ ,  $\eta^2=0.43$ ), but

**b.** Significant differences between the 2<sup>nd</sup> and 3<sup>rd</sup> assessments ( $F=10.06$ ,  $p=0.019$ ,  $\eta^2=0.63$ ) (Figure 1).

**a. FVC:** The repeated measures ANOVA revealed significant differences between the 3 assessments ( $F=8.58$ ,  $p=0.005$ ,  $\eta^2=0.589$ ). The within subjects' contrasts revealed No significant differences between the 1<sup>st</sup> and 2<sup>nd</sup> assessment ( $F=0.13$ ,  $p=0.73$ ,  $\eta^2=0.02$ ), but

**b.** Significant differences between the 2<sup>nd</sup> and 3<sup>rd</sup> assessments ( $F=8.68$ ,  $p=0.026$ ,  $\eta^2=0.59$ ) (Figure 2).

**c. 6-MWT:** Repeated measures ANOVA revealed significant

**Table 3:** Means and standard deviations of MIP, MEP, FEV<sub>1</sub>, FVC and 6-MWT in the inspiratory muscle training group (n=7), at the three-time points evaluated.

Variables	Visit 1	Visit 2	Visit 3	ICC	p
Respiratory pressure					
MIP (cm H <sub>2</sub> O)	-	38.14 (11.8)	48.14 (18) <sup>*</sup>	0.927	0.003
MEP (cm H <sub>2</sub> O)	-	35.14 (7.38)	39.57 (8.3) <sup>*</sup>	0.943	0.001
Respiratory function					
FEV <sub>1</sub> (l)	1.34 (0.62)	1.19 (0.52)	1.58 (0.7) <sup>*</sup>	0.957	0
FVC (l)	1.66 (0.86)	1.7 (0.73)	2.75 (1.2) <sup>*</sup>	0.84	0.004
Functional walking ability 6-MWT (m)	246.63 (146.73)	254.5 (145.89)	289.56 (150.7) <sup>*</sup>	0.994	0

Visit 1: time point zero (prior to the respiratory therapy program); Visit 2: time point one (after a follow-up period of 49 days); Visit 3: time points two (after a follow-up period of 90 days); IMT: inspiratory muscle training; MIP: maximal inspiratory pressure; MEP: maximal expiratory pressure; and PEF: peak expiratory flow. <sup>\*</sup>Significant differences between the three assessments p<0.05.

differences between the 3 assessments ( $F = 8.86, p = .004, \eta^2=0.596$ ). The within subjects' contrasts revealed

- No significant differences between the 1<sup>st</sup> and 2<sup>nd</sup> assessments ( $F=2.25, p=0.18, \eta^2=0.27$ ), but
- Significant differences between the 2<sup>nd</sup> and 3<sup>rd</sup> assessments ( $F=8.04, p=0.03, \eta^2=0.57$ ) (Figure 3).

## Discussion

### First phase

The present study examined the validity and reliability of the 6-MWT in a sample of preschool children with spastic CP. As we know, the validity and reliability of measures varies by sample [39]. Researchers should always present updated validity evidence of the measures used [39] and avoid generalizing validity to all samples examined, which can lead to misinterpretation of their findings [24]. A systematic review of Soloway et al. [30] provided an overview of the measurement properties of walking tests and concluded that the 6-MWT was the most extensively researched, best tolerated and most reflective of activities of daily living. Also, 1-MWT represents a valid and reliable method of assessing functional ability in children with CP aged 4 to 16 years [40,41].

The results of the present study revealed that 6-MWT is a valid and reliable test. Specifically, a strong correlation between 1-MWT and 6-MWT was found. Furthermore, children with typical development showed higher performance compared to children with spastic CP in 6-MWT. This indicates that CP as pathology affects the functional walking ability and can be ascertained when compared with a TD peer group. The results are consistent with Leunkeu et al. [27] who referred high validity ratios for this test.

High reliability was found for the 6-MWT measures in the present study that is consistent with the reliability level found in the studies of Fitzgerald et al. [31], Leunkeu et al. [27], Thompson et al. [42], Maher et al. [32] and Vinchhi et al. [43]. The results of Fitzgerald et al. [31] showed that the 6-MWT differentiated four different test groups of children (aged 4 to 18 years) classified in GMFCS levels I, II, and III and TD, according to walking ability. Leunkeu et al. [31] reported good reproducibility and validity of this test in children with CP functioning at GMFCS levels I and II in the age range 10 to 16 years. Thompson et al. [42] showed high test-retest reliability in children with CP classified in GMFCS levels I to III aged 4 to 18 years ( $ICC=0.98$ ). Maher et al. [32] as well found high test-retest reliability in children with CP classified in GMFCS levels I to III aged 11 to 17 years ( $ICC=0.98$ ), respectively. The study of Vinchhi et al. [43] indicates that test retest reliability was found to be excellent in CP

children of age group 4 to 14 years. Average group scores in these studies vary from 305 to 455 meters in children with CP and from 471 to 677 meters children that are TD [31].

Timed walking tests may offer a less expensive, simpler, and safe alternative to laboratory testing for children with CP [31]. Among such tests, the 6-MWT is a safe, standardized, self-paced walking test, requires a minimum of time, and doesn't need expensive and specialized equipment, commonly used to assess functional ability in terms of activities of daily living in children with CP [27,31]. In a Delphi study, the 6-MWT has been recommended as a sub-maximal exercise test for children with CP of GMFCS levels I to III [44]. Further, the 6-MWT may predict cardiorespiratory fitness in both healthy and severely disabled children, whether used with or without gas collection [27].

In research, the use of a valid timed walking test for preschool children with spastic CP will strengthen the internal validity and help researchers to compare both, individuals and groups of children with CP. They may also be used to track changes over time in response to growth and potential clinical interventions [31]. The 6-MWT provides a simple, reliable, and valid measure of the efficiency of gait [27].

### Second phase

In the second phase, the effect of IMT program on the MIP, MEP, FEV<sub>1</sub>, FVC and 6-MWT were examined in pre-school children with CP. Findings of the current study showed improvement of all variables after 6 weeks of respiratory training. Structural and biochemical adaptations to the inspiratory muscles through IMT are evident within 6 weeks [45].

Following the 6-week IMT intervention, the children with CP had an improvement in MIP and MEP which is in accordance with the findings of Keles et al. [46]. The study of Keles et al. [46] was conducted in children with CP between the ages of 7 to 14 years, GMFCS I and/or II. The treatment group received IMT at 30% of MIP. Children trained for a total of 30 min per day, 7 days per week, for 6 weeks. The main findings of their study were that IMT improved respiratory muscle strength in children with CP.

The aforementioned findings are supported also by the results of previous studies [15-23] with similar intervention programs, in different special populations. In particular, Fry et al. [15] used a 10-weeks IMT program in individuals with multiple sclerosis, Gosselink et al. [47] used IMT in severely disabled multiple sclerosis patients, Ray et al. [22] also used a 5-week combined progressive resistance IMT program in multiple sclerosis. Tout et al. [19] trained

with IMT COPD patients, Zeren et al. [20] trained with IMT patients with atrial fibrillation for 12 weeks, Sutbeyaz et al. [18] studied an IMT program in patients with sub acute stroke for 6 weeks, Gurses et al. [16] studied the effects of IMT on respiratory muscle strength and pulmonary functions in children with bronchiectasis, Lima et al. [17] investigated the effect of IMT and respiratory exercises in children with asthma, de Jong et al. [21] used the IMT in patients with cystic fibrosis and Smith et al. [23] examined the effect of IMT in a child with nemaline myopathy and organ transplantation.

IMT is a technique used to increase the strength of the diaphragm and accessory muscles of inspiration [20,48]. Since the inspiratory muscles, are morphologically and functionally skeletal muscles, they respond to training in the same way as any muscle if applied the appropriate physiological load [49,50]. Thus, it is not surprising the improvement in respiratory muscle strength as indicated by the increase in MIP and MEP with the appropriate training.

In the present study, FEV<sub>1</sub> and FVC improvements were achieved by participants from 2<sup>nd</sup> to 3<sup>rd</sup> measurement. This improvement might have been attributed to the intervention program due to the increase in the respiratory muscle strength. Previous findings in children with neuromuscular disorders confirmed that respiratory muscles dysfunction reduces pulmonary function [51]. The VC reflects the combined effect of weakness and the static load on the respiratory muscles [52]. According to the recent literature [7,53,54] VC is limited in children with CP by weakness of both inspiratory muscles and expiratory muscles [2,55,56], preventing full inflation and expiration. For this reason, improvement in the FVC in this study likely relates to improvement in MEP.

These findings are consistent with previous ones reported by Lee et al. [2] who investigated the effect of feedback respiratory training on pulmonary function of children with cerebral palsy aging 6 to 12 years, using a device known as SpiroTiger (Idiag AG, Volketswil, Switzerland) instead of IMT. Their training protocol included a total of 45 mins (15 mins feedback respiratory training and 30 mins conventional rehabilitation therapy) per day, with a 10-min rest period halfway through the session. They observed significant improvements in FVC and FEV<sub>1</sub> after IMT in the experimental group. Other researchers published controlled studies on pulmonary exercise interventions and reported improvement of FEV<sub>1</sub> and FVC in disabled persons after implementing similar IMT programs [15-20]. Nevertheless, Keles et al. [46] in their study didn't observed improvements in the pulmonary function test after the IMT.

a. To our knowledge, there is no published research investigating the effect of IMT on the 6-MWT for pre-school children with CP. In the present study a significant 6-MWT effect was observed, despite the absence of exercise training for the lower extremities. Specifically, according to the three repeated measurements performed, there was no significant difference between the 1<sup>st</sup> and the 2<sup>nd</sup> measurement and

b. A significant improvement from the 2<sup>nd</sup> to 3<sup>rd</sup> measurement for the 6-MWT.

Similar results have been reported in the study of Keles et al. [46] who applied IMT in children with CP. They observed marked improvement in the 6-MWT distance after IMT. Furthermore, similar findings have been documented in studies that implement IMT to patients with heart failure [57], atrial fibrillation [20] and chronic obstructive pulmonary disease [58]. According to these

studies [20,57,58], the increase of respiratory muscles strength delays the development of diaphragmatic fatigue, increases ventilator efficiency and reduces the blood flow required by the respiratory muscles during exercise [20,57]. Therefore, functional capacity may be increased. The same mechanism may be providing an increase in functional capacity in children with CP reported in the present study.

IMT is a technique used to increase the strength or endurance of respiratory muscles [59,60]. The pulmonary training protocols used by previous authors [2,15-21,46] differed somewhat from the training protocol used in this study. Regarding the exercise intensity, it was defined according to the recommendations of previous investigations [16-18,20]. The duration and frequency were defined according to the weekly program of the children in their rehabilitation context. Additionally, the supervision by the primary researcher monitored the proper implementation of the program and overall the internal validity.

Applying the framework of the International Classification of Functioning, Disability, and Health (ICF), CP clinical practice and research go beyond describing the anatomy and physiology of individuals with CP to considering their ability to participate in daily activities [61]. It is a general fact that respiratory dysfunction can affect walking ability, which together result in disturbance of normal motor development and restriction of functional activities in daily life [2,25,62-64]. The potential interactions among respiratory function, functional walking ability and daily activities of children with CP are receiving growing attention [46,2,20]. Effectiveness of respiratory training in MIP, MEP, FEV<sub>1</sub>, FVC and walking ability of children with CP is an important clinical issue.

This is the first study to apply an IMT program on respiratory function and functional walking ability in pre-school children with spastic CP. Early intervention in respiratory function is a key issue. IMT provides an effective way of exercising inhalation muscles in children with CP from pre-school age and could be included in their physiotherapy sessions. The inspiratory muscle trainer used in this study, Threshold IMT, is an inexpensive device, simple to use, without known side effects and could easily be incorporated into daily rehabilitation sessions in the clinic and then carried over in the home with minimal training. Inspiratory muscles training may be beneficial for the management of the pulmonary function, as FVC and FEV<sub>1</sub> increased via IMT. Improving the respiratory function of children with CP will lead to a better quality of life, reduction of functional restrictions and participation in physical activities.

a) The strength of the present study lies in: The sample used: pre-school children with spastic CP,

b) The absence of withdrawals and no baseline differences,

c) The internal validity, assured by supervision of the IMT program and blinding for the assessors and

d) The use of valid and reliable 6-MWT.

## Conclusion

In conclusion, present study clearly supported the effectiveness of IMT program on respiratory function and functional walking ability in pre-school children with CP. The inclusion of an inhalation muscle exercise program in the rehabilitation context of children with CP had a positive effect. Data analysis showed that the exercise of the inspiratory muscles resulted in a significant improvement of each of the research variables. These findings are consistent with the

international literature. IMT may be included in the rehabilitation context of children with CP.

## Limitations

Despite positive findings, limitations of this study exist. First, generalizability and external validity of the results are limited by the small sample size and absence of control group, due to difficulty in recruiting children with CP. Second, selection bias may have affected the results as the participants do not broadly reflect all children with cerebral palsy. Third, to participate in this study subjects had to be ambulatory. Therefore, results may not be generalizable to non-ambulatory children with CP. Fourth; the measures of pulmonary function are largely dependent on the level of the subject's effort and motivation.

1. Future studies may consider the above limitations and examine the mechanism which IMT affect the physiological indicators;
2. Long-term effects of IMT with increasing resistance;
3. Efficacy of IMT according to GMFCS level; and
4. Measuring changes in speech, cough, and number of respiratory infections.

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