Critical Review:
What are the effects of intensive speech therapy intervention for speech outcomes in children with cleft lip and palate?

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Abstract: This critical review examines the speech outcome of children with cleft palate in relation to the intensity of speech therapy received. Overall, research suggests that intensive speech therapy may be more beneficial than conventional speech therapy at reducing articulation errors. Furthermore, there is support in the literature that intensive speech therapy may reduce the cost of treatment and decrease the total hours of speech therapy required. The findings of this review includes suggestions for further research and clinical implications relevant to practicing speech-language pathologists.

Introduction

Cleft lip and/or palate (CLP) is the most common congenital abnormality of the cranial facial complex (Watson, 2001), affecting approximately 400-500 children born in Canada each year (Pavri & Forrest, 2011). These children commonly have articulation problems, 40-50% of which require speech therapy (Enderby & Philipp, 1986; Cleft Palate Foundation, 2007). Research has shown that weekly therapy has failed to adequately improve articulation performance in some children (Albery & Enderby, 1984).

Children with articulation disorders resulting from CLP often initiate communication less frequently and fail to add or elaborate on a topic in conversation (McWilliams, Morris & Shelton, 1990). Negative communication patterns can develop as a result of deviant speech from the CLP (McWilliams et al., 1990). Effective early intervention is crucial to enhance speech development in children with CLP. A role of Speech-Language Pathologists (SLPs) is to determine which service delivery model is most appropriate and maximize the effectiveness of speech therapy.

Currently, weekly articulation therapy is the most frequent service delivery model used when treating children with CLP (Pamploma, Ysunza, Patino, Ramirez, Drucker, & Mazon, 2005). Research has shown that intensive speech therapy is a more effective treatment method than conventional weekly therapy for children with reading disabilities (Torgesen et al. 2011), autism (Sallows & Graupner, 2005), language delays (Barratt, Littlejohns, & Thompson, 1992), disfluent speech (Druce, Debney & Byrt, 1997) and other speech and language deficits (Barratt et al., 1992). Despite the results from published studies demonstrating that intensive treatment can be more effective than conventional weekly therapy when treating children with CLP, many SLPs fail to adopt this approach (Pamploma et al. 2005). The importance of adopting an intensive treatment approach for this population has many benefits, including the brevity of treatment and monetary implications. These are important factors that need to be considered when planning treatment programs.

Objectives

The primary objective of this review was to critically evaluate existing literature, regarding the effectiveness of intensive speech therapy when compared to conventional weekly speech therapy, at reducing articulation errors in children with CLP. This paper also aims to establish whether predicted improvements were maintained during follow up. The final objective is to propose evidence-based practice recommendations and clinical implications for SLPs and facilities treating children with CLP.

Methods

Search Strategy: Computerized databases including PubMed, SCOPUS, Medline and OVID were used to find articles related to the

topic of interest. The following search strategies were employed:
[(cleft lip) OR (palate) AND (intensive) OR (articulation) OR (speech therapy)]
[(cleft lip) OR (cleft palate) AND (intervention) AND (speech)]
[(cleft lip) OR (cleft palate) AND (speech camp) AND (intensive)]
The search was limited to articles in English.

Selection Criteria: Studies selected for inclusion in this critical review paper were required to investigate speech outcomes of children with CLP or velo-pharyngeal insufficiency (VPI), who participated in an intensive speech therapy program. With the exception of the presence of a CLP or VPI, as well as involvement in intensive daily speech therapy, no limitations were set on the demographics of the research participants.

Data Collection: Results of the literature search yielded four articles congruent with the aforementioned selection criteria. Two of these studies were randomized clinical trials, one employed a single subject design and the last article was a case study.

Results

The following section will describe briefly the results, and the strengths and weaknesses of four studies investigating the effectiveness of intensive treatment compared to traditional weekly treatment for articulation disorders resulting from CLP.

Randomized Clinical Trials – Between Groups

A randomized clinical trial (RCT) was conducted by Albery and Enderby (1984) to investigate whether six weeks of intensive speech therapy significantly improved articulation in children with CLP. The authors compared this improvement with a control group that received conventional weekly therapy once a week for one year Forty-six children (mean age of 8;7) were randomly divided into the experimental group or the control group. The experimental group (n=26) had three 30 minute sessions of speech therapy daily over a six week period. Two of these treatment sessions were individual and one was a group session. The control group (n=21) continued weekly therapy of two one-hour sessions. The Edinburgh Articulation Test was used to assess both groups after the investigation period. Results showed, that six weeks of intensive articulation therapy produced greater improvements than the weekly therapy. Comparing the final scores at two years post-trial of control (7.1 errors) and experimental (2.6 errors) ($p<0.05$). These improvements were maintained up to 24 months after the study was concluded.

This study used a well-defined set of inclusion criteria and ensured groups were both randomized and equally balanced, providing level one evidence. This label is given to studies that provide the "gold standard" of scientific evidence and uses rigorous methods when investigating participant outcomes. While the study reported a high level of evidence, several limitations existed. The authors highlighted the difficulties encountered when running an intensive treatment protocol in a hospital setting, including extensive planning and availability of one full time SLP. Additionally, parents may find it a disadvantage for their young children to be away from home during the week, only coming home on weekends. Despite the lack of extensive clinical testing, this study used a randomized design and valid and reliable articulation tests. Therefore the results from this study and their clinical implications can be considered suggestive.

Pamploma et al. (2005) conducted a between-group RCT to investigate whether intensive treatment was more beneficial than conventional weekly therapy at enhancing articulation in CLP children with a compensatory articulation disorder (CAD). Forty-five children with repaired CLP exhibiting CAD between the ages of 3;0 and 10;0 years old, attended a three-week speech summer camp for four hours per day, five days per week. A comparison group (n=45) received conventional speech therapy, which consisted of one-hour sessions, twice a week, for 12 months. Both programs targeted a variety of speech and language modalities including articulation, reading and writing. A Fisher exact test was used to demonstrate that there was not a significant difference ($p>0.05$) between the

distributions of the severity of CAD between the participant groups.

At the end of their respective treatment periods, both groups demonstrated a significant decrease in their CAD. The majority of participants in both groups had completely corrected or showed mild CAD. A chi-square test demonstrated that both groups of participants showed a significant ($p<0.05$) decrease in the severity of CAD, with no significant difference in the degree of severity between the groups ($p>0.10$). Although the results for both treatment groups were similar, the total hours of speech therapy received was significantly different. The traditional approach involved 104 hours of therapy per participant, whereas participants attending the speech summer camps received 60 hours of speech therapy. The intensive treatment protocol cost $100 per participant, while traditional therapy cost $412 per participant. These findings have important clinical implications that could guide cost effective and efficient speech therapy programs.

This study used a stringent set of inclusion criteria and ensured that both groups were randomized and matched on variables, such as age, gender, age of repair of the secondary palate, age of tympanostomy tubes, educational level and social-economic status. This study provides level one research evidence. This RCT was not without limitations, as highlighted by the authors, the most prominent of which was the lack of post-treatment follow up to determine the durability of the treatment effect. Additionally, there was limited analysis and statistical data provided and no inclusion of reliability measures. Furthermore, the results in the study are presented in descriptive statistics only. The authors acknowledge the reduced number of participants and the homogeneity of the groups as a limitation that does not allow for significant conclusions to be made. The research by Pamploma et al. (2005) provide suggestive evidence to support intensive speech program for children with cleft palate.

Single Subject Design N-of-1

Van Demark and Hardin (1986) conducted a single subject design (N-of-1) to determine the effectiveness of an intensive articulation therapy program for children with CLP and whether the improvement was maintained following termination of the program. Thirteen children between the ages of 6;8 – 12;0 attended a six-week summer speech program and received a follow-up assessment nine-months later. They employed a highly motivating and empirically sound articulation remediation program.

The authors commented on the difficulty controlling variables in this population, such as the age of the child at initiation of therapy. Stringent criteria for entrance into the program created a small and unrepresentative sample, which limits the power of the study to detect potential differences and makes it difficult to generalize results to clinical practice.

Despite these limitations, this study provides detailed results of improvements in the number of correct sounds and a decrease in the frequency of errors in single words and repeated sentences after six weeks intensive articulation therapy. Analysis of variance and Duncan Range Testing indicated that pre- and post-therapy word articulation test scores were significantly different ($p<0.05$). The average percentage of phonemes produced correctly increased from 66 to a mean percentage score of 82 immediately following therapy. Follow-up examination indicated that few subjects made significant progress during the nine-month period of weekly speech therapy in school and some children actually regressed. These results indicate that traditional weekly therapy failed to maintain gains made during the intensive program. This study provides level one evidence. These results and their clinical implications provide suggestive evidence for intensive programs as the most effective method of treatment for articulation errors in children with CLP.

Case Study

A case study conducted by Grunwell and Dive (1988) reviewed the effectiveness of intensive therapy for children with CLP having articulation disorders. Six children aged 4;0-10;0 participated in the two-week course. The children stayed at the hospital during the week and returned home on weekends. The emphasis of the treatment protocol was articulatory placement and production. Each child spent a total of two hours a day in individual therapy and three hours in group therapy. The aim of these sessions was to increase the children's awareness of natural sound classes and phonemic contrasts. Two children were selected and their results were described in detail to support the program.

Despite some persistent articulatory errors, intensive speech therapy targeting both articulation and phonological systems showed improvements. Additionally, findings show that intensive programs can facilitate reorganization and expansion of previously static speech errors that appeared resistant to traditional once weekly therapy.

This study by Grunwell and Dive (1988) present level four evidence. Case studies are useful to describe novel treatment environments or analyze characteristics of a single individual. They cannot be used to draw cause and effect conclusions, and their results are not readily generalized. The children in this study were referred with persistent speech problems, resistant to traditional therapy and were not typical of the CLP population. An additional drawback was the lack of statistical analysis provided by the authors. All data was presented as descriptive statistics with no provision of means, standard deviation or effect size. Due to poor statistical analyses and lack of controlled variables within the study, the validity and reliability of these results should be questioned. The results from this study and their clinical implications can be considered equivocal.

Discussion

A review of the four studies above indicates that intensive therapy improves speech outcomes in children with CLP. The number of treatment hours of speech therapy required was significantly different between the treatment modalities, with intensive therapy requiring approximately half as many hours as traditional weekly therapy to achieve similar results. Additionally, there was a significant decrease in the costs associated with intensive therapy when compared to traditional weekly therapy. Intensive treatment reduces the financial burden placed on families and increases available speech services by enhancing treatment efficacy, making it a more attractive service delivery model.

While all studies illustrated a reduction in articulation errors, not all studies found a significant difference between intensive and weekly speech therapy. This may be accounted for by some of the limitations in these studies. These include; (1) failing to address the potential impact of co-occurring syndromes; (2) lack of detailed statistical analysis; (3) failing to account for normal developmental errors during assessments; and (4) small sample size.

It is important to consider the high rate of co-occurring deficits (e.g. micrognathia) (Solot, Knightly, Handler, Gerdes, Mcdonald-Mcginn, Moss, Wang, Cohen, Randall, Larossa, Driscol, Emanuel, & Zackai, 2000) in some children where CLP occurs secondary to an overarching syndrome (e.g. Pierre Robin Sequence) (Caouette-Laberge, Bayet, & Laroque, 1994). In this case, the syndrome may present concomitant anomalies or difficulties that impact speech outcomes, making it difficult to make appropriate clinical decisions. Albery and Enderby (1984) and Pamploma et al (2005), used a well-defined set of inclusion criteria, including age, gender, education level and timing of repair of the secondary palate. They ensured both groups were randomized and equally balanced. In contrast, Van Demark and Hardin (1986) did not specify any exclusion criterion, which reduces the possibility for

accurate comparison to other populations. Additionally, some studies failed to report any statistical findings. Grunwell and Dive (1988) and Pamploma et al (2005) reported their findings in descriptive statistics. This makes it difficult to make accurate predictions based on their results.

The average age of the participant when conducting follow-up assessments for each of the studies varied from preschool to school aged children. Pamploma et al. (2005) reported follow-up assessments on children as young as three years of age. Speech assessments of young children can be difficult interpret and make clear predictions about potential long-term speech deficits (Lewis, Freebairn, & Taylor, 2000). Additionally, articulation errors at an early age may be attributed to developmental errors that may self correct as the child develops. Albery and Enderby (1984) followed children for two years post-treatment when participants were a mean age of 10;7. Assessing older children may reduce the impact of typical developmental speech errors, as speech characteristics at that age are more enduring and may be more predictive of long term articulation difficulties (Lewis et al, 2000).

Studies included in this critical review varied in their sample sizes, with some failing to include an adequate number of participants to make accurate predictions about treatment effects. Grunwell and Dive (1988) included six children in their study. The low number of participants and uneven group distribution limits the generalizability of the researchers' findings and reduces the probability of detecting existing differences. The small sample size in this study may provide an explanation as to why the researchers failed to detect a significant difference between the intensive speech therapy program and the traditional once weekly approach.

It is recommended that further research on this topic be completed to examine the speech outcome of children with CLP in relation to the intensity of speech therapy received. In order to improve upon the evidence provided by the existing literature, it is recommended that future studies address the limitations discussed above. An important criterion not incorporated into many of the studies was inclusion of a detailed description of the treatment programs. Failing to report this information makes it difficult accurately replicate the study. A well-designed study should allow for an independent researcher to replicate the experiment, under the same conditions, and achieve the same results. Without this information one may question the reliability and validity of the findings.

Although there is clearly a need for additional research in this area, the studies included in this review provide suggestive evidence for improved articulation, decreased service costs, reduced total therapy hours, and long-term maintenance in children with CLP enrolled in intensive therapy.

Clinical Implications

The following clinical recommendations are based on the critical review of the available literature regarding the use of intensive treatment for children with CLP:

a. There is suggestive evidence indicating intensive speech therapy results in a significant decrease in articulation errors in children with CLP.
b. Although intensive speech therapy increases the amount of hours children spend in treatment over a short period of time, research shows that intensive therapy may lead to a decrease in the total number of hours required to achieve similar speech outcomes.
c. Evidence suggests that traditional weekly speech therapy may be twice as expensive to implement as intensive speech therapy.
d. Intensive programs have been shown to facilitate improvements of previously static articulatory errors that appeared resistant to traditional once weekly therapy.

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