

Critical Review:
Effectiveness of Telehealth Delivery of the Lidcombe Program for Preschool Children Who Stutter

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This study reports a critical review examining the effectiveness of telephone-based and webcam-based telehealth delivery of the Lidcombe Program in reducing frequency of stuttering in preschool children. Studies evaluated included two randomized controlled trials, two case studies, and one single group pre-test post-test design. Significant, long-term reductions in stuttering frequency were observed across all studies. Overall, findings suggest that telehealth delivery of the Lidcombe Program appears to offer a viable alternative to standard delivery of the program for preschool children who stutter

Introduction

Approximately 5% of children begin to stutter with about 74% of these individuals recovering naturally within two years of stuttering onset (Lewis et al., 2008). However, current research does not enable the speech-language pathologist (SLP) to predict whether a child will recover naturally or will continue to stutter in the absence of intervention (Lewis et al., 2008). Therefore, it is essential that children who stutter receive intervention during the preschool years because stuttering becomes less responsive to treatment as children increase in age (Lewis et al., 2008).

The Lidcombe Program is a two-stage behavioral treatment program developed for children under the age of six who stutter (O'Brian et al., 2014). During Stage 1 of the program, parents attend one-hour weekly sessions at the clinic with their child (Lewis et al., 2008). The SLP trains parents to present three verbal contingencies for stutter-free speech (acknowledgement, praise, request for self-evaluation) and two verbal contingencies for unambiguous stuttering (acknowledgement, request for self-correction) (Onslow et al., 2003). The parent implements these contingencies when communicating with their child in everyday situations (Lewis et al., 2008). The SLP measures the child's stuttering frequency in percentage of syllables stuttered (%SS) during each clinic visit (Lewis et al., 2008). In addition, parents are trained to measure stuttering each day on a ten-point severity rating scale in which 1 = no stuttering, 2 = extremely mild stuttering, and 10 = extremely severe stuttering (O'Brian et al., 2014). Stage 1 of the program is concluded when stuttering is absent or extremely mild (Lewis et al., 2008). This is classified as a stuttering frequency measure that is below 1%SS and a severity rating of 2 or lower which is maintained for three consecutive weeks (Onslow et al., 2003). During Stage 2, parents gradually withdraw treatment

and attend fewer clinic visits while the maintenance of treatment gains are monitored (O'Brian et al., 2014).

The Lidcombe Program has been found to be an effective early intervention program for preschool children who stutter, resulting in reductions of stuttering frequency to near-zero %SS which are maintained for two to seven years post-treatment (Wilson et al., 2004). However, the Lidcombe Program may not be accessible to all children who stutter that may benefit from it. Attendance at weekly clinic sessions may be a barrier for families who live remotely from speech-language pathology services (O'Brian et al., 2014). This is particularly the case for rural areas of large countries that are not densely populated, such as Canada and Australia. It can also affect families that have limited access to transportation, chronic health problems, work commitments, or are dealing with multiple children. In addition, people who have limited access to SLP services are more likely to receive lower quality services (Wilson et al., 2004).

The World Health Organization recognizes telehealth as a potential solution to these issues (World Health Organization, 1998). Telehealth is defined as "the use of information technology and telecommunication to support or deliver health services to remotely located sites" (Wilson et al., 2004). The technology currently in use includes videoconferencing, telephone communication, and Internet communication (Wilson et al., 2004). There has been growing evidence for the efficacy of telehealth delivery of a range of healthcare services, including the treatment of stuttering (O'Brian et al., 2014). Therefore, it is important to examine the existing literature to determine whether telehealth delivery of the Lidcombe Program is an efficacious alternative to standard delivery of the program to ensure that there is equitable access to quality speech-language pathology services worldwide.

Objectives

The primary objective of this paper was to critically review the existing literature regarding the effectiveness of telehealth delivery of the Lidcombe Program in reducing frequency of stuttering in preschool children.

Methods

Search Strategy

Computerized databases including PubMed, PsychInfo, and CINAHL as well as Google Scholar were searched using the following search strategy: [(telepractice) OR (telehealth) OR (telerehabilitation) OR (telemedicine)] AND [(stuttering) OR (stutter)] AND (child) AND (Lidcombe Program).

Selection Criteria

Studies selected for inclusion in this review paper were required to investigate telehealth delivery of the Lidcombe Program for preschool children who stutter, deliver treatment to subjects by phone or webcam, and include an outcome measure of %SS. Some studies also included outcome measures of treatment efficiency and parent satisfaction; however, the results of these additional measures were not included in this review.

Data Collection

Results of the literature search yielded five articles that met the selection criteria. The articles included two randomized controlled trials (Bridgman et al., 2016; Lewis et al., 2008), two case studies (Harrison et al., 1999; Wilson et al., 2004), and one single group pre-test post-test study design (O'Brian et al., 2014).

Results

Telephone-Based Telehealth Delivery

The papers cited here all investigated the effectiveness of telephone-based telehealth delivery of the Lidcombe Program for preschool children who stutter.

Harrison et al. (1999) investigated the effectiveness of telephone delivery of the Lidcombe Program in a case study involving a 5 year 10 month old male classified as having a severe stutter (12.4-17.7%SS). An adapted version of the Lidcombe Program was administered. These adaptations included: (1) scheduled telephone consultations in place of clinic sessions, (2) mailed recordings of the child's speech in everyday speaking situations for measurement of fluency outcomes to replace online measurements in the clinic, (3) indirect, delayed observation of parents implementing verbal contingencies through review of mailed recordings with feedback provided during the following consultation, (4) minimal contact between the child and SLP. Measures of %SS and syllables per minute (SPM) were obtained from audio and video recordings by the treating SLP and a blinded specialist in stuttering

immediately pre-treatment and 12, 19, and 23 months post-treatment. A significant decrease in %SS and SPM, indicating near-zero stuttering levels, was achieved after a 9-month treatment period. These treatment effects were maintained at a 23-month follow-up.

A strength of this study is its clear description of Lidcombe Program adaptations and data collection methods. However, results from this study are limited due to modification of methodology over the course of treatment, such as the frequency of telephone consultations. Additional limitations include a small sample size, lack of appropriate statistical analyses, and use of a case study design. Overall, this study provides equivocal evidence that telephone delivery of the Lidcombe Program is effective in reducing frequency of stuttering in preschool children.

Wilson et al. (2004) presented five case studies investigating the fluency outcomes of preschool children receiving telephone delivery of the Lidcombe Program. Participants were selected if they had been diagnosed with a stutter by an SLP, were between two and six years of age, and were reported by parents to have been stuttering for at least two months prior to treatment. The Lidcombe Program was administered in accordance with the treatment manual with the addition of the adaptations implemented by Harrison et al. (1999). In addition, Wilson et al. (2004) provided supplemental video training materials and information sheets which were mailed to parents at various points throughout the course of treatment. Audio or video recordings were collected for each participant at 2 months, 1 month, and 1-week pre-treatment and at 1 week, 1, 2, 4, 6, 8, and 12 months post-treatment. These recordings were presented in a randomized order to an SLP specializing in stuttering who counted %SS and SPM for each sample. Appropriate statistical analyses were performed. All five participants reached the clinical criteria for completion of Stage 1 (%SS of less than or equal to 1 and daily severity rating of 1 or 2). Two children scored a mean of less than 1%SS and two children scored a mean of less than 2%SS at 12 months post-treatment. One child experienced a clinically significant relapse for a period during Stage 2, which was subsequently brought under control. This relapse was associated with parental withdrawal of treatment procedures and noncompliance.

Strengths of this study include clearly stated inclusion criteria, blinding of the SLP collecting %SS measures, and use of appropriate statistical analyses. However, the results of this study are limited by its small sample size and the uncontrolled nature of the study design. Overall, this study is suggestive that telephone delivery

of the Lidcombe Program effectively decreases frequency of stuttering.

Lewis et al. (2008) conducted a parallel group, open plan randomized controlled trial to investigate the efficacy of telephone-based telehealth delivery of the Lidcombe Program for preschool children who stutter. Participants were included if they were between 3 years to 4 years 6 months of age, had been stuttering for longer than six months, had received no previous or current treatment for stuttering, had a history of normal development apart from stuttering, and both the child and parents were proficient in English. This resulted in 22 children being selected. Participants were randomly assigned to receive either telephone delivery of the Lidcombe program by a trained SLP or no treatment. Children were treated with procedures as similar as possible to those in the Lidcombe Program treatment manual, incorporating the necessary adaptations made during the Harrison et al. (1999) and Wilson et al. (2004) studies. Measures of %SS were obtained from audio recordings of the children's speech in everyday situations by the treating SLP and a blinded specialist in stuttering at 1-week pre-randomization, 9-months post-randomization, and 12-months after Stage 1 completion. %SS scores at 9-months post-randomization were 1.1 for the treatment group and 1.9 for the no treatment group. Appropriate statistical analyses were completed. An ANCOVA demonstrated a 73% decrease in %SS in the treatment group immediately post-treatment compared to the no treatment group. Only one child was noted to be stuttering 12-months after completion of Stage 1.

Strengths of this study include clear inclusion criteria, high inter-rater reliability (.96), and a strong study design involving randomization of participants to treatment group prior to inclusion decision. A potential limitation of this study is its inclusion of a child outside the age criteria and the presence of more girls than boys within the control group. Overall, this study provides compelling evidence that telephone delivery of the Lidcombe Program is an effective treatment for preschool children who stutter.

Webcam-Based Telehealth Delivery

The papers cited here all investigated the effectiveness of webcam-based telehealth delivery of the Lidcombe Program for preschool children who stutter.

O'Brian et al. (2014) investigated the effectiveness of webcam delivery of the Lidcombe Program using a single group pre-test post-test design. Participants were two males and one female ages 3 years 6 months, 4 years 3 months, and 4 years 9 months, respectively. All children had been stuttering for longer than 6 months,

had not received any previous speech therapy, were diagnosed with a stutter during a webcam assessment by the SLP, and had access to the Internet and a webcam. The treatment process followed the Lidcombe Program treatment manual, with the following necessary adaptations for webcam: (1) measures of %SS were collected weekly over webcam at the beginning of the session while talking with the child or observing the child talking with a parent in view of the camera, (2) more time was spent describing procedures and role playing with parents rather than direct demonstration with the child, (3) SLP spent less time, overall, directly communicating with the child. Speech assessments were conducted at pre-treatment, on completion of Stage 1, and 6-months after Stage 1 completion. Audio recordings of each child's speech were collected and scored by two blinded SLPs who measured %SS. Appropriate statistical analyses were completed. At 6-months post-Stage 1, all children demonstrated reductions in stuttering to less than 1%SS with overall reductions from pre-treatment to 6-months post-treatment ranging from 69-99%.

Strengths of this study include high inter-rater reliability (.89), use of appropriate statistical analyses, and consistent participant characteristics, with the exception of one participant with a mild language delay. However, the use of a single group pre-test post-test design limits results of this study as a direct comparison cannot be made to standard delivery of the Lidcombe Program. This study is further limited by its small sample size and lack of long-term follow-up. Overall, this study is suggestive that webcam delivery of the Lidcombe Program is an effective treatment for reducing %SS in preschool children who stutter.

Bridgman et al. (2016) conducted a parallel group, open plan randomized controlled trial to investigate the efficacy of webcam delivery of the Lidcombe Program compared to traditional, clinic-based delivery of the program. Participants were selected if they were between the ages of 3 years and 5 years 11 months and had been stuttering for longer than 6 months, a diagnosis of stuttering by the parent and assessing SLP, home access to the Internet and webcam with adequate audio and visual quality, and functional child and parent English language skills. Children with attention-deficit/hyperactivity disorder or intellectual disability were excluded. 49 children were randomly assigned to either an experimental group receiving the Lidcombe Program in their homes via webcam or a control group receiving standard delivery of the program in a clinic setting. All participants were treated according to the Lidcombe Program treatment guide. Audio recordings were collected at pre-randomization, 9-months post-randomization, and 18-months post-randomization from

which %SS scores were obtained for each child by two blinded SLPs specializing in stuttering. Appropriate statistical analyses were completed. Results indicated that 62% of children in the control group at 9-months post-randomization and 94% at 18-months post-randomization had completed Stage 1. In the experimental group, 67% at 9-months post-randomization and 100% at 18-months post-randomization had finished Stage 1. In addition, the data demonstrated that there was insufficient evidence of a post-treatment difference between the standard and webcam delivery of the Lidcombe program in %SS at 9 months and 18 months post-randomization.

Strengths of this study include clear inclusion and exclusion criteria, high intra-rater (.99) and inter-rater (.83) reliability, and its random allocation of participants to treatment condition. However, the results may be limited due to the use of the same treating SLP for all participants in all groups as the SLP was unable to be blinded to treatment condition and may introduce a bias towards a particular delivery model. Overall, this study provides compelling evidence that webcam delivery of the Lidcombe Program is similar in effectiveness to standard delivery of the program in reducing frequency of stuttering.

Discussion

Across studies, it was found that all participants receiving the Lidcombe Program via telehealth, whether by telephone or webcam, reached near-zero stuttering levels (%SS of less than 1). In addition, long-term maintenance of these treatment effects was observed in each study. Taken together, the results of the five reviewed studies provide equivocal to compelling evidence that the Lidcombe Program is an effective alternative to standard delivery of the program in terms of reducing frequency of stuttering.

The study by Harrison et al. (1999) provides equivocal evidence due to its low-level study design, extremely small sample size, modification of methodology during implementation, and lack of appropriate statistical analyses. Wilson et al. (2004) and O'Brian et al. (2014) provide suggestive evidence as they provided a clear description of methods and employed appropriate statistical analyses. However, these studies were still limited by their small sample sizes and the uncontrolled nature of their study designs. Due to the likelihood of natural recovery within the stuttering population, larger samples are needed to ensure treatment effects are a result of the treatment itself rather than from natural recovery. Studies conducted by Lewis et al. (2008) and Bridgman et al. (2016) provide the most compelling evidence due to their use of randomized controlled trials, enabling a direct comparison to be made between

groups. Lewis et al. (2008) provides a direct comparison between telephone delivery of the Lidcombe Program and no treatment, allowing them to conclude that the significant reduction in stuttering frequency observed in the treatment group is greater than that which may be seen from natural recovery. Similarly, Bridgman et al. (2016) directly compared webcam and standard delivery of the Lidcombe Program, allowing them to conclude that the lack of a post-treatment difference found between groups suggests that these delivery methods are similar in their effectiveness.

Some of the studies in this review included secondary outcome measures of treatment efficiency. Preliminary results from this data suggests that, while telehealth delivery of the Lidcombe Program has been found to successfully reduce stuttering frequency, it may be less efficient when compared to standard delivery. Both Wilson et al. (2004) and Lewis et al. (2008) explored the efficiency of telephone delivery of the Lidcombe Program in terms of number of weeks, number consultations, duration of consultations, and total clinician time and found that, overall, telephone delivery was two to three times less efficient than standard delivery. In contrast, there were inconclusive results regarding the efficiency of webcam delivery of the Lidcombe Program. O'Brian et al. (2014) found that while webcam delivery was more efficient than telephone delivery, it was still two times less efficient than standard delivery. However, Bridgman et al. (2016) found that webcam delivery of the program was equal or more efficient than standard delivery. However, none of these studies controlled for stuttering severity in their participants. It has been widely documented that higher pre-treatment stuttering frequency may impact the number of consultations required to meet Stage 1 criteria of the Lidcombe Program (O'Brian et al., 2014). Therefore, these findings warrant further study to clarify the relative costs and benefits of telehealth delivery of the Lidcombe Program.

Future Research Considerations

It is recommended that further research be conducted to clarify and confirm the effectiveness of telehealth delivery of the Lidcombe Program. In order to improve the level of evidence provided by the existing literature, it is suggested that future research:

- a) Employ study designs that offer a stronger level of evidence, such as randomized controlled trials comparing telehealth and standard delivery of the Lidcombe Program, to determine whether these methods are equally effective.

- b) Recruit larger sample sizes and use clear inclusion and exclusion criteria such that results can be more appropriately applied to clinical practice.
- c) Involve more than one SLP in delivering treatment to participants to ensure that treatment effects are independent of the clinician implementing therapy.
- d) Continue to include outcome measures of stuttering frequency and treatment efficiency to confirm the relative costs and benefits of telehealth delivery.

Conclusion

Overall, both telephone and webcam delivery of the Lidcombe Program were found to result in significant, long-term reductions in stuttering frequency. Therefore, telehealth delivery of the Lidcombe Program appears to offer a viable alternative to standard delivery of the program for preschool children who stutter.

Clinical Implications

As technology continues to advance, telepractice may be a useful tool for clinicians in ensuring equitable access to speech-language pathology services. However, while current research suggests that telehealth delivery of the Lidcombe Program is effective in reducing stuttering frequency, it is recommended that clinicians be cautious when implementing the findings of these studies into clinical practice. Due to limitations in study design and sample size as well as the possibility of decreased treatment efficiency, a telehealth delivery model should only be considered as a treatment option when standard delivery is not accessible to the family. In addition, it may be necessary to determine eligibility for telehealth services on a case-by-case basis to ensure that the family will be able to comply with the program requirements.

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