Critical Review: The Effects of Intrathecal Baclofen Therapy on Speech in Individuals with Cerebral Palsy

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Methods

Search Strategy

The following computerized data bases were searched; AMED, Embase, CINAHL, Medline, PubMed and Proquest Nursing.

Keywords used include:

(Intrathecal Baclofen) AND ((Speech) OR (Intelligibility)) AND (Cerebral Palsy)

The OVID Databases (AMED, Embase, CINAHL and Medline) were useful in obtaining articles that were relatively specific to the topic at hand. PubMed and Proquest Nursing databases were useful in obtaining more general articles addressing the different treatments for CP. These articles only briefly and subjectively mention the possible effects of ITB on speech and do not attempt to support this claim through means of outcome measures.

Selection Criteria

The studies included in the critical review were required to investigate the effects of ITB on speech in individuals with CP, using some form of outcome measure. Articles that did not use any form of outcome measure were excluded from the review.

Data Collection

Results of the literature search yielded two abstracts and three articles pertaining to ITB therapy and its effects on speech. Other articles not meeting the above criteria were used for informational purposes only. Three of these articles obtained subjective evidence through the administration of an interview or questionnaire and looked at speech outcomes as a subcomponent of the study. The other two studies obtained objective evidence through the administration of the Assessment of Intelligibility of Dysarthric Speech (AIDS) (Yorkston & Beukelman, 1981). Both of these studies employed a singlesubject design and focused mainly on speech outcomes.

Results

For a brief outline of articles with regards to speech, see Appendix A.

Studies that Administered a Subjective Measure of Speech Outcome

Albright, Barry, Shafron and Ferson (2001), sought to "evaluate the effects of intrathecal baclofen on patients with severe generalized dystonia." This study looked at several outcomes, including speech. With regards to speech, a telephone questionnaire was administered. Sixty-two out of seventy-seven participants were contacted and responded. The participants were asked several questions including whether speech had improved, been unchanged or worsened. Thirty-three percent reported an improvement in speech, 63% reported no change, and 4% reported that their speech had worsened. No specific conclusions regarding speech were made by the researchers.

Motta, Buonaguro, Stignani, and Condurso (2004) assessed the efficacy of ITB therapy in treating dystonia in children with CP. The study included 22 participants obtained from a convenience sample, and focused on any resulting changes in the dystonia. With regards to speech, a subjective patient's questionnaire was administered following pump implantation. Fifty percent of participants reported speech improvements as a result of the reduced dystonic movements. Again, no specific conclusions regarding speech were made by the researchers.

Bjornson, McLaughlin, Loeser, Nowak-Cooperman, Russel, Bader and Desmond (2003) sought to clarify what issues, if any, related to oral motor control are affected by ITB. Thirty children and their families were part of a convenience sample for this study. An interview tool containing 33 questions was designed for this study. At the time, it had not yet been tested for validity, reliability or responsiveness. The interview was administered in person, to the child and parent, at a mean time of 2.1 years post pump implantation. Twenty-three of the s aip

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speaking rate slowed from 50 words per minute at baseline to 29 words per minute for both the 25 and "100mgs" doses. The intelligible words per minute increased from a baseline mean of 9 to a post-trial mean of 17 (Fisher exact: p<0.001). The participant's intelligibility of single words did not show any significant improvement. The results from the Frenchay Dysarthria Assessment (Enderby, 1983) indicated that volume and 'in speech' measures from the laryngeal section improved significantly (both changing from 2 to 4.5). The authors concluded that there was a significant change in the intelligibility of speech after administration of intrathecal baclofen.

Leary, Gilpin, Lockley, Rodriguez, Jarret, & Stevenson, (2006) completed a single-subject study in order to determine the effect of ITB on the speech of an ambulant adult with CP. Their participant was a 40 year old male with severe spastic dysarthria whose speech was "characterized by a harsh and effortful voice with intermittent phonation breaks and probable laryngeal spasm." In addition, the participant reported tightness and pain in the throat along with diaphragmatic tension when talking. The participant had a reduced intelligibility of speech.

The AIDS was used as the outcome measure for speech and was performed by one observer and scored by another independent observer. Observers were not blinded to treatment status. Outcome measures were obtained at baseline, post trial and 6 months post-pump implantation. Study results indicated improvements following the trial of 25μ g of ITB. Outcome measures at 6 months post implantation indicated sustained improvements. From the AIDS, intelligibility for single words changed from 88% to 84% to 94% from baseline, to post-trial to post-implant, respectively. His correct complete sentences changed from 9.1% to 31.8% to is reliable. The test also purports to be sensitive to change. Intelligibility is a good outcome to measure because it may be considered the universal consequence of dysarthric speech, and may serve as global index of severity (Yorkston & Buekelman, 1981). Intelligibility is considered by Yorkston and Buekelman (1981) to be the "sum of the interacting processes that are involved in speech production." So, if breath support is improved as a result of ITB, this will in turn improve intelligibility, or if laryngeal spasticity is reduced as a result of ITB, this will improve intelligibility, and so on. Improvements seen in intelligibility will mean that improvements have been made in speech.

Despite this major strength in the two singlesubject studies, both studies had several weaknesses which result in caution needing to be taken when interpreting their results. The major weakness of both studies was the use of a single-subject design. This study design leads to major flaws such as a lack of power and an inability to generalize results to the larger population at hand.

The study by Mason et al (1998) had multiple weaknesses which call into question the validity of the results. First, little information regarding the participant and methods were given, further affecting the generalizability of the results. Second, the authors did not mention whether the two pretests and two posttests were rated by the same individual. The same observer needs to rate both tests in order to maintain reliability of the AIDS (Duffy, 2005). If not, this could result in instrumentation, a threat to internal validity. In addition, the researchers only give the mean intelligible words per minute, so the Intelligibility of Dysarthric Speech (CAIDS) or the Preschool Speech Intelligibility Measure (Morris, Wilcox, Schooling, 1995), and acoustic measures such as jitter and shimmer. Acoustic measures can be used on all individuals while the intelligibility test chosen will depend on the age, mental abilities, and literacy skills of the subjects included in the study. Intelligibility and/or acoustics should be measured pre ITB, post ITB trial, and a couple of times post ITB pump implantation to monitor for continued change.

In addition to the quasi-experimental design, future research should also include an analysis regarding the characteristics of those subjects whose speech does improve verses those who experience no change. This will help future professionals to better predict those individual's whose speech will benefit from ITB therapy and those who won't.

Following completion of the above research recommendations, future research may also focus on comparing improvements in individuals treated for spasticity verses those individuals treated for dystonia.

Conclusions

It is concluded that each article on its own appears to provide support for the notion that ITB may improve speech in individuals with CP. Although the three studies containing subjective evidence reported that 33-50% of subjects experienced an improvement in speech, this information is descriptive in nature and cannot be used to establish causality. The two studies that obtained objective evidence reported that there was an improvement in speech. However, both of these studies were single-subject studies, causing them to lack power and making the results not generalizable to the larger population at hand.

Regardless of these major flaws, it is concluded