

## **Critical Review:**

### **In children with cerebral palsy, what treatment methods are effective in improving speech production?**

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This critical review examined the evidence regarding effective treatment methods for improving speech production in children with cerebral palsy. Study designs include single subject “n of 1” design (3), single group pre-post test (2) and a case study. Overall, the evidence summarized in this review is considered inconclusive and it is recommended that additional research be completed.

#### ***Introduction***

Cerebral palsy (CP) affects approximately 1-2/1000 births each year, making it one of the most common motor disorders amongst children (Paneth & Hong, 2006). Cerebral palsy has recently been defined as an umbrella term that describes a movement or posture disorder that is caused by a non-progressive interference, lesion, or abnormality in the immature brain. It is a permanent, yet unchanging disorder that persists throughout one’s life. (Rosenbaum, Paneth, Leviton, Goldstein & Bax, 2007) Because the underlying cause of CP is not curable, children must cope with the disorder for the remainder of their life. Some characteristics of CP may include difficulties in speech and language, and/or communication (Pennington, Goldbart, & Marshall, 2005).

Speech problems associated with cerebral palsy may include articulation errors, oral motor difficulties, rate or prosody errors, and dysarthria. Dysarthria is defined as “speech disorders resulting from disturbances in musculature control over the speech mechanism due to damage of the central or peripheral nervous system. It designates problems in oral communication due to paralysis, weakness, or in coordination of the speech musculature” (Duffy, 2005). Dysarthria can greatly reduce the intelligibility of one’s speech, and make it difficult for those with dysarthria to communicate with others. A study on the characteristics of speech in children with cerebral palsy by Kiran, Üstüner Atik, Dursun, & Topbaş (2004) reported that 60.4% of children with cerebral palsy had disturbances in intelligibility by others, and 37.5% of children had decreased intelligibility by family members. Additionally, Kiran et al. (2004) reported errors contributing to decreased intelligibility: 66.7% of children had a disturbance of oral motor functions and phonology, 50% had difficulty with articulation, 47.9% had prosody disturbances, and 33.3% had respiration disturbances. These impairments may be of great difficulty for children with cerebral palsy as many children prefer to use natural speech when possible over augmentative and alternative communication devices (AAC).

Children with cerebral palsy may also experience negative impacts on their educational, social, and familial relationships due to their decreased intelligibility of speech. A study by Lass, Rucello, and Lakawicz (1988) investigated listener perceptions of children with articulation disorders and dysarthria secondary to either cerebral palsy or other developmental disorders. Results indicated that normal speakers were rated more favourably than dysarthric speakers on nearly all aspects of speaking tasks.

Traditionally, speech-language pathologists have focused on helping children with cerebral palsy to maximize their communication skills. Therapy concentrating on speech production and improved intelligibility may address laryngeal, velopharyngeal, respiratory and articulatory incompetencies. Despite widely accepted dysarthria therapy techniques, very little evidence has been put forth to prove efficacy of treatment methods for this population (Pennington, et al. 2005). This review will critically examine multiple treatment methods for improving speech production in children with cerebral palsy. For the purposes of this paper improvement in speech production will include any method that proposes to improve speech intelligibility or articulation of specific phonemes.

#### ***Objectives***

The primary objective of this paper is to critically review treatment methods that improve speech production in children with cerebral palsy and examine their effectiveness. Recommendations for future research in this area, as well as clinical implications will also be discussed.

#### ***Methods***

##### **Search Strategy**

Online databases including Scopus, Pubmed, and Embase were searched using the following search criteria:

“Cerebral palsy” AND “child\*” OR “young adult” AND “articulation”

“Cerebral palsy” AND “child\*” OR “young adult”  
AND “dysarthria”  
“Cerebral palsy” AND “child\*” OR “young adult”  
AND “intelligibility”

Reference lists from applicable articles were also used to identify further articles. The search was limited to articles in English.

#### Selection Criteria

Articles were selected for review if they focused on improvements in intelligibility or articulation in children with cerebral palsy. Treatment methods had to target speech or articulation directly, and could not use any AAC methods concurrently. Only those articles pertaining to treatment methods within the scope of practice of a Speech Language Pathologist were chosen for review.

#### Data Collection

Following exclusion of articles that did not meet selection criteria limits, results of the search yielded the following types of articles: single subject “n of 1” design (3), single group pre-post test (2) and case study (1).

### **Results**

#### Single subject designs

A study using single subject design is an appropriate method to examine a new hypothesis about the management of a disorder. This is a strong design for preliminary research in the area of treatment methods for improving speech production in children with cerebral palsy. With this design the patient acts as their own control and results are interpreted based on temporal measures. Although results may indicate an active intervention they must be interpreted with caution as causality may not be directly due to the intervention alone.

Pennington, Smallman, and Farrier (2006) used a single subject design to investigate the effects of intensive dysarthria therapy in 6 students between the ages of 10 and 18 with cerebral palsy. Participants took part in individual therapy 5 times a week for 5 weeks, in which treatment focused on a systems approach. Therapy included activities used to control breathing, to regulate intensity and mark stress. Each student was able to progress through therapy at their own rate of progress, thus allowing a more tailored individual approach. Following completion of therapy, the student’s speech intelligibility was measured using the Children’s Speech Intelligibility Measure as well as through a story formulation task. This data was compared to

original baseline data collected using the same measures. Results indicate that 4 students improved intelligibility scores at the single word level following completion of therapy, although results were not statistically significant. Additionally, 2 of the 6 students had improved intelligibility scores in connected speech, but again, the results were not statistically significant. Despite improvements immediately following the completion of the intervention, all but one of the participant’s intelligibility scores showed a regression at 7 weeks following the termination of therapy.

Various participant and methodological strengths were noted in this study. First, the participants were of an older age, thus reducing maturation effects on the results. Secondly, each participant was well described and extensive information about their speech disorders was available to the reader. This is important to understand individual results in relation to severity or type of the presenting speech disorder. Furthermore, the researchers ensured that therapists providing treatment and assessment were randomized, thus decreasing experimenter bias.

Despite the strengths of this study, some apparent weaknesses were noted within the methods and results. One being that the actual method of treatment was not well described. This makes it difficult to reproduce results in further studies, and thus strengthen the results this study. Another methodological weakness is that the procedures within the therapy were very contrived, having students say single words in a clinical setting. This is not reflective of realistic speaking contexts and thus, the applicability of the results must be considered. A weakness noted within the methodology of the study was that the statistical method of calculating significant differences and interpreting results was not indicated. This is a limitation to the results as it is not possible to know how the researchers came to their conclusions. Additionally, it would have been beneficial for the results to have been grouped by the participant’s type or severity of cerebral palsy. By also analyzing trends within these subgroups, the results would have been strengthened and thus more complete. Finally, with such a small group of participants, no control group, and no multiple baselines, it is very difficult to confirm if changes in intelligibility were due to the intervention.

Overall, this study piloted research for treatment using a systems approach to improve speech production in children with cerebral palsy. Although the results were not statistically significant this study gives insight into an individual treatment method that may be successful in improving speech production. Given the strengths

and weaknesses of this study, the level of evidence for this study is suggestive.

Another single subject, multiple baseline study was performed by Marchant, McAuliffe, and Huckabee (2007). This article examined the effectiveness of two treatment approaches, phonetic placement therapy and sEMG biofeedback therapy, to improve intelligibility in a 13 year old female with spastic hemiplegic cerebral palsy. The subject received 6 weeks of therapy in which there was a 2 week period of no treatment in between the 2 intervention approaches. Identical speech recordings were taken for perceptual and acoustical analysis and physiologic results from sEMG measures were completed prior to and after each intervention. Findings concluded that there was no significant differences in sEMG amplitudes however improved stability in amplitude measures was noted post sEMG treatment. Intelligibility was significantly improved at the single word level following PPT therapy and this was maintained following sEMG treatment. A perceptual rating did not indicate any change to articulatory parameters or overall intelligibility following completion of both components. Furthermore, there was no reported difference in the participant's self perception of speech.

This article presents strengths in terms of its participant, methodology, and results. The participant was of an older age which reduces maturation effects on the results of the study. The participant's speech was well described within the article and she underwent in depth sensory and cognitive testing prior to intervention. Using an ABACA method was an effective way to differentiate results from the two treatment methods and thus eliminating learning effects or confounding results. Both the treatment methods and assessment methods were extremely well documented and clear to the reader, as well, they were both justified by previous scientific evidence. Furthermore, appropriate reliability measures were reported to eliminate possible tester effects or biases. The results of the study were described both qualitatively and quantitatively, which provided information about the changes and improvements in the subject. Statistical measures used to infer results were appropriate based on the type of type of study and measurement results. The participant also completed a survey regarding perceptions of her speech throughout study. This adds another dimension within the results and contributes to this study's strengths.

Regardless of the numerous strengths, the study by Marchant, McAuliffe, and Huckabee (2007) has one weakness to be considered. The study has a very low 'n' value and thus external validity is queried.

Overall this study boasts various strengths in terms of its design, methodology, and both quantitative and qualitative results, despite the low sample size. This study is judged to provide compelling evidence to prove that phonetic placement therapy is effective in improving intelligibility at the word level.

Wu, and Jeng (2004) completed a single subject design that examined the effects of two articulatory intervention approaches to improve speech production in children with cerebral palsy. The study included 2 Mandarin speaking participants of similar age and severity. Each participant was randomized into one of 2 treatment groups: a phonetically based approach or a phonological based approach. Both children participated in 4 months of therapy with two 30 minute sessions each week, targeting one phoneme per month. Therapy targeted 4 phonemes that the participants could not produce at the beginning of the study. Pre and post test measures of articulation were completed. Results indicate that the phonetically based program allowed the participant to make greater gains than the subject in the phonological based intervention. Additional measures indicated that both participants showed a regression on outcome measures at 2 months following treatment.

The participant selection and methodology of this single subject design has a number of strengths. The first being that the participants included in the study were matched and randomized into their treatment group. This allows results to be contrasted and reduces group allocation bias. Furthermore, extensive qualitative information about each intervention method was included within the article. This makes the study easily replicated and interpreted for future research.

Although the above strengths are noted, the article by Wu and Jeng (2004) contains weaknesses pertaining to the results. First, the results were not examined statistically. This is a drawback to the study as the treatments may not have caused any significant changes in the individual's speech production, and thus the results are less conclusive. Also, the study only examined articulation at the single syllable level. This fairly narrow level of speech is judged to have limited ecological validity and may not generalize to higher levels of speech and communication.

Overall, this paper provides suggestive evidence that phonetic placement therapy can improve speech production in children with cerebral palsy.

Single group pre-post test designs

The single group pre-post test design is an appropriate method to investigate effects of a treatment in improving speech in children with cerebral palsy. It is advantageous because of the increased sample size which improves the validity of the results. However, results should be inferred with caution as there may be variability between participants particularly in a heterogeneous group such as cerebral palsy.

A single group study involving a pre-post test design was conducted by Puyuelo and Rondal (2005). This study investigated two treatment approaches to improve speech production in 10 Spanish-speaking children with CP over a four year period. The participants were all between the ages of 3 and 3.5 at the beginning of the study and they had differing severities of cerebral palsy. Each subject participated in 2 years of therapy targeting oral praxis, but due to limited success, the intervention method changed to include 2 more years of therapy centered on phonation, prosody, and sound duration. The second treatment was provided in parallel with communication training for parents and teachers, as well as neurodevelopmental treatment to improve sensorimotor functioning. Prior to intervention and following both types of intervention a dysarthria specific questionnaire was administered in order to track changes in intelligibility. Following the completion of the entire study, each participant had their speech examined through spectrographic analysis. Data was analyzed statistically using the Friedman test and the Wilcoxon test. Results indicate that after the first intervention targeting oral praxis, the only significant difference between baseline and intervention measures was an improvement in voice control. Following the second intervention significant differences were noted on all aspects of dysarthric characteristics (oral, respiration, voice, articulation, intelligibility, and prosody) except for oral praxis.

This single group study contains various strengths in the participant selection, methodology and results. The participants in the study were well described in terms of type of cerebral palsy and their language skills prior to intervention. The article also includes a thorough explanation of procedures used to improve speech production. As well, qualitative data was provided to describe persisting speech errors for each of the subjects in the study. By including this information the results are strengthened, as individual participant differences are accounted for within the heterogeneous group. Additionally, the appropriate statistical measures were used to yield their results which were extensively described within the article.

However, several weaknesses were noted in terms of the participants in the article by Puyuelo and Rondal

(2005). One limitation is that the severity of each child's cerebral palsy was not noted within the article. Similarly, articulation errors and characteristics of the participant's speech were only labeled as "unintelligible" and their speech was not qualitatively described. Not knowing the severity of the speech disorder at the beginning of the treatment makes it difficult to interpret results accordingly and with confidence. In addition, the participants were only of an age of 3.5 at the beginning of the study and a control group was not included. These last two concerns make it difficult to distinguish changes due to intervention or from changes that may be due to maturation effects.

Further weaknesses are present within the methodological design and results interpretation. One weakness is that within the second intervention the participants were involved with a "neurodevelopmental treatment". This treatment was not defined within the article and may have been an additional source of improvement for the children in the study. Without knowing the exact procedures of a parallel treatment or attempting to control for parallel effects, it is difficult to state that improvements in speech production were caused by the intended intervention. Another area of weakness is that the speech language pathologist providing treatment is the same person to provide all assessments. This may have introduced several sources of experimenter bias into the study. For example, the experimenter's familiarity with the children's speech over the four year period of the study may have had an important impact on the consistency and reliability of intelligibility ratings. Finally, the article lacks a description of the parameters of the improved areas, as well as any information about results found in spectrographic analysis for the group of children, thus making the results incomplete.

Overall this paper provides equivocal evidence that the treatment methods improve speech production in children with cerebral palsy. A possible maturation effect, in combination with weaknesses in participant and methodological description makes the results difficult to interpret with confidence.

A second article by Ray (2001) used a pre-post test design to examine the functional outcomes of children with cerebral palsy who received orofacial myofunctional therapy. The experiment included 16 children between the ages of 7 and 10 with mild to moderate spasticity due to cerebral palsy. All participants had been learning English as a second language since the age of 3 years. Subjects underwent orofacial examination and speech testing pre and post testing. This included measures of diadochokinetic rates, examination of speech anatomy, examination of

the function of the tongue, lips and jaw, intelligibility testing, and examination of the types of articulatory errors. Subjects completed 4 months of therapy in English, with 25 minute sessions 5 days of the week. These sessions included 15 minutes of individualized orofacial myofunctional treatment, and 10 minutes of group treatment. Exercises included those to improve resting postures of the articulators and strengthening of the tongue, lips and jaw. No traditional articulation therapy was provided. Participants were also provided with home based exercise programming. Results were evaluated using the Wilcoxon Signed rank test, Spearman's correlation coefficient, and t-tests. The results indicate that 15/16 subjects showed functional improvement in lip and tongue postures. Additionally, overall improvement in the functioning of the tongue, lips and jaw was noted and intelligibility scores at the word level improved significantly without direct intervention. The correlation between lip/jaw functioning and intelligibility scores was not significant however a significant correlation between the tongue function and intelligibility scores was noted.

Various strengths were noted in the article by Ray (2001) in terms of its participants, methods and results. This is the only study within this critical review to include more than 10 participants. This gives the study relatively stronger validity and predictive value and suggests a stronger level of evidence for the reported intervention. The methods were well described and specific exercises and procedures were outlined in an appendix which allows for replication of the study. The results were analyzed using appropriate statistical procedures and correlation measures were also utilized to provide further information about the results. Additionally, the author reports on interjudge reliability scores, helping to account for experimenter differences.

In contrast to the above strengths, some limitations in the article by Ray (2001) were apparent within the methods of the study. The intervention method targeted within the study employed the use of home programming in English, however it is not reported if the parents are competent in this language. This may have been an extraneous variable contributing to results of the study. In addition, all of the participants had been attending a school for students with cerebral palsy and it was reported that within this setting the students may have had additional instruction regarding speech production. This may have been a confounding variable within the study, thus making results open to interpretation. Finally, intelligibility ratings were measured using a speech sample of 20 one and two syllable words. This level of speech is judged to have limited ecological validity and may not represent improvement in speech at higher level.

The overall evidence for using orofacial myofunctional therapy as an intervention method for children with cerebral palsy is suggestive. Although the study has relatively strong power and reliability, more intensive intelligibility testing and a control group for comparison would improve evidence to compelling.

#### Case study

A final study by Gibbon and Wood (2003), examined the effects of using electropalatography (EPG) to treat an articulation disorder in an 8 year old boy with mild spastic cerebral palsy. The boy participated in 15 clinical based sessions lasting 45 minutes over 4 months to improve articulation of velar sounds. During sessions the participant used real-time visual feedback of tongue-palate contact patterns to establish velar placement for /k/ and /g/. He also used a portable training unit at home after the 5<sup>th</sup> session. Comprehensive EPG measures analyzing centre of gravity (COG) and duration of stop closures were taken pre and post treatment and were changes in data were analyzed using t-tests. Results showed that following therapy there was a significant difference in the COG values when the participant was producing a /k/, and that he was not longer using velar fronting.

This case study demonstrates several strengths. The first being that the methodology of the study was well documented and were based upon a previous study targeting articulation improvements. Strengths were also noted within the results of this study. Appropriate statistical analyses were used to analyze the data collected, and clear graphs of EPG articulation measures pre and post therapy helped to describe the data more qualitatively. Also, results were measured using computerized results regarding points of articulation and force applied by the tongue, thereby reducing experimenter error.

The Gibbon and Wood (2003) article also demonstrates some limitations. There was only one participant in the study, which makes the evidence for EPG therapy to treat articulation disorders in children with cerebral palsy less compelling. Furthermore, EPG equipment and expertise is not widely available to the practicing SLP clinician, thus reducing this article's importance to evidence based practice literature. Finally, the participant may have learned articulation techniques from prior therapy with the same clinician that carried over to results within the study.

Overall, evidence for EPG therapy to improve speech production in children with cerebral palsy is suggestive.

### ***Recommendations***

This literature review examined several different treatment approaches to improve speech in children with cerebral palsy. Based on the literature, it is concluded that there is limited evidence to endorse any type of treatment approach to improve speech in this population of children. There is, however, a trend within this subset of literature to support the use of a phonetic based approach to improve speech production. Studies by Marchant, McAuliffe, and Huckabee (2007) as well as Wu, and Jeng (2004) yielded results indicating that this method of intervention improved speech in children with cerebral palsy. Although these studies generated similar results, they were both judged to have weaknesses regarding external validity, ecological validity and completeness within their results both quantitatively and qualitatively. Further studies involving larger samples and stronger study designs are required to strengthen the evidence for using a phonetic based approach to improve speech in children with cerebral palsy.

The studies examined within this literature review are preliminary studies piloting research in new treatment approaches to improve speech in children with cerebral palsy. The importance of these studies to this area of research is compelling, as they are the first of their kind to study each intervention method. However, of these studies there were many areas of weakness in terms of participant selection, methodology, and results. Reliability and validity within each study were judged to be weak, and thus the results should be interpreted with caution.

With regard to future research in the area of treatment methods to improve speech in children with cerebral palsy, it is recommended that the following be considered:

- 1) Participants should be grouped by both type and severity of cerebral palsy in order to identify treatment methods and trends pertinent to each group.
- 2) Control for maturation effects by including older children and also by incorporating untreated control groups.
- 3) Future studies should consider the use of conversational speech tasks in the assessment and treatment procedures in order to increase the ecological validity of the results.

- 4) Larger sample sizes and better experimental designs are required to improve the strength of the evidence in this area of clinical research.

### ***Clinical Implications***

This review does not provide compelling evidence for the use of one specific method for the treatment of speech in children with cerebral palsy. At this time, the evidence is judged to be fairly weak and does not provide support for the use of any of the above methods in the treatment of speech in individuals with cerebral palsy.

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