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ORIGINAL ARTICLE

Feasibility of a standardized protocol for respiratory training with intermitted positive pressure breathing ventilator application in dysphonia and dysarthria

Cecilia PERIN [1, 2], Miryam MAZZUCCHELLI [1, 2] *, Daniele PISCITELLI [1, 3], Giacomo BRAGHETTO [1], Roberto MERONI [1, 2, 4], Cesare M. CORNAGGIA [1, 2], Cesare G. CERRI [1, 2]

[1]School of Medicine and Surgery, University of Milan - Bicocca, Monza, Monza e Brianza, Italy; [2]Istituti Clinici Zucchi, Carate Brianza, Monza e Brianza, Italy; [3]School of Physical and Occupational Therapy, McGill University, Montreal, QC, Canada; [4]Department of Physiotherapy, LUNEX International University of Health, Exercise and Sports, Differdange, Luxembourg

*Corresponding author: Miryam Mazzucchelli, School of Medicine and Surgery, University of Milan - Bicocca, Monza (MB), Italy.
E-mail: m.mazzucchelli@campus.unimib.it

ABSTRACT

BACKGROUND: Brain damage can affect several functions related to speech production leading to dysphonia and dysarthria. Most rehabilitation treatments focus on articulation training rather than on pneumophonic coordination and respiratory muscle strength. Respiratory training using an intermitted positive pressure breathing (IPPB) ventilator can be used for this last purpose; no agreement on a standard protocol has been reached to date.
AIM: To evaluate the feasibility and the effectiveness of a standardized incremental protocol of respiratory training using IPPB to treat dysphonia and dysarthria.
DESIGN: Case series study.
SETTING: Neuropsychological Rehabilitation Unit in an Italian Neurorehabilitation Division.
POPULATION: Thirty-two subjects with dysphonia and dysarthria resulting from neurological lesion.
METHODS: Participants were assessed using clinical evaluation scales (GIRBAS scale of dysphonia, Robertson dysarthria profile), respiratory function test, and arterial blood gas analysis in air. The evaluations were performed at baseline and after 20 sessions of respiratory training with IPPB. The protocol provided a default increment of ventilator parameters. All subjects also underwent a standard speech and language therapy treatment. A satisfaction survey to assess acceptability and the Goal Attainment Scale were applied.
RESULTS: All participants fulfilled the protocol. No complications or discomfort were reported. Subjects' satisfaction at survey was 97.7%. After respiratory training, all respiratory function parameters increased, but only maximal voluntary ventilation (MVV), maximum inspiratory pressure (MIP), and maximum expiratory pressure (MEP) were statistically significant ($P<0.05$). Clinical evaluation scales significantly improved ($P<0.05$). Correlation between respiratory function parameters and clinical evaluation scales showed a moderate correlation between MVV, MEP, MIP, and Robertson dysarthria profile ($P<0.01$). A weak correlation was found between MIP, MVV, and GIRBAS scale ($P<0.05$).
CONCLUSIONS: Our protocol showed to be practical and well-tolerated. After respiratory training, MVV, MIP and MEP improved in significantly. Clinical scale scores improved in all participants.
CLINICAL REHABILITATION IMPACT: Respiratory training using IPPB ventilator can be useful in implementing speech and language treatments in subjects with dysphonia and dysarthria linked to brain injury.

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KEY WORDS: Breathing exercises; Dysphonia; Dysarthria; Brain injuries.

Dysphonia and dysarthria are acquired neurological communication disorders associated with decreased health-related quality of life.[1, 2] Dysarthria occurs in approximately 10-60% of survivors of traumatic brain injury[1] and affects about 20-40% of post-stroke individuals.[3-5] In degenerative conditions (*e.g.*, Parkinsonian disorders), dysphonia and dysarthria incidence ranges from 70% to 100%.[2] Their impact affects physical and psychosocial

features, resulting in a severe disability worsening patients' activity and social participation.[6]

Brain damage may compromise several speech production functions, such as respiration, phonation, articulation, and prosody; therefore, some authors use the term "dysarthrophonopneumia."[7]

The main goal in treating speech disorders is improving articulation communication abilities, considering all dimensions of the International Classification of Functioning, Disability and Health (ICF) Framework.[1, 8] Several speech rehabilitation therapies, such as traditional behavioral approaches and new therapeutic options, have been proposed to address impaired function of speech production.[1, 9, 10] However, according to recent reviews, the evidence is lacking about the best treatment options, and as well as indications on the optimal duration or intensity of standard care are missing.[1, 9, 10]

An essential aspect to consider is respiratory/phonatory dysfunction.[11, 12] According to some authors,[13] the recovery of post-stroke dysarthria is dependent on the improvement of respiratory function because the basis of sound production is supported by the volume and control of respiratory airflow.

Post-stroke individuals often experience dysarthria associated with disordered breath control.[14] Stroke may lead to reduced respiratory muscle strength[15, 16], prolonged bed rest and pulmonary complications (*e.g.*, pneumonia), with high risk of mortality.[17] Individuals with Parkinson's disease (PD) may also experience significant respiratory changes, including increased chest wall rigidity and decreased respiratory muscle strength and coordination. These impairments affect mechanisms related to speech and breathing, leading the individual to rely more on active muscle forces. The increased use of active muscle forces makes speech less effective, more effortful, and fatiguing.[18, 19]

For these reasons, an efficient coordination pattern between breath and speech and increased strength of respiratory muscles would be recommended.[13] Traditional respiratory/phonatory interventions include respiratory/phonatory exercises with postural adjustments, appropriate speech-breathing patterns, and effort closure techniques to ameliorate pneumophonic coordination.[20]

Respiratory muscle strength training focuses on increasing the force-generating capacity of inspiratory and expiratory muscles.[21] Previous studies demonstrated the effectiveness of respiratory muscle strength training in increasing respiratory muscle strength[22, 23] and improving speech breathing[19] in neurological conditions. Various devices and protocols have been used to improve respiratory muscle strength.[24] To our knowledge, there are no reports that considered the use of an intermittent positive pressure breathing (IPPB) ventilator with an incremental protocol for treating dysphonia and dysarthria.

IPPB ventilatory systems have great importance in respiratory rehabilitation and have been used to treat pulmonary complications in veterans.[25] In the last 20 years, the IPPB ventilator has been used in respiratory deficits related to chest wall deformation (kyphoscoliosis),[26] degenerative respiratory muscles deficits[27] and chronic obstructive pulmonary disease.[28]

IPPB is used in clinical practice primarily to improve lung volume and to reduce breathing work.[28] The mechanism of action is based on the tidal volume (Vt) increase. The device delivers a low positive pressure level during spontaneous breathing; inspiratory flow and the end-inspiratory pressure can be set to improve patient comfort. An interesting feature is the fact that the trigger for respiratory act follows spontaneous breathing, and this may help the breathing pattern by encouraging coordination and stimulating muscle strength.

This poses the basis for the potential use for speech and language therapy. Interestingly, respiratory training with IPPB may help patients to improve their respirator pattern by enhancing pulmonary compliance and increasing respiratory muscle strength, in line with other approaches like the Lee Silverman Voice Treatment (LSVT®) that aims at maximizing phonatory and respiratory functions.[29] However, to the best of our knowledge, there is no agreement on a standard protocol for its use in dysphonia and dysarthria. Therefore, the first aim of this study was to evaluate if a standardized incremental protocol of respiratory therapy with IPPB is feasible in clinical practice for treating dysphonia and dysarthria linked to neurological conditions. The second aim was to investigate the effects of this respiratory treatment combined with traditional speech therapy on respiratory function parameters and clinical outcomes.

Materials and methods

Participants

This study included subjects suffering from dysphonia and/or dysarthria due to traumatic brain injury, stoke or extrapyramidal disorders (Parkinson, MSA). Study participants were recruited among consecutively admitted subjects to the Neuropsychological Rehabilitation Unit of Istituti Clinici Zucchi in Carate Brianza from 2016 to 2017.

To be included in the study, subjects needed to be in stable neurological and cardiopneumological conditions (assessed with neurological and cardio-pneumological examination, ECG, chest x-rays, blood pressure, pulse and saturation monitoring) and able to express consent to take part in the experimental protocol. Exclusion criteria were the following: assisted respiration, in home-treatment with respiratory machines, non-treated PNX, intracranial pressure over 15mmHg (primitive or secondary), history of heart failure, trachea-esophageal fistula, recent esophageal surgery, hemoptysis of unknown origin in 3 months before the treatment, nausea, aerophagia, hiccups, active untreated tuberculosis, radiographic evidence of bleb.[30]

The study was conducted following the declaration of Helsinki and was approved by the local Ethics Committee. All participants signed an informed consent form prior to participation.

Outcomes

The following outcome measures were collected.

Demographic data were obtained at the baseline assessment. Dysarthria was quantified using the Robertson dysarthria profile that contains eight domains (respiration, phonation, facial musculature, diadochokinesis, oral reflexes, articulation, intelligibility, prosody), each including 5-20 items (rated on a 4-point scale) evaluating the subject's speech performance ability. The total score ranges from 0 to 280 (the greater the score, the better the speech performance).[31]

Dysphonia was assessed using the GIRBAS scale. It provides a perceptual evaluation of voice according to five parameters: Grade (overall impression), Roughness, Breathiness, Asthenicity and Strain. Each parameter is rated on a 0 to 3 scale, where 0 indicates normality; 1, a slight deviance; 2, a moderate deviance; and 3, a severe deviance from normal.[32] An expert speech therapist (>20 years of experience) administered both scales.

Cognitive impairment was tested with Mini-Mental State Examination (MMSE).[33] The Zung Self-Rating Depression Scale[34] was also used to account for possible psychological interference in compliance. The Goal Attainment Scale (GAS)[35] was used to select each patient's rehabilitation treatment goals and to calculate the extent to which a patient's goal was met.

Spirometry examination was performed using a portable spirometer (Spirobank by CareFusion). Maximum inspiratory pressure (MIP) and maximum expiratory pressure (MEP) evaluation were carried out using a portable manometer (Micro RPM by CareFusion). Arterial blood gas analysis in air (Hemogasanalizer Osmetech Opti CCA) was done to evaluate arterial pH, pO_2 and pCO_2.

TABLE I.—*Satisfaction survey.*

Satisfaction survey
Number of treatments completed
Number of treatments not completed
Did you enjoy the treatment?
Did you find the treatment interesting?
Did you find the treatment easy to perform?
Did you find the treatment boring?
Did you find the treatment painful?
Did you find the treatment strenuous?
Did you experience short breath after the treatment?
Did you find the treatment made you feel dizzy?
Did you find the treatment caused nausea?
Did you find the treatment caused headache?
Did you find the treatment caused vertigo?
Did you find the treatment caused heart rate increment?
Did you find the treatment caused hiccough?
Would you like to participate to other rehabilitation programs like the one you concluded?

A satisfaction survey was administrated to assess the protocol acceptability. A questionnaire was developed on purpose, using a Likert Scale to evaluate the patient's experience. Each item scored from 1 to 7, where 1 represents strongly disagree and 7 strongly agree. The survey items are reported in Table I.

Interventions

Respiratory training consisted of 20 non-invasive ventilatory therapy sessions with IPPB lasting 15 minutes each, three times/week. Each session consisted of 4 series of 10 respiratory acts each, with a pause of 2 minutes within each series. The treatment was considered clinically relevant if the patient tolerated at least 25 hPa (minimum effective value considered for rehabilitation in pneumology according to clinical practice). IPPB Ventilator Alpha 2000 was used for the treatment. This device allowed four different expiratory resistances with linear progression. Participants wore a nose clip during treatments and were connected with the IPPB machine using a snorkel or a face mask. To monitor the intervention safety, blood pressure, heart rate, and blood oxygen saturation were measured before and after each session.

The respiratory protocol was defined with a pneumologist specialized in respiratory physiopathology targeting respiratory muscle strength training and measuring respiratory function parameters to prove its effectiveness. The first two sessions were pre-training sessions, with only five breaths for each series. Protocol parameters could

TABLE II.—*Description of incremental protocol for respiratory training with IPPB.*

Treatment day	Maximum inward pressure (hPa)	Trigger pressure (hPa)	Air flow (L/min)	Expiratory resistance
1-2	15	-1	40	1.0
3	15	-2	40	1.5
4	16	-2	37	1.5
5	17	-2	34	1.5
6	18	-2	31	1.5
7	19	-2	28	1.5
8	20	-2	25	1.5
9	20	-2	40	2
10	21	-2	37	2
11	22	-2	34	2
12	23	-2	31	2
13	24	-2	28	2
14	25*	-2	25	2
15	25*	-2	40	2.5
16	26	-2	37	2.5
17	27	-2	34	2.5
18	28	-2	31	2.5
19	29	-2	28	2.5
20	30	-2	25	2.5

*Minimum value to consider the treatment clinically relevant.

be adaptable, as a subject may tolerate a certain pressure. Whenever the patient could not tolerate higher pressures, the positive pressure level was set back to the previous session threshold, and it was maintained for all the following sessions until the conclusion of the protocol. Table II depicts the evolution of ventilator parameters.

Resident physiatrists administered the intervention.

All participants received a traditional speech and language treatment of 45 minutes, three times/week focused on oro-facial musculature movement and articulation.[36] Patients performed lip, jaw, tongue exercises without phonation and then progressed to articulation exercises ranging from single phoneme utterances to more complex utterances including those with consonant clusters, words of increasing length and eventually to phrases, sentences and continuous speech, and prosodic exercises.

Statistical analysis

Normality of data distribution was assessed through visual inspection of QQ plots and the Kolmogorov-Smirnov test. Continuous variables are presented as means and standard deviations or median with first and third quartile, whereas categorical variables are reported as numbers and percentages.

Given the skewness of the data distribution, we used the paired-sample Wilcoxon signs-rank test to detect difference in respiratory parameters and clinical scales. Spearman's Rho correlation coefficient was used to test the correlation between respiratory parameters and clinical scales. The correlations were assessed as follow: $\rho>0.70$ strong correlation, $0.50<\rho<0.70$ moderate correlation, and $\rho<0.50$ weak correlation.[37]

The sample size was calculated to detect two-point variation in the GIRBAS scale, 15% variation at Robertson dysarthria profile and 5% variation in at least one of the measured respiratory parameters (MVV, MIP, MEP). A minimum of 30 subjects was required. Statistical analysis was performed using SPSS statistical software v. 25 (SPSS Inc., Chicago, IL, USA).

Results

Thirty-two subjects (age: 67.47±12.43 years; 25 male and seven female) were recruited and assessed. The demographic characteristics of participants are shown in Table III. Eighteen participants were affected by stroke (three hemorrhagic and 15 ischemic), three by traumatic brain injury, nine by multifocal degenerative pathologies (PD) and two had a mixed etiology (stroke on Parkinson's disease). Fifteen subjects were in acute/subacute neurological conditions (≤90 days) and seventeen in chronic phase (>90 days).

After assessing the clinical scale for dysarthria and dysphonia, four participants were affected by dysarthria only,

TABLE III.—*Demographic and clinical characteristics of study participants.*

Characteristics	Value
N. participants	32
Age (years)	67.47±12.43
Sex	
Males	25 (78.1%)
Females	7 (21.9%)
Etiology	
TBI	3 (9.4%)
Stroke	18 (56.2%)
Extrapyramidal disorders (Parkinson, MSA)	9 (28.1%)
Mixed	2 (6.2%)
Onset	
≤90 days	15 (46.9%)
>90 days	17 (53.1%)
Subjects with dysarthria	30 (93.8%)
Subjects with dysarthria only	4 (12.5%)
Subjects with dysphonia	28 (87.5%)
Subjects with dysphonia only	2 (6.2%)
Mini Mental State Examination	24.58±2.55
Zung Self-Rating Depression Scale	47.02±11.69

Values are expressed as mean±SD for continuous data and count (percentage) for categorical data.
BMI: Body Mass Index; TBI: traumatic brain injury; MSA: multiple system atrophy.

and two participants were affected by dysphonia only. Twenty-six were affected by both conditions.

All enrolled subjects fulfilled the training protocol without side effects or discomfort. No respiratory alkalosis was reported. Oxygen saturation increased from a pre-treatment average of 96%, to a 98% after the session.

The satisfaction questionnaire submitted after treatment showed a patient satisfaction value of 97.7%. At the Goal Attainment Scale, participants expressed the wish to "speak better" in different situations (*e.g.*, talk on the phone, have an intelligible language being understood from other people). Thirteen patients achieved the expected level, setting the score at 0, while seven patients experienced an improvement more than expected (score 1.00), ten patients improved significantly much more than expected (score 2.00) and two subjects experienced an excellent improvement (score 3.00).

All participants showed an increase in all the respiratory function parameters, but only MVV, MIP, and MEP were statistically significant. Also, clinical evaluation scales significantly improved (Table IV).

The Spearman rank correlation test was used to correlate significantly improved respiratory parameters (MVV, MIP, MEP) and clinical evaluation scales. A moderate significant correlation between MVV, MEP, MIP and Robertson dysarthria profile ($P<0.01$) was detected. A weak significant correlation between MIP, MVV and GIRBAS scale ($P<0.05$) was found. Correlation with MEP and GIRBAS scale was not statistically significant (Table V).

Discussion

The first aim of this study was to verify the feasibility and applicability in clinical practice of a standardized incremental protocol of respiratory training with IPPB for treating dysphonia and dysarthria linked to neurological conditions. The training with IPPB associated with standard speech therapy showed to be safe, easy, and well-applicable. All patients were able to perform the protocol reaching maximum pressure without side effects. Participants expressed a high degree of satisfaction (97.7%) after the treatment. Moreover, all participant achieved their rehabilitation goal. Notably, about 60% of individuals reported an improvement more than expected on the Goal Attainment Scale. These results strongly suggest that the primary endpoint has been achieved.

The second aim was to investigate the effects of this respiratory treatment combined with traditional speech therapy on respiratory function parameters and clinical outcomes. Respiratory training with IPPB ventilator combined with conventional speech and language therapy lead to an improvement in all respiratory function parameters; especially MVV, MIP and MEP were statistically significant. For an efficient vocal production, a correct volume

TABLE IV.—*Respiratory function parameters and clinical scales pre- and post-treatment.*

Parameters	Before (T0)	After (T1)	P value
Respiratory function parameters			
FVC	2.43 (1.74; 3.20)	2.42 (1.82; 3.32)	0.550
FEV_1	1.80 (1,45; 2,38)	1.85 (1.50; 2.40)	0.384
PEF	2.97 (2.24; 4.58)	3.41 (2.49; 4.37)	0.070
MVV	40.50 (28.88; 53.20)	49.35 (31.40; 59.88)	0.001*
MIP	32.00 (17.50; 47.50)	38.50 (21.25; 56.00)	0.015*
MEP	50.00 (39.25; 69.25)	59.00 (44.00; 83.00)	<0.01*
Clinical evaluation scales			
Robertson Profile	183 (154.00; 209.50)	215.5 (193.5; 241.00)	<0.01*
GIRBAS Scale	8.00 (5.00;12.00)	3.00 (1.00; 7.00)	<0.01*

Values are expressed as median (25%; 75%).
FVC: forced vital capacity; FEV_1: forced expiratory volume in one second; PEF: peak expiratory flow; MVV: maximal voluntary ventilation; MIP: maximum inspiratory pressure; MEP: maximum expiratory pressure.
*Statistically significance difference considering $P<0.05$.

TABLE V.—*Correlation between respiratory function parameters and clinical scales.*

Parameters	MVV		MIP		MEP	
	ρ	P value	ρ	P value	ρ	P value
Robertson profile	0.666	<0.01*	0.637	<0.01*	0.657	<0.01*
GIRBAS scale	-0.444	0.018*	-0.452	0.016*	-0.337	0.079

MVV: maximal voluntary ventilation; MIP: maximum inspiratory pressure; MEP: maximum expiratory pressure.
*Statistically significance difference considering $P<0.05$.

and control of air flow is required.[7] The improvement of these variables indicates that our treatment addressed the elements necessary for an efficient vocal production. Our results are in with recent literature that underline the importance of respiratory training in treating dysarthria and dysphonia linked to brain damage.[13, 19, 22]

The Robertson dysarthria profile and GIRBAS scale showed significant score improvements after treatments. These findings highlighted that the association of speech and language treatment with respiratory muscle strength training lead to clinically detectable modification, even using qualitative scales. The strong correlation found between respiratory function parameters and clinical evaluation scales score confirmed that the association of both trainings could be adopted.

Moreover, our results suggest-that the rehabilitation treatment addressed impairment and activity and participation dimensions of the International Classification of Functioning, Disability and Health (ICF) Framework,[8] as recommended from recent literature.[1]

According to a recent review, there is uncertainty about the best device, intensity and duration for dysarthria and dysphonia treatments.[12, 24] To our knowledge, nobody proposed an incremental protocol for respiratory training with IPPB in treating dysarthria and dysphonia.

The use of an incremental protocol is important for increasing tolerance, as suggested by AARC clinical practice guidelines.[30] IPPB ventilator seems to be adequate in training dysphonia and dysarthria linked to neurological condition, requiring an active patient participation since the device is triggered by voluntary respiratory act. Future studies are needed to clarify whether a passive automatic trigger cue can be more efficient than letting the patient actively trigger the device.

The feasibility of respiratory training using IPPB ventilator was confirmed, even with the inclusion of subjects with mild cognitive impairment and mild depression in our sample. Finally, our study included acute and chronic neurological subjects. The latter may have developed a chest stiffness and/or a less efficient respiratory dynamic. However, the variables collected showed an increase of pulmonary function-parameters and better clinical evaluation scores in all participants, regardless of the onset of the disease. These data suggest that a significant improvement was possible even in chronic subjects.

The importance of addressing respiratory/phonatory dysfunction in dysarthria and dysphonia have been addressed in recent literature. The strength of this study is the proposed use of IPPB ventilator with an incremental protocol with the aim to ameliorate pulmonary function parameters and clinical evaluation scales in dysphonia and dysarthria of neurologic origin.

Limitations of the study

We acknowledge that the observational nature of the study may produce some flaws. For instance, there was not a long-term follow-up and speech and language treatment was not blind. Further case/control studies with a sham group and blinding are needed. Future researches should also investigate differences between a mechanical ventilatory incentive and a passive incentive ventilation. Another comparison could be conducted using an alternative treatment associated to speech therapy, *e.g*., conventional active pulmonary training or manual treatment of respiratory muscles. Moreover, we are aware that acoustic and perceptual measures of voice and speech could provide more objective results; nevertheless, we used clinical scales with Italian validation and good psychometric proprieties[38, 39] and we obtained objective measures of respiratory function. Finally, we agree that our results refer to a specific sample and generalizability of our results should be verified with other studies; however, the aim of the study was primarily to investigate the applicability of respiratory training with IPPB with an incremental protocol in clinical practice.

Conclusions

Dysarthria and dysphonia assessment should consider the evaluation of respiratory/phonatory dysfunction. Respiratory training using IPPB ventilator can be a useful resource in implementing speech and language treatments in subjects with dysphonia and dysarthria linked to brain injury.

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