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ORIGINAL ARTICLE

Effects of Lee Silverman Voice Treatment (LSVT LOUD) on Swallowing in Patients with Progressive Supranuclear Palsy: A Pilot Study

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Objectives: Progressive supranuclear palsy (PSP) is an uncommon progressive neurodegenerative disease with no effective cure at present. The initial symptoms resemble those of Parkinson's disease; however, the prevalence of PSP is about one-tenth that of Parkinson's disease. In many cases, dysphagia is severe, and the development of dysphagia is an early predictor of life expectancy. The aim of the current study was to define the effects of Lee Silverman Voice Treatment (LSVT LOUD) on swallowing and voice/speech in seven patients with PSP. **Methods**: Each patient underwent swallowing and voice/speech evaluations before and after 4 weeks of LSVT. Swallowing motility disorders were defined, temporal measures of swallowing were determined by videofluoroscopic evaluation, and voice measures of maximum phonation and speech intelligibility in reading and monologue were examined. **Results:** After LSVT, the median duration of opening of the upper esophageal sphincter (from the beginning of the posterior movement of the bolus to upper esophageal sphincter opening) on videofluoroscopy was significantly shortened from 0.42 to 0.38 s (Wilcoxon signed-rank test P=0.016). The oral transit duration was decreased in five patients, but the decrease was not significant. Voice changes after LSVT included increases in voice intensity and in sustained duration were not significant. **Conclusion:** In this small study, it was found that LSVT may improve swallowing functions in patients with PSP.

Key Words: dysphagia; LSVT; progressive supranuclear palsy; voice treatment

INTRODUCTION

Progressive supranuclear palsy (PSP) is a Parkinsonism-related neurodegenerative disease the features of which include onset in middle age or older, a tendency for progressive Parkinsonism, frequent falls, supranuclear gaze palsy, and dementia. Patients have a high risk of falling when frozen gait and postural instability are prominent. As the disease progresses, articulation and swallowing disorders, dementia characterized by impaired memory recall and slowness of thought, and decreased attentiveness become evident. The prevalence of PSP is reportedly 10–20 per 100,000, about one-tenth of that of Parkinson's disease (PD); however, PSP has become increasingly evident in surveys in Japan (Japan Intractable Disease Information Center).[1)]

In PSP, there is a loss of neurons in the globus pallidus,

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subthalamic nucleus, cerebellar dentate nucleus, red nucleus, substantia nigra, and brainstem tegmentum. Abnormally phosphorylated tau protein accumulates in neurons and glial cells; however, the etiology is unclear, and medication is not effective.

Diagnosis is based on the National Institute of Neurological Disorders and Stroke and the Society for PSP diagnostic criteria 1996[2] and the Movement Disorder Society PSP criteria 2017.[3] The initial symptoms of PSP are often parkinsonian symptoms. Compared with PD, regular falls, unintelligible speech, and cognitive impairment occur earlier in PSP.[4] Furthermore, swallowing and speech disturbances occur earlier, progress more rapidly, and become more severe in PSP than in PD, and swallowing problems are associated with a worse prognosis.[5] The early onset of dysphagia (within 2 years), cognitive impairment, older age at onset, and time to disability have been found to be predictors of shorter survival.[6,7]

Based on a search of the literature, we found no evidence of any effective treatment for dysphagia in PSP. The early stage of the clinical presentation of swallowing disorders is similar to that in PD; however, there is no specific pattern of dysphagia among other parkinsonism disorders.[8,9]

Speech disturbance in PSP is of mixed type with hypokinetic and spastic components, and is different from the hypokinetic speech performance in PD.[10] Moreover, primary progressive aphasia of nonfluency/agrammatism is characteristic, and nonsense words and frontal lobe symptoms are seen in PSP.[11]

In PD, short- and long-term efficacy data have been published supporting the effectiveness of the Lee Silverman Voice Treatment program (LSVT LOUD, hereafter LSVT).[12–15] In early-stage PD, there is high-level evidence of the effectiveness of LSVT for speech.[13] The LSVT program was designed to improve the perceptual characteristics of the voice by targeting the underlying motor disorder associated with the disease. LSVT programs include a combination of targeting increased amplitude (loudness in the speech motor system) and a focus on sensory recalibration to help patients recognize that movements with increased amplitude are within normal limits, even if they "feel too loud."

This study was conducted because there is no evidence of the effectiveness of treatment for dysphagia in PSP, but in PD, there are studies showing the effectiveness of LSVT for dysarthria and dysphagia.[16,17] Furthermore, in PSP, one study showed that LSVT is effective for speech.[10] The current study was done to evaluate whether LSVT improves swallowing in patients with PSP.

METHODS

Subjects

The participants included in this study were seven patients with PSP diagnosed using the diagnostic criteria[2,3] between April 2017 and March 2019 who visited our hospital regularly. These patients had dysphagia or dysarthria, a Mini Mental State Examination (MMSE) score greater than 20, no head or neck cancer, and no other neurological disorders that may affect swallowing.

LSVT Therapy

The patients underwent LSVT four times a week for 4 weeks from an LSVT-certified speech–language–hearing therapist (SLHT). Therapy was given for 50–60 min at each session, and self-treatment was performed every day.

During each LSVT therapy session, patients practiced daily exercises, including drills aimed at enhancing the maximum duration of sustained vowel phonation, the maximum fundamental frequency range, and the maximum functional speech loudness. Patients were also trained to use a louder voice while speaking, to accurately judge their loudness, and "to feel effort, feel loudness—that is what it needs to feel like when you talk so people understand you." In addition, all patients did daily homework and carry-over exercises focusing on "think loud."

Evaluations of Swallowing and Voice/Speech

Evaluations of swallowing and voice/speech were performed before starting LSVT and immediately after 4 weeks (16 sessions) of LSVT in each patient.

Swallowing

The swallowing function was evaluated independently by two SLHTs using videofluoroscopy (VF). These evaluations included qualitative evaluation (residue, aspiration/penetration) and quantitative evaluation (time-phase analysis). The VF study of oropharyngeal swallowing was completed using the standard protocol of the Japanese Society of Dysphagia Rehabilitation (JSDR).[18] The VF examination was conducted with the patients sitting in a chair or wheelchair. The protocol consisted of drinking 3 ml of barium liquid, swallowing 3 ml of barium crushed jelly, and swallowing yogurt (paste) coated with barium. During this radiographic study, patients were viewed in the lateral plane. The videofluoroscopic studies were recorded on a digital video recorder for later analysis.

Swallowing measurements: VF video images were re-

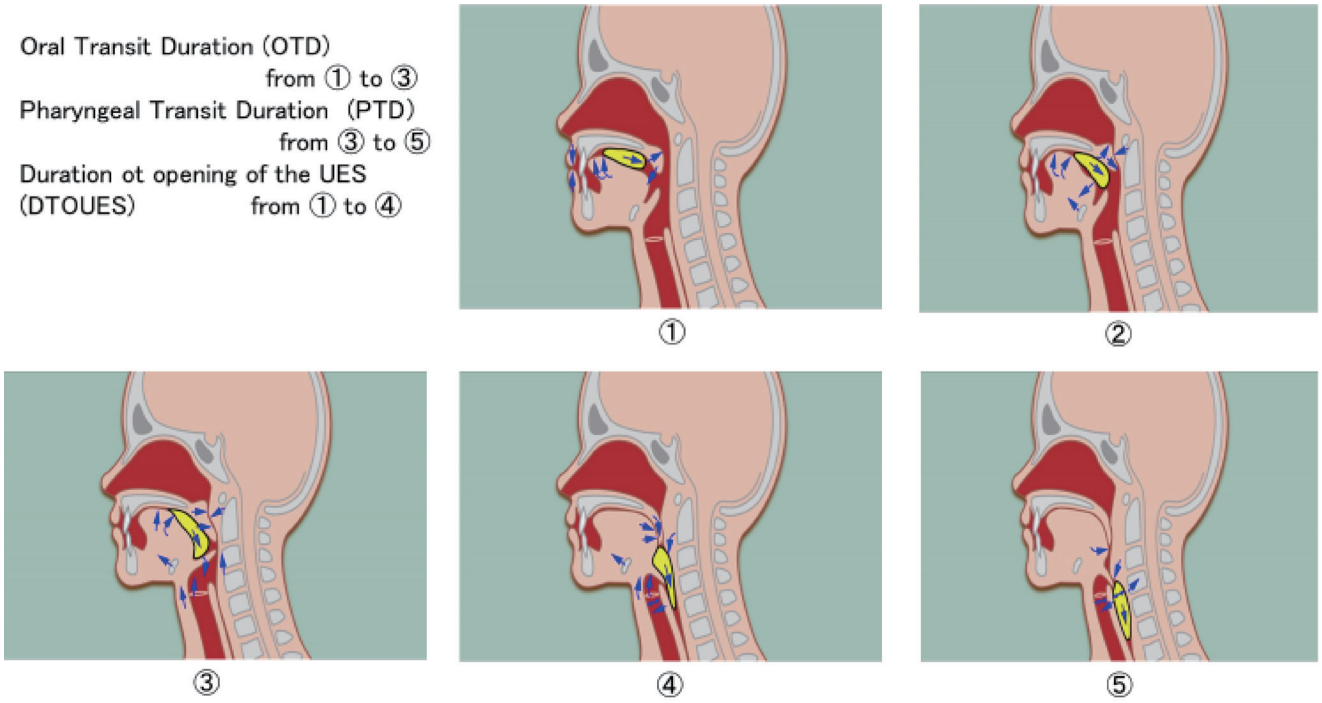

Fig. 1. Quantitative measurement methods for VF.
OTD: from when the bolus first begins posterior movement in the oral cavity to when the head of the bolus first reaches the ramus of the mandible. PTD: from when the head of the bolus first reaches the ramus of the mandible to when the tail, or the end of the bolus, passes through the upper esophageal sphincter (UES). DTOUES: from when the bolus first begins posterior movement in the oral cavity to when the UES opens.

corded, and data reduction involved two types of analysis: qualitative evaluation and quantitative evaluation, using standard JSDR protocols.[18] Qualitative evaluation involved three scores in which residue in the oral and pharyngeal cavity and identification of penetration/aspiration during oropharyngeal swallow were evaluated for jelly, yogurt, and liquid, and the worst score was used. Quantitative evaluation consisted of temporal measurements of the oropharyngeal swallowing of 3 ml of liquid. Motility disorders were identified by reviewing the VF video of each swallow. Temporal measurements and observations were completed for each swallow according to the method developed by Lof and Robbins.[19] Quantitative measurements consisted of the oral transit duration (OTD, measured from when the bolus first begins posterior movement in the oral cavity to when the head of the bolus first reaches the ramus of the mandible), the pharyngeal transit duration (PTD, measured from when the head of the bolus first reaches the ramus of the mandible to when the tail, or the end of the bolus, passes through the upper esophageal sphincter [UES]), and the duration to opening of the upper esophageal sphincter (DTOUES, measured from when the bolus first begins posterior movement in the oral cavity to when the UES opens) (**Fig. 1**).

Two SLHTs analyzed the VF data independently. No clinician who provided LSVT therapy was involved in the swallowing analysis.

Voice/Speech

The evaluations consisted of assessment of voice function and speech intelligibility before and after 4 weeks of LSVT. Tasks completed during each voice recording session were maximum phonation of the vowel /a:/, with maximally sustained phonation of the vowel with sufficient phonatory effort; reading the “North wind and sun” (in Japanese); and a monologue during which the patients talked about their happiest day.

Maximum phonation for the vowel /a:/ was examined as the sustained duration and sound pressure level analyzed using Praat (speech analysis software that was designed and continues to be developed by Paul Boersma and David Weenink of the University of Amsterdam). The intelligibility of speech (reading and monologue) was evaluated by four

Table 1. Patient characteristics

Case number	Age	Sex	Disease duration (years)	ADL Modified Rankin scale
1	59	F	10	4
2	74	M	4	3
3	74	M	5	4
4	79	F	4	5
5	80	M	7	4
6	80	M	4	3
7	92	M	2	4

ADL, activities of daily living.

SLHTs using a 5-point rating scale (1: intelligible to 5: not intelligible).[20] The mean value of the rating scale of the four SLHTs' evaluations was used.

Statistical Analysis

Comparisons of VF qualitative data and quantitative data consisting of time phase, voice, and speech intelligibility over time were performed using a non-parametric, paired, two-group, Wilcoxon signed-rank test comparing each pair of values in the control group at 0 weeks and in the treatment group at 4 weeks. Considering the issue of multiplicity resulting from pairwise multiple comparisons, P values were adjusted using the Bonferroni method. Probabilities were calculated using accurate P value computations, and statistical testing was two-tailed with a significance level of 0.05. In preliminary analyses, normality testing of variable data was performed using the Shapiro-Wilk test. Statistical analysis was performed using IBM SPSS Statistics 24.0 for Windows (IBM Corporation, Chicago, IL, USA).

Ethics

This study was approved by the institutional review board (Kansai Rosai Hospital, number 16c35g). The Ethical Guidelines for Medical and Health Research Involving Human Subjects were followed.

RESULTS

Seven patients (age range 59–92 years) with PSP according to the diagnostic criteria were referred for LSVT by the neurologist and participated in this study (**Table 1**). The disease duration was 2–10 years, and the modified Rankin Scale score was 3–5. All study patients provided written, informed consent to undergo VF examination and participate in this study.

No patient's medication regimen or physiotherapy rehabilitation program was changed during the 4 weeks of participation in this study. Patients continued being treated with LSVT alone or at least without other combination therapies that may affect dysphagia (e.g., conventional speech and swallowing therapy).

Swallowing on VF

On qualitative analysis, oral residue decreased in two subjects but increased in one. The penetration/aspiration score worsened in two subjects and was unchanged in the others. Pharyngeal residue decreased in two subjects but increased in two subjects. These changes were not significant. On quantitative analysis (**Fig. 2**), the median DTOUES on VF was significantly shorter, from 0.42 to 0.38 s (Wilcoxon signed-rank test P=0.016) after LSVT. OTD was shorter in five of seven subjects, but not significantly so. PTD was shorter in only two subjects.

In the present study, one participant fell during the 4-week period of LSVT and broke some ribs; however, he continued the LSVT drills uninterrupted with taking analgesics. This subject's OTD and DTOUES were shortened.

Voice and Speech

The duration of maximum phonation for the vowel /a:/ increased in 6 patients, but the change was not significant (**Fig. 3**). Neither the sound pressure level of maximum phonation nor speech intelligibility showed significant improvement (**Fig. 3**). In one participant, we could not evaluate the duration and the sound pressure of maximum phonation for the vowel /a:/, because his voice was very small before and after LSVT (not changed).

DISCUSSION

This is the first report of the effectiveness of LSVT for dysphagia in patients with PSP. After 4 weeks of LSVT, the me-

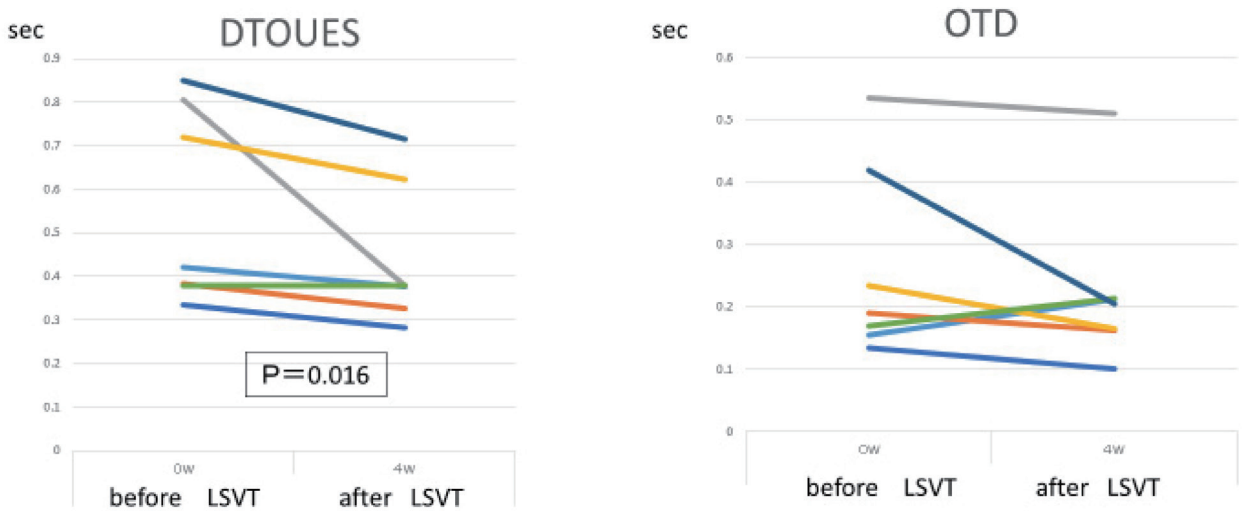

Fig. 2. The duration of opening of the upper esophageal sphincter (DTOUES) and the oral transit duration (OTD) before and after LSVT in patients with PSP. After LSVT, the median DTOUES on VF was significantly shorter (Wilcoxon signed-rank test P=0.016), and OTD was shorter in five of seven patients, but the difference was not significant.

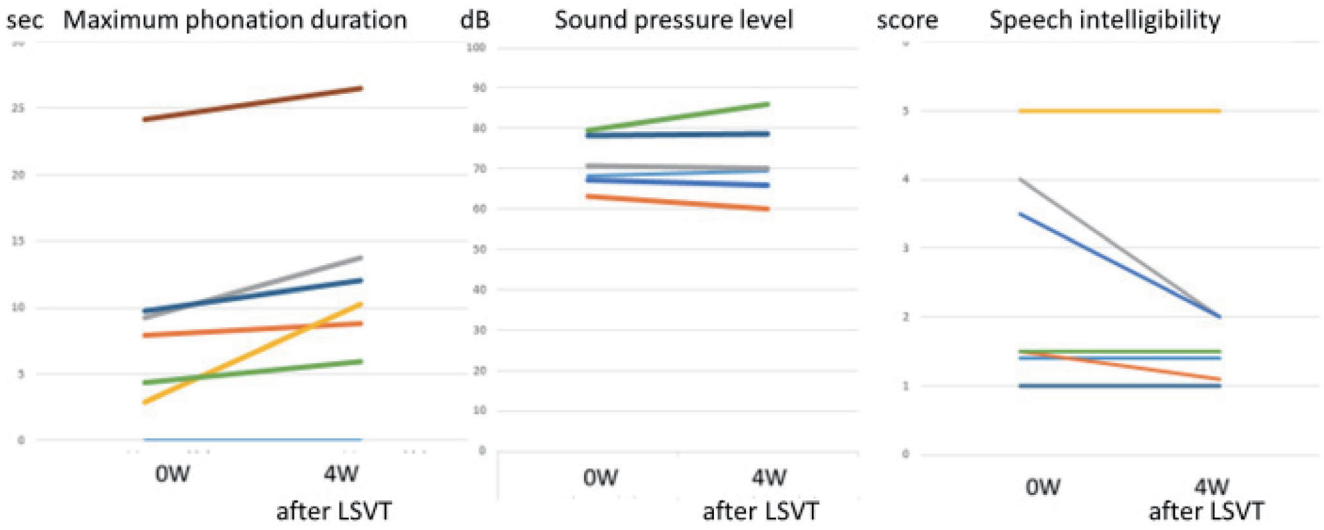

Fig. 3. Maximum phonation duration, the sound pressure level, and speech intelligibility in reading and monologue before and after LSVT in patients with PSP. The duration of maximum phonation for the vowel /a:/ increased in 6 patients, but not significantly.

dian DTOUES on VF was significantly shortened; however, changes in voice and speech after LSVT were not significant.

The initial symptoms of PSP resemble those of PD. For speech and dysphagia in PD, there is evidence of the effectiveness of LSVT.[13,16,17] For speech, in a previous PSP study, LSVT increased the maximum phonation duration and the volume of the voice.[10] Consequently, this study was conducted to evaluate whether LSVT improves swallowing in patients with PSP.

In this small study, voice and speech did not improve significantly. PSP involves widespread tau pathology, and patients have a mixed type of dysarthria with both hypokinetic and spastic components and frontal lobe symptoms. Therefore, speech in PSP differs from the hypokinetic speech performance of PD patients.[10,11] These complicating factors may explain why LSVT was not significantly effective for voice and speech in the present study.

For swallowing, DTOUES was significantly shortened, and there was a tendency for oral phase disturbances to improve more than pharyngeal phase disturbances. These results are

consistent with those of previous LSVT studies in PD.[15–17]

A change in brain activity during voluntary swallowing after LSVT was reported in a positron emission tomography study.[21] That study found that the therapeutic effect of LSVT in PD resulted from a shift in cortical activity to the right hemisphere. In another study, the sensorimotor cortex displayed strong degrees of interhemispheric asymmetry in volitional swallowing in healthy volunteers.[22] In our view, these mechanisms contribute to the improvement of swallowing in the oral phase (voluntary swallowing) rather than the pharyngeal phase. In the current study, we believe that pharyngeal residue and aspiration did not change because of the small effect of LSVT on pharyngeal movement.

In PD, other than LSVT, effective dysphagia treatment was reported in 20 PD patients after 4-week rhythm training using a metronome. OTD was shortened in a cross-over study.[23] Another study involved randomized 4-week expiratory muscle strength training in 60 PD patients, and it showed that the penetration/aspiration score on VF improved.[24] These treatments are worth investigating in PSP patients in the future. Furthermore, in PSP, abnormal eating behaviors (for example, keeping food in the mouth and spitting out food) induce more severe dysphagia. In the future, it will be necessary to develop more effective treatments for the eating habits of PSP patients.

Limitations

PSP is an uncommon disease and has features of Parkinsonism. Diagnostic criteria have been developed, but it is difficult to diagnose PSP in the early stage because of its fast progression. Consequently, only a few PSP patients could be recruited for the current LSVT study.

Conclusion

This is the first report of the effect of LSVT on dysphagia in PSP. PSP is an uncommon progressive neurodegenerative disease that tends to be diagnosed at an advanced stage. Therefore, it was difficult to accumulate evidence about the effect of the treatment on dysphagia, especially in the early stage of PSP. In the present study, it was found that LSVT may be effective in improving swallowing function. Further research is needed to accumulate evidence for the effect of LSVT on swallowing and to determine whether LSVT changes dysphagia in early-stage PSP.

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CONFLICTS OF INTEREST

The authors have no conflicts of interests directly relevant to the content of this article.

REFERENCES

1. Japan Intractable Disease Information Center: 2020. [in Japanese] https://www.nanbyou.or.jp/entry/4115 Accessed 7 Nov 2020
2. Litvan I, Agid Y, Calne D, Campbell G, Dubois B, Duvoisin RC, Goetz CG, Golbe LI, Grafman J, Growdon JH, Hallett M, Jankovic J, Quinn NP, Tolosa E, Zee DS: Clinical research criteria for the diagnosis of progressive supranuclear palsy (Steele-Richardson-Olszewski syndrome): Report of the NINDS-SPSP International Workshop. Neurology 1996;47:1–9. DOI:10.1212/WNL.47.1.1, PMID:8710059
3. Höglinger GU, Respondek G, Stamelou M, Kurz C, Josephs KA, Lang AE, Mollenhauer B, Müller U, Nilsson C, Whitwell JL, Arzberger T, Englund E, Gelpi E, Giese A, Irwin DJ, Meissner WG, Pantelyat A, Rajput A, van Swieten JC, Troakes C, Antonini A, Bhatia KP, Bordelon Y, Compta Y, Corvol JC, Colosimo C, Dickson DW, Dodel R, Ferguson L, Grossman M, Kassubek J, Krismer F, Levin J, Lorenzl S, Morris HR, Nestor P, Oertel WH, Poewe W, Rabinovici G, Rowe JB, Schellenberg GD, Seppi K, van Eimeren T, Wenning GK, Boxer AL, Golbe LI, Litvan I, Movement Disorder Society-endorsed PSP Study Group: Clinical diagnosis of progressive supranuclear palsy: The Movement Disorder Society criteria. Mov Disord 2017;32:853–864. DOI:10.1002/mds.26987, PMID:28467028
4. O'Sullivan SS, Massey LA, Williams DR, Silveira-Moriyama L, Kempster PA, Holton JL, Revesz T, Lees AJ: Clinical outcomes of progressive supranuclear palsy and multiple system atrophy. Brain 2008;131:1362–1372. DOI:10.1093/brain/awn065, PMID:18385183
5. Nath U, Ben-Shlomo Y, Thomson RG, Lees AJ, Burn DJ: Clinical features and natural history of progressive supranuclear palsy: a clinical cohort study. Neurology 2003; 25; 60(6):910–916. DOI:10.1212/01.wnl.0000052991.70149.68.

6. Glasmacher SA, Leigh PN, Saha RA: Predictors of survival in progressive supranuclear palsy and multiple system atrophy: a systematic review and meta-analysis. J Neurol Neurosurg Psychiatry 2017;88:402–411. DOI:10.1136/jnnp-2016-314956, PMID:28250027
7. dell'Aquila C, Zoccolella S, Cardinali V, de Mari M, Iliceto G, Tartaglione B, Lamberti P, Logroscino G: Predictors of survival in a series of clinically diagnosed progressive supranuclear palsy patients. Parkinsonism Relat Disord 2013;19:980–985. DOI:10.1016/j.parkreldis.2013.06.014, PMID:23968651
8. Sulena GD, Gupta D, Sharma AK, Singh B: Clinical profile of dysphagia in patients with Parkinson's disease, progressive supranuclear palsy and multiple system atrophy. J Assoc Physicians India 2017;65:32–37. PMID:28799303
9. Johnston BT, Castell JA, Stumacher S, Colcher A, Gideon RM, Li Q, Castell DO: Comparison of swallowing function in Parkinson's disease and progressive supranuclear palsy. Mov Disord 1997;12:322–327. DOI:10.1002/mds.870120310, PMID:9159726
10. Sale P, Castiglioni D, De Pandis MF, Torti M, Dall'armi V, Radicati FG, Stocchi F: The Lee Silverman Voice Treatment (LSVT®) speech therapy in progressive supranuclear palsy. Eur J Phys Rehabil Med 2015;51:569–574. PMID:26138088
11. Goetz CG, Leurgans S, Lang AE, Litvan I: Progression of gait, speech and swallowing deficits in progressive supranuclear palsy. Neurology 2003;60(6):917–922.
12. Ramig LO, Countryman S, O'Brien C, Hoehn M, Thompson L: Intensive speech treatment for patients with Parkinson's disease: short- and long-term comparison of two techniques. Neurology 1996;47:1496–1504. DOI:10.1212/WNL.47.6.1496, PMID:8960734
13. Ramig LO, Sapir S, Fox C, Countryman S: Changes in vocal loudness following intensive voice treatment (LSVT) in individuals with Parkinson's disease: a comparison with untreated patients and normal age-matched controls. Mov Disord 2001;16:79–83. DOI:10.1002/1531-8257(200101)16:1<79::AID-MDS1013>3.0.CO;2-H, PMID:11215597
14. Ramig LA, Fox C: The integral role of speech production in the science and treatment of Parkinson's disease. Mov Disord 2012;27:811–813. DOI:10.1002/mds.25046, PMID:22674515
15. Ramig LO, Sapir S, Countryman S, Pawlas AA, O'Brien C, Hoehn M, Thompson LL: Intensive voice treatment (LSVT) for patients with Parkinson's disease: a 2 year follow up. J Neurol Neurosurg Psychiatry 2001;71:493–498. DOI:10.1136/jnnp.71.4.493, PMID:11561033
16. Sharkawi AE, Ramig L, Logemann JA, Pauloski BR, Rademaker AW, Smith CH, Pawlas A, Baum S, Werner C: Swallowing and voice effects of Lee Silverman Voice Treatment (LSVT): a pilot study. J Neurol Neurosurg Psychiatry 2002;72:31–36. DOI:10.1136/jnnp.72.1.31, PMID:11784821
17. Miles A, Jardine M, Johnston F, de Lisle M, Friary P, Allen J: Effect of Lee Silverman Voice Treatment (LSVT LOUD®) on swallowing and cough in Parkinson's disease: a pilot study. J Neurol Sci 2017;383:180–187. DOI:10.1016/j.jns.2017.11.015, PMID:29246611
18. The fourth version of VF standard procedure (detailed version) [in Japanese] Japanese Dysphagia Rehabil 2014;18(2):166–186. https://www.jsdr.or.jp/wp-content/uploads/file/doc/VF18-2-p166-186.pdf
19. Lof GL, Robbins J: Test-retest variability in normal swallowing. Dysphagia 1990;4:236–242. DOI:10.1007/BF02407271, PMID:2209099
20. Itoh M, Furukawa M: Speech intelligibility of glossectomized patients with reconstruction using myocutaneous flap. Jpn J Logop Phoniatr 1991;32:347–353. DOI:10.5112/jjlp.32.347
21. Narayana S, Fox PT, Zhang W, Franklin C, Robin DA, Vogel D, Ramig LO: Neural correlates of efficacy of voice therapy in Parkinson's disease identified by performance-correlation analysis. Hum Brain Mapp 2010;31:222–236. DOI:10.1002/hbm.20859, PMID:19639554
22. Hamdy S, Rothwell JC, Brooks DJ, Bailey D, Aziz Q, Thompson DG: Identification of the cerebral loci processing human swallowing with $H_2(15)O$ PET activation. J Neurophysiol 1999;81:1917–1926. DOI:10.1152/jn.1999.81.4.1917, PMID:10200226
23. Nozaki S, Matsui T, Umaki Y, Yoshikawa H, Kaneto T, Sugishita S, Imai T, Fukuoka T, Oguro D, Inokawa M, Domen K, Daimon T: Rhythm therapy with a metronome to treat dysphagia in patients with Parkinson's disease. Deglutition 2012;1:400–412.
24. Pitts T, Bolser D, Rosenbek J, Troche M, Okun MS, Sapienza C: Impact of expiratory muscle strength training on voluntary cough and swallow function in Parkinson disease. Chest 2009;135:1301–1308. DOI:10.1378/chest.08-1389, PMID:19029430