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Case Report

Clearance Device Application with Laryngeal Stimulation in Post-Stroke Dysphonia: A Case Report

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Abstract:

Post stroke dysphonia refers to speech and voice disorders caused by paralysis of vocal cords. This case report explored results obtained during speech therapy treatment of a patient with neurological dysphonia through the use of a device based on high frequency oscillatory vibrations and intermittent negative pressure (Simeox), currently indicated for treatment of chronic respiratory diseases with mucus hypersecretion. The results of the acoustic and spectrographic analysis at baseline, during and after treatment showed significant improvements, particularly for voice sonority and stability.

This unique study revealed the potential benefit of combining oscillatory vibrations and intermittent negative pressure, both delivered by a dedicated device, for the treatment of dysphonia secondary to cerebral-vascular stroke event. Further evidence is required to explore efficacy and feasibility of such approach for rehabilitation activities.

Key words: dysphonia; rehabilitation; physiotherapy; speech therapy

Introduction

Neurological dysphonia is a frequent and disabling condition after stroke due to impaired vocal cord function. The severity and specific characteristics of dysphonia can vary depending on the location and extent of the stroke, adversely affecting communication and social activities [1]. Current optimal medical treatment is based on speech therapy which may ensure complete recovering in most patients, however with no or partial response in selected cases. Among emerging techniques, neuromuscular electrical stimulation (NMES) showed a positive effect on voice quality and phonation [2], and coupling other forms of laryngeal stimulation with noninvasive mechanical ventilation (NIMV) intervention derived from the respiratory rehabilitation setting, may represent a new approach. This case report is based on the combined adoption of high frequency oscillatory vibrations and intermittent negative pressure, simultaneously delivered by a dedicated device (Simeox), currently indicated for chronic respiratory diseases with mucus hypersecretion, in a post-stroke patient during intensive rehabilitation treatment.

Case presentation

A 46 years old male of Arabic origin, with no previous medical history, presented to our emergency department following loss of consciousness. On neurological evaluation, a Glasgow Coma Scale (GCS) score of 4, decerebration to pain stimulus and snoring breath were detected. A brain CT scan revealed thrombosis of right vertebral artery at level of foramen magnum, complete thrombosis of basilar artery and filiform left vertebral artery, coupled with subcortical hypodense area in the right parieto-occipital site of ischaemic origin. He then underwent endotracheal intubation and successful mechanical thrombectomy was performed.

On post-operative day 1, the patient was transferred to Intensive Care Unit (ICU) and extubated. The respiratory assessment showed anhypovalid cough since extubation with suboptimal respiratory gas exchanges, requiring high-flow nasal cannula (HFNC) oxygen therapy. On day 3, endotracheal intubation and invasive mechanical ventilation (IMV) were performed due to hypoxia, absence of valid cough, and thick tracheal secretions. On day 7, percutaneous tracheostomy with bronchoscopic guidance was packed. On day 9, he was placed on spontaneous breathing with one-way valve PEEP 5 and FiO2 31%. On day 10, oxygen therapy was set via heat moisture exchanger (HME) at 2 l/min.

On day 3, a second brain CT scan revealed ischaemic areas in the right temporo-occipital and ipsilateral posterior thalamo-capsular territories, as far as a small initial left occipital hypodensity of similar nature. On day 11, physiotherapy was initiated for residual right hemiplegia, and on day 18 speech therapy intervention was started for post-stroke swallowing difficulty and dysphonia. At that time, the patient presented plegia in the right upper limb and severe paresis in the right lower limb, being able to maintain a sitting position in a wheelchair. He could eat homogeneous diet, showing score 4 for nutrition and 0 for hydration according to the International Dysphagia Diet Standardisation Initiative (IDDSI) [3], and score 5 according to the Functional Oral Intake Scale (FOIS-IT) [4]. He also tolerated fluids in small sips, while maintaining a capped cannula 24-hour. On day 47, he was decannulated without complications.

Evaluation of dysphonia

Before the acute event, the patient had no previous difficulties other than nasal voice. No voice disorders were also reported in the family. The patient's work profession involved a mild daily voice load (< 2h/day) but the patient was subjected to intense and prolonged sound stimuli (which is why he uses headphones), abrupt temperature changes, and contact with dust and gases. He was a current smoker (1 pack/day up to one year), having consumed alcohol until about 5 years ago, and taking no medications. The patient described post-stroke vocal problems as follows: "...the voice does not come out well and is most disabling during telephone interview...". Results of the voice handicap index (VHI) questionnaire [5] indicated a minimal amount of handicap with a score of 17 out of 120.

The clinical voice assessment showed nasal breathing, normal mobility of the diaphragmatic- abdominal, and a reduced mobility of the costo-diaphragmatic and sterno-costal. Apical breathing was not evident at rest. Minimal dyspnea was perceived during spontaneous speech. The patient also presented a light central deficit of the right 7th cranial nerve, without significant reductions of phono-articulatory abilities. Voluntary cough was elicitable and audible.

The fiberoptic endoscopic evaluation of swallowing (FEES) [6] showed no impairments on larynx structure. The respiratory space was preserved, and vocal cords showed streaks and minimum accumulation of saliva in the anterior portion. During phonation with the vocalization /i/, incomplete adduction of the vocal cords was observed, along with localized air escape at the posterior third.

The larynx had traces of secretions in the piriform sinuses, marginally covering the mucosa (Level II according to the Yale Pharyngeal Residue Severity Rating Scale [7]) and potentially cleared with voluntary swallowing acts. Epiglottic vallecula were adequately cleared (Level I according to the Yale Pharyngeal Residue Severity Rating Scale).

In phonation, on vocalization tests, hypotonia of vocal folds was observed (Figures 1a,b,c), with incomplete vocal cords adduction and air leakage.

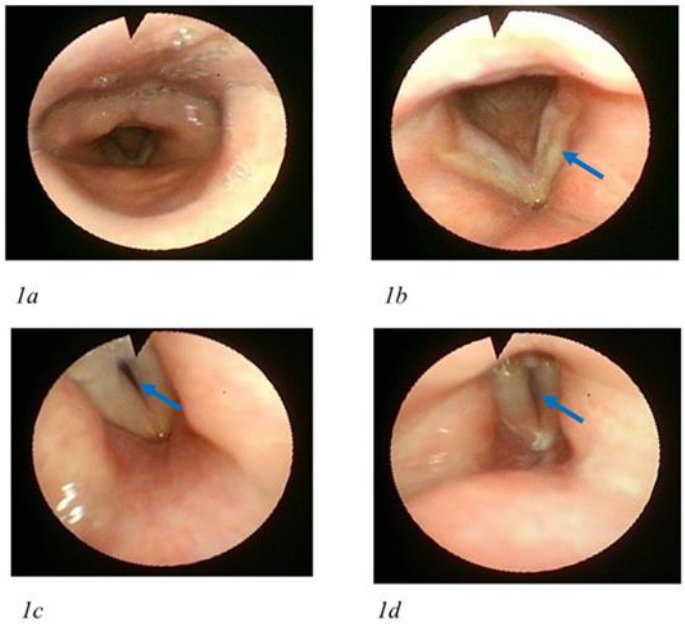


Figure 1: Patient's fiberoptic endoscopic evaluation of swallowing (FEES) examinations at baseline and after treatment

Description. 1a: FEES at baseline, laryngeal aditus; 1b: FEES at baseline, focal folds in abduction at rest, presence of vergeture; 1c: FEES at baseline, vocal folds in adduction during phonation, incomplete adduction; 1d: FEES at end of treatment, persistence of minimal incomplete adduction (arrows indicate the findings)

The breathiness, asthenia, and strain (GIRBAS) scale [8] revealed an overall degree of severe dysphonia (G3) with greater impairment at the level of voice instability (I3), hoarseness (R3) and voice blowing (B3); however, mild impairment was observed at the parameters of asthenic voice (A1) and choked voice (S1).

An audio analysis of voice and speech was performed by the PRAAT software package [9], considering the following indices: Phonatory duration time (sec); Average intensity (dB); Vocal intensity range (dB); Fundamental frequency (Pitch); Jitter; Shimmer; Diplo-phonic voice; AVQUI; Spectrographic classification according to Yanagihara; Spectrogram. All tested indices were found out of standards.

Treatment of dysphonia

In view of the elapsed time and the patient's compliance, it was decided to use the Simeox device within the speech therapy treatment as a tool to stimulate laryngeal motility and sensitivity.

Simeox is a CE marked electro-medical device designed for the bronchial airway clearance through the mechanical effect of very short-lasting negative pressures with frequencies equal to 6 and 12 Hertz, and with increasing intensity from -25 cmH20 to -45cmH20 that act at the level of the bronchial tree, both in its proximal and distal parts. Its mechanical effects are applied during the expiratory phase by increasing the

exhalation flow rate, allowing the mobilization of secretions in the bronchial tree. Current evidence supports its effectiveness in patients with chronic lung diseases associated with mucus hypersecretion and mucus retention, such as bronchiectasis, COPD (Chronic Obstructive Pulmonary Diseases) and Cystic Fibrosis [10-12].

The off-label choice to use this device, after informed patient consent, was made because several studies indicate that the application of negative pressure within the airways during exhalation, both in healthy individuals and in those with neuromuscular disease, may lead to hypopharyngeal constriction with adduction of the vocal cords, with greater beneficial effect for higher pressures [13-15]. According to this preliminary evidence, the use of the Simeox device was considered, in view of its capability to generate intermittent increasing negative pressures and to mechanically stimulate the vocal apparatus. As a final effect, an improvement of mobility of vocal cords was expected without safety concerns.

To date, there are no experiences in the literature regarding the treatment of dysphonia with devices that directly and "locally" stimulate the functionality of the larynx. Among the most frequently cited rehabilitative approaches, indirect stimulations through the adoption of unusual but facilitating postures for vocal emission [15] and the vibration rehabilitation technique using the 'NOVAFON' device [16-19] are reported. The latter involves a medical device placed externally on the neck at the level of the larynx, which produces vibrations ranging from 50 Hz to a maximum of 100 Hz, reaching a depth of 6 cm in the tissues. This vibratory stimulation of the larynx is different and cannot be compared to the administration of Simeox, where stimulation is delivered through a mouthpiece and transmitted directly into the airways.

In this clinical scenario, Simeox was used in the following way: patient in sitting position was required to inspire from the nose at tidal volume (Vt), then after a brief teleinspiratory pause, was required to exhale for 3-4 seconds. During this expiratory phase, Simeox was activated by the patient using of a remote control, and the patient himself was asked to perform a vocalization (/o/) in order to optimize laryngeal recruitment. A Simeox session included 1 cycle of 10 respiratory acts (8 exhalations at 12Hz and 2 exhalations at 6Hz) at increasing power from – 25cmH20 to -45 cmH20.

Treatment involved multidisciplinary team, with joint speech therapist and physical therapist sessions. At the beginning and conclusion of each administration, a voice sample of vocalizing (/a/) and speaking voice (i.e. the Italian phrase “Le mie aiuole sono belle”) was recorded.

Treatment outcome

The patient completed 11 sessions without any adverse event and with total adherence to the program.

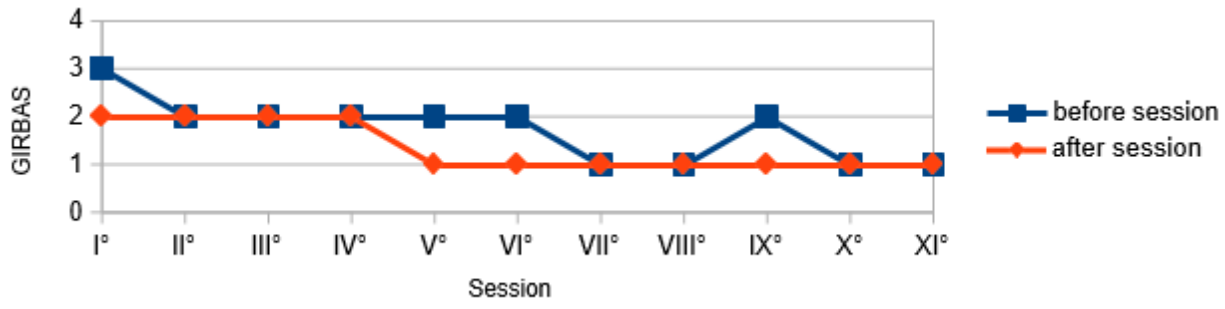
At the end of treatment, FEES showed improvement in chordal adduction in phonation (Figure 1d).

The GIRBAS scale showed an improvement in the overall degree of dysphonia (G1), from 3 (starting of the 1st Simeox session) to 1 (end of the last session) (Figure 2a). Improvements were observed at each session, for all variables, and maintained over time (Table 1).

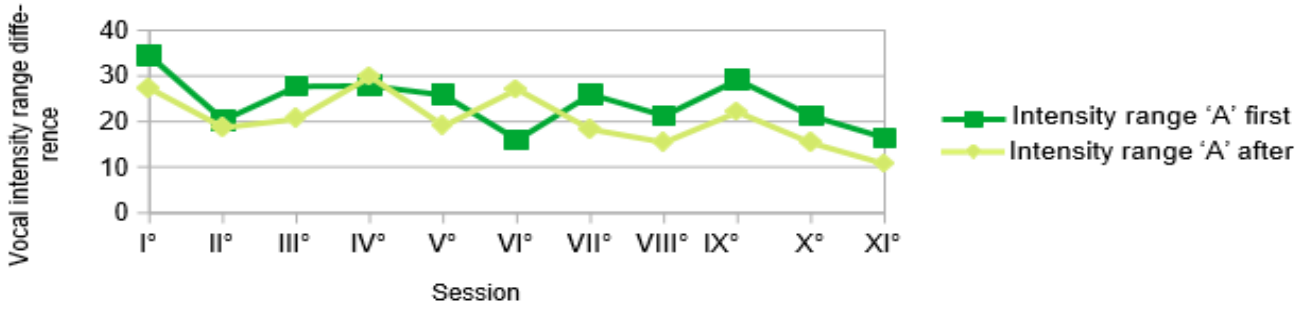
Session	/a/ before SIMEOX T0	/a/ after SIMEOX T0	/a/ before SIMEOX T10	/a/ after SIMEOX T10
G (General Grade of Dysphonia)	3	2	1	1
I (Instability)	3	2	1	1
R (Roughness)	3	3	1	1
B (Breathy)	3	2	1	1
A (Asthenic)	1	0	0	0
S (Strained)	1	1	0	0

Table 1: GIRBAS subscores before and after the first (T0) and the last (T10) Simeox session

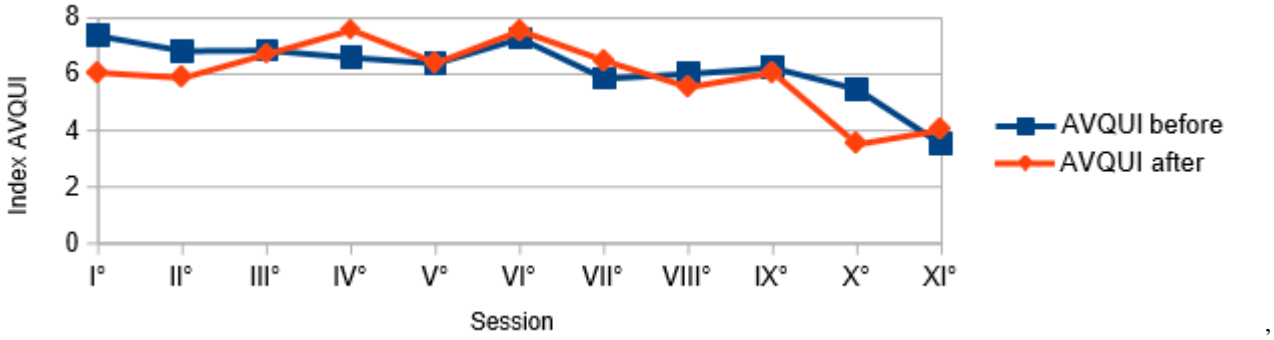
Data obtained by PRAAT analysis are shown in figures 2b and 2 c. A first relevant finding, strongly perceived acoustically and objectified by PRAAT analysis, was the increase in the Mean Intensity Index at vocalizing /a/ immediately after the first use of Simeox; however, this increase appeared not to be stable over time. A stable reduction of the range of vocal intensity throughout the sessions was conversely observed (Figure 2b). The Acoustic Voice Quality Index (AVQUI) [20] is an acoustic measure of voice quality and combines 6 acoustic markers (i.e. Smoothed central peak prominence 'CPPS', Harmonics- to- noise ratio, Shimmer local, Shimmer local dB, Slope of LTAS, Tilt of trendline through LTAS) from speaking voice and sustained vocalizations, thus obtaining a single significant measure of dysphonia severity (euphonic voice AVQUI<2.35). Importantly, AVQUI correlates with auditory-perceptual judgments of voice quality, has high test-retest reliability, and demonstrates high sensitivity to changes in voice quality through voice therapy. The AVQUI index decreased sharply over time, thus indicating an increase in voice quality, and at final evaluation was close to the <2.35 cut-off for normality (Figure 2c).



2a



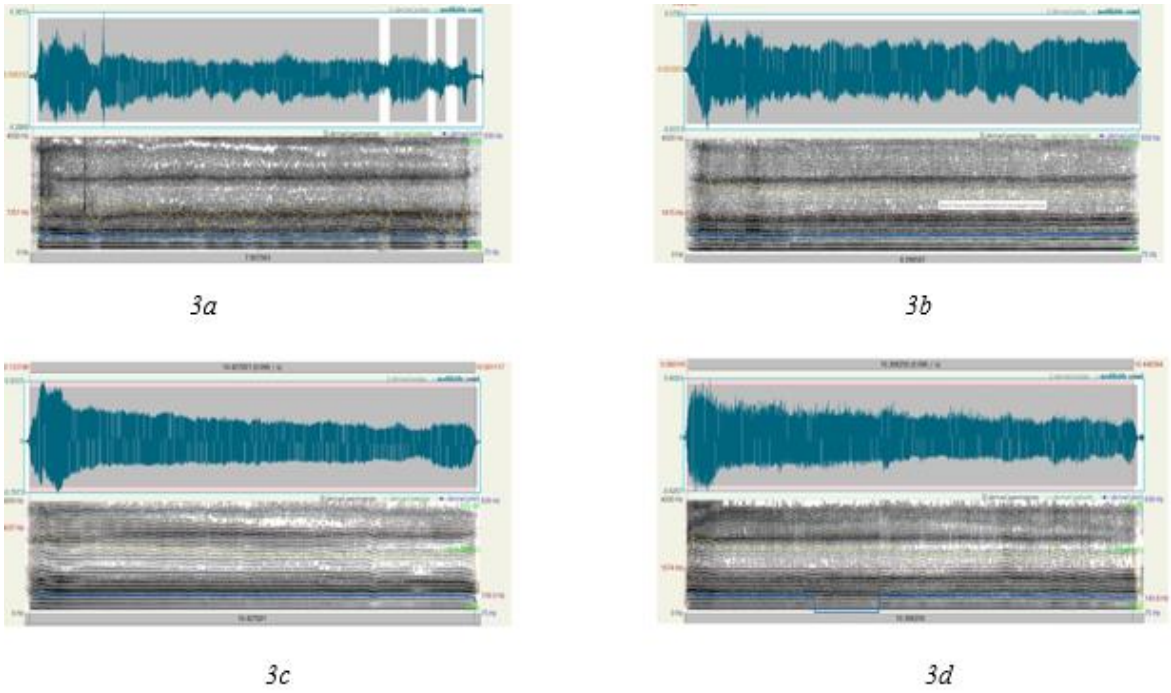
2b



2c

Figure 2: Trend of GIRBAS scores (2a), vocal intensity range trend (dB) (2b), and AVQUI (Voice Quality Index) (2c) during Simeox sessions

The spectrographic representation of the vocalizations confirmed these positive results (Figure 3). The comparison of the two spectrograms recorded on the first day, before and after Simeox execution, showed an immediate improvement in vocal sound and a marked reduction in the noise component. In particular, the spectrogram after Simeox revealed the harmonics up to a frequency of 1415 Hz, which was an increase of about 100 Hz from the starting frequency of 1351 Hz.



3a *3b*

3c *3d*

Figure 3: PRAAT vocalizing A' before and after Simeox treatment

Description. 3a: before first session; 3b: after first session; 3c: before last session; 3d: after last session

The patient continued to rate his voice as 'quite good' and he recognized the improvement as compared to the beginning of speech therapy treatment. At final evaluation by means of the Index of self-assessment of dysphonia, scores obtained on the P, F and E subscales is 10/120 indicated a degree of mild severity (i.e. minimal amount of handicap).

Discussion

Post stroke dysphonia refers to speech and voice disorders caused by paralysis of vocal cords. Speech therapy, often delivered during the course of rehabilitation programs, is the primary treatment for post-stroke dysphonia and is traditionally based on voice therapy, exercises to strengthen the vocal cords and muscles, breathing exercises, vocal hygiene, and compensation strategies [21,22]. Among emerging therapies to treat post stroke dysphonia, several case series have reported improvements with neuromuscular electrical stimulation (NMES) [23], in absence of randomized clinical trials (RCT) giving recommendation for its use during rehabilitation. Moreover, robust evidence is lacking for the application of transcutaneous electrical nerve stimulation (TENS) in the rehabilitation of post stroke dysphonic patients [24]. As a consequence, there is growing interest for other modalities of laryngeal stimulation, aimed at favoring muscle contraction, increasing vocal fold tension, and ultimately improving voice performance.

This case report arises from the joint experience in the field of speech therapy and respiratory physiotherapy, testing the utilization of a device normally used in chronic respiratory diseases for airway clearance to improve speech function. The idea of adopting negative pressure to stimulate laryngeal sensitivity and perception is not new and is based on the "sensory bombardment principle" of the phonatory apparatus. As a novelty, Simeox may deliver both oscillatory vibrations and intermittent negative pressure, leading to increased proprioception and ability of the dysphonic subject to "listen to himself inside". This therapeutic approach could open new opportunities to reinforce traditional rehabilitation programs for post-stroke dysphonia, with the aim to reduce current rates of no-responder.

The use of Simeox for the treatment of dysphonia has never been explored before and therefore in this first experience it was not possible to use pre-existing protocols for outcome analysis. We adopted a pragmatic approach by avoiding complex techniques (such as the recording of voice collection) and excluding indices at high risk of bias (e.g. Fundamental Frequency, Jitter, Shimmer, Diplo-phonic Voice, and Spectrographic Classification according to Yanagihara).

However, our findings suggest an important clinical benefit, with an immediate perceived change in the intensity and quality of the voice. This improvement seems to be confirmed by objective data from endoscopic examination, the spectrographic and the scales of perceived dysphonia by the patient. Moreover, benefits are maintained over time, as indicated by the AVQUI value and voice stability data.

Conclusion

The adoption of intermittent negative pressure with high frequency oscillatory vibrations during exhalation, administered at time intervals, for laryngeal stimulation seems to result in voice improvement in terms of intensity, stability and quality in a subject diagnosed with neurological dysphonia. Further research is needed to evaluate the effectiveness of this adjuvant approach to current speech treatments in larger patient populations and to formally include it among core components of post stroke rehabilitation.

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