Singing in groups for Parkinson's disease (SING-PD): A pilot study of group singing therapy for PD-related voice/speech disorders

Ludy C. Shih a,*, Jordan Piel b, Amanda Warren b, Lauren Kraics a, Althea Silver a, Veronique Vanderhorst a, David K. Simon a, Daniel Tarsy a

a Department of Neurology, Beth Israel Deaconess Medical Center and Harvard Medical School, Boston, MA 02215, United States
b Department of Rehabilitation Services, Beth Israel Deaconess Medical Center, Boston, MA 02215, United States

1. Introduction

Parkinson's disease (PD) related voice and speech impairment significantly impact quality of life. The majority of PD patients will experience a PD-related voice/speech disorder [1]. These and other non-motor impairments have been shown to affect overall quality of life ratings. LSVTLOUD (Lee Silverman Voice Therapy) has shown to benefit PD-related voice impairment [2].

Common characteristics are typically observed in parkinsonian speech, secondary to dysfunction of the extrapyramidal system, but also due to various sensorimotor deficits which may also play a role. Numerous studies have observed significant physiologic changes in the voice and speech mechanism including decreased neural drive to laryngeal muscles, decreased range of motion of lip and tongue muscles, decreased glottic and velopharyngeal port closure, reduced respiratory capacity and coordination of movements, and reduction in size and peak velocity of jaw movements [3–7]. These in turn result in decreased loudness, breathy vocal quality, short phonation time, and reduced speech intelligibility. Decreased ranges of pitch and loudness limit inflection and the ability to convey emotion in speech. Importantly, impaired laryngeal sensorimotor processing, self-perceived vocal loudness, internal cueing, and vocal vigilance all play critical roles in the overall mechanism of parkinsonian voice dysfunction [6,8].

Multiple well-executed studies on LSVT show that subjects with PD can improve parameters of voice production and communication function including glottal closure, vocal loudness, speech intelligibility, and voice handicap [9] by means of regular, high-intensity vocal exercises, and that these improvements may last for up to two years and longer [2]. LSVT is an intensive, high effort training program which may be effective by means of activity-dependent neural plasticity enabling retraining of movement amplitude scaling [6]. Four 1-h long sessions four times weekly over four weeks is the standard treatment frequency in LSVTLOUD but may be given less frequently over an extended period of time in the LSVT Extended version. The main goal of LSVT is increased healthy vocal loudness, measured as the sound pressure level (SPL). LSVTLOUD encourages maximal effort and multiple repetitions of sustained vowel sounds during basic and more complex vocal movements with increasing complexity of words, phrases, sentences, reading passages and ultimately conversation [6]. Training vocal loudness appears to have benefits on other physiologic voice parameters such as articulation [10], intonation [11], and other important functions such as facial expression [12] and swallowing [13]. In addition, LSVT retraining sensory perception and internal
cuing and is administered with dosage and intensity consistent with principles that drive activity-dependent neural plasticity [14]. However, despite the existence of effective, evidence-based voice and speech treatment programs such as LSVT, perhaps as few as 3–4% of PD patients with voice and speech complaints actually participate in treatment, though this estimate was made well over a decade ago [11]. Common barriers to participation in speech therapy for PD patients may include the perception that speech therapy programs are too intensive or may not be engaging enough to sustain a long-term commitment to practice and exercise at home, as well as limited clinician availability to deliver individual therapy. However, it appears that recent efforts to incorporate telepractice, software-based training programs, and expanded efforts to certify and train clinicians online may help maximize delivery of therapy [15,16].

Music based rehabilitative strategies have been used in other neurologic conditions which impair language and speech, such as autism and aphasia resulting from stroke [17,18]. The rationale for music-based training emphasizes improvements in auditory perception and feedback in autism, whose sensory feedback and perception is known to be impaired. In the case of aphasia resulting from hemispheric stroke, reassignment of language areas to remaining healthy brain in the unaffected hemisphere may be responsible, though it remains to be seen whether these processes are relevant for PD patients whose voice and speech dysfunctions may be more subcortically based and may involve more motor than cognitive impairment. Potential therapeutic mechanisms of singing that may be more relevant to PD are based on observations that singing may naturally promote and intensify aspects of voice and speech production [19]. Singing may encourage louder voice production than does regular speech and may help patients learn to develop and train their respiratory capacities. Additionally, pitch variability, range, and different song tempos singing may improve intonation, timing and speech rates [19]. Singing may also impact the entire integration and coordination of the respiratory, phonatory and articulation aspects of communication, since singing involves slower syllable and word articulation compared to speaking [17]. Additionally, singing may activate the limbic system, whose connections to various subcortical networks involved in the regulation of vocal intensity control [6].

Organized singing groups for PD have been reported in the lay press to be popular and may provide an effective alternative to standard therapies. There have been two prior studies on the use of singing in treatment of voice and speech disorders associated with Parkinson’s disease. Haneishi et al. found that a music therapy voice protocol did not significantly improve speech intelligibility and vocal intensity in 4 subjects with PD who were given 12–14 h of Music Therapy Voice Protocol (MTVP), in three 1-h long sessions weekly for about 4–5 weeks [19]. MTVP consisted of vocal exercises for 20 min including deep breathing, followed by 15 min of singing, usually 2–3 songs of varying range, word complexity and phrase length. The rest of the sessions consisted of other non-singing phonation and speech exercises. Additionally, maximum duration, vocal range, fundamental frequency and fundamental frequency variability did not show any improvement following the 12–14 session treatment when corrected for multiple comparisons. A recent study by Di Benedetto et al. of 20 subjects with PD who underwent 20 h of collective speech therapy, consisting of two 1-h session weekly, combined with 26 h of choral singing, consisting of one 2-h sessions weekly over 13 weeks showed that voice treatment which included choral singing helped improve respiratory function, phonation time, and prosody, but did not examine measures of well-accepted markers of PD speech function, such as vocal loudness. None of the other acoustic speech and voice parameters that they examined improved [20].

In light of the popularity of singing-based groups as an alternative or complementary therapy to improve speech and voice impairment in PD, we sought to examine the potential therapeutic effectiveness of this form of therapy with outcome measures comparable to those examined by the clinical studies of LSVT. We conducted a pilot open-label efficacy trial of a 12 week group singing intervention, primarily focused on singing, administered once weekly for 90 min, on a cohort of subjects with idiopathic PD to determine if singing therapy improves vocal loudness as measured by SPL at 50 cm.

2. Subjects and methods

Subjects were included in the trial if they had idiopathic PD based on UK PD Brain Bank Criteria assessed by a movement disorders neurologist, Hoehn and Yahr stages 1–5, complained of voice/speech impairment, scoring > 8 points on the Voice Handicap Index (VHI), had a Mini Mental Status Examination (MMSE) score > 24 and were able to commit to a 12 week singing intervention as well as all assessment visits. All subjects signed the informed consent for the study, which was approved by the Beth Israel Deaconess Medical Center Institutional Review Board and registered on www.clinicaltrials.gov.

Voice analysis was performed at baseline and at 1 and 12 weeks post-intervention. Acoustic measures captured included SPL at 50 cm distance from the microphone during reading of the “Rainbow Passage” (Connected speech 1) and during a description of the cookie theft picture (Connected speech 2), maximum cued volume (db), maximum phonation time (s), average speaking fundamental frequency (Hz), pitch range, and l/t ratio. Except for sound pressure level, which was measured once during each study visit, all acoustic measures were sampled and measured three times, with the median measure calculated for that test session. Baseline acoustic measures were carried out by a voice/speech/language pathologist (A.W.) on a Computerized Speech Lab (CSL) acoustic analysis system, Model 4510B (RayPentax, Lincoln Park, NJ). Statistical analysis was performed on Stata 11.1 (StataCorp, College Station, TX). Baseline VHI and Voice Related Quality of Life (VRQOL) measures were used to assess subject-rated outcomes of voice impairment. VHI is a 30-item questionnaire, range of scores from 0 to 120, higher values reflecting more handicap [10]. VRQOL is a 10-item questionnaire measured as index of 0–100, higher values reflecting better quality of life [11].

Choral therapy sessions were designed to target similar behaviors/goals to those in evidence-supported treatments for PD including kinesthetic awareness, calibration to loudness and phonatory effort, increased respiratory excursion, increased vocal loudness, increased pitch range, and increased movement of articulators. Sessions generally consisted of 10 min of stretching and gross-motor movement exercises, 10 min of breathing training and structured vocal exercises, and approximately 70 min of singing popular songs. These sessions were carried out by a separate voice/speech/language pathologist and singing instructor experienced in the treatment of patients with PD, who was blinded to baseline measures (J.P.). Subjects were required to attend at least 10 of the 12 choral therapy sessions and were assessed at 1 week and 12 weeks after the intervention. Subjects were not permitted to be engaged in voice therapy or a separate singing group at anytime between recruitment and after their 12 week assessment. Subjects were provided with exercise handouts, audio recordings, and lyric sheets and guided to practice everyday at home.

The study was originally designed to have 80% power to detect a 4 dB difference in the primary outcome measure, requiring 32 subjects total divided between a singing intervention arm and a non-singing intervention arm consisting only of group breathing and stretching exercises. Due to the group nature of the intervention and poorer than expected recruitment, the study was modified to become a single-arm open-label pilot study of preliminary efficacy of the singing intervention. Statistical analysis was performed on STATA 11 (StataCorp, College Station, TX). Kruskal–Wallis tests were used to compare pre- and post-intervention acoustic voice measures.

3. Results

Fifteen subjects enrolled in the study. One subject dropped out of the study after attending 5 classes due to loss of interest, and another subject dropped out prior to any classes. Thirteen subjects, 11 male and 2 female, were included in the analysis (Table 1). Mean age was 66 (SD = 7.9) and mean disease duration was 9 years (SD = 8.5). Mean MMSE and total UPDRS scores were 28.8 (SD = 1.8) and 45.5 (SD = 22.0). Mean Hoehn and Yahr stage 2.2, with 10 out of the 13 participants in H & Y stages 1–3.

Objective voice acoustic analysis measures as a group showed no significant improvement on the primary outcome of SPL at
versus the poor correlation with UPDRS III motor subscale (did not correlate with any of the UPDRS subscales, with notably significant threshold of 4 dB on our primary outcome. Both subjects had baseline SPL at 50 cm of 70 dB or greater. Secondary outcome measures were analyzed, including SPL at 50 cm during a description of the cookie theft picture (Connected speech 2), maximum cued volume, maximum phonation time, fundamental frequency, pitch range — a surrogate marker for inflection, and s/z ratio — an indicator of the ability to emit a voiced sound using the laryngeal apparatus compared to a voiceless sound, the “z” sound versus the “s” sound. There was no significant difference on these measures from baseline to post-intervention.

Baseline VHI and VRQOL scores reflected a wide range of impairments. Mean pre-treatment VHI composite score was 43.7 ± 22.2 immediately after intervention was 47.0 ± 15.4 and 13 weeks after the end of the intervention was 47.9 ± 21.9 (p = 0.84, Fig. 2). VHI emotional, functional, and physical subscales were not significantly different from pre-treatment to immediately after the 12 week intervention (p = 0.69, 0.62, and 0.76, respectively). Mean baseline VRQOL score was 69.2 ± 18.0 compared to 70.6 ± 19.4 immediately after treatment and 68.1 ± 24.0, 13 weeks after the intervention (p = 0.90, Fig. 3). VRQOL social/emotional and physical subscales were also not significantly improved (p = 0.75 and p = 0.95) from pre-treatment to immediately after the 12 week intervention.

We also examined correlation of acoustic measures and voice-related disability measures with UPDRS subscales. Only the UPDRS I subscale showed a significant correlation with any of our voice-related measures. Composite VHI scores significantly correlated with UPDRS I (r = 0.771, p = 0.04, Bonferroni-adjusted p value), while VRQOL total score did not correlate with UPDRS I (r = −0.686, p = 0.20), suggesting validity of the VHI in measurement of PD-related speech impairment. In contrast, SPL measures did not correlate with any of the UPDRS subscales, with notably poor correlation with UPDRS III motor subscale (r = −0.31, p = 1.00). None of the VHI or VRQOL total scores or subscales correlated with the UPDRS motor subscales or total score.

4. Discussion

To our knowledge this is the largest study of the impact of a choral singing intervention on objective measures of vocal loudness, the primary outcome measure significantly improved by LSVTLOUD. The results of the study suggest that 12 weeks of once weekly 90 min singing sessions with a trained voice/speech therapist is not helpful in improving loudness during connected speech. A previous pilot study of 20 patients with PD used collective speech therapy, combined with choral singing measured outcomes on various respiratory and voice variables but not vocal loudness.

In our study, we analyzed a number of objective acoustic measures, including those measures which, in aggregate, are thought to most influence speech intelligibility. Maximum phonation time, maximum volume, and voicing ability as measured by s/z ratio were unchanged. Measures that approximate quantification of prosody, including fundamental frequency range, also were not improved. Finally, neither of the subject-rated voice-related quality of life measures improved after the 12 week intervention.

We also performed post hoc analysis examining correlation between objective acoustic measures voice-related disability scores and UPDRS scores to explore the relationship of voice impairment and overall and specifically PD-related motor impairment, as suggested by some recent studies in the field [21,22]. These studies have been undertaken to explore the utility of automated quantitative voice and speech analysis to serve as useful biomarkers of PD disease progression although older studies have shown mixed results when attempting to correlate dopamine-responsive motor impairments and voice parameters affected in parkinsonian speech [4,6,23]. In our study, SPL, VHI and VRQOL scores did not correlate with UPDRS motor subscales or total scales, though VHI scores correlated with UPDRS I (non-motor) scores. Our data suggests that

![Fig. 1. Sound pressure level (SPL) during reading of the Rainbow Passage (Connected speech 1) did not improve among the cohort from baseline to either 1 week or 13 weeks post-treatment (Kruskal–Wallis chi-square = 0.697, p = 0.71).](image1)

![Fig. 2. Voice handicap index (VHI) scores did not decrease with treatment compared to baseline (Kruskal–Wallis chi-square = 0.21, p = 0.90). Higher scores indicate greater handicap.](image2)

<table>
<thead>
<tr>
<th>Table 1: Subject characteristics.</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Mean</strong></td>
</tr>
<tr>
<td>Sex</td>
</tr>
<tr>
<td>Age</td>
</tr>
<tr>
<td>Years since dx</td>
</tr>
<tr>
<td>MMSE</td>
</tr>
<tr>
<td>Beck depression</td>
</tr>
<tr>
<td>UPDRS I</td>
</tr>
<tr>
<td>UPDRS II</td>
</tr>
<tr>
<td>UPDRS III</td>
</tr>
<tr>
<td>UPDRS IV</td>
</tr>
<tr>
<td>Total UPDRS</td>
</tr>
<tr>
<td>Hoehn and Yahr</td>
</tr>
</tbody>
</table>
voice impairment as captured by sound pressure level and patient-perceived disability is often independent of motor impairment as measured by the UPDRS motor subscale, which may support the finding that other factors besides dopamine-responsive cardinal symptoms are the main contributors to voice and speech impairment, though thorough analysis via automated algorithms of other specific biomarkers of speech was not undertaken in this study.

There are several potential explanations for why this study intervention failed to show significant improvements across the entire cohort. Our study sample reflected a wide range of speech and voice impairment as shown by both objective and subjective measures. Two individuals who did demonstrate a clinically meaningful improvement of greater than 4 dB in SPL at 50 cm had higher baseline vocal loudness at baseline (greater than 70 dB), suggesting that subjects who are more mildly impaired might benefit from this type of intervention. However, there were insufficient numbers of subjects to allow a definitive subgroup analysis to address this finding. It is also possible that our own data was subject to variability that was not well characterized due to lack of repeated test–retest measures.

Additionally, we consider that the intensity and frequency of the singing intervention may not have been sufficient to achieve a meaningful benefit. Our intervention provided 16 therapy hours spread over 12 weeks and did not quantify the amount of home practice. LSVT<sup>LOUD</sup> by contrast consists of 4 h per week over 4 weeks with more specific prescription of exercises to be completed at home. LSVT-X (Extended) consists of 2 h a week over 8 weeks. The longer gaps between sessions for our particular singing intervention may have failed to consistently provide enough reinforcement of gains achieved at each session. LSVT has been proposed to enhance neural plasticity by means of a training program with high complexity, saliency and intensity [24]. Singing is also likely to incorporate complex vocal motor training with dual-task loading and emotional saliency but delivered at the doses given here, lacks the intensity that LSVT<sup>LOUD</sup> provides. Although we did assign exercises to be practiced at home between sessions, we did not require subjects to provide daily records of the amount of practice they performed at home, and noncompliance with these practice sessions also may have reduced the effectiveness of the weekly therapy. We conclude then that intensity is both a major factor towards the effectiveness of speech/voice therapy but may also be a considerable barrier towards patients’ ability to receive this type of therapy face to face with a trained therapist.

Singing-based therapy, individually or in groups, may continue to be utilized by many people with PD as an alternative form of voice and speech therapy, perhaps in part due to limited access to LSVT therapy for some patients. Despite our negative findings, several participants provided feedback that they enjoyed social aspects of the treatment sessions and found the experience subjectively beneficial. Proponents of LSVT concede that “the frequency of treatment can become an obstacle to providing LSVT due to mobility problems or scheduling conflicts [25].” These access problems have motivated the development of extended LSVT treatment, which requires less frequent visits over an extended period of time, and LSVT treatment by proxy such as by videophone or computer [15,16,26]. Future studies of singing-based strategies in PD-related voice and speech impairment might explore the effectiveness of different intensities and frequencies of therapy.

Acknowledgments

We acknowledge the Michael J. Fox Foundation for funding this study and the National Parkinson Foundation for their support of the Center of Excellence at BIDMC. We also thank the RJG Foundation for their support of our wellness research programs. Dr. Shih was supported by Clinical Investigator Training Program, Harvard/MIT Health Sciences and Technology- Beth Israel Deaconess Medical Center, in collaboration with Pfizer and Merck & Co. We also thank Gottfried Schlaug and Nancy Mazonson for their assistance with obtaining funding and subjects for the study.

References


