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RESEARCH ARTICLE

Lung Inflation by Glossopharyngeal Breathing and "Air Stacking" in Duchenne Muscular Dystrophy

ABSTRACT

Bach JR, Bianchi C, Vidigal-Lopes M, Turi S, Felisari G: Lung inflation by glossopharyngeal breathing and "air stacking" in Duchenne muscular dystrophy. Am J Phys Med Rehabil 2007;86:295–300.

Objective: To compare the use of glossopharyngeal breathing (GPB) and air stacking to increase lung volumes and cough peak flows (CPF), and GPB to increase ventilator-free breathing ability (VFBA), for patients with Duchenne muscular dystrophy.

Design: A case series of all referred patients with declining vital capacity (VC). Seventy-eight patients underwent training in and monitoring of the efficacy of air stacking (retaining consecutively delivered volumes of air delivered via manual resuscitator and held by glottic closure) to maximum insufflation capacity (MIC). GPB also was demonstrated to all 78 patients, and 32 were formally trained and prescribed GPB as their VCs decreased below 400 ml. To be successful, the MIC or GPB maximum single-breath capacity (GPmaxSBC) had to exceed VC. Improvements in VFBA were determined by requiring fewer ventilator-assisted breaths per minute. CPFs were measured by peak flow meter.

Results: Seventy-four (94.9%) of the patients could air stack (MIC $>$ VC), and, thus far, 21 (27%) are able to GPB. Fifteen could GPB sufficiently to delay onset of daytime ventilator use and, later, to require 1.9 fewer ventilator assisted breaths per minute. For the 47 patients with multiple data points, as VC deteriorated from 1080 $\pm$ 870 to 1001 $\pm$ 785 ml, MIC increased from 1592 $\pm$ 887 to 1838 $\pm$ 774 ml. For 21 patients, GPmaxSBC significantly exceeded VC (824 $\pm$ 584 *vs.* 244 $\pm$ 151 ml, respectively, $P < 0.001$). The ability to increase lung volume by air stacking (MIC) was better retained than was the ability to increase lung volume by GPB (GPmaxSBC). Air stacking also permitted assisted CPF to exceed unassisted CPF: 289 $\pm$ 91 and 164 $\pm$ 76 liters/m, respectively ($P < 0.001$).

Conclusions: GPB and air stacking can increase lung volumes and, thereby, cough flows. GPB also can be used in many cases to delay and decrease daytime ventilator use.

Key Words: Glossopharyngeal Breathing, Cough, Duchenne Muscular Dystrophy, Respiratory Therapy, Noninvasive Mechanical Ventilation, Life Expectancy

The key to the successful long-term use of noninvasive mechanical ventilatory support is in effectively expelling airway secretions when necessary.[1,2] To do so, the augmentation of lung air volumes can be crucial to optimize cough peak flows (CPF).[3–6] In 1981, it was reported that 47 Duchenne muscular dystrophy (DMD) patients reached a maximum (plateau) vital capacity (VC) between ages 10 and 12 (range 9–16); in the general population, VC plateaus at age 19.[7] Subsequently, VC decreases by 5–10% per year in patients with DMD.[7,8] This contributes to the decrease in unassisted CPF. Two methods of lung-inflation therapy that can result in increased cough flows are air stacking and maximum-depth glossopharyngeal breathing (GPB).

Air stacking involves the use of a manual resuscitator or volume-cycle ventilator to deliver volumes of air that are consecutively held by glottic closure until no more air can be retained.[9] The maximum lung volume that can be held by air stacking is the maximum insufflation capacity (MIC).

Dail first described GPB as gulping air into the lungs for the purpose of providing deep lung volumes to increase the cough flows of five postpoliomyelitis patients.[10,11] Use of GPB for ventilator-free breathing has been reported for high-level traumatic tetraplegia patients and poliomyelitis patients,[11–13] but not for DMD patients.[13] Indeed, DMD patients who are not taught and equipped with respiratory muscle aids to avoid respiratory failure often undergo tracheotomy before they are able to benefit from GPB for ventilator-free breathing ability (VFBA).[1]

The VFBA is considered limited when patients need intermittent positive pressure ventilations to supplement unassisted minute ventilation to prevent distress and to maintain baseline blood-gas levels. When absent, cessation of ventilator use results in immediate distress and blood-gas deterioration.

Advances made during the last two decades in noninvasive mechanical ventilatory support and mechanically assisted coughing have greatly improved survival without resort to tracheotomy.[1] This makes air stacking and GPB increasingly important for the autonomous augmentation of cough flows, voice volume, and VFBA. GPB mastery also can eliminate fear of ventilator dysfunction or disconnection. Thus, we prospectively determined whether the ability to air stack and GPB could improve with practice and result in improved VFBA and cough flows.

MATERIALS AND METHODS

This work was approved by the hospital ethics committee. Seventy-eight consecutive males who had visited a clinic since 1996 were studied after VC plateau. The initial appointment for 16 patients had been before 1996. The diagnosis was based on clinical, enzymatic, electromyographic, and biopsy assessments. Gene-deletion studies were positive for 54 of 70 patients. All except one lost the ability to walk by age 11. Four of the 78 patients had taken glucocorticoids.

All patients were trained in air stacking and were given a demonstration of GPB after the plateauing of the VC. The age and magnitude of the plateau was documented by pre- and post-VC measurements below a maximum (plateau) for 24 patients, or it was assumed for the others because of their age and because subsequent data points were lower than the initial VC measurement. All 74 self-directed patients mastered air stacking when initially introduced to it (MIC $>$ VC) and were asked to practice it two to three times per day, 10–15 maneuvers each time, from then on. Air stacking was performed by the patient receiving consecutively delivered volumes of air from a manual resuscitator or a volume-cycled ventilator via a mouthpiece, nasal, or oral–nasal interface and retaining as much as possible with a closed glottis (MIC).[9] The volume was then measured spirometrically to determine MIC.

Each patient was asked about his practice efforts at every visit. Thirty-one of 47 who returned for at least one follow-up visit reported performing it at least twice daily.

GPB was demonstrated after VC plateau at every clinic visit, and each patient was screened for the ability to exceed VC by GPB. Once a patient demonstrated mastery (maximum single-breath capacity [GPmaxSBC] $>$VC), he was asked to practice it three times a day. If he had not mastered GPB by the time his VC had decreased to 400 ml (the point at which many patients with DMD begin to use daytime ventilatory assistance), a demonstration videotape was dispensed, and GPB practice was formally prescribed.[14] GPB practice was considered optional before this point because it could not be used for VFBA early on, and air stacking was easier to master and the ability easier to retain over time for deep lung expansion, as typified by the patient record in Figure 1.

GPB was taught by having the patient take a deep breath and hold it, then imitate the clinician and take 15–20 gulps, and then blow the volume into a spirometer. The GPmaxSBC was the maximum volume that could be gulped in. Training was facilitated by monitoring gulp efficiency, which was defined as (GPmaxSBC $-$ VC) $\div$ (number of gulps to a maximum insufflation) in milliliters per gulp.[15] If initially unsuccessful, the patient's nostrils were sealed to demonstrate to him the need for the soft palate to seal off the nasopharynx. Even then, nine patients could not master the glottic movements necessary for successful GPB. On obser-

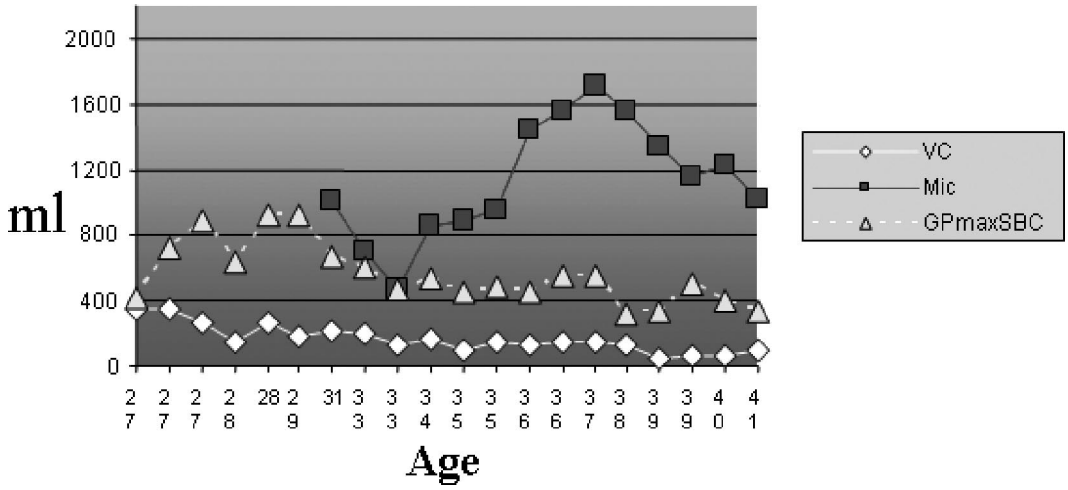

FIGURE 1 *Graphic for a patient with 20 data points. VC, vital capacity; MIC, maximum insufflation capacity; GPmaxSBC, glossopharyngeal maximum single-breath capacity.*

vation, they did not have as much rostral–caudal movement of the glottis as did patients whose GPmaxSBC exceeded VC, nor did they have the glottic closure gulp (click) that is heard with successful GPB. VC, MIC, and GPmaxSBC were measured spirometrically (Mark 14 spirometer; Ferraris Development and Engineering Co, Ltd, London, UK).

Assisted CPF are defined as CPF augmented by an abdominal thrust that is timed to glottic opening after air stacking or GPB to deep lung inflation. Unassisted and assisted CPF were measured by Access Peak Flow Meter (Health Scan Products Inc, Cedar Grove, NJ) at every visit.

The maximum of four or five measurements was recorded for the initial (post-VC plateau) VC, MIC, GPmaxSBC, and CPF; the maximum observed assisted CPF, MIC, and GPmaxSBC; and the most recent data. A standard oral–nasal mask was used for spirometry and CPF measurements.

Diminished VFBA was defined by lack of autonomous ability to breathe without using GPB, or by a difference in need for mouthpiece intermittent positive-pressure ventilations during 5-min periods when using GPB *vs.* periods when it was not used, with no change in end-tidal carbon dioxide or oxyhemoglobin saturation. Patients took these assisted breaths to avoid dyspnea. Delayed onset of daytime ventilatory assistance was recognized when patients who normally used GPB throughout waking hours became dyspneic and hypercapnic when breathing without it, to the extent that only resumption of GPB or deep, ventilator-assisted breaths could maintain them.

Statistical Analysis

Descriptive statistics included mean and standard deviations. Comparisons between mean values of MIC and GPmaxSBC with VC at the initial, plateau, and most recent evaluations were made by *t* test using the Bonferroni correction for six comparisons. This warranted a P value <0.008 for statistical significance.

RESULTS

All 78 patients cooperated for VC measurements, but four were too cognitively impaired to learn lung-inflation techniques. Twenty-four DMD patients' VCs plateaued at mean age 12.7 ± 3.1 (range 10.5–16.1) yrs, at 2026 ± 555 (940–2510) ml.

The 74 remaining self-directed patients (mean age 20.6 ± 3.1 yrs) were taught air stacking after their VC was on the decline. Their mean initial VC, and MIC by air stacking, were 987 ± 631 and 1501 ± 618 ml, respectively, and unassisted and assisted CPF were 145 ± 112 and 250 ± 84 liters/m, respectively. The VC, MIC, and CPF changes over time for the 47 patients with multiple data points during a 7- to 169-mo follow-up are illustrated in Figure 2. For 31 of the 47 patients who reported practicing air stacking at least twice a day, the MIC increased over time despite diminishing VC. For these 31, MIC plateaued at 21 ± 18 (2–51) mos after initial training. Eight of the 47 patients' post-plateau VCs increased by 35–70 ml for one or more follow-up visits after practicing lung-insufflation therapy. Two patients lost the ability to air stack (MIC = VC) at ages 26 and 38.

Ten patients had mastered GPB on their own and presented with GPmaxSBC >VC. Twenty-two others were formally taught GPB. The VC and GPmaxSBC changes over time for 19 of the 32 patients who mastered GPB and had two or more data points are illustrated in Figure 3. Eighteen of the 19 reported using GPB daily. For 11 of the 19, GPmaxSBC increased with practice and plateaued 13 ± 16 (range 3–49) mos after initial mastery

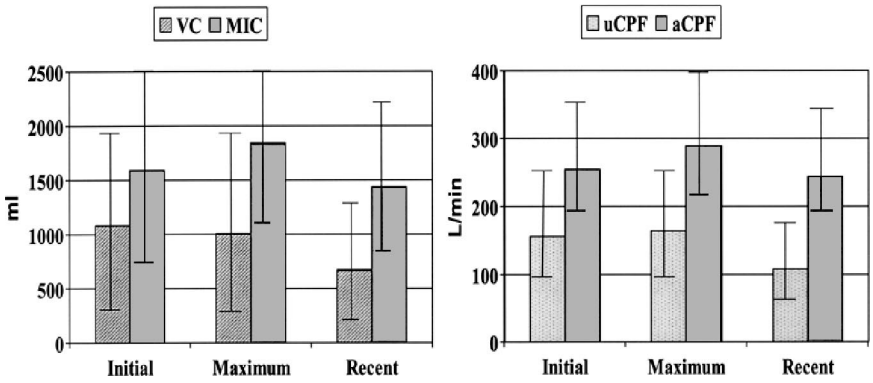

FIGURE 2 *Changes in vital capacity (VC), maximum insufflation capacity (MIC), and unassisted and assisted cough peak flows (CPF) over time for 47 patients with two or more data points; age initially 20.6 ± 4.1 (13.1–33) yrs, at maximum 21.9 ± 4.5 (13.6–37.2) yrs, most recently 24.9 ± 4.5 (13.7–43.1) yrs.*

(Figure 4). Gulp capacities were from 8 ± 9 to 70 ± 34 ml, depending on whether GPB was being used for normal minute ventilation or for maximal lung inflation, with the latter gulps being much smaller.

Fifteen patients eventually used GPB throughout daytime hours such that respiratory distress and hypercapnia developed when ceasing GPB. Thus, GPB delayed the need for daytime ventilator use. At the most recent evaluation, these 15 required 5.3 ± 1.7 mouthpiece intermittent positive-pressure ventilations per minute (1200 ml each) when not using GPB and 3.4 ± 1.7 ventilations per minute when using it. All used volume-cycled ventilators on assist/control mode with delivered volumes of 850–1500 ml and a backup rate of 10–12 per minute. Nine of the 32 patients did not master GPB, and two had GPmaxSBC exceed VC by 80 and 240 ml at initial mastery but have not yet returned.

Whereas 74 of 78 (95%) patients with DMD mastered air stacking, only 21 of 78 (26.9%) have mastered GPB thus far, and at least nine are likely to never master it. Some of the remaining 42 may master it as their VCs decrease below 400 ml. Thus, for at least 27% of DMD patients, GPmaxSBC can exceed VC, and for most of these, it can delay the need for daytime ventilator use. For the 74 patients as a whole, and for the patients in Figures 2–4, the MIC and GPmaxSBC values significantly exceeded VC and assisted CPF exceeded unassisted CPF ($P < 0.008$).

The mean age of beginning nocturnal noninvasive ventilation was 19.1 ± 3.3 yrs. Although all of our GPB users lost the ability to breathe unaided by ventilator use, none have tracheostomy tubes. Six patients died during the course of the study:

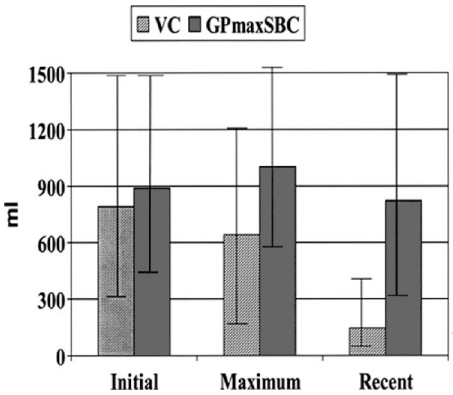

FIGURE 3 *Changes in vital capacity (VC) and glossopharyngeal breathing maximum single-breath capacity (GPmaxSBC) over time for 18 patients, including eight whose GPmaxSBC did not increase with training. Age at initial GPB mastery 24.1 ± 4.3 (17.5–31.1) yrs, maximum GPmaxSBC 25.4 ± 4.8 (18–33.5) yrs, most recent evaluation 29.3 ± 5.7 (23.2–43.1) yrs of age.*

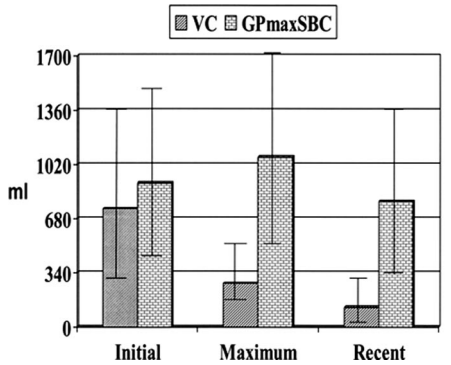

FIGURE 4 *Changes in vital capacity (VC) and glossopharyngeal breathing. Maximum single-breath capacity (GPmaxSBC) for 11 patients whose GPmaxSBC increased over time. Age at initial GPB mastery 24.4 ± 4.3 (17.1–31.0) yrs, at GPmaxSBC maximum 27.8 ± 4.3 (17.5–33.5) yrs, at most recent evaluation 30.2 ± 5.6 (24.9–43.1) yrs of age.*

five from overt cardiac failure, and one, who had no assistance at home, from respiratory failure during a respiratory tract infection.

DISCUSSION

The VC plateau that we found for 24 patients occurred months later than the figure reported previously.[7] The fact that 4 of our 24 patients received glucocorticoid therapy may be one of the reasons for this.

In a previous study, it was reported that three DMD patients used GPB to inflate the lungs to two to three times VC. The authors did not report any effect on cough flows or on VFBA.[16] The delayed need to use daytime ventilatory support and, subsequently, the fewer assisted breaths required when using GPB than when not using it, provided some security to the patients because they could survive longer by using GPB in the event of ventilator failure, and GPB saved them some effort in having to rotate the neck and grab the mouthpiece as much as would have been necessary otherwise. Also, because 90% of episodes of respiratory failure and death for conventionally managed DMD patients occur as a result of ineffective coughing during intercurrent upper–respiratory tract infections,[17] and because cough flows correlate with (pre-) cough volumes,[9] it is important for patients to be able to autonomously increase lung volumes when they need to cough. Lung-inflation therapy also helps maintain both dynamic and static pulmonary compliance.[18] GPB, like air stacking via a volume ventilator, permits this. Because both GPmaxSBC and MIC can improve and plateau over a wide range of time (2–51 mos) despite declining VC, both should be monitored regularly, and practice should be encouraged. Pressure-cycled ventilators such as BiPAP machines do not permit air stacking and should not be used for these patients.

The slight, temporary increase in VC for eight patients was probably attributable to an improvement in pulmonary compliance brought about by regular lung-expansion therapy. Although we only measured the effect of deeper lung volumes and abdominal thrusts on assisted coughing, deep lung insufflations and abdominal thrusts have been separately shown to increase CPF almost equally, with the greatest increases when they are combined.[9,12,13]

The GPmaxSBCs and gulp volumes in DMD patients tend to be lower than those reported for postpolio and spinal cord–injured patients, who often have gulp capacities over 100 ml, GPmaxSBCs over 3000 ml, and many hours of VFBA despite having little or no VC.[5,12,13] This is because bulbar-innervated muscles tend to be spared for the latter but become increasingly dysfunctional in DMD. Thus, although DMD patients are usually able to speak clearly, take food by mouth, and maintain good vocal-fold mobility and glottic closure even after 40 yrs of age,[1] they lose the ability to close the glottis tightly enough to hold deep lung volumes. Whereas most patients with bulbar amyotrophic lateral sclerosis lose the ability to air stack, and GPB and cough become physically impossible for them,[19] we have observed losses of air-stacking ability in only 2 of 74 DMD patients, despite some being over age 40.

When air stacking and GPB are suboptimal, lung-insufflation therapy can still be performed using a manual resuscitator with the expiratory valve blocked, by delivering high volumes from a volume-cycled ventilator, or by using the CoughAssist machine (J. H. Emerson Company, Cambridge, MA) at insufflation pressures of 40 cm H_2O or more via an oronasal interface. Indeed, as assisted CPF and air-stacking ability decrease, our patients use CoughAssist machines for both mechanically assisted coughing and for maximal lung insufflations. No complications have been associated with this therapy for the >1000 patients with neuromuscular diagnoses who have been treated in this manner in the last 28 yrs.[20]

Comparing GPmaxSBC and MIC permits the evaluation of oropharyngeal muscle groups. When the GPmaxSBC is greater than the MIC achieved by air stacking via a mouthpiece (as often occurs in postpoliomyelitis patients), then the glottis is sufficiently intact to hold deep lung volumes, but the lips and buccal muscles are too weak to permit air delivery past the vocal fold. Some such patients can air stack better via nasal interfaces. Likewise, when MIC is greater than GPmaxSBC, as for these DMD patients, the hypopharyngeal musculature is weakened and laryngeal mobility is impaired. Some patients can only GPB or air stack with the nose plugged because the soft palate is unable to seal off the nasopharynx.

Although we and others strongly recommend that GPB be taught to patients with neuromuscular disorders,[21] few clinicians are familiar with the technique. Some patients learn it on their own. This is probably a consequence of cerebral imprinting of the phylogenetic distribution of lung-ventilation mechanisms. Aspiration breathers, like mammals, ceased GPB shortly after aspiration breathing had evolved.[22] Indeed, at least four of our patients who learned it on their own and used it spontaneously were hypercapnic and hypoxic. It is possible that the mechanism for GPB is the same as that observed in experimental studies in lunged amphibians for whom buccal pumping activity increases during hypoxia and hypercapnia.[23,24] Lung packing by GPB is used by many breath-hold divers to increase lung air volumes by up to 5.6 liters over VC to permit longer submersion, and by many competitive swimmers to increase thoracic volumes and buoyancy.[22–27] The transpulmonary pressures generated by maximal-depth GPB in swim-

mers have been reported as high as 80 cm H_2O.[28,29] These are similar to the pressures we use for maximum-depth air stacking. Despite such high pressures, no barotrauma or other complications have been reported.

Many postpolio, spinal cord–injured, and DMD patients can use GPB instead of a ventilator for ventilatory support when awake.[12,13,15,30] With deteriorating inspiratory and bulbar-innervated muscle function, however, DMD patients eventually use GPB to take fewer ventilator-assisted breaths. Once a DMD patient's VC is inadequate to breathe without continuous ventilator use (VC <200 ml), bulbar-innervated musculature is inadequate to use GPB for complete ventilator-free breathing.

GPB has been included among the techniques that are not well adapted to producing bronchial clearance, because "it cannot be mastered by all patients."[31,32] However, it seems that this is rarely even attempted. The clinician should at least identify those patients who already perform it and should help them improve their technique by spirometric feedback. With training and practice, many DMD patients can exceed their VCs three- to tenfold (Fig. 4)—increases comparable with those of spinal cord–injured and postpolio patients.[5,12,13] Both air stacking and GPB can improve with practice, even when VC is deteriorating. GPB provides a vital advantage for those who master it. It should no longer be ignored for patients with DMD.

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