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Experimental Article

The effect of incentive spirometer training on oromotor and pulmonary functions in children with Down's syndrome

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الملخص

أهداف البحث: فحص تأثير التدريب بمقياس التنفس المحفز على الوظائف العضلية للفم، ووظائف التنفس عند أطفال متلازمة داون.

طرق البحث: تم تقسيم ٣٤ طفلا مصابين بمتلازمة داون من الجنسين من عمر ٦ ـ٢١ سنة عشوائيا لمجموعتين. المجموعة أ تلقت تمرينات عضلية للفم، والمجموعة ب تلقت تمرينات عضلية للفم بالإضافة إلى التدريب بمقياس التنفس المحفز. تم استخدام قياس التنفس المحوسب ذي شاشة رئيسة لاختبار وظيفة الرئة لقياس وظائف التنفس (القدرة الحيوية القسرية، وتدفق الزفير القسري في ثانية واحدة والزفير)، واستخدام التقييم العضلي الفموي بالدرجات لتقييم الوظائف العضلية للفم قبل وبعد تلقي العلاج.

النتائج: نتائج ما بعد العلاج أوضحت فرقا إحصائيا واضحا في الوظائف العضلية للفم ووظائف التنفس في كل من المجموعتين ولكن لم يوجد فرق واضح بين المجموعتين في النتائج.

الاستنتاجات: التمرينات العضلية للفم أكثر تأثيرا من التدريب بمقياس التنفس المحفز في تحسين كلا من وظائف التنفس والوظائف العضلية للفم لدى أطفال متلازمة داون.

الكلمات المفتاحية: متلازمة داون؛ وظائف التنفس؛ الوظائف العضلية للفم؛ القدرة الحيوية القسرية؛ التقييم العضلي الفموي بالدرجات

Abstract

Objectives: This study investigated the effect of incentive spirometry training on oromotor and pulmonary functions in children with Down's syndrome.

Methods: Thirty-four children with Down's syndrome were randomly divided into two groups; the children were of both sexes and aged between 6 and 12 years. Group A received only oromotor exercises, while Group B received oromotor exercises and incentive spirometry training. The pulmonary function test was performed using computerized spirometry model master screen that assessed pulmonary functions (peak expiratory flow, forced vital capacity, and forced expiratory volume in 1s), while the orofacial myofunctional evaluation with score (OMES) was used to evaluate oromotor function before and after treatment.

Results: The post treatment results showed significant difference in oromotor and pulmonary functions within both groups, but no significant differences were found between the two groups.

Conclusions: Oromotor exercises are more effective than incentive spirometry training in improving both pulmonary and oromotor functions in children with Down's syndrome.

Keywords: Down's syndrome; Oromotor functions; Orofacial myofunctional evaluation with score; Peak expiratory flow; Pulmonary functions

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Introduction

Down's syndrome, also known as trisomy 21, is the most prevalent chromosomal disorder and the commonest cause of intellectual disabilities.[1] Children with Down's syndrome often have impaired oral motor control due to dyspraxia and muscle weakness.[2,3]

The stomatognathic system plays a role in both feeding and homogenous breathing. To fulfil these functions, it relies on a sophisticated sensorimotor system which is under the control of various subcortical and cortical regions.[4] Orofacial myofunctional disorders interfere with this system, disrupting a patient's appearance and causing dysfunctions in their lips, cheeks and mandible, respiration, mastication, swallowing, speech, and tongue mobility. Many oral diseases can lead to such disorders, including genetic, congenital, acquired, and degenerative disorders, as well as mouth breathing, malocclusion, and temporomandibular disorders.[5,6]

In Down's syndrome, several orofacial structural anomalies occur, such as incomplete development of the midfacial region, relatively small bones in the upper jaw and nose bridge, ligamentous laxity in the temporomandibular joint, and hypotonia of the facial and masticatory muscles. In addition, patients with Down's syndrome tend to have a small oral cavity and a hypotonic tongue that rests between the dental arches and, protrudes from the mouth, giving the appearance of macroglossia. This leads to tongue protrusion and tongue thrust during eating and speech.[7]

Oromotor exercises are nonspeech activities that involve sensory stimulation or actions of the lip, jaw, tongue, soft palate, larynx, and respiratory muscles to improve their functions. They may include sensory stimulation, passive exercise, active muscle exercise, and muscle stretching.[8]

Children with Down's syndrome have poorer lung function than healthy, age-matched controls.[9] Indeed, children with intellectual disabilities have decreased abdominal strength and endurance, probably because they tend to participate in fewer vigorous recreational activities than children of the same age.[10]

However, no studies have yet sufficiently addressed the effect of incentive spirometer training on oromotor functions or the effect of oromotor exercises on pulmonary function in children with Down's syndrome. Thus, the novel purpose of the present study was to investigate the effect of incentive spirometer training on oromotor and pulmonary functions in children with Down's syndrome.

Materials and Methods

Study design

This was a randomized clinical trial that was conducted from 2017 to 2019. Thirty-four children were assigned randomly to two groups using closed envelopes to avoid selection bias; 15 children received oromotor exercises plus incentive spirometry, while 19 children received oromotor exercises only. The power of the study was measured *post hoc* using G*Power 3.1 software, with a sample size of 34 subjects, a 0.05 type I error (2 tailed), and a critical Z of 1.95; the power was 0.81.

The procedures of the study were explained to the parents, all of whom signed consent forms.

Participants

At the beginning of the study, 40 children with Down's syndrome were recruited. However, six did not continue: two were excluded for non-compliance, two refused to perform pulmonary function testing, one was suffering from morbid obesity, and one failed to attend the re-evaluation. Ultimately, 34 children completed the study: 14 boys and 20 girls aged between 6 and 12 years (Figure 1).

The children were recruited from the National Institute of Neuromotor System. All of them had both Down's syndrome and orofacial myofunctional disorders, were able to follow instructions, and had a body mass index between the 5th and the 95th percentile, according to a growth chart. Children were excluded from the study if they had visual or hearing defects, acute or chronic respiratory diseases, or a serious medical condition, such as untreated cardiac problems.

Instrumentation

Computerized spirometry model master screen pulmonary function testing was used in the current study, which was performed at National Research Centre. Spirometry is the most common pulmonary function test; it measures flow, as well as the volume of air that could be inhaled and exhaled. The orofacial myofunctional evaluation protocol with scores (OMES) was used to evaluate appearance, tongue posture and mobility, lips, jaws and cheeks, mastication, deglutition, and respiration.

Procedures

Evaluation

Pulmonary functions. One pulmonary function assessment was performed by a blinded examiner. Forced vital capacity (FVC), forced expiratory volume in 1 s (FEV1), and peak expiratory flow (PEF) were measured. The children sat on a chair with their head and trunk straight and their hip and knee joints flexed at a right angle. They were then instructed to take deep breaths as they blew a piece of paper. A clean mouthpiece connected to the flow sensor was introduced into their mouth, and a nasal clip was placed around their nose to prevent air escaping. They then took a deep breath and held it long enough to seal their lips tightly around the mouth-piece. Finally, they expired hard and as forcibly as possible, until they could no longer expel any air. This task was repeated three times by each child, and the best trial was recorded. The pulmonary function test was performed at the initial assessment and repeated after treatment ended. Both times, the assessment was conducted pre-bronchodilator and post-bronchodilator to detect reversibility.

Oromotor functions. The OMES was used to evaluate oromotor functions.[5] Regarding appearance/posture, the children's lips, tongue, mandible, cheeks, face (symmetry), and hard palate were evaluated. To assess mobility, the children were asked to perform four to six movements each with their lips, tongue, cheeks, and mandible. The children's mode of breathing were observed at rest and during mastication and assessed as nasal, or oronasal (light dysfunction), or oronasal (severe dysfunction). Deglutition was assessed, first using a liquid bolus and then using a solid bolus; in particular, the experimenters observed lip behaviour, tongue behaviour, and other behaviours, as well as signs of alteration (jaw sliding, tension in the facial muscles, food escape, movement of the head or other parts of the body, choking, or noise). The efficiency of deglutition was assessed in terms of the number of swallowing repetitions. With regards to mastication, the children's bites were observed and their masticatory type evaluated in terms of the percentage of chewing strokes occurring on each side of the oral cavity. Their chewing time was also recorded.

Total score ranged from 32 to 104, with the lowest value indicating the worse orofacial myofunctional condition, and the highest value the best degree of myofunctional disorder.

-Ruler was used for measuring the distance of the mouth opening from a mark placed on the highest point of the upper border of the upper lip till the mark placed on the lowest point of lower border of the lower lip.

Intervention

Oromotor exercises

To improve proprioception, tone, and mobility, the following oromotor exercises were performed: (1) Sensory stimulation of cheeks, lips, gums, and tongue using an electric toothbrush to the cheeks, as well as lip massage, cheek massage, and tapping of the cheeks, lips, and gums. Such oral motor therapy facilitates normal oral motor patterns and increases sensory awareness, mouth closure, jaw stability, tongue mobility, and strength. Fast tapping and vibration increase tone; (2) Lip and cheek exercises were conducted by instructing the children to purse their lips, smile, hold for 10 s, relax for 10 s, and repeat five times. They were then asked to press on a tongue depressor with their lips, hold for 10 s, relax for 10 s, and repeat 10 times. Next, they were requested to puff their cheeks with their lips closed, hold for 10 s, relax for 10 s, and repeat five times. The children also performed exercises using horns and bubbles. Next, they performed lip protrusion exercises to the right and left, with five repetitions for 10 s on each side, followed by 10 s of relaxation. Cheek compression exercises were conducted with five repetitions, holding for 10 s, with 10 s of relaxation; (3) With regards to tongue exercises, the children were asked to elevate their tongue to the alveolar ridge and hold it there for as long as possible. They were then told to push against the elevated tongue with a lollipop stick or tongue depressor, hold for 10 s, relax for 10 s, and repeat 10 times; (4) To exercise their jaws, the children were expected to close their jaws against resistance; that is, bite on a tongue depressor, hold for 10 s, relax for 10 s, and repeat 10 times; (5) Finally, the children were asked to drink using a straw, which helps to retract the tongue, seal the lips, and compress the cheeks.

Incentive spirometry

In this test, the patients sat on the chair in a comfortable position, holding the incentive spirometer in an upright position. They were asked to expire quietly, close their lips firmly around a mouthpiece, and inhale slowly and deeply until the ball in the spirometer lifted (the spirometer pistons rise with inspiration and descend with expiration). Next, they were instructed to hold their breath for as long as possible, and then breathe out slowly. Corresponding to the inspiratory flow, the balls lifted and remained suspended, which served as visible feedback.

All children received the treatment sessions for 1 h weekly over 3 months, for a total treatment time of 12 h. In addition, they performed a home program, including drinking from straws, playing with horns and bubbles, and repeating the exercises from the session, 2–3 times per day.

Statistics

SPSS for Windows, version 20 (SPSS, Inc., Chicago, IL) was used to conduct the statistical analysis. Significance was set at a p-value of <0.05. The t-test was used to compare general characteristics between the two groups.

Data were screened for normality, homogeneity of variance, and the presence of extreme scores. The Shapiro–Wilk test for normality showed that appearance, mobility, function, total score, and pulmonary function score were not normally distributed, so the Mann–Whitney U test was used to compare these groups. The Wilcoxon test was used to compare within-group differences before and after the study. Data are expressed as median and interquartile range.

Results

Results revealed no significant differences between the two groups in terms of mean age, BMI, or IQ ($p > 0.05$ in all cases; Table 1). Regarding the total OMES score, no significant difference in either pre- or post-study total score was observed between the two groups ($p > 0.05$ in both cases; Table 2). However, in both groups, a significant difference did occur between the pre- and post-study OMES scores ($p < 0.05$), with a percentage improvement of 10.4% in Group A the oromotor only group and that of 10% in Group B the oromotor and spirometer group. Concerning the ruler measurement of the mouth opening, there was no significant difference in either pre- or post-study values between the two groups ($p > 0.05$ in both cases; Table 3). However, in both groups, there was a significant difference between the pre- and post-study values ($p < 0.05$), with a percentage improvement of 54.5% in Group A and that of 63.6% in Group B. With regards to FVC, FEV1, and PEF, there was no significant difference in either pre- or post-study values between the two groups ($p > 0.05$). However, in both groups, a significant difference was found between the pre- and post-study values ($p < 0.05$). FVC showed a percentage improvement of 54.3% in Group A and of

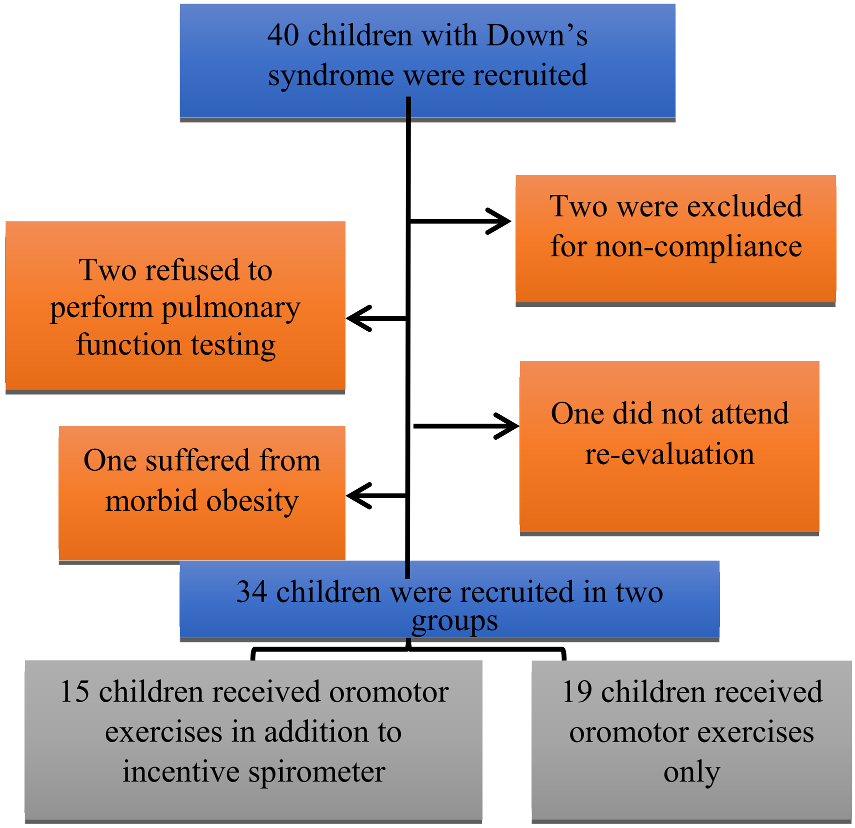

Figure 1: Flow chart.

Table 1: General characteristics of children.

Groups	General characteristics		
	Age (yrs) Mean ± SD	BMI (kg/m^2) Mean ± SD	IQ
Group A	7.9 ± 1.4	19.2 ± 2.4	55.2 ± 7.6
Group B	9 ± 1.9	19.4 ± 3.2	59.8 ± 9.1
t-value	−1.8	−0.14	−1.6
p-value	0.07.3	0.886	0.114

*SD: standard deviation, p: probability.

Table 2: Comparison between pre- and post-study values of orofacial myofunctional within and between groups.

Item	Pre-study Median (range)	Post-study Median (range)	z-value	p-value	% improvement
Total score					
Group A	77 (61−92)	85 (72−97)	3.59	0.001	10.4%
Group B	80 (70−93)	88 (77−104)	2.9	0.004	10%
z-value	−1.2	−1.1			
p-value	0.224	0.266			

*range: interquartile range.

Table 3: Comparison between pre- and post-study mean values of ruler measure within and between groups.

Ruler measure	Pre-study Median (range)	Post-study Median (range)	z-value	p-value	% improvement
Group A	1.1 (0.5−1.5)	0.5 (0.4−0.9)	−3.07	0.002	54.5%
Group B	1.2 (0.8−1.5)	0.5 (0.2−1.1)	−3.18	0.001	63.6%
z-value	−0.889	−0.527			
p-value	0.374	0.598			

Table 4: Comparison between pre- and post-study values of forced vital capacity, forced expiratory flow for 1 s, and peak expiratory flow within and between groups.

Item	Pre-study Median (range)	Post-study Median (range)	z-value	p-value	% improvement
FVC					
Group A	46 (38−79)	71 (65.6−91.5)	3.38	0.001	54.3%
Group B	67 (51.6−77)	83 (74.8−91.3)	3.4	0.001	23.8%
z-value	−0.954	−0.885			
p-value	0.340	0.376			
FEV1					
Group A	51.7 (40.2−90)	75 (60−95)	3.34	0.001	45%
Group B	78.8 (59−81)	89 (80−103)	2.86	0.004	12.9%
z-value	−1.3	−1.1			
p-value	0.193	0.245			
PEF					
Group A	40.2 (33−76)	62 (51−72)	2.7	0.006	54%
Group B	65 (54−72)	75.9 (58−92)	2.35	0.019	16.8%
z-value	−1.8	−1.6			
p-value	0.059	0.099			

23.8% in Group B. FEV1 improved by 45% in Group A and by 12.9% in Group B, while PEF showed a percentage improvement of 54% in Group A and that of 16.8% in Group B (Table 4).

Discussion

One of the major signs of Down's syndrome is hypotonia in the orofacial muscles, which causes a permanently open mouth. Patients with Down's syndrome also have a protruding tongue, which rests exposed on an everted lower lip; they also show dysfunction in mastication, deglutition, and speech.[11] In the current study, oromotor exercises improved oromotor functions in both groups, with no significant difference between the groups. Specifically, the exercises improved the appearance of the lips and tongue, indicating improved strength and muscle tone, and they improved the patients' ability over time to seal their lips tightly and retrude their tongue. These results confirm that the orofacial therapy by using neuromuscular stimulation are techniques consisting of physical therapy of the oral structures and appliance therapy for stimulating the tongue and lips. Their main aim was to improve muscle tone and thus promote healthy oral function and morphology. To achieve these goals, patients must accomplish a resting tongue position behind the incisors and correct their habit of opened mouth.[12] Oromotor therapy can significantly improve tongue posture at rest, tongue elevation strength, and tongue position during swallowing of solid food.[13]

In the present study, incentive spirometer training conferred no significant improvement on oromotor function. Renata et al.,[14] stated that the incentive spirometer can be used to train the inspiratory muscles, as well as to improve the strength of the expiratory muscles, expiratory flow, and lung capacity through expiratory training.

During swallowing, the hyolaryngeal complex is elevated to open the upper oesophageal sphincter. The muscles in this complex are essential to orofacial motoricity, and according to Troche et al.,[15] their function and mobility can be improved through training using an incentive spirometer. For this reason, we assumed in the present study that using the incentive spirometer requires tight lip sealing, and that tongue retrusion can improve the strength of these muscles, open-mouth posture, and tongue protrusion in children with Down's syndrome. Importantly, no previous research has addressed this point. Our results showed that the incentive spirometer did not significantly improve oral motor functions, but that oral motor exercises did have a significant effect, perhaps because muscle weakness is improved much more with repeated oromotor exercises, or because the children's intellectual disability led to improper use of the incentive spirometer, even though the researchers spent a long time familiarizing the children with the device, using different methods of demonstration and ensuring the children could keep their lips sealed for a long time around the mouthpiece.

In the current study, the incentive spirometer had no important role in improving pulmonary function in children with Down's syndrome. Multiple studies have identified that incentive spirometry can enhance pulmonary function (PEFR, FVC, FEV1, and maximal voluntary ventilation [MVV]) by increasing lung volume, decreasing resistance to air flow, expanding collapsed areas, and improving deep diaphragmatic breathing. Furthermore, the method gives visual feedback for diaphragmatic training.[16]

Khalili and Elkins[19] found that children with intellectual disabilities have decreased lung function, which may imply that intervention can improve lung function in this population and subsequently reduce respiratory infection. As only a few published studies have investigated pulmonary function in children with Down's syndrome, we evaluated the effect of incentive spirometer in such children in the present study. The results showed that post-treatment pulmonary function was significantly improved in both groups, but that there was no significant difference between the two groups, indicating that it was the oromotor exercises that improved lip sealing and nasal breathing, rather than the incentive spirometer training. In contrast, according to a study by Ahmed,[20] incentive spirometry can improve ventilation in children with

Down's syndrome by enhancing respiratory muscle strength and endurance, increasing the number of perfused alveoli, and maintaining positive pressure in the airways. In addition, that study found an increase in FVC, FEV1, PEF, and MVV. These differences may have occurred because the children in the present study were younger, or because the children in the Ahmed study received other modalities of chest physiotherapy while those in our study received only incentive spirometer and oromotor exercises.

The results of the present study showed that oromotor exercises improve pulmonary functions, including FVC, FEV1, and PEF. In one study by Cassir et al.,[21] a large number of patients acquired correct and physiological nasal breathing after oral motor therapy because they had gained increased labial muscle tone, labial seal at rest, and proper tongue position on the anterior palate. This study implied that oral breathing usually results from weak orofacial musculature and a low resting tongue position. The goal of oromotor therapy is to normalize the muscle patterns involved in oral movements during normal swallowing and nasal breathing, emphasizing lingual and labial muscle tone and mobility to maintain lip closure at rest. The treatment provides the patient with the essential conditions to maintain nasal breathing, which is essential for controlling respiratory diseases.

Study limitations

The intellectual disability of the children with Down's syndrome made it difficult for them to follow instructions, thus, we spent a long time teaching them to follow the instructions well.

Conclusion

Oromotor exercises improve both oromotor and pulmonary functions more than incentive spirometer training in children with Down's syndrome.

This study emphasizes that oromotor exercises should be applied to treat children with Down's syndrome to improve their oromotor and pulmonary functions, and that oromotor exercises are more effective than incentive spirometer training in this regard.

Recommendations

Future investigations should continue to study the effect of incentive spirometer training and oromotor exercises on pulmonary and oromotor functions in other populations, such as in those with cerebral palsy.

Source of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

No conflicts of interest to declare.

Ethical approval

The present study was approved by the Ethical Research Committee of the Faculty of Physical Therapy, Cairo University, Egypt (P.T.REC/012/001632).

Consent

The procedures of the present study were explained to the participant's parents, who signed consent forms before the study procedures were applied.

Authors contributions

AFI, F.H.A., and E.E.S. formulated the idea and implemented the study design; N.E.G. was responsible for the clinical evaluation of the children; AFI was involved in the clinical application of the intervention and in the writing of the manuscript; AFI and F.H.A. carried out the data analysis and interpretation; F.H.A., E.E.S., and N.E.G. revised the final draft; all authors confirmed the final article manuscript and were responsible for the content and similarity index of the manuscript. All authors have critically reviewed and approved the final draft and are responsible for the content and similarity index of the manuscript.

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