Critical Review: Fetal alcohol spectrum disorder in Aboriginal children: Does a language profile emerge?

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This critical review examines the language profile of Aboriginal and non-Aboriginal children with fetal alcohol spectrum disorder (FASD) in five studies. A literature search was conducted and the following study designs were included: four quasi-experimental studies (case control) and one qualitative study. Based on the critical review, a specific language profile for this population did not emerge. Overall, evidence revealed that all children with FASD demonstrate a variable language profile, which may fluctuate with age. Additional studies involving more subjects, cognitively matched control group, culturally sensitive assessment approach and a more comprehensive battery of measures is recommended in order to obtain more consistent results.

Introduction

In 1973, a group of children born to mothers who consumed excessive amounts of alcohol during pregnancy were described by Jones and Smith (1973). They termed the pattern of deficits observed in these children as Fetal Alcohol Syndrome, which was later changed to Fetal Alcohol Spectrum Disorder (FASD). In this study, they described a pattern of congenital malformations that included growth deficiencies, central nervous system dysfunction, mental deficiencies, and abnormalities of internal organs and the skeletal system (Jones & Smith, 1973).

The prevalence of FASD in the general Canadian population is estimated to be approximately 1%. This translates into about 300 000 people in Canada living with FASD. In addition, it is estimated that approximately one out of every 500 to 3000 births will have FASD (Health Canada, 2001). Within an Aboriginal population, prevalence estimates are suspected to be much higher. It must be noted that accurate prevalence rates are difficult to obtain due to the diversity of the population and a general mistrust of mainstream society (Szlemko, Wood, & Thurman, 2006). Health Canada (2001) suggests that FASD can be as high as one in every five births in some Aboriginal communities, and a study conducted in an Aboriginal community in northern British Columbia in 1987 (Robinson, Conroy, & Conroy 1987), identified 22 of 116 children with FASD.

Since there is a high prevalence of FASD within an Aboriginal population, the need for evidence-based guidelines in speech-language assessments is critical. In addition, Aboriginal children often present with a language profile that is different than that of a non-Aboriginal population due to cultural differences. With these differences in mind, one could expect that Aboriginal children with FASD will also present with a different language profile compared to non-Aboriginal children with FASD. With research to support this premise, a language profile for an Aboriginal FASD population could be discovered. This profile would be critical to help guide the speech-language pathologist in developing appropriate assessment and treatment approaches.

Objectives

The primary objective of this paper is to evaluate the existing literature that examines the language characteristics of Aboriginal and non-Aboriginal children with FASD. From this evaluation of the literature, a summary of expressive and receptive language characteristics will be developed for an Aboriginal population. The secondary objective is to provide an appropriate recommendation for speech-language pathologists regarding assessment of an Aboriginal population with FASD.

Methods

Search Strategy

Computerized databases, including ProQuest Education Library, CINAHL, PsycINFO, SCOPUS, Science Direct, and the University of Western Ontario libraries search engine, were searched using the following key terms and search strategies:

> ((language) OR (linguistic) OR (communicat*) AND (fetal alcohol) AND (aboriginal) OR (Indian) OR (first nation) OR (native)).

The search strategy was also employed without the terms "aboriginal", "Indian", "first nation", and "native" to locate studies in a non-Aboriginal population.

Selection Criteria

Studies included in this critical review were selected if they were conducted on Aboriginal FASD and/or non-Aboriginal FASD child populations, and if their goal was to evaluate expressive and receptive language.

Data Collection

Results of the literature search yielded five studies that met the above selection criteria: four quasiexperimental designs (case control study) and one qualitative research design.

Results

The following studies are organized according to strength of the results in relation to the objective of this critical review.

Study #1 Becker, Warr-Leeper, and Leeper (1990) conducted a case control study to describe and compare the speech and language characteristics of six Aboriginal children with FASD (age range: 4 years, 6 months to 9 years, 6 months) to culturally and cognitively matched, non-FASD Aboriginal children. Children were matched based on non-verbal cognitive ability. The authors used a Mann-Whitney U test to evaluate differences between the FASD group and the typically developing group. This test was used to compare means between groups of different sizes. To evaluate quantitative differences in language development (i.e. typical language development, but acquisition is at a slower rate), an analysis of scores was conducted based on chronological age. Similarly, the same analysis was conducted based on mental age to evaluate qualitative differences in language development (i.e. language differences that cannot be accounted for by cognitive ability). All subjects were evaluated on a nonverbal cognitive measure, grammatical measures, semantic measures, and memory measures.

The results relevant to this review indicated that Aboriginal children with FASD scored significantly below typically developing children of the same age in terms of comprehension of syntactic forms and semantics at the single word level. These children also produced fewer grammatically complete and accurate sentences in spontaneous speech than the younger control group. This data suggested that there are significant quantitative differences in grammatical ability (both comprehension and production) and semantic comprehension of single words based on chronological age. In addition, there were no qualitative differences in language development observed in this study. A major limitation of this study was the small sample size selected to evaluate language in an Aboriginal FASD population, although this sample size is typical for this population. This limitation will impact the power of results obtained within this population. In addition, the use of culturally biased measures to evaluate syntactic and semantic development may have skewed the results. That is, Aboriginal children may score lower on tests designed for mainstream society, since items and testing methods may not be familiar to them. In addition, norms are often based on children from the United States representing different cultural backgrounds, providing an invalid comparison for an Aboriginal population. This discrepancy was noted in the article and the authors accounted for culturally biased testing materials with the use of derived scores. A further limitation of this study was that some subjects (i.e. FASD and controls) did not complete all tests, further limiting the validity of the results due to the increasing small sample size for analysis. Although, this study displays several weaknesses, the design of this study was appropriate for the population due to the rarity of the disorder within a minority population, such as Aboriginal children. In addition, the use of cognitively matched subjects on the basis of non-verbal intelligence scores eliminates language differences on the basis of intelligence. The authors also used a comprehensive battery of tests to assess syntactic and semantic development, which may provide further direction for speech-language pathologists when assessing an Aboriginal population with FASD.

Results from this study suggest that Aboriginal children with FASD display difficulty in comprehension and production of syntactic forms and comprehension of semantics, as compared to their culturally matched peers. Given the level of evidence for this study is rated moderately-low, results are suggestive and should be implemented in clinical practice with caution. The results can be used to guide the clinician when selecting possible areas of assessment (i.e. syntactic and semantic knowledge).

Study #2 Carney and Chermak (1991) conducted a case control study to examine and compare the expressive and receptive language abilities of 17 typically developing Aboriginal children and ten Aboriginal children with FASD (age range: 4 years to 12 years, 11 months). The subjects were matched based on age and the control subjects were determined to be typically developing by observations made by a teacher. The researchers used the *TOLD-I* and *TOLD-P* to assess receptive and expressive language skills.

The authors used the independent t-test to compare means between the different age groups and the control groups. The results indicated that younger Aboriginal children with FASD (age range: 4 years to 8 years, 11 months) performed significantly below the control group in both receptive and expressive areas of language. This indicates that younger Aboriginal children with FASD display more global language deficits. The older Aboriginal FASD children (age range: 8 years, 11 months to 12 years, 11 months) performed significantly below mean performances of the control group on measures of comprehension and production of syntax.

A major limitation of this study was the use of a small sample size. This limitation will impact the power of results obtained within this population. It should also be noted that the age groups differed in size (i.e. 7 subjects in the younger FASD group and 3 subjects in the older FASD group), making comparison between the groups difficult. In addition, the small size of the older group made the results from this group less reliable. Method of selection for the control group was a further limitation. This group was selected based on teacher observation, therefore not ensuring the control group was typically developing. This made the results difficult to interpret as language deficits could have been a function of FASD or depressed cognitive function, which was discussed by the authors. It should also be noted that one control subject fell below the 16th percentile on three subtests. The authors suggested this could have been the result of culturally biased testing or the lack of intelligence testing to ensure a typically developing control group. These results suggest that the testing methods used are inadequate to measure language development in this population as Aboriginal non-FASD children score significantly below test norms on these tests. This further highlights the need for culturally sensitive testing within this population. This study also used a limited battery of assessment tests. making results exploratory and less comprehensive. The authors expressed caution in interpreting results due to the limitations of the statistical analysis, particularly when considering management of language problems in this population. It was also noted that differences in cultural and environmental experiences and cognitive ability make interpretation of the results difficult.

Although this study has various limitations, it does provide some guarded support for a possible language profile of Aboriginal children with FASD. This study suggests that there may be language differences as the child ages. That is, younger Aboriginal children with FASD tend to show more global language deficits while older Aboriginal children with FASD tend to show more deficits in syntactic ability. Given the level of evidence for this study is rated moderately-low, results are suggestive and should be implemented in clinical practice with caution. The results do provide some guidance for the clinician when selecting possible areas of assessment.

Study #3 Engelhart (1992) conducted a case control study to examine and compare the expressive and receptive language abilities of 21 Aboriginal children with FASD to 21 Aboriginal children without FASD. The age range of these children was from 4 years, 4 months to 12 years, 8 months. This researcher used the *TOLD-I* and *TOLD-P* to assess receptive and expressive language skills.

The author used the independent t-test to compare means between the different age groups and control groups. The results indicated that younger (age range: 4 years, 4 months to 8 years, 6 months) Aboriginal children with FASD had significantly lower receptive and expressive language skills than the non-FASD Aboriginal group. However, results of the older (age range: 8 years, 6 months to 12 years, 8 months) Aboriginal children with FASD showed no significant difference in receptive or expressive language skills as compared to a non-FASD Aboriginal population.

A major limitation of this study was the small sample size selected, although this study was the largest in comparison to the other studies discussed. This limitation will impact the power of results obtained within this population. Also, control subjects were not cognitively matched to the FASD population. The author used the Peabody Picture Vocabulary Test (PPVT) to provide a common language reference point in lieu of IQ scores. This test is not an appropriate measure to match children on language ability as it only measures the child's receptive vocabulary. The lack of control for cognitive ability makes it difficult to determine if language deficits are a function of FASD or cognitive ability. A further limitation, which was noted by the author, was that the younger and older FASD groups were uneven (i.e younger group had 16 subjects and older group had 5 subjects), making the comparison and interpretation of the results difficult.

Overall, the results of this study suggest that younger Aboriginal children with FASD tend to show more global language deficits while older Aboriginal children with FASD appear to exhibit language profiles similar to their non-FASD peers. Given the level of evidence for this study is rated moderately-low, results are suggestive and should be implemented in clinical practice with caution. Due to the level of evidence and limitations of this study, results should be interpreted with caution. In addition, clinicians should be cautious of results from this study, since it has not been published and therefore not peer reviewed. With the above considerations, this information can provide direction and cautions for future researchers when evaluating language profiles of Aboriginal children with FASD.

Study #4 Hamilton (1981) conducted a case control study to examine the linguistic system (i.e syntax, semantics, pragmatics, and short-term memory) of ten children with FASD (age range: 4 years, 5 months to 6 years, 10 months) as compared to a group of typically developing children of the same age level (control group 1), to a younger group of typically developing children of the same linguistic level (control group 2), and to a group of children with Prader-Willi syndrome of the same age level and linguistic level (control group 3).

The author used a two tailed t-test for paired samples when comparing the FASD group with the three control groups. Generally, FASD children performed like the younger typically developing group in terms of comprehension of syntactic forms and produced less complex grammatical forms in comparison to this same group. In terms of semantic development, FASD children produced and comprehended single words much like the younger typically developing group and had similar semantic complexity of their utterances as the younger group. Overall, results from this study concluded that children with FASD score significantly below mean performance of typically developing age matched subjects for syntactic and semantic language development and were developing these language functions at a slower rate than younger typically developing children of similar linguistic level.

A major limitation of this study was the use of a small sample size. This limitation will impact the power of results obtained within a FASD population. In addition, the researcher used more assessment measures of production than comprehension, which they noted as a caution in the article. This makes comparison of the results difficult since the results of language comprehension may be less comprehensive than that of language production. The researcher also used a group of children with Prader-Willi syndrome for a control group that was of the same age level and linguistic level. This group was used to determine if language differences were a function of intelligence or FASD. Using a control group characterized by a syndrome may present confounding factors since this group may present characteristics of language and intelligence that are unique to that syndrome making comparison with the FASD group invalid. Although this study displayed

various limitations, it also had strengths to support the validity of the results. To account for linguistic level, the researcher matched the subjects according to mean length of utterance (MLU). They also used a comprehensive battery of assessment methods, making the assessment results more reliable. In addition, they employed inter-judge reliability to confirm accuracy of results for all measures (i.e. language sample transcription and all formal tests).

Overall, the results of this study suggest that children with FASD display deficits in comprehension and production of syntactic structures and semantic complexity. Given the level of evidence for this study is rated moderately-low, results are suggestive and should be implemented in clinical practice with caution. Due to the limitations and level of evidence of this study, the results provide exploratory evidence in the field of FASD and language deficits and will help guide the speech-language pathologist in selecting areas of assessment and analyzing expected outcomes in treatment. In addition, clinicians should be cautious of results from this study, since it has not been published and therefore not peer reviewed.

Study #5 Church et al. (1997) conducted a qualitative study to evaluate speech and language problems in 22 patients with FASD (age range: 3 years, 8 months to 26 years, 11 months).

The researchers concluded that all FASD subjects who participated in language testing exhibited deficits in both receptive and expressive language, usually performing below the 5th percentile. Specifically, 18 of the 22 (82%) subjects displayed receptive language deficits and 17 of 21 (76%) subjects displayed expressive language deficits. The authors defined a deficit if the child scored below the 13th percentile on at least one of the measures. For the purposes of this critical review, subjects over 13 years of age were removed from the analysis to allow for comparison across studies and to remove subjects' scores where test norms were violated (i.e. tests were administered to subjects that did not fall into the age range for that test). After applying this exclusion criterion, 11 of 15 (73%) subjects exhibited receptive language deficits and 7 of 14 (50%) subjects exhibited expressive language deficits as expressive and receptive language deficits were operationalized by the authors.

A major limitation of this study was the use of a small sample. This limitation will impact the power of results obtained within a FASD population. In addition, the qualities that make up the group (i.e. composed of a majority of African American subjects) do not allow for extension of results to a mainstream society population. A further limitation was that not all subjects were measured on all language tests. Only 8 of 22 subjects were evaluated on all 5 measures of receptive and expressive language. The authors also considered deficits (either receptive or expressive) when the subject scored below at least the 13th percentile on at least one of the tests, even if they scored within normal limits on other measures of receptive or expressive language. In addition, some of the language measures selected were violated. Some of the measures they used only extend up to 17 years of age and they used these measures on subjects over the age of 18, making comparison to norms impossible. They also reported that there was no apparent racial or gender issues but do not state how they determined this finding. In addition, the authors do not discuss limitations of their study, how it related to other research, or how their findings can be clinically implicated.

Overall, results from this study should be interpreted with caution and the results should only be used to help guide future research. Given the level of evidence for this study is rated low, results are equivocal and should not be implemented in clinical practice. Due to the level of evidence, limitations and weakness of this study, the results obtained should not be used to guide clinical decisions when evaluating a child with FASD.

Discussion

Overall, the literature collected demonstrates several limitations. Each study utilized small sample sizes, which limits the power of the results obtained within the studies. Also, control subjects in one study (Engelhart (1992)) performed significantly poor in some areas of language, limiting the homogeneity of the control groups. Also in this study, some FASD subjects scored at the floor of available norms, suggesting testing methods were not appropriate for this unique population. This limitation further supports the need for culturally sensitive testing methods. Some studies also used undesirable methods of selection for the control group (Carney and Chermak (1991), Engelhart (1992), Hamilton (1981)). Subjects were not cognitively matched or displayed either characteristics of a particular disorder. This limits the ability to attribute language deficits found to a FASD population. Uneven groups were also observed in studies that examined age differences (Carney & Chermak (1991), Engelhart (1992)). The uneven groups, likely the result of small sample sizes, makes evaluation of language differences across age levels difficult to determine.

Due to the lack of quality research with both an Aboriginal FASD population and a non-Aboriginal

FASD population and the variable results of the research available, one cannot make a conclusion regarding language differences between these two populations. Despite the limitations of the above studies, what can be concluded (supported by Engelhart (1992), Carney & Chermak (1991), Becker, Warr-Leeper, Leeper (1990)) is that FASD Aboriginal children in general, tend to have more global language deficits when they are younger but as they grow older, their language profiles become more variable, sometimes showing greater difficulty with syntactic development. In addition, these younger children tend to have deficits that are quantitative (i.e. acquisition of language in appropriate developmental sequence, but slower) in nature, not qualitative (i.e. disordered language). No specific comparison between an Aboriginal population and non-Aboriginal population in terms of a language profile can be made at this time.

In order to further investigate the language differences of Aboriginal children with FASD, future research should focus on greater control of environment, culture, cognitive variability, and assessment methods. Future case control studies should include larger sample sizes to increase the power of the results. If evaluating differences across age levels, one should select larger sample sizes and a younger and older group should be selected making every effort to ensure sample sizes are relatively equal. In studies that do not evaluate language differences across age levels, cognitively matched groups would be essential to rule out language differences due to cognitive ability. This group should not be characteristic of any other disorder and should be matched on non-verbal cognitive ability to account for language differences.

Culturally sensitive assessment approaches should also be utilized to evaluate an Aboriginal population with FASD. This type of approach would be essential with this population since each Aboriginal community varies from one to the other. Differences exist in language experiences and exposure, social experiences, pragmatics, home life, and dialect. Culturally sensitive assessment methods attempt to account for these differences. For example, a dynamic assessment method that utilizes a battery of tests administered in both a standardized and non-standardized way to evaluate specific areas of language and obtain different kinds of information is a more culturally sensitive approach. Administering tests in a non-standardized manner may allow the clinician to adjust the way they administer the items so that it is more in line with cultural practices and remove items that are inappropriate for that particular culture.

Future research could also focus on case studies within a large sample and determine trends between the cases. This type of research may provide more valuable clinical knowledge than some experimental designs. Another suggestion would be to conduct multiple case control studies that replicate results across the studies (increases the power of results obtained). This type of research may be a more useful way to examine this type of population due to the diversity. An additional future research suggestion would be to conduct qualitative studies and then look for ethnographically triangulated results. This type of research can provide valuable knowledge as it focuses on observing the same results across studies that are conducted by different researchers. This type of research is critical for an Aboriginal population with FASD due to the diversity of the population.

Clinical Implications

The research from the above studies contains several weaknesses but due to the lack of research available, it can provide a speech-language pathologist a general guide in assessing an Aboriginal population with FASD. It can guide the clinician to assess general areas of possible dysfunction with more appropriate measures population. language for this unique of Recommendations that emerged from the research reviewed are to use culturally sensitive assessment methods to account for cultural differences. This is the most critical element in obtaining accurate results. This can be accomplished through informal testing and dynamic assessment methods. The clinician should also utilize a wide range of language tests that tap multiple language systems, considering the younger child with FASD tends to have more global language deficits. Tests selected for the older child with FASD should include analysis of comprehension and production of syntactic skills, since these children may have deficits in this area.

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