Critical Review:
Effectiveness of LSVT LOUD in Children with Dysarthria Secondary to Cerebral Palsy

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Approximately 80% of individuals with cerebral palsy have an accompanying motor speech disorder, such as dysarthria, which greatly affects the clarity, audibility, and intelligibility of their speech (Reed et al., 2017). This critical review examines the current evidence base regarding the effectiveness of Lee Silverman Voice Treatment (LSVT) LOUD in children with dysarthria secondary to cerebral palsy (CP). Studies include single-subject, between group, and mixed-group designs, examining both the therapeutic effects and physiological brain changes found immediately after, and several weeks following treatment. The evidence gathered from this review is both suggestive and promising. Recommendations for future research and clinical practice are provided.

Introduction

Currently, there is little evidence to support a single effective treatment approach for pediatric motor speech disorders, including childhood apraxia of speech and dysarthria. Children with dysarthria secondary to cerebral palsy (CP) are highly underrepresented in intervention research in the field of speech-language pathology (Levy et al., 2012). Therefore, there is a strong need for efficacy data and well-controlled research studies in this area.

Traditional treatment for dysarthria consists of addressing each of the affected subsystems involved in speech production, including respiration, phonation, articulation, and resonance. (Levy et al., 2012). Lee Silverman Voice Treatment (LSVT) LOUD is an evidence-based and frequently used treatment protocol for adults with dysarthria secondary to Parkinson's Disease (PD). In contrast to traditional methods, LSVT LOUD is a single-focus treatment approach which follows established principles of motor learning and neuroplasticity. These principles include intensive treatment involving active repetitive practice and movement-associated sensory feedback (Boliek et al., 2012).

A concern about delivering an intensive motor treatment to children with neuromuscular disorders is their ability to withstand the specified treatment protocol. Prior research suggests that children with CP can tolerate intensive interventions with a reduction in physical fatigue throughout the course of treatment. These studies also found effectiveness for intensive repetitive practice on gross motor function in children with CP, suggesting similar potential effectiveness for intensive practice on the motor speech system (Boliek et al., 2012).

This review includes information regarding therapeutic and physiological treatment effects of LSVT LOUD, as well as the maintenance of gains several months following intensive treatment.

Objectives

The primary objective of this paper is to critically evaluate existing literature regarding the effectiveness of LSVT LOUD in children with dysarthria secondary to CP. The secondary objective is to provide clinical implications and recommendations based on the current evidence.

Methods

Search Strategy

The reviewed articles were found using several computerized databases including Western Libraries, CINAHL, and PubMed.

The search terms used included:

> (LSVT LOUD) OR (Lee Silverman Voice Treatment) AND (cerebral palsy) AND (dysarthria) OR (motor speech disorders) AND (children) OR (child)

The search was limited to articles written in English between 2010 and 2020 for a more recent review. Moreover, research on the use of LSVT LOUD in the pediatric population has only began to surface in the last 10 years.

Selection Criteria

Articles selected for review were required to investigate the effects of LSVT LOUD in children with dysarthria secondary to cerebral palsy only.

Data Collection

Results of the literature search yielded five articles which included the above selection criteria. One study used a single-subject design, two included a between-group design, and two articles used different measures to analyze results from the same mixed non-randomized clinical trial.

Results

Single-Subject Design

Single-subject designs are an appropriate starting point for testing hypotheses interested in examining possible treatment effects. It is also considered appropriate as the population size of children with spastic CP and dysarthria is relatively small. Caution should be taken when interpreting results due to the small sample sizes.

Boliek and Fox (2012) conducted a phase 1 treatment study using a nonconcurrent multiple baseline single-subject design with replication across subjects in order to examine the therapeutic effects of LSVT LOUD in children with spastic CP and dysarthria. Four children between 5 and 7 years of age with a medical diagnosis of spastic CP underwent a full dose of the LSVT LOUD protocol, consisting of 4 one-hour sessions a week for 4 consecutive weeks, for a total of 16 treatment sessions. Outcome measures on parent rating forms, listening, voice, and speech tasks, were taken over at least 4 baseline recordings (BASE), as well as 1-week following treatment (POST), and again 6-weeks following treatment (FUP). Results indicated listener preference for POST over BASE data for all participants, as well as improved parent ratings from BASE to POST. All participants also demonstrated gain in at least one acoustic measure. Maintenance of gains at FUP varied across participants.

Despite large heterogeneity in this population, the authors attempted to control for age, sex, ability to follow directions, vocal fold pathology, medication stability, velo-pharyngeal incompetence, structural disturbances of the speech mechanism, concomitant speech disorders, and maturational changes during the study period. This does limit their participant pool as it reduces the participants to those with CP and dysarthria without additional factors or disabilities which may affect treatment outcomes and that many children with CP and dysarthria may not have. As such, this also helps with generalizability of findings, as many of the varying factors in this participant population have been controlled for. To account for these factors, the researchers included an age- and sex- matched control group of typically developing children, as well as one untreated child with CP. All participants with CP also presented with a similar dysarthria severity level, making generalizability of findings specific to moderate severity, however the authors did not state this. Moreover, Boliek and Fox (2012) outlined their treatment protocols, outcome measures, and analysis procedures clearly and thoroughly, so they are easily replicable for future studies. The researchers' established an appropriate baseline by including a minimum of 4 data points for each participant. The treatment phase of the study was equal to or longer than each of the baseline conditions, which is also considered appropriate. Including multiple baseline conditions strengthens the single-subject design overall, as opposed to a basic or experimental design only. Moreover, the researchers included a control group for comparison, which is not required in phase 1 studies and provides greater statistical power.

The outlined research methods were thorough and adequate, including a participant screening with clearly defined inclusion and exclusion criteria, random assignment of participants to baseline conditions of varying durations, and two recording sessions at both POST and FUP. Data collection procedures were identical for BASE, POST and FUP recording sessions. Additionally, the investigator who delivered treatment did not collect POST or FUP data, which allows for more objective data collection. Sentences for testing were not trained during the treatment phase and were randomized for data collection. However, the sentences were not pre-recorded in order to standardize the task.

Other weaknesses of the study included a small but appropriate sample size, borderline appropriate inter-rater reliability calculations for some of the outcome measures, and intra-rater reliability was also lower than optimal for the preferred listening task. In addition, auditory-perceptual variables were not clearly defined for the SLP listeners, which may indicate highly variable subjective analysis in the task results. In regard to data analysis, the researchers used appropriate statistical tests for single-subject data but did not report effect sizes or confidence intervals (CI), which limits credibility of statistical results.

The level of evidence for this study is suggestive due to reported conflict of interest for the researchers, some questionable reliability findings, and missing effect sizes and CIs in the statistical analysis. However, an appropriate design, outcome measures, and statistical tests were used. Based on the findings of the study, LSVT LOUD may result in some gains for children with CP, particularly in the perceptual qualities of their speech following treatment.

Between-Group Design

Between-group designs are appropriate for comparing outcomes between different treatment approaches or determining possible treatment effects of one approach. However, more participants and data points from each condition are required to achieve adequate statistical power than for single-subject designs.

Levy, Ramig, and Camarata (2012) conducted a phase 1 study using a small group pre-post intervention design to explore the effects of a more "traditional" subsystems intervention approach versus the single-focus LSVT LOUD intervention in three children with dysarthria secondary to CP. Two children received a full dose (4 one-hour sessions per week for 4 consecutive weeks) of LSVT LOUD following the established protocol, while the third child received traditional therapy for 50 minutes twice per week for four weeks. Outcome measures included a functional impact questionnaire for caregivers, a standardized articulation assessment, and listener preference ratings for informal picture naming of contrastive words and spontaneous speech during play. The authors reported that both treatment approaches had positive effects on speech function, with LSVT LOUD exhibiting change in the SPL acoustic measure, and traditional therapy exhibiting change in standardized articulation measures.

Levy and colleagues (2012) described their methods and data collection procedures thoroughly. Some strengths of the methods were that intervention providers did not collect data at POST so as to eliminate potential bias, assessment scoring and listening tasks were blinded to the treatment condition also to reduce bias, and baseline stimuli was presented in a counterbalanced order during the listening task. Randomization to treatment conditions was not employed, but the allocation process was appropriate considering participant life constraints and the more time-consuming nature of LSVT versus traditional intervention.

However, this study presented with several weaknesses and limitations. Not only was the sample size extremely small for a between-group design, but all of the children were females, which limits generalizability of findings to the female population only. Moreover, the authors did not outline or employ any specific inclusion or exclusion criteria during participant selection, resulting in the participants significantly varying in age, severity of dysarthria, and concomitant speech, language and/or cognitive difficulties. All of these factors limit the generalizability of findings as there is a small sample size with extremely large heterogeneity. The researchers also did not establish an appropriate baseline as only two data points were collected. In addition, the baselines for SPL were highly variable so the reported improvements in this area from LSVT intervention should be taken with extreme caution. Treatment conditions also differed on several variables, such as the traditional intervention being delivered by two master's students, whereas the LSVT intervention was delivered by a certified SLP. These two groups differ greatly on clinical expertise and experience and may therefore indicate differences in the quality of treatment delivery. Moreover, the total number of treatment sessions differed greatly, and only the LSVT intervention included daily carryover assignments. These differences make it difficult to determine which specific aspects of the interventions resulted in different or improved therapeutic effects. Additionally, no measurements were taken to determine maintenance of gains over time. Lastly, no statistical or reliability measures were used, as the authors only provided descriptive results. While also unnecessary in phase 1 studies, a lack of external control further limits overall statistical power.

While results are somewhat promising for the traditional approach, the overall level of evidence for this study is equivocal due to the poor methodology, participant selection procedures, and complete lack of statistical analysis. However, the authors provided a rationale for further research in this area with more optimally controlled studies.

Boliek and Fox (2016) conducted a phase 1 treatment validation study using a small between-group pre-post treatment design with a slightly greater age range than in their previous 2012 study. Their aim was to validate previous findings and expand the evidence base for treatment outcomes following LSVT LOUD in children with dysarthria secondary to CP. Seven children with spastic CP and dysarthria between 6 and 10 years of age underwent a full dose of LSVT LOUD and were compared to a matched group of typically developing children who did not receive treatment. Outcome measures included qualitative and quantitative measures of communication and social functioning, and acoustic features taken once at PRE, POST, and 12-weeks following treatment (FUP). Results indicated improved voice quality and articulation at FUP versus PRE, and increased parent ratings at both POST and FUP. Improvements on acoustic measures were found at POST, with variable maintenance at FUP. Single word intelligibility improved at POST but was not maintained at FUP.

Similar to their previous study, Boliek and Fox (2016) clearly described their methods, outcome measures, data collection procedures, and statistical analyses for easy replication. Their sample size was slightly larger than in their previous study and relatively adequate for a between-group design drawing from a

rare population. They included appropriate inclusion and exclusion criteria attempting to control for age, sex, native language, presence of a speech or voice disorder, hearing impairment, vocal fold pathology, ability to follow directions, medical stability, velopharyngeal incompetence, and structural disorders of the speech mechanism. Another strength of the study was that interventionists did not collect data at POST or FUP to maintain objectivity. While fewer expert SLPs served as listeners compared to their 2012 study, the authors included an additional 54 naïve listeners so that 10 listeners evaluated each child's data at all three time points. This change may be an improvement for providing more practical, real-world findings. Inter- and intra-rater reliability calculations were reported and strong for acoustic measurements. Results were analyzed in replication of their 2012 study and appropriate statistical measures were used. However, they still did not report on effect sizes or CIs, thus limiting statistical confidence.

Similar limitations of this study to their previous one included that the data collectors were not blind to treatment status of the participants, no specific definitions were provided to listeners for auditory-perceptual measures, and there was a reported conflict of interest. In contrast to the 2012 study, dysarthria of participants ranged from mild to severe which may make generalizability of findings even more difficult given the small sample size of such a heterogenous population. For the above reasons, the level of evidence of this study is suggestive and does validate some of the previous findings for LSVT LOUD in children with dysarthria and CP.

Mixed Group Design

Mixed designs are often used when the researchers wish to examine both within- and between-group data in their statistical analysis, as was the case for the following studies. Similar to a between-group design, even more participants and data points from each condition are required to achieve adequate statistical power.

Reed, Cummine, Bakhtiari, Fox, and Boliek (2017) conducted a mixed non-randomized clinical trial with 8 children with dysarthria and CP between 7 and 16 years of age in order to examine physiological brain changes as a result of treatment. The researchers included an age- and sex-matched control group of typically developing children. Each child with CP completed a full dose of LSVT LOUD in addition to a 12-week maintenance program in order to examine slow-phase neural changes. Children were recorded at PRE, POST, and 12-weeks FUP, and included both trained and untrained tasks. Outcome measures included determining white matter integrity using diffusion tensor imaging (DTI) to measure slow and fast phase changes in fractional anisotropy (FA) for two motor tracts and five association tracts, as well as acoustic measures of voice and speech. The authors stated that the CP group showed an increase in FA in several motor and association tracts at POST and FUP. Acoustic data on untrained tasks were correlated with changes in FA detected at POST and FUP.

A strength of this study in contrast to the previous three, was that no data were collected or analyzed by individuals associated with LSVT Global, the treatment protocol, or its delivery. However, they were still involved in the writing and publication of the research article. Reed and colleagues (2017) also demonstrated sound inclusion and exclusion criteria to help control for variability in the patient population. They also reported high intra- and inter-measurer reliability, and clearly defined their outcome measure selection rationale, data collection, and analysis procedures for easy replication. Moreover, naïve listeners passed a hearing screening and did not have a background in speech and language training. To ensure reported effects were not a reflection of a development confound, correlations between dependent measures and age were calculated, with no significant correlations or outliers identified. As hypothesized, the typically developing group did not show changes in FA that met either fast or slow phase criteria indicating significant change, which increases the credibility of findings in the CP group.

This study also had several limitations, including a relatively small sample size for a mixed design, and a large age range for participants, which makes generalizability more difficult. The authors also reported on several statistical trends in addition to significant findings, however they did not clearly state that these trends do not indicate statistically significant change. This made results look more promising than they were, as none of the association tracts met criteria for fast phase change and only one motor tract reached statistical significance for fast phase changes. Additionally, none of the motor tracts reached significance for slow phase changes, and a couple of association tracts showed an increase in FA, possibly indicating slow phase change. Evidently, the results are much more limited than the authors concluded in their paper. Moreover, the typically developing children also demonstrated changes in vocal loudness and diadochokinetic (DDK) performance over the three recording sessions, which makes the same reported improvements for the CP group equivocal. Lastly, part of the authors' rationale for looking at white matter tracts was previously reported altered integrity of white matter in children with CP compared to TD peers. However, no major differences were found at PRE between the two groups, which brings into question the

validity of the outcome measure itself. As such, the findings of this study are somewhat suggestive, and do provide the first preliminary neuroanatomical evidence for neuroplasticity changes following LSVT LOUD in this patient population.

Bakhtiari, Cummine, Reed, Fox, Chouinard, Cribben, and Boliek (2017) tested the same 8 children and used the same general design as the previous study, but this time aimed to examine potential neural changes using fMRI to demonstrate post-treatment connectivity changes using graphical models. 16 bilateral brain regions of interest based on previous speech, language, and neuroanatomical literature were selected for examination. Results demonstrated reduced neural activity in regions associated with decreased motor system effort, and increased activity in a region associated with contribution to decision making processes for the CP group. Post-treatment changes in connectivity between areas related to the motor speech feedback system suggests greater recruitment of this system and less reliance on the feedforward control system, which is a desired outcome for this kind of neuroplasticity treatment.

The authors of this study clearly defined their methods, outcome measures, data collection, and analysis procedures for replication. Similar research has also been conducted on Parkinson's patients following LSVT LOUD, so this type of outcome measure allows for a more direct comparison to the original treatment population. Moreover, the authors provided a rationale for looking at both sides of the brain based on recent research findings of a more bilateral language network in children compared to adults. They also used the DIVA model proposed in previous speech and language literature to make inferences about the functional meaning of the fMRI findings on speech processing and output. The authors employed appropriate statistical tests for the data, however they did assume normal distribution without reporting having calculated this. Moreover, the researchers averaged the data for each control participant over the three recording sessions in order to increase statistical power based on the assumption that there would be few to no brain activity changes across the same time span for the typically developing group.

Unfortunately, the authors still did not report on effect sizes or confidence intervals, which continues to limit overall statistical confidence. There was also still a reported conflicted of interest for some of the authors involved. Another large limitation of examining neuroanatomical effects versus direct therapeutic outcomes as a result of treatment is the larger amount of inferencing and subjective analysis involved. For example, observed change on an acoustic measure such as sound pressure level (SPL), indicates a change in loudness possibly as a result of treatment. Whereas, observed change on a brain measure such as increased activation of a certain brain area during a particular speech task leaves much more up to interpretation on behalf of the authors. Consequently, the level of evidence for this study is suggestive due to promising, but potentially biased findings. Results from the study are consistent with previous findings and do provide further evidence for treatment-based neuroplasticity.

Discussion

The biggest limitation of the current evidence base as a whole is that the research in this area is still in phase I validation. In contrast, research on LSVT LOUD in Parkinson's Disease (PD) has reached the highest level of validation using three randomized control trials, which increases confidence in those findings. Despite the individual limitations for each article, mostly all of the papers used appropriate designs, methods, and statistical analyses for a phase 1 trial, so there is some confidence in the current evidence base. Even small sample sizes and a lack of control group is considered appropriate for this phase of research.

The largest limitation in terms of the LSVT LOUD treatment protocol itself are mixed findings of maintenance of gains at FUP (follow-up) and carry-over into everyday speech. This is evident both in the research base with PD patients and in the articles included in this review. When choosing a treatment approach, it is desired that it will be functional for the individual in their daily lives and will last beyond the treatment phase. Currently this clinical confidence does not exist for LSVT LOUD, especially with this patient population as the research is still early on and limited.

Overall, the current review serves as suggestive preliminary evidence for therapeutic effectiveness of LSVT LOUD in children with dysarthria secondary to CP. This review also provides a compelling rationale for continued and expanded research into the effects of this treatment approach in this population.

Clinical Implications

Evidence-based therapy approaches for children with motor speech disorders continues to be a neglected area in research, and many speech-language pathologists are left to draw their own conclusions about the efficacy of differing treatment approaches. Currently, LSVT LOUD certified SLPs may take an advanced course to provide this therapy to children with cerebral palsy and down syndrome. LSVT Global Inc.'s offering of this advanced course for clinicians may be premature given the current level of evidence. However, the motor learning and neuroplasticity principles on which the

LSVT LOUD approach is based may be important principles for SLPs to include in their therapy toolkit when serving this patient population.

Future Research Considerations

While it may not necessarily be realistic for this rare of a population, larger sample sizes are recommended for future studies to increase statistical power. Moreover, utilizing smaller age ranges when possible, will allow for easier generalizability of findings. It is also recommended that future researchers report on all important statistical measures, including confidence intervals and effect sizes, in order to increase confidence in statistical findings. Moreover, it is highly recommended that future research is also conducted and written by individuals not associated with LSVT Global Inc. in order to eliminate potential bias. Based on the current findings, additional research exploring the impact of the same versus increased dosage of the adult LSVT LOUD protocol in the pediatric population may be beneficial. Lastly, it may be more useful for future research to focus on therapeutic effects by using outcome measures such as acoustic measures and perceptual listener ratings, as opposed to neurophysiological evidence, in order to simplify the interpretation of findings.

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