

Critical Review: Short and Long-term Outcomes of Intensive Treatment for School-age Stuttering

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This critical review examines the short and long-term outcomes of intensive treatment for school-age individuals who stutter. A literature search was conducted and study designs included one controlled trial and five quasi-experimental studies. Overall, there is strong research evidence that fluency shaping approaches to intensive treatment for school-age stutters result in positive short and long-term outcomes. Evidence for stuttering modification approaches is more guarded. While not addressed empirically, all studies stressed the importance of a strong maintenance component to any intensive treatment program.

Introduction

Stuttering is a relatively common childhood communication disorder that typically develops between the ages of 2-4 years old (Laiho & Klippi, 2007). While various studies have shown that anywhere from 20-80% of pre-school children naturally recover from stuttering (Andrews, Guitar & Howie, 1980; Yairi & Ambrose, 1992), the prevalence of school age children who stutter is still estimated at 1%, falling slightly to 0.8% after puberty (Andrews et al, 1983; Craig, 2000). There is significant evidence that school age children are much less likely to recover spontaneously from stuttering (Craig, 1996; Yairi and Ambrose, 1992). For those individuals who continue to stutter, the disorder can have lasting social, emotional, and even vocational impacts (Craig, 1990; Craig, 2000). Thus it is imperative that we are able to provide effective and efficient treatment for these individuals.

Intensive treatment programs for stuttering involve high frequency and intensity treatment (often 5-6 hours a day for anywhere from 1-3 weeks) and are typically based on one of two general approaches: fluency shaping or stuttering modification (Guitar, 2006). Fluency shaping approaches use operant conditioning to reinforce fluent speech (often beginning at a slowed rate) with the ultimate goal of entirely fluent speech (Guitar, 2006). Treatment programs that may use this approach include smooth speech, gradual increase in length and complexity of utterance (GILCU), electromyography (EMG) feedback, and delayed auditory feedback (DAF). Stuttering modification therapies aim to reduce the tension and struggle associated with stuttering as well as reducing negative emotions such as shame and fear. Specific techniques used in stuttering modification include pull-outs, cancellations, and preparatory sets (Guitar, 2006).

Intensive programs offer advantages over non-intensive treatment for both the client and the clinician. From the client's perspective, one of the major benefits may be increased motivation and decreased attrition. Motivation is one of the greatest problems in stuttering therapy (Fawcus, 1970). Intensive programs can increase client and family motivation by providing nearly immediate results; access to fluent speech is often achieved in the first few days of treatment. This rapid access to fluent speech reinforces the client and family efforts and may result in a

lower drop-out rate (Druce & Debney, 1997). Drop-out rate may also be decreased simply due to the shorter treatment schedule, there is less of a chance that major life events might interfere with treatment (Druce & Debney, 1997). Intensive programs also offer multiple opportunities to practice developing fluency skills in a range of speaking situations. From the clinician's perspective, knowing that the majority of a client's treatment can be carried out within days or weeks (versus years) may help in management of caseloads. (Druce & Debney, 1997; Fawcus, 1970).

There is a considerable body of research regarding the short and long-term effects of intensive treatment approaches with adults who stutter (Andrews et al, 1980; Howie, Tanner, & Andrews, 1981; Boberg & Kully, 1994; Blomgren, Roy, & Callister, 2005). The body of literature regarding the impacts of intensive treatment programs on school-age children is much less established. Conture and Guitar (1993) and Schwartz (1993) describe a number of challenges facing the school-age population. School-age individuals who stutter have been stuttering for a longer time than pre-school children and thus the behaviours, thoughts and emotions are much more "entrenched" (Conture & Guitar, 1993). School-age individuals are also exposed to increasing influence from their peer group, who are often much less forgiving than pre-school playmates (Conture & Guitar, 1993; Schwartz, 1993). Given the unique characteristics represented by this population, it is a group that should not be overlooked (Conture & Guitar, 1993).

Objectives

The purpose of this paper is to critically examine the existing literature regarding the short and long-term effects of intensive treatment programs for school-age individuals who stutter. Evidence based recommendations regarding the use of intensive treatment programs for school-age individuals who stutter will be developed, and future research directions will be suggested.

Methods

Search Strategy

Computerized databases including SCOPUS, MEDLINE, CINAHL, EMBASE, AMED, PubMed,

PsycINFO, ComDis Dom, and Cochrane Library were searched using the following search terms: ((fluency) OR (stuttering)) AND ((intensive treatment) OR (intensive therapy))
A manual search was conducted to locate any studies heavily referenced in articles from the computerized search.

Selection Criteria

Studies selected for inclusion in this critical review were required to investigate the outcomes of intensive treatment for school-age individuals who stutter.

Data Collection

Results of the literature search yielded one controlled trial and five quasi-experimental studies. Three articles from the British Journal of Disorders of Communication published in the 1970's were also reviewed (Fawcus, 1970; Rustin, 1978; Watts, 1971); although they reported some positive results, they were primarily descriptive in nature and thus were used for informational purposes only. The six studies considered in this review used one of two broad fluency therapy approaches: fluency shaping or stuttering modification.

Results

A randomized controlled trial (RCT) is often considered the gold standard in research design, however in certain instances, using an RCT can be inappropriate or impossible (Greenhalgh, 2006). All of the studies included in this review were subject to practical and ethical constraints relating to blinding procedures and withholding treatment. Double-blinding was not possible as subjects participated in their own treatment and thus were necessarily aware of which treatment they were receiving. An ethical concern faced by researchers in this field is how to create a control group without withholding treatment to eligible individuals. A commonly accepted practice is to use individuals who are on the waiting list for treatment to form a control group. This practice does not allow for true random assignment, however it is considered an acceptable compromise given the ethical constraints (Greenhalgh, 2006). Given that all of the researchers were faced with the same practical and ethical challenges, the studies in this review will be analyzed within these constraints.

Intensive Treatment Programs Using a Fluency Shaping Approach

Craig and Hancock (1996) conducted a multi-centre trial with school-age children (9-14 years) comparing three treatments where Intensive Smooth Speech (ISS) (n=27) and Electromyography Feedback (EMG) (n=25) were offered at one clinic and Home Based Smooth Speech (HBSS) (n=25) was offered at another clinic. Subjects were assigned to treatment groups based on when and where they sought treatment. The control group (n=20) was recruited from the waiting list at the second clinic. ISS and EMG were intensive approaches and involved small group treatment sessions of 6.5 hours/day for five consecutive days with six periodic maintenance sessions over the next year. Pre and post treatment assessment results were

analyzed with a multivariate analysis of variance (MANOVA) and subsequent ANOVAS, which was appropriate for the group design. Results of the study showed that when compared with pre-treatment measures, all treatment groups showed a significant decrease in percent syllables stuttered (%SS) and anxiety, and a significant increase in naturalness and syllables per minute (SPM), at all points of assessment. The greatest changes were seen immediately post-treatment with slight increases in %SS up to 12 months. Significant differences were noted between the control group and each of the treatment groups, however there were no significant differences noted between the treatment groups on any of the outcome measures. Using Pearson's product-moment correlation coefficient, r , researchers found a positive, statistically significant correlation between pre-treatment severity and treatment outcomes at 12 months.

Craig and Hancock (1996) carried out a thorough, well-designed study. Despite the inherent practical and ethical limitations discussed above, the authors made efforts to limit possible sources of bias and error. One potential concern was that subjects were not randomly assigned to treatment groups, and thus may have differed on some important characteristics even before they received the treatment. The authors strengthened the validity of their findings by showing that the groups did not differ significantly on age, sex, or pre-treatment %SS. Although double-blinding was not possible, experimenter bias was limited by blinding the rater to both the subject's group and the time of the assessment. Finally, although the study lacked a control group at the 12 month assessment, the authors pointed to a lack of significant change in the control group over three months, as well as other research showing little spontaneous recovery in school-age stutters as evidence that differences in the treatment groups at 12 months were due to the interventions. Overall, Craig and Hancock (1996) conducted a well-controlled study, considered multiple variables, and a variety of outcome measures. Their study provides strong evidence for the effectiveness of intensive treatment for stuttering in school-age individuals.

Craig and Hancock (1998) conducted a follow-up to their original study (1996) to examine whether the treatment effects found at 12 months were sustained in the long-term (from two to six years). Of the original subjects, 19/25 in the EMG group, and 21/27 in the ISS group were assessed at anywhere from two to six years after the intensive therapy. Attrition was due to a refusal to participate and an inability to contact subjects. There were no significant differences between the results of the 2-3 year follow-ups and the 4-6 year follow-ups, thus the data was analyzed together. Data were analyzed with a multivariate analysis of variance (MANOVA) and subsequent ANOVAS, which was appropriate for the experimental design. The authors found no significant differences between or within groups on %SS or anxiety from the 12 month assessment to the long-term follow-up. There was a statistically significant increase in SPM in all groups, and while child and clinician ratings of naturalness were not significantly different, parents rated their children's speech as slightly less natural at the long-term follow-up.

The Craig follow-up study used the same rigorous methods to limit bias and error as the original study (Craig and Hancock, 1996). One new challenge faced by this follow-up study was that of subject attrition. Subject attrition in the treatment groups was 24% and 22% (EMG and ISS respectively), with the possibility that subjects no longer a part of the study were systematically different in some way. To address this concern, the authors offered evidence that the 12-month outcomes of the subjects who remained in the study were no different than the subjects who dropped out of the study. Overall, the study offers considerable evidence of sustained long-term improvements in stuttering behaviours of individuals who attend intensive treatment programs.

Boberg and Kully (1994) conducted a quasi-experimental within-groups study to assess long-term outcomes in adolescents (ages 11-17 years, $n=25$) [and adults (ages 18-36, $n=17$), whose results were not considered in this review] who attended the intensive treatment programs at the Institute for Stuttering Treatment and Research (ISTAR) over a two year period. The Comprehensive Stuttering Program (CSP), a smooth speech program with cognitive components, was administered seven hours/weekday for three weeks. Speech samples were collected through pre- and post-treatment telephone calls within the clinic, as well as two surprise phone calls to subject's homes during the two year follow-up period. Outcome measures included %SS and the Speech Performance Questionnaire (SPQ) to assess subjects' perceptions of their stuttering. Speech samples were presented to independent judges, who were blinded to the purpose of the study, and demonstrated an inter-rater reliability of 0.971. Descriptive statistics were used to analyze the data. The results showed a dramatic decrease in %SS of adolescents immediately post-treatment, with increases over the next 24 months. Authors noticed the increases appeared to be largely due to 6/25 adolescent subjects. When those subjects were removed and the means re-calculated, there was little evidence of relapse. Only 14 of 25 subjects returned the SPQ but results supported the %SS data. Post-hoc analyses at the follow-up assessments showed "no systematic relationship" between attendance at ISTAR refresher clinics and long-term outcomes (Boberg and Kully, 2004).

The Boberg and Kully study (1994) had a number of strengths, including excellent inter and intra-rater reliability, low attrition, blinding of raters, and long-term follow-up. Still, there are some weaknesses that limit the implications of their results. One limitation was that because no formal statistical procedures were used, it was not possible to determine which changes in the data were statistically significant. Additionally, researchers did not describe which methods they used in their post-hoc analysis of a possible relationship between attendance at refresher clinics and long-term outcomes. Another limitation, acknowledged by the authors, was that only one speaking context (telephone) was considered, thus there was no guarantee those results would generalize to other speaking situations. Finally, there was also no measure of naturalness or SPM, meaning it was not possible for the authors to draw conclusions about the quality of the subjects' post-treatment speech. Although the study does have some

limitations, it provides moderate empirical support for the use of intensive treatment with school-age stutterers.

Druce and Debney (1997) conducted a quasi-experimental within-groups study to assess short and long-term outcomes of Gradually Increasing Length and Complexity of Utterance (GILCU) treatment on 15 children (6.9-8.1 years). Treatment was in small groups, 6.5 hours/day for five consecutive days, as well as seven hours of maintenance over three months. Parents received two one-hour workshops and daily 45 minute observations of children's sessions. Researchers assessed %SS, SPM, naturalness and overall severity on seven visits (from one month pre-treatment to 18 months post-treatment). Raters were blinded to the purpose of the study and speech samples were presented in random order. Inter-rater reliability was 0.97. Differences between pre and post treatment measures were analyzed using paired t-tests with a Bonferroni correction. Results showed a significant decrease in %SS from immediate pre-treatment to one month post treatment, with small but not statistically significant increases at each of the follow-up assessments. Pearson's product-moment correlation coefficient, r , was used to examine linear relationships between variables. Severity and naturalness were highly correlated with %SS, and all three variables showed strong positive correlations with long-term outcomes. Statistical procedures used were appropriate given the experimental design. Additionally, authors identified a sub-group of five subjects with "poor outcomes", who showed significant relapse over the follow-up period.

Druce and Debney (1997) conducted a well-designed study. They considered numerous relevant variables, used appropriate statistical analyses to demonstrate significant findings, implemented blinding procedures, and measured results over a long-term period. One limitation of the study was that all measures were taken within the clinic, thus it was not clear how treatment gains would generalize to settings outside of the clinic. Overall, the study provides substantial support for the use of intensive treatment with young school-age stutterers.

Intensive Treatment Programs Using a Stuttering Modification Approach

Laiho and Klippi (2007) conducted a quasi-experimental study where 21 school aged stutterers (6.8-14 years) received non-avoidance based intensive therapy based on Van Riper's (1973, as cited in Laiho & Klippi, 2007) and Dell's methods (1979, as cited in Laiho & Klippi, 2007). Subjects were split into two groups; the "child" group (6-9 years) received 14 days of therapy (35.5 hours) and the adolescent group (10-14 years) received 18 days of therapy (52.5 hours). Parents in both groups had 7.5 hours of supervised practice with their children. Researchers analyzed pre- and post-treatment conversational speech samples for %SS, quality and length of stutters, and escape and avoidance behaviours using paired t-tests with a Bonferroni correction which was appropriate given the research design. Results showed significant within-group decreases in all outcome measures immediately post-treatment. Pearson's product-moment correlation coefficient, r , was also used to examine relationships between variables. Researchers found

significant positive correlations ($p < 0.01$) between the use of techniques to repair or prevent stutters, and %SS, muscle tension, struggle behaviour, blocks and moments of stuttering. A survey was used at post-treatment and at 9 months after treatment to assess long-term outcomes and was analyzed using the Mann-Whitney test. Survey data showed no significant differences between immediate post-treatment results and 9 month follow up. The authors inferred no significant relapse perceived by the subjects and their parents during the follow up period, on the variables addressed in the survey. Post-hoc analysis of survey results identified continued therapy and home practice during the follow-up period as positively correlated with long-term outcomes. The authors also noted greater improvements in the adolescent group, however no statistical analysis was provided.

The Laiho and Klippi (2007) study had a number of methodological weaknesses. Firstly, one of the authors analyzed both the pre- and post-treatment assessment data and there was no information given regarding any blinding procedures, leading to the possibility of experimenter bias. Other raters did analyze portions of the data to establish inter-rater reliability for each of the variables with coefficients ranging from 0.8-0.96 which were considered "acceptable" by the authors. Another potential weakness of the study was that 11/21 participants had %SS < 2% at the outset of the study, however some researchers do not even consider individuals with a %SS < 2% to be stutters (Craig 1996). The authors conceded that it may have been difficult to see a significant change in %SS in many of these subjects. However, the authors did note qualitative changes (such as a reduction in avoidance behaviours) in the stuttering behaviours of those subjects whose %SS did not decrease (Laiho and Klippi, 2007). A final concern, acknowledged by the researchers, was that no objective measures of stuttering behaviours were collected at the long-term follow up, forcing the researchers to draw conclusions using only subjective survey data. Although the study has a number of limitations, it does provide some guarded evidence for the use of intensive treatment for school-age stutters.

Nilsen and Ramberg (1999) conducted a quasi-experimental within-groups study to evaluate the effectiveness of an intensive stuttering treatment program, based on methods from Van Riper, Stromsta and Sheehan (As cited in Laiho & Klippi, 2007). Of the 15 adolescents (13.0-17.09 years) first enrolled in the study, two subjects withdrew from the program due to unrelated personal problems and were not followed. The treatment program was broken into three parts, totaling 21 days over six months. Treatment outcomes included stuttering severity (mild, moderate, or severe), stuttering/communication problems, and social skills, as rated by an independent listener, the therapists, and youth leaders involved in therapy. Therapists and youth leaders used visual analogue scales (VAS) when making their judgments. No formal statistical procedures were reported. Group means were not discussed, however individual results showed 12/13 subjects had significant improvements on at least one outcome measure and 7/13 subjects had significant improvements on at least two outcome measures. Using informal observations, authors noted improved outcomes

for older adolescence (defined as older than 15 years) (Nilsen and Ramberg, 1999). A follow up questionnaire sent to participants two weeks post-treatment was returned by 12/13 subjects. Results showed that 9/13 were satisfied with the help they received through the program and all respondents felt that the program was important or very important to them.

A major limitation of the Nilsen and Ramberg (1999) study was the methods used to assess treatment outcomes. There was no information regarding statistical procedures used in data analysis. In addition, while an independent listener was used to analyze some of the data, treating therapists also provided input, introducing the possibility of experimenter bias into the results. Informal observations of the data suggest that the youth leaders, responsible for rating social skills of the subjects, were extremely inconsistent in their ratings of subjects, however no information on inter-rater reliability was provided. In fact, the authors did not report any reliability data in the study, making their findings less useful. Another limitation of the study was that the authors did not control for subjects with concomitant speech and language difficulties, which could act as confounding variables. A final and significant limitation of this study was the lack of long-term follow-up to determine if treatment gains were maintained for any length of time after the intervention. Given the multiple limitations of the study, it provides limited evidence of the effectiveness of intensive treatment for school aged individuals who stutter.

Challenges Common to All Studies

Of the six studies included in this review, there were a number of limitations common to all of the research designs. All of the studies used convenience sampling to select their subjects, increasing the possibility of a non-representative sample. Small sample size was also a concern in the majority of the studies, with group sizes ranging from 15-27. However given the tendency of fluency therapy to result in a large effect size (e.g. Andrews, Guitar, & Howie, 1980; Craig & Hancock, 1996), the studies likely had sufficient power to detect treatment effects. Finally, since none of the quasi-experimental studies (5 of 6) included control groups, it is possible that the effects in the treatment groups were due to something other than the intervention, such as reactivity (the Hawthorne Effect), maturation, spontaneous recovery, or an uncontrolled variable (Greenhalgh, 2006). To address concerns regarding spontaneous recovery, many of the authors cited literature showing that spontaneous recovery is extremely uncommon in school-age stutters (Andrews & Harvey, 1981; Yairi & Ambrose 1992; Yairi et al, 1996).

Discussion

The studies included in this review used a variety of treatment programs based on either fluency shaping or stuttering modification approaches. The subjects all received different amounts of therapy, for different periods of time. Despite the variety of intensive interventions, all of the studies showed positive outcomes. The evidence supporting intensive fluency shaping programs is more compelling, however given the existing evidence, it is not

possible to determine if there are any significant differences in outcomes of fluency shaping and stuttering modification approaches.

A number of the studies (Boberg & Kully, 1994; Craig, 1996; Craig, 1998; Druce & Debney, 1997) identified a sub-group of individuals who demonstrated relapse in outcome measures (although rarely to pre-treatment levels). Interestingly, in three of four cases, the proportion of subjects who demonstrated significant relapse was near 25%.

All of the studies that considered long-term outcomes found that treatment results were generally maintained, with some deterioration of fluency over time. A few of the studies provided conflicting evidence as to whether continued therapy resulted in improved long-term outcomes (Boberg and Kully, 1994; Laiho and Klippi, 2007), however the evidence presented in both studies was limited, with no statistical analyses presented. Despite limited empirical evidence, authors of all of the studies pointed to the importance of maintenance programming of some kind in order to maintain fluency outcomes achieved in intensive therapy.

Only two studies in this review compared non-intensive with intensive treatment programs (Craig, 1996; Craig 1998). The authors found no statistically significant difference in short or long-term outcomes. Given that this evidence is based on only one group of subjects, it is not possible to draw definitive conclusions about any differences between intensive and non-intensive treatments.

Other factors shown to be positively correlated with long-term outcomes included pre-treatment severity and increased age. Craig (1996), Craig, (1998), Druce and Debney (1999), and Boberg and Kully (1994) all found statistically significant relationships between increased pre-treatment severity and poorer long-term outcomes. Laiho & Klippi (2007) and Nilsen & Ramberg (1997) noted improved outcomes for older adolescents, however neither study offered statistical analysis to support these assertions.

Recommendations

The following are clinical recommendations based on a critical review of the available literature regarding the use of intensive treatment for school-age stutterers:

1. There is strong evidence that fluency shaping approaches to intensive treatment for school-age stutterers result in positive short and long-term outcomes.
2. Positive outcomes were also found for stuttering modification approaches, but the research evidence was limited.
3. Intensive and traditional non-intensive treatments were equally effective in one well-controlled study. Further studies are needed to validate these findings.
4. While not addressed empirically, all of the studies concluded that maintenance planning and

programming was an important component of intensive treatment programs.

Directions for Future Research

Further research may help clinicians maximize the effectiveness of intensive treatment for school-age stutterers by examining: differences in outcomes between fluency shaping and stuttering modification approaches, qualities that make an individual a good candidate for an intensive treatment program, characteristics of treatment programs that impact its effectiveness (e.g. length, amount of instruction), and qualities of effective maintenance programs.

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