The effectiveness of the Lee Silverman Voice Treatment (LSVT) for improving speech and voice production at 12 and 24 months post-treatment in patients with Parkinson's disease: A Critical Review of the literature

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This critical review examines the long term (12 and 24 month) effectiveness of LSVT on improving speech and voice production in patients with Parkinson's disease. Study designs reviewed included two randomized control trial studies, two single subject multiple baseline studies, and one single group pre-posttest study. Results provided suggestive evidence supporting the long term effectiveness of LSVT in improving some aspects of speech and voice in the clinical setting; however, evidence supporting the generalization of treatment effects to functional communication in natural environments is inconclusive.

Introduction

Nearly 100,000 Canadians (Parkinson Society Canada, 2007) and 1.5 million Americans live with Parkinson's disease (PD) (Ramig, Sapir, Countryman, Pawlas, O'Brien, Hoehn & Thompson, 2001). As the North American population ages, these numbers will increase. Speech and voice disorders associated with the disease exist in over 75% of effected individuals (Ramig, Bonitati, Lemke & Horii, 1994) and are characterized by reduced voice volume (hypophonia), poor voice quality (dysphonia), reduced pitch inflection (hypoprosody), reduced range of articulatory movements (hypokinetic articulation), and short rushes of speech (Sapir, Ramig & Fox, 2008). These disorders of speech and voice, collectively termed hypokinetic dysarthria, can result in the reduced ability to communicate (Sapir, Ramig & Fox, 2008). This is considered to be one of the most difficult aspects of PD, with deleterious effects on an individual's psychosocial well-being and quality of life (Fox, Morrison, Ramig & Sapir, 2001).

Lee Silverman Voice Treatment (LSVT) is one therapy program currently identified in the literature as having generated short and long term efficacy data on its ability to improve voice production as well as impact the overall communication function of individuals with PD. Introduced by Ramig and colleagues in 1987 (Ramig et al., 2001), LSVT is an intensive, high effort treatment with focus on increasing phonatory effort and vocal fold adduction as well as on improving sensory perception of effort (Ramig, Horii & Bonitati, 1991). Treatment is delivered 4 times a week over 4 weeks. Therapy tasks completed in all sessions include drills of maximum duration of a sustained vowel and maximum fundamental frequency range. These tasks are thought to increase a patient's phonatory effort such that voice is produced with maximum efficiency. 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 (Ramig et al., 1991).

Published reviews of earlier studies (Fox, Morrison, Ramig & Sapir, 2001; Yorkston, Hakel, Beukelman & Fager, 2007), generally support the immediate effectiveness of LSVT in improving aspects of speech and voice that have been relatively untreatable by traditional speech therapy and are inconsistently managed by pharmacological and surgical treatments (Dromey, Kumar, Lang & Lozano, 2000). Following recommendations of these reviews, recent research has examined the long-term (12 and 24 months) outcomes of LSVT. It is essential to critically evaluate the methodology and statistical analyses of this research so that clients may be provided with the most current and accurate information when deciding upon suitable treatment options.

Objectives

The primary objective of this review is to provide a summary and critical evaluation of existing literature on the long-term (12 and 24 months) effectiveness of LSVT for improving speech and voice production in individuals with Parkinson's disease. A secondary objective is to provide evidence based practice recommendations regarding LSVT as a speech and voice therapy option for this population. Opportunities for future research will also be discussed.

Methods

Search Strategy

Computerized databases, including PubMed, CINAHL, Scopus and the University libraries search engine were searched using the following search strategy: (Parkinson's disease) AND ((LSVT) OR (Lee Silverman Voice

Treatment) OR (Voice therapy)). The search was limited to articles written in English. There was no limitation on the date of articles. An examination of the reference section of articles returned yielded further studies for inclusion.

Selection Criteria

Studies selected for inclusion in this critical review paper were required to investigate the long term, (12 and 24 months), effects of LSVT in improving speech and voice production in patients with Parkinson's disease. No limits were set on the etiology of Parkinson's disease, time since diagnosis, stage, or severity of the disease in research participants.

Data Collection

Results of the literature yielded the following types of articles congruent with the aforementioned selection criteria: Randomized control trial studies (3), single subject ABA designed studies (2) and a within-group pre-posttest study (1).

Results

Randomized control trial studies

In a randomized control trial study, Ramig, Countryman, O'Brien, Hoehn and Thompson (1996) evaluated the short and long term (6 and 12 months) effects of LSVT compared with a placebo (respiration) treatment on the speech and voice deficits common to PD. The participants, thirty-five patients with idiopathic Parkinson's disease (IPD) were randomly assigned to one of the two groups after being stratified on nine variables.

Both forms of treatment were administered in 16 sessions within a 1 month time period. Specifics of the LSVT program, including therapy techniques and goals, were consistent with those outlined above. Participants in the respiratory group also received intensive, high effort therapy; however the focus was on improving respiratory muscle activity for inspiration and expiration. Speech and non-speech tasks, clinician feedback and daily homework were present in both groups. Two clinicians delivered both forms of treatment and were randomly assigned to participants.

Statistical analysis (MANOVA) was completed for acoustic measures of vocal intensity, measured in sound pressure level (SPL), fundamental frequency variation, measured in semitone standard deviation (STSD), fundamental frequency (F0), and forced vital capacity (FVC) from before to after treatment, before to 6 months after treatment and before to 12 months after treatment. Data was collected during sustained vowel phonation, reading of the phonetically balanced "Rainbow Passage" and in 25-30 seconds of conversation. Also obtained and analyzed at 12 months post-treatment were scores on the motor section of the Unified Parkinson's Disease Rating Scale (UPDRS), scores of cognition, self-ratings of depression, and self perception of sickness impact on communication and social interaction.

Results indicated that at 12 months post-treatment, only the LSVT group demonstrated a statistically significant increase on measures of vocal intensity, (during sustained vowel phonation [$p < 0.0001$] and reading [$p = 0.009$] only) and STSD (in conversation [$p = 0.020$] only). No statistically significant time-by-treatment group interactions were observed for measures of F0 or FVC. No statistically significant differences were found in either group at 12 months post-treatment for measures of severity, cognition, depression or sickness impact on communication and social interaction. The authors concluded that these findings support the long term effectiveness of LSVT for improving vocal intensity in patients with PD.

In a study by Sapir, Ramig, Hoyt, Countryman, O'Brien and Hoehn (2002), thirty-five participants diagnosed with IPD were randomly allocated to an LSVT group or a high effort respiratory treatment (RET) group after stratification on 5 variables. The authors hypothesized that LSVT would be more effective in improving loudness and overall voice quality in participants at 12 months post-treatment as judged perceptually by six listeners (three certified speech-language pathologists and three non-expert listeners). Tape recordings of each participant reading the "Rainbow Passage" were made pre-treatment and at 12 months post-treatment. These recordings were presented as a pair to each listener with the order of the samples randomized. The listener was then asked to determine which sample in the pair was "better quality" and which sample sounded "louder". Researchers used a chi-squared test to analyze the data and the results indicated that statistically significant improvements in loudness [$p < 0.0001$] and quality [$p < 0.0001$] occurred in the LSVT group at 12 months post-treatment but not in the RET group based upon listener's perceptual ratings. These findings support the researchers' hypothesis.

In the only study currently published in literature to investigate the impact of LSVT at 24 months post-treatment on vocal function in patients with PD, Ramig et al. (2001) used a randomized control trial study to compare the effects of the LSVT and RET specifically on measures of SPL and STSD. Thirty patients were randomly assigned to one of the two groups. The groups

were found to be comparable pre-treatment on age, time since diagnosis, speech severity rating, voice severity rating and stage of disease. Data was collected by a primary investigator blind to the form of treatment each participant received. SPL and STSD measures were obtained as participants performed maximum duration of sustained vowel, reading of the "Rainbow Passage" and in 25-30 seconds of monologue. Results were analyzed using repeated measures analysis of variance (ANOVA) and revealed that at the 24 month follow-up, mean SPL was significantly higher for the LSVT group compared to the RET group during maximum sustained vowel phonation [p=0.000] and reading [p=0.0460], but not in conversation. Mean STSD was also significantly higher for the LSVT group compared to the RET group during reading only [p=0.016] when retested at 24 months. When compared to pre-treatment results, the LSVT group significantly improved SPL and STSD scores across all tasks at 24 months while the RET group did not.

The strengths of these studies are found in their design. Randomized control trial studies are considered the gold standard when attempting to draw causal inferences about treatment effects in groups of patients with certain disorders and thus provide the highest level of experimental evidence (Dollaghan, 2007). The random allocation of participants following extensive stratification in the three studies evaluated ensured no systematic differences between intervention groups in factors that may affect outcome. Therefore, it can be concluded with reasonable confidence that all outcomes were the result of the type of treatment received. Furthermore, the statistical analysis applied to the data was appropriate for each study's design and purpose. While the majority of studies examining the long-term efficacy of LSVT focus primarily on obtaining objective, acoustic measures, the perceptual effects assessed in the study by Sapir et al. (2002) adds to the overall evaluation of LSVT.

The methodologies of the aforementioned studies however were not without flaws and their results should be interpreted with caution. In Ramig et al. (1996) the authors failed to specify whether or not therapy providers were aware of the researchers' hypothesis. It is therefore possible that their unconscious expectations could have influenced the results. Furthermore, in this study, as well as in Ramig et al. (2001) data collected in only one task (conversation) was representative of natural speech. It must be noted that in the study by Ramig et al. (1996) it was during conversational speech that participants in the LSVT group did not show a significant increase in vocal intensity in comparison to a placebo group at 12 months post-treatment, or at 24 months as indicated in the Ramig et al. (2001) study. Similarly, in the study by Sapir et al., (2002) all results were based on reading tasks only. As Sapir and colleagues themselves pointed out, it cannot yet be concluded that the long term effects of LSVT are perceptible in more typical, natural speaking situations. The addition of intelligibility as a treatment outcome measure should be considered for future replication of these studies in order to give a more representative evaluation of treatment effects. Overall, these studies demonstrated that at 12 and 24 months post-treatment, LSVT was effective in improving only some acoustical measures of voice and in limited contexts that did not represent natural speaking situations.

Within-group pre-posttest studies

Ramig and colleagues (1994) evaluated the short- and long-term effectiveness of LSVT on improving speech and voice production in patients diagnosed with IPD who were concurrently involved in multidisciplinary rehabilitation. While both acoustic and perceptual measures were obtained immediately following therapy, only the following acoustic measures were collected at 12 months post-treatment from fifteen of the original forty participants: maximum duration of a sustained vowel (seconds), fundamental frequency variation (STSD), mean fundamental frequency (hertz), fundamental frequency range (semitones). Data were collected as the participants performed maximum duration of a sustained vowel and read the "Rainbow Passage". Seven of the fifteen patients re-assessed at the 12 month time period received an additional 3 months of therapy after the initial 1 month-long LSVT program (group 1), while eight patients received no further treatment (group 2). Analysis of variance (ANOVA) revealed a statistically significant improvement from pre- to post- to 6- to 12 months post-treatment on measures of maximum and mean duration of a sustained vowel [$p<0.002$, $p\leq0.001$], and fundamental frequency range [$p\leq0.011$] There were no significant effects for treatment groups indicating that improvement on these variables were maintained whether or not patients had received additional therapy. Results on measures of mean fundamental frequency and STSD were considered separately for males and females. Therefore, statistical analysis of data collected on these measures at 12-months post- treatment could not be performed due to unequal and limited sample sizes.

Limitations of this study lie in its methodology. Participants received LSVT in the context of intensive multidisciplinary rehabilitation. The extent to which these concurrent therapies could have influenced the short- and long-term LSVT efficacy data is unknown. This group of participants constitutes an unrepresentative

sample of the general population of patients with PD, as it would not be typical for most patients to receive multiple intensive therapies and educational sessions. Although the authors explain that many participants were from out of town and returned home immediately following LSVT, a further limitation of this study was the relatively small sample size from which data was obtained at 12 months post-treatment, making statistical analysis of some measures impossible. Data on outcome measures obtained immediately following treatment by the subgroup of participants re-tested at 12 months were not analyzed in comparison to collective results obtained immediately post-treatment from participants who were not retested at 12 months. It is therefore possible that this subgroup differed significantly on outcomes measured immediately following therapy and thus, was unrepresentative of the initial group.

As was problematic with studies reviewed earlier in this paper, outcome measures in the current study were obtained in tasks that were not representative of natural speech. Reading of the "Rainbow Passage" was the only task in this study that required participants to produce connected speech. While the authors did attempt to probe the impact of therapy related changes on functional communication through the use of perceptual rating scales, these measures were obtained only in the short term at not at the 12 month time period. Inclusion of long-term perceptual data would have helped to strengthen their conclusion that LSVT is an effective voice therapy for patients with PD. In conclusion, while this study can be considered to provide a relatively high level of evidence (level 2b) due to its design, its results must be interpreted with caution because of the exclusion of functional, meaningful speaking tasks and outcome measures as well as a small and possibly unrepresentative sample.

Single-subject ABA studies

Countryman and Ramig (1993) evaluated the short- and long-term effects of LSVT on improving the speech and voice characteristics of one individual who had undergone bilateral thalamotomy one and half years prior to the start of treatment. The patient, a 65 year old female in Stage III on the Hoehn and Yahr Scale for Parkinson's disease, was previously a semi-professional opera singer and choral soprano with a Master's degree in voice performance. At the start of LSVT, perceptual characteristics of her speech and voice included: reduced loudness, intermittent hoarseness, abnormally low pitch, vocal fry, monotonicity, mildly impaired articulation, intermittent harsh, raspy voice, intermittent vocal straining, reduced respiratory support, slight slowing of speech and mild vocal tremor. These deficits were believed to be a direct result of lesioning to the right ventral lateral nucleus of the thalamus during her second surgery, a right radio frequency thermothalamectomy. An intelligibility rating of 90% during conversational speech was made by a speech-language pathologist pre-LSVT. Tests of cognition given at this time revealed mild impairment in her ability to learn new, nonverbal information and mild-moderate impairment in sustained attention and concentration. All other cognitive and psychosocial testing yielded results within normal limits. An oral motor exam indicated reduced range of motion in the tongue and a slightly slowed diadochokinetic rate. Data was collected pre-treatment, post-treatment and at 6 and 12 months after treatment on acoustic measurements of maximum duration of sustained vowel phonation, fundamental frequency of sustained vowel phonation, intensity, jitter, shimmer, harmonic-to-noise ratio, coefficient for variation of amplitude and frequency, vocal fold adduction (analyzed using the custom-built software program EGGW), maximum fundamental frequency range, and semitone standard deviation. Also collected at each time point were the patient's self-perceptual ratings of loudness, monotonicity and intelligibility, recorded on a visual analogue scale. Finally, perceptual ratings were made by three speech-language pathologists on a 5 point scale for measures of tremor/unsteadiness, vocal fry, articulatory precision and overall quality.

Wilcoxon ranked-sum tests were performed to compare pre-treatment variables with immediate post-, 6 months post- and 12 month post-treatment variables. Results revealed that statistically significant improvements at the 12 month time period were made on only four of the eleven acoustical measures: maximum duration of sustained vowel phonation [$p < 0.04$], fundamental frequency of sustained vowel phonation [$p < 0.005$], vocal intensity during sustained vowel phonation only [$p < 0.04$], and frequency modulation [$p < 0.043$]. Although not statistically analyzed, observation of the patient's self-perceptual ratings indicated an immediate impression of improvement. This impression continued throughout the 12 month time period but to a lesser extent. Data on the speech-language pathologists' perceptual ratings at the 12 month follow-up were not made available. However, judgments made at this point in time were reported to be consistent with those made immediately post-treatment in which the patient's improvement during sustained phonation and short sentence reading was judged to be greater than in paragraph reading. The authors conclude that while the patient in this study demonstrated significant pre- to post-treatment improvement in voice production, she was unable to carry over treatment techniques into connected, functional speech or into the long-term.

Dromey, Ramig and Johnson (1995) investigated the effects of the LSVT on voice and speech production in one patient with PD immediately following treatment as well as at 6 and 12 month follow-ups. The authors hypothesized that the patient would increase his vocal intensity as a result of treatment and were specifically interested in evaluating changes in articulation and underlying phonatory mechanisms associated with an increase in intensity. The patient, a 49 year old male in Stage II on the Hoehn and Yahr Scale for Parkinson's disease, was selected for study because he was representative of early stage Parkinson's disease and his activities of daily living required adequate oral communication. Prior to the start of the LSVT, the patient reported that his voice was soft and at times raspy, however he believed his speech to be understandable most of the time. Neuropsychological testing revealed mild attentional difficulties at the start of the study which did not progress over the year in which he was followed. The patient had not received previous speech or voice therapy. Data on measures of acoustic, aerodynamic, respiratory and articulatory acoustic variables were collected immediately post-, 6 and 12 months post-treatment while the patient performed the following tasks: tidal volume, forced vital capacity, maximum duration sustained vowel phonation, maximum fundamental frequency range, a series of /pae/ syllables, reading the "Rainbow Passage", reading 70 individual words and a 30-second monologue. Statistical analysis was not applied to the data; rather, visual inspection was conducted to examine trends over time. For the purpose of this review, outcome measures of vocal intensity, mean fundamental frequency, fundamental frequency variation, jitter and shimmer at 12 months post-treatment will be reported. Based upon observation of data, all outcome measures indicated improved values at the 12 month time period in comparison to pre-treatment levels. Furthermore, all data collected at this time were at or near values obtained immediately post-treatment. Measures of mean fundamental frequency and fundamental frequency variation showed greater improvement during reading than in conversation. As well, vocal intensity was significantly greater for sustained vowel duration and in repetition of the /pae/ syllable than in reading and monologue.

Due to the unique histories of patients, as was particularly evident in the study by Countryman and Ramig (1993), generalization of treatment results from single-subject design studies to a population of patients cannot be made, nor can these results be solely used to prove or disprove the long term effectiveness of LSVT in improving speech and voice production in patients with PD. These results can however, be analyzed for trends consistent with larger scale studies. It was interesting to note that outcomes observed in both patients examined in these two studies were similar with the outcomes reported in studies evaluated earlier in this review; that is, the lack of improvement in the long term on tasks that more closely represented natural speech. While the application of inferential statistical techniques to single-subject design studies has been debated, the lack of statistical analysis in Dromey, Ramig and Johnson (1995) limits the use of their results. After visual inspection of their data it was noted that on some measures, only slight improvement was made. Without statistical analysis it cannot be known whether this slight improvement was due to chance or to the effects of therapy. Both Countryman and Ramig (1993) and Dromey, Ramig and Johnson (1995) acknowledge potential performance variability associated with PD due to tremor, fatigue and uncontrollable responses to medication. It was for this reason that data was collected twice pre- and immediately post-treatment. At the 6 and 12 month time points however, data was collected only once; therefore it is with less certainty that these results represented the patient's true abilities. Despite the high level of evidence these two studies are considered to provide due to their experimental design, their results must be interpreted and generalized with extreme caution.

Discussion

While the authors of the studies reviewed in this paper generally conclude that LSVT is effective in improving various aspects of speech and voice production in patients with PD at 12 and 24 months post-treatment, a critical appraisal of the research reveals a set of limitations and methodological flaws that warrant caution when interpreting the stated results. Of high concern was the limited use of speech tasks that represented natural, functional communication for the collection of the data. Improvement on acoustic measures during production of maximum sustained vowel phonation for example, does not allow one to conclude that improvement has occurred in meaningful communication which is of greater significance and value. A further limitation was the lack of appropriate outcome measures to assess change in overall communicative ability and the perceptions of patients and their conversation partners in regards to treatment effectiveness. Another major concern was that the collection of all outcome measures was completed in the clinical setting. This common practice is problematic as it is possible the client is able to "turn on" a louder, better quality voice in response to the testing environment. Therefore, speech used during clinical evaluations may overestimate and be unrepresentative of the patient's natural speaking tendencies in daily, meaningful interactions. Due to the development of

devices such as the voice accumulator and the voice dosimeter for monitoring speech and voice production in natural conversational situations, it should now be relatively easy to collect objective outcome measures outside the clinic environment to evaluate the carry-over of treatment effects. (Adams & Dykstra, 2009). These results would supplement those obtained in the testing environment and should weigh heavily in the determination of treatment efficacy. Finally, it must be pointed out that Dr. Ramig was the author/co-author on all studies evaluating the long-term efficacy of LSVT in patients with PD. Dr. Ramig is the founder of the LSVT program and it is therefore possible that her personal biases regarding the effectiveness of the program influenced the research results.

Conclusion

There is suggestive evidence that the LSVT is effective in improving *some* aspects of speech and voice production in *limited contexts* at 12 and 24 months post-treatment in patients with PD. More research is needed to determine whether improvement generalizes to more functional speaking tasks in ecologically valid situations outside of the clinic room.

Recommendations

Based on the limitations of the current research discussed above, it is recommended that further research be conducted to investigate the long-term efficacy of LSVT and should include the following:

a) Collection of data in speech tasks that represent functional, meaningful communication
b) Outcome measures chosen with particular relevance to patients and their communicative partners
c) Acoustic measures of participants following the LSVT obtained in natural environments

Clinical Implications

Speech and voice symptoms common to PD have been relatively untreatable by traditional speech therapy and are inconsistently managed by pharmacological and surgical intervention (Dromey, Kumar, Lang & Lozano, 2000). While further research is needed to address whether improvements resulting from LSVT are transferred into situations outside the clinic room, current research provides suggestive evidence supporting improvement of some aspects of speech and voice in limited contexts. Therefore, LSVT should continue to be considered, albeit with caution, as a therapy option for improving speech and voice production in patients with PD.

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