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RESEARCH ARTICLE

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Dysarthria treatment for Parkinson's disease: one-year follow-up of SPEAK OUT!® with the LOUD Crowd®

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ABSTRACT

Introduction: SPEAK OUT! with The LOUD Crowd is a standardized speech therapy program typically consisting of 12 one-on-one treatments and ongoing weekly group maintenance sessions for patients with dysarthria due to Parkinson's disease (PD). It is based upon the hypothesis that increased attention to speech, which is a goal-directed motor activity, may compensate for the impairment in automatic sequential motor behaviors often demonstrated in patients with PD. We present results on the 1-year response to treatment.
Methods: Forty individuals with idiopathic PD received SPEAK OUT! delivered in 12 one-on-one 40-min treatment sessions 3 times per week for four consecutive weeks in addition to ongoing group maintenance sessions called The LOUD Crowd. Evaluations occurred 3 times at baseline, within one and six weeks after completion of the SPEAK OUT! sessions ($N = 40$) and 1-year later ($N = 35$). Assessments included mean speech intensity and intonation from reading and monolog, the voice quality acoustic measure called cepstral peak prominence (CPP), and scores on the voice-related quality of life questionnaire.
Results: The significant improvements achieved in all outcome measures from baseline to completion of SPEAK OUT! were maintained 1-year later. Participation throughout the year in regular group maintenance sessions (The LOUD Crowd) was positively correlated with level of improvement at 1 year for all measures except patient perception of voice.
Conclusions: These long-term data contribute evidence of the effectiveness of this speech therapy program for improving communication for individuals with PD and emphasize the importance of regular and ongoing group sessions to sustain therapeutic gains.

Introduction

Dysarthria occurs in as many as 90% of people with Parkinson's disease (PD), advancing in severity as the disease progresses, causing significant problems in being understood by others [1,2]. Dysarthria in PD is characterized by reduced loudness, abnormal rhythm, reduced pitch range, prosodic loss resulting in monotonous and non-emotional speech, inappropriate pauses, and consonant and vowel imprecision [3,4]. These alterations in speech negatively affect patients' self-perception as communicators even at early stages of the disease [5]. Such changes can lead to psychosocial isolation and significantly reduced quality of life [6], suggesting that early intervention is important. While pharmacological approaches have not rendered good results in improved speech in PD [7], mounting evidence supports the efficacy of speech therapy [8–11].

We present data on the long-term (1 year) follow up of SPEAK OUT! with The LOUD Crowd, an 8-h standardized speech-rehabilitation program that combines one-on-one treatment (SPEAK OUT!) with ongoing weekly group maintenance sessions (The LOUD Crowd). This program is based upon motor learning theory, with the focus on "speaking with intent," training patients to be thoughtful and purposeful during speech production. This "intentional" focus is based upon the hypothesis that speech, a goal-directed motor activity, may benefit from increased attention and mindfulness to compensate for the impairment in automatic sequential motor behaviors characteristic of PD [12]. Major features of SPEAK OUT! with The LOUD Crowd are (1) it is brief intervention, consisting of 12 sessions of 40 min each; (2) the group maintenance component, the LOUD Crowd, is integrated into the SPEAK OUT! treatments and does not require separate training or certification of the speech-language pathologist; and (3) warm-up and practice exercises are provided in a courtesy workbook for each patient [11].

Our earlier study reported significant positive outcomes immediately after 12 SPEAK OUT! sessions [11]. A retrospective study reported positive outcomes immediately after intervention and maintained at 1 year [13] but the retrospective design did not allow for assessment of the effect of The LOUD Crowd maintenance component. Therefore,

long-term prospective outcomes data are necessary to evaluate the entire program's efficacy. The primary aim of this study was to evaluate acoustic data and patient self-perception of voice 1 year after completion of individual therapy sessions (SPEAK OUT!). Specifically, we asked the question: "To what extent are the gains achieved immediately after completion of SPEAK OUT! maintained one year later?" The secondary aim was to determine the effect of participation in The LOUD Crowd group maintenance program.

Methods

Patients

Men and women who had been diagnosed with PD and who had come for a speech therapy evaluation at the Parkinson Voice Project in Richardson, Texas, USA and were invited and assessed for eligibility to participate in the study. Eligibility criteria included: a diagnosis of idiopathic PD and referral for speech therapy, no history of deep brain stimulation, no speech therapy within the prior two years, no other neurological diseases or medical conditions affecting speech production, and sufficient cognitive abilities and English language skills to fully participate in therapy.

A total of 166 patients were screened for participation, and 47 subjects were enrolled into the study. A sample size of 35 participants was calculated to be enough to identify an increase in speech intensity of at least 9 dB [13] with an alpha of 0.05 and 80% power [14]. Taking into account a potential loss to follow-up of 10% of subjects, a sample of 40 subjects was deemed necessary to detect a statistically significant and clinically meaningful finding in this study.

Intervention outcomes were obtained on 40 subjects at the six-week follow-up and on 35 subjects at the 1-year follow-up. The details of the enrollment and follow-ups are presented in Figure 1, and the demographic data for the subjects who were assessed at the six-week and 1-year follow-ups are reported in Table 1. For a full description of the original sample recruitment and procedures please see Behrman et al. [11]

Approval for this research was obtained by the Western Institutional Review Board for participation of the patients at the Parkinson Voice Project and by the City University of New York Institutional Review Board for analysis of de-identified patient data by the second author and her research team. All patients in this study participated in the informed consent process.

Speech therapy: SPEAK OUT! with the LOUD Crowd

SPEAK OUT! is a standardized speech rehabilitation intervention of 12 one-on-one 40-min sessions (3 times per week for four consecutive weeks), with twice-daily home speech exercise practice (once per day on therapy days). It is paired with The LOUD Crowd, a group-based speech rehabilitation maintenance program. Participants were encouraged to attend at least one session of The LOUD Crowd during the third or fourth week of the SPEAK OUT! sessions as a standard of care, followed by a minimum of once per week group attendance after finishing the
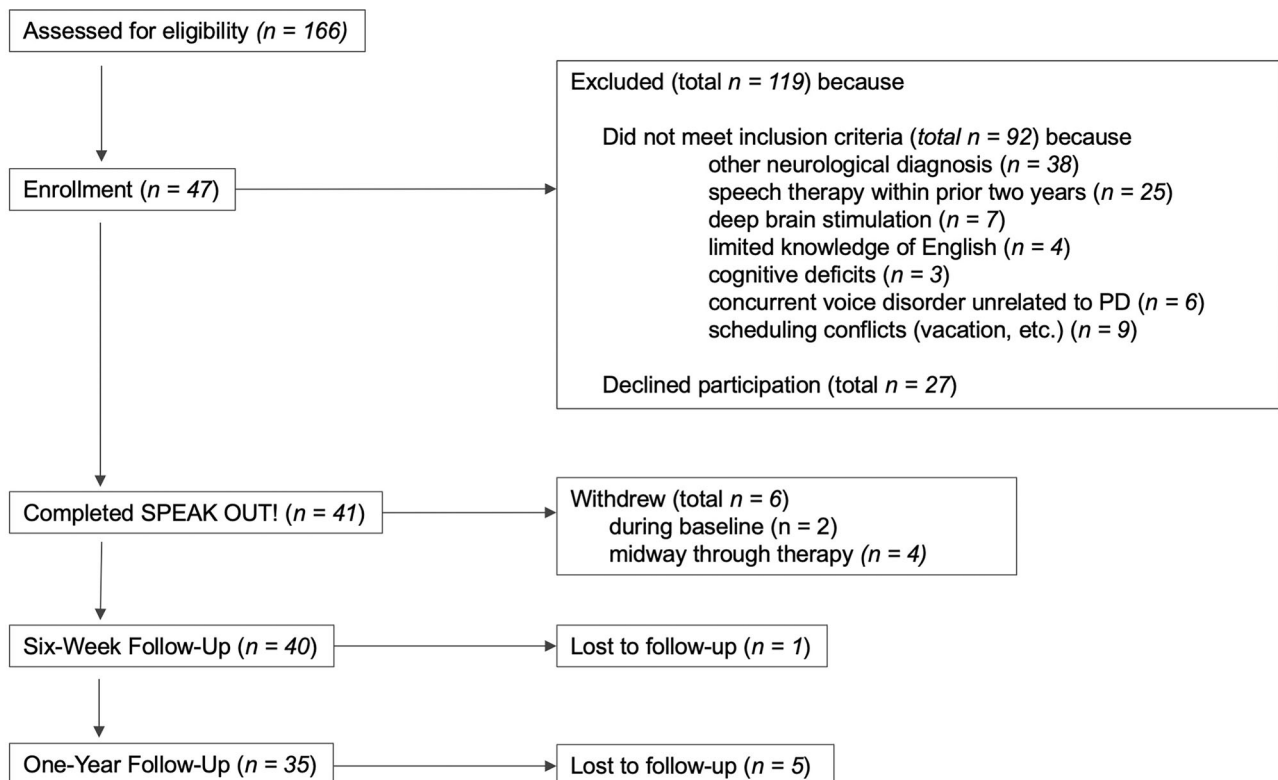

Figure 1. The enrollment, treatment, and assessment subject numbers are shown in this flowchart.

Table 1. Participant demographics for the 40 patients (25 men and 15 women) who completed the 12 weeks of individual therapy sessions (SPEAK OUT!) and the 34 patients (21 men and 14 women) who were assessed at 1-year after therapy.

	Age (in years)		Time elapsed (in months) between diagnosis of PD and first baseline measures		Score on Hoehn and Yahr Scale [8]	
	Men	Women	Men	Women	Men	Women
Participants who completed SPEAK OUT!						
Mean	69.6	66.7	48	58.8	2.4	2.2
Range	47 − 84	46–82	1–237	3–189	1–3	1–3
Median	70	68.2	38	40.6	2	2
SD	7.8	8.7	53.5	55.0	0.6	0.8
Participants assessed at 1 year after SPEAK OUT!						
Mean	65.8	65.9	45	56.2	2.2	2.6
Range	47–84	46–82	1–237	3–189	2–3	2–4
Median	69	65.5	35	39.2	2	2
SD	8.0	9.8	52.1	53.0	0.7	0.9

Table 2. SPEAK OUT! with The LOUD Crowd program of speech therapy for Parkinson's disease (PD).

Parameter	SPEAK OUT! with The LOUD Crowd
Dosage	SPEAK OUT! Individual treatment sessions of 40 min, 3 d per week for 4 weeks for a total of 12 sessions (8 h total) plus home exercise program (explained further below). The LOUD Crowd Weekly group sessions of 40 min ongoing indefinitely.
Focus	Patients are instructed to speak with intent for all utterances within the session. Increase cognitive focus upon goal-directed, intentional behavior and minimize automatic motor behavior.
Verbal Cues	Speak with intent. Be deliberate. Speak out. Say it like you mean it. Say it purposefully. Speak with your intentional voice, not your automatic voice.
Therapy Protocol	Parkinson's information session: Prior to initiating individual treatment (SPEAK OUT!), patient and family attend a group session to learn about basic neurophysiology of PD, intentional and automatic motor behaviors, home practice expectations, and need for ongoing group maintenance, The LOUD Crowd. Six SPEAK OUT! components: 1. Warm-Up: produce connected vocalizations using nasal phoneme-initial words; 2. Vowel: sustain/ɑ/ with good quality voice for maximum of 10 s; 3. Glides: sustain/ɑ/ and glide up the scale starting and ending with modal pitch; 4. Numerical Sequences: count aloud pausing after every three to five numbers; 5. Reading: start with phrases and progress to paragraphs; 6. Cognitive-linguistic exercises: Structured activities designed to elicit novel responses. The task was designed to improve word retrieval and cognitive processing speed, while focusing upon intentional speech. Conversational speech was interwoven throughout the session to facilitate transfer to communication in daily life. The LOUD Crowd: Patient attends one to two LOUD Crowd sessions during week three or four of SPEAK OUT! (depending upon scheduling) to become acquainted with the group sessions, followed by weekly sessions upon completion of SPEAK OUT! The same six components and conversational speech are performed in a group format. Upon completion of SPEAK OUT! the patient continues daily home practice and regularly attends a weekly group on an ongoing basis for ongoing accountability, practice, support, and encouragement.
Therapy materials	SPEAK OUT! Workbook is provided to each patient in the U.S. by Parkinson Voice Project; SPEAK OUT! Workbook and Therapy Flash Cards are provided to each SLP who completes training.
Home exercise program	15-minute home practice session using SPEAK OUT! Workbook: Frequency: During individual therapy (SPEAK OUT!): once a day on treatment days; twice a day on non-treatment days. After completion of SPEAK OUT!: ongoing once daily.
Shaping techniques	SLP uses modeling as well as verbal and visual cues to elicit intentional speech. SLP prompts patient self-monitoring and self-generated internal cueing for increased intentional speech. The patient was asked to consciously and purposefully use intentional speech consistently. When intentional speech was not used, the SLP immediately cued the patient to resume use of intentional speech. In this way, the distinction between automatic and non-automatic was emphasized, and patient self-monitoring and self-generated internal cueing were trained.
Clinical data collection	Objective: • Intensity per sound level meter for 10 responses within each therapy component • Duration (up to 10 s) for sustained/ɑ/ Subjective: • Level of cueing. • SLP perception of vocal quality and use of intentional speech.
Adjuvant program (not assessed in this protocol)	SPEAK OUT! Refreshers: Patient returns for a reevaluation every six months; additional treatment sessions are scheduled, as needed, with the goal of returning dB SLP levels for reading and conversation to original discharge status.
Discharge criteria	SPEAK OUT!: Goal attainment (per clinical data collection above): typically within 10–12 sessions (for this study, all participants received 12 sessions). The LOUD Crowd: No discharge. Participation is recommended on a regular basis to maintain therapeutic gains to the extent possible.

individual therapy, along with continued daily practice of speech exercises. The full program is described in Table 2.

Assessment procedures and instrumentation

All assessments and research data collection were done at the Parkinson Voice Project. Two speech-language pathologists conducted the SPEAK OUT! therapy sessions for four weeks. Baseline assessments were conducted before intervention, at one- and six-week post-intervention, and again at the 1-year follow-up. Subjects were scheduled for their assessments during the "ON state" of their dopaminergic medications for all encounters as a standardized procedure. Notably, most research shows little effect of such medications on speech [15], consistent with the hypothesis that PD-related dysarthria may result from factors other than dopamine depletion [16].

Speech tasks included a one-minute monologue and reading of a shortened version of "The Caterpillar" [17] for each assessment. The monolog was elicited with the instructions to "Tell me about a recent vacation, or the place where you grew up, or what you did for a living before retirement." Digital audio recordings (Zoom H4N audio recorder, 44.1 kHz sampling rate) with a head-mounted AKG (C520) cardioid condenser microphone at 30 cm from the mouth, were obtained in a low ambient noise level room. Cueing was not used during the recordings. The research team (led by the first author) who analyzed the anonymized data had no direct patient contact.

Outcome measures

Acoustics outcome measures included mean speech intensity, intonation (standard deviation of fundamental frequency), and voice quality measured by cepstral peak prominence (CPP), an acoustic index that has shown strong correlation with the listener's judgment of dysphonia severity in people with dysarthria related to PD [18]. A score on the 10-item voice-related quality of life [19] questionnaire identified the subjects' perception of their voice and the impact on quality of life during the recent past.

Intensity, calculated for each speaking task, was calibrated with a 1 kHz tone of known intensity. Calculation of intonation was obtained using Praat acoustic analysis software [20]. Both intensity and intonation were calculated from 45 s of the monologue and the entire reading passage. CPP was calculated with Praat using the phrase "We were away a year ago," an all-voiced sentence recommended for this measure [21].

The acoustic data from 10% of the subjects were randomly drawn for reliability re-measurement using intra-class correlation (ICC) analyses. ICC model (1, 1), a one-way random model, was used for intra-rater reliability and ICC (2, 1) a two-way random model, was used for inter-rater reliability. ICC analyses indicated strong inter-rater (ICC = 0.92, $p < .001$) and intra-rater reliability (ICC = 0.97, $p < .001$) for intensity and strong inter-rater (ICC = 0.92, $p < .001$) and intra-rater reliability (ICC = 0.93, $p < .001$) for fundamental frequency.

Statistical analysis

Normal distribution for all parameters was determined using a Kolmogorov–Smirnov test. To assess the primary aim of this study – the outcome 1 year after completion of SPEAK OUT! – paired t-tests were used to compare the value of each outcome variable between the 6-week and 1-year follow-ups. Paired t-tests were also used to compare values between baseline and the 1-year follow-up to determine the extent of any increase or decrease in values from baseline in case, the 1-year data changed significantly from the 6-week follow-up. To assess the secondary aim – the effect of The LOUD Crowd maintenance sessions – the change score between the 6-week and 1-year follow-ups were regressed on the number of maintenance sessions using ordinary least squares regression. The Wilcoxon test was used to analyze the voice-related quality of life scores. To adjust for multiple post hoc pairwise comparisons, the Bonferroni correction factor was applied when interpreting these results with $\alpha = 0.0042$.

Results

Comparison of speech at 6 weeks post-treatment with the 1-year follow-up

The change in levels (Δ) from 6-week post-SPEAK OUT! to the follow-up at 1-year post-treatment demonstrated statistically significant improvement for intensity in reading ($\Delta = 0.8$, SD = 2.7) [t(3.88), $p = .0005$] and in monologue ($\Delta = 0.85$, SD = 1.8) [t(3.7), $p = .0008$], intonation in reading ($\Delta = 0.28$, SD = 0.9) [t(2.89), $p = .007$] and in monologue ($\Delta = 0.78$, SD = 1.6)[t(3.53), $p = .0013$], and the voice quality measure of CPP ($\Delta = 2.1$, SD = 0.6)[t(3.58), $p = .001$.]. That is, the change in mean scores on all acoustic measures from 6 weeks to 1 year was statistically significant although numerically small. Statistically significant improvement was not observed for the change in scores of the voice-related quality of life ($\Delta = 0.2$, SD =6.4) [t(1.09), $p = .28$], although on average, the scores did not deteriorate from 6 weeks to 1 year.

Comparison of speech at baseline with the 1-year follow-up

The change in levels (Δ) from baseline to the follow-up at 1-year post-treatment demonstrated statistically significant improvement for intensity both in reading ($\Delta = 8.94$, SD = 4.9) [t(10.72), $p < .0001$] and in monologue ($\Delta = 7.25$, SD = 6.7) [t(10.72), $p < .0001$]; and in intonation both in reading ($\Delta = 8.74$, SD = 5.9) [t(8.65), $p < .0001$] and in monologue ($\Delta = 9.48$, SD = 7.2) [t(7.7), $p < .0001$]. Statistically significant improvement was also demonstrated in CPP (the acoustic measure that correlates with voice quality) ($\Delta = 6.7$, SD = 5.6) [t(6.9), $p < .0001$] and also in scores of the voice-related quality of life ($\Delta = 10.22$, SD = 15.3) [t(3.90), $p = .0004$]. Graphical results of the distribution of scores on all outcome variables at baseline, 6 weeks after completion of SPEAK OUT! and at the 1-year follow-up are presented in Figure 2.
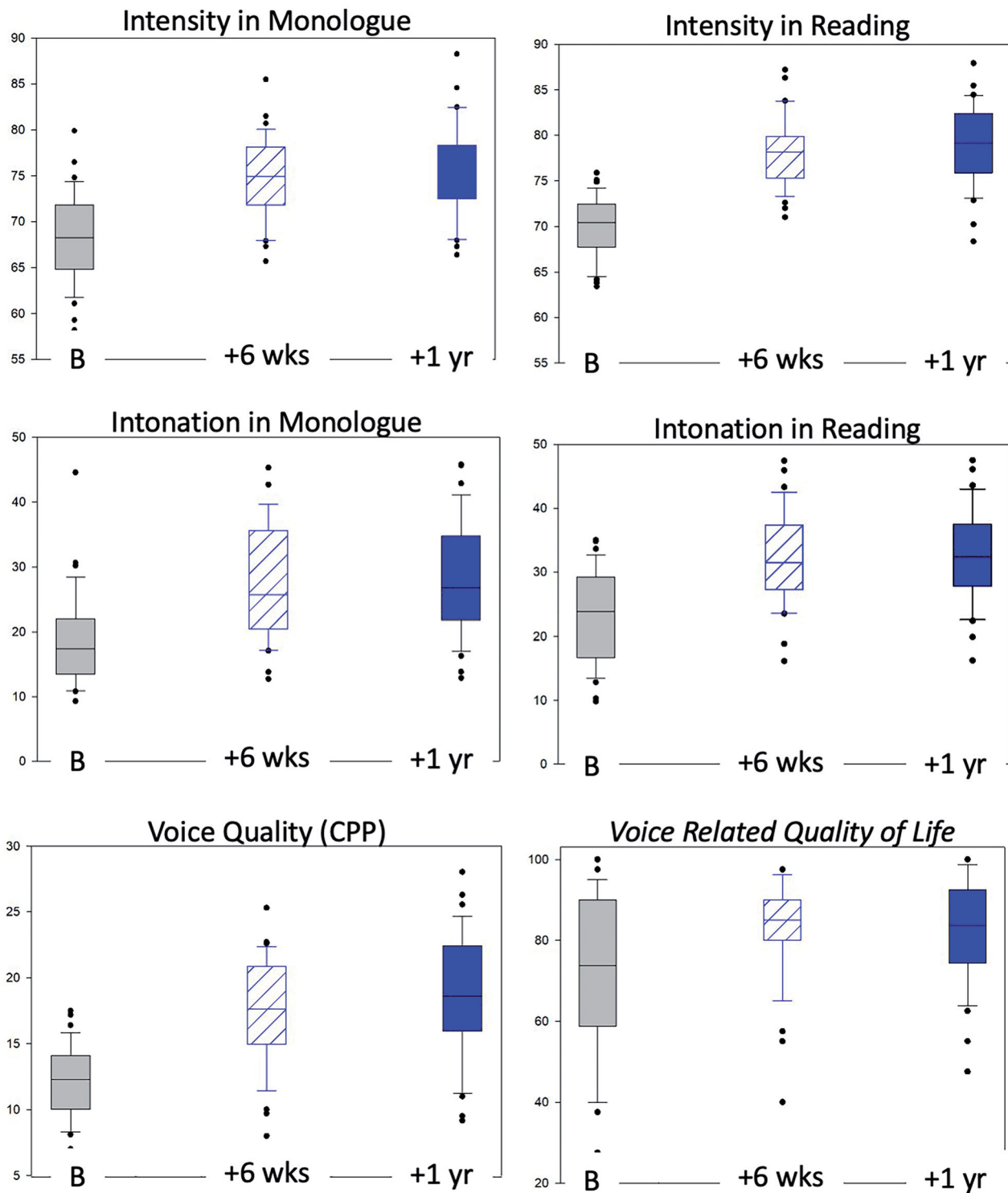
Figure 2. Distribution of scores for each of the outcome variables. B = baseline, +6 weeks = assessment after completion of the 12 individual therapy sessions of SPEAK OUT!®, +1 year = 1-year assessment after baseline assessment. Note that, in a box-and whisker plot, the boundary of the box closest to 0 indicates the 25th percentile and the boundary of the box farthest from 0 indicates the 75th percentile. Whiskers above and below the box indicate the 10th and 90th percentiles. The horizontal line with the box marks the median.

Effect of participation in the LOUD crowd group maintenance sessions at the 1-year follow-up

On average, patients attended 21 sessions of The LOUD Crowd (range 0–41, SD 13). Regression analysis was conducted to estimate the effect of participation in The LOUD Crowd on the outcome variables at the 1-year follow-up. Participation in these group maintenance sessions was significant for levels of intensity in reading $[F(1, 32) = 15.06, p < .0005]$ and in monologue $[F(1, 32) = 13.66, p \leq .008]$, levels of intonation in reading $[F(1, 32) = 8.32, p = .0007]$ and
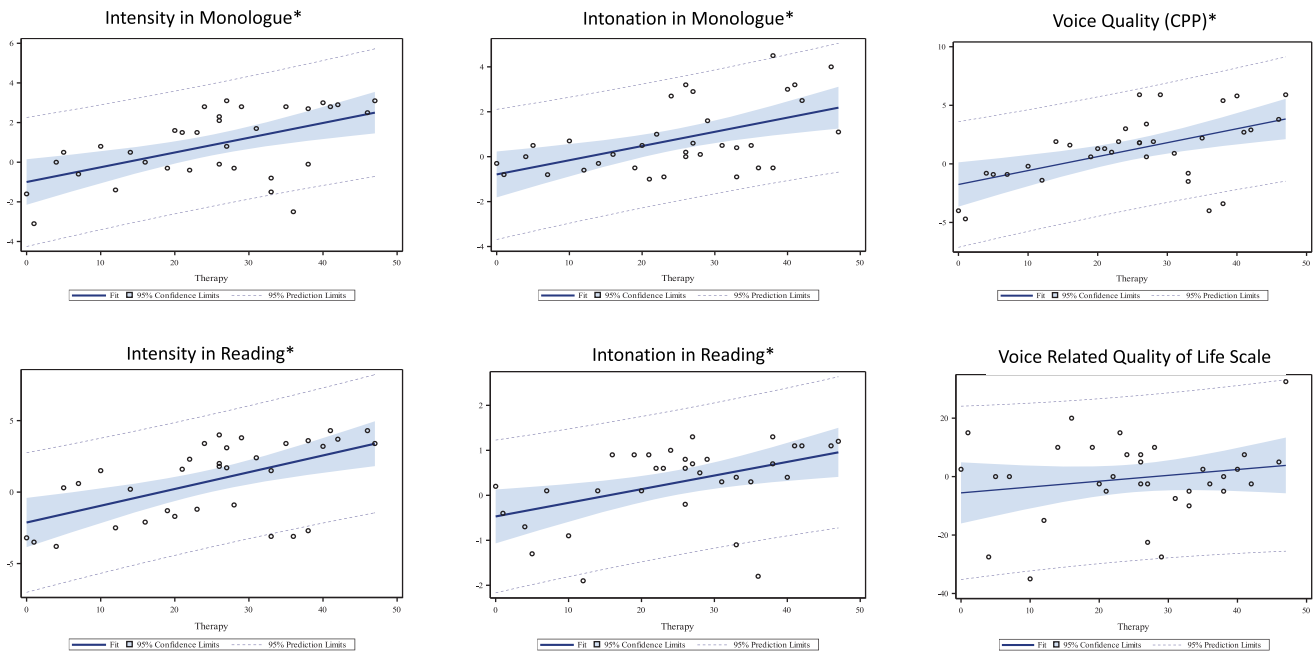
Figure 3. Regression curves with confidence intervals of the relationship between number of The LOUD Crowd® group maintenance sessions attended between the follow-up immediately upon completion of the initial 12 sessions of SPEAK OUT!® therapy and the 1-year follow-up for all outcome variables. Asterisks denote statistically significant relationships.

in monologue [$F(1,\ 32) = 12.43$, $p = .001$], and CPP – the acoustic measure of voice quality [$F(1,\ 32) = 12.84$, $p = .001$]. Participation did not significantly affect scores on the voice-related quality of life [$F(1,\ 32) = 1.18$, $p = .28$]. The regression curves with confidence intervals for all the outcome variables are presented in Figure 3.

Discussion

The data obtained in this project describe the long-term 1-year results of a standardized dysarthria treatment program for patients with PD consisting of a total of 12 40-min sessions of one-on-one treatments across 4 weeks (SPEAK OUT!) together with ongoing weekly group maintenance sessions (The LOUD Crowd). The major finding was that the statistically and clinically significant improvements in speech intensity, intonation, CPP, and the voice-related quality of life scale that were achieved upon completion of SPEAK OUT! [11] was maintained 1 year later. Furthermore, continued participation in The LOUD Crowd group maintenance sessions was a significant factor in the preservation of acoustic gains made from the individual therapy sessions.

It is hypothesized that speaking with intent resulted in greater muscular activity of both the respiratory and laryngeal systems, thereby increasing effort of the speech production system overall, improving intensity. The overflow of increased muscular activity from a specific speech behavior to the entire speech production system has been reported previously in other types of dysarthria [22]. The intensity improvement in reading (∼9 dB) and monologue (∼7 dB) and its maintenance at the 1-year follow-up are clinically

significant since it has been described that a 10-dB increase in speech intensity is perceived twice as loud by the listener [23].

The gains in intensity reported in this study were greater than those reported at 1-year post-therapy in Watts' [13] retrospective study of SPEAK OUT! However, in that study, less than 40% of the original patients were included at 1 year, and control for participation in The LOUD Crowd was not reported.

The increase in intonation obtained with the use of intentional speech and maintained at the 1-year follow-up were clinically significant. When averaging these gains across speaking tasks by gender, increases of approximately 6.5 semitones (equal to notes on the Western musical scale) in men, and approximately 4 semitones in women were achieved. These gains would be perceived easily by listeners.

It is posited that the gains achieved in intonation 1-year post-therapy were related to overcoming the habituation of decreased range of motion that often characterizes movement accuracy in PD. Speaking with intent emphasizes increased effort and attention to movement of the oral articulators. Use of intentional speech may have spread to the phonatory system and increased the dynamics of laryngeal muscle activity, resulting in increased intonation [24].

Improved intonation may also have been related to the patients' outlooks about their speech production. Emotional outlook [25] and overall communicative abilities impact intonation in individuals with dysarthria [26]. It is possible that the improved speaking abilities that resulted from participation in the rehabilitation program generated improved emotional state, as reflected in the higher scores of the voice-related quality of life post-therapy. These rationales

for improved intonation are untested and additional research is necessary to support or refute these hypotheses.

The post-therapy CPP values obtained in this study at the 1-year follow-up are typical of healthy speakers of a similar age group [27], highlighting the long-term efficacy of the intervention. It is hypothesized that the improvement could be due to enhanced vocal fold closure associated with the augmented speech intensity [28]; however, further studies incorporating videolaryngeal stroboscopy as an outcome measure would be necessary to corroborate this point.

The approximately 10-point improvement in scores of the voice-related quality of life scale that was achieved immediately upon completion of SPEAK OUT! were maintained at 1 year (Figure 2), but the scores were not influenced by the frequency of participation in The LOUD Crowd (Figure 3). As reported previously, despite the improvement, scores remained below those of healthy age-matched controls immediately post-therapy [11]. Although speaking with intent improved the acoustic characteristics of the patients' voices as well as the patient's perception of voice quality and associated quality of life, the underlying etiology of the dysarthria for these patients remains unchanged. Therefore, we hypothesize that the patient's perception of vocal abilities may be negatively influenced by the awareness of persistent dysarthria; however, the impact of other psychosocial variables cannot be ruled out.

It is notable but not surprising that participation in The LOUD Crowd group maintenance sessions enabled patients, on average, to maintain the acoustic gains that had been achieved immediately upon completion of the individual SPEAK OUT! therapy sessions. We believe that, in the case of PD, speech therapy provides patients with an adaptive, compensatory strategy that must be continually practiced and consciously employed given the deficits in both sensory awareness and automated motor behaviors, and the progressive nature of the disease.

The necessity for ongoing practice is further supported by a study of patients diagnosed with PD and treated with another program, Lee Silverman Voice Treatment [8]. In that study, large gains were made in average intensity in monologue one month after completion of the therapy, but at seven months after therapy, approximately half of those gains were lost. In other research on a different modality, it has been suggested that continued physical exercise is necessary for maintaining physical benefits for individuals living with PD [29]. Of course, comparisons across different studies, treatments, and modalities must be considered with caution. Nevertheless, our findings contribute evidence to support the necessity of ongoing structured practice for patients living with PD to help them maintain initial therapeutic gains in speech production.

Examination of individual data points from each of the outcome variables in this study revealed that not every patient achieved similar success in maintaining or improving speech at the 1-year assessment. The box plots of Figure 2 reflect the spread of data points. However, no pattern or specific variables could be discerned between 6-weeks and 1-year post SPEAK OUT! that correlated with improvement beyond participation in The LOUD Crowd. Deeper understanding of the factors that help each patient to find success with therapy programs should be further explored, perhaps through single-subject experimental designs.

A limitation of this study is that the possible mechanisms underlying the improvement with the use of intentional speech are unknown, specifically, the degree to which speaking with intent results in increased activity of the goal-directed basal ganglia circuits or how intentional speech may override or compensate for the impairment in automatic sequential motor behaviors. Further research into these areas is needed.

It would be worthwhile to explore outcomes of SPEAK OUT! and The LOUD Crowd in patients who speak languages other than English. Although a large multilingual study showed no language-based differences in clinical parkinsonian phenotypes [30], it is unknown whether the effect of speaking with intent is language dependent, particularly regarding intonation, given prosody-related differences among languages.

In conclusion, this study contributes evidence for speech-language pathologists, neurologists, and patients' families that therapeutic gains in speech therapy for hypokinetic dysarthria secondary to idiopathic PD can be achieved with ongoing practice and regular group attendance, despite the progressive nature of the disease. Furthermore, this study confirms the long-term effectiveness of the SPEAK OUT! and The LOUD Crowd program.

Disclosure statement

Alison Behrman is an unpaid member of the Scientific Advisory Board of Parkinson Voice Project. Jennifer Cody was a paid employee of Parkinson Voice Project during the conduct of this study. Samantha Elandary is the CEO of Parkinson Voice Project and a member of its Board of Directors. Shilpa Chitnis is an unpaid member of the Scientific Advisory Board of Parkinson Voice Project.

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