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The Lee Silverman Voice Treatment (LSVT®) speech therapy in progressive supranuclear palsy

P. SALE [1], D. CASTIGLIONI [1], M. F. DE PANDIS [2], M. TORTI [3], V. DALL'ARMI [1], F. G. RADICATI [3], F. STOCCHI [3]

[1]Department of Neurorehabilitation, IRCCS San Raffaele Pisana, Rome, Italy
[2]San Raffaele Cassino, Cassino, Frosinone, Italy
[3]Institute for Research and Medical Care, IRCCS San Raffaele Pisana, Rome, Italy

Background. **The Lee Silverman Voice Treatment (LSVT®) was specifically created and tested to comply with the needs of individuals with Parkinson's disease (PD) and other neurological problems. This is a high effort intensive treatment that aims at increasing vocal intensity through the increase of subglottal air pressure, i.e. respiratory effort, for a better cordal adduction and vibration, following the motto "think loud".**
Aim. **The main goal of this study is to inspect the efficacy of LSVT® treatment in progressive supranuclear palsy (PSP) patients.**
Design. **Longitudinal study.**
Setting. **Rehabilitative inpatient unit.**
Population. **Sixteen patients with PSP and 23 patients with idiopathic PD as control were enrolled in the study.**
Methods. **All patients underwent a training consisting in16 sessions of speech therapy following the LSVT® protocol. Initially the two groups of patients had similar voice problems, *i.e.* low volume and bad articulation of speech.**
Results. **A statistically significant improvement was found among the data collected before and after treatment in the PSP and Parkinson groups. Increase in maximum phonation duration and volume of voice in reading were similar in the two groups. Improvement in quality of voice and articulation were more significant in the PD group as compared to the PSP group.**
Conclusion. **These results, along with previous findings, add further support to the generalized therapeutic impact of intensive voice treatment on respiratory and laryngeal functions in individuals with PSP.**
Clinical Rehabilitation Impact. **The positive results, the absence of dropout and collateral effect following this clinical treatments with LSVT technique encouraged to use this technique in PSP patients.**

KEY WORDS: Supranuclear palsy, progressive – Voice disorders - Rehabilitation of speech and language disorders.

Corresponding author: F. Stocchi, Institute for Research and Medical Care, IRCCS San Raffaele, Via della Pisana 235, 00163 Rome, Italy. E-mail: fabrizio.stocchi@sanraffaele.it

Progressive supranuclear palsy (PSP) is a rare progressive neurodegenerative disorder that causes serious and progressive problems with control of gait and balance, along with complex eye movement and thinking problems, swallowing, limb function, and subsequent limitation of activities of daily living.[1, 2] Speech problems are present in almost the totality of PSP patients and they are characterize by a harsh and strained voice with frequent articulatory errors, stuttering, palilalia and variable intensity and rate of speech along with or even outweighing the monotonous speech of Parkinsonism.[3] Moreover the neurogenic shuttering is abnormal dysfluency deriving from damage to the central nervous system and clinically is disorders in the rhythm of speech in which the individual knows precisely what he or she wishes to say but at the time is unable to say it because of an involuntary repetition, prolongation or cessation of a sound [4, 5] and often exhibit executive dysfunction such as difficulties with shifting mental set, problem solving, and abstract thinking.[6] The involvement of brainstem and midbrain structures in PSP probably influences the

clinical manifestation of dysarthria, which is often an early and prominent symptom of PSP.[7] In PSP, dysarthria is a common and often early and prominent sign and is, as opposed to the hypokinetic dysarthria in Parkinson's disease (PD), characterized by signs of spasticity, for example, monotony, harsh voice, imprecise articulation, and slow rate.[8] In particular, in PSP subjects the speech abnormality was more severe comparing to other neurodegenerative diseases such as PD and multisystemic atrophy (MSA), and also had a mixed pattern of hypophonic and spastic components.[4-6] A longitudinal study conducted by Goetz *et al.* monitored a well-characterized PSP population in which unintelligible speech occurred at median disease duration of 71 months, median 44 months after first consultation. Moreover as a composite endpoint, speech/gait accounted for 98% of the sample's first key motor impairment. There are three broad speech symptoms, which may be experienced singly or altogether. These deficits have a negative impact on quality of life and cause considerable impact on patients' social and emotional well-being.[7] In general four approaches to speech therapy are available: behavioral treatment techniques (drill, exercise), instrumental aids including prosthetic and augmentative devices, medication, and surgical procedures.[9-11] In the past years Ramig *et al.* developed the Lee Silverman Voice Treatment (LSVT®) a standardized, research-based speech treatment protocol with established efficacy for PD.[12] LSVT® trains the target of vocal loudness in order to: 1) enhance the voice source, consistent with improving the carrier in the classic engineering concept of signal transmission; 2) use vocal loudness as a trigger for distributed effects (*e.g.*, improved articulation, vocal quality and intonation, and reduced rate) across the speech production system; 3) recalibrate sensorimotor perception of improved vocal loudness; and 4) train a single self-cue and attention to action to facilitate generalization of treatment effects into functional communication.[12] Until now no clinical rehabilitative speech and voice trials were conducted in PSP patients. The objective of this study is to evaluate the efficacy of LSVT® techniques for speech as an innovative approach to train patients with PSP.

Materials and methods

The study was design to have a group of PSP patients and a group of PD patients to prove the validity of the LSVT technique. Only patients complaining of voice or speech problems were enrolled in the study. Eligible PSP and PD patients on stable doses of medication prior to study from inpatient and outpatient rehabilitation centre were recruited. The inclusion criteria were: 1) diagnosis of idiopathic PD by UK Brain Bank criteria, without other significant neurological problems for PD subjects;[16] 2) diagnosis of PSP according to the National Institute of Neurological Disorders and Stroke Society for PSP International Workshop without other significant neurological problems for PSP subjects;[17] 3) between the ages of 30-80 years. The following exclusion criteria were identified; 4) unable to understand instructions required by the study (Informed Consent Test of Comprehension); 5) chronic and on-going alcohol or drug abuse, active depression, anxiety or psychosis that might interfere with use of the equipment or testing; 6) diagnosis of other atypical parkinsonian syndrome. The Ethics Research Committee of the Institute approved the study and the patients underwent the study after signing an informed consent according to the Italian law.

Procedures

The patients were divided into two groups: PD Group (CG) and PSP Group (EG) according to clinical diagnosis.

Clinical assessments

Trained professionals not involved in the research treatment and blind to patients' treatment performed all instrumental and clinical assessments. All outcome assessments were collected in ON phase one hour and half (90 minutes) after the oral assumption of the usual dose of oral PD medication at the start of the treatment (T0) and at the end (T1). Clinical and instrumental outcomes were performed using valid and reliable tools for PD and PSP that include: PSP rating scale, UPDRS and clinical neurological and cognitive evaluation.

Instrumental assessment and training

To assess and to train the hertz reached and the decibel emission we used a commercial Korg OT-12 Orchestral Tuner and a Digital Sound Pressure

Level Meter SM4. The Korg OT-12 Orchestral Tuner has a broad tuning spectrum that covers the pitch ranges of all sounds. The OT-12 features a dual display with both a VU-style meter for accurate pitch indication whit an LCD screen for excellent visibility to use as visual feedback. The broad range of calibration starts from 27.50 Hz (A0) and ends at 4186 Hz (C8). Untrained human voices range from the 2nd to the 5th octave. The second system used is a Digital Sound Level Meter SM4. The SM-4 meter is used to measure sound pressure level (SPL) and to assess the frequency response, the efficiency, and the sound radiation or the noise level. The system is also fit to consider the human hearing characteristics.

Therapeutic intervention

All subjects underwent a preliminary one-hour individual session of experimental LSVT®. In the past years Ramig and colleagues developed the LSVT® a standardized, research-based speech treatment protocol with established efficacy for PD [12] treatment. The LSVT® program utilizes intensive high phonatory effort exercises in order to increase vocal fold adduction. In particular the patients underwent 16 one hour sessions of speech therapy following the LSVT® treatment by a trained and certified therapist (4 times a week for 4 weeks, *i.e.* 4 subsequent days of treatment/three days without treatment with "homework"). All rehabilitative LSVT training was conducted in individual session. The LSVT® treatment is described by Ramig *et al.*[18-23] The individual rehabilitative program includes: 1) enhancing the vocal source (adduction); 2) using phonation as a trigger to increase effort and coordination through stimulating the "loud" global variable (respiratory support); 3) retraining sensory processing during the speech production (increasing fundamental frequency range). Patients are encouraged to maximize phonatory effort and are given frequent encouragement to "think loud" during sustained phonation tasks, reading, and conversational speaking tasks. Attention is given to the respiratory system in the form of general reminders for subjects to take deep breaths "to be loud." The respiratory system is indirectly stimulated during all "think loud" speech tasks. All these tasks follow a gerarchic order of increasing difficulty. All rehabilitative training was conducted in individual session.

Statistical analysis

The Wilcoxon signed-rank test for paired data and the Wilcoxon rank-sum/Mann-Whitney tests were therefore used to test all recorded parameters for differences pre- and post-treatment between PD and PSP patients, and for the presence of pre-post changes in both groups. Statistical significance was set to a P-value <0.05. The analyses conducted on this study were explorative, thus no adjustment for multiple comparisons was applied.

Results

A total of 96 patients were screened, and 39 were enrolled in the study and 57 patients were excluded because not meeting inclusion criteria or declined to participate at the study. Twenty-three subjects (59%) were affected by PD and the remaining 16 subjects (41%) were affected by PSP. Clinical characteristics of the study subjects (N.=39) are reported in Table I. Within each group, no dropouts were recorded during the study period and all the subjects were compliant to treatment. The averages of disease duration were 8 years for PD and 4 years for PSP patients. The mean values at the start of the treatment (T0) and at the end of the treatments were report in Table II. In particular both groups of patients had similar voice problems, i.e. low volume and bad articulation of speech with difference (between the groups) in hertz reached and decibel emission and in the frequency response, efficiency, and sound radiation. At T0 only one significant difference between PD and PSP patients was found in High Fre-

TABLE I.—*Clinical characteristics of the subjects enrolled in the study.*

	PSP-group	Parkinson-group
Subjects (M/F)	11/5	17/6
UPDRS part II scores		26.27±5.65
PSP scores	41±12	

TABLE II.—*Evaluation of the speech therapy: improvement in Parkinson's Disease (PD) patients and in progressive sopraneclear paralysis (PSP) patients (a. p-value from the Wilcoxon signed-rank test for paired data), and difference in PD and PSP groups before and after the treatment (b. p-value from the Wilcoxon rank-sum/Mann-Whitney test).*

	PD			PSP			Difference between groups	
	Pre-Treatment	Post-Treatment	Change P-value[a]	Pretreatment	Post-Treatment	Change P-value[a]	Pretreatment P-value[b]	Post-treatment P-value[b]
High frequencies Max value (Hz)	349.23; 376.85±121.18	381; 414.07±126.52	0.111	261.63; 308.99±110.51	329.63; 359.28±153.28	0.081	0.036†	0.162
High frequencies Mean value (Hz)	311.13; 328.5±104.29	349.23; 378.73±114.6	0.016†	233.47; 269.87±94.44	311.13; 333±138.29	0.029†	0.062	0.215
Low frequencies Min value (Hz)	123.47; 133.48±35.21	103.83; 119.69±31.82	0.663	138.59; 139.26±27.92	123.47; 126.87±20.02	0.061	0.500	0.223
Low frequencies Mean value (Hz)	130.81; 142.2±35.15	113.27; 129.02±30.76	0.304	146.83; 149.53±33.9	130.81; 135.8±25.91	0.259	0.497	0.342
"ah" Emission Min value (dB)	70; 70.61±7.33	80; 81.04±2.87	<0.001*	71; 1.13±5.51	80; 80.63±4.53	<0.001*	0.830	0.863
"ah" Emission max value (dB)	86; 82.83±7.7	94; 93.39±2.82	<0.001*	85; 84.69±5.85	92.5; 92.19±4.18	<0.001*	0.548	0.490
"ah" Emission min value (sec)	7; 7.65±4.11	10; 11.3±5.67	<0.001*	5; 6.13±3.4	9.5; 8.69±4.47	0.038†	0.234	0.178
"ah" Emission max value (sec)	12; 12.87±5.5	16; 16.96±8.43	0.002†	12; 12±4.94	14; 14.19±6.01	0.049†	0.688	0.399
Sentence min value (dB)	77; 75.96±4.12	79; 79.13±3.65	0.002†	79.5; 77.94±5.84	81; 78.88±6.85	0.230	0.132	0.448
Sentence max value (dB)	86; 85.91±5.56	89; 88.57±4.79	0.007†	89; 88.19±4.52	90.5; 89.31±5.62	0.233	0.182	0.465
Reading min value (dB)	74; 71.7±8.22	77; 77.87±5.49	0.006†	68; 68.13±6.44	81; 79.75±8.16	<0.001*	0.063	0.100
Reading max value (dB)	80; 81.26±6.43	85; 85.35±5.09	0.004†	82.5; 81.38±5.04	87.5; 86.38±6.01	<0.001 *	0.943	0.283
Speaking min value (dB)	71; 70.82±5.35	74; 75.17±4.06	0.001†	73; 72.07±6.67	79; 76.83±4.34	0.018†	0.494	0.195
Speaking max value (dB)	79; 78.64±5.47	82; 82.35±4.45	0.008†	81.5; 80.86±6.14	83.5; 83.08±4.78	0.234	0.235	0.393

Hz: Hertz;
dB: decibels;
sec: seconds. Summary Statistics: median;
mean±standard deviation. Statistical significance: P<0.05. †0.001≤P<0.05;
*P<0.001.

quencies Value (P=0.036). The other evaluations did not show statistical difference at T0 between PD and PSP patients. The PSP patients showed significant improvements in High Frequencies Mean Value (Hz) emission (P=0.029) and in "ah" Emission minimum and maximum Value (dB) (P<0.001). Also during the "ah" Emission the minimum (P=0.038) and maximum (P=0.049) value (sec) showed a significant improvement. The Reading Minimum and Maximum Value (dB) (P<0.001) and the Speaking Minimum Value (dB) (P=0.018) showed statistical improvements. Similar to PSP patients, the PD patients showed significant improvements in: High Frequencies Mean Value (Hz) (P=0.016), "ah" Emission Minimum and maximum Value (dB) (P< 0.001), "ah" Emission Minimum and maximum Value (sec) (P< 0.001), Sentence Minimum (P=0.002) and Maximum (P=0.007) Value (dB) and in Reading (P=0.006) and Speaking (P=0.008) minimum and maximum value.

Discussion

LSVT proved to be efficacious also in patients with PSP. The LSVT programs for individuals with PD have been developed and researched over the past 20 years beginning with a focus on the speech motor system (LSVT LOUD).[19] In particular LSVT® helps individuals with PD to "re-calibrate" their motor and perceptual systems so that they are less inclined to under scale (reduce amplitude) speech movement parameters. This multisystemic up scaling of motor output clearly results in larger amplitude of speech that more closely resembles normal speech.[5] LSVT LOUD is a standardized, research-based speech treatment protocol with established efficacy for PD [16-18] but considering the small patient numbers in the trials published, there is insufficient evidence to support or refute the efficacy of any form of SLT over another to treat speech problems in patients with Parkinson's disease.[12, 17-23] Our choice to treat the PSP patients with LSVT technique was secondary to the good results in term of recovery of speech disability that we obtained during the clinical use in PD and in a small number of PSP patients. In particular the absence of dropout and collateral effect following the first clinical treatments of PSP patients with LSVT technique encouraged us to use in this first trial this technique in PSP patients. Recently, basic science research in animal models has documented the value of exercise for improving motor performance and potentially slowing progression of motor symptoms and neural degeneration. Accredited theories of brain reorganization and recovery support the use of early, intensive, repetitive, and context-related exercise as optimal strategies to promote motor relearning and minimize motor deficit. In order to improve the motor and non-motor function, the paradigm of rehabilitation strategies is focused on finalized and task specific training. Even if there is not a widely accepted protocol for speech rehabilitation in PSP patients, the treatments proposed until now vary in type, duration, intensity, and frequency. The present paper is the first study showing the evidence by clinical data that attest the efficacy of a highly intensive and specific treatment to improve the voice and speech impairments in individuals with PSP. The use of PD patients as control is aimed at having an internal procedural control to validate the efficacy of the treatment. The statistical improvements from T0 and T1 obtained in PSP patients in term of mean emission value in Hz and mean emission reading and speaking value in dB are more interesting and promising for future studies. It can be argued that intelligibility is the most critical outcome to be measured in speech and language therapy trials. Our choice to use an instrumental assessment to measure the gain and the recovery is aimed at getting over the present limitation on speech outcome measurement. The reproducibility of our results in Parkinson confirms the validity of our method and our therapists in administering the treatment.

Study limitation

The very small sample size and the absence of a control group do not allow having a high significance in the results, but the global improvement of assessments could encourage to plan a large RCT. Moreover a cognitive evaluation assessed by a MOCA or Frontal Assessment battery are recommended and may be add in the future studies to correlate the cognitive state and the effective gain duo to the LSVT therapy.

It should be noticed, however, that there are some important elements, especially in PSP patients, which cannot be measured objectively, but nevertheless have a significant impact, possibly negative, on the patients' vocal and speech performance. These are individual neuropsychological reactions to the

effort of voice emission and articulation mainly during repeated reading of functional phrases, which somehow represent a "test" for the patient, and may cause stress.

Conclusions

These results, along with previous findings, add further support to the generalized therapeutic impact of intensive voice treatment on respiratory and laryngeal functions along with improvement of speech intelligibility in individuals with PSP individual.

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Conflicts of interest.—The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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