

ORIGINAL RESEARCH

# Inspiratory Muscle Training and Physical Fitness in Children With Down Syndrome: A Randomized Controlled Trial



Mohamed Ali Elshafey, PhD,<sup>a</sup> Reham Saeed Alsakhawi, PhD<sup>a,b</sup>

From the <sup>a</sup>Department of Physical Therapy for Pediatrics, Faculty of Physical Therapy, Cairo University, Cairo, Egypt; and <sup>b</sup>Department of Rehabilitation Sciences, College of Health and Rehabilitation Sciences, Princess Nourah bint Abdulrahman University, Riyadh, Saudi Arabia.

## Abstract

**Objective:** To investigate the effect of inspiratory muscle training (IMT) on physical fitness in children with Down syndrome (DS).

**Design:** Randomized control trial.

**Setting:** The study was conducted in a private physical therapy center.

**Participants:** Forty boys and girls with DS aged between 7 and 10 years were enrolled in this study (N=40). They were randomly and equally divided into 2 groups (A and B).

**Interventions:** All participants received 30 minutes of aerobic exercise training 3 times per week for 12 weeks, whereas group B received an additional 30 minutes of IMT before each aerobic exercise session.

**Main Outcomes Measures:** Maximal inspiratory pressure (MIP); maximal expiratory pressure (MEP); submaximal aerobic endurance; muscular strength; endurance.

**Results:** Among the group undergoing IMT, there were significant improvements in MIP, MEP, and submaximal aerobic endurance using the six-minute walk test (6MWT); strength and endurance using the curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, and flexed-arm hang; and back saver sit and reach tests ( $P > 0.05$ ).

**Conclusions:** Inspiratory muscle training and aerobic exercise training demonstrated more significant improvements in physical fitness than in those children who received only aerobic exercise training.

Archives of Physical Medicine and Rehabilitation 2022;103:2279–87

© 2022 by the American Congress of Rehabilitation Medicine.

Down syndrome (DS) is one of the most common chromosomal causes of developmental disability in humans, with an estimated incidence of 1 in 800 live births<sup>1</sup> and a marked increase in prevalence with advanced maternal age.<sup>2,3</sup> Deficits associated with DS include muscle hypotonicity, congenital disabilities, joint hypermobility, intellectual disability, visual and auditory problems,

obesity, heart defects, facial features, and respiratory system and cardiovascular abnormalities.<sup>4</sup>

In most cases, respiratory abnormalities are responsible for morbidity and hospitalization in children with DS.<sup>5</sup> The factors contributing to respiratory abnormalities and complications in children with DS include hypotonia, limited lung surface area, lower airway volume, and significant airway compression, which limit physical fitness.<sup>6</sup>

Physical fitness is a set of elements related to physical activity.<sup>7,8</sup> There are 3 main types of physical fitness<sup>7-9</sup>: physiological fitness, such as metabolic, morphologic, bone integrity; health-related fitness, such as flexibility, body composition, cardiovascular fitness, and muscular strength and endurance; and skill-related fitness, such as agility, speed, balance, reaction time, coordination, and power. Patients with DS have low physical fitness, including aerobic fitness

Clinical Trial Registration No.: NCT04767412.

Ethical consideration: All the ethical consideration was fulfilled before conducting the study. The study was conducted after obtaining ethical approval from the IRB institutional review board, Faculty of Physical Therapy, Cairo University (P.T.REC/ 012/001922) (Clinical trial registration no.: NCT04767412). All methods were performed in accordance with the relevant guidelines and regulations. The parents of all the children were provided with a written informed consent form to sign regarding their willingness to participate in the study. The procedures and purposes of the study were described in detail to the mothers before they voluntarily agreed to have their children participate. All parents were assured of the confidentiality of their information.

Disclosures: none

and balance, compared with those without DS.<sup>8-13</sup> The low physical fitness of individuals with DS limits their participation in physical and social activities and their ability to work and perform daily living activities, resulting in poorer health-related quality of life than that in individuals without DS.<sup>8,14-16</sup>

Several studies on individuals with various medical conditions have suggested that increasing the level of physical fitness by improving the respiratory function increases social participation and the level of motivation to be physically active.<sup>17</sup> Furthermore, numerous studies have indicated positive effects of physical activity on individuals with DS; for instance, individuals with DS who participated in the Special Olympics were physically active and had a higher aerobic capacity, muscular strength, and power than sedentary individuals with DS.<sup>18</sup> Other studies have also shown that physical activity or exercise training programs targeting individuals with DS improve strength and balance,<sup>19-21</sup> cardiovascular fitness and endurance,<sup>20</sup> and peak aerobic capacity.<sup>18</sup> Physical activity or exercise training programs have also been used to improve respiratory function in children with DS.<sup>22-26</sup> Inspiratory muscle training (IMT), which is a particular type of respiratory muscle training, has been widely used in patients with stroke,<sup>27,28</sup> spinal cord injury,<sup>29</sup> chronic obstructive pulmonary disease,<sup>30,31</sup> Parkinson disease,<sup>32</sup> and multiple sclerosis.<sup>33</sup> IMT aims to improve exercise capacity, inspiratory muscle strength and endurance, and dyspnea.<sup>27-34</sup> Studies have reported improvements in quality of life, activities of daily living, cardiorespiratory fitness, and walking ability in patients receiving IMT.<sup>27,35,36</sup>

Few researchers have studied the effects of respiratory muscle training on physical fitness, particularly among children with DS. Therefore, there is a need to investigate the effect of IMT on physical fitness in children with DS.

The objectives of the current study were to evaluate the effect of aerobic exercise on physical fitness and that of IMT on respiratory muscle strength in children with DS. Furthermore, this study aimed to compare the effectiveness of the combination of aerobic exercise and IMT with aerobic exercise in improving the physical fitness of children with DS.

## Methods

### Study design

A randomized controlled trial was conducted to determine whether a combination of IMT and aerobic exercise improves physical fitness in children with DS.

### Participants

In this study, 40 boys and girls with DS, aged between 7 and 10 years (mean, 7.8±1.5 years) were recruited from a private

physical therapy center in Egypt. The inclusion criteria were as follows: (1) children with a body mass index (BMI [calculated as weight in kilograms divided by height in meters squared]) in the 5th percentile to <85th percentile and (2) children with the ability to understand and follow verbal commands and instructions used during training and tests. Children (1) with neurologic, visual, or auditory defects, (2) with asthma or any chronic chest diseases, (3) with significant tightness or deformities in the lower limbs (all the participating children underwent a quick flexibility assessment of the musculoskeletal system in the lower limbs), (4) with obesity, or (5) who performed regular physical activity were excluded. Because of the lack of previous studies on the effect of IMT on physical fitness in children with DS and the consequent inability to calculate the effect size, a pilot study was conducted on a sample of 10 patients. The statistical analysis test (2 × 2 mixed design multivariate analysis of variance) conducted on this sample size revealed a Pillai value of 0.42, which was used to detect the effect size using the G\*power program (G\*power 3.0.10). Power analysis revealed that 33 patients were sufficient to produce a power level of 95%, with a detected effect size of 0.72. With the attrition of patients during the 3-month study period, we examined all the available patients during the entire study period.

This study was approved by the ethics committee (No: P.T. REC/012/001922) and was conducted in accordance with the Declaration of Helsinki guidelines. The authors provided a detailed consent form describing the procedures and purpose of the study to the parents and legal guardians of potential participants. In the beginning, the authors conducted the study with the parents of children to explain the purpose, potential benefits, and procedure of the study with written informed consent and described the methods, procedures, and aim of the study in detail before participation. Instruments were administered to children after rapport building, and the parents of participants were guaranteed the confidentiality of their children's information (fig 1).

## Measurements

### Body mass index

To determine BMI, the weight (kg) and height (m) of each participant were measured using an electronic weight and height scale, respectively; children with a BMI ≤ 25 were included. An accurate electronic platform scale with a capacity of 150 kg and sensitivity of 100 was used to measure weight.<sup>a</sup> A stadiometer fixed to the wall with a millimeter scale was used for height measurements.<sup>b</sup> The participants stood up with an erect posture, feet together, and heels touching the wall. The apex of the ear and outer corner of the eye remained parallel to the ground, forming an angle of 90° to the bar of the stadiometer. The horizontal bar of the stadiometer was lowered and laid on the head, allowing reading in centimeters.<sup>37</sup>

### Six-minute walk test

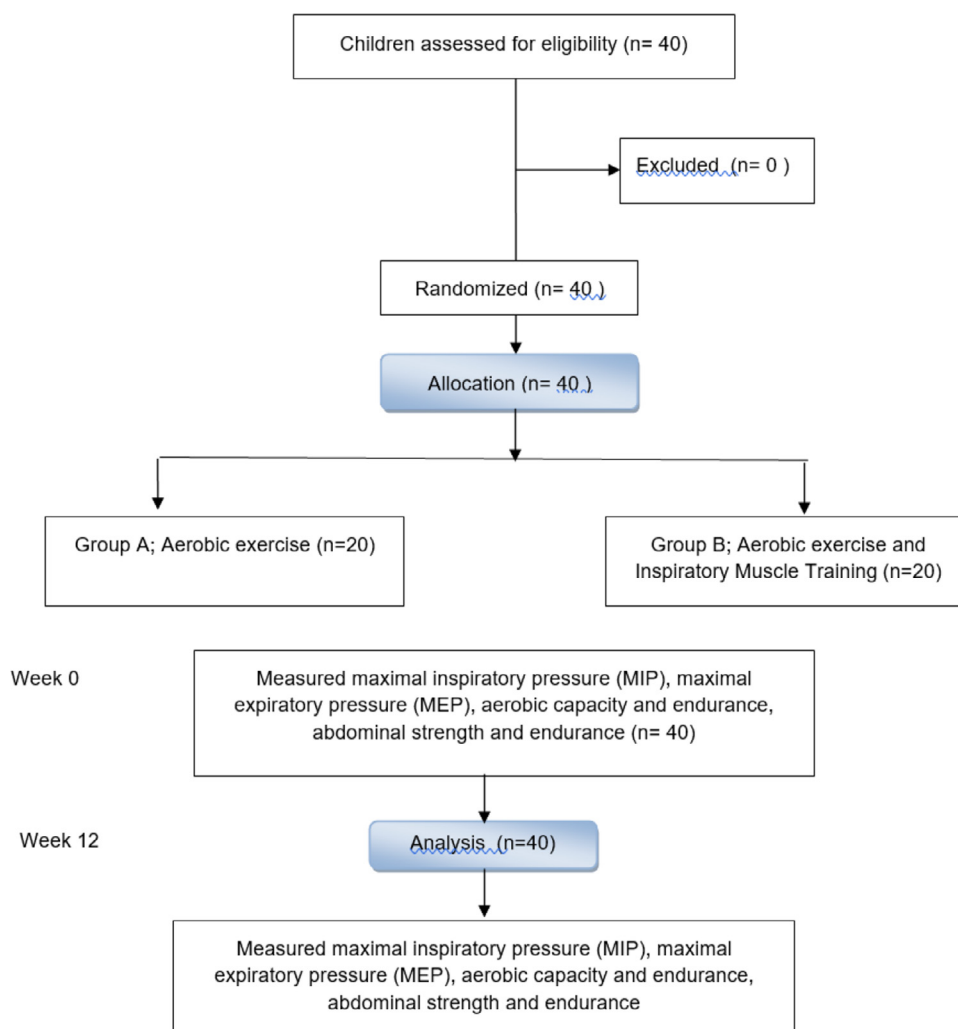
The 6-minute walk test (6MWT) assesses aerobic capacity.<sup>38</sup> The 6MWT showed good test-retest reliability and increased the walking distance after 2 practice walks in children with DS.<sup>39</sup>

### Gio Digital Pressure Gauge

The Gio Digital Pressure Gauge<sup>c</sup> was used to detect the intensity of IMT and to reflect the improvement in respiratory muscle strength after the training program.<sup>40,41</sup> Respiratory muscle strength was assessed by determining maximal inspiratory

#### List of abbreviations:

<b>BMI</b>	<b>body mass index</b>
<b>BPFT</b>	<b>Brockport Physical Fitness Test</b>
<b>DS</b>	<b>Down syndrome</b>
<b>IMT</b>	<b>inspiratory muscle training</b>
<b>MEP</b>	<b>maximal expiratory pressure</b>
<b>MIP</b>	<b>maximal inspiratory pressure</b>
<b>6MWT</b>	<b>6-minute walk test</b>



**Fig 1** Consolidated Standards of Reporting Trials flow diagram.

pressure (MIP) and maximal expiratory pressure (MEP). A mouthpiece was attached to the Gio Digital Pressure Gauge for measurement, and participants were asked to exhale into the mouthpiece.

#### Pulse oximeter

During the evaluation, pulse rate and oxygen saturation were measured using a pulse oximeter.<sup>42,d</sup>

#### Brockport Physical Fitness Test

The Brockport Physical Fitness Test (BPFT) measures physical fitness and has provided significant results in the health-related assessment of aerobic functioning in children with disabilities. This test is sufficiently valid and reliable for use with young people with selected disabilities. The BPFT includes 4-7 test items: aerobic functioning, body composition, and at least 2 test items for musculoskeletal functioning. In this study, musculoskeletal function test items were selected. BPFT includes children from the general population with health-related needs, intellectual disabilities, and mild limitations in physical fitness.<sup>43,44</sup>

#### Procedure

We collected the name and sex of all participants and imported the data into an electronic program (SPSS), which randomly and

equally divided participants into 2 groups: group A and group B. This study was conducted from October 2020 to December 2020.

First, all children were assessed before (week 0) and after (week 12) the intervention by blinded independent assessor-blinded participant allocation. For the assessment, we calculated the BMI using the following formula and excluded children with obesity:  $BMI = \text{weight (kg)} / \text{height (m)}^2$ .

Second, the 6MWT was performed on a long, flat, straight, and enclosed corridor with a hard surface. If the weather was excellent, the test was performed outdoors. All the children in this study were assessed outdoors. The walking course for the test was 30 meters in length. The length of the corridor was marked at 3-m intervals. Turnaround points were marked with cones. Children were asked to rest by sitting in a chair located near the starting position for at least 10 minutes before the test started. Children walked as far as possible for 6 minutes. They walked back and forward along the corridor. It was acceptable to slow down, stop, rest for a while, and then resume walking as soon as possible.

Third, MIP was measured using the Gio Digital Pressure Gauge, and 50% of MIP was set as the initial resistance for assessing respiratory muscle strength. It was used to measure the maximum static pressure (cmH<sub>2</sub>O) generated during exhalation. Participants were asked to inhale and exhale 3 times and the highest value of static pressure was recorded.

Fourth, the maximum heart rate was calculated using the following equation: Age-predicted maximum heart rate=220–age. We then calculated the training heart rate using the Karvonen formula: Age-predicted maximum heart rate–resting heart rate=heart rate reserve; training heart rate or aerobic training intensity=50%–70% heart rate reserve+resting heart rate.

Fifth, musculoskeletal functioning was assessed using the BPFT. The following test items were selected for the assessment: (1) The curl-up test assessed abdominal endurance by recording the number of completed curl-ups performed in the prescriptive time. (2) The dumbbell press measured the strength and endurance of the upper limbs: children were asked to lift a 1-kg dumbbell by their dominant hand as many times as possible until feeling tired; the assessor recorded the total number of dumbbell lifts within 30 seconds. (3) Trunk lift to test back muscle strength: children were asked to lift their upper body by using the back muscles and to hold this position for 1 second; the assessor recorded the best score after 3 trials of lifting. (4) Standing long jump to assess the explosive strength of the lower limbs: children were asked to jump up from a standing position with 2 feet as far as possible, and the assessor recorded the best score after 3 trials of jumping. The distance jumped (cm) was measured. (5) Pull-up: children were asked to pull up their body while grasping an overhead bar and raise their chin above the bar followed by their back to extend their arms completely. (6) Seated push-up: children were asked to push their body up and out of a chair using the upper extremities and holds this position for as long as possible, and the assessor recorded the time of pushing up. (7) Flexed-arm hang: children were asked to hang on a bar with their chin above it, and the assessor recorded the duration of how long the child could hang. (8) Back saver sit and reach individually measured the flexibility of the right and left legs. Children performed this by bending 1 leg.

All participants received 30 minutes of aerobic exercise training 3 times per week for 3 months. In contrast, group B received an additional 30 minutes of IMT, followed by a 30-minute rest period before each aerobic exercise session.

Researchers met the children several times to familiarize them and discussed each procedure at the beginning. Researchers followed all the following steps for each child during the therapeutic session and ensured the presence of people whom the children trusted, such as mothers, during the therapeutic sessions.

### Aerobic exercise

Both the groups complied with the same parameters of aerobic exercise.<sup>22,45</sup> Exercise was performed for 30 min/session 3 times/wk for 12 weeks. The exercise sessions included the following phases: (1) Warm-up phase (5 minutes): flexibility and range of motion exercises, such as half knee bending, arm circles, running in place, and toe raises. The child began with a warm-up to increase the heart rate and muscle temperature and improve blood flow. (2) Activity phase (20 minutes): moderate-intensity treadmill walking<sup>e</sup> at 50%–70% of age predicted maximum heart rate.<sup>46</sup> (3) Cooldown phase (5 minutes): Slow flexibility exercises to decrease heart rate and metabolism of the child to near normal.

### Inspiratory muscle training

IMT has been widely applied to improve the inspiratory muscle strength, endurance, exercise capacity, and dyspnea. Such treatment could improve patients' activities of daily living, quality of life, walking ability, and cardiorespiratory fitness.<sup>28</sup>

In group B, participating children were treated with IMT using the threshold IMT,<sup>f</sup> 3 times per week for 12 weeks.<sup>8,47,48</sup>

**Table 1** Comparison between both groups in mean age, z score, weight and height, and resting heart rate

Item	Group A, Mean ± SD	Group B, Mean ± SD	P Value
Age (y)	7.76±1.5	7.86±1.9	.53
z score of BMI	0.87	0.82	.23
Resting heart rate	70±11	68±12	.34

Threshold IMT is a spring-loaded valve that can adjust inspiratory resistance. This training lasted for 30 minutes. During the first 10 minutes of practice, threshold IMT was used in 5 series of 60 seconds each, separated by rest periods of 60 seconds, to develop muscle strength. During the final 20 minutes, the equipment was used uninterruptedly to develop endurance. Before the beginning of each session, the pressure threshold load throughout the practice was 50% of MIP calculated during the evaluation of children.<sup>47,48</sup>

### Data analysis

Statistical analysis was conducted using SPSS version 23.<sup>g</sup> The variable of interest was the group-by-time interaction at a priori  $\alpha$  level of 0.05 and 95% CI. Intension-to-treat analysis was performed for statistical analysis.

### Demographic characteristics

There was no significant difference between the groups in terms of mean age, z score of weight and height, and resting heart rate ( $P>.05$ ), as illustrated in table 1. There was also no significant difference in the sex ratio between the groups ( $\chi^2=0.14$ ,  $P>.05$ ).

### Results

Data were normally distributed for the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests, as assessed by the Shapiro-Wilk test ( $P>.05$ ). There were no univariate or multivariate outliers, as evaluated by the box plot and Mahalanobis distance ( $P>.05$ ), respectively. There were linear relationships, as assessed by the scatterplot, and no multicollinearity. Homogeneity of variances ( $P>.05$ ) and covariances ( $P>.05$ ) were assessed by the Levene's test of homogeneity of variances and Box's M test, respectively.

Two-way multivariate analysis of variance for outcome measurement indicated a statistically significant effect of group ( $F=4.825$ ,  $P=.001$ , partial  $\eta^2=0.682$ ), time ( $F=253.412$ ,  $P=.001$ , partial  $\eta^2=0.991$ ), and group-by-time interaction ( $F=31.46$ ,  $P=.001$ , partial  $\eta^2=0.933$ ). Post hoc within-group analysis revealed a statistically significant difference ( $P<.05$ ) for all measured variables (MEP, MIP, and the results of the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests) in the 2 groups. After the intervention, group B showed significant improvement in MEP, MIP, and the results of the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests compared with group A ( $P<.05$ ), as shown in tables 2-4. Within- and

**Table 2** Descriptive and inferential statistics of the dependent variables in the experimental and control groups pre and post the 3-month study period

Variable	Time	Group A (n = 20)	Group B (n = 20)	P Value*
6MWT (m)	Pretreatment	237.6±29.34	235.9±22.43	.838 <sup>NS</sup>
	Post treatment	253.8±18.48	267.3 ±19.24	.029 <sup>S</sup>
	P value <sup>†</sup>	.002 <sup>S</sup>	.0001 <sup>S</sup>	
MEP (cmH <sub>2</sub> O)	Pretreatment	80.1±3.24	80.4±3.10	.767 <sup>NS</sup>
	Post treatment	90.95±3.79	104.8±8.15	.0001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
MIP (cmH <sub>2</sub> O)	Pretreatment	86.45±5.54	86.9±5.90	.805 <sup>NS</sup>
	Post treatment	96.95±6.46	109.55±8.02	.0001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Curl-up test (times)	Pretreatment	5.6±1.6	5.8±1.57	.693 <sup>NS</sup>
	Post treatment	8.05±1.66	10.25 ±2.22	.001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Dumbbell press (times)	Pretreatment	5.9±1.77	5.75±2.33	.82 <sup>NS</sup>
	Post treatment	8.65±1.72	10.75±2.80	.007 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Trunk left (times)	Pretreatment	5.15±1.56	5.25±1.55	.84
	Post treatment	7.7±1.75	9.9±3.56	.003 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Stand jump (cm)	Pretreatment	9.7±3.24	9.9±3.56	.854 <sup>NS</sup>
	Post treatment	12.5±3.31	14.95 ±3.21	.023 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Seated push-up (times)	Pretreatment	8.6±1.95	8.35±2.23	.708 <sup>NS</sup>
	Post treatment	11.55±2.39	14.85±2.71	.0001 <sup>S</sup>
	P value <sup>†</sup>	0.0001 <sup>S</sup>	0.0001 <sup>S</sup>	
Pull-up (times)	Pretreatment	4.4±1.63	4.25±1.58	.77 <sup>NS</sup>
	Post treatment	6.95±1.70	9.15±2.15	.001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Flexed arm hanging (s)	Pretreatment	4.1±1.33	3.9±1.11	.61 <sup>NS</sup>
	Post treatment	6.3±1.65	9.1 ±1.25	.0001 <sup>S</sup>
	P value <sup>†</sup>	0.0001 <sup>S</sup>	0.0001 <sup>S</sup>	
Back saver right (cm)	Pretreatment	10±2.95	9.75±2.88	.788 <sup>NS</sup>
	Post treatment	13.05±2.48	16.15±2.08	.0001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	
Back saver left (cm)	Pretreatment	9.8±2.74	9.7±2.59	.906 <sup>NS</sup>
	Post treatment	12.9±2.26	16.05±2.56	.0001 <sup>S</sup>
	P value <sup>†</sup>	.0001 <sup>S</sup>	.0001 <sup>S</sup>	

Abbreviations: NS,  $P > .05$ =nonsignificant; S,  $P < .05$ =significant.

\* Intergroup comparison.

† Intragroup comparison of the results pre- and posttreatment.

between-groups differences were present at 95% CI for the effects of interventions for all dependent variables.

## Discussion

Children with DS often have low peak aerobic capacity, heart rate, and muscle strength, which are primary factors contributing to lower physical fitness than their peers without DS.<sup>26</sup> According to previous studies, children with DS respond to aerobic endurance training, and this has improved work capacity.<sup>49</sup> Therefore, this study aimed to investigate the effects of aerobic exercise programs and IMT on physical fitness in children with DS.

Both groups were homogeneous in their demographic characteristics and baseline data prior to intervention; there was no significant difference between both groups in MEP, MIP, and the results of the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back

saver sit and reach tests. After intervention there was significant improvements for all measured variables (MEP, MIP, and the results of the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests) in the 2 groups in favor of group B.

Posttreatment improvement of 6MWT, MEP, MIP, curl-up, dumbbell press, trunk left, standing jump, seated push-up, pull-up, flexed arm hang, and back saver tests in a group of children who received aerobic exercise could be attributed to improvement of ventilatory functions and general circulation, in addition to the reduction of workload on the heart through strengthening of the respiratory muscles. The capacity to use oxygen in several ways would increase while promoting improved respiratory fitness in relation to physical fitness.<sup>22</sup>

The results of the aerobic exercise group were improved by enhancing the muscle tone throughout the body, reducing resistance to airflow in the long run, enhancing fast airflow in and out of the lungs through increasing pumping efficiency and strength of

**Table 3** Between-group differences and their associated 95% CIs for outcome measures

Measure	Time	Group	Mean Difference (Group A–Group B)	95% CI for Difference		
				Lower Bound	Upper Bound	
6MWT (m)	Pretreatment	Group A	Group B	1.700	–15.020	18.420
	Post treatment	Group A	Group B	–13.500*	–25.580	–1.420
MEP (cmH <sub>2</sub> O)	Pretreatment	Group A	Group B	–.300	–2.331	1.731
	Post treatment	Group A	Group B	–13.850*	–17.920	–9.780
MIP (cmH <sub>2</sub> O)	Pretreatment	Group A	Group B	–0.450	–4.117	3.217
	Post treatment	Group A	Group B	–12.600*	–17.265	–7.935
Curl-up (times)	Pretreatment	Group A	Group B	–.200	–1.218	0.818
	Post treatment	Group A	Group B	–2.200*	–3.458	–0.942
Dumbbell press (times)	Pretreatment	Group A	Group B	0.150	–1.178	1.478
	Post treatment	Group A	Group B	–2.100*	–3.592	–0.608
Trunk left (times)	Pretreatment	Group A	Group B	–0.100	–1.098	0.898
	Post treatment	Group A	Group B	–2.200*	–3.586	–.814
Standing jump (times)	Pretreatment	Group A	Group B	–0.200	–2.383	1.983
	Post treatment	Group A	Group B	–2.450*	–4.542	–0.358
Seated push-up (times)	Pretreatment	Group A	Group B	0.250	–1.093	1.593
	Post treatment	Group A	Group B	–3.300*	–4.940	–1.660
Pull-up (times)	Pretreatment	Group A	Group B	0.150	–0.881	1.181
	Post treatment	Group A	Group B	–2.200*	–3.444	–0.956
Flexed arm hanging (s)	Pretreatment	Group A	Group B	0.200	–0.588	0.988
	Post treatment	Group A	Group B	–2.800*	–3.740	–1.860
Back saver right (cm)	Pretreatment	Group A	Group B	0.250	–1.619	2.119
	Post treatment	Group A	Group B	–3.100*	–4.567	–1.633
Back saver left (cm)	Pretreatment	Group A	Group B	0.100	–1.611	1.811
	Post treatment	Group A	Group B	–3.150*	–4.700	–1.600

\* 95% CI for Difference

the heart, allowing more blood flow through the body, leading to an improved oxygen transportation from the lungs to the heart rapidly and to all parts of the body, including the respiratory system.<sup>22</sup> This explains the effect of anaerobic exercise in children with DS.<sup>50-55</sup>

Aerobic capacity is a factor related to functional ability. It is also low in individuals with DS, leading to a loss of primary function because of poor physical fitness.<sup>56-59</sup> Aerobic exercise positively affects pulmonary function and cardiopulmonary fitness, particularly in children with DS.<sup>20,21,60,61</sup>

A study by El Kafy and Helal suggested the combination of IMT and aerobic exercise as the treatment to improve cardiopulmonary function and fitness in children with DS.<sup>22</sup> In group B, MEP, MIP, and the results of the 6MWT, curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests were significantly improved compared with those in group A. These outcomes may be explained by an increased maximal power output and maximal oxygen intake in group B. Improvement in pulmonary function may have a mechanical basis for the improvement in MEP and MIP, and the results of the 6MWT are observed mainly because of an increase in thoracic cage dimensions, that is, vertical, lateral, and anteroposterior diameters of the chest.<sup>62</sup> The improvement in MEP and MIP and the results of the 6MWT mainly indicate increased mechanical efficiency of the respiratory muscle for generating power and deep inspiration. This might have improved pulmonary function, exercise capacity, endurance, and physical fitness in group B.

Our results revealed that exercise tolerance among children with DS improved significantly after the IMT and aerobic exercise programs, which is in agreement with Strasseles et al.<sup>63</sup> They

showed that lower limb exercise training and respiratory muscle training improved the quality of life. Other studies have also indicated a relationship between IMT and improved inspiratory muscle strength in different patient populations.<sup>64-67</sup>

Posttreatment improvements in the results of the curl-up, dumbbell press, trunk lift, standing long jump, seated push-up, pull-up, flexed-arm hang, and back saver sit and reach tests in group B may be explained by the improvement in trunk control. Inspiratory muscles, particularly the diaphragm, which cause the adjustment of intra-abdominal pressure, play a significant accessory role in postural control, specifically, core stability.<sup>68</sup> Other studies have also reported diaphragmatic contraction for the stability of the trunk earlier than trunk extension, rapid arm movements, and flexion of all extremities.<sup>67-73</sup> We hypothesize that the improvement of balance control during dynamic situations and physical fitness resulted from inspiratory muscle strength, leading to aggravation of core stabilization after IMT. These improvements may be linked to an increased trunk control, exercise capacity, and participation after IMT, and some factors directly communicate and significantly affect the quality of life. The progressive incapacity of exercise may allow a child to share a longer playing time with peers. In this way, it could increase socialization and improve quality of life.

### Study limitations

This study had several limitations. First, it had a small sample size because of excluding children who engaged in regular physical activity. Additionally, enrollment of children only aged 7-10 years might limit the generalizability of the results. Second, the assessor was not blinded to the children's allocation to the groups, and they

**Table 4** Within-group differences and their associated 95% CIs for outcome measures

Variables	Groups	Time	Mean Difference (I-J)	95% CI for Difference*		
				Lower Bound	Upper Bound	
6MWT (m)	Group A	Pretreatment	Post treatment	-16.200*	-26.035	-6.365
	Group B	Pretreatment	Post treatment	-31.400*	-41.235	-21.565
MEP (cmH <sub>2</sub> O)	Group A	Pretreatment	Post treatment	-10.850*	-13.772	-7.928
	Group B	Pretreatment	Post treatment	-24.400*	-27.322	-21.478
MIP (cmH <sub>2</sub> O)	Group A	Pretreatment	Post treatment	-10.500*	-12.985	-8.015
	Group B	Pretreatment	Post treatment	-22.650*	-25.135	-20.165
Curl-up (times)	Group A	Pretreatment	Post treatment	-2.450*	-3.018	-1.882
	Group B	Pretreatment	Post treatment	-4.450*	-5.018	-3.882
Dumbbell press (times)	Group A	Pretreatment	Post treatment	-2.750*	-3.236	-2.264
	Group B	Pretreatment	Post treatment	-5.000*	-5.486	-4.514
Trunk left (times)	Group A	Pretreatment	Post treatment	-2.550*	-3.196	-1.904
	Group B	Pretreatment	Post treatment	-4.650*	-5.296	-4.004
Standing jump (times)	Group A	Pretreatment	Post treatment	-2.800*	-3.723	-1.877
	Group B	Pretreatment	Post treatment	-5.050*	-5.973	-4.127
Seated push-up (times)	Group A	Pretreatment	Post treatment	-2.950*	-3.831	-2.069
	Group B	Pretreatment	Post treatment	-6.500*	-7.381	-5.619
Pull-up (times)	Group A	Pretreatment	Post treatment	-2.550*	-3.063	-2.037
	Group B	Pretreatment	Post treatment	-4.900*	-5.413	-4.387
Flexed arm hanging (s)	Group A	Pretreatment	Post treatment	-2.200*	-2.678	-1.722
	Group B	Pretreatment	Post treatment	-5.200*	-5.678	-4.722
Back saver right (cm)	Group A	Pretreatment	Post treatment	-3.050*	-4.040	-2.060
	Group B	Pretreatment	Post treatment	-6.400*	-7.390	-5.410
Back saver left (cm)	Group A	Pretreatment	Post treatment	-3.100*	-4.000	-2.200
	Group B	Pretreatment	Post treatment	-6.350*	-7.250	-5.450

\* 95% CI for Difference

participated in the intervention. Third, this study only included outcome measures related to the body structure, function, and activity capacity of the International Classification of Functioning, Disability, and Health. Fourth, the effect of the interventions was measured immediately after 3 months of intervention. Therefore, the outcomes of this study do not indicate the sustainability of the improvements observed after treatment.

## Recommendations

Based on the results of this study, we recommend that researchers should investigate the effect of IMT on the quality of life in future studies. In addition, it could measure the effect of IMT on long-term goals in children with DS.

## Conclusions

Aerobic exercise and IMT significantly improved physical fitness in children with DS. However, the combination of aerobic exercise and IMT demonstrated even more significant improvement in physical fitness in children with DS than aerobic exercise alone.

## Suppliers

- Platform scale; Tanita.
- Bodimeter 208; Seca.
- Gio Digital Pressure Gauge DPG409 Series Omegadyne.
- ANP100 Finger Pulse Oximeter with LED Display; Anapulse.
- Valiant 2 Pediatric Treadmill.

- Inspiratory muscle training; Respironics.
- SPSS; IBM.

## Keywords

Aerobic exercise; Brockport physical fitness test; Down syndrome; Maximum inspiratory pressure; Six-minute walk test

## Corresponding author

Reham Saeed Alsakhawi, PhD, Assistant Professor, Pediatrics Physical Therapy Department, Faculty of Physical Therapy, Cairo University, Egypt, Assistant Professor, Rehabilitation Sciences Department, College of Health and Rehabilitation Sciences, Princess Nourah Bint Abdulrahman University, Riyadh, Saudi Arabia. *E-mail address:* rsm211\_pt@yahoo.com.

## Acknowledgment

We thank all participating children and their parents.

## References

- Roizen NJ, Patterson D. Down's syndrome. *Lancet* 2003;361:1281-9.
- Irving C, Basu A, Richmond S, Burn J, Wren C. Twenty-year trends in prevalence and survival of Down syndrome. *Eur J Hum Genet* 2008;16:1336-40.

3. Morris JK, Alberman E. Trends in Down's syndrome live births and antenatal diagnoses in England and Wales from 1989 to 2008: analysis of data from the National Down Syndrome Cytogenetic Register. *BMJ* 2009;339:b3794.
4. Pitetti KH, Rimmer JH, Fernhall B. Physical fitness and adults with mental retardation. An overview of current research and future directions. *Sports Med* 1993;16:23–56.
5. Ram G, Chinen J. Infections and immunodeficiency in Down syndrome. *Clin Exp Immunol* 2011;164:9–16.
6. Bull MJ. Pulmonary complications and chronic conditions of Down syndrome during childhood: an agenda for clinical care and research. *J Pediatr* 2011;158:178–9.
7. U.S. Department of Health and Human Services. Physical activity and health: a report of the surgeon general. Atlanta: Centers for Disease Control and Prevention; 1996.
8. Li C, Chen S, Meng How Y, Zhang AL. Benefits of physical exercise intervention on fitness of individuals with Down syndrome: a systematic review of randomized-controlled trials. *Int J Rehabil Res* 2013;36:187–95.
9. Bouchard C, Shepherd R, Stephens T. Physical activity, fitness and health. Leeds: Human Kinetics; 1994.
10. Temple VA, Frey GC, Stanish HI. Physical activity of adults with mental retardation: review and research needs. *Am J Health Promot* 2006;21:2–12.
11. Lloyd M, Burghardt A, Ulrich DA, Angulo-Barroso R. Physical activity and walking onset in infants with Down syndrome. *Adapt Phys Activ Q* 2010;27:1–16.
12. Black DP, Smith BA, Wu J, Ulrich BD. Uncontrolled manifold analysis of segmental angle variability during walking: preadolescents with and without Down syndrome. *Exp Brain Res* 2007;183:511–21.
13. Shields N, Taylor NF, Dodd KJ. Effects of a community-based progressive resistance training program on muscle performance and physical function in adults with Down syndrome: a randomized controlled trial. *Arch Phys Med Rehabil* 2008;89:1215–20.
14. Sutherland G, Couch MA, Iacono T. Health issues for adults with developmental disability. *Res Dev Disabil* 2002;23:422–45.
15. Lahtinen U, Rintala P, Malin A. Physical performance of individuals with intellectual disability: 30 year follow-up. *Adapt Phys Activ Q* 2007;24:125–43.
16. Verstegen RH, van Gameren-Oosterom HB, Fekkes M, Dusseldorp E, de Vries E, van Wouwe JP. Significant impact of recurrent respiratory tract infections in children with Down syndrome. *Child Care Health Dev* 2013;39:801–9.
17. Kohl HW III, Cook HD. Educating the student body: taking physical activity and physical education to school. Kohl HW III, Cook HD, editors. Washington (DC): National Academy of Sciences; 2013. p 69.
18. Balic MG, Mateos EC, Blasco CG, Fernhall B. Physical fitness levels of physically active and sedentary adults with Down syndrome. *Adapt Phys Activ Q* 2000;17:310–21.
19. Tsimaras VK, Fotiadou EG. Effect of training on the muscle strength and dynamic balance ability of adults with down syndrome. *J Strength Cond Res* 2004;18:343–7.
20. Rimmer JH, Heller T, Wang E, Valerio I. Improvements in physical fitness in adults with Down syndrome. *Am J Ment Retard* 2004;109:165–74.
21. Carmeli E, Kessel S, Coleman R, Ayalon M. Effects of a treadmill walking program on muscle strength and balance in elderly people with Down syndrome. *J Gerontol A Biol Sci Med Sci* 2002;57:M106–10.
22. El Kafy EM, Helal OF. Effect of rowing on pulmonary functions in children with Down syndrome. *Pediatr Phys Ther* 2014;26:437–45.
23. Andriolo RB, El Dib RP, Ramos LR. Aerobic exercise training programmes for improving physical and psychosocial health in adults with Down syndrome. *Cochrane Database Syst Rev* 2005;3:CD005176.
24. Fernhall B, Pitetti KH, Rimmer JH, et al. Cardiorespiratory capacity of individuals with mental retardation including Down syndrome. *Med Sci Sports Exerc* 1996;28:366–71.
25. Khalili MA, Elkins MR. Aerobic exercise improves lung function in children with intellectual disability: a randomised trial. *Aust J Physiother* 2009;55:171–5.
26. Mendonca GV, Pereira FD. Influence of long-term exercise training on submaximal and peak aerobic capacity and locomotor economy in adult males with Down's syndrome. *Med Sci Monit* 2009;15:CR33–9.
27. Xiao Y, Luo M, Wang J, Luo H. Inspiratory muscle training for the recovery of function after stroke. *Cochrane Database Syst Rev* 2012;5:CD009360.
28. Jung JH, Kim NS. The effect of progressive high-intensity inspiratory muscle training and fixed high-intensity inspiratory muscle training on the asymmetry of diaphragm thickness in stroke patients. *J Phys Ther Sci* 2015;27:3267–9.
29. Sheel AW, Reid WD, Townson AF, Ayas NT, Konnyu KJ. Spinal Cord Rehabilitation Evidence Research Team. Effects of exercise training and inspiratory muscle training in spinal cord injury: a systematic review. *J Spinal Cord Med* 2008;31:500–8.
30. Gosselink R, De Vos J, van den Heuvel SP, Segers J, Decramer M, Kwakkel G. Impact of inspiratory muscle training in patients with COPD: what is the evidence? *Eur Respir J* 2011;37:416–25.
31. Lötters F, van Tol B, Kwakkel G, Gosselink R. Effects of controlled inspiratory muscle training in patients with COPD: a meta-analysis. *Eur Respir J* 2002;20:570–6.
32. Inzelberg R, Peleg N, Nisipeanu P, Magadle R, Carasso RL, Weiner P. Inspiratory muscle training and the perception of dyspnea in Parkinson's disease. *Can J Neurol Sci* 2005;32:213–7.
33. Fry DK, Pfalzer LA, Chokshi AR, Wagner MT, Jackson ES. Randomized control trial of effects of a 10-week inspiratory muscle training program on measures of pulmonary function in persons with multiple sclerosis. *J Neurol Phys Ther* 2007;31:162–72.
34. Enright SJ, Unnithan VB, Heward C, Withnall L, Davies DH. Effect of high-intensity inspiratory muscle training on lung volumes, diaphragm thickness, and exercise capacity in subjects who are healthy. *Phys Ther* 2006;86:345–54.
35. Britto RR, Rezende NR, Marinho KC, Torres JL, Parreira VF, Teixeira-Salmela LF. Inspiratory muscular training in chronic stroke survivors: a randomized controlled trial. *Arch Phys Med Rehabil* 2011;92:184–90.
36. Sutbeyaz ST, Koseoglu F, Inan L, Coskun O. Respiratory muscle training improves cardiopulmonary function and exercise tolerance in subjects with subacute stroke: a randomized controlled trial. *Clin Rehabil* 2010;24:240–50.
37. Lohman TG, Roche AF, Martorell R. Anthropometric standardization reference manual. Champaign: Human Kinetics; 1988.
38. Li AM, Yin J, Yu CC, et al. The six-minute walk test in healthy children: reliability and validity. *Eur Respir J* 2005;25:1057–60.
39. Casey AF, Wang X, Osterling K. Test-retest reliability of the 6-minute walk test in individuals with Down syndrome. *Arch Phys Med Rehabil* 2012;93:2068–74.
40. Jalan NS, Daftari SS, Retharekar SS, Rairikar SA, Shyam AM, Sancheti PK. Intra- and inter-rater reliability of maximum inspiratory pressure measured using a portable capsule-sensing pressure gauge device in healthy adults. *Can J Respir Ther* 2015;51:39–42.
41. Caruso P, Albuquerque AL, Santana PV, et al. Diagnostic methods to assess inspiratory and expiratory muscle strength. *J Bras Pneumol* 2015;41:110–23.
42. DeMeulenaere S. Pulse oximetry: uses and limitations. *J Nurse Pract* 2007;3:312–7.
43. Winnick JP, Short FX. Conceptual framework for the Brockport Physical Fitness Test. *Adapt Phys Activ Q* 2005;22:323–32.
44. Winnick JP, Short FX. Brockport Physical Fitness Test manual-2nd edition with web resource: a health-related assessment for youngsters with disabilities. Canada: Human Kinetics Publishers; 2014.
45. Gaowgzeh RM, Chevidikunnan MF, Khan FR. Impact of pulmonary rehabilitation program on respiratory function and exercise tolerance in subjects with Down syndrome. *J Dent Med Sci* 2017;16:68–72.
46. Ordonez FJ, Rosety MA, Camacho-Molina A, Fornieles G, Rosety M, Rosety-Rodriguez M. Aerobic training at moderate intensity reduces



- protein oxidation in adolescents with Down syndrome. *Scand J Med Sci Sports* 2012;22:91–4.
47. Değer U, Uzuner S, Tuzun EH, Eker L, Mihcioglu S, Malkoc M. Pulmonary functions, respiratory muscle strength and motor skills in children with down syndrome: a comparative study. *Eur Respir J* 2017;50:PA2566.
  48. Human A, Corten L, Jelsma J, Morrow B. Inspiratory muscle training for children and adolescents with neuromuscular diseases: a systematic review. *Neuromuscul Disord* 2017;27:503–17.
  49. Mendonca GV, Pereira FD, Fernhall B. Reduced exercise capacity in persons with Down syndrome: cause, effect, and management. *Ther Clin Risk Manag* 2010;6:601–10.
  50. Yang D, Park S, Kang J, Kim J, Kim S. Effects of inspiratory muscle exercise using biofeedback on inspiratory muscle activity and pulmonary function in patients with stroke. *J Kor Phys Ther* 2015;27:287–91.
  51. Keles MN, Elbasana B, Apaydina U, Aribas Z, Bakirtas A, Kokturk N. Effects of inspiratory muscle training in children with cerebral palsy: a randomized controlled trial. *Braz J Phys Ther* 2018;22:493–501.
  52. Farid R, Ebrahimi A, Khaledan A, Talaei-Khoie M. Effect of aerobic exercise training on pulmonary function and tolerance of activity in asthmatic patients. *Iran J Allergy Asthma Immunol* 2005;4:133–8.
  53. Saudo E, Oliga E, Matsuse T, Teramoto S. The effects of pulmonary rehabilitation combined with inspiratory muscle training on pulmonary function and inspiratory muscle strength in elderly patients with chronic obstructive pulmonary disease. *Nippon Ronen Igakkai Zasshi* 1997;34:929–34.
  54. Harver A, Mahler D, Baird C. Descriptors of breathlessness in healthy individuals. *Chest* 2000;118:679–90.
  55. Tomohiro O, Yoshio N, Kiyoji T. Effects of exercise intensity of physical fitness and risk factors for coronary heart disease. *Obes Res* 2003;11:1131–9.
  56. Sutbeyaz S, Koseoglu B, Gokkaya N. The combined effects of controlled breathing techniques and ventilatory and upper extremity muscle exercise on cardio-pulmonary responses in patients with spinal cord injury. *Int J Rehabil Res* 2005;28:273–6.
  57. Emiel F. Approaches to improving health status in chronic obstructive pulmonary disease. *Am Thorac Soc* 2006;3:262–9.
  58. Carmeli E, Barchad S, Lenger R, Coleman R. Muscle power, locomotor performance and flexibility in aging mentally-retarded adults with and without Down's syndrome. *J Musculoskelet Neuronal Interact* 2002;2:457–62.
  59. Carmeli E, Kessel S, Bar-Chad S, Merrick J. A comparison between older persons with down syndrome and a control group: clinical characteristics, functional status and sensorimotor function. *Downs Syndr Res Pract* 2004;9:17–24.
  60. Pitetti KH, Boneh S. Cardiovascular fitness as related to leg strength in adults with mental retardation. *Med Sci Sports Exerc* 1995;27:423–8.
  61. Heller T, Hsieh K, Rimer J. Barriers and supports for exercise participation among adults with Down syndrome. *J Gerontol Soc Work* 2002;38:161–78.
  62. Vicente-Rodriguez G, Ara I, Perez-Gomez J, Dorado C, Calbet JA. Muscular development and physical activity as major determinants of femoral bone mass acquisition during growth. *Br J Sports Med* 2005;39:611–6.
  63. Tylor NF, Dodd KJ, Shields N, Bruder A. Therapeutic exercise in physiotherapy practice is beneficial: a summary of systematic reviews 2002–2005. *Aust J Physiother* 2007;53:7–16.
  64. Ajith S, D'Sa IP, Faisal CKM, Anandh V, Sreejith N. Physiological Cost Index (PCI) in patients with chronic obstructive pulmonary disease (COPD) before and after giving two commonly used breathing exercises. *Int J Curr Res Rev* 2011;3:41–8.
  65. Strasseles S, Smith D, Mahajan P. The costs of treating COPD in the United States. *Chest* 2001;119:344–52.
  66. McConnell A. Respiratory muscle training e-book: theory and practice. Elsevier Health Sciences; 2013.
  67. Silva IS, Fregonezi GA, Dias FA, Ribeiro CT, Guerra RO, Ferreira GM. Inspiratory muscle training for asthma. *Cochrane Database Syst Rev* 2013;2013:CD003792.
  68. Houston BW, Mills N, Solis-Moya A. Inspiratory muscle training for cystic fibrosis. *Cochrane Database Syst Rev*. John Wiley & Sons, Ltd 2008;4:CD006112. <https://doi.org/10.1002/14651858.CD006112.pub2>.
  69. Pollock RD, Rafferty GF, Moxham J, Kalra L. Respiratory muscle strength and training in stroke and neurology: a systematic review. *Int J Stroke* 2013;8:124–30.
  70. Hodges P, Cresswell A, Thorstensson A. Preparatory trunk motion accompanies rapid upper limb movement. *Exp Brain Res* 1999;124:69–79.
  71. Hodges PW, Gandevia SC. Changes in intra-abdominal pressure during postural and respiratory activation of the human diaphragm. *J Appl Physiol* 2000;89:967–76.
  72. Kolar P, Sulc J, Kyncl M, et al. Stabilizing function of the diaphragm: dynamic MRI and synchronized spirometric assessment. *J Appl Physiol* 2010;109:1064–71.
  73. Hudson AL, Butler JE, Gandevia SC, De Troyer A. Role of the diaphragm in trunk rotation in humans. *J Neurophysiol* 2011;106:1622–8.