Critical Review:
Do therapy protocols focusing on breath support help to improve intelligibility in children with dysarthria resulting from cerebral palsy?*

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This critical review examines the effectiveness of therapy targeting breath support on improving intelligibility in school-aged children with dysarthria secondary to cerebral palsy. Four articles were included in the review. All of the studies included were of a single subject design. Overall, the studies of this review demonstrate preliminary evidence that breath support therapy is an effective intervention to improve the intelligibility of children with dysarthria resulting from cerebral palsy. Recommendations for clinical practice and future research are discussed.

Introduction

What is Cerebral Palsy?

Cerebral palsy (CP), is defined as an encompassing term for motor impairment syndromes that are non-progressive, but changing, and caused by lesions of the brain incurred in the early years of life (Sankar & Mundkur, 2005). The global incidence CP is approximately 2-2.5 in every 1000 births and is therefore a significantly prevalent syndrome worldwide (Rosen & Dickinson, 1992).

Communication deficits, most commonly dysarthria, exist in approximately 50% of children with CP (Kennes et al., 2002). Dysarthria is characterized by deficits of the muscles used for speech that may impact multiple systems of the speech mechanism, such as respiration and articulation, resulting in less intelligible speech production (Pennington, Smallman, & Farrier, 2006). Therefore, when considering the prevalence of CP in children and the frequency of dysarthria resulting from CP, it is important for Speech Language Pathologists (SLPs) to know the best practice protocols of how to treat the children of this population.

It has been observed that those with dysarthria have a shallower breathing pattern that interferes with intelligibility because they attempt to produce more syllables quickly on a short supply of air (Hodge & Wellman, 1999). When adults with acquired dysarthria present with the same concerns that are having an impact on intelligibility, such as shallow breathing, low breath support, or increased rate of speech, therapies using a systems approach are recommended (Yorkston, Beukelman, Strand, & Bell, 1999). Although a lack of intelligible speech often directs clinicians to use articulation therapies with children, articulation therapy is only appropriate when other speech systems, including laryngeal, velopharyngeal, and respiration, are functioning optimally (Strand, 1995). Therefore, when considering therapy with children diagnosed with dysarthria secondary to cerebral palsy, the underlying speech systems, such as respiration and rate of speech, must be addressed before articulation therapies can commence. Furthermore, breath support therapies targeting subsystems may generalize and improve intelligibility without focusing on articulation by providing adequate breath supply to produce longer utterances and slowing rate of speech which allows for more precision in placement.

Objectives

The primary objective of this paper is to evaluate the available literature concerning the effectiveness of breath support treatment on intelligibility in children with dysarthria secondary to cerebral palsy. The secondary objective of this appraisal is to provide SLPs recommendations on the implementation of breath support therapy with children who have a diagnosis of cerebral palsy and dysarthria.

Methods

Search Strategy

The computerized databases used to obtain the peer-reviewed articles selected included: Google Scholar, PubMed, and Scholars Portal Journals. The following were used as keywords: [(cerebral palsy) AND (breath support) OR (LSVT) OR (voice therapy) AND (intelligibility)].

Selection Criteria

The studies selected for inclusion in this critical appraisal required the use of a therapy protocol that

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focused on increasing respiratory and phonatory functioning with children who had a diagnosis of dysarthria secondary to cerebral palsy. Furthermore, the studies had to have outcomes measures in intelligibility. For the selection of these articles, the age parameter was 18 years and under. No limitations were placed on the study design, dysarthria severity, year published, or type of cerebral palsy.

Data Collection

The literature search yielded four articles that corresponded with the selection criteria. All four research studies used a single subject design.

Results

Single Subject Design

Single subject designs provide flexibility for individual differences because each participant acts as his/her own control. Single subject designs are therefore appropriate for examining children with cerebral palsy since it takes into account varying types and severities in the diagnosis. Furthermore, two of the four studies (Pennington, Miller, Robson, & Steen, 2010 & Pennington et al., 2013) included multiple baselines in the design which improves the internal validity of the experiment because it controls for factors such as maturity that may influence the changes seen in the participants.

Levy, Ramig, and Camarata (2013) compared the effects of two voice therapies on intelligibility of 3 children (ages 3-9) with mild to moderate dysarthria secondary to spastic cerebral palsy. Two participants completed 16 sessions of Lee Silverman Voice Treatment (LSVT LOUD) while the remaining participant received 8 sessions of traditional voice therapy based on a systems approach (Pennington et al., 2010) due to the inability to commit to the intensity of LSVT LOUD. Both interventions lasted for 4 weeks. Intelligibility was rated by naïve listeners who compared within-participant intelligibility of pre- and post-intervention recordings of single words and spontaneous speech samples.

The authors concluded that the treatments improved the intelligibility of the participants because the post-intervention speech samples were chosen as "easier to understand" more often than the pre-intervention samples.

The primary limitation of this study is the lack of statistical analyses used to quantify the data collected on changes in intelligibility. For example, the authors could have used inter-rater reliability measures to ensure that the ratings between the naïve listeners were consistent. This study also lacks intervention reliability as the therapy sessions were not monitored for consistency of administration. The two therapies were also administered by different researchers of varying qualifications from speech and language pathology students to a certified LSVT LOUD therapist.

The strengths of this study include the number of stimuli for single words (120 words) and spontaneous samples (30 sentences), and the use of well-defined therapy protocols. The authors also screened the listeners' hearing and blinded them to the time of the collected samples. This study offers equivocal evidence for the use of breath support therapies with children diagnosed with dysarthria secondary to CP, but offers a foundation for other exploratory studies.

Pennington et al. (2006) conducted an exploratory study on the effects of a systems approach on intelligibility with 6 children (ages 10-18 years) diagnosed with different types of cerebral palsy. The therapy focused on breath control, speech intensity, and marking stress. Intervention was administered 5 days a week for 5 weeks and each session was approximately 20-30 minutes in duration. Listeners were given a closed set of choices for single words and transcribed the connected speech samples which were compared to the recording for accuracy. One measurement was obtained pre intervention (1 week pre-therapy) and two measurements were collected post intervention (1 and 7 weeks post-therapy) using naïve listeners.

The authors reported that 4 of the 6 participants experienced an increase in intelligibility for single words and connected speech immediately after intervention, but these increases in intelligibility were not statistically significant and most participants returned to pre-intervention intelligibility at the 7 week post-intervention measurement. The statistical analyses used were not accurately presented.

Similar to the Levy et al. (2013) study, the primary limitation of this study is a lack of statistical analyses that could have provided more detailed information concerning the results, such as inter-rater reliability scores for the naïve listeners. Furthermore, this study did not report on the reliability of treatment administration which indicates that the participants could have been receiving slightly different therapy sessions. The therapy was also administered by more than one individual which further deteriorates the fidelity of the intervention. None of the participants completed all 25 sessions and not all students received the same amount of sessions. Additionally, the listeners were given rote samples (i.e. the days of the week, months) to listen to before hearing the study samples

which may have had an acclimatization impact on the listeners to the voices of the participants.

Despite these limitations, this study also had strengths including a well-defined therapy plan with strict progression criteria, which allows for replicability, and appropriate inclusion and exclusion criteria for participants. The listeners were also blinded to the time of the sample. Given the strengths and weaknesses of this study, it provides equivocal evidence for the use of breath support therapies with child diagnosed with CP.

Pennington et al. (2010) investigated whether a systems approach focusing on stabilizing respiration and phonation effort, speech rate, and phrase length would improve the intelligibility of 16 students (ages 11-18 years) with moderate to severe dysarthria secondary to cerebral palsy of varying types. Intelligibility was rated by familiar and unfamiliar listeners using a closed set of responses for the single word samples, and transcription for the connected speech samples which were compared to the recordings. Each student received individual therapy at school three times per week with a research speech and language therapist. Each session was approximately 35 to 40 minutes in duration.

The appropriate number of measures were taken at each time interval, and a stable baseline was established for each of the participants. The treatment took place over 6 weeks, with data collection occurring 1 and 6 weeks pre-intervention as well as 1 and 6 weeks post-intervention. Appropriate statistical analyses of repeated-measures ANOVA indicated a statistically significant increase in intelligibility of single words but more varying results in connected speech with most children experiencing a statistically significant increase. Increases in intelligibility were noted in both familiar and unfamiliar listeners. Furthermore, the results demonstrated that the changes in increased intelligibility were maintained at the 6 weeks post-treatment data collection.

Pennington et al. (2010) identified three limitations within their study. First, the therapy was not monitored for intervention reliability, indicating that the participants may have been administered slightly differing interventions. Second, three students did not receive the same amount of sessions as the other participants. And third, the maintenance of acquired skills from intervention was only tested at 6 weeks post-treatment, therefore the long term impacts of the therapy cannot be statistically determined.

Issues beyond the authors' concerns were also identified in the inter-rater reliability scores. The inter-rater reliability for familiar listeners was low in connected speech (0.31) and moderate in single words (0.53). These reliability scores may have been influenced by the amount of time the familiar listeners spent with the participant. The inter-rater reliability score for unfamiliar listeners for connected speech was also moderate (0.67). The combined inter-rater reliability scores for connected speech may have had an impact on the final reporting of increases in intelligibility in connected speech. In connection with inter-rater reliability scores, it was also found that the agreement score between familiar and unfamiliar listeners was moderate (0.67). A statistical difference was found between improved intelligibility of single words and connected speech for unfamiliar listeners, with single words being significantly higher.

Despite the limitations, this study demonstrated many strengths including appropriate exclusion criteria for selecting participants, a well-defined therapy protocol with specific progression criteria allowing for replicability, and a pool of 120 unfamiliar adult listeners. The authors also blinded the listeners to the time point of the recordings and blinded the unfamiliar listeners to participant identity. Furthermore, there was a high agreement score between familiar and unfamiliar listeners for single words. Based on the strengths of this study, it offers suggestive evidence that therapy focusing on stabilizing breathing, phonation, speech rate, and phrase length may improve intelligibility of single words with unfamiliar listeners in older children with dysarthria secondary to cerebral palsy.

Pennington et al. (2013) continued their exploration in the effects of breath support therapy on intelligibility in 15 children with different types of cerebral palsy as well as moderate to severe dysarthria (ages 5-11 years). Familiar and unfamiliar listeners were given a closed set of responses for single word samples and asked to transcribe connected speech samples. The children were provided 35-40 minutes of intervention three times a week for six weeks (Pennington et al., 2010).

The appropriate number of measures were taken at each time interval and no changes were observed in the baseline collections. The data collection points followed the same timeline as Pennington et al. (2010) and added a fifth collection at 12 weeks post-intervention. After applying appropriate ANOVA analyses, a statistically significant increase in intelligibility was found for most of the participants in single words and connected speech with familiar and unfamiliar listeners. Additionally, the researchers found no decline in intelligibility at the 12 week measurement which suggested that the behavioural changes made by the intervention had been maintained without continuing therapy. Although an overall improvement in intelligibility was discovered,

the differing responses to therapy of the individual children was also noted.

The Pennington et al. (2013) study possessed some of the same limitations as the Pennington et al. (2010) study such as a lack of intervention administration reliability measures and differing total sessions administered to each student. Furthermore, the Pennington et al. (2013) study also experienced some limitations concerning inter-rater reliability. The inter-rater reliability for familiar listeners was low for single words (0.47) as well as connected speech (0.31). Additionally, the second recording was being rated consistently lower in intelligibility than the first recording of the same collection time by unfamiliar listeners.

Despite the limitations, this study demonstrated some of the same strengths as the Pennington et al. (2010) study including appropriate selection criteria for participants, a well-defined therapy protocol, and a large unfamiliar listener pool. There was also the blinding the listeners to the time point of the recordings and the blinding of participant identities to unfamiliar listeners. High agreement scores were observed between familiar and unfamiliar listeners. In addition, the Pennington et al. (2013) study revealed high inter-rater reliability scores in both single words and connected speech for unfamiliar listeners as well as maintained intelligibility in a longer term follow-up examination (12 weeks post-intervention). Based on the strengths of this study, it offers compelling evidence that therapy focusing on stabilizing breathing, phonation, speech rate, and phrase length may impact intelligibility of single words and connected speech in younger children with dysarthria secondary to cerebral palsy.

Discussion

Breath support techniques are used to establish the subsystems of the speech mechanism in order to improve intelligibility in children with dysarthria secondary to cerebral palsy. This paper appraised 4 single subject design studies to determine if breath support intervention is an appropriate approach in increasing the intelligibility of children with dysarthria. It is important to note that small sample sizes and individual characteristics of the participants must be considered when determining the generalizability of the results of these studies. It is also important to bring attention to the fact that 3 of 4 of the analyzed studies were in relation to Dr. Lindsay Pennington's laboratory which may also bias the results slightly. All 4 studies demonstrated that children's intelligibility of single words and connected speech increased post treatment and furthermore the Pennington et al. (2010) and Pennington et al. (2013) studies demonstrated that these changes in behaviour could be maintained over 6-12 weeks. This offers suggestive evidence that breath support therapies help to improve intelligibility in children with dysarthria secondary to cerebral palsy.

A common limitation among all of the studies was intervention administration reliability which may have had an impact on the results concluded from each study. The Pennington et al. (2010) and Pennington et al. (2013) studies also demonstrated discrepancies in inter-rater reliability scores among familiar listeners. Levy et al. (2013) and Pennington et al. (2006) did not demonstrate the use of appropriate statistical analyses within their studies, did not control for external influences on intelligibility by using multiple baseline measurements, and had small unfamiliar listener groups. The aforementioned limitations of the 4 appraised studies indicates that the results must be interpreted with caution and that these studies can be improved upon to offer stronger evidence for the use of breath support intervention with children with dysarthria.

All 4 of the studies shared two strengths in that they all followed very structured therapy protocols with specific progression criteria and blinded the unfamiliar listeners to the identity of the participants as well as the time points of the samples being presented. Furthermore, the Pennington et al., (2010) and Pennington et al., (2013) intervention protocols were described in detail which increases the replicability. The Pennington et al. (2010) and Pennington et al. (2013) studies also helped increase the statistical strength of their findings by including multiple baselines to control for external influences, using appropriate statistical analyses, as well as recruiting large unfamiliar listeners groups. Pennington et al., (2006). Pennington et al., (2010), and Pennington et al. (2013) also incorporated children with various types of cerebral palsy which improves the generalizability of this intervention within the CP population. The strengths of these studies and the fact that the participants experienced increases in intelligibility immediately post-treatment as well as maintained results demonstrates suggestive evidence for the use of breath support therapies for improving intelligibility in children with dysarthria secondary to cerebral palsy.

Clinical Implications

The therapy protocol used in 2 of the studies (Pennington et al., 2010 & Pennington et al. 2013) is well described which makes it replicable in clinical settings. However, this therapy protocol was researched as being administered three times a week in 35 to 40 minute sessions for 6 weeks in the participant's

attending school. A review of speech and language services in Ontario schools by Deloitte (2013) revealed that students usually receive 10 week blocks of therapy and are regularly seen once a week for 30- 45 minute sessions. Furthermore, Deloitte (2013) also reported that the majority of speech and language services are provided to students below grade 4. And since Ontario only provides free services to children who qualify for the Preschool Speech and Language Program, private therapy of this intensity would be very expensive. These service delivery limitations interfere with the ability to replicate this therapy protocol into school speech and language services as well as private therpay. Furthermore, the heterogeneity of the cerebral palsy population needs to also be considered when determining if breath support therapy is an appropriate intervention for a client.

While therapies focusing on breath support offer preliminary evidence of effectiveness, it is suggested that future research endeavours include the following to strengthen the research and extend clinical applicability:

1.1 The effectiveness of breath support therapy with once weekly session compared to sessions three times per week

1.2 A comparison of breath support therapy to traditional articulation therapy in improving intelligibility

1.3 Intervention fidelity measures for the therapy protocol developed by Pennington et al. (2010)

1.4 The effectiveness of breath support intervention on varying severities of dysarthria

1.5 Comparison of cerebral palsy types and effectiveness of breath support therapy on intelligibility

1.6 Larger sample sizes

In conclusion, the current literature leads to suggestive evidence of the effectiveness of breath support therapies on improving the intelligibility of children with dysarthria secondary to cerebral palsy.

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