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Language in Children With Fetal Alcohol Spectrum Disorder

By ©

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Abstract

Language abilities were studied in eight participants with a diagnosis of FASD; all participants had age appropriate nonverbal problem-solving abilities (NVIQ > 80), and were between the ages of 8;0 and 10;0. Nonverbal problem-solving abilities were measured using the Test of Nonverbal Intelligence-3 (TONI-3); language structure and content were measured using the Clinical Examination of Language Fundamentals-4 (CELF-4), and pragmatic language was measured using the Test of Pragmatic Language (TOPL) and the Children's Communication Checklist-2 (CCC-2). The mean CELF-4 and TOPL scores were significantly below their mean TONI-3 scores, indicating that general and social language impairments are present in this population even when they do not display cognitive impairment.

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Introduction

Prenatal exposure to alcohol has a teratogenic effect on the developing fetus, altering the course of typical fetal development (Autti-Rämö, 2002; Cordero, Gerberding, & Floyd, 2004). This exposure affects each individual differently, depending on the amount and concentration of alcohol taken by the mother during the different stages of fetal development. The most extreme effects can be seen in individuals diagnosed with Fetal Alcohol Syndrome (FAS), who all exhibit delayed growth, facial dysmorphia, and CNS damage. More subtle effects of prenatal alcohol exposure also occur and can be damaging (Cordero et al., 2004). Many individuals who were exposed to alcohol *in utero* and have deficits in at least one of the main areas do not meet full diagnostic criteria for FAS. A number of terms have been used in the past thirty years to describe these conditions, such as partial FAS (pFAS), Fetal Alcohol Effects (FAE), Alcohol-Related Birth Defects (ARBD), and Alcohol-Related Neurological Disorder (ARND). Because there was no common set of criteria for diagnosing FAS and related disorders such as pFAS, FAE, ARBD etc., the term Fetal Alcohol Spectrum Disorder (FASD) is commonly used when referring to any individual affected by prenatal alcohol exposure.

The birth defects associated with FASD include behavioural, emotional, cognitive, and social dysfunctions that stay with the affected individual throughout his or her life (Canadian Center on Substance Abuse, [CCSA] 2005; Lockhart, 2001; O'Leary, 2003; Ryan, 2006; Sokol, Delaney-Black, & Nordstrom, 2003). One area of functioning that is often reported to be impaired in this population is language ability. However, little research has been done to compare how language skills in children with FASD are affected when compared to overall cognitive ability (Cone-Wesson, 2005; Sparks, 1984). Furthermore, social language skills, such as the ability to adapt one's language based on the communication partner and the context, have rarely been assessed in this population (Gentry, Griffith, Dancer, Davis, Eaton, & Schulz, 1998), despite the fact that anecdotal and clinical reports often mention that children affected by prenatal exposure to alcohol present with impaired social communication skills (Abkarian, 1992). Impaired social communication ability, if undiscovered and untreated, can affect an individual's ability to develop interpersonal relationships and this, in turn, makes it difficult to succeed in education and employment settings. It is important to understand how language abilities are affected in children with FASD so that caregivers and professionals can be more informed about their children's difficulties and deliver more effective intervention. This project examines communicative abilities in children with FASD, in an attempt to better understand the difficulties that affect this population.

Literature Review

An Introduction to FASD

Etiology of FASD

Studies have shown that 9-13% of pregnant mothers report having consumed alcohol at some point during their pregnancy, and one in eight women of childbearing age (18 through 44) reports consuming a large amount of alcohol in the past month (Cordero et al., 2004). This is a distressing statistic, as many women are likely to engage in alcohol consumption early in their pregnancy when they are not aware of their condition, and studies have shown that high blood alcohol levels during the first few weeks of pregnancy can lead to very severe problems (Autti-Rämö, 2002).

It is often reported that there is no safe amount of alcohol consumption during pregnancy (CCSA, 2005; Lockhart, 2001). However, this is not necessarily the case, and this 'misconception' is partly due to the way that alcohol consumption is reported in the literature. For instance, a woman who has one drink every day and a woman who has seven drinks in one day, once a week, can both be said to have an average alcohol consumption of seven drinks per week, but the levels of fetal alcohol exposure in each case are quite different (Armstrong & Abel, 1999). A number of studies indicate that a maternal history of 'binge' drinking – defined as five or more drinks on any one occasion – has a more pronounced effect on fetal development than do smaller doses, due to a higher alcohol concentration (Autti-Rämö, 2002; Cordero et al., 2004; Lockhart 2001). However, the same pattern and amount of alcohol exposure can lead to different outcomes in different cases, so there must be additional factors that contribute to FASD (Lockhart, 2001). A number of studies have shown that the following risk factors are often associated with a diagnosis of FASD: a pattern of repeated binge drinking, low socio-economic status (SES), smoking, malnutrition, environmental pollutants, maternal age, maternal stress levels, and the presence of other drugs (Abel & Hannigan, 1995; Armstrong & Abel, 200; Burd, Marsolf, Klug, & Kerbeshian, 2003).

Prevalence of FASD

Estimates of the prevalence of individuals with prenatal exposure to alcohol are unreliable because of sampling problems, poor access to cases, different methodological techniques, and changing diagnostic criteria over time and between countries (Cordero et al., 2004; May & Gossage, 2001; O'Leary, 2003). Canadian prevalence rates of FAS are estimated to be approximately 1 per 1000 (Warwick, 2003). The U.S. Center for Disease Control indicates that the United States has similar prevalence rates, with estimates ranging from 0.2 to 1.5 cases per 1000 births (in Cordero et al., 2004). A review of studies up to 2001 indicates that the prevalence of all FASDs in the U.S. could be close to 10 per 1000 births for FASD, depending on diagnostic methods and criteria (May & Gossage, 2001). There has been a large increase in the rate of FAS since the disorder was first described. An examination of medical records showed that prevalence rates in the U.S. rose from 1-2 per 10,000 in 1979 to 3.7 to 6.7 per 10,000 in 1992 and 1993 (May & Gossage, 2001); however, that jump is likely due to increasing awareness of the disorder, and not an increase in incidence.

Even within the United States and Canada, the prevalence rate differs across groups within the general population. Minority groups typically have higher FAS prevalence rates than Caucasians. For instance, prevalence rates of FAS among Canadian Aboriginal groups range from 7-16% (CCSA, 2005). In his summary of prevalence, Abel (1995) indicated that in the United States, Caucasian prevalence rates were 0.26 per 1000, while African-American rates were 2.9 per 1000, over ten times higher. However, these figures should not be taken as evidence that there is an ethnic predisposition for prenatal alcohol toxicity. Since members of minority groups are more likely to fall below the poverty line than Caucasians, and since low SES is highly associated with FAS, increased prevalence in ethnic minorities is likely due to environmental, rather than racial or cultural, factors (Abel, 1995).

Diagnosis of FASD

As mentioned previously, the umbrella term FASD is applied to individuals with a wide variety of conditions and disorders. Due to a lack of standard definitions, diagnostic labels were based on clinical impressions that lacked accuracy and precision. There were no objective scales to measure the key diagnostic features, and there was no 'common language' that clinicians and researchers could use when discussing FASD (Astley, 2004). In 1997, the *4-Digit Diagnostic Code* (Astley & Clarren, 2000) was created by the FAS Diagnostic and Prevention Network at the

University of Washington, in an attempt to compensate for the above weaknesses. The 4-Digit Diagnostic Code assesses each of the following 4 diagnostic dimensions: (1) growth deficiency, (2) the distinctive FAS facial phenotype, (3) central nervous system (CNS) disturbance, and (4) exposure to alcohol. Detailed descriptions of each dimension appear below; within each, a score is applied to rank the severity of impairment for the individual. A score of 1 indicates no or unlikely impairment, 2 indicates mild or possible impairment, 3 indicates moderate or probable impairment, and 4 indicates severe or definite impairment. Since there are 4 different diagnostic criteria that can be ranked from 1 to 4, the 4-digit Diagnostic System contains 256 possible 4-digit codes, which are then separated into 22 unique Clinical Diagnostic Categories. Three of these categories include codes that meet the diagnostic criteria for a diagnosis of FAS, defined as a profile with ratings of 2 or greater on all 4 diagnostic features. Other categories contain codes that do not meet criteria for FAS, but which are equivalent to the now obsolete diagnoses of FAE, ARND, and ARBD, as well as additional categories describing conditions on the spectrum that previously have not been adequately described and diagnosed (Astley, 2004).

Defining Features of FAS and FASD

Growth Deficiency

Many studies have observed some degree of growth retardation in individuals with FAS; however, the measurement criteria used in these studies has often varied, leading to inconsistent diagnoses. Areas of variability include the time of measurement, area measured (height, weight, head circumference), and cut-off level (Cordero et al., 2004). The 4-Digit Diagnostic Code measures growth deficiency by examining the patient's growth records for measures of the individual's height and weight, adjusted for age and gender. Height and weight are each ranked A, B, or C based on where the individual falls on a normalized growth curve; A represents falling above the 10th percentile and C represents falling below the 3rd percentile, while B represents falling between the 3rd and 10th percentile ranks. The codes for height and weight are then converted, using criteria specified in the manual (Astley, 2004) to a single digit from 1 to 4, where 1 indicates no growth deficiency, 2 indicates mild deficiency, 3 represents moderate deficiency, and 4 indicates severe deficiency.

The FAS Facial Phenotype

Individuals with FAS present with a specific facial dysmorphia. Prenatal alcohol exposure disrupts cellular migration along the midline of the face (Johnson, Swayze, Sato, & Andreasen, 1996), leading to a pattern of dysmorphia that has simultaneous expression of three facial features: small palpebral fissures or eye openings, which gives the appearance of 'slit eyes'; a smooth philtrum, i.e. the normally grooved region between the nose and upper lip; and a thin upper lip (Astley, 2004; Cordero et all, 2004; Sokol et al., 2003). Additional features may also be present, such as the presence of folds of skin at the corners of the eyes, a low nasal bridge, a short nose, and a small, flat midface (Cordero et al., 2004; Warren & Foudin, 2001). For the purposes of the 4-Digit Diagnostic Code, each of the three key features can either be measured directly with a ruler or indirectly measured through computerized analysis of a digital photograph. Each feature is ranked against typical growth charts and individually scored as A, B, or C in a similar manner to growth deficiency, with A being the least severe expression of the feature and C being the most severe. These letter scores are then converted into a single digit representing FAS Facial Features in the 4-Digit Diagnostic Code, using criteria specified in the manual (Astley, 2004). Consistent with the growth deficiency dimension, 1 represents no expression of the features, 4 represents severe expression, and 2 and 3 represent mild and moderate expression, respectively.

CNS Abnormalities

The central nervous system is particularly sensitive to alcohol exposure, leading to cognitive and behavioural problems that can adversely affect the exposed individuals throughout their lives (Cordero et al., 2004; O'Leary, 2003). Prenatal exposure to alcohol can lead to a number of structural, functional, or neurological abnormalities, depending on the amount of alcohol consumed and the timing during pregnancy.

For this dimension of the 4-digit code, a rating of 4 means definite structural and / or neurological abnormality, and is given when the individual presents with clear abnormalities determined by direct observation or imaging techniques (Astley, 2004). In the case of an individual without definite CNS damage, ranks from 1 to 3 are used to indicate *both* the likelihood of CNS damage and the level of CNS dysfunction. Likelihood of damage / dysfunction is indicated by standardized assessment tools or clinically significant information that has been interpreted by appropriate professionals. Each level of CNS dysfunction is described below.

As previously mentioned, a rating of 4 ("definite" CNS damage) is given if there is documented CNS damage from at least one clinically significant piece of structural or neurological evidence. Examples of adequate structural evidence are: 1) microcephaly, or diminished overall head circumference of 2 or more standard deviations below the norm, and 2) significant neural abnormalities (discernible through imaging techniques) of pre-natal origin. Examples of adequate neurological evidence are: 1) seizures that cannot be attributed to postnatal influences such as head trauma or disease, and 2) other 'hard neurological signs' (e.g., cranial nerve problems leading to blindness or deafness, etc.).

A rating of 3 (significant dysfunction and "probable" CNS damage) is indicated when standardized tests or clinically significant information indicate significant impairment in three or more of the following domains of brain function: cognition, academic achievement, adaptive behaviour / social skills, motor / sensory integration, attention, behavioural problems, psychiatric conditions, and language / social communication. Significant impairment is generally defined by performance at least two standard deviations below the norm on a standardized test. A rating of 2 (mild to moderate dysfunction and "possible" CNS damage) is indicated in two cases: 1) those who have not yet had the necessary testing to confirm or rule out a rating of 3, typically because they are too young to be tested by the necessary standardized assessment tools or 2) those who have evidence of some brain dysfunction, but in less than 3 of the above areas of function or at a level that is less severe than 2 standard deviations below the norm. Rank 1 classification (no dysfunction and no CNS damage) is given to patients when no observable functional problems are found.

Alcohol Exposure

The final diagnostic feature used when diagnosing FASD is maternal alcohol exposure, rated according to the quantity, timing, frequency, and certainty of exposure (Astley, 2004). A rating of 4 indicates confirmed exposure to high levels of alcohol early in pregnancy, i.e. a blood alcohol concentration greater than 100 mg/dL (the equivalent of 6-8 servings of alcohol) weekly. A rating of 3 indicates confirmed exposure to alcohol, but the amount is either unknown or at lower levels than necessary for a rating of 4. A rating of 2 indicates unknown exposure, neither confirmed absence nor confirmed presence of alcohol, such as would be found in cases of adoption with closed records, or when the mother has been known to drink but was never observed drinking during pregnancy. Finally, a rating of 1 indicates confirmed absence of exposure, and is relatively rare in the general population.

Summary of the Diagnostic Criteria

Once all the diagnostic criteria have been assigned a diagnostic rank, they are combined to make a 4-Digit Diagnostic Code. As mentioned earlier, there are 256 possible codes, grouped into 22 Diagnostic Categories, as indicated in the 4-Digit Diagnostic Code manual (Astley, 2004). For example, an individual with moderate growth deficiency (3), severe facial dysmorphia (4), definite structural CNS damage (4), and confirmed alcohol exposure (4) would have the code 3444, which corresponds to Diagnostic Code A – Fetal Alcohol Syndrome (alcohol exposed). A patient with severe growth deficiency (4) and FAS facial phenotype (4), but only mild CNS dysfunction / "possible" CNS damage (2) and unknown alcohol exposure (2) would have the code 4422, which corresponds to Diagnostic Code M – Sentinel physical finding(s) / neurobehavioural disorder (alcohol exposure unknown). At the Glenrose Rehabilitation Hospital, where participants for this study were recruited, the term FASD is used when a patient presents with confirmed alcohol exposure (Rank 3 or 4) and a level of brain dysfunction ranging from mild to severe (Rank 2 to 4) that cannot be explained by other genetic, prenatal, or postnatal factors (e.g. poor nutrition, trauma, etc.; Andrew, personal communication). Using these criteria, FASD is indicated for 102 Diagnostic Codes, or 7 of the 22 Diagnostic Categories.

The 4-digit method of diagnosis shows high degrees of intra-rater and inter-rater reliability. When the authors of the code examined 20 randomly selected patients that had been previously diagnosed using this method and re-diagnosed them up to 4 years later, the new codes for all 20 patients matched the previous codes, showing inter-rater and intra-rater reliability of 100% (Astley & Clarren, 2000). In a second study, when records for 16 patients were compared between regional clinics and the University of Washington clinic, the authors found an exact 4-digit Diagnostic Code match for 15 out of the 16 patients, and a Diagnostic Category match for all 16 patients, giving inter-rater reliability measures of 94% and 100% respectively (Astley & Clarren, 2000).

Language and Communication in FASD

Most of the research regarding children with FASD is concerned with medical complications and general cognitive abilities: the language abilities of individuals in this population have received comparatively little attention. Although there is general consensus that individuals with FAS typically present with some language difficulties, language abilities within the wider umbrella of FASD are not as well-established.

Language Ability in Fetal Alcohol Syndrome

Language ability is typically described in one of two ways. Firstly, language can be separated into the three domains of form, content, and use. Language form describes how the language is put together, and includes phonology (the sounds of a language and the rules that govern how they can be combined), morphology (the way in which the words of a language are constructed through roots and affixes, e.g., the root "*run*" can be combined with the suffix "*-s*" to form "*runs*"), and syntax (the rules that control the way in which sentences are constructed). Language content consists of semantic knowledge; that is, the meaning of words and sentences. Language use is termed pragmatics, and refers to how language is used in social contexts to achieve different goals, such as requesting desired objects and sharing feelings, and to manage interactions, e.g., a child asking his mother politely for a cookie while saying "Gimme!" to a peer. Another way to describe language is to divide it into receptive and expressive components. Receptive language describes an individual's ability to understand language, while expressive language describes an individual's ability to generate language.

These two ways of describing language are often combined: for example, one can have appropriate syntactic skills receptively but difficulty with expressive syntax. Most of the studies of children with FAS that looked at language ability have shown that these individuals present with deficits in language form and content, in both expressive and receptive domains (Becker, Warr-Leeper, & Leeper, 1990; Carney & Chermak, 1991; Church, Eldis, Blakley, Bawle, 1997; Gentry et al., 1998; Janzen, Nanson, & Block, 1994), but very few have examined language use or pragmatics.

In 1990, Becker and colleagues compared the language abilities of six Native Canadian children with FAS, ages 5;6 (5 years, 6 months) to 9;6 (9 years, 6 months) to six matched control participants. The participants were matched on nonverbal cognitive ability as measured by the Raven's Coloured Progressive Matrices (CPM) and family situation (adoptive home or foster care). They were individually given a large battery of standardized tests of both language form (syntax) and content (semantics). The results showed that the participants had delays in both grammar and semantic ability relative to their chronological age. However, all the FAS participants had exposure to multiple living environments and exposure to another language (Ojibway) during early development. These factors may have negatively affected their test scores, making it difficult to attribute poorer language skills to FAS.

Carney and Chermak (1991) studied 10 Native Canadian children with FAS who had a mean age of 8;3 (SD 2;3) and compared them to 17 typically developing culturally matched peers with a mean age of 9;2 (SD 3;5). The mean IQ of the FAS participants, as measured by the Wechsler Intelligence Scale for Children-Revised

(WISC-R), was 79; the IQ scores of the control participants were not measured but reported to be appropriate to their grade level. The participants' language abilities were measured on the Test of Language Development-Primary (TOLD-P) or Intermediate (TOLD-I), depending on their chronological age. Composite scores for all FAS participants were lower than their non-FAS peers. This study shares the same language and home environment limitations as Becker et al. (1990), but is also limited by the possible difference in cognitive ability between the two groups. If the IQ measurements of the participants with FASD are accurate, they had a mean IQ of 79, indicating at least some cognitive impairment, while the children in the control group were reported to have age-appropriate cognitive abilities. In this case, lower language scores could be the result of lower levels of cognitive ability. On the other hand, for a person who may have language delay, measuring IQ with a verbally biased test such as the WISC is inappropriate and likely to underestimate cognitive ability, so it is difficult to have confidence in the IQ measures that are reported. Furthermore, there is a one-year difference between the mean ages of the two groups. Thus, in this study, we are unable to tell whether the comparisons are made between equivalent populations.

As part of a general study of neuropsychological ability in preschoolers with FAS, Janzen and colleagues (1994) compared ten children with FAS (age 3;6 to 5;0) to ten typically developing participants matched on age, gender, ethnicity, and SES (in this case, lower-middle class). In each of the two groups, eight of the participants were Native Canadian, and the remaining two were Caucasian. The Test of Early Language Development (TELD; Hresko, Reid, & Hammill, 1981) was used to measure receptive and expressive language abilities in both the form and content domains. The results showed that the FAS group scored significantly lower on the

TELD than the matched control participants. However, since a mental-age control group was not included in this study, it is difficult to know if the language delays in the FAS participants were due to the effects of prenatal exposure to alcohol on the developing brain, or general developmental delay.

Church et al. (1997) conducted a study of the hearing, vestibular, dentofacial, and speech / language deficits of 22 individuals with FAS aged 3;8 to 26;11. In terms of language skills, the participants completed a battery of standardized tests that measured their receptive and expressive language abilities, as well as a comprehensive measure of language ability. The results of this study indicated that 82% of the participants had deficits in receptive language and 76% had deficits in expressive language. However, since there was no measure of non-verbal IQ, it is not clear which or how much of the language deficit was due to general developmental delay, i.e., participants may have had language skills commensurate with their general cognitive skills. Additionally, the wide age range of the participants in this study makes it difficult to generalize the results.

Finally, Gentry and colleagues (1998) described the communication, behaviour, and intelligence of three children diagnosed with FAS. The participants' language abilities were measured using the Clinical Evaluation of Language Fundamentals-3 (CELF-3), which measures receptive and expressive language form and content, and the Test of Pragmatic Language (TOPL). The participants were also given the Test of Nonverbal Intelligence (TONI) to obtain a nonverbal intelligence score. On the TONI, the first two participants scored 111 and 105, indicating average intelligence, while the third participant scored 69, indicating moderately delayed nonverbal intelligence. Scores on the language tests were markedly lower. The third child exhibited severe delays on both language tests. The other two participants scored in the low-average range on the CELF-3 (88 and 85), and the second participant also scored in the low average range on the TOPL (88). Even though these scores were still in the normal range, they appear delayed compared to their nonverbal intelligence scores. The small sample size limits the generalizability of these findings. However, this study is notable for the inclusion of a pragmatic language measure, something that none of the previously mentioned studies included in their measures of language ability in FAS. Furthermore, as this study compared the participants' language abilities to their non-verbal cognitive abilities, it was able to indicate how much of their language impairment might be explained by general cognitive skill.

Language Ability in FASD

Despite the weaknesses identified in the above studies, there is a consensus that individuals with a diagnosis of FAS generally present with some kind of language dysfunction. There is much more uncertainty as to whether or not prenatal alcohol exposure can lead to language disorders in individuals without a diagnosis of FAS but with a diagnosis of FASD (Mattson & Riley, 1998). Some studies find no evidence of language impairment in individuals with FASD (Greene, Ernhart, Martier, Sokol, & Ager, 1990; Kaplan-Estrin, Jacobson, & Jacobson, 1999; Nulman, Rovet, Kennedy, Wasson, Gladstone, Fried, & Koren, 2004), while others indicate that this population may indeed present with some language difficulties (Russell, Czarnecki, Cowan, McPherson, & Muldar, 1991; Streissguth, Barr, Olson, Sampson, Bookstein, & Burgess, 1994).

Greene and colleagues (1990) examined children from 369 mothers who had a risk of alcohol-related problems. Language development in these participants was measured: (1) by administering the Sequenced Inventory of Communication

Development (SICD), a test that evaluates language acquisition and development, at ages 1, 2, and 3, and (2) by taking a language sample when the participants were 2 years of age. In this study, the children's language abilities were more closely correlated to the effect of a disadvantaged home environment than to maternal drinking. They concluded that there was little support for an effect of FASD on language development, except perhaps in a small number of cases or extremely severe cases, such as those with Fetal Alcohol Syndrome.

Another study by Kaplin-Estrin et al. (1999) examined language development in 92 participants, at ages 1;1 and 2;2. The participants did not have a diagnosis of FAS, but, based on their mother's interviews, were at risk for other effects of prenatal alcohol exposure: their mothers reported absolute alcohol consumption rates of at least 0.5 oz per day at conception. Two language-specific measures were used as part of the study: the MacArthur Communication Development Inventory-Words (CDIW), a parental checklist of the child's expressive vocabulary, and the Noncanonical Commands Test, which assesses comprehension of multi-word utterances. Although the participants scored lower than their age norms on the CIDW, there were no significant relationships between drinking during pregnancy or at conception and either of the language scores.

In 2004, Nulman et al. studied 51 children whose mothers sought counselling for binge drinking during pregnancy and then compared them to a matched control group of children. While the participants' ages were not specifically reported, the authors noted that most of the participants were "relatively young". Language ability in the participants was measured using the Reynell Developmental Language Scales, which measure both receptive and expressive language in children aged 15 months to 7 years 6 months. The study showed that the two groups of children scored comparably on the test of language, but that subtle behavioural changes (including social interaction) characterised the group with FASD.

In each of these three studies it was suggested that in the absence of FAS, maternal alcohol consumption had little effect on their children's language ability. On the other hand, the participants in this study were "relatively young", and the literature indicates that the impact of FASD becomes more evident as children grow older (Nulman et al., 2004). It is possible that due to the participants' young ages, the language abilities most severely affected by FASD had not yet developed, and therefore the study could not have measured the more severe effects of maternal alcohol consumption on language skill.

On the other hand, other studies have indicated that individuals with FASD have impaired language abilities. Russell et al. (1991) conducted a prospective study of 186 children; they found that maternal drinking during pregnancy was associated with lower language abilities in children. In this study, mothers were administered questionnaires that yielded two measures of alcohol consumption: the Prior to Pregnancy Absolute Alcohol per day (PPAA), and Indications of Problem Drinking (IPD). These measures were then compared to measures of their children's growth, dysmorphology, and cognitive development. Receptive language ability in the child participants was measured by the Token Test, which measures receptive syntax, and measures of Verbal and Performance IQ were assessed using the Wechsler Preschool and Primary Scale of Intelligence (WPPSI). Children whose mothers were identified as problem drinkers (IPD>1) had significantly lower Verbal IQ and Token Test scores.

Another prospective study by Streissguth and colleagues (1994) examined over 500 children born to mothers who abused alcohol during pregnancy. They found that measures of alcohol abuse, particularly measures of binge drinking, were significantly negatively correlated (ranging from -.0677 to -.1513) with Word Attack scores from the Woodcock Reading Mastery Tests (WRMT). Both of these studies had large sample sizes, making their conclusions more credible than some of the previous studies. However, the measures of language ability were limited: in the study by Russel et al. (1991) only receptive syntax was examined, and in the study by Streissguth et al. (1994), only Word Attack. Overall, research on language difficulties in FASD has been both limited and inconclusive; some studies seem to indicate that there is no evidence of language delay or dysfunction in individuals within the broader umbrella of FASD, while others support the opposite conclusion.

Pragmatic Language in FASD

Language pragmatics have been studied very little in individuals with FASD. Only one study (Gentry et al., 1998) attempted to measure overall levels of pragmatic language in children with FASD, using a standardized measure. This is unfortunate, considering that individuals with FASD have been reported to have impaired pragmatic ability. Even when they display fluent grammatical and semantic language abilities, they have been described as "intrusive, loquacious, and over-inquisitive", all of which indicate difficulty with appropriate pragmatic language use (Abkarian, 1992). Shaywitz, Caparulo, & Hodgson, (1981) described two children (ages 5;11 and 5;4) with prenatal exposure to alcohol but without a diagnosis of FAS and found that they displayed socially inappropriate interpersonal skills and poor discourse abilities. Older children with FAS show difficulty in planning and telling oral narratives. Compared to chronologically-age-matched peers, their narratives lack cohesion, coherence, and key story elements (Coggins, Friet, & Morgan, 1998). These individuals also have difficulties with other social behaviours, including attachment, adaptive functioning, and socialization (Kelly, Day, & Streissguth, 2000; Nulman, 2004; Olswang, 1998; Timler & Olswang, 2001). Even though pragmatic language deficits have been reported anecdotally in this population, these abilities have rarely been studied systematically. Because so few studies have focused on pragmatic language abilities in FASD, there is a lack of evidence documenting possible impairments in the social communication skills of these individuals. Since pragmatic language is crucial for group interaction and social functioning, there could be serious consequences if these impairments are overlooked in clinical settings.

In most of the studies reviewed thus far, language has been assessed using standardized tests. These formal tests are typically used for assessing language form and content, but do not focus on language use or pragmatic language skills. There are many reasons for this. Because of its fluid nature, pragmatic ability is difficult to quantify, and there are limited data on developmentally appropriate pragmatic norms (Adams, 2002). Furthermore, successful use of pragmatic language skills involves changing one's behaviour across situations. Because standardized tests are administered in a single, structured situation, they are often unable to obtain a satisfactory measure of more complex, context-based behaviours. In addition, the increased structure of the formal setting and concrete instructions given to individuals during standardized tests can lead to higher, possibly inflated, performance when compared to information gathered from a naturalistic setting (Bishop, 1998).

Despite the limitations of using standardized tests to assess pragmatic language ability, some formal measures have been developed. These assessment tools have typically focussed on the assessment of skills that are acquired through the course of normal development. One such instrument is the Test of Pragmatic Language (TOPL; Phelps-Terasaki & Phelps-Dunn, 1992). The TOPL assesses a child's pragmatic language ability by sampling for typically developing pragmatic skills. The child is shown a series of pictures and is asked to either supply an appropriate statement for the situation or to explicitly state the implied meaning of an utterance. For example, one picture shows Tom and Mike walking along a sidewalk where Tom does not see a patch of wet cement and is about to step in it. Mike says to Tom, "I guess you like walking in wet cement", and the child is asked what Mike really meant by what he said. An example of a correct response is "Watch out for the wet cement!", while an incorrect response might be "Tom likes walking in wet cement." Correct answers are scored as 1, while incorrect answers are scored as 0.

The TOPL has been shown to be effective in separating clinical groups from typically developing children (Young, Diehl, Morris, Hyman, & Bennetto, 2005), but, as a standardized test, still has a number of limitations. Because the child is judged as to whether he or she provides the same type of appropriate responses as other children of the same age, the TOPL measures *quantitative* differences in pragmatic ability; that is, whether or not an individual exhibits the same behaviours during social communication as typically developing children, such as initiating conversations or turn-taking. However, some clinical populations can exhibit *qualitative* differences in pragmatic ability (such as bizarre utterances, e.g. describing a triangle filled with dots as "horse spotted") that typically developing children do not (Bishop, 1998). The TOPL does not measure such differences in social communication, and therefore these salient behaviours would be unaccounted for when using this assessment tool. Additionally, the dichotomous scoring system does not take into account varying degrees of quality in response (Young et al., 2005). Finally, the high degree of structure and concrete instructions that are involved in administering the TOPL could lead to a higher performance that is not reflective of their general level of skills (Adams, 2002).

The Children's Communicative Checklist-2 (CCC-2; Bishop, 2003) is a parent checklist that is designed to alleviate the above difficulties. It is used both as a screening measure for language form and content, and to detect pragmatic impairment in children aged 4 and older who speak in multi-word sentences. Parents are asked how often (less than once a week or never, at least once a week but not every day, once or twice a day, several times a day or always) their child engages in certain communication behaviours, some of which are typical for children of that age and some of which are not. The CCC-2 has a number of advantages over formal standardized assessment tools such as the TOPL. Firstly, by using someone who is familiar with the child's language ability as an informant, it is more likely that the information gathered will accurately reflect the child's typical level of functioning. Secondly, it avoids the problems associated with measuring context-dependent behaviour in a single standardized setting, and it provides a more comprehensive picture of an individual's pragmatic language ability, by having parents rate their child's language in their natural environment (Adams, 2002). Thirdly, the CCC-2 takes less time to complete and score than a more traditional standardized test. Lastly, because the checklist samples both typical and atypical behaviours, the CCC-2 takes into account both *qualitative and quantitative* pragmatic irregularities. The CCC-2 has been shown to successfully separate a number of clinical subgroups, including Specific Language Impairment (SLI), Pragmatic Language Impairment (PLI), and Autism Spectrum Disorder (ASD) from typically developing controls (Norbury, Nash, Baird, & Bishop, 2004).

Summary

Overall, language problems are commonly reported in individuals with FASD; however, comparatively little research has been done that focuses specifically on language and communication skills relative to other cognitive domains (Abkarian, 1992; Cone-Wesson, 2005; Sparks, 1984). Furthermore, it is difficult to draw any strong conclusions from what little research has been done because of changes in diagnostic criteria, small sample sizes, and variations in the age and ability of the participants among the studies (Abkarian, 1992). Lastly, as social communication skills have been assessed in only one study (Gentry et al., 1998), there is almost no empirical information about pragmatic language in individuals with FASD.

Purpose

This research study focused on a number of questions about language and communication impairment in individuals with FASD. There is a general agreement in the literature that children with FAS demonstrate language impairment, but many of the studies are limited by the absence of adequate cognitive assessments. Participant IQ was not consistently measured or reported, but when it was, it was often based on assessments relying on verbal skills. For children who have language impairments, such tests may underestimate their cognitive abilities because they lack the ability to frame their responses appropriately or because they misunderstand the question, rather than because they have reduced cognitive skill. Only one study (Gentry, 1998) measured nonverbal cognition and compared language test results to this more appropriate cognitive measure, but Gentry's sample was extremely small (n=3).

The participants in this study were all selected to have nonverbal IQs greater than or equal to 80 (as assessed by the Test of Nonverbal Intelligence-3, or TONI-3, Brown, Sherbenou, & Johnsen, 1997) in order to assess whether language and communication impairment existed in the presence of typical or near-typical cognitive skills. In addition, some authors have indicated that pragmatic language ability in this population may be delayed relative to structural language (Abkarian, 1992; Coggins et al., 1998), so the pragmatic domain was a particular focus.

The first research question this study addresses is:

1. How does the performance of high-functioning (non-verbal IQ $\geq$ 80) children aged 8;0 to 10;0 with FASD on (1) a standardized test of language structure and content (i.e., the CELF-4) and (2) a standardized test of pragmatic language (i.e., the TOPL) compare to their nonverbal IQ?

Based on Gentry's (1998) findings, we expected that participants' scores on the CELF-4 and the TOPL would be lower than their nonverbal IQ, indicating language and pragmatic deficits that exist beyond those that are attributable to general cognitive delay.

The second research question this study addresses is:

2. Are there significant differences between language form and content (as indicated by quotient scores on the CELF-4) and pragmatic language (as indicated by quotient scores on the TOPL) in high-functioning (non-verbal IQ > 80) children aged 8;0 to 10;0 with FASD?

Based on the anecdotal reports of pragmatic language impairment in individuals with FASD even when their structural language abilities are normal, we expected that pragmatic language quotients as measured by the TOPL would be significantly lower than measures of language structure and content, as indicated by quotients on the CELF-4.

The third research question this study addresses is:

3. Does the CCC-2 detect pragmatic language impairment in a greater proportion of high-functioning (non-verbal IQ > 80) children aged 8;0 to 10;0 with FASD than does the TOPL?

Because the CCC-2 is designed to detect both quantitative and qualitative deficits in pragmatic language, we hypothesized that it would identify pragmatic language impairment in a higher proportion of the participants with FASD than would the TOPL.

Methods

Participants

We attempted to recruit twenty participants for this study. Initially, staff at the Glenrose Rehabilitation Hospital provided contact information for the guardians of fourteen possible participants. Contact information was out of date for two of those participants, leaving a pool of 12. The guardians of three participants declined to have them participate; one other participant showed initial interest in participating but continually had to cancel or re-schedule appointments, and so was unable to be involved. In the end, eight participants (6 M, 2 F) were recruited. All eight were diagnosed at the Glenrose Rehabilitation Hospital as having one of the seven Diagnostic Categories that fall under the FASD spectrum. All participants were between the ages of 8;0 and 10;0 to ensure that all participants met the criteria for administration of the diagnostic tools and in an attempt to optimize homogeneity. All eight of the participants had a non-verbal IQ higher than 80, as measured on the TONI, to ensure that any significant language delay discovered would not be due to a significant cognitive delay. Participants were excluded from the study if they had a history of hearing loss or if English was not their primary language. Two of the participants lived in adoptive homes, while the rest were living with foster families; all of the participants' had been living in their current home environment for at least two years. Two of the participants were fraternal twins.

Procedures

After explaining the study to the guardian and obtaining informed consent (see Appendices 1 and 2) from the participant's legal guardian, a battery of four assessment instruments was administered in the child's home environment. Tests were administered in the home in order to put the child at ease and avoid the effects of an unfamiliar testing environment. Most of the testing took place in one session that typically lasted between two and two-and-a-half hours; the twin siblings completed the assessments in two one-and-a-quarter hour sessions by their foster mother's request. It has been reported that this population sometimes presents with attentional problems (Coles, 2001; Lockhart, 2001), so in order to reduce participant fatigue and increase compliance, the subjects were allowed to take breaks between tests and between different sections of the tests as needed. All children were instructed to indicate their desire for a break when they felt they needed one, and all of them took advantage of this arrangement. The appointments were arranged at the convenience of the child's family, and all measures were administered by the author, a trained speech-language pathologist.

The CCC-2 was delivered to the caregiver at the time of the assessment; instructions on how to complete the response form were provided by the author, and in most cases the forms were completed while the participants were completing the standardized tests. In the cases where the caregivers were unable to complete the

CCC-2 during the assessment, the guardian and the author made arrangements to collect the forms at a later date. Seven of the eight caregivers completed and submitted their CCC-2 forms: repeated attempts to contact the eighth and final caregiver met with no success. The completed CCC-2 forms were then scored by the author.

Measures

A measure of the participants' nonverbal IQ was obtained by administering the Test of Nonverbal Intelligence-3 (TONI-3; Brown et al., 1997). The TONI-3 measures cognitive performance by assessing abstract reasoning and problem-solving skills using nonverbal content. It can be used with subjects from 6;0 through 89;11 and consists of a number of stimuli, each depicting a pattern of geometrical shapes with one shape missing and an array of options. The subject is asked to select from the array the shape that would best complete the pattern. Test administration requires approximately 15 minutes. Raw scores were converted to quotient scores (mean=100, SD=15). The TONI-3 was normed on 3451 people in the U.S. from a representative sample and demonstrated high internal consistency ($r = .89$ and .90 between alternate test forms). Inter-scorer reliability was calculated at .99. Validity was measured by correlating the TONI-3 with measures of school achievement and positive relationships were reported (coefficients to .76).

Language form and content were measured by the Clinical Examination of Language Fundamentals-4 (CELF-4; Semel et al., 2003). The CELF-4 has been normed for use with individuals from ages 5 to 21 and is widely used to assess language ability in both clinical and research contexts. The CELF-4 has two different test forms: Form A is used with participants from 5 to 8 years of age and Form B with participants from 9 to 21. The test consists of a number of subtests that can assess language form and content, in both receptive and expressive domains. Four subtests combine to give a Core Language Score (CLS), which is used to indicate whether or not there is a language disorder. For Form A, the subtests that make up the CLS are Concepts & Following Directions, Word Structure, Recalling Sentences, and Formulating Sentences; when calculating the CLS for Form B, the Word Structure subtest is replaced with Word Classes: Total. CLS scores are comparable between the two different forms (A and B). The composite scores are expressed as a quotient score (mean=100, SD=15). The total time needed for administration of the test is approximately 45-60 minutes. Reliability and validity of the CELF-4 was established using 320 subjects from the standardization sample. In this smaller sample, corrected test-retest stability ratings of the different subtests across all ages ranged from adequate to excellent (from .78 to .90) and stability ratings of the composite scores were also excellent (from .88 to .92). Furthermore, inter-rater agreement for trained raters ranged between .88 and .99, and average standard error of measurement (SEM) ranged from 3.22 to 5.46 on composite scores. Measures of subtest intercorrelation show that the CLS has a high correlation with most of the language indices and subtests, indicating a good degree of internal validity (Semel et al., 2003).

Two instruments were used in this project to study pragmatic language skill. The first was the Test of Pragmatic Language (TOPL; Phelps-Terasaki & Phelps-Dunn, 1992). The TOPL assesses pragmatics by sampling a range of typically developing pragmatic abilities. It was normed on 1016 typically developing children between the ages of 5 and 13. During administration of the test, a child is shown pictures of different social situations, and is asked to provide a response or interpret the meaning of a statement for one of the individuals in the picture. A pragmatically correct response is scored as 1, while a pragmatically incorrect response is scored as

0. Raw scores were converted to a quotient score (mean=100, SD=15). A trained professional can administer the test in 30-45 minutes. A measure of inter-rater reliability, calculated by the two authors independently scoring 30 students' test results, yielded a coefficient of .99. Teacher ratings of pragmatic language skill were correlated with TOPL students for children in the standardization sample and yielded criterion-related validity of .82.

The second assessment tool that was used to measure pragmatic language skills in this study is the Children's Communication Checklist-2 (CCC-2; Bishop, 2003, 2006). The CCC-2 is a parent checklist that is designed to detect and analyze both qualitative *and* quantitative pragmatic impairment in children aged 4 and older who speak in multi-word sentences. It was first designed for use in the United Kingdom, but was later standardized for use in the United States in 2005 using a sample of 950 children (Bishop, 2006). The checklist consists of 70 multiple-choice items from 10 different scales. The first four scales sample traditional structural language skills (A-D: Speech, Syntax, Semantics and Coherence), while the remaining scales sample for pragmatic abilities that are difficult to sample through standardized tests (E-J: Initiation, Scripted Language, Context, Nonverbal Communication, Social Relations, and Interests). Each item rates the frequency of occurrence of a given behaviour, ranking "less than once a week (or never)" as 0, and "several times (more than twice) a day (or always)" as 3. Two composite scores can be obtained using the CCC-2. The General Communication Composite (GCC) is calculated by summing the scaled scores of the first eight subtests (Speech, Syntax, Semantics, Coherence, Initiation, Scripted Language, Context, and Nonverbal Communication) and converting that to a standard score, and is designed to identify any possible communication problems. The Social Interaction Difference Index

(SIDI) is calculated by taking the sum of the standard scores of the first four subtests (those that deal with traditional structural language skills) and subtracting it from the sum of the standard scores for Initiation, Nonverbal Communication, Social Relations, and Interests. Thus, the SIDI indicates disproportionate pragmatic language impairment, i.e. pragmatic impairment that is not attributable to structural or semantic language delay. The raw GCC score can be converted to a quotient score (mean=100, SD=15). Clinically, the SIDI scores are used in conjunction with the GCC scores to indicate whether or not the child may have pragmatic language difficulties; however, this process does not easily lend itself to statistical or research analyses. For this study, a conservative interpretation of SIDI scores was applied where SIDI scores of less than zero were interpreted as indicating pragmatic impairment. A negative SIDI indicates that a child's social communication abilities are poorer than his general communication abilities. The CCC-2 can take up to 15 minutes for parents to complete, and another 15 minutes to score. Test-retest reliability data for the U.S. version were obtained by administering the CCC-2 to the parents of 98 children, and was found to be between .86 and .96. Across the entire standardization sample of 950 children, internal consistency was excellent (between .94 and .96) for the GCC. Validity data were obtained by administering the CCC-2 to 134 children with communication problems as well as 20 typically developing control participants. There were significant differences between the clinical groups and the control group on all 10 subscales, and there was very little overlap between the clinical groups and the control group on the GCC. Comparing parent and professional ratings on the children's SIDI scores yielded an inter-rater reliability of .79.

Results

Data Analysis

First, quotient scores for all tests for each participant are shown in Table 1. Second, means and standard deviations for the quotient scores on the CELF-4 CLS, the TOPL, and the TONI were displayed in a box-plot graph and are shown in Figure 1; the means and standard deviations for the GCC of the CCC-2 were displayed with the CELF-4 CLS in Figure 2. Visual inspection was used to assess differences between the participants' language test scores and their NVIQ, as well as comparing their scores to the chronological age-expected scores of 100 (SD =15) on the two language tests. Across all measures, a cut-off standard score of 80 was used as the level at or below which a person's skills would be considered impaired. This is consistent with standard definitions of what constitutes a language impairment (1.25 SDs below the mean; Paul 2001), with the minimum NVIQ established for inclusion in the study, and is very close to the level recommended (79) by the authors of the TOPL (Phelps-Terasaki & Phelps-Dunn, 1992). In the current data, results were the same whether a score of 80 or 79 was used as the cut-off for the TOPL.

In order to answer the first research question (whether or not the participants' CELF-4 and TOPL scores differed from their nonverbal IQ scores), visual, descriptive, and parametric statistical analyses were used. First, the participants' scores were plotted on box-plot graphs in order to see if there were any immediately noticeable differences in the scores. Second, paired sample t-tests were carried out to see if there were significant differences between the participants' TONI scores and their CELF-4 scores and TOPL scores. Third, descriptive analysis of the participants scores were briefly summarized.

In order to answer the second research question (whether there were significant differences between the subjects' CELF-4 and TOPL quotient scores), the participants' standard scores for the two tests were compared through a paired-sample t-test in order to determine if the mean scores were significantly different. The same analysis was performed to see whether there were any significant differences between the subjects' CELF-4 and CCC-2 GCC scores.

In order to answer the third research question (if the CCC-2 detects pragmatic language impairment in more subjects than the TOPL), the number of subjects with pragmatic language impairment as indicated by an SIDI score of less than zero on the CCC-2 were compared to the number of subjects with pragmatic language impairment as indicated by the TOPL (quotient score less than or equal to 80) in a 2x2 matrix, to see whether different numbers of participants were judged to have pragmatic language impairment on the two assessments.

Research Findings

The means and standard deviations for all the participants' test results are reported in Table 1; each participant's individual results are reported in Table 2. Based on the results of the TONI-3, no participants fell below the inclusion criteria of 80 as shown in Table 2; i.e., none of the participants presented with nonverbal cognitive impairment. However, general language impairment (as measured by the CELF-4 CLS) and pragmatic language impairment (as measured by the TOPL) were discovered in 62.5% (5 out of 8) of the participants. The mean standard scores of the TONI-3 and CELF-4 CLS have a difference of over 15 standard score points (TONI: 95.38, CELF-4: 78.38; see Figure 1), which is statistically significant ($t(7) = 2.808$, $p=.026$), and the effect size is large (d=0.993). Thus, this group of children with FASD presented with greater language impairment than would be expected from their age-appropriate nonverbal cognitive abilities. The mean standard scores of the TONI-3 and TOPL have a difference of over 20 standard score points (TONI: 95.38, TOPL: 73.75; see Figure 1), which is also statistically significant ($t(7) = 3.131$, $p = .017$), and again, the effect size is large ($d = 1.107$). Thus, these children with FASD also presented with impairment in social communication that was greater than would be expected from their age-appropriate cognitive abilities. Overall, this suggests that children with FASD present with language and pragmatic deficits that are beyond those that are attributable to general cognitive delay.

Also of interest was the profile of subtest performance on the CELF-4. As shown in Figure 3, when the profiles of subtest scores for the seven participants who were eligible to use Form B of the CELF-4 were examined, all seven participants scored at or below the 16^{th} percentile rank on the Formulating Sentences subtest (a task that examines expressive language), and at or below 9^{th} percentile rank on the Concepts & Following Directions subtest (a task that examines receptive language).

To determine whether pragmatic skills were more impaired than structural and semantic skills, we examined the CELF-4 CLS quotients to the TOPL quotient scores. Half the participants presented with *both* general and pragmatic language difficulties (see Table 2). The mean difference between CELF-4 CLS and TOPL scores is 4.63, which is not a statistically significant difference ($t(7) = 1.04$, $p = .334$; see Figure 1). Thus, contrary to expectations, these children with FASD did not demonstrate greater pragmatic deficits than those in language form and content.

To determine whether the CCC-2 was a more comprehensive test of pragmatic language than the TOPL, we examined whether it identified a greater proportion of participants as demonstrating language impairment. As shown in Table 3, the TOPL and the CCC-2's SIDI each indicated that four participants (of the seven with complete CCC-2 data) presented with social communication difficulties. Therefore, contrary to expectations, the CCC-2 did not identify a greater proportion of participants with pragmatic impairment than the TOPL. In addition, each test identified somewhat different people as pragmatically impaired.

On the other hand, for the seven children with complete CCC-2 data, their mean standard scores on the CELF-4 CLS and CCC-2 GCC had a difference of less than 3 points, and the difference is not statistically significant ($t(6)=.563$, $p=.581$; see Figure 2). This indicates that both the CCC-2 and the CELF-4 detect general language impairment in this population.

Discussion

Participants in this study were specifically chosen to have typical or close to typical levels of nonverbal cognition in order to ensure that any language deficits could not be attributable to low IQ. Results indicated that the participants exhibited structural and pragmatic language impairments despite their typical nonverbal cognitive skills. Thus, our results suggest that children with FASD display language and communication deficits beyond what could be accounted for by their level of cognition.

Although the scores on most of the CELF-4 subtests covered a wide range of scores, there were two subtests that were difficult for all of the participants. The first was Concepts & Following Directions, which evaluated the participants' ability to interpret, recall, and execute oral commands of increasing length and complexity (Semel et al., 2003). The second was Formulated Sentences, which examined the participants' ability to create sentences when given both semantic (within the context of a picture) and syntactic constraints (a target word) (Semel et al., 2003). One possible reason for the participants' poor performance on these tests could be due to problems with attention or memory. Although it was not known if any of the participants presented with attentional deficits, impaired attention is often observed in cases of FASD (Coles, 2001; Lockhart, 2001). Both of the subtests in question require attending to instructions and recalling verbal information. However, many of the other CELF-4 subtests where the participants showed more success also rely on attention and memory: e.g. for the Recalling Sentences subtest, the participants were required to repeat back a series of sentences increasing length, and in Understanding Spoken Paragraphs participants listened to a story and were then asked questions about that story. As many of the other subtests rely on memory and attention, problems in these areas cannot account for all of the difficulty the participants had on these two subtests. One possibility for future research could be to see if this pattern of impairment holds with a larger group of participants, and how this compares between groups with different FASD profiles and to other clinical populations.

Contrary to most other research in FASD, pragmatic language skills were a specific focus. Our results showed that 75% of the participants in the current study were judged to have pragmatic language difficulties on at least one of the two pragmatic language tests (the TOPL or the CCC-2), which lends support to the notion that these children often have pragmatic language difficulties that could be overlooked. Indeed, 50% of the participants had both general language impairment *and* pragmatic language impairment. Even though the TOPL quotients were lower, on average, than the CELF-4 composites, the difference was not statistically significant. This finding was contrary to expectations, which had predicted that pragmatic scores would be significantly lower than structural and semantic scores. Significant differences might occur in a study where more participants were available. Both the CELF-4 and the CCC-2 indicated similar levels of general language impairment in the participants, lending support to the notion that the CCC-2 is an appropriate instrument for screening for general language impairment.

Contrary to expectations, the CCC-2 did not identify more participants as having social communication impairments than did the TOPL. This might be explained by the fact that the two instruments measure slightly different things. As previously mentioned, the TOPL measures quantitative differences from the typical development of social communication, while the CCC-2 also takes into account qualitatively different behaviours. The two instruments may have measured different aspects of each participant's pragmatic language abilities, which would have led to different scores. Furthermore, the criterion we used in this study to identify pragmatic impairment on the CCC-2 was very conservative, and could have contributed to the difference in the findings. Which, if either, of the two instruments is the better, or more accurate, remains unknown, since each identified some different participants as demonstrating pragmatic impairment and we still lack a "gold standard" assessment of pragmatic function against which to compare.

This study has a number of advantages over previous studies that looked at language in children with FASD. First of all, by using the 4-Digit Diagnostic code, the participants were selected using more stringent and standardized criteria than in some of the previous studies that used non-standardized, clinical judgements for diagnosis. As all the diagnoses were made using exactly the same methodology, this allows for more accurate comparisons between subjects. Furthermore, using this standardized method of diagnosis improves chances for appropriate replicability of this study.

Another major advantage of this study is that it includes measures of pragmatic language. Pragmatic language difficulty has been anecdotally and clinically noted in the individuals with FASD, but only one previous study (Gentry et al., 1998) that examined language ability in individuals with FASD. The results of the current study indicate that the majority of children with FASD do indeed have pragmatic language impairment, and that this important area of skill should not be ignored in evaluation of a child's ability to function in his or her community.

Other advantages of this study include the fact that it included a non-verbal cognitive measure, so that each participant's language ability could be compared to an appropriate measure of cognitive status. Furthermore, all the participants spoke English as their primary language, which eliminates complications that can arise when participants speak or grew up speaking other languages. In addition, because all the participants were within a narrow range of chronological ages (8;0 to 10;0), the group is more homogeneous and clearly defined.

There are also significant limitations, the largest of which is the small number of participants. Initially, the study sought to examine the language abilities of twenty children with FASD; however, only eight participants meeting the inclusion criteria were found. Additional efforts were made to try and recruit participants from the FASD Clinic in Camrose, Alberta, but these efforts were unsuccessful. There are many possible reasons for the low rate of recruitment. Firstly, it is sometimes difficult to recruit participants for studies of FASD, as the child's diagnosis brings with it an element of social stigma for the parent(s). Even if the participants come from adoptive or foster family situations, the perceived stigma could cause caregivers to deny consent for their child to participate in the study. Secondly, those caregivers who *are* willing to allow their children to participate in research studies find that their children are often recruited for many different research projects; this over-sampling of a small group of children could cause otherwise-willing caregivers and social workers to decline participation in order to give their child a break from constant observation. Lastly, as the recruitment criteria for the study were stringent in order to ensure a relatively homogeneous sample and control for potentially confounding variables, the number of possible participants was limited. Since the current study has a small number of participants, generalizability to the overall population is reduced. However, other studies of language in this population also suffer from the same limitation and it is notable that this project has more than twice the number of participants than the only previous study examining pragmatic impairment which looked at 3 participants (Gentry et al., 1998). Furthermore, the large effect sizes, associated with statistically significant differences between both structural and pragmatic language and participant nonverbal IQ scores, indicate that the differences are stable even with a limited sample size.

In addition, the participants came from a wide range of home environments, including foster families, adopted families, and biological families. As language ability can be affected by being raised in a low SES environment, or from inconsistent home environment when young (as can be experienced with foster care), it is possible that the effects of the home environment could contribute to some of the participants' language difficulty. Originally, we had hoped to avoid this difficulty by recruiting only participants from adopted, middle-class family environments. This recruitment criterion was dropped at the suggestion of staff at the Glenrose Rehabilitation hospital, who suggested that it was unrealistic to expect to recruit that narrowly. Still, all of the participants in this study had been in the environment in which they were tested for at least 2 years, so they had relative stability for that period of time.

Also, although all the participants were diagnosed to have FASD using the 4-Digit Diagnostic Code, we did not have access to the specific 4-digit codes, so we were unable to determine how many of the participants had *different* diagnoses on the spectrum. It is possible that different clinical categories present with different profiles of cognition, language, and communication. Our purpose was to determine whether high-functioning children with FASD exhibited language and social communication impairments, but in future, research should address whether there may be specific profiles attached to particular sub-groups.

Finally, all of the testing was done by the author, who, while a trained speech-language pathologist, was not blind to the study's hypotheses. Budget restrictions precluded hiring blinded testers. Still, testing protocols and scoring procedures as mandated by the manuals were followed rigorously. In addition, results from the parent-completed CCC-2 GCC composite were very close to those obtained for the CELF-4, adding credibility to this author's testing.

Despite these limitations, the results of this study are promising. They indicate that children with FASD do present with general and pragmatic language impairments even when their non-verbal cognitive abilities are age-appropriate. Further research in this area is needed to discover whether or not children with FASD present with a particular profile of pragmatic skills, or whether sub-types of FASD might demonstrate specific patterns. Future research could also indicate whether the pattern of general, structural language impairment indicated by the participants' CELF-4 subtests holds for a larger group of participants, or for groups of participants with specific FASD profiles.

Table 1. Summary of Participant Data

	Mean	*Standard Deviation*
Chronological Age	9.31	0.27
TONI-3 Quotient Score	95.38	15.67
CELF-4 CLS Score	78.38	11.28
CELF-4 CLS Score *	75.86	9.442
TOPL Quotient Score	73.75	12.27
CCC-2 GCC Score *	78.43	12.58

* - For the seven participants with CCC-2 data

Table 2. <u>Individual Participant Data</u>

Participant Number	*Sex*	*Chronological Age (yr.; mo.)*	*TONI-3 Quotient Score*	*CELF-4 CLS Standard Score*	*TOPL Quotient Score*	*CCC-2 GCC Standard Score*	*CCC-2 SIDI Score*
1	F	9;1	91	70	58	72	10
2	M	9;2	130	91	81	62	5
3	M	9;8	83	79	68	72	7
4	M	9;8	84	60	70	96	-5
5	M	9;2	88	90	90	77	-13
6	M	8;11	95	96	78	*n/a**	*n/a**
7	F	9;5	87	73	87	95	-6
8	M	9;5	105	78	58	75	-5

* - CCC-2 data unavailable

Table 3. Pragmatic Impairment on TOPL and CCC-2*

	Impaired (≤80)	*Not Impaired (>80)*
TOPL Score	4	3
CCC-2 SIDI Score	4	3

* - For the seven participants with CCC-2 data

Figure 1. Comparison of TONI-3, CELF-4 CLS, and TOPL Scores

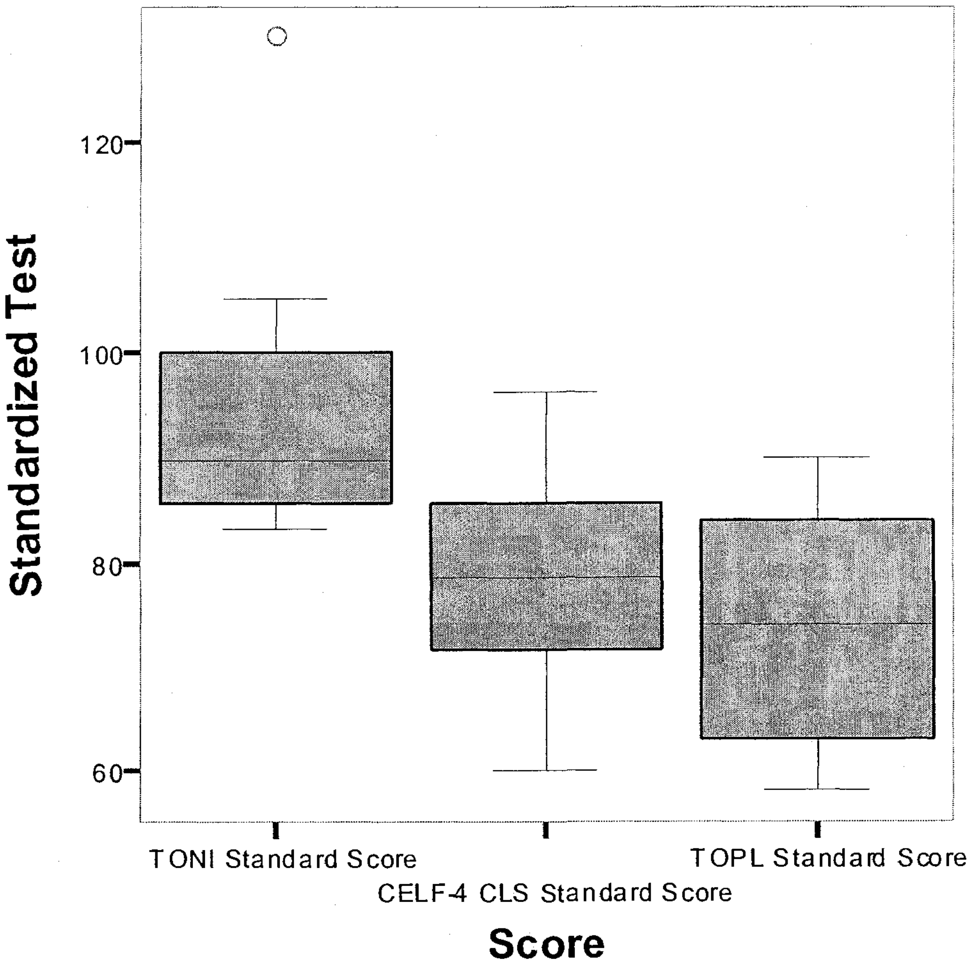


Figure 2. <u>Comparison of CELF-4 CLS and CCC-2 GCC Scores</u>

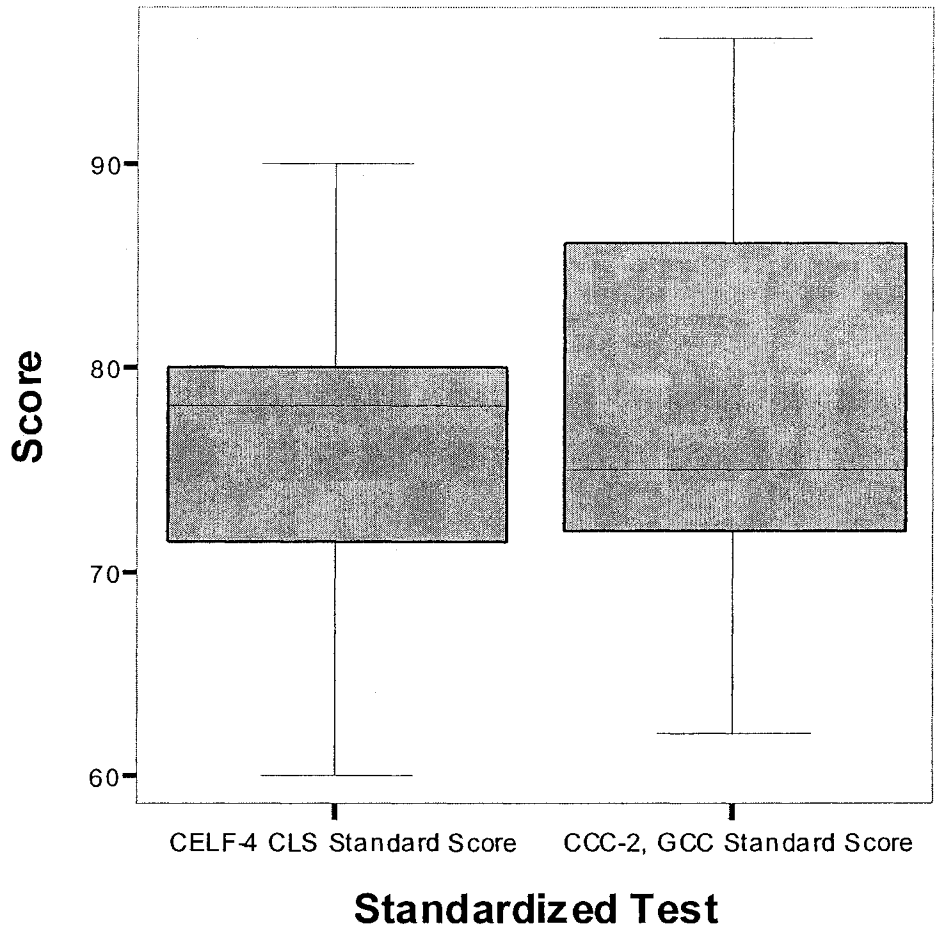
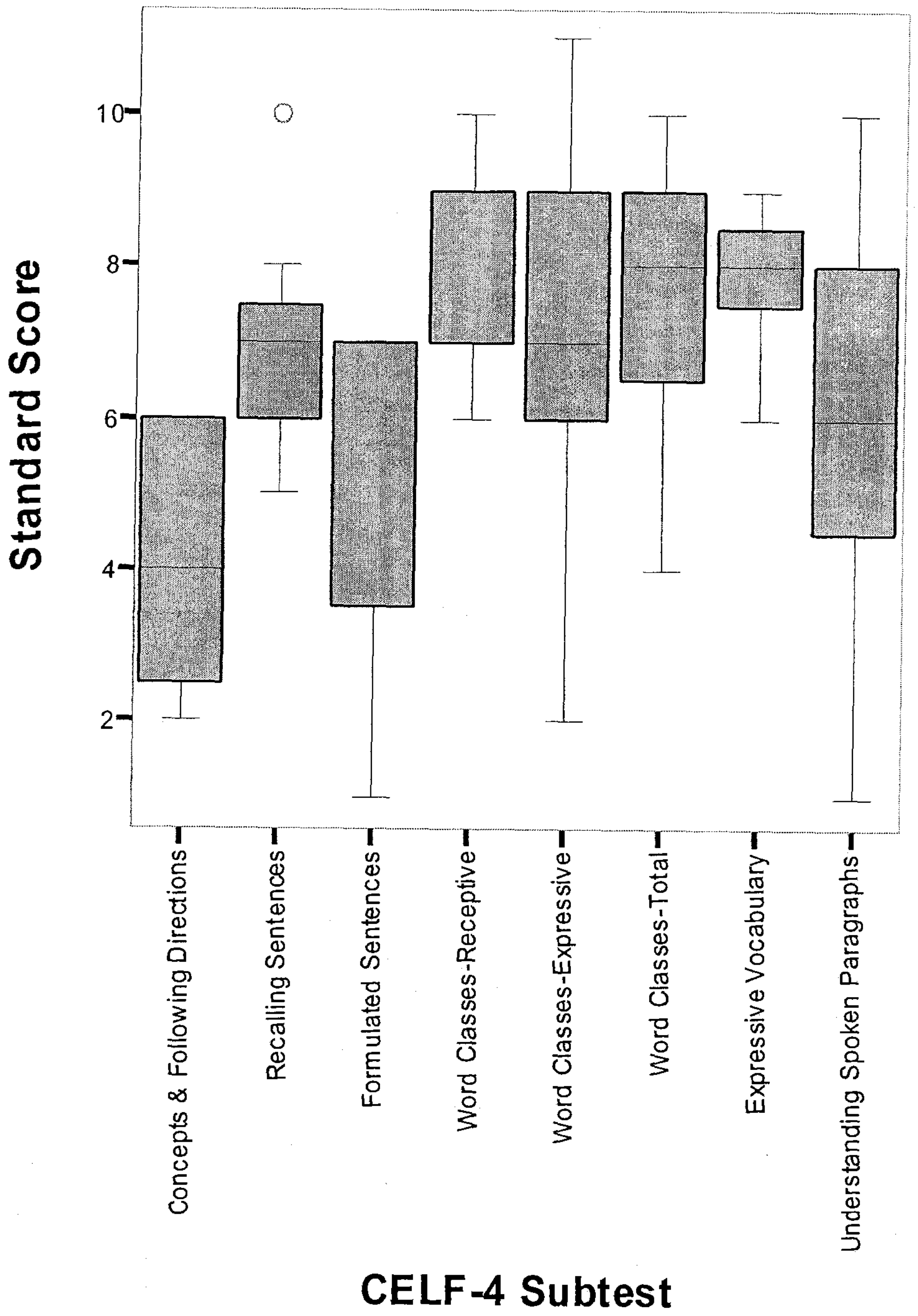


Figure 3. Comparison of CELF-4 Subtest Scores



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Appendix 1: Parent Information Sheet

Title of Research Study:
Pragmatic Language Ability in Fetal Alcohol Spectrum Disorder

Principal Investigator: Joanne Volden, Ph.D., S-LP (C), R. SLP
Associate Professor, Speech Pathology & Audiology
Secondary Investigator: Devin R. Bruce, B.Sc.
M.Sc. Thesis Student, Speech Pathology & Audiology
Phone: (780) 492-7235

Background: Children with Fetal Alcohol Spectrum Disorder (FASD) can have a number of problems. Some have language and communication problems. This study will look at how children with FASD communicate. Your child has been asked to participate because he / she has a diagnosis of FASD and is between 8 and 10 years old.

Procedures: Your child will complete three different tests. One test will measure your child's thinking ability, another test will measure your child's grammar and vocabulary, and the last test will measure your child's social language. The tests take from 75 minutes to 105 minutes. The tests will be done in two sessions at your home. You will also complete a checklist that asks questions about how your child communicates. This checklist takes 15 minutes to complete.

Possible Benefits: There are no direct benefits in participating in this study. Any information gained in this study may be used to perform more research. This may help other children with FASD in the future.

Possible Risks: There are few risks in participating in this study. Your child could become frustrated with the tests or become tired after completing the tests. To reduce stress, the tests will be completed over two sessions, and your child can take a break in a session if he / she wishes.

Confidentiality: Your child's personal records will be kept confidential. Research data collected during this study will not identify your child by name. Data files will use initials and a coded number. The files will be kept in a locked cabinet inside a locked office for at least five years. Your child's file will not be used for further research without another ethics review. Any reports published as a result of this study will not identify your child by name.

Voluntary Participation: You can remove your child from the research study at any time. This will not affect your child's care in any way.

Contact Names and Telephone Numbers:
If you have questions about your child's rights, please call the Patient Relations Office of Capital Health, at 407-1040. The researchers are not connected with this office.

Please contact any of the individuals identified below if you have any questions or concerns:

Paul Hagler, Ph.D
Associate Dean, Graduate Studies and Research
Faculty of Rehabilitation Medicine
(780) 492-9674

Appendix 2: Parent Consent Form

PARENT CONSENT FORM

Title of Project: **Pragmatic Language in Fetal Alcohol Spectrum Disorder**

Principal Investigator: Joanne Volden, Ph.D., S-LP (C), R. SLP Phone Number: (780) 492-0651

Secondary Investigator: Devin R. Bruce, B.Sc. Phone Number: (780) 492-7235

Part 2 (to be completed by the research subject):

	Yes	No
Do you understand that your child has been asked to participate in a research study?	☐	☐
Have you read and received a copy of the attached Information Sheet?	☐	☐
Do you understand the benefits and risks involved in taking part in this research study?	☐	☐
Have you had an opportunity to ask questions and discuss this study?	☐	☐
Do you understand that you are free to withdraw your child from the study at any time, without having to give a reason and without affecting your child's future care?	☐	☐
Do you understand who will have access to your child's records, including Personally identifiable health information?	☐	☐
Do you want the investigator(s) to inform your child's family doctor, pediatrician or Speech Pathologist that your child is participating in this research study?	☐	☐

Name ______________________ **Profession** ______________________

Who explained this study to you? ________________________________

Child's Name ________________________

I agree for my child to take part in this study: YES ☐ NO ☐

Signature of Parent or Guardian ______________________ Date & Time __________

(Printed Name) ______________________________

Signature of Witness ______________________________ Date & Time __________

Signature of Investigator or Designee __________________ Date & Time __________

THE INFORMATION SHEET MUST BE ATTACHED TO THIS CONSENT FORM AND A COPY GIVEN TO THE RESEARCH SUBJECT