Critical Review: Evidence of the Presence of Literacy Difficulties in Children Who Have Childhood Apraxia of Speech

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This critical review examines the presence of literacy disorders among children who have been diagnosed with childhood apraxia of speech (CAS). Overall, research suggests that children diagnosed with CAS are at risk for having literacy difficulties at school age however this evidence is guarded due to weaknesses in the literature. The findings of this review have implications for researchers and practicing clinicians in the field of speech-language pathology.

Introduction

Apraxia of speech in children is classified by several different names. Some more common names given in the literature are Childhood Apraxia of Speech (CAS), Developmental Verbal Dyspraxia, Developmental Apraxia of Speech, Developmental Articulatory Dyspraxia and Dyspraxia. For simplicity, within this paper the concept of apraxia of speech in children will be referred to as CAS, because this is the most recent term.

There is a long lasting controversy regarding the presence of CAS and its diagnostic label within the literature of communication disorders (Love, 2000). CAS, when initially identified presents differently in different children, but it is hypothesized that all children with CAS have a difficulty with planning or programming the motor-speech movements to produce speech (Ozanne, 2005). The controversial diagnosis of CAS is long-standing as some of the characteristics of CAS are also seen in children with other speech disorders. Therefore distinguishing it from these other speech disorders can be challenging. There is debate over whether CAS is a motor planning problem only, or a type of phonological disorder (Velleman, 2003). Over many years criteria for the diagnosis of CAS has changed, and many characteristics have been identified that assist in the diagnosis of CAS. In the literature, there is little evidence regarding the best treatment practices for children with CAS. Perhaps this is due to the challenge in identifying CAS in the first place.

CAS is a disorder that occurs during child development so it is important to recognize the progression of the disorder and how it affects other areas of child development besides speech development alone. It has been proposed that children with CAS present with various symptoms that change over time in each individual (Velleman & Strand, 1994). Love (2000) highlighted that some researchers have suggested that linguistic impairment is a characteristic of CAS. This suggests that children with CAS could be at risk for a language disorder. Stackhouse and Snowling (1992a) identify that prior research failed to acknowledge the impact that CAS has on a child's developing language system. In summary, according to researchers there is a possibility that the presence of CAS is correlated with language disorders in children, therefore the development of any linguistically demanding task could be at risk. The literacy tasks: reading and spelling, place high demands on an individual's speech and language abilities, and therefore these children with CAS might have an increased risk in developing disorders in those areas. Moriarty and Gillon (2006) identified that traditional treatment approaches for CAS focus little on literacy difficulties. This might not be a large issue if children with CAS do not have literacy difficulties, however, if they do in fact have literacy difficulties, there should be implications on the child's speech and language treatment.

Both spoken and written language are important in today's literate society. In schools there is a strong emphasis on developing literacy skills, in order to equip children for success later in life (Gillon & Moriarty, 2007). Children with literacy difficulties are likely to have difficulty in school as literacy is central to classroom learning, particularly in higher grades when classroom demands require that the children read to learn. Thus there is great value in children developing strong literacy skills.

Objectives

The primary objective of this paper is to critically evaluate the existing literature pertaining to evidence of the presence of literacy difficulties, including reading and spelling, in children diagnosed with apraxia of speech. The secondary objective is to

propose evidence-based recommendations for future practice and research regarding literacy impairment in children who have CAS.

Methods

Search Strategy

The following computerized databases were searched: CINAHL, Cochrane, ComDis Dome, Medline, and PsychInfo.

The criteria used to search these databases were:

((apraxia of speech) OR (dyspraxia)) AND ((literacy) OR (read*) OR (writ*) OR (spell*) OR (phonological awareness)) AND (child*)

Reference lists of the articles selected were also searched for further relevant articles.

Selection Criteria

Studies selected for inclusion in this review were required to investigate reading or spelling abilities of school-age children with apraxia of speech. No articles including participants with an intellectual or neuromotor disorder were included (i.e. cerebral palsy or down syndrome). Also studies were not included if the participants used augmentative and alternative communication as their primary method of communication. Further, studies that focused solely on the treatment methods of reading and spelling were not included in this review.

Data Collection

Results of the literature search yielded the following study types: a case-control study, a cohort study, and a case study. One informational article was used for background information within this paper.

Results

In their case-control study, Snowling and Stackhouse (1983) investigated reading and spelling strategies in school-age children with CAS. The study compared two groups; four children with CAS and four reading-age matched controls who had normal articulation. The groups of children were compared during three activities; an oral imitation and word writing task, an oral reading task, and a word copying task. Audio recordings of oral responses were scored for phonetic accuracy. A descriptive analysis was completed as opposed to a statistical analysis. Children with CAS were found to have more difficulty with tasks of imitation, spelling and reading than the control group. Reading difficulty in CAS was recognized to be less striking than imitation and spelling errors. For the children with CAS, spelling errors were more common on final consonants than initial consonants. It was found that there was poor agreement between the articulation and spelling errors of children with CAS, suggesting that spelling errors were not easily explained by a child's speech errors.

In a cohort study, Lewis, Freebairn, Hansen, Iyengar and Taylor (2004) examined the differences in speech/language and written language skills between school-age children with CAS and children with other speech-sound disorders. They also looked at the change and progression in these skills over time. In this study 10 children with suspected CAS were compared with two control groups; a group of 15 children with speech disorders and a group of 14 children who had speech and language disorders. The three groups of children were tested at preschool age and at school age. Preschool testing included assessment of articulation/phonology, oral motor skills, language and conversational speech. At school-age follow-up these were measured again in addition to written spelling, reading decoding, reading comprehension and intelligence assessments. Using statistical analysis, comparisons were made within the CAS group over time as well as between the three groups. Results indicated that children with CAS continued to have deficits in speech although there was some improvement over time. They also had language deficits showing little improvement between preschool and school age. Children with CAS were reading, spelling, and academically impaired relative to the speech disordered control group as well as normative standards (one or more standard deviations below the mean). Children with CAS were found to spell significantly poorer than children in the speech and language disordered control group. Finally, results suggested that the spelling of children with CAS was inferior to their reading.

Stackhouse and Snowling (1992b) investigated two case studies of school-age children with CAS for links between phonological difficulties and reading and spelling skills at two points in time approximately four years apart. There was no control group established in this study, however the two children's measures throughout the study were compared to test results from either an articulation-matched, reading-matched or spelling-matched group of normal children established in a previous study. During both testing periods a series of standardized speech and language tests were conducted as well as detailed testing of auditory-phonological processing, reading and spelling. The results of the reading and spelling testing were analyzed descriptively. Results indicated that both participants in the study had

pervasive phonological difficulties. Reading difficulties were evident by weaknesses in letter knowledge, oral reading strategies and silent reading skills. The participants were also found to have difficulty with spelling. Over the four year period there was some improvement in intelligibility, letter knowledge, oral reading strategies and spelling. Spelling non-words was difficult for the participants and little improvement was observed over four years. At the second testing period standardized tests showed only marginal improvements in reading and spelling. After four years of intensive speech therapy, a phonic teaching regime and being taught letter-sound translation rules the participants were still unable to apply rules to reading and spelling. It was found that there were serious persisting deficiencies in phonological spelling. In summary the findings of this study suggested that children with CAS may have difficulty acquiring literacy skills.

Discussion

Subject Selection and Characteristics

Diagnosis of CAS varied across the studies analyzed. Lewis et al. (2004) utilized a rigorous criterion for selecting children with CAS. In this criterion, participants were selected based on diagnosis of CAS from the child's speech-language pathologist (SLP), they were then screened for motor programming aspects in their speech disorder and then were required to demonstrate at least four commonly reported characteristics of CAS from a list of eight. Snowling and Stackhouse (1983), and Stackhouse and Snowling (1992b) specified criteria for CAS, however the diagnosis of CAS was provided by each child's personal SLP. Therefore, due to different SLPs diagnosing the children, the identification of children may not have been consistent or reliable in these two studies. If participants in the studies were misidentified, the results of the study might not have been valid, therefore having a well defined diagnostic procedure for CAS is imperative.

Although all children in the studies were identified as having CAS, there were also some differences amongst the individual children. All of the children with CAS were identified as having typical intelligence at the initiation of each study, except for one child from the Snowling and Stackhouse (1983) study. Attention deficit hyperactivity disorder and learning disabilities were identified in some participants in the Lewis et al. (2004) study. Phonological awareness difficulties and a history of hearing problems were identified as potential interfering disorders in the Stackhouse and Snowling (1992b) study. The presence of these additional disorders could affect the generalizability of the studies to children who have CAS only.

All the studies analyzed lacked important details regarding the selection method for participants in the studies. None of the studies described the process for recruiting children with CAS, nor did they mention any random selection of children for the control groups. Random selection of participants could not be applied in these studies due to the small sampling population. Thus, if an individual met the participant criteria, they would be included in the study. Because the method for participant selection was not clearly identified it is possible that there could have been selection bias in these studies which could affect the generalizability of the results.

The studies varied in their sample size as the studies included two, four or ten children diagnosed with CAS. The small sample sizes could be due to the limited occurrence of CAS in the population. Sample size may have also been affected by the CAS diagnostic criteria used in the studies that may have only recognized moderate to severe CAS in children. Whatever the reason, small sample sizes leave much to chance, limit generalizability and reduce the likelihood of seeing existing effects.

Method

All studies focused on articulation, reading and written spelling abilities in children with CAS. Although some studies also measured auditory-phonological processing, oral-motor skills, language, intelligence and copying abilities, these are not the focus in this review. Each study reviewed used a different design and some study designs delivered more convincing evidence than others.

One commonality among all the studies reviewed was that there was no mention of blinding. None of the studies specify who administered the tests to the participants. There is no way of knowing if the researchers administered the tests, or different clinicians administered the same test to different participants. In order to increase the study quality these should be controlled carefully. By blinding the assessors, bias could be reduced and the results of the study could have been stronger and more reliable. Inter-rater reliability was assessed and found to be adequate on transcriptions of articulation on these tests; however it was not assessed on all of the tests including tests of spelling and reading.

The studies by Snowling and Stackhouse (1983), and Stackhouse and Snowling (1992b) both

involved descriptive analysis. Descriptive analysis was necessary and relevant for the purposes of the studies in order to expose detailed patterns and draw conclusions about reading and spelling abilities. It should be recognized that because it is not a technical procedure it would be difficult to duplicate. Researcher competence is key in type of research; therefore, the methodology of the studies should give clear descriptions of who the researchers are and describe their qualifications, which was not done in either of these studies.

All of the analyzed studies lack important information describing the speech and language treatment that the children with CAS had prior to the study and over the course of the study. Treatment over the non-testing years of the Lewis et al. (2004) and Stackhouse and Snowling (1992b) studies was neither tracked nor controlled for, and could have influenced the reading and spelling results on the follow-up assessments.

Another weakness that was discovered in the Lewis et al. (2004) study is that some of the tests that compared the children from preschool to school-age differed. For example the expressive and receptive language test done at school-age was the CELF-4 but the test done at preschool age was the TOLD-P:2. Due to different norming populations on these tests it would be difficult to compare their results. Of the tests that were changed, none included analysis of reading or spelling, therefore this potential weakness is not an issue in this analysis. Reading and spelling conclusions drawn from the study would not be affected by this.

In the Stackhouse and Snowling (1992b) study the first testing period lasted much longer in duration than the second testing period. The study should have indicated why the initial testing took an extended period of time and should have highlighted any possible implications this could have had on the study results. There was also a weakness in this study because it did not have one consistent control group. If there was a consistent control group participating in this study, it could have had a more favourable level of evidence. A final weakness in the method of this study was that it included the administration of some non-standardized tests that had questionable construct validity.

Statistical Analysis

Snowling & Stackhouse (1983) did not use statistics to draw conclusions or establish significance. Perhaps it was due to small sample size that they just compared raw scores of the children with CAS to the control group and explained them descriptively. This is a weak method of comparison, however doing a descriptive analysis allowed them to describe trends in the data, and this contributed to the study's results. With few participants in each group the study likely would not have enough power to reject the null hypothesis; therefore it was appropriate that a descriptive approach to this study was used rather than statistical analysis.

The issue of small sample size was also seen in the study by Stackhouse and Snowling (1992b). In this study, statistics were not used throughout the article to demonstrate significance, however, some of the subtests in the study were analyzed using statistics. A t-test was appropriately used to compare the output phonology of the two participants to the mean of an articulation-matched control group. On the oral reading strategies subtest, a z-test was employed to assess each participant's performance compared to the reading-age matched control group. On other tests, comparison with the control group entailed searching to see if the participants' scores fell within the range of the control groups' scores. Overall, it is important to note that the majority of the study included detailed descriptive analysis of reading and spelling abilities which could not be evaluated statistically.

Lewis et al. (2004) used chi squared tests and analysis of variance (ANOVAs) to examine the group differences in age, gender and socioeconomic status. A suitable between groups ANOVA was used to compare the three groups for articulation/phonology, oral-motor skills and language at preschool age. ANOVA was also used to compare the groups' performance on tests of articulation/ phonology, oral-motor skills, language, spelling and reading at school age. The group main effects that were significant were appropriately followed up by employing the Tukey HSD test as this compares all possible pairs while maintaining the type one error. Bonferroni corrections were made within each domain for multiple comparisons. Analysis of covariance (ANCOVA) was employed to examine whether the degree of change over time within each group was different across groups. The ANCOVA compared groups for articulation and language tests at school age and their corresponding test at preschool age was the covariant. The ANOVA done at school age is most relevant for the purposes of this paper and it revealed moderate to large effect sizes on reading and spelling tasks indicating that the reading and spelling abilities of children with CAS were significantly poorer than those of the comparison group(s).

Summary Statement

The studies discussed ranged in level of evidence, reliability and validity. The Lewis et al. (2004) study was a well-designed cohort study with suggestive validity and compelling importance. Although the design of the Stackhouse and Snowling (1992b) study was a case study reflecting expert opinion, it was also found to have suggestive validity and compelling importance. Finally, the case-control study by Snowling & Stackhouse (1983) was found to have equivocal validity but compelling importance. Regardless of weaknesses in study designs the overall importance of the findings should not be disregarded. All of the analyzed studies found that children with CAS had difficulties spelling. Spelling non-words was notably difficult for these children. Reading difficulties were also a common finding among the studies, although they were less conspicuous than spelling difficulties.

Recommendations

It is difficult to have complete confidence in the research findings due to concerns regarding, subject selection, study design and statistical analysis. However, based on the critical review of the available literature, there is evidence suggesting that school-age children with CAS are at risk for having literacy difficulties in the areas of reading and spelling. As such, the following research recommendations and clinical implications should be considered:

1) More evidence about the language and literacy disorders in children with CAS is needed.
2) Future studies regarding children with CAS should include a well defined rigorous selection criterion, larger sample sizes, blinding, and an increased variety of measurement tools.
3) Further research regarding the prevalence of literacy impairment among children with CAS is needed.
4) Clinicians should be aware that children with CAS could be at risk for literacy difficulties, and therefore are encouraged to monitor these children more closely, and/or implement early intervention targeting early literacy skills.
5) Clinicians should also be aware that guarded evidence suggests that spelling non-words may be particularly difficult for children with CAS.

Conclusions

The present literature suggests that children with CAS are at risk for having literacy difficulties in the areas of reading and writing. Despite weaknesses in the literature reviewed, the evidence discovered was found to be of great importance. This information is important for clinicians to consider when developing intervention plans for children with CAS. Further research is needed to lend more support to the conclusions put forth in the current literature.

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