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Acute Effects of Air Stacking Versus Glossopharyngeal Breathing in Patients with Neuromuscular Disease

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Authors' contributions

This work was carried out in collaboration between all authors. Author RTC designed the study, wrote the protocol and wrote the first draft of the manuscript. Authors RVU, LV and HP managed the literature searches, data collection and analysis. Author JV wrote the manuscript. All authors read and approved the final manuscript.

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ABSTRACT

Objective: To compare the effects of Air Stacking (AS) and Glossopharyngeal Breathing (GPB) on the Maximum Insufflation Capacity (MIC) in patients with Neuromuscular Disease (NMD).
Methods: We design a randomized cross-over study. Children and adolescents with NMD who were users of non-invasive mechanical ventilation were recruited. Vital capacity (VC) and MIC were measured before and after the intervention with AS and GPB. Values were compared pre- and post-intervention and were considered statistically significant if $p < 0.05$.
Results: We selected 14 patients with a median age of 12.5 years (range 9-18) with the following diagnoses: Duchenne Muscular Dystrophy (7), Spinal Muscular Atrophy Type II (3), Spinal Cord

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Injury (1) and Congenital Myopathies (3). The median baseline VC was 1325 ml (CI 1084-1594 ml). AS improved the VC to 1930 mL (CI 1630-2434 mL, p<0.001), and GPB increased the VC to 1600 mL (CI 1370-1960 mL, p=0.001). There was a significant difference of 290 mL (CI 168-567 mL, p<0.002) between both techniques.

Conclusion: The air stacking and glossopharyngeal breathing were both effective in increasing the maximum insufflation capacity, but air stacking resulted in a greater increase in MIC.

Keywords: Neuromuscular diseases; maximum insufflation capacity; vital capacity; air stacking; glossopharyngeal breathing.

1. INTRODUCTION

In neuromuscular diseases (NMD), progressive weakness of the respiratory muscles causes changes in the cough mechanism and prevents the elimination of secretions, which is the primary cause of morbidity and mortality in this population [1].

Improving the cough response and achieving adequate secretion removal is required to increase lung volume and optimize the peak cough flow (PCF) [2]. It has been shown that a high PCF decreases respiratory complications in NMD [3]. Vital capacity (VC) and maximum insufflation capacity (MIC) are the most important factors in increasing the PCF and, consequently, obtaining a more efficient cough [2]; thus, optimizing the lung volumes to achieve an adequate cough flow is essential for effective bronchial hygiene. Currently, manual techniques are used to increase the MIC to produce an effective PCF [4].

The most important manual techniques used to improve the MIC and, consequently the PCF, are air stacking (AS) and glossopharyngeal breathing (GPB). These have the advantages of low cost, patient autonomy and relative ease of learning compared to other techniques using electromechanical devices (e.g., volumetric ventilators and mechanical cough assist devices), whose main disadvantages are the high cost and complexity of use [5]. AS involves delivering multiple breaths into a manual resuscitation bag and holding the insufflation volumes with the momentary closure of the glottis, which is repeated until the MIC is reached [6]. GPB uses the glossopharyngeal muscles to introduce successively small amounts of air into the lungs to induce coughing and assist the weak inspiratory muscles [7]. These techniques have been useful in treating different NMD, such as Spinal Muscular Atrophy (SMA) Type II, Duchenne Muscular Dystrophy (DMD) and Spinal Cord Injury (SCI) [5,8,9].

The aim of this study was to compare the effects of two low-cost techniques, AS and GPB, on the MIC in children and adolescents with NMD. The hypothesis of this investigation was that although both techniques are effective, using AS results in a higher MIC than GPB.

2. METHODS

We design a randomized cross-over study. Child and adolescent beneficiaries of domiciliary programs of noninvasive ventilation in Santiago, Chile, were selected. The inclusion criteria were a diagnosis of NMD, age between 5 and 18 years without respiratory exacerbation in the past 30 days, no prior use and no knowledge of the AS or GPB techniques, ability to understand instructions and signed consent to participate. Patients were excluded from the study if they were tracheostomized or had incomplete glottis closure. Glottis closure was measured during the initial insufflation maneuver for the total lung capacity with a ventilometer (FERRARIS Wright® MK 8. Louisville, USA) connected online. Patients who exhibited expiratory flow during the maneuver were excluded. The selection process of patients is explained in the flowchart (Fig. 1).

2.1 Pulmonary Function Measurements

Spirometry data were collected using the ndd EasyOne Spirometer (ndd Medical Technologies, Zurich, Switzerland), which was chosen for its portability and level of accuracy. The percentual and absolute values reported were the FEV_1, FVC, as well as the FEV_1/FVC. To measure MIP and maximal expiratory pressure (MEP), an analog vacuum manometer (DHD Healthcare, New York, NY, USA) was used, calibrated from -120 to 0 cmH_2O and from 0 to 120cmH_2O. The interface used was a rigid plastic flanged mouthpiece. To prevent air leakage, a nose clip was used. The pressure was registered with the system occluded at total pulmonary capacity for MEP and residual volume for MIP. The record of the peak pressure obtained after the first second following initiation of the forced maneuver and sustained for at least 1 second was used for analysis. The results of spirometry and respiratory muscle pressures were expressed as a percentage of the reference value [10,11].

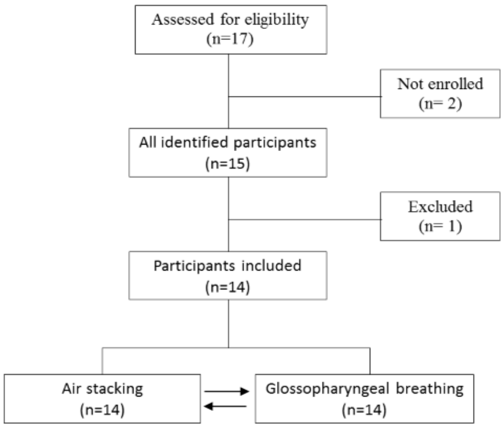

Fig. 1. Flowchart of subject selection

Baseline vital capacity (VC) and maximum inspiratory capacity (MIC) for each maneuver, were evaluated using a portable ventilometer device (FERRARIS Wright® MK 8. Louisville, CO, USA), to assess volumes, often used in cough assistance studies. Baseline VC was measured by a maximal inspiration followed by a maximal expiration. Ten minutes later, we realized the first maneuver (AS or GPB), and the MIC was measured (depending on the randomization), and 40 minutes later we realized the second maneuver, and the MIC was again measured (Fig. 2). To minimize the risk of bias, the order of execution of each cough assistance technique was performed randomly for each participant. Randomization was performed by a free software (for www.randomization.com) specifically designed for generating random number lists.

AS was performed with the subject seated in his wheelchair using a manual resuscitation bag (LIFESAVER® model 5345, Hudson, Temecula, USA) connected to a corrugated tube with an internal diameter of 22 mm, a one-way valve, and a pipette. The maximum capacity of the bag was 1600 mL. A chest physiotherapist insufflated the patient during the inspiratory phase, requesting that they inspire as much air as possible. GPB was also performed with the subject seated in his wheelchair and performing successive maneuvers of "swallowing air" until the maximum volume achieve was maintained. Then, the patient was instructed to breathe through ventilometer to register the MIC. Three measurements for each of the techniques were performed, and the highest reading was recorded. A difference of $\leq$10% between the measurements was used as the repeatability criterion. The results were expressed as a percentage of the reference value [10]. Between each assessment, the patients rested for 5minutes. Before the completion of each maneuver, the patient was trained in the execution of each technique for 10 minutes. All evaluations were made on the same day.

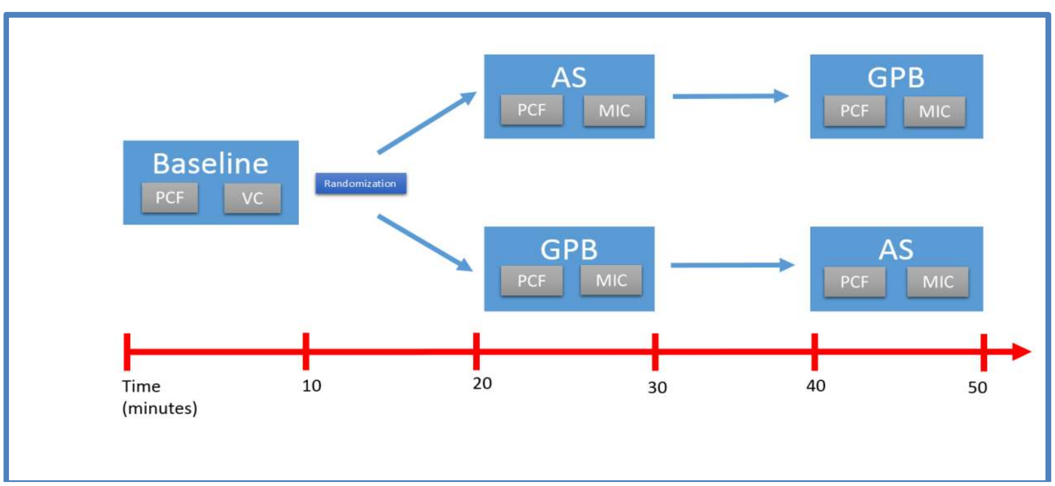

Fig. 2. Schematic representation of the intervention

VC: Vital Capacity; PCF: Peak Cough Flow; MIC: Maximum Insufflation Capacity; GPB: Glossopharyngeal Breathing; AS: Air Stacking

Baseline PCF was assessed using a peak flow meter (MiniWright®, Clement Clarke International, Essex, England) graduated in L/min. Baseline PCF assessment was performed with the patient seated and with a respiratory physiotherapist monitoring the seal between the entire surface of the mask and the flowmeter to avoid any leakage. The subject was instructed to cough as hard as possible into the flowmeter connected to his/her mouth. For an evaluation to be considered repeatable, we obtained, at least, three efforts with a difference ≤ 10% and recorded the highest value as the PCF. Ten minutes later, the PCF was measured after AS and GPB. To minimize the risk of bias, the order of execution of each cough assistance technique was performed randomly for each participant.

The protocol was implemented in the patients' homes by a trained chest physiotherapist [4]. All devices were new, and they had the manufacturer calibration. The informed consent was signed by the child's parents, who also gave informed verbal assent. The research project was approved by the Ethics Committee for Research Involving Human Beings, Faculty of Medicine, University of Chile.

2.2 Statistical Analysis

The statistical analysis was performed using STATA 11.1 software (Stata Corp. College Station, USA). The descriptive statistics was presented in mean ± standard deviation or median and interquartile range depending on distribution. The Shapiro-Wilk test was used to evaluate the normality of the sample. Student's t-test was used for data with a normal distribution and the Wilcoxon test for nonparametric data. A p-value of <0.05 was considered to be statistically significant.

For the sample size calculation, we referenced a similar article that assessed the MIC in SCI patients undergoing an assisted cough protocol. We accepted an alpha risk of 0.05 and a beta risk of 0.2. Thirteen subjects were required to detect a difference ≥500 mL. A standard deviation of 350 mL was assumed. We estimated a loss-to-follow-up rate of 30% [12]. In addition, a post hoc power calculation based on the number of participants was calculated using the statistical software of G*Power [13].

3. RESULTS

We evaluated 14 patients with NMD (8 men and 6 women) with a median age of 12.5 years (range 9-18 years). The patients had the following diagnoses: 7 with DMD, 3 with SMA type II, 2 with Nemaline Myopathy, 1 with SCI (with a complete lesion at C5) and 1 with Centronuclear Myopathy. All of the patients were using nocturnal non-invasive ventilation in bilevel mode (Table 1).

Table 1. Baseline patient´s characteristics

Age (years)	**12.4±2.6 (range 8 – 18)**
Age at diagnosis (years)	9.9±3.4 (range 2-16)
Duration of NIV (months)	25.6±8.7 (range 14-40)
FVC (ml)	1529±517
FVC (% Reference value)	62.2±31.9
FEV_1 (ml)	1243±502
FEV_1 (% Reference value)	64.1±31.4
FEV_1/FVC	82.7±15.5
MIP (cmH_2O)	57.5±10.8
MIP (% Reference value)	57.9±13.9
MEP (cmH_2O)	40.7±22.2
MEP (% Reference value)	31.5±21.3
Use of Wheelchair	14/14

NIV: Noninvasive Ventilation; FVC: Forced Vital Capacity; FEV_1: Forced Expiratory Volume in one second; MIP: Maximal Inspiratory Pressure; MEP: Maximal Expiratory Pressure

The median baseline VC was 1325 mL (CI 1084-1594 mL), which increased after both maneuvers with a MIC of 1930 mL (CI 1630-2434 mL) after AS and 1600 mL (CI 1370 -1960) with the GPB technique. Both techniques demonstrated statistically significant differences from the baseline VC with a value of p <0.001. The median MIC obtained using AS was 290 mL (CI 168-567 mL) greater than that achieved using GPB and had a statistically significant difference with p =0.002 (Fig. 3).

The median baseline PCF was 175 L/min (CI 130-200 L/min). The PCF increased to 220 L/min (CI 178-278 L/min, p <0.001) after AS and to 195 L/min (CI 158-267 L/min, p =0.017) after GPB (Fig. 4). Both techniques showed significant differences from the baseline, but no between them.

4. DISCUSSION

Our results suggest that in the studied patients, the low-cost techniques GPB and AS are able to generate significant changes in the MIC in children and adolescents with NMD with complete glottis closure.

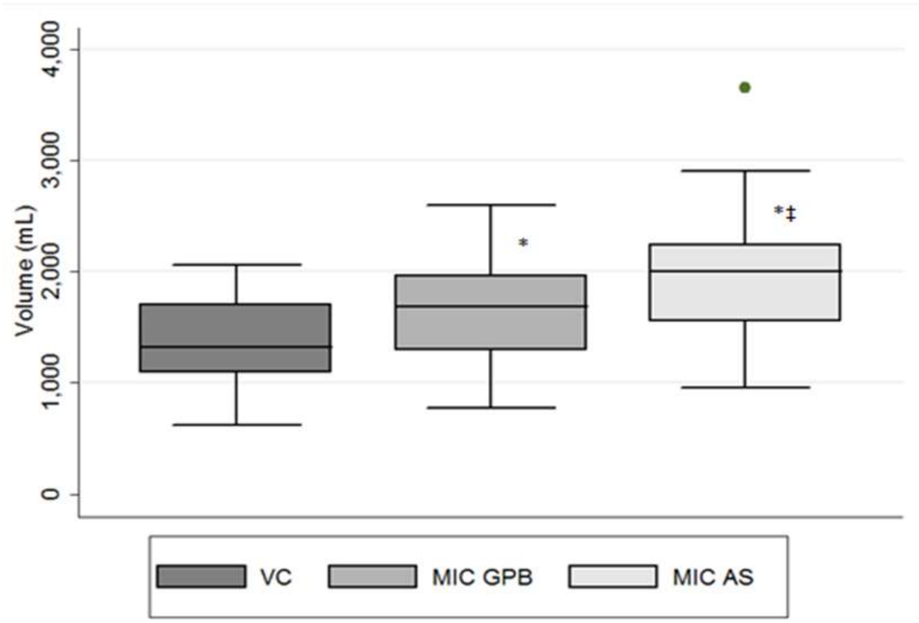

Fig. 3. Vital capacity and maximum insufflation capacity

*Boxplot graph with values obtained for each maneuver. VC: Vital Capacity; MIC: Maximum Insufflation Capacity; GPB: Glossopharyngeal Breathing; AS: Air Stacking; *VC vs MIC GPB or MIC AS: p < 0.001; ‡MIC GPB vs MIC AS: P < 0.01*

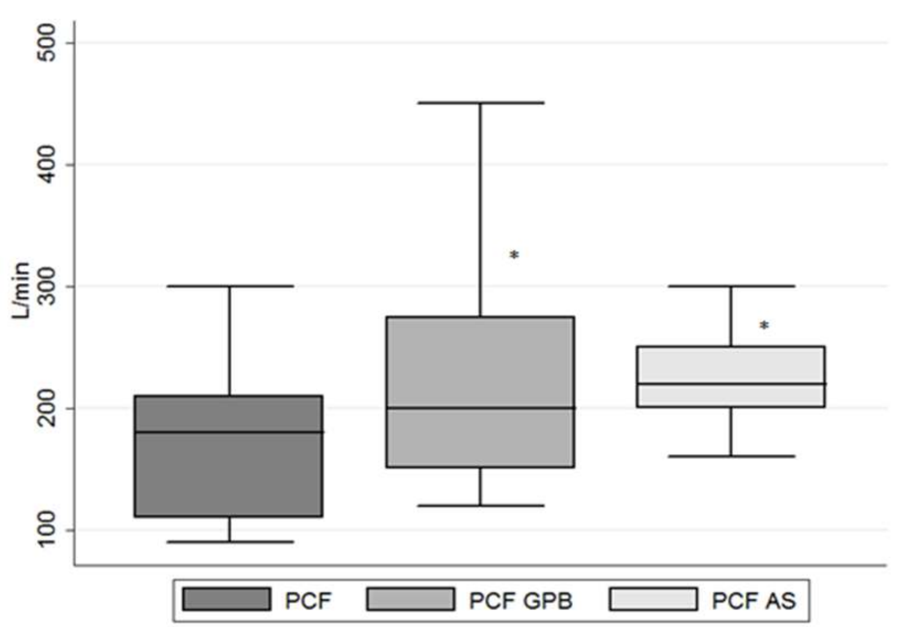

Fig. 4. Peak cough flow

*Boxplot graph with the values of Peak cough flow (PCF) obtained for each maneuver. GPB: Glossopharyngeal Breathing; AS: Air Stacking; *PCF vs PCF GPB or PCF AS: p<0.02*

In the studied patients, the best results were obtained with the AS maneuver. Probably, because GPB may be more difficult to learn and implement, especially due to the active role of the patient when inhaling air. In AS, inflation is performed by an outside operator, and the patient has a passive role in the execution of the technique.

In respect of the AS, our results show an increase in MIC and the PCF. Despite being an acute intervention, the results are consistent with other studies that also held a similar methodology. For example, Brito and coworkers observed in a study of 28 DMD patients with FVC <60% of predicted, with nocturnal non-invasive mechanical ventilation, a significantly increased of the baseline PCF from 171±67 L/min to 225±80 L / min with the use of AS technique [14]. These results have been replicated in long-term protocols, and they have even shown improvements in other parameters such as forced vital capacity, which justifies the use of these techniques to prevent respiratory complications [15].

In respect of the GPB, Nygren-Bonnier performed a protocol of series of 10 cycles, four times a week for eight weeks, in 25 patients with cervical SCI. They observed a significant improvement in the VC by 25% [9]. Furthermore, these authors, in a group of 11 children with SMA type II applied the same protocol and demonstrated positive effects increasing VC and chest expansion significantly [8].

Another similar study in patients with DMD by Bach et al. [5] shows that both AS and GPB have a positive effect on MIC, despite the progressive reduction of VC characteristic of this disease. Our protocol included only one session, whereas Bach used a number of long-term cases and demonstrated positive results with an increasing MIC. These results were consistent with the data obtained in our study that proved the therapies were effective in the first session. Furthermore, Bach also used cycled volume ventilators [4]. Our sample used only a manual resuscitation bag, which resulted in a considerable reduction in costs.

The literature has reported the usefulness of these techniques in increasing the MIC in NMD patients. Kang and Bach [6] conducted a study in which patients with DMD, amyotrophic lateral sclerosis (ALS), SMA and post-polio syndrome were subjected to a program of insufflation with AS to a maximum of 10-15 breaths three times daily for six months. Most subjects increased their MIC by 22%. These results agree with those obtained in our study and confirm that both techniques are effective in increasing the MIC and thus improving the effectiveness of a cough.

At baseline, our sample presented with a VC mean 47% of the reference value. After AS, VC exceeded 75% of the reference value. These results suggest that after AS, the VC can reach values close to normal, which will slow the progression of a restrictive pattern [5]. Additionally, Rochester found that effective cough-facilitation techniques should be part of NMD pulmonary rehabilitation programs and should be evaluated for their usefulness for other diseases [16].

Recently, a group of respiratory management experts for NMD patients [16] recommended the routine evaluation of the cough and the application of techniques designed to increase the PCF as essential elements in the treatment of patients with NMD. The recommendation emphasizes AS with a grade of 1B for patients with VC of less than 80%, a common feature found in our sample. While our population consisted of only of patients who could perform glottic closure, the AS technique can also be performed in patients with a glottal dysfunction [16], which is another advantage of AS compared to GPB.

There are mechanical devices that have been shown to improve the cough in patients with NMD [17-21]. While these devices are effective, they are not available to the entire population due to their cost, such as the case of Latin America. As shown in a recent study by Senent et al. [22], some of these devices may be indicated in patients who have glottic dysfunction, such as advanced stages of ALS. Consequently, it is necessary to find low-cost therapeutic strategies for use in patients with NMD and neurological diseases by determining which technique is more cost-effective and bring to the professional team different options to decide the best option for each patient. For these reasons, we explored portable and low-cost techniques to assess and treat patients considering that neuromuscular disease patients are commonly treated at home setting by physical therapist where the availability of costly devices are scarce in most Latin American countries.

Therapeutic techniques to increase MIC, improve lung compliance, decrease micro atelectasis generation and improve voice volume decrease progressive thoracic deformity and increase cough effectiveness [13]. These factors are fundamental in contributing to an improvement in the quality of life and decrease in respiratory morbidity and mortality in NMD patients.

One of the main limitations of this study is the small number of subjects evaluated. Despite the small patient number, the sample size calculation determined that only 13 patients were needed to show significant changes because previously published studies have demonstrated that these techniques are very effective at increasing the MIC. Also, the power analyzed after data collection was 98%. The study design, quasi-experimental, without a control group, was another limitation. Future studies including a control group are necessary to obtain more consistent results. Another limitation is that our intervention was only one session. Based on the positive results, it is necessary to determine to the outcomes of these interventions in the medium-and long-term. An important limitation was that our design does not incorporate a sham intervention, which would have decreased the risk of bias. The above is important to consider because the effectiveness of the technique depends on the professional training that applies, and could lower inter or intra-observer reliability of the measured variables.

5. CONCLUSION

Finally, we concluded that, in the studied patients, both air stacking and glossopharyngeal breathing are effective in increasing the maximum insufflation capacity in children and adolescents with neuromuscular diseases, but air stacking is the most effective of the two techniques.

DISCLAIMER

This manuscript was presented at the conference "European Respiratory Society International Congress, 2015, At Amsterdam, The Netherlands" available link is "http://www.researchgate.net/publication/282573763_Effects_of_air_stacking_versus_glossopharyngeal_breathing_on_maximum_insufflation_capacity_in_children_and_adolescents_with_neuromuscular_disease" date 26 –30 September 2015, details Volume: 46

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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