

Critical Review: Is Telehealth Delivery of Fluency Intervention Effective for People who Stutter?

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This critical review investigates evidence regarding the efficacy of fluency intervention programs delivered via a high- or low-tech telehealth format. Study designs include expert opinion, case reports, single subject experimental designs, and randomized controlled trials. Overall, the evidence supports the use of this therapy delivery format as an effective alternative to face-to-face contact with specific fluency intervention program and populations. Clinical recommendations are discussed.

Introduction

Stuttering is a distressing communication disorder that usually begins around 3-4 years of age (Lewis *et al.*, 2008). Of the approximately 5% of children who go through a period of disfluency, about 75% will recover naturally within a couple of years (Lewis *et al.*, 2008). Despite the likelihood that natural recovery will occur, it is generally recommended that treatment begin during the pre-school years, as stuttering becomes less responsive to treatment as children get older (Guitar, 2006).

Established treatment methods for childhood stuttering can include parent-delivered programs such as the Lidcombe Program (Harrison *et al.*, 1999), in which parents are trained to deliver verbal contingencies for their child's speech. Acknowledgment, praise and self-evaluation are used for stutter-free speech; acknowledgement and request for self-corrections are used for unambiguous stutters (Wilson *et al.*, 2004).

Persistent stuttering is generally much less responsive to treatment (Guitar, 2006). Adults who stutter are also more likely to exhibit social maladjustment, phobias, or under-realized occupational achievement (Carey *et al.*, 2009).

Currently used techniques for treatment of chronic stuttering include speech-restructuring techniques that alter speech at the levels of breathing, phonation, and articulation (Carey *et al.*, 2009). Speech restructuring programs usually include use of prolonged speech, such as in the Camperdown Program (O'Brian *et al.*, 2008; Carey *et al.*, 2009). The Camperdown Program provides clients with spoken models of prolonged speech, and they are instructed to use whichever features of the speech they require in order to control their fluency (O'Brian *et al.*, 2008).

Regardless of age or fluency intervention method used, treatment for stuttering usually requires intense and/or prolonged contact with a speech-language pathologist

(Sicotte *et al.*, 2003). In addition, generalization and maintenance of therapy gains can be difficult, especially in adult populations (Theodoris, 2008). These attributes of fluency intervention can be problematic for clients who live in remote locations, or who cannot access services for mobility, transportation, or financial reasons (Wilson *et al.*, 2004; Theodoris, 2008). This is a particular problem in countries such as Canada, where the majority of SLP services are clustered in urban centres (Kully, 2002; Carey *et al.*, 2009).

Telehealth is the use of information technology and telecommunications to support or deliver health services (PRHCIT, 1996). Telehealth can be either low- or high-tech, depending on the medium employed. Low-tech delivery generally refers to telephone contact, whereas high-tech delivery includes an interactive visual medium, such as videoconferencing (O'Brian *et al.*, 2008). Telehealth has been shown to be a viable treatment modality in several areas of speech and language (Theodoris, 2008). Telehealth has good potential for fluency intervention, where in-clinic improvements often fail to carry over to other settings (O'Brian *et al.*, 2008). If effective, this service-delivery model has the potential to ameliorate some of the difficulties associated with delivering stuttering therapy to remote or isolated clients (Lewis *et al.*, 2008).

Objectives

The primary objective of this review was to critically evaluate the evidence surrounding the efficacy of telehealth delivery of fluency intervention on stuttering outcomes in children and adults. A secondary objective was to develop recommendations for clinicians considering implementing this service delivery model.

Methods

Search Strategy: Computerized databases including CINAHL, PubMed, Proquest, Medline, and OVID were searched. The following key terms were targeted:

[(stuttering) OR (fluency disorders) OR (disfluency)] AND [(telehealth) OR (telemedicine) OR (distance intervention)]

The search was limited to articles written in English.

Selection Criteria: Studies selected for review were required to report on clinical applications of high- or low-tech telehealth adaptations of traditional fluency interventions. No limits were placed on the subject demographics or specific intervention program designs.

Data Collection: Results of the literature search yielded 8 articles that met the selection criteria. These included the following study designs: expert opinion (2), case study (1), replicated single subject experimental design (3), and randomized controlled trials (2). The intervention programs reflected in the studies included the following: Parent-delivered Lidcombe Program for children (3), Camperdown speech-restructuring program for adults (2), and Unspecified (3).

Results

There are inherent differences in treatment approaches for young children versus older children or adults who stutter. Therefore, whenever possible, the parent-delivered Lidcombe program will be considered separately from speech-restructuring programs.

Expert Opinion

Respected experts in the field can make valuable judgments about the efficacy and feasibility of new treatment options. However, without explicit critical appraisal, the work is open to subjective bias.

In 2000, Kully began reporting on the use and benefits of telehealth to treat adult stutterers at the Institute for Stuttering Treatment and Research (ISTAR) in Edmonton, AB. Her group regularly used telephone contact for fluency maintenance sessions. They expanded to the use of video-conferencing for maintenance with a 38-year-old man, and reported positive clinician and client experiences anecdotally. However, this client had previously been seen face-to-face, so his success cannot be attributed solely to therapy via telehealth.

In 2002, Kully reported more extensively on ISTAR's use of telehealth. She described the use of low- and high-tech options for serving children undergoing a parent-delivered program similar to Lidcombe, and adolescents/adults learning speech restructuring techniques. All of the clients served this way were unable to attend traditional sessions. Although informal reports were positive, objective data was not reported, and Kully reported that the clients chosen for non-traditional therapy were highly stimutable at intake.

Both of these early ventures into telehealth may be cautiously interpreted as justification for further exploration of this treatment modality.

Lidcombe

Case Study

Case studies are useful to describe novel treatment situations or provide an in-depth look at a single individual. However, they cannot be used to draw cause and effect conclusions, and their results are not readily generalized.

Harrison, Wilson, and Onslow (1999) reported a case study involving a 5-year-old boy treated entirely via low-tech telehealth using the parent delivered Lidcombe program. Results showed that the subject achieved and maintained near-zero stuttering levels up to 23 months post-treatment, with an associated increase in speaking rate (syllables per minute).

The client and adaptations to the Lidcombe program for telehealth delivery were described in adequate detail to allow replication by an outside investigator. Intra- and inter-rater reliability was carefully established on all measures at all sampling points.

This was a well-designed case study. However, bias was likely as neither the client nor the investigators were blinded. The family actively sought out this treatment and was noted to be particularly motivated, introducing selection bias. The evidence provided by this report is suggestive, but the results cannot be readily generalized.

Single Subject Design

Single subject experimental designs can be very powerful, especially if well designed with frequent sampling. Generalizability is enhanced if the results are replicated across a number of subjects.

In 2004, Wilson, Onslow and Lincoln described an extension of their research with the Lidcombe adaptation. They reported on five single-subject experimental cases involving children ages 3;5 to 5;7 who were followed up to a year post-treatment. In all cases, visual inspection indicated that criteria were met for the children to complete stage one, which is defined as <1 Percent Syllables Stuttered (%SS) and daily parental severity ratings of 1-2 on a 10-point scale. In addition, at 12-months post-treatment %SS remained at or near criteria (range from 0.6-1.4 %SS) for 4/5 clients.

Wilson *et al* (2004) thoroughly described further refinements to the Lidcombe program required for the telehealth format, and the methods would be feasibly replicated. Each subject is described in detail, including stuttering severity. Variability was addressed by sampling across at least 3 speaking situations at each time point. Covert recordings were included in an effort

to reduce the potential influence of recording on speaking performance. Each sample was quantified by a blinded rater, and a portion were independently verified for reliability.

Although the single subject results were replicated across 4/5 cases, data is missing for the 5th participant beyond one week into treatment. There was an unusually high number of families (13/18) recruited for the study who dropped out prior to the start of data collection, indicating decreased desirability of this format for some clients. Additionally, preliminary examination of treatment efficiency suggested that telehealth delivery required more clinician time than standard intervention.

Overall, these results are highly suggestive of treatment efficacy. However, the clinical importance may be limited by the decreased treatment efficiency.

Randomized Control Trial (RCT)

RCTs are the most powerful of all study designs as they allow comparison between groups. However, conclusions can be constrained by threats to validity or reliability of the study design.

Lewis, Packman, Onslow, Simpson and Jones (2008) provided further evidence to support the efficacy of their Lidcombe adaptation. They reported on the results of a parallel group RCT with multiple blinded outcome measures. At 9-months post randomization, their experimental group (n=9) showed a significantly larger decrease in %SS compared to the no-treatment control group (n=13). At 9-months post-randomization, children in the control group were offered the same therapy as the experimental group had received. At 18 months post-randomization, 7/8 subjects in this cohort were re-assessed, and showed a significant decrease in %SS.

Lewis *et al*'s (2008) study features systematic subject and methodological descriptions. Randomization was blinded, and groups were considered representative. Subsequent analysis using appropriate analysis of covariance (ANCOVA) showed that the experimental and control groups were not similar at baseline in regards to stuttering severity. A paired t-test suitably established a within-group difference for the control group who subsequently received treatment. Outcome measurement was blinded, and reliability was rigorously established. Treatment fidelity was verified by an independent observer familiar with the procedures.

While the study design was sound, it would be prudent to limit interpretation based on this small sample size. The authors employed intention-to-treat analysis when a child older than the inclusion criteria was mistakenly randomized to a group. However, they did not include data from 15/37 clients and families who chose not to

participate after having been originally recruited. These included 5 who obtained SLP services locally rather than participating in a telehealth program. Again, this may indicate a client preference for traditional therapy delivery. In addition, visual inspection of data related to clinician workload indicated that the adapted program required an average of 3x the resources of traditional therapy. These results are compelling due to their strong validity and reliability. However, once again, the clinical importance is limited by decreased treatment efficiency.

Speech Restructuring Single Subject Designs

Sicotte, Lehoux, Fortier-Blanc and Leblanc (2003) investigated the use of videoconferencing for fluency intervention and maintenance with 6 clients in Quebec, Canada. Data were collected at baseline, following a 12 session treatment period, and at 6-months post treatment. Results from qualitative data collection suggested that clinicians were satisfied with most of the technical and clinical aspects of the sessions and that clients and their families had seen some reduction in stuttering frequency and concomitant acquisition of better communication skills. Visual inspection of %SS pre- and post-treatment revealed that all patients showed reduction in stuttering that was at least partly maintained until the end of the follow-up period.

All patients completed the study with 100% attendance in therapy. All 6 clients showed similar trends across pre- and post-therapy measures, and data for %SS was averaged across at least two samples at each collection. Sicotte *et al* took a broad approach to outcome assessment, including questionnaires to assess patient and clinician perceptions in addition to quantifiable stuttering measurements.

Despite adequately defined inclusion criteria, subjects were not described in detail for variables such as baseline stuttering severity or exposure to previous treatment. Variability in %SS was not reported across different speaking situations, and the clinical environment may have biased speech patterns of the subjects during sample collection. The fluency treatment program utilized was described only as "currently accepted and well-used procedures", and no attempt was made to compare the outcomes to a control group. Only short term outcomes were assessed, and there were no efforts to establish reliability of data collection. Furthermore, the clients ranged in age from 4-19 years old, which suggests that the approach and activities chosen would have varied considerably from case to case. These factors put the validity and reliability of the results into question.

O'Brian, Packman and Onslow (2008) reported the results of a trial to establish the viability of telehealth

delivery of the Camperdown Program. Eight adults took part, and although there was considerable individual variability, they showed an average 82% reduction in %SS immediately post-treatment. This reduction remained at 74% at 6-month follow up. O'Brian *et al* also gathered data regarding speech rate, showing an average Syllables Per Minute (SPM) increase from 184 SPM to 228 SPM. Self-rated severity ratings improved for the majority of clients in most situations. Finally, "naturalness ratings", as judged by naïve listeners, were comparable to a control group of speakers who had not undergone speech restructuring.

O'Brian *et al* provided excellent descriptions of their participants and the adaptations made to traditional Camperdown program. All participants completed the trial. Sampling was thorough and considerable effort was made to reduce the influence of the clinician on speech performance during collection. The authors also took care to include holistic outcome measures designed to capture many facets of fluency; % SS in naturalistic situations, self-ratings of severity for 5 different scenarios, and naïve-listener perceptions of naturalness. Samples were quantified by blinded judges, and a portion were re-checked for inter and intra-judge reliability. Informal analysis of clinician contact time revealed that telehealth delivery of the Camperdown program was *more* efficient than face-to-face therapy.

This trial had only a small number of participants, and no comparison was made either to traditional service delivery or to a no-treatment group. Long-term outcomes were not established. In addition, the heavily self-directed nature of this service delivery model may have decreased treatment fidelity, and no attempt was made to establish consistency amongst participants. This study provides suggestive preliminary evidence for the efficacy of telehealth delivery of the Camperdown Program. The clinical importance is enhanced by the finding that telehealth is more efficient than traditional delivery.

Randomized Control Trial

An extension of the Camperdown adaptation was reported in 2009 by Carey, O'Brian, Onslow, Block, Jones and Packman. They carried out a parallel group RCT comparing telehealth delivery of the Camperdown Program (n= 20) to traditional face-to-face therapy (n=20). Results of an ANCOVA accounting for differences in baseline %SS indicated no significant differences between the groups at 9-months post randomization, immediately post-treatment, 6-months post treatment, or 12-months post treatment. Secondary outcome measures included self-reported severity across several scenarios, speech naturalness, treatment satisfaction and a measure of efficiency. A 2 sample t-test indicated that both groups experienced similar reductions in self-reported severity. There was also no

significant difference between the telehealth and face-to-face groups as judged by a naïve-listener for speech naturalness. However, both groups were rated as significantly less natural in comparison to a non-stuttering control group. A treatment satisfaction survey showed no differences between groups with one exception; the telehealth group was significantly more likely to rate the therapy as "extremely convenient". Analysis of treatment efficiency showed the telehealth group required an average 221 minutes less contact time (95% C.I. = 387-56 min, p= 0.01) than the face-to-face group.

This study featured well-described participant selection and methodology. Groups were judged to be representative and similar on key variables. Randomization was blinded and all but 3 clients completed the trial, for whom intention-to-treat analysis was completed. Outcome measures were comprehensive, including naturalistic sampling, and showed high intra- and inter judge reliability. Participants were also followed up to 1-year post treatment, extending previous findings that this model of service delivery can result in long-term fluency gains.

This RCT would have been stronger had there been larger participant numbers. The authors note that four participants who met inclusion criteria at recruitment failed to demonstrate %SS criteria at randomization, but do not indicate which group these clients were in, which could have introduced bias. All client contact was managed by the same primary Speech Pathologist, who was necessarily not blind to group allocation. Overall, this study has strong validity and reliability, making it compelling evidence that telehealth Camperdown delivery is not inferior to traditional face-to-face therapy. In addition, formal measures of treatment efficiency make the findings clinically important.

Discussion

There are difficulties in studying the use of telehealth with stuttering intervention. There is a relatively small population compared to other speech disorders and the nature of the interventions used often preclude double-blinding and random allocation. In spite of this, an emerging evidence base has provided consistent suggestive to highly-suggestive evidence that telehealth delivery of stuttering intervention can be an effective alternative to traditional face-to-face service delivery models. All of the studies reviewed concluded that telehealth fluency intervention was effective for the specific individuals or groups they targeted. However, the field of evidence is limited by small samples and lack of large-scale comparisons to either matched case-controls or groups receiving traditional therapy or no-treatment. In addition, most previous studies have

included only clients for whom traditional face-to-face therapy was not an option.

Fluency intervention programs are variable and only two telehealth adaptations (Lidcombe and Camperdown) have been studied in detail. Both of these programs were developed by the same research group, and the majority of evidence available thus far comes from their own publications. There are inherent differences between the two main therapy programs studied as well as the specific populations that each program targets, making it difficult to draw broad conclusions. Findings show that Lidcombe can be successfully implemented via telehealth, but the clinical implications are limited by trends towards decreased efficiency compared to a face-to-face model. On the other hand, the Camperdown Program has been shown to be both efficacious and efficient in terms of clinical resources required. This suggests that it may be the nature of the intervention itself, not the service delivery model that contributes to this aspect of clinical importance.

The evidence base also reveals several cautions required for clinicians considering using telehealth as part of their fluency practice:

- a) The nature of disfluent speech often encompasses non-verbal struggle behaviors that can be difficult to capture via audio-only assessment.
- b) Comprehensive outcome measures should be employed to capture aspects of disfluency that transcend %SS.
- c) The technology of telehealth is not uniform and is bound to change with time. Findings from previous studies may not be generalizable across technologies.
- d) Evidence regarding client satisfaction with a telehealth alternative is equivocal.

Conclusion

Broader study of the application of telehealth to fluency intervention is required, especially incorporating comparisons to matched case-controls, groups receiving traditional therapy, or no-treatment groups. However, with case-by-case consideration of client variables such as age and access to services, as well as careful implementation of an established adapted fluency intervention program, telehealth delivery can be an effective alternative to face-to-face intervention.

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