Critical Review:

In children with cerebral palsy and a diagnosis of dysarthria, what is the effectiveness of speech interventions on improving speech intelligibility?

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This critical review examines literature on the impact of speech interventions on increasing speech intelligibility in children with cerebral palsy and dysarthria. Five experimental studies, including single-subject designs and single-group studies, were evaluated in this review. Overall, majority of the literature demonstrated statistically significant increases in intelligibility, providing suggestive evidence to support the positive impact of speech interventions for children with cerebral palsy and dysarthria. Therefore, implementing speech interventions to this population may be a viable option for therapy given the specific needs and abilities of the child. However, limitations observed in these studies suggest a need for further research in this area.

Introduction

Cerebral palsy (CP) is one of the three most common developmental disabilities, present in over 2 in 1000 live births worldwide (Sankar & Mandkur, 2007). These individuals have a range of deficits in gross and fine motor function and in cognitive abilities, which contribute to communication impairments in about 60% of this population (Hustad, Gorton, & Lee, 2010). The communication problems have commonly been addressed with speech and language therapy focusing on expressive communication (i.e. use of gestures, facial expressions, and augmentative and alternative devices) (Pennington, Smallman, & Farrier, 2006).

As a result of low oral-motor skills, about 35% of children with CP are also diagnosed with dysarthria, a speech disorder involving abnormal strength, speed, and accuracy of movement of the speech system (Hustad et al., 2010; Pennington et al., 2013). The oro-motor deficits associated with dysarthria impede intelligibility, reducing the individual's ability to effectively communicate using spoken language (Pennington, Miller, Robson, & Steen, 2009). Therefore, therapy focusing on improving speech production is being provided to children with CP and dysarthria who have adequate language skills to use speech as a means of communication (Pennington et al., 2006). Various interventions have been implemented in order to improve perceptual intelligibility. Among these include relaxation of the speech musculature, phonetic placement therapy, improving breath support, coordinating phonation and breathing, improving speech prosody, and slowing speech rate (Marchant et al., 2008; Pennington et al., 2009). A majority of therapies have been found to significantly improve intelligibility. while others have not been as successful. The efficacy of speech interventions as a means to improve speech intelligibility will be reviewed in this paper.

Objectives

The objective of this paper is to provide a critical review of the current literature on the effectiveness of speech interventions on improving perceptual intelligibility for children with cerebral palsy who have been diagnosed with dysarthria.

Methods

Search Strategy

The computerized databases, including PubMed and Google Scholar, were used to search for relevant articles.

The keywords incorporated in searching the databases included:

[(("cerebral palsy/rehabilitation") AND "children") AND "speech"]

[(("Speech Disorders/therapy") AND cerebral palsy) AND "children"]

Selection Criteria

The papers included in this review were required to provide outcome measures on intelligibility and to target a population of children (0-18 years) with a diagnosis of cerebral palsy and dysarthria. All severities and types of CP and dysarthria were included in the search.

Data Collection

The literature search revealed experimental articles that included two single-subject designs and three singlegroup pre-posttest studies.

Results

Single-Subject Designs

Marchant, McAuliffe, and Huckabee (2008) used a single-subject design to assess the effectiveness of phonetic placement therapy (PPT) and sEMG-facilitated biofeedback relaxation treatment on improving intelligibility in single words and continuous speech in a child (13 yrs) with spastic hemiplegic cerebral palsy and spastic dysarthria. The child received two weeks of PPT, two weeks of treatment withdrawal, then two weeks of relaxation therapy. PPT targeted individual speech sounds and sEMG relaxation therapy targeted relaxation of facial muscles. Speech samples were collected and analyzed pretreatment (3 times) and immediately following each treatment type using standardized tests and reading passages commonly employed in this research area. Visual inspection of percent intelligibility measures in single words revealed an increase in intelligibility immediately following PPT as compared to baseline, and immediately following sEMG as compared to baseline. There was no change in intelligibility post-sEMG when compared to post-PPT.

This paper provided a clear description of the execution of the study, including the assessment protocol and the intervention procedure, allowing for accurate study replication. Blinding was achieved by randomly presenting the child's speech samples to two Speech-Language Pathologists (SLPs) who were not involved in conducting the treatment. Acceptable levels of interrater reliability were reported. One limitation of the present study was that no measurement was taken right before the sEMG relaxation therapy. As a result, it was unclear whether the maintenance of the increased intelligibility after the relaxation therapy was due to the long-term effects of PPT, or if it was due to the positive effects of sEMG treatment. Another possible limitation was the use of only raters familiar with dysarthric speech, so it is unclear whether intelligibility would have increased for unfamiliar listeners. Finally, the absence of statistical analysis limits the validity of the findings because no quantitative measures of improvement could be reported regarding gains in intelligibility.

This single-subject design offers suggestive evidence to support the efficacy of PPT intervention and mildly suggestive evidence for sEMG relaxation therapy in a child with cerebral palsy and dysarthria.

Pennington, Smallman, and Farrier (2006) examined the effects of a five-week speech intervention targeting breath support and prosody on improving intelligibility in single words and connected speech using a single-subject pre-posttest design with six children (aged 10-18) with mild-severe dysarthria and cerebral palsy. Intervention moved sequentially, first targeting breath support, and then moving to speech prosody once the use of breath support was achieved. Standardized tests

that are commonly used to assess intelligibility in single words and connected speech were administered one week prior to intervention, one week post-intervention, and seven weeks post-intervention. Four of the children were observed to have increases in intelligibility in single-words immediately following intervention, however, appropriate use of Wilcoxon signed ranks test revealed no statistically significant changes in any speech intelligibility measures after intervention compared to baseline measures.

A strength of this study was the detailed description of each child's skills pre-intervention, including cognitive, language, and motor speech abilities. Also, listeners transcribed unidentified speech samples in a random order, preventing listener familiarly and inflation of intelligibility scores. A drawback to the study was that the actual statistical values were not provided. As well, no inter-rater reliability was reported. Without this data, it is unclear whether the intelligibility ratings were consistent between raters, thus possibly skewing the data. Furthermore, the description of the intervention was not sufficiently expressed, and therefore would impede accurate replication of the study.

Overall, this study provides mildly suggestive evidence that intensive speech therapy focusing on breath support and prosody effectively improves intelligibility in single-words in children with cerebral palsy and dysarthria.

Single-Group Pre-Posttest Studies

In a single-group pre-posttest study, Pennington, Miller, Robson, and Steen (2009) evaluated the efficacy of a "systems approach" on improving speech intelligibility in single words and connected speech in 16 children (aged 11-19) with a diagnosis of cerebral palsy and moderate-severe dysarthria. Intervention concurrently focused on increasing respiratory and phonatory control, slowing speech rate, and maintaining breath support during speech. Measures commonly used to assess intelligibility in single words and connected speech were administered pre- and post-intervention. The appropriate use of ANOVA and multilevel modeling revealed statistically significant increases in single-word intelligibility measures for familiar and unfamiliar listeners and increases in connected speech intelligibility for familiar listeners.

A strength of this article was its well-described intervention protocol, which included the hierarchy exercises and the criterion required to proceed to the next step of the hierarchy. Also, listener bias during analysis was limited through blinding and randomization of speech samples. The unclear documentation of the study's clinical findings and significant results was a major drawback to this article and limits the article's clinical value. Also, the increases in intelligibility were found by comparing the average of the two pre-intervention assessments (1 and 6 weeks prior) and the average of the two post-intervention assessments (1 and 6 weeks post). Therefore, it cannot be determined whether the increased intelligibility was only observed immediately following intervention, or if it was also observed 6-weeks later. As well, there were low inter-rater reliability scores between familiar listeners (0.53 and 0.31), possibly contributing to skewed results.

This study provides suggestive clinical evidence to support the efficacy of the "systems approach" for improving intelligibility in children with cerebral palsy and dysarthria.

Using a single-group pre-posttest study design, Pennington et al. (2013) assessed the effectiveness of speech therapy targeting breath support, phonation, and speech rate on improving speech intelligibility in single words and continuous speech in 15 children (aged 5-11) with a diagnosis of cerebral palsy and moderate-severe dysarthria. The intervention simultaneously targeted respiratory and phonatory control, slowing speech rate, and maintaining breath support during speech. Standardized assessments commonly used to measure speech intelligibility and communication/participation changes were administered pre- and post-intervention. ANOVA revealed statistically significant increases in intelligibility for single-words and connected speech post-intervention for familiar and unfamiliar listeners. No significant improvements in intelligibility were found at the 12-week post-intervention assessment.

Strength of this study was in the randomization of speech samples and blinding of listeners, limiting listener bias. Another strength was the clear description of the participants' motor function, dysarthria type, and language abilities. A possible drawback of this study is in the variability of language skills exhibited by the participants, potentially causing differences in the length of collected speech samples between individuals. This variability was not addressed or controlled for during the analysis of the speech samples, possibly resulting in inconsistent ratings of intelligibility. Another weakness was in the way the long-term (12week post-intervention) measure of intelligibility was compared and analyzed to the previously collected data. Rather than comparing baseline measures to the 12week post-intervention measures, the baseline measures and the measures collected at 1 and 6 weeks posttreatment were averaged and compared to the 12-week post-intervention measures. This may have contributed to the lack of statistically significant increase in intelligibility calculated at the 12-week assessment.

Other weaknesses include low levels of inter-rater reliability across the familiar listeners and limited description of the intervention protocols.

This study provides suggestive evidence to support the efficacy of therapy targeting breath support, phonation, and speech rate in increasing intelligibility in children with cerebral palsy and dysarthria.

Puyuelo and Rondal (2005) used a longitudinal singlegroup pre-posttest study design to assess the effectiveness of two speech interventions on improving intelligibility, and other speech parameters beyond the scope of this review, in ten children (3 yrs) with a diagnosis of cerebral palsy and dysarthria. The first twoyear intervention focused on the mobility and sensitivity of articulators, food mastication, and expiratory control, and the second focused on the co-ordination of speech breathing and improving speech prosody, with one month of treatment withdrawal between interventions. Neurodevelopmental treatment was provided to these children throughout the four years of the study. Appropriate use of ANOVA and Wilcoxon Matched-Pair Test revealed significant increases in speech intelligibility on the Robertson's Dysarthria Profile (translated into Spanish) following the second intervention compared to baseline measures and compared to measures gathered after the first intervention. No significant increases were revealed after the first intervention compared to baseline.

A strength of this study was in the administration of preintervention language assessments to ensure all participants had similar expressive language skills. A major drawback of this study was in the possible biasing of the assessment measures due to the lack of blinding and randomization of the speech samples and the subjectivity of the assessment tool. Furthermore, the author of the paper was the only one administering the assessments for each child, further contributing to rater bias. Because this study took place over four years, maturation of the children could have played a role in the improvement of their intelligibility. Therefore, a lack of control group was another drawback. Description of the children's severity and type of dysarthria was not reported, preventing clinicians from determining the subpopulation who may benefit from the intervention. Finally, it is difficult to determine if the significant increase in intelligibility was a result of the speech intervention or a result of the neurodevelopmental intervention because both were being administered concurrently throughout the study. Overall, this study provides mildly suggestive evidence supporting the effectiveness of speech intervention focusing on speech breathing and prosody in increasing intelligibility in young children with cerebral palsy and

dysarthria.

Discussion

Review of the literature suggests that speech intervention can be effective in improving speech intelligibility in children with cerebral palsy and dysarthria.

While most of these studies exhibited statistically significant changes in intelligibility following intervention, the functionality of these observed improvements should be considered, as the articles did not address the clinical significance of the improvements (Marchant et al., 2008; Pennington et al., 2009; Pennington et al., 2013; Puyuelo & Rondal, 2005). Therefore, it is unclear whether there was enough improvement in intelligibility to impact the children's ability to communicate with spoken language. Also, most of the increases in intelligibility were noted to be at the single-word level, while outcome measures in connected speech did not show significant improvement as often (Marchant et al., 2008; Pennington et al., 2009; Pennington et al., 2006). Improvements in single word intelligibility may be beneficial to children who speak at the single-word level, but not to those who are able to communicate with longer utterances. This level of individualized analysis wasn't offered in the papers, limiting their ability to determine the functionality of these outcomes. Another weakness in these studies was that the interventions were only administered for a few weeks (2-6), perhaps contributing to the limited improvement in connected speech (Marchant et al., 2008; Pennington et al., 2009; Pennington et al., 2006). These children may have shown further improvement in connected speech following a longer intervention period. Finally, these studies all had small sample sizes or were single-subject designs, thus limiting generalization of the findings to a larger population. Gathering further information about the individual's ability to functionally communicate by means of speech following improvements in intelligibility would be useful information to validate the value of the speech interventions.

There are challenges involved in designing studies for this population and for this field of research. One such challenge is the heterogeneity of the children involved in the treatment groups. This population exhibits a wide variety of abilities, due to the number of factors that contribute to the severity and type of the cerebral palsy and dysarthria, making it difficult to gather a homogeneous group of individuals. Due to the variability of symptoms that each child exhibits, using single-subject designs can be beneficial because it allows for more control over the study variables. The limited population of children makes it difficult to have a control group along with the treatment group. Without these control groups, it is more challenging to determine whether the speech interventions are accountable for the gains in intelligibility or if these gains are due to extraneous factors, such as the maturation of the children.

More research is warranted in this field in order to further validate the positive findings of these interventions. Further research could explore the effects of speech intervention provided to individuals over an extended period of time. Because this population could be on a caseload for longer than just one short block of therapy, it is realistic to be able to provide a prolonged duration of intervention. Further information could also be obtained reflecting the long-term effects of therapy following extended intervention time.

Conclusions and Clinical Implications

Based on the suggestive evidence provided in the reviewed studies, speech interventions have the potential to be a successful therapy approach for improving intelligibility in children with cerebral palsy and dysarthria. After consideration of the cognitive and language abilities of the participants, appropriately individualized speech interventions may be implemented to aid in increasing intelligibility in order for the child to use spoken language as an effective and efficient means of communication.

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