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Prosodic Improvement in Persons with Parkinson Disease Receiving SPEAK OUT!® Voice Therapy

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Keywords
Parkinson disease · Dysarthria · Speech therapy · Outcomes

Abstract
Background/Aims: Hypokinetic dysarthria in Parkinson disease (PD) hinders the ability to verbally communicate and interferes with activities of daily living. SPEAK OUT!® is a therapy program designed to improve functional communicative ability. In contrast to the Lee Silverman Voice Treatment program, SPEAK OUT!® promotes speaking with intent to effect loud speech. This study evaluated the efficacy of SPEAK OUT!® in persons with idiopathic PD in 3 domains: self-reported voice handicap, clinical ratings of dysarthria and prosody, and acoustic analysis of prosody. ***Participants and Methods:*** Pre-/post-therapy data included PD participants' scores on the Voice Handicap Index (VHI) and the Voice-Related Quality of Life (V-RQOL) questionnaire, audio recordings, perceptual evaluation scores, and demographic data, such as age, sex, handedness, diagnosis, and onset of PD. ***Results:*** Participants achieved a statistically and clinically significant improvement in speech intensity, pitch range, normalized pairwise variability index for pitch, sustained vowel duration, reading intelligibility, and vocal quality after SPEAK OUT!® training, consistent with both of the self-report voice scores, i.e., the VHI and the V-RQOL, and with the perceptual speech evaluation scores. Longer PD duration was associated with lowered efficacy. ***Conclusions:*** SPEAK OUT!® is effective and should be administered as early as possible after disease onset.

Introduction

Parkinson disease (PD) is a progressive neurodegenerative disorder affecting 1 in 100 adults over the age of 60 years. 1.5 million people in the United States currently

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have PD, and about 60,000 Americans are newly diagnosed each year [1, 2]. Fifty percent more men are affected than women [3]. It is estimated that by 2040 twice as many people will be diagnosed with PD [4].

A majority of persons with PD manifest hypokinetic dysarthria, which is characterized by reduced loudness, breathy voice, mono-pitch, voice tremor, intermittent rapid rushes of speech, and imprecise production of consonants [5, 6]. These speech deficits hinder their ability to verbally communicate and interfere with activities of daily living. Persons with PD may report difficulty being understood on the phone, at a drive-through restaurant, or at a social event where there is background noise [7–9]. Some people have essentially resigned themselves to the fact that their loved ones must speak for them or order for them. Living with PD and its associated speech impairment, in particular, has psychosocial and emotional impact. Patients often report a tendency to withdraw from friends and family, feeling isolated and depressed [7–9].

As for treatment, patients may benefit from medical treatment, including administration of pharmaceuticals, such as levodopa (L-dopa) or dopamine agonists, or deep brain stimulation. While pharmacological and surgical treatments have remarkable effects on limb motor control, it is fair to say that the effects on speech have often been less impressive [10–12]. As a result, behavioral treatment is still the mainstay for the management of hypokinetic dysarthria [13]. Within this approach, the Lee Silverman Voice Treatment (LSVT) has become the treatment of choice. Introduced by Ramig et al. [14] in 1987, the LSVT program has been evaluated in numerous published investigations. LSVT is an intensive, high-effort treatment with focus on increasing phonatory effort, vocal fold adduction, and sensory perception of effort [15, 16]. Treatment is delivered 4 times a week over 4 weeks. Treatment tasks within each session include drills of sustained vowels as well as phonation over one's maximal frequency range. Patients are then taught to generalize their techniques to functional phrases, reading, and conversational speech. Throughout treatment, patients are instructed to think "loud" and complete tasks with a "loud, good quality" voice [14].

The efficacy of LSVT has been widely investigated in studies that have included randomized control trials [14, 16]. Results of these studies show efficacy on objective outcome measures, including vocal intensity measured in sound pressure level (SPL) and fundamental frequency variation, as well as perceptual measures pertaining to speech quality and loudness. A recent review of efficacy studies of LSVT concluded that there is general support in the literature that LSVT improves aspects of speech and voice in the long-term (12 and 24 months) outcomes [14, 17]. Even so, some caution is warranted as the aforementioned efficacy studies are not without limitations. This is because outcome data in these studies may not well generalize to natural functional communication as they are frequently derived from speech samples, such as short utterances, and maximum performance tasks, including maximum sustained vowel phonation. In addition, these tasks do not lend themselves to evaluate the possibly deleterious effect LSVT may have on prosody. This is because the focus of LSVT is on loudness, which is but one acoustic dimension of prosody besides pitch and duration. Since prosody entails controlled variability in loudness, pitch, and duration, the emphasis on loud speech could well diminish variability as in mono-loudness rather than enhance it and possibly reduce variability in pitch and duration as well.

In more recent years, diminished vocal effort in persons with PD has also been treated with other voice therapy approaches whose clinical goals relate to a similar focus on increased motor amplitude. One such regimen, called SPEAK OUT!®, has begun to see more application in clinical practice. SPEAK OUT!® is a voice therapy program designed to improve functional communicative ability. In contrast to the LSVT program, this program promotes speaking with intent as a vehicle to effect loud speech. Cues such as "speak with authority," "use your CEO voice," and "say it with gusto" are associated with the concept of "intent" and utilized during treatment sessions [18]. Confirmation of speaking intent includes an increase in vocal loudness combined with variation of intonation, which approximates more natural speech prosody during connected speech utterances [18]. The SPEAK OUT!® program runs over 12 sessions, 3 sessions per week for 4 weeks. Each session is usually completed in 45 min and addresses 6 component activities. They include vocal warm-ups, vowel prolongation exercises, intonation/gliding exercises, verbal delivery of automatic sequences (e.g., counting numbers, days, and months), oral reading of sentences or passages, and cognitive tasks (e.g., talking about one's happiest childhood memory or most recent vacation; listing 3 things to consider before buying a car or going on a trip; describing 5–10 strategies that help manage PD; or providing 5–10 typical items on a grocery list). Homework assignments include a twice a day speech exercise regimen for 25 consecutive days starting on the patient's first day of individual therapy. A sound level meter is provided to each patient in the first

2–3 weeks so they can measure their speech intensity during practice. After this period, patients are encouraged to use dB apps on smart devices, such as smart phones, iPads, or tablets. Additionally, each patient receives a personal workbook with step-by-step instructions for daily therapy and home exercises. It can be seen that SPEAK OUT!® and LSVT, while similar, differ in 2 respects. One is the focus on speaking with intent so as to achieve loud speech. Speaking with intent is defined and modeled as a purposeful cognitive focus on increasing vocal loudness and intonation variability during speech [19]. The rationale for this is rooted in clinical observations by Boone [pers. commun.] where he noticed that if individuals with PD speak with intent they often speak louder and clearer. Boone attributes this effect to a greater reliance on the pyramidal system by his patients speaking with intent and a lesser dependence on (i.e., bypassing to a degree) their faulty extrapyramidal system where dopamine depletion causes loss of some automatic movements.

The efficacy of SPEAK OUT!® has only been evaluated in 2 rather small investigations by the same group [20, 21] and 1 larger-scale study [18]. Levitt [20] obtained efficacy data in 6 male participants with PD when they were off medication at baseline and after SPEAK OUT!® administration (at 4 weeks). In addition, he conducted follow-up evaluations when participants were enrolled in a maintenance program at 8 and 12 weeks, respectively. Dependent measures in this study included vocal intensity during sustained phonation (6 s) of the corner vowels /a/, /i/, and /u/ and frequency of F1, F2, and F3. In addition, perceptual measurement of vocal improvement as measured by the Voice-Related Quality of Life (V-RQOL) questionnaire was obtained. Results revealed a significant increase in intensity during sustained phonation ranging from 13 to 17 dB_{SPL}. Gains were maintained at 8 and 12 weeks, except in 1 participant whose average intensity dropped 4 and 6 dB during these time intervals, respectively. Descriptive analysis of the vowel formants showed changes suggestive of vowel expansion with the greatest change shown at 4 weeks after therapy. The patients' perception of vocal functioning changed significantly (from fair at baseline to good at 4 weeks) for physiological dimensions on the V-RQOL but remained statistically similar on the social-emotional dimensions of this scale. Levitt et al. [21] conducted a follow-up study with 12 participants (4 females and 8 males), which in addition to sustained vowel prolongation of the corner vowels also included gliding from the lowest to the highest pitch level and vowels extracted from 3 sentences. Results revealed a 14- to 20-dB_{SPL} increase across tasks. Sex was shown not to influence the results. Pre- versus post-V-RQOL differences were not reported in this study. Watts [18] conducted a retrospective study of 78 patients (52 males and 26 females) who completed at least 12 SPEAK OUT!® treatment sessions before post-treatment measures were collected. The primary outcome variable was vocal intensity (dB_{SPL}) measured in 3 different speaking conditions: sustained vowel, reading, and conversation. Results showed that mean intensity increased by approximately 17 dB, 9 dB_{SPL}, and 6 dB_{SPL} for sustained vowels, reading, and conversation, respectively. Overall, it appears that SPEAK OUT!®, not unlike LSVT, is an effective treatment for reduced vocal intensity in individuals with PD. That said, few, if any, data exist that bespeak the influence of SPEAK OUT!® on prosody.

Prosody is increasingly being considered an important dimension in the differential diagnosis of motor speech disorders. Liss et al. [22] used several rhythm indices, including standard deviation of mean vocalic (ΔV) and intervocalic duration (ΔC) in an utterance, the coefficient of variation for these intervals (VarcoV, VarcoC), the overall percentage of vocal intervals (%V), and the difference in duration between successive pairs of vowel and consonant intervals as in the pairwise variability indices (e.g., normalized pairwise variability index [nPVI] for vowel and raw PVI for consonant), and found that different combinations of them can reasonably well distinguish the dysarthrias from each other and from normally healthy speakers. Even though this level of differentiating potential was not replicated in a smaller study by Levitt [20], it seems that the objective evaluation of prosody remains an important component of treatment efficacy, particularly if the evaluation is comprehensive and includes the quantification of the dimensions of intensity and pitch in addition to duration. Evaluation of prosody is further supported in light of recent evidence that shows that therapeutic improvement in prosody in persons with PD is associated with increased intelligibility [23].

Purpose

The aims of this study were to evaluate the efficacy of the SPEAK OUT!® program in persons with idiopathic PD by examining pre- versus post-differences in 3 domains: participants' self-reported voice handicap, clinical ratings of dysarthria and prosody, and acoustic analysis of prosody.

Table 1. Demographic and clinical information of participants with PD (n = 16)

Sex	
Male	11
Female	5
Mean age, years	71.6±6.7
Mean duration of PD, years	7.9±8.0
VHI score	
Pre-test	57.9±28.3
Post-test	36.2±28.9
V-RQOL score	
Pre-test	24.8±10.9
Post-test	17.9±7.2
Hypokinetic dysarthria severity rating, pre-test/post-test	
None	0/1
Mild	7/12
Moderate	6/2
Marked	2/0
Severe	1/1

Values are means ± standard deviations or *n*. PD, Parkinson disease; VHI, Voice Handicap Index; V-RQOL, Voice-Related Quality of Life.

Table 2. Overview of outcome measures

Self-report	VHI score V-RQOL score
Clinician assessment	intelligibility speech rate intonation intensity dysarthria severity voice quality
Acoustic measures	intensity pitch range nPVI for pitch nPVI for intensity nPVI for duration

VHI, Voice Handicap Index; V-RQOL, Voice-Related Quality of Life; nPVI, normalized pairwise variability index.

Methods

Participants

All participants gave their consent before participating in the study which was approved by the Institutional Review Board at the University of Oklahoma Health Sciences Center. Data were collected on 16 participants with PD (11 males and 5 females; mean age 71.6 ± 6.7 years; mean duration of PD 7.9 ± 8.0 years) (Table 1).

Procedure

Participants were seen pre- (1–3 weeks before therapy) and post-therapy (1–3 weeks after therapy) at INTEGRIS Jim Thorpe Rehabilitation Center. Demographic and clinical variables included age, sex, ethnicity, handedness, diagnosis, and onset of PD.

At each visit, participants were asked to read the "My Grandfather Passage" (GP) and to produce conversational speech elicited with prompts, such as "What are your plans for this weekend?" or "What did you have for breakfast?," along with follow-up questions or requests for details (e.g., "Tell me more about ..."). A target sentence from the GP ("We often urged him to walk more and smoke less, but he always answers, 'Banana oil!'") was selected for acoustic analysis.

Measurements

Participants were asked to complete the Voice Handicap Index (VHI) and the V-RQOL questionnaire [24, 25] before and after treatment. To supplement self-reported outcomes, perceptual evaluation was conducted by eliciting independent ratings of participants' paragraph reading from a clinically certified speech-language pathologist (fourth author) and a research assistant with extensive background in motor speech disorders (second author); ratings used a 5-point rating scale ranging from –2 to 2 (see Appendix 1 for scoring guidelines). Finally, dysarthria severity (based on the GP reading) was rated using a 5-point scale (0: none, 1: mild, 2: moderate, 3: marked, 4: severe). Raters were blinded as to pre-treatment versus post-treatment status, and audio samples were reviewed in a random order generated by the third author. Listening was conducted in a soundproof room. Inter-rater reliability was calculated using a percent agreement for 2 raters (Table 2).

For all audio elicitations, recordings were made using a Zoom H1 Handy digital recorder with a TUBE-preamp and a headset microphone (AKG C430) placed 8 cm away from the participant's mouth. Settings were held constant for all participants and recording sessions. Pre- versus post-therapy differences in acoustic prosody parameters, including amplitude, pitch, and nPVI for duration, intensity, and pitch, were analyzed for the target sentence in the GP. All acoustic measurements were conducted in Praat version 5.1.07 [26].

Analysis

Pre- versus post-therapy differences in acoustic measures were assessed using paired t tests, whereas those involving rating scales were statistically analyzed using Wilcoxon signed-rank tests. All statistical tests were performed in SAS® version 9.4 and/or R version 3.3.1.

Results

Self-Reported Results

Post-therapy VHI score (36.2 ± 28.9; p = 0.0009) and V-RQOL score (17.9 ± 7.2; p = 0.02) were significantly

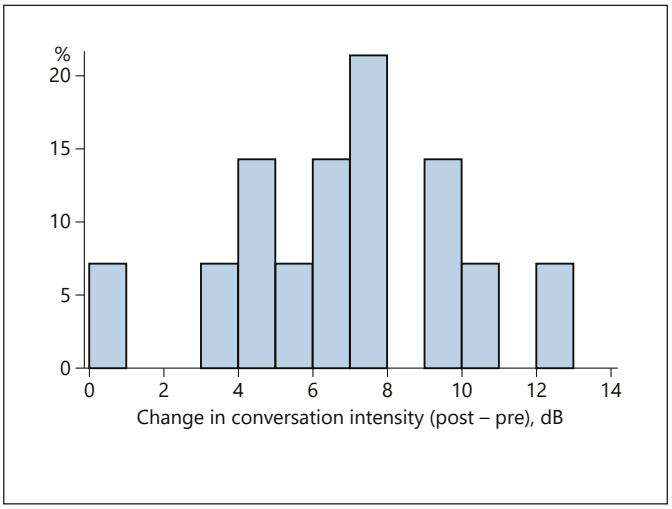

Fig. 1. Change in conversational intensity (post-therapy – pre-therapy) in dB_{SPL}. SPL, sound pressure level.

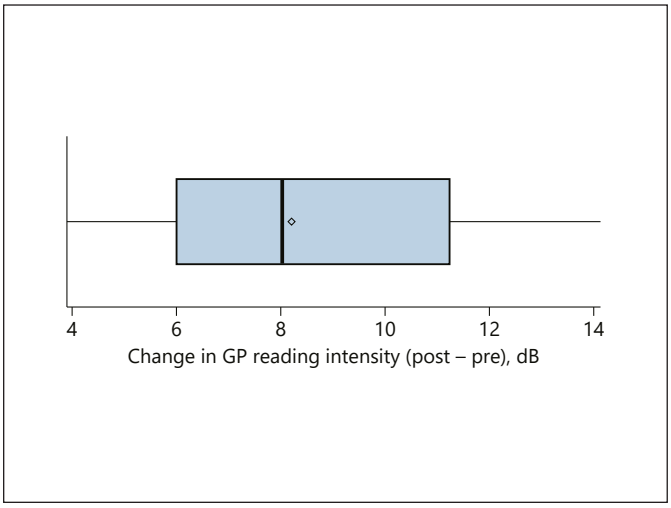

Fig. 2. Change in GP reading intensity (post-therapy – pre-therapy) in dB_{SPL}. GP, "My Grandfather Passage"; SPL, sound pressure level.

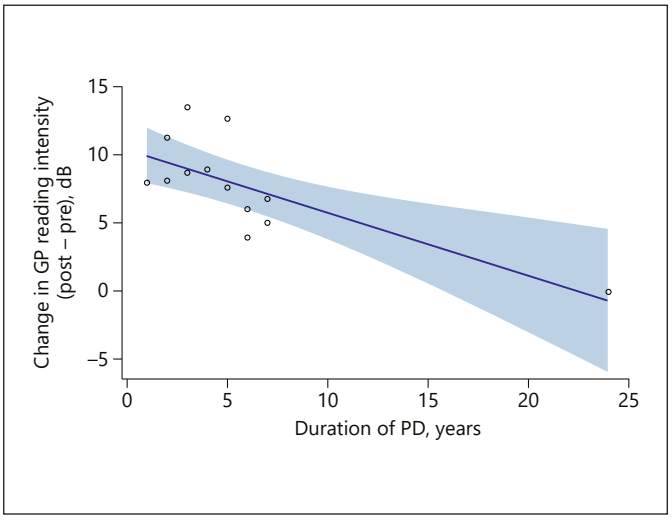

Fig. 3. Intensity improvement (dB_{SPL}) by duration of PD. PD, Parkinson disease; SPL, sound pressure level.

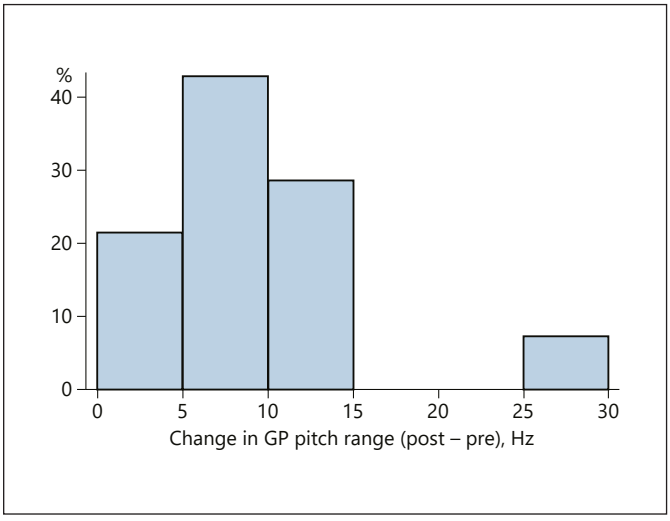

Fig. 4. Change in GP reading pitch range (post-therapy – pre-therapy) in Hz. GP, "My Grandfather Passage."

lower than pre-treatment scores (pre-VHI mean 57.9 ± 28.3; pre-V-RQOL mean 24.8 ± 10.9) (Table 1). Participants scored an average of 21 points lower (95% CI 11–32) on post-therapy VHI and scored an average of 6 points lower (95% CI 1–11) on post-therapy V-RQOL. In other words, VHI changed from "severe" to "mild" after therapy, while V-RQOL remained "fair to good."

Clinician Assessments

Analysis of perceptual ratings by the 2 raters revealed that post-treatment intensity was significantly improved compared to pre-treatment intensity in the GP reading ($p = 0.0005$; inter-rater reliability 75%).

Post-treatment reading intonation in the GP was judged as significantly less monotone ($p = 0.0008$; inter-rater reliability 70%), while reading intelligibility was sig-

nificantly increased (p = 0.0070; inter-rater reliability 81%). Presence of hoarseness (whispery voice) was significantly reduced compared to pre-treatment in the GP reading (p = 0.046; inter-rater reliability 88%). Finally, post-treatment overall dysarthria severity (p = 0.010; inter-rater reliability 94%) was significantly lower with a mean decrease of 0.53 points.

Acoustic Measures

Post-treatment intensity (dB_{SPL}) was significantly greater compared to pre-treatment intensity in the conversational ($p < 0.0001$; Fig. 1) and GP reading ($p < 0.0001$; Fig. 2) tasks. Specifically, participants gained an average of 7.0 ± 3.1 dB_{SPL} in conversational speech and 8.2 ± 4.0 dB_{SPL} in the GP reading. A greater duration of time since diagnosis of PD was associated with decreased improvement in intensity in both conversational speech (p = 0.016) and GP reading (p = 0.004; Fig. 3).

Post-therapy pitch range in the GP reading (mean 28.6 Hz, 95% CI 23.0–34.2) was significantly greater than before treatment (mean 18.1 Hz, 95% CI 14.6–21.6; $p < 0.0001$). Pitch range improvements in the GP reading did not vary by sex (p = 0.9399) and age (p = 0.089). Participants' pitch range in the GP reading expanded by an average of 10.0 Hz (95% CI 6.6–13.5; Fig. 4).

Post-treatment nPVI for pitch was significantly increased (p = 0.028), with a mean increase of 3.91 Hz (95% CI 4.51–9.72), but nPVI for intensity (p = 0.136) and nPVI for duration (p = 0.538) did not change significantly.

Post-therapy speech rate during GP reading was significantly slower (post-therapy 4.3 ± 0.4 syllables/s, pre-therapy 4.6 ± 0.8 syllables/s; p = 0.041), having been reduced by an average of 0.31 (0.01–0.60) syllables/s.

Discussion

Participants achieved a statistically and clinically significant improvement in speech intensity, pitch range, nPVI for pitch, sustained vowel duration, reading intelligibility, and vocal quality after SPEAK OUT!® training, consistent with both of the self-reported voice scores, i.e., the VHI and the V-RQOL, and with perceptual speech evaluation scores. The mean 8.2 dB_{SPL} increase in passage reading corresponds to an approximate doubling in perception of loudness [27]. This increase is similar to that reported by Watts [18]. As the participants read the GP louder, they also used a wider pitch range, indicating improvement in prosody. That said, while speech rate was slower statistically, it was only marginally slower in a clinical sense. The speech rate difference was about half of that observed by Martens et al. [23] who directly targeted slowing of speech rate (along with intonation) in what they labeled "SPRINT" therapy. Even so, the slower speech rate may well have contributed to increased articulatory precision and intelligibility while minimally interfering with naturalness. It is a limitation of the present study that these dimensions were not assessed. Future efficacy studies of treatments that target prosody either indirectly (SPEAK OUT!®) or directly in one or more of its dimensions (LSVT, SPRINT) could well be served if they evaluated the dimensions of prosody (pitch, intensity, and duration) comprehensively as was recently done by Boutsen et al. [28]. In this study, the acoustic multidimensional prosody index was used to determine whether persons with different types of dysarthria rely more or less on any of these prosodic dimensions in their implementation of prosody.

Longer PD duration was associated with lowered pre-/post-loudness difference in the efficacy outcome measurements. While not quantified statistically by Martens et al. [23], inspection of the raw data suggests that they are somewhat at variance with our observation and that more research is needed. Even so, our results comport with the general clinical working principle that speech deficits in PD should be treated as early as possible after disease onset. In addition, they also support the notion that while PD is progressive and degenerative in nature, speech impairments associated with the disease respond favorably to rehabilitation. Overall, it can be concluded that the SPEAK OUT!® program is a viable treatment for individuals with PD that improves not only their speech intensity but most likely also other prosodic features.

Disclosure Statement

The authors declare that there are no conflicts of interest.

Appendix 1

Dysarthria Evaluation Form

The "My Grandfather" passage

Rate the speaker's performance on the "My Grandfather" passage:

	-2	-1	0	1	2
Intelligibility: (circle one)	Very unintelligible	Less intelligible than average	Normal	More intelligible than average	Very intelligible
Speech Rate: (circle one)	Too slow	Slower than average	Normal	Faster than average	Too fast
Intonation: (circle one)	Flat or monotone	Less variable than average	Normal	More variable than average	Too variable
Intensity: (circle one)	Very quiet	Quieter than average	Normal	Louder than average	Very loud

Dysarthria severity (circle one)	None	Mild	Moderate	Marked	Severe
Voice quality: (circle as many as needed)	Breathy	Hoarse	Normal	Harsh	Strained or strangled

Audio sample number: _____ Evaluator: _____

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