



Breathing pattern and muscle activity using different inspiratory resistance devices in children with mouth breathing syndrome

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In children with mouth breathing syndrome, adding inspiratory load using a nasal interface improves chest wall volumes and respiratory muscle activity, and a flow resistance device increases lung volume more than a pressure threshold device <https://bit.ly/3lzlayK>

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Abstract

Aim The aim of this study was to evaluate the acute effects of different inspiratory resistance devices and intensity of loads *via* nasal airway on the breathing pattern and activity of respiratory muscles in children with mouth breathing syndrome (MBS).

Methods Children with MBS were randomised into two groups based on inspiratory load intensity (20% and 40% of the maximal inspiratory pressure). These subjects were assessed during quiet breathing, breathing against inspiratory load *via* nasal airway and recovery. The measurements were repeated using two different devices (pressure threshold and flow resistance). Chest wall volumes and respiratory muscle activity were evaluated by optoelectronic plethysmography and surface electromyography, respectively.

Results During the application of inspiratory load, there was a significant reduction in respiratory rate ($p<0.04$) and an increase in inspiratory time ($p<0.02$), total time of respiratory cycle ($p<0.02$), minute ventilation ($p<0.03$), tidal volume ($p<0.01$) and scalene and sternocleidomastoid muscles activity (root mean square values, $p<0.01$) when compared to quiet spontaneous breathing and recovery, regardless of load level or device applied. The application of inspiratory load using the flow resistance device showed an increase in the tidal volume ($p<0.02$) and end-inspiratory volume ($p<0.02$).

Conclusion For both devices, the addition of inspiratory loads using a nasal interface had a positive effect on the breathing pattern. However, the flow resistance device was more effective in generating volume and, therefore, has advantages compared to pressure threshold.

Introduction

Mouth breathing is characterised by a shift in the physiological type of exclusively nasal breathing to oral or mixed breathing [1] for a period of >6 months [2]. It is more common in childhood and owing to its various causes is classified as mouth breathing syndrome (MBS) [3].

A modification from nasal to oral breathing will be accompanied by functional and morphological adaptive transformations [4] and determines changes in stomatognathic, musculoskeletal and respiratory systems [2]. Studies also report other MBS repercussions such as: learning deficits [5], nutritional imbalances [6], auditory loss [7], behavioural changes [8] and impairment of respiratory and exercise capacity [9].



Children with MBS have an anteriorisation of the head that generates an increase in thoracic kyphosis. This change in posture negatively influences the contraction of the diaphragm and abdominal muscles (*i.e.* reducing their ability to contract), impacting their synergism and leading to reduced chest wall expansion and weakness of the respiratory muscles [1, 2]. There is also failure in filtration, humidification and heating of the inspired air, which results in an excessive presence of leukocytes in the blood, therefore increasing the lungs' hypersensitivity and decreasing their volumes and capacity [10].

In a previous study, DE HELD *et al.* [11] performed respiratory muscle training using nasal breathing in children with MBS and reported improvement in respiratory muscle strength and nasal respiratory flow. However, these authors used simplistic methods of assessment and did not quantify lung volume. Recently, MEDEIROS DA FONSÊCA *et al.* [12] evaluated the acute effects of inspiratory loads using different interfaces (nasal and oral) in children with MBS and observed that the use of the nasal interface was more effective in increasing chest wall volumes and activating the respiratory muscles.

The application of inspiratory load aims to increase endurance and respiratory muscle strength in order to favour the respiratory system [13]. The following respiratory muscle training modalities have been reported in the literature: voluntary isocapnic hyperpnea, flow resistive loading and pressure threshold loading [14]. However, the specific impacts of each type of inspiratory load in the generation of lung volume are still not clear.

Considering the respiratory changes due to mouth breathing and the possible benefits of using inspiratory loads *via* the nasal airway, we hypothesised that applying the load using a nasal interface would improve the breathing pattern, the variation of chest wall volumes and the muscle activity of respiratory muscles in children with MBS.

The assessment of the diaphragm muscle (main respiratory muscle) using surface electromyography is uncertain due to its deep location, which makes it difficult to have the myoelectric signal captured superficially. Owing to this fact, the accessory respiratory muscles, scalene (SCL), sternocleidomastoid (SCM), intercostal (IC) and rectus abdominis (RA), were selected for assessment. Furthermore, this study was designed to also evaluate the responses of the aforementioned variables when subjected to different inspiratory resistance devices.

Methods

Type of study and subjects

This research was a cross-sectional study, and it was approved by the Hospital Research Ethics Committee (number 1.251.451/2015). The participants' legal guardians signed a consent form drafted according to the Helsinki Declaration.

Children with diagnosis of MBS assisted by the Otorhinolaryngology Outpatient Clinic of Onofre Lopes University Hospital/Brazilian Hospital Services Company (HUOL/EBSERH) were recruited for the study. Selected subjects were of both sexes, with ages from 6 to 13 years, body mass index classified according to the World Health Organization normality standard [15], and relation between forced expiratory volume in 1 s and forced vital capacity (FEV_1/FVC) >80% of predicted [16]. Subjects who failed to perform the tests or voluntarily removed themselves from the study were excluded.

Randomisation and study design

The sample was randomised and allocated into two groups by way of a simple draw. They were grouped according to the intensity of load applied, 20% or 40% of the maximal inspiratory pressure ($MIP_{20\%}$ and $MIP_{40\%}$, respectively). Assessments were carried out in two stages: 1) clinical, spirometric and respiratory muscle strength evaluation; and 2) chest wall volumes concomitant to the activity of respiratory muscles. During stage 2, the assessment consisted of three steps of 30 s each: 1) quiet breathing (QB); 2) breathing against inspiratory load using a nasal interface (Load); and 3) recovery (Rec) (figure 1). The nasal interface was used in all steps of stage 2. Each step of stage 2 was limited to 30 s so the optoelectronic plethysmography (OEP) and surface electromyography (sEMG) signals could be registered in a synchronised mode. The load intensities were applied using two different devices: pressure threshold and flow resistance. The order of application was also randomised following a simple draw. Participants were asked to breathe exclusively through the nose during all steps of this protocol.

Pulmonary function and respiratory muscle strength

Spirometry was performed using the KoKo DigiDoser® spirometer (NSPIRE Health, Longmont, CO, USA). The evaluations were performed according to the criteria of acceptability and reproducibility of the

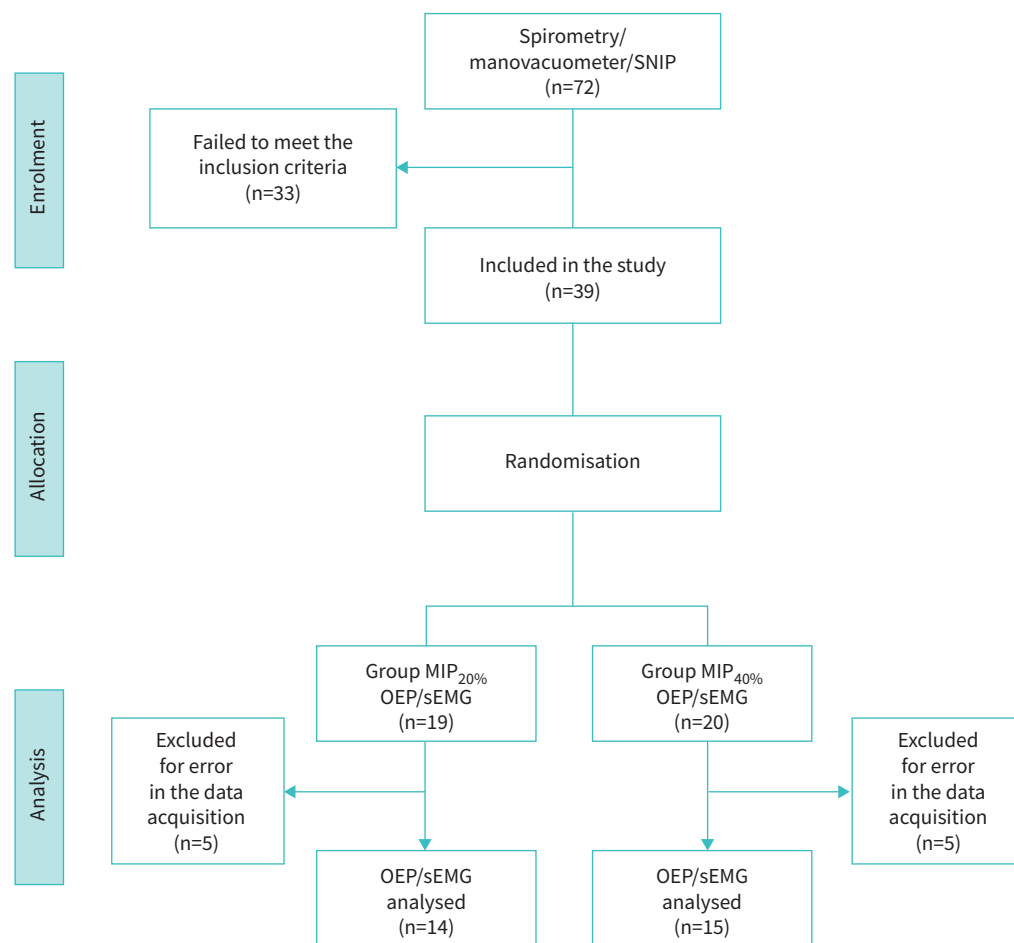


FIGURE 1 Study design. SNIP: sniff nasal inspiratory pressure; MIP: maximal inspiratory pressure; OEP: optoelectronic plethysmography; sEMG: surface electromyography.

American Thoracic Society/European Respiratory Society (ATS/ERS) [17] and the reference values derived according to the recommendations of the Brazilian Guidelines for pulmonary function test [16].

Respiratory muscle strength was determined by measuring maximal inspiratory pressure (MIP), maximal expiratory pressure (MEP) and sniff nasal inspiratory pressure (SNIP) using a digital manovacuometer (NEPEB-LabCare/UFGM, Belo Horizonte, Brazil). The evaluations were performed according to the ATS/ERS [17] acceptability and reproducibility criteria. Reference values previously published by LANZA *et al.* [18] were used for MIP and MEP, whereas the reference values of STEFANUTTI and FITTING [19] were used for the SNIP.

Optoelectronic plethysmography

The measurement of the volumes of the chest wall and its compartments, *i.e.* pulmonary rib cage (RC_p), abdominal rib cage (RC_a) and abdomen (Ab), was performed by OEP (BTS®, Milan, Italy) in which volumes were obtained following an experimental model according to the Gauss theorem [20]. Before each data acquisition, the equipment was calibrated at a frequency of 60 Hz. Six photosensitive cameras positioned around the subject (three in the anterior region and three in the posterior region) captured the movement variation of 89 reflexive markers fixed at specific points of the thorax [21].

From OEP data, the following variables for the chest wall and its compartments were analysed: tidal volume (tidal volume in chest wall ($V_{T,CW}$), tidal volume in pulmonary rib cage ($V_{T,RCp}$), tidal volume in abdominal rib cage ($V_{T,RCa}$) and tidal volume in abdomen ($V_{T,Ab}$)), end-inspiratory volumes (end-inspiratory volume in chest wall (EIV_{CW}), end-inspiratory volume in pulmonary rib cage (EIV_{RCp}), end-inspiratory volume in abdominal rib cage (EIV_{RCa}) and end-inspiratory volume in abdomen (EIV_{Ab})),

end-expiratory volumes (end-expiratory volume in chest wall (EEV_{CW}), end-expiratory volume in pulmonary rib cage (EEV_{RCp}), end-expiratory volume in abdominal rib cage (EEV_{RCa}) and end-expiratory volume in abdomen (EEV_{Ab})), respiratory rate, inspiratory time (T_I), expiratory time (T_E), inspiratory flow, expiratory flow, minute ventilation (V'_E) and total time of respiratory cycle (T_{TOT}).

Surface electromyography

The sEMG was performed following the recommendations of the International Society of Electrophysiology and Kinesiology (ISEK) [22]. Myoelectric signals from the SCL, SCM, IC and RA were recorded using the electromyographic TeleMyo DTS Desk Receiver® (Noraxon USA Inc., Scottsdale, AZ, USA) and four wireless sensors Clinical DTS (Noraxon USA Inc.) with 20–500 Hz pass filter-band, 1000 gain, 16-bit resolution and a common mode rejection rate >120 dB. Bipolar double-trace Ag/AgCl (Miotec, Porto Alegre, Brazil) passive surface self-adhesive electrodes were placed on these muscles: SCL at a distance of 5 cm from the sternum-clavicular joint and 2 cm above this point [23], SCM in the lower third of the distance between the mastoid process and the sternum-clavicular joint [24], IC over the second intercostal space and 3 cm from the sternum, and RA at 4 cm from the umbilical scar [25]. All electrodes were placed on the right side of the body to minimise electrocardiogram (ECG) interference. Before placing the electrodes, the skin region was shaved and cleaned with alcohol to reduce the impedance for capturing the electrical signal. Data acquisition and analysis were performed using the MR 3.2 (Noraxon, Inc.). Some filters were applied to process the obtained signals: 1) ECG-reduction algorithm to remove the cardiac electrical signal; 2) rectification of the full wave type to convert the negative signal to positive; and 3) root mean square (RMS) smoothing algorithm using a 50-ms window to eliminate non-reproducible signals. Raw data was analysed by means of RMS and normalised from respiratory baseline values [26].

Inspiratory load

The inspiratory loads were applied with a nasal interface using two different inspiratory resistance devices: 1) an inspiratory mechanical device providing a pressure threshold load (Threshold™ IMT; Philips Respironics, Inc., Murrysville, PA, USA); and 2) an electronic device with variable flow resistive load (POWERBreathe®, KH5; HaB International Ltd, Southam, UK).

Threshold™ IMT is a mechanical inspiratory pressure threshold device. The resistance is spring-imposed with a predetermined pressure, where sufficient inspiratory muscular effort is required to overcome the resistance and promote a valve opening for air passage. This is maintained as long as the pressure threshold is being reached. The POWERBreathe® KH5 is an electronically controlled variable flow resistance device, in which an absolute initial load is assigned and successively reduced depending on the inspiratory flow generated by the subject; its valve is adjusted dynamically (100 times per second), in real time, to accommodate within-breath changes in inspiratory flow rate.

Figure 2 demonstrates the variation of the tidal volume of the chest wall during the three evaluation steps (QB, Load and Rec) and during the use of both inspiratory resistance devices (pressure threshold and flow resistance).

Sample size and statistical analysis

Sample size was established considering the tidal volume as the main variable. Five subjects in each group (total of 10 subjects) were evaluated using hypothetical one-way ANOVA during all three steps (QB, Load and Rec). A sample size mean of 11 subjects per group was estimated using the following: an alpha error of 0.05 with bilateral distribution and a test power of 80%.

Data normality was verified using the Shapiro–Wilk test. The ANOVA one-way and Friedman tests were used to analyse the parametric and non-parametric variables respectively, for the steps of QB, Load and Rec. Dunn's *post hoc* was applied in the case of a significant difference. The comparisons between groups ($MIP_{20\%}$ and $MIP_{40\%}$) and between devices (pressure threshold and flow resistance) for non-parametric variables were performed using Mann–Whitney and Wilcoxon tests, respectively. The same comparisons were analysed using the multiple comparison ANOVA for repeated measures (*post hoc* Bonferroni) when variables were parametric. The power (β) and effect size were estimated and are detailed in the Results section of this study.

For data analysis, the GraphPad Prism 6.0 program for Windows (GraphPad Software, San Diego, CA, USA) was used. The sample size calculation, β and effect size of the study were calculated using GPower software version 3.1.9.2 (University of Düsseldorf, Kiel, Germany). For all statistical analyses, a level of significance $p < 0.05$ with bilateral distribution was adopted.

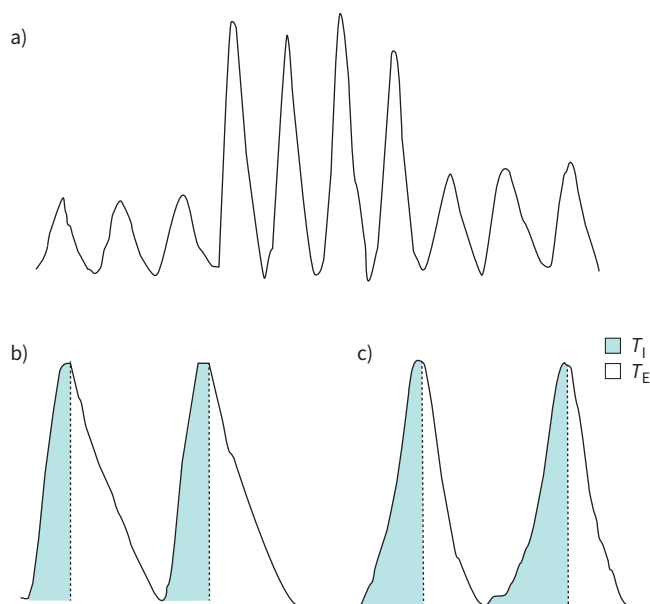


FIGURE 2 a) Demonstration of tidal volume variation during the periods of quiet breathing, use of inspiratory load and recovery. Comparison of volume curves during increment of load with b) pressure threshold and c) flow resistance devices. T_i : inspiratory time; T_E : expiratory time.

Results

We recruited 72 patients, 39 of whom were screened to participate in this study. 10 subjects were excluded due to low quality of data acquisition, resulting in a final sample of 29 subjects who were randomised and categorised into two groups ($MIP_{20\%}$: 14 and $MIP_{40\%}$: 15) (figure 1). The description and characterisation of the sample regarding clinical, spirometric and respiratory muscle strength data are shown in table 1.

The variables from optoelectronic plethysmography and sEMG are exposed as the variation from quiet spontaneous breathing (Δ Load-QB and Δ Rec-QB) and will be demonstrated below.

Chest wall volumes

As shown in figure 3, regardless of the imposed load, significant increases in the tidal volume of the chest wall ($V_{T,CW}$) were found during the Load step when compared to QB and Rec steps ($p < 0.01$). These changes occurred in the rib cage compartments (RCp and RCa) in the $MIP_{20\%}$ group and in all compartments in the $MIP_{40\%}$ group. The generation of $V_{T,CW}$ was greater when using the flow resistance

TABLE 1 Anthropometric, pulmonary function and respiratory muscle strength characteristics of both groups

Variable	$MIP_{20\%}$ group	$MIP_{40\%}$ group	p-value
Sex n (F/M)	7/7	9/6	NS
Age years	11±2	11.6±1.9	NS
Weight kg	35.8±9.7	40±10.5	NS
Height m	1.43±0.16	1.45±0.12	NS
BMI $\text{kg}\cdot\text{m}^{-2}$	17.2±2.26	18.8±2.26	NS
FVC % pred	103.1±13.3	107.9±12.7	NS
FEV ₁ % pred	95.3±12.9	99.6±10.2	NS
FEV ₁ /FVC % pred	91±4.9	91.9±6.6	NS
MIP % pred	80.8±18.3	87.1±16.5	NS
MEP % pred	81.7±18.2	89±10.5	NS
SNIP % pred	68.4±15.9 [#]	78.9±14.6 [#]	NS

Data are presented as mean±SD unless otherwise stated. MIP: maximal inspiratory pressure; F: female; M: male; NS: nonsignificant; BMI: body mass index; FVC: forced vital capacity; % pred: percentage of predicted value; FEV₁: forced expiratory volume in 1 s; MEP: maximal expiratory pressure; SNIP: sniff nasal inspiratory pressure. [#]: there is no predictive equation defined for SNIP in female children, so data are only shown for males.

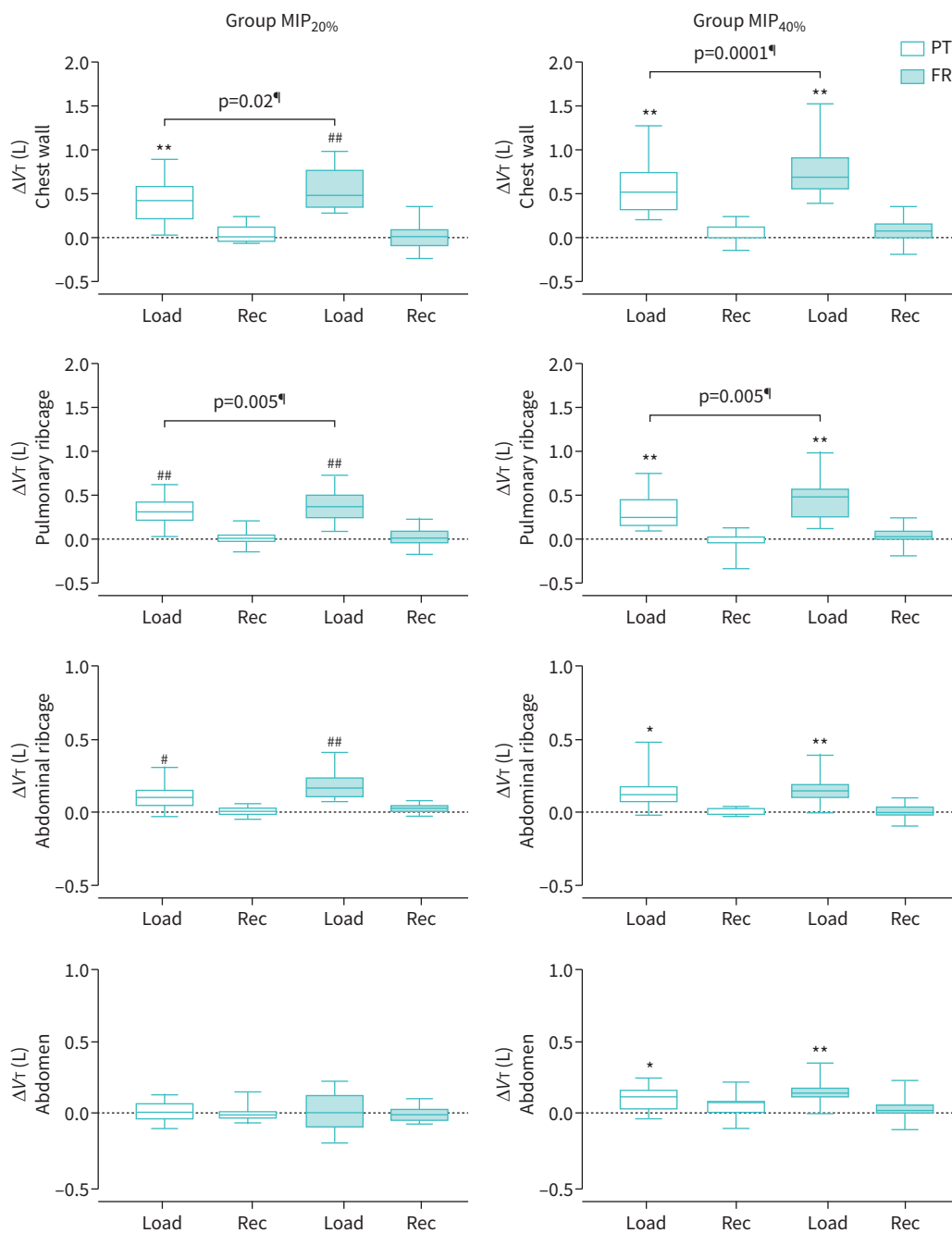


FIGURE 3 Variations of tidal volume of chest wall and its compartments. MIP: maximal inspiratory pressure; V_T : tidal volume; Load: breathing against inspiratory load; Rec: recovery; PT: pressure threshold; FR: flow resistance. *: $p < 0.05$, **: $p < 0.01$ for one-way ANOVA (*post hoc* Bonferroni); and #: $p < 0.05$, ##: $p < 0.01$ for Friedman test (*post hoc* Dunn's) for comparison between quiet spontaneous breathing, breathing against inspiratory load and recovery. ¶: $p < 0.05$ for Wilcoxon test in the comparison between flow resistance and pressure threshold devices.

versus pressure threshold device (MIP_{20%} $p = 0.02$ and MIP_{40%} $p = 0.01$). The MIP_{40%} group presented significant increase in the volume of abdominal compartment when compared to the MIP_{20%} group during the application of load in both devices (pressure threshold and flow resistance, $p = 0.01$).

Regarding the operational volumes, a significantly higher end-inspiratory volume (EIV_{CW}) ($p < 0.01$) was observed during the Load step when compared to QB and Rec regardless of load or type of device applied.

The rib cage compartments were the main determinants for this increase. In addition, a significant decrease in end-expiratory volume (EEV_{CW}) (pressure threshold, $p=0.01$ and flow resistance, $p=0.03$) was observed when 40% of load was imposed, and the abdomen compartment was responsible for this result. When comparing devices, the flow resistance device promoted a greater increase in EIV_{CW} compared to the pressure threshold device during load application (MIP_{20%}: $p=0.01$; MIP_{40%}: $p=0.02$), with no differences in EEV_{CW} observed in this comparison. The 40% of MIP load generated EEV_{CW} reduction when compared to 20% of MIP load in both devices (pressure threshold and flow resistance, $p=0.01$). Changes in operating volumes are shown in figure 4.

In both groups and devices, during inspiratory load, the breathing pattern showed a decrease of respiratory rate and an increase of total time of T_{TOT} , T_I , duty cycle and V'_E . During the recovery step these variables returned to baseline values. T_E did not present significant variations. Although not statistically significant, the use of the flow resistance device presented an additional increase of 0.51 s in T_{TOT} and 0.49 s in T_I compared to the pressure threshold device. These analyses are shown in table 2.

Electrical activity of respiratory muscles

Surface electromyography signals were normalised, and their representations are shown in figure 5. During Load step, the electrical activity of SCM and SCL muscles increased in comparison to QB and Rec steps ($p<0.01$). However, the IC muscle only showed this same behaviour with the intensity of 40% of MIP load ($p=0.01$). RA muscle did not present significant changes. There was no significant difference between inspiratory resistance devices. In the intergroup analysis, an increase in %RMS of the SCM (pressure threshold: 243.5% RMS, flow resistance: 291.8% RMS), SCL (pressure threshold: 100.6% RMS, flow resistance: 223.2% RMS), IC (pressure threshold: 39.8% RMS, flow resistance: 31.54% RMS) and RA muscles (30% RMS) was observed during loading of 40% versus 20% of MIP; however, this was not statistically significant.

Power and effect size

Table 3 summarises the effect size and power test for chest wall volumes and the sEMG RMS values of the SCM and SCL muscles during QB, Load and Rec steps. Table 4 shows the comparison between flow resistance and pressure threshold devices during the application of inspiratory load.

Discussion

We studied the acute effects of applying different inspiratory resistance devices and load intensities using a nasal interface on the breathing pattern variations and respiratory muscle activation in children with MBS. The main findings were: 1) acute inspiratory loading using a nasal interface increased chest wall volumes and respiratory muscle electrical activity; 2) the effects in chest wall volumes were more in evidence when using the variable flow resistance device compared to the pressure threshold device; and 3) confirmation of respiratory muscles weakness in children with MBS.

In order to evaluate the pulmonary function of children with MBS, we performed spirometry and measured maximal respiratory pressures, but unlike the results found in the literature [27], we did not find reduced values in our sample. However, we confirmed the respiratory muscle weakness of these subjects, a result supported by several authors [1, 9, 28]. Reduction of pulmonary volume is a characteristic of restrictive respiratory disorders and occurs as a consequence of decreased respiratory muscle strength. This leads to more superficial respiratory cycles and/or reduction of costal grid expandability, leading to impairment of pulmonary function. These changes contribute to the formation of atelectasis and retention of secretions, which predispose to respiratory tract infections, as well as may negatively reflect tolerance and exercise capacity due to hypoventilation.

Respiratory repercussions resulting from oral breathing persists in adulthood [29], impacting the respiratory system and functional exercise capacity. Thus, early diagnosis is important for proper intervention in order to avoid anomalies [30]. CUCCIA *et al.* [31] reported that when regularisation of this breathing occurs (from oral to nasal) until adolescence, normalisation of the craniocervical posture tends to follow.

The literature is limited regarding respiratory interventions in children with MBS. We found benefits on the breathing pattern and muscle activity when applying inspiratory load using a nasal interface. A protocol of respiratory muscle training and nasal breathing performed in children with MBS by DE HELD *et al.* [11] resulted in improvement of respiratory muscle strength, with MIP reaching its predicted values and also improvement in nasal respiratory flow. The authors explained the importance of nasal respiratory training for the re-establishment of lung volume and nostril elasticity. This promoted an increase of nasal airflow evidenced by greater nasal inspiratory flow peak values. BARBIERO *et al.* [32] observed

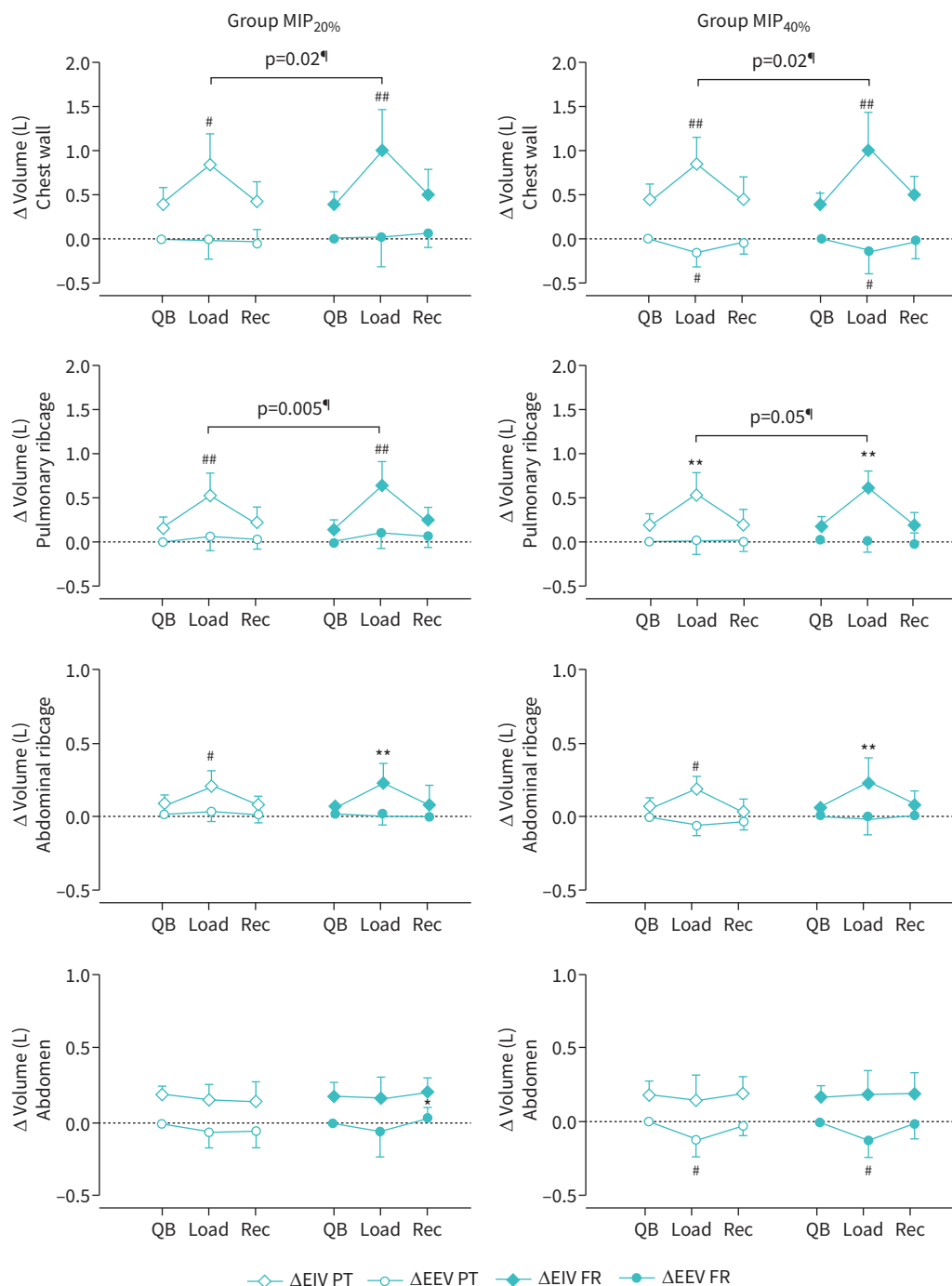


FIGURE 4 Variations in operational volumes of chest wall and its compartments. MIP: maximal inspiratory pressure; QB: quiet spontaneous breathing; Load: breathing against inspiratory load; Rec: recovery; ΔEIV: end-inspiratory volume variation; ΔEEV: end-expiratory volume variation; PT: pressure threshold; FR: flow resistance; *, p<0.05, **, p<0.01 for one-way ANOVA (*post hoc* Bonferroni); and #, p<0.05, ##, p<0.01 for Friedman test (*post hoc* Dunn's) for comparison between quiet spontaneous breathing, breathing against inspiratory load and recovery. †: p<0.05 for Wilcoxon test in the comparison between flow resistance and pressure threshold devices.

improvements in forced vital capacity, Tiffeneau score, respiratory muscle strength and daily habits in mouth breathers after therapy with re-expansive respiratory exercises associated with respiratory biofeedback. However, both studies used simplistic methodologies to evaluate pulmonary function and

TABLE 2 Time variables of respiratory pattern

	MIP _{20%} group		p-value	Δ FR/PT Load	MIP _{40%} group		p-value	Δ FR/PT Load	Δ 40%/20% Load
	Load	Rec			Load	Rec			
T_{TOT} s				0.49				0.53	
PT	1.77±1.78 (0.75–2.8)	−0.55±0.77 (−0.49–0.4)	0.002 [#]		2.16±2.51 (0.76–3.45)	0.42±1.10 (−0.19–1.02)	0.02 [¶]		0.39
FR	2.26±2.47 (0.83–3.68)	−0.21±0.65 (−0.58–0.16)	0.002 [#]		2.69±3.51 (0.75–4.67)	0.60±1.15 (−0.04–1.22)	0.02 [#]		0.43
T_I s				0.47				0.5	
PT	1.4±1.47 (0.55–2.24)	0.12±0.51 (−0.18–0.41)	0.01 [¶]		1.58±1.26 (0.58–1.97)	0.08±0.58 (−0.24–0.4)	0.001 [¶]		0.18
FR	1.87±1.5 (1–2.73)	−0.10±0.39 (−0.32–0.13)	0.002 [#]		2.08±2.4 (0.75–3.4)	0.24±0.92 (−0.27–0.75)	0.007 [#]		0.21
T_E s				0.01				0.02	
PT	0.38±0.85 (−0.11–0.87)	−0.16±0.56 (−0.49–0.16)	ns		0.58±1.57 (−0.3–1.45)	0.34±0.88 (−0.15–0.83)	ns		0.2
FR	0.39±1.2 (−0.32–1.09)	−0.11±0.57 (−0.44–0.22)	ns		0.60±1.57 (−0.27–1.47)	0.35±0.76 (−0.07–0.78)	ns		0.21
V_E L·min^{−1}				1.55				1.55	
PT	2.83±2.72 (1.26–4.4)	0.74±1.69 (−0.24–1.71)	0.006 [#]		4.87±5.15 (2.01–7.72)	0.29±2.37 (−1.02–1.6)	0.004 [#]		2.04
FR	4.38±7.34 (1.2–9.1)	0.40±2.16 (−0.85–1.65)	0.03 [#]		6.42±7.74 (2.14–10.7)	0.22±2.30 (−1.05–1.49)	0.001 [#]		2.04
RR bpm				1.35				0.81	
PT	−6.46±10 (−8.42–−3.5)	0.22±2.81 (2.76–1.51)	0.003 [¶]		−5.74±8.54 (−9.83–−1)	−2.13±8.74 (−3.64–0.81)	0.02 [#]		0.18
FR	−5.11±4.51 (−11.4–1.49)	−0.18±3.60 (−0.05–2.12)	0.007 [#]		−4.93±7.42 (−9.68–−0.8)	−0.68±4.03 (−0.05–2.49)	0.04 [#]		0.72

Data are presented as mean±SD (95% CI), unless otherwise stated. PT: pressure threshold; FR: flow resistance; MIP: maximal inspiratory pressure; T_{TOT}: total time of respiratory cycle; T_I: inspiratory time; T_E: expiratory time; ns: nonsignificant; V_E: minute ventilation; RR: respiratory rate; bpm: breaths per minute. [#]: p<0.05 one-way ANOVA (*post hoc* Bonferroni); [¶]: p<0.05 Friedman test (*post hoc* Dunn's) for comparison between quiet spontaneous breathing, breathing against inspiratory load (Load) and recovery (Rec).

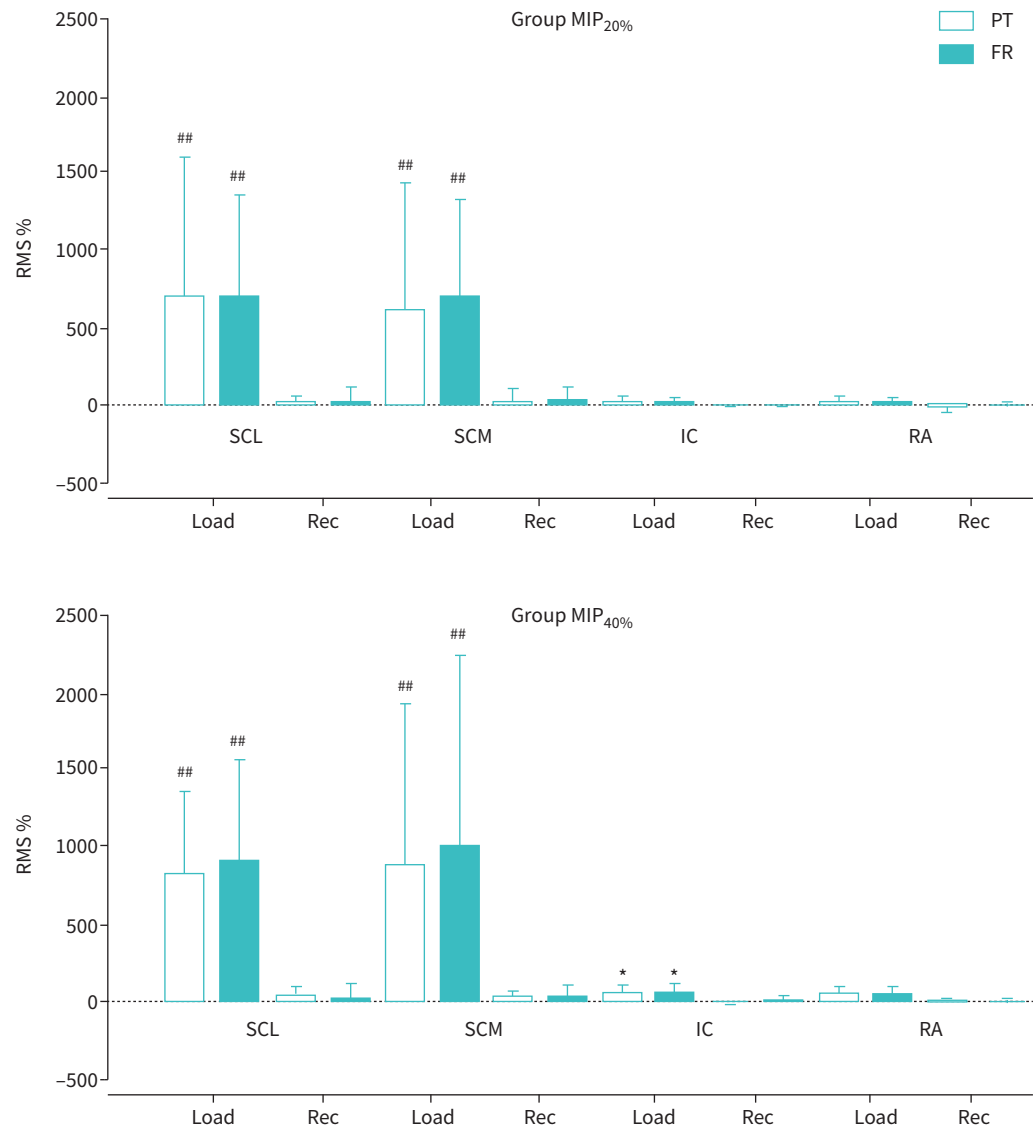


FIGURE 5 Percentage of respiratory muscle activation. MIP: maximal inspiratory pressure; RMS: root mean square; PT: pressure threshold; FR: flow resistance; Load: breathing against inspiratory load; Rec: recovery; SCM: sternocleidomastoid; SCL: scalene; IC: parasternal portion of the intercostal; RA: abdominal rectum. *: $p < 0.05$ for one-way ANOVA (*post hoc* Bonferroni); #: $p < 0.01$ for Friedman test (*post hoc* Dunn's) for comparison between quiet spontaneous breathing, breathing against inspiratory load and recovery.

were not clear regarding the effects on respiratory muscle activity. MEDEIROS DA FONSÉCA *et al.* [12] observed an increase in V_T and EIV, in addition to an increase in the activity of the SCL and SCM muscles, during the use of acute inspiratory loads in children with MBS, and these effects were more effective with the use of a nasal interface compared to an oral one.

The application of inspiratory loads for muscle training has the potential to improve respiratory muscle strength and endurance. We observed that the application of acute inspiratory loads promoted an increase of $V_{T,CW}$ due to the recruitment of rib cage compartments (RCp+RCa) in lower intensity loads (20% of the MIP) and recruitment of all compartments in higher loads (40% of the MIP). Tidal volume is the product resulting from the variation between operational volumes (EIV and EEV), which behaved differently depending on the intensity of load imposed. Initially, at lower intensities, the volume generation was entirely due to EIV expansion in the rib cage. And, at higher load levels, there was an additional increase of the EEV in the abdomen.

TABLE 3 Effect size and power test for comparison of chest wall volumes and percentage of respiratory muscle activation intensity between quiet spontaneous breathing, breathing against inspiratory load and recovery periods

	MIP _{20%} group		MIP _{40%} group	
	Effect size partial Eta η_p^2	Power	Effect size partial Eta η_p^2	Power
V_T				
Pressure threshold	0.74	0.99	0.80	0.99
Flow resistance	0.86	0.99	0.90	0.99
EIV				
Pressure threshold	0.68	0.98	0.63	0.97
Flow resistance	0.79	0.99	0.74	0.99
EEV				
Pressure threshold	0.1	0.13	0.1	0.14
Flow resistance	0.24	0.31	0.28	0.42
SCM				
Pressure threshold	0.42	0.63	0.43	0.7
Flow resistance	0.6	0.93	0.48	0.79
SCL				
Pressure threshold	0.43	0.66	0.73	0.99
Flow resistance	0.57	0.89	0.7	0.99

MIP: maximal inspiratory pressure; V_T: tidal volume; EIV: end-inspiratory volume; EEV: end-expiratory volume; SCM: sternocleidomastoid; SCL: scalene.

TABLE 4 Effect size and power test for comparison between flow resistance and pressure threshold devices during inspiratory load

	MIP _{20%} group		MIP _{40%} group	
	Effect size Cohen's d_z	Power	Effect size Cohen's d_z	Power
V_{T,CW}	6.5	0.99	2	0.99
EIV	1.36	0.99	1.3	0.99
EEV	0.2	0.11	0.2	0.11
SCM	0.07	0.06	1.19	0.99
SCL	0.57	0.51	0.94	0.92

MIP: maximal inspiratory pressure; V_{T,CW}: tidal volume in chest wall; EIV: end-inspiratory volume; EEV: end-expiratory volume; SCM: sternocleidomastoid; SCL: scalene.

During the application of 20% of MIP load, we found a constant EEV in all compartments and increased EIV values only in the rib cage compartments, indicating that the increase in V_{T,CW} at this load level was exclusively due to the action of inspiratory muscles. On the other hand, when 40% of MIP load was applied, we still found EIV increased in the rib cage compartments, but EEV was constant only in the rib cage and reduced in the abdomen. This behaviour is responsible for the reduced EEV_{CW} and indicates that both the rib cage and abdominal muscles contributed to increase V_{T,CW} at greater demands of work.

The reduction of EEV in the abdomen associated to its constant value in the rib cage is a mechanism favourable to the action of the diaphragm muscle. It increases the diaphragm pre-inspiratory length and prevents its excessive shortening during inspiration. Similarly, EIV increased in the chest wall and constant in the abdomen facilitates the mechanics of chest wall inspiratory muscles, increasing their shortening during inspiration and preventing their excessive pre-inspiratory stretching [33]. ROMAGNOLI *et al.* [34] and ALIVERTI *et al.* [33] evaluated hypercapnic stimulation and exercise, respectively, in healthy subjects and found variations in chest wall kinematics similar to our studies. They concluded that thoracic and abdominal muscles are used as a pressure generator and that coordinated action of these muscles minimises the pressure produced by the diaphragm, which then acts primarily as a flow generator.

A more efficient ventilatory pattern during load imposition was also observed in our study, regardless of load level applied or device used. We found an increase in T_I which is related to the lung filling time and

promotes an increase in EIV. We also found an increase in T_E ; this works as an effective mechanism to allow sufficient time to empty the lungs avoiding EEV increase, and thus not causing dynamic hyperinflation. Lastly, we found a respiratory rate reduction as a result of longer respiratory cycles due to T_{TOT} increase, which facilitates the ventilation.

The sEMG helped us to interpret the results. There was an increase in inspiratory muscle activity simultaneously to an increase in lung volumes. When the 20% of MIP load was imposed, the SCM and SCL muscles were recruited to generate volume in the rib cage. In the 40% of MIP load, additional activation of IC muscle occurred and, although not significantly expressed, the RA muscle presented an increase of 30% RMS in its activity compared to the MIP_{20%} group. This result reinforces that at initial loads only the rib cage muscles are responsible for the volume generation, whereas at higher loads, the abdominal and rib cage muscles act together. HAWKES *et al.* [35] evaluated the acute effects of submaximal inspiratory load on MIP, diaphragm and intercostal activation of healthy subjects. Similarly to our results, they found incremental responses in muscle recruitment. They identified this behaviour as a way to optimise respiratory muscle synergy for strength generation.

In our study two different inspiratory resistance devices were used: pressure threshold and flow resistance. The flow resistance device generated greater lung volume when compared to the pressure threshold, although the electromyography did not present any difference between devices. HOSTETTLER *et al.* [36] refute our results, finding no difference in lung volume variation between pressure threshold and flow resistance devices when evaluating healthy subjects. However, LANGER *et al.* [37] evaluated the effect of inspiratory muscle training with pressure threshold and variable flow resistance in COPD patients and concluded that subjects who used the flow resistance device tolerated higher training loads and obtained greater improvements in inspiratory function when compared to pressure threshold. In another recent study WALTERSPACHER *et al.* [38] evaluated inspiratory muscle activity in three modes of inspiratory muscle training: pressure threshold, inspiratory flow resistive load and voluntary isocapnic hyperpnea in healthy adult subjects, and their results corroborate with ours when presenting similar respiratory muscle activation between pressure threshold and flow resistance devices.

LANGER *et al.* [37] associated the better results using a flow resistance device with its ability to monitor the unsupervised training sessions and to ensure that subjects executed the sessions satisfactorily. However, this hypothesis is refuted by our study, since in our protocol both interventions were supervised. Another explanation proposed by these authors is related to the differences in the loads applied. The flow resistance device is an electronic valve that adjusts dynamically and in real time to the inspiratory flow being generated. Its mechanism reduces the initial load in order to promote a better volume–pressure relation of inspiratory muscles. Therefore, it maintains a resistance throughout the inspiratory period, allowing the EIV to get closer to total lung capacity.

In addition, we observed a better respiratory pattern during the use of a flow resistance *versus* pressure threshold device. We found higher values of T_{TOT} and particularly of T_i , proving that flow resistance promotes longer inspiratory periods, which are responsible for the increase in EIV. This could be associated with the different inspiratory effort performed with each device. With a pressure threshold, the inspiratory flow starts only after isovolumetric contraction of the inspiratory muscles, and it is only maintained while they are capable of generating the predetermined threshold pressure. Conversely, with variable flow resistance, the inspiratory flow is ongoing throughout the inspiratory effort, varying its intensity as the volume–pressure relationship of the inspiratory muscles. This mechanism allows the muscles to maintain the work of breathing during the entire inspiratory phase resulting in increased pulmonary volume.

As a limitation of this study, we point out the choice of the RA muscle to measure abdominal activity by sEMG. This may have masked our results, since this muscle is located under an adipose layer, making it difficult to capture the signal. However, our results add new perspectives regarding the application of inspiratory loads using a nasal interface, considering the increase in lung volume and inspiratory muscle activity achieved in children with MBS. Furthermore, the generation of pulmonary volume was more effective when the flow resistance device was applied. Therefore, the application of this loading modality favours the respiratory system, given that an increased pulmonary volume is a protective factor against respiratory tract infections. We suggest that longitudinal studies should be carried out to identify the effects of inspiratory muscle training *via* nasal airway in children with MBS.

Conclusion

The results of this study suggest that the application of inspiratory loads using a nasal interface has a positive effect on the breathing pattern of children with MBS. In both devices, we observed an increase in

chest wall volume without hyperinsufflation and an increase in the electrical activity of the inspiratory muscles. However, the flow resistance device was more effective in generating volume and thus has some advantages compared to the pressure threshold device. Therefore, the use of flow resistance modality favours functions in children with MBS by increasing chest wall volume.

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Conflict of interest: J.D. Medeiros da Fonsêca has nothing to disclose. A. Aliverti is co-inventor of optoelectronic plethysmography, the patent rights to which are held by his institution, the Politecnico di Milano. K. Benício has nothing to disclose. V.S. de Farias Sales has nothing to disclose. L. Fontes Silva da Cunha Lima has nothing to disclose. V.R. Resqueti has nothing to disclose. G.A. de Freitas Fregonezi has nothing to disclose.

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